An Exploration of the Psychological Aspects of Cluster Headache

Helena Whitley

Candidate Registration Number:100153902

March 2025

Word Count: 37,050

Primary Supervisor: Dr Elisabeth Norton

Secondary Supervisor: Dr Fergus Gracey

Patient and Public Involvement (PPI) Representative: Mr Wayne Nolan

Doctoral Programme in Clinical Psychology

University of East Anglia

Faculty of Medicine and Health Sciences

It should be acknowledged that some of the material in this thesis portfolio was taken from the ClinPsyD Thesis Proposal

Table of Contents

List of Figures and Tables

Figures

Tables

List of Portfolio Appendices

Acknowledgments

I would like to thank Mr Wayne Nolan for introducing me to the world of Cluster Headache and giving me the opportunity to dedicate my thesis portfolio to supporting moving understanding of this debilitating condition forwards. His continuous enthusiasm and reflective personal experience, both in meetings and also via email, has been incredibly motivating throughout the research process. I would also like to thank my supervisors, Dr Elisabeth Norton and Dr Fergus Gracey, for their expert advice throughout the whole process. I would like to thank OUCH for allowing this research to be advertised on their website.

I would also like to thank my friends- you know who you are. Thank you for the cups of tea, words of wisdom, and lunch-time walks to break up the writing process. I would also like to thank my parents for their continuous support, love, and guidance. Thank you for entertaining and indulging my interests and curiosity through my long and winding road of education.

Finally, I would like to thank the participants who I interviewed for the empirical paper of this portfolio; I found it remarkable how open and courageous they were in sharing their difficult experiences. I hope their words, presented through this portfolio, will catalyse further research into Cluster Headache.

Thesis Portfolio Abstract

Background

Cluster Headache (CH) is cited as one of the most painful experiences known to humankind. This thesis portfolio aimed to provide a greater insight into the psychological aspects of CH. A systematic review accumulated evidence of rates of depression and suicidality in individuals living with CH and an empirical paper explored the psychological experience of living with CH.

Methods

A systematic review and meta-analysis was conducted to determine rates of depression and suicidality amongst individuals with CH compared to non-headache controls and individuals with other primary headache conditions (Migraine or Tension-Type Headache (TTH)). Secondly, 13 interviews were conducted with individuals living with CH and this data was analysed using Reflexive Thematic Analysis.

Results

Meta-analyses of 20 studies showed that compared to non-headache controls, adults with CH had much higher levels of depression and suicidality. However, there was no significant difference in depression levels between CH and Migraine individuals. Comparing individuals with CH and TTH, the initial meta-analysis found no significant difference in depression levels, but a sensitivity analysis showed TTH individuals having higher levels of depression. Considerable heterogeneity and publication bias were present. Reflective Thematic Analysis identified five themes relevant to the CH experience: "Darkness", "Battling", "Shifting", "Control", and "Despair". There were differences within these themes based on whether a person was in the moment of pain or between attacks, whether they had the chronic or episodic form of CH, and how long they had lived with CH.

Conclusions

This portfolio highlighted that psychological aspects of CH include increased depression and suicidality. Increased depression was also present for the other primary headache disorders. The empirical paper identified various psychological processes important in the experience of CH which could be the target of psychological treatment.

Chapter One: Thesis Portfolio Introduction

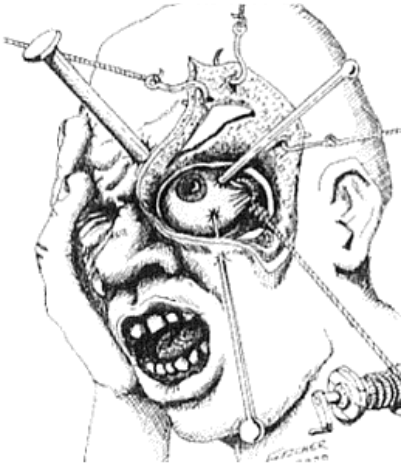
Headache disorders are under-recognised and under-treated (World Health Organisation, 2011). Cluster Headache (CH) is a severe primary headache disorder and is the most common of the trigeminal autonomic cephalgias (Headache Classification Committee of the International Headache Society (IHS), 2018). A primary headache disorder means it is not caused by, or a symptom of, another illness. Other primary headache disorders include Tension-Type Headache (TTH) and Migraine (IHS, 2018). CH is relatively rare, with lifetime prevalence being reported as 0.12% of the global population (Fischera et al., 2008; Wei et al., 2018); although true rates are hard to determine due to frequent misdiagnosis (Ahmed, 2019; Wei et al., 2018). Despite the relatively low prevalence rates, CH is reported to be one of the most severe and enduring conditions and is commonly seen in clinical specialist headache centres (Cronin et al., 2007; Nesbitt & Goadsby, 2012). It is anecdotally more painful than childbirth and painful conditions such as pancreatitis (Burish et al., 2021). The level of pain means the condition has been nicknamed “the suicide headache” due to sufferers presenting with suicidal ideation in a bid to stop the pain (Rossi et al., 2018; Wei & Goadsby, 2021). CH has been associated with compromised quality of life, high co-morbidity with anxiety and depression, work-related disability, and socio-economic burden (D’Amico et al., 2020; Freeman et al., 2022; Ji et al., 2019).

The International Classification of Headache Disorders (ICHD-3) classifies CH as strictly unilateral severe pain, and associated autonomic involvement, and/or restlessness (Hoffman & May, 2018; IHS, 2013; 2018). The pain is typically localised to the unilateral orbital, peri-orbital, and/or temporal areas (British Medical Journal (BMJ), 2024, IHS, 2018). Painful attacks last between 15-180 minutes and one must have at least five attacks before receiving a diagnosis of CH (IHS, 2013; 2018). Frequency of attacks range from every other day, to eight per day, if left untreated (IHS, 2018). Pain is subjective, made clear by the

definition of pain being "an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage" (International Association for the Study of Pain (IASP), 2020; IASP Subcommittee on Taxonomy, 1979; Raja et al., 2020). Therefore, qualitative accounts are helpful to try and understand one's experience. Qualitative accounts of CH have described the pain as similar to a "red-hot poker" being pushed "through your eyeball" (Andre & Cavers, 2021, p. 422) and as "incapacitating, relentless, and paralysing" (Palacio-Cena et al., 2016, p. 1179). See a visual representation of the pain drawn by a sufferer in Figure 1 (Organisation for the Understanding of Cluster Headache (OUCH), 2024). As aforementioned, the level of pain has been suggested to give rise to suicidal thoughts as a mechanism to stop the pain (Rossi et al., 2018; Rozen & Fishman, 2012). Associated with the pain is often ipsilateral autonomic features, such as running drooping eyes, facial sweating, and constriction of the pupils (IHS, 2013; 2018). Furthermore, psychological-behavioural symptoms of agitation, restlessness, and pacing during attacks are also reported (Clinical Knowledge Summaries (CKS), 2024; IHS, 2013; 2018).

Figure 1

Visual Depiction of the Experience of Cluster Headache Pain (OUCH, 2024).



CH is categorised into two forms based on frequency of the attacks (Ahmed et al., 2019; IHS, 2018). The first is the "episodic" form (ECH), which is more common (85-90% of sufferers) and involves a period (weeks or months), often termed a "bout", when one has frequent attacks, which often occur at the same time each day. ECH sufferers then have a period of pain-free remission which is at least three months in length and is often more than a year. Typically, individuals have one or two bouts each year, which often start in the spring or autumn, although the cause of this circannual rhythm is not well understood (IHS, 2018). The less common form is "chronic" CH (CCH; 10-15% of sufferers). This is characterised by attacks occurring for at least a year, with no significant remission period (less than 3 months). CCH normally evolves out of ECH (May, 2013).

The rarity of the condition (0.12% of the global population, Fischera et al., 2008) means it is not well known amongst clinicians, and so diagnosis is often missed or delayed (Schindler & Burish, 2022). The peak age of onset is between 20 and 40 years (Ahmed et al., 2019; Mazoni et al., 2016; Russell, 2004) but case reports have shown individuals as young as two years old (Majumdar et al., 2009) and as old as 83 years old (Evers et al., 2002) presenting with the condition. It is reported to be around three times more common in men than women (Mazoni et al., 2016), which sets it apart from other primary headaches (i.e. Migraine and TTH) that are more common in women (Delaruelle et al., 2018). However, the observed gender gap in CH has reduced in recent years (Ekbom et al., 2002), potentially due to increased diagnosis amongst women, rather than a change in actual rates (CKS, 2024; Hoffman & May, 2018). Importantly, a meta-analysis of 16 population-based studies across four continents (Fischera et al., 2008) highlighted the limited evidence in countries south of the equator, meaning the above epidemiological statistics may not apply in the southern hemisphere (Hoffman & May, 2018).

The exact cause of CH is unknown (Wei et al., 2018). Current theories are based on animal, clinical, and neuroimaging studies and implicate three systems; the hypothalamus, the trigeminovascular system, and the autonomic system (Hoffman & May, 2018; Wei et al., 2018; Wei & Goadsbey, 2021). The hypothalamus is the coordinating centre of the body, linking the nervous and endocrine systems, and is vital for regulating the body's circadian rhythm. It is hypothesised that the hypothalamus is responsible for generating headache attacks and plays a role in the regularity of bouts at a certain time of year (circannual) and attacks at a certain time of day (circadian) (Wei & Goadsbey, 2021). The involvement of the trigeminovascular system is thought to explain the severe pain in the unilateral trigeminal distribution. Finally, the involvement of the autonomic system is thought to explain symptoms such as drooping and tearing of the eye. There is a link with genetics, with research reporting that a first degree relative of a CH sufferer has between a five and 39 times increased risk of developing the condition (Belin et al., 2023). A systematic review (Elbadawi et al., 2021) examined risk factors associated with CH and supported the hereditary link, as well as indicating that smoking, alcohol consumption, head trauma, and being male are other risk factors.

In terms of prognosis and treatment of CH, it is considered a lifelong condition, but evidence indicates that frequency of attacks reduces, and period of remission increases, as one ages (Wei et al., 2019). Treatment recommendations within the NHS are all dedicated to medical approaches, none of which were specifically created for CH (Ander & Cavers, 2021; Andersson et al., 2017; National Institute for Health and Care Excellence (NICE), 2022). Treatments are separated into abortive techniques, for the acute attack, interim treatments to reduce level of pain and frequency of attacks, and preventative techniques (Wei & Goadsbey, 2021). Abortive techniques include triptans, and short-burst oxygen therapy, interim treatments include corticosteroids, and preventative medications include verapamil and

lithium (NICE, 2022; Wei & Goadsby, 2021). Neuromodulation devices have also been trialed for preventative techniques, such as invasive occipital nerve stimulation, and non-invasive vagus nerve stimulation (Wei et al., 2019). Efficacy for all these medical approaches is modest, and their mechanism of action incompletely understood (Andersson et al., 2017; Kandel & Mandiga, 2023; Peng & Burish, 2023). Furthermore, pharmacological treatments come with side-effects, require monitoring, and are often only a short-term solution due to drug resistance (Freeman et al., 2022; Kandel & Mandiga, 2023). Neuromodulation devices come with surgical complications such as infection, and pain at the generator site, and non-invasive vagus nerve stimulation is contra-indicated for CCH, and individuals with various co-morbid medical conditions (Schindler & Burish, 2022). Furthermore, all these medical solutions are difficult to access, due to cost and complex referral processes (Freeman et al., 2022). Because of the above, a review concluded that even with the best available treatment, individuals with CH experience significant burden (Grinberg et al., 2021) and the European Headache Alliance criticised the current management of CH among healthcare services (Rossi et al., 2018). This indicates that research is required to improve the management of the condition CH and so alleviate suffering for this group.

CH is coined “the suicide headache” and has been associated with psychopathology (D’Amico et al., 2020; Freeman et al., 2022; Ji et al., 2019; Rossi et al., 2018). Despite this, there are no psycho-behavioural guidelines for treatment. This is contrary to the other primary headache conditions, Migraine and TTH, where the NICE guidelines include psychological interventions (NICE, 2022; NICE, 2024). A review of the literature called for a greater understanding of the debilitating nature of CH, concluding that “it is surprising so little research has examined psychological and behavioral treatments to improve the lives of people with cluster headache” (Grinberg et al., 2021, p.3). This was corroborated by a second review (Schenck & Andrasik, 2019).

Outline of Thesis Portfolio

This thesis portfolio aimed to provide a greater understanding of the psychological aspects of CH to then inform support for the CH community. Chapter Two of the thesis portfolio is a systematic review which accumulated evidence on psychological outcomes of CH compared to other primary headache conditions, and non-headache controls. Chapter Three explains how further exploratory research is required to understand the psychological processes present for individuals with CH which influence psychopathology. This acts as a bridging chapter to Chapter Four which is the main empirical paper. The empirical paper aimed to promote deeper understanding of psychological processes involved in the CH experience using qualitative techniques, as a step towards developing psychological interventions. The focus of the empirical paper was suggested by a Patient and Public Involvement (PPI) representative, who has the chronic form of CH and felt they would have benefitted from psychologically-informed care. Chapter Five provides additional commentary on the methods used in the empirical paper. Finally, Chapter Six brings together the whole portfolio. This chapter critically evaluates the work, and then presents recommendations for clinical practice and ongoing research.

Chapter Two: Depression and Suicidality in Individuals with Cluster Headache; a Systematic Review and Meta-analysis

Submitted to the International Journal of Clinical Practice

Word count: 6,900

Miss Helena Whitley, University of East Anglia

Dr Fergus Gracey, University of East Anglia

Miss Annabel Fellows, University of Bath

Miss Karina Pazvantova, University of Bath

Dr Elisabeth Norton, University of East Anglia

Abstract

Background

The primary headache disorder, Cluster Headache (CH), has been associated with psychopathology. However, research in the area is inconsistent and often limited by small sample sizes. This review systematically explored the evidence regarding depression and suicidality levels amongst adults with CH compared to adults without headaches and adults with Migraine or Tension Type Headache (TTH).

Methods

The review was registered with PROSPERO (registration number: CRD42024489570). Four databases were searched for papers from inception to November 2024. Papers had to be empirical research, available in English, and compare adults with CH to a control in terms of depression and/or suicidality symptoms. Outcomes with sufficient data were pooled using random-effects meta-analysis. The quality of studies was assessed using an adapted Newcastle-Ottawa Scale.

Results

229 studies were screened by title and abstract, 76 were read in full, and 20 articles were included in the review. The quality of 95% of studies was rated as medium to poor. Meta-analyses showed that compared to non-headache controls, adults with CH had significantly higher levels of depression (n= 16; SMD= 0.92; 95% CI: 0.74-1.11) and suicidality (n=4; SMD= 0.71; 95% CI: 0.46-0.95). When comparing adults with different primary headaches, meta-analyses reported no significant difference in depression levels. However, after sensitivity analysis, TTH adults had moderately higher depression levels than CH individuals. It was not possible to explore suicidality between primary headache individuals as there was not enough research.

Conclusions

This review found that CH individuals had higher levels of depression and suicidality than non-headache controls. Depression levels amongst CH adults were not higher than adults with Migraine, and adults with TTH may have higher levels. Considerable heterogeneity and publication bias mean caution should be taken when interpreting these results. However, findings suggest that clinicians could consider assessing the mental health of individuals presenting with CH.

Key Words: Cluster Headache, Primary Headache Disorders, Depression, Suicidality, Systematic Review, Meta-Analysis

Introduction

Cluster Headache (CH) is a severe primary headache disorder which affects approximately 0.12% of the population worldwide (Fischera et al., 2008). CH has been rated as one of the worst pains a person can experience (Burish et al., 2021; Grinberg et al., 2021). The condition results in recurring headache 'attacks' which can occur daily and last from minutes to hours. There are two sub-types of CH; Episodic CH (ECH) and Chronic CH (CCH) (Ahmed et al., 2019; IHS, 2018). The former involves periods where an individual will suffer frequent attacks of pain, and then remission periods which are pain-free, and at least three months in length. CCH, on the other hand, involves no significant period of remission. Treatments for the condition are limited at best (Grinberg et al., 2021). Research exploring persistent pain conditions has shown that it is important to consider pain from a biopsychosocial perspective (Gatchel et al., 2007). All systematic reviews related to CH have focused on the bio-medical features of the condition, such as pharmacological treatment (Robbins et al., 2016) and examination of risk factors such as head trauma, specific biomarkers, familial links, smoking, and alcohol consumption (Elbadawi et al., 2021; Long et al., 2021; Søborg et al., 2024; Waung et al., 2020). There is a growing evidence base related to non-biological factors which may be relevant to CH. This systematic review aimed to explore such evidence, specifically psychological aspects of the condition.

The extreme level of the pain means CH has been coined the name "the suicide headache" (Wei & Goadsby, 2021). Sufferers have reported that suicidal ideation results as a short-term solution to escape the experience of pain. Some research has also indicated that the condition is related to higher rates of depression (Grinberg et al., 2021). This is perhaps unsurprising, considering the level of pain resulting from CH, combined with its recurrence, delay in diagnosis, limited treatment, and reported negative impacts on various aspects of a person's life (Grinberg et al., 2021). Indeed, a large US survey study with over 1,000 CH

participants found that 20% of individuals lost their jobs due to the condition (Rozen & Fishman, 2012). Furthermore, CH has been found to impact one's ability to engage in social relationships, complete cognitive tasks, and drive (Rozen & Fishman, 2012). Whilst CH has been related to both suicidality and depression, the research is inconsistent and has not been systematically explored.

Depression and Cluster Headache

Depression is a common and leading cause of disability (WHO, 2021), characterised by feelings of hopelessness, having little interest in life, and associated symptoms of disruption in sleep, appetite, cognitive deficits, and suicidal ideation (NICE, 2024). Depression has been associated with increased pain sensitivity (Hermesdorf et al., 2016). Furthermore, depression has been associated with poor health behaviours, such as lower adherence to medication, smoking, and alcohol use, potentially due to the symptoms of deactivation, poor motivation, and feelings of hopelessness (An & Xiang, 2015; Grenard et al., 2011; Jang et al., 2020). The reciprocal association between depression and pain (Vadivelu et al., 2017) and depression's association with poor self-management, means it may be that CH individuals with co-morbid depression have increased suffering. Therefore, it is important to have an understanding of the true rates of depression amongst CH individuals.

Rates of depression have been reported as low as 6% to as high as 57% in CH studies, with there being variability based on the definition of depression and population studied (Grinberg et al., 2021). Two large European studies, each with over 1,500 individuals living with CH, reported that over 40% of individuals met the criteria for depression (Donnet et al., 2007; Pohl et al., 2020). However, these studies did not compare to a control group, meaning it is difficult to determine that the CH diagnosis was the cause of the increased depression, rather than another variable. This is particularly relevant when considering concepts such as depression, because depression is common in the general population (WHO, 2021). Some

studies have included a control group, albeit with small sample sizes of 40 (Gómez-Mayordomo, 2020) and 14 (Mitsikostas & Thomas, 1999) CH individuals. These studies reported higher rates of depression in a CH group compared to non-headache controls. These small samples are characteristic of much CH research. Interestingly, the study by Mitsikostas and Thomas (1999) also recorded depression levels amongst individuals with the other two primary headache disorders: Migraine and Tension-Type Headache (TTH). Authors reported that rates of depression appeared similar across the primary headache disorders (Mitsikostas & Thomas, 1999).

Suicidality and Cluster Headache

The most extreme result of depression could be argued to be suicidal ideation and behaviour (WHO, 2021). To encompass both ideation and behaviour, the current review will use the term "suicidality". The first mention of the relationship between CH and suicidality was in 1939: "our patients were disabled by pain so severe that several had to be constantly watched for fear of suicide" (Horton, 1939). Some more recent research has explored this proposition. A survey of over 1,000 CH individuals reported that 55% of individuals had suicidal ideation, which is significantly higher than general population rates, whereas suicidal behaviours were similar to the general population (Rozen & Fishman, 2012). However, this study did not involve verification of CH diagnosis and contrasts to another study which reported higher rates of suicidal behaviour as well as ideation during a CH attack (Ji Lee et al., 2019).

Overall, CH has been related to both depression and suicidality. However, research findings are inconsistent, lack systematic exploration, and have low power due to research often having involved small sample sizes (Markley & Buse, 2006). Furthermore, related research did not always include a control group, or ensure study participants had a genuine diagnosis of CH. It is important to review rates of psychopathology in the CH population,

because of the likely reciprocal relationship between psychopathology and pain. Additionally, current NICE guidelines (2022) for CH all relate to medical management. This is contrary to the other primary headache conditions, Migraine and TTH, where psycho-behavioural interventions such as relaxation and Cognitive Behavioural Therapy (CBT) are recommended (NICE, 2022; NICE, 2024). This could be because there is more rigorous research examining psychological factors in these headaches; for example, a systematic review concluded that Migraine is associated with increased risk of depression (Amiri et al., 2019), and another concluded that psychological interventions could be helpful for TTH (Qin et al., 2024).

Purpose of the Review

This review aimed to collate the evidence which exists about depression and suicidality in individuals living with CH compared to a control group, either a healthy non-headache group, or individuals with other primary headache conditions (Migraine or TTH). It is hoped such evidence will be helpful in determining whether future research should explore psychological avenues for treatment for CH. This is important, because current approaches to managing CH, which are purely biomedical, are limited in effectiveness (Landmark et al., 2024; Lund et al., 2023).

Review Questions

1. Is there evidence that levels of depression and suicidality are elevated amongst adults with Cluster Headache compared to adults without head pain?
2. Is there evidence that levels of depression and suicidality are different amongst adults with Cluster Headache compared to adults with Migraine or Tension-Type Headache?

Methods

Cochrane guidelines were used to guide this systematic review, and reporting of the review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses

(PRISMA) (Higgins et al., 2019; Page et al., 2021). See Appendix A for PRISMA Checklist. The systematic review was registered with PROSPERO (registration number: CRD42024489570) and the protocol can be accessed on the PROSPERO website. A slight amendment was submitted, and accepted, by PROSPERO, to make the review specifically focused on depression and suicidality rather than psychopathology as a whole.

Eligibility

The PECOS framework (Dekkers et al., 2019; Morgan et al., 2018; Paul & Leeflang, 2021) was used to define the eligibility criteria and objectives of the review.

Population

The population of interest were adults (≥18 years of age) living anywhere in the world. Animal studies were excluded. Research was excluded if participants were undergoing a medical operation, such as having an invasive occipital nerve stimulator implanted, as an invasive procedure may introduce a large confounding variable.

Exposure

The study sample had to include at least 10 individuals diagnosed with CH, either the episodic or chronic form. This meant that case studies and series were excluded. Individuals with CH were excluded if they had another headache e.g., comorbid Migraine. If the headache sample was mixed with participants with other headache conditions, at least 80% of the sample needed to be comprised of CH individuals, or the data regarding CH individuals needed to be separated.

Comparison

Studies had to include a comparison between the exposure group and another group, either a healthy control with no headache or participants with another primary headache condition (Migraine or TTH).

Outcome

Studies had to include a measure of depression and/or suicidality. Suicidality referred to either suicidal ideation or behaviour. The method of measure which was acceptable was flexible and included a valid and reliable questionnaire, a clinical interview, and/or registered diagnosis. A valid and reliable questionnaire was preferential as this indicated the level of symptoms. In terms of suicidality, questionnaire measures had to be focused on suicidality rather than have only one question related to it. This was to ensure sufficient depth and reliability in assessing this complex construct. For example, the Patient Health Questionnaire 9 (PHQ-9) was not acceptable, despite question nine being related to suicidal ideation (Kroenke et al., 2001), as one single response related to suicidality was not deemed to provide enough information about this concept. Suicidality may be a feature of depression itself (WHO, 2021), and so there may be some overlap in these experiences for people living with CH and resulting questionnaire scores. As this was the first review of its kind, limits were not placed on whether depression/suicidality was seen as a risk factor or a consequence of CH.

Study Design

Peer-reviewed articles from any period were included in the review, assuming they were available in English. The study had to be primary research. Therefore, books and reviews were excluded. There were no further strict criteria in terms of acceptable study design.

Search Strategy

Searches took place on MEDLINE, PsycINFO, Scopus, and Cumulative Index to Nursing and Allied Health Literature (CINAHL). The search involved the key terms "Cluster Headache" OR "Trigeminal Autonomic Cephalalgia" being combined with the operator AND with “Depression” OR “Suicide” OR “Suicidal Ideation”. Exploded terms and Medical

Subject Headings (MESH)/index terms were used to enhance results. See Appendix B for full set of electronic search terms.

Study Selection

Papers identified using the above search strategy were screened in three stages, as recommended by the PRISMA 2020 flowchart (Page et al., 2021). Records identified from the database search were imported into Endnote21. Duplicates were removed, and then all papers were screened based on the above inclusion/exclusion criteria based on their title/abstract. The first author (HW) screened all papers, and two external reviewers screened 25% of studies (AF and KP). Papers included after screening were then read in full to determine eligibility by the first author (HW), and again 25% were screened by two external reviewers (AF and KP). Having a complete dual review process was deemed important, as this can increase the number of relevant studies included (Stoll et al., 2019). Importantly, the external reviewers were assistant psychologists who had previous research experience, but were unaware of the review topic, so reducing risk of bias in study selection, whilst maintaining accuracy.

Data Extraction

After studies were selected, the included papers were read in full again and the key information was extracted manually into a Microsoft Excel document. The search function on the desktop was also used to improve accuracy. Two external reviewers (AF and KP) extracted data from 25% of the papers to check accuracy. Specifically, the following information was collected: study characteristics (author, country, design), the CH and comparison sample information (sample size, age, gender), method of ascertainment of CH diagnosis, measure of depression/suicidality, and relevant qualitative findings. When available, continuous data regarding the level of depression/suicidality was collected, with Means and Standard Deviation being the first choice. If multiple continuous scales were used,

the one deemed to focus most on depression/suicidality was used. For example, if research presented the Beck Depression Inventory (BDI; Beck et al., 1961), and the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983), the BDI data was collected, as the HADS also measures anxiety. If continuous data was not available, other measures, such as diagnosis, or quantitative data of above cut-off symptom scores were collected. This approach was adopted from a comparable systematic review comparing depression levels in a clinical and non-clinical group (Gambadauro et al., 2019).

Quality Assessment

The Newcastle-Ottawa Scale (NOS) for Assessing the Quality of Nonrandomized Studies in Meta-Analysis, case-control version (Wells et al., 2014), was employed to methodologically assess quality of selected studies. The NOS was chosen as it is the most used tool for this type of study (Ma et al., 2020) and can be adapted based on the topic, which is recommended by the Cochrane Handbook (Higgins, 2011; Kalfas et al., 2022). The tool is separated into three areas: (1) selection, (2) comparability, and (3) outcome, and studies are awarded a certain number of 'stars' based on quality. An adapted version of the checklist was created in which a total of eight stars could be awarded per study, with more stars indicating higher quality (See Appendix C). Studies allocated eight stars were judged low risk of bias, six to seven stars medium risk, and five stars or below indicated high risk of bias (Muka et al., 2020). The primary researcher (HW) completed 100% of quality checks, and two external reviewers (AF and KP) and the primary research supervisor (EN) assessed 30% of papers in total.

Synthesis of Results

Relevant qualitative results were presented in a summary table for each study. Quantitative data regarding depression/suicidality were collected and collated into a Microsoft Excel spreadsheet for the meta-analysis. Meta-analysis was conducted using

"metafor" packages in RStudio (R Core Team 2019). It was predicted that there would be a large between-study heterogeneity and so random-effects model was used, as this is a more conservative approach (Kim et al., 2001).

Depression/suicidality symptoms are most commonly measured as continuous data using various available scales. Therefore, Standardized Mean Difference (SMD) was used as the effect size for this meta-analysis (Lipsey & Wilson, 2001). There was a preference for calculation of Cohen's d effect size using Means and Standard Deviation (SD) when available (Higgins et al., 2023). If research provided Median and Interquartile Range data, a calculator was used to convert this into Means/SD (Abbas et al., 2024). For the studies which measured depression/suicidality in a dichotomous way, Odds Ratios (ORs) were used, or calculated from proportion data, and then converted into a Cohen's d using an online calculator (Lin, 2024). When adjusted and unadjusted ORs were provided, the unadjusted ratio was used, as studies may have adjusted for different factors, meaning the ORs would not be comparable and inappropriate for combination in meta-analysis (Chang & Hoaglin, 2017).

Individual effect sizes were pooled using random-effects meta-analysis to estimate the pooled difference in depression/suicidality rates amongst CH vs comparison groups. Forest plots were created to display the meta-analysis results visually. Heterogeneity was assessed using the Cochran's Q test and I^2 statistics for each analysis (Higgins et al., 2013; Higgins & Thompson, 2002; Huedo-Medina et al., 2006). Prediction intervals were also calculated to report on the range in which the true effect size of 95% of future studies would fall (Higgins et al., 2009). Predication intervals allowed clinical interpretation of heterogeneity (Inthout et al., 2016). When interpreting the Cohen's d effect size, ≤ 0.39 was considered small, 0.40-0.69 moderate, and ≥ 0.70 large (Cohen, 1988; Schünemann et al., 2023).

Several sensitivity analyses were planned to gauge the stability of findings. Firstly, it was planned to remove any studies which appeared to be outliers when inspecting forest

plots. Secondly, meta-analyses were re-run which excluded studies which were identified as high risk of bias from the NOS quality assessment. Thirdly, only studies which reported Means/SDs in their results were included. Publication bias was examined by inspecting funnel plots, and asymmetry of the funnel plot was tested using Egger's test (Sadeghi, 2024). The non-parametric trim-and-fill method was also applied (Sadeghi, 2024).

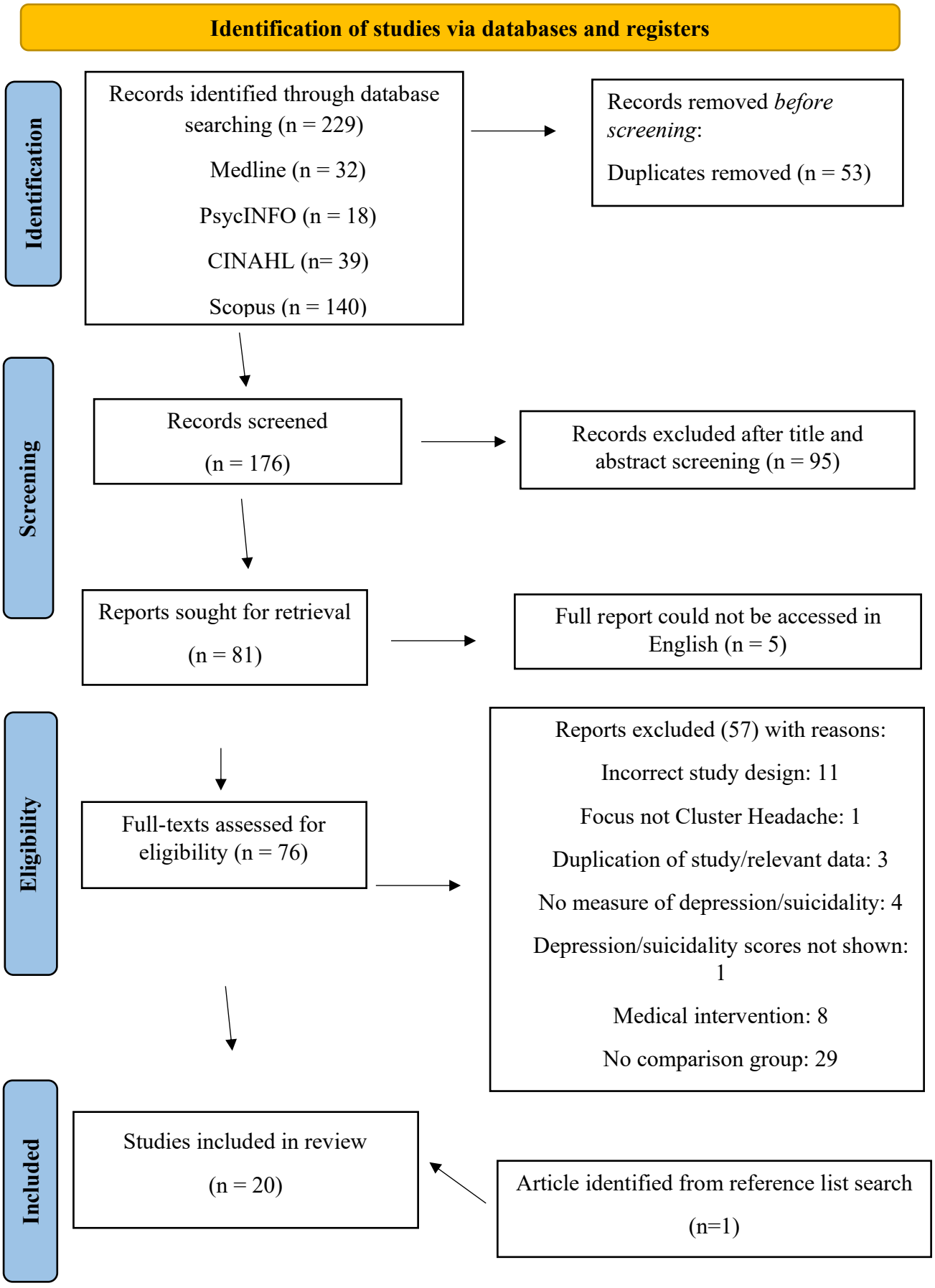
Results

Study Selection

The database search was carried out on 12/11/2024. An initial search identified 229 studies across the four databases. 176 remained after 53 duplicates were removed. 81 remained after titles/abstracts were screened. Most papers were excluded at this stage due to them being reviews, case studies/series, or not measuring depression/suicidality. 81 papers were then read in full, of which 19 were identified as eligible. See PRISMA flow diagram in Figure 2 which details why papers were excluded at this stage. One final paper was identified (Mitsikostas & Thomas, 1999) when reading around the area, meaning a total of 20 papers were included. External reviewers (AF and KP) screened 44 papers' abstracts/titles (25%) and 26 papers in full (32%). No discrepancies occurred in decisions made, indicating high inter-rater reliability. See Figure 2 which details this process through a PRISMA flow diagram.

Figure 2

PRISMA Flow Diagram



Study Characteristics

Table 1 gives a summary of the 20 included studies' general characteristics and relevant qualitative findings. See Appendix D for a table which includes more detailed study characteristics (i.e. age and sex of study participants) and individual study summary statistics (Means/SD or ORs). The papers were published between 1999 and 2024. All research was conducted in the northern hemisphere; with 60% of the studies originating in Europe, 20% in Asia, and 20% the United States. The majority (95%) of the studies measured depression/suicidality symptoms in a cross-sectional way, with only Liang and colleagues (2013) employing a longitudinal design. The number of relevant participants per study ranged from 34 to 3,892,260. Notably, the largest study, with nearly 4 million individuals, used population data for the control (Crespi et al., 2022). The median sample size, of relevant study subjects, in a study was 236. The total number of individuals in each group across the 20 included studies was as follows: 11,775 individuals with CH, 3,924,308 headache-free controls, 3,952 Migraine controls, and 698 TTH controls.

CH diagnosis was confirmed in 95% of studies by either a pre-registered diagnosis or assessment by a specialist clinician, using direction from established criteria (IHS, ICD, or ICHD). The exception being one paper, where the diagnosis followed the ICD classification, but this was not confirmed by a specialist (Koo et al., 2021).

Depressive symptoms were measured using a continuous questionnaire for 70% of studies: five studies used the Hamilton Depression Rating Scale (HAM-D; Hamilton, 1980), two studies used the Patient-Health Questionnaire (PHQ-9; Kroenke et al., 2001), four studies used the BDI (Beck et al., 1961), and three studies used the HADS (Zigmond & Snaith, 1983). The remaining 30% of studies recorded depression in a dichotomous way using health insurance claims (one study), diagnosis (three studies), clinical interview (one study), and self-report (one study). Suicidality was recorded in a continuous way for one study (Suicidal

Behaviour Questionnaire; SBQ-R; Soman et al., 2001), and the remaining used dichotomous methods: health insurance claims (one study), clinical interview (one study), and diagnosis (one study).

Table 1

Summary Table of Study Characteristics

First Author (Year)	Country	Study Type	CH Group	CH Diagnosis	Control Group	Depressive /Suicidality Measurement	Relevant Findings
Anagnostou (2017)	Greece	Cross-sectional study	N= 4 CCH, 17 ECH	ICHD-II	27 Migraine individuals. 20 TTH individuals	HAM-D	Significantly more depressive symptoms in CH and TTH group than Migraine group
Ballesta-Martínez (2022)	Spain	Cross-sectional study	N= 31 ECH	ICHD-III	20 headache-free controls; age-, gender-, and education-matched	HADS-D	Significantly more depressive symptoms in CH group compared to control group
Chen et al (2010)	Taiwan	Cross-sectional study	N=51 CH	ICHD-II	772 Migraine individuals. 218 TTH individuals	HADS	No significant difference in depression scores between head pain conditions
Choong (2017)	U.S	Observational retrospective database study	N= 7,589 CH	CH ICD-9 code in database	30,341 headache-free controls, matched for age and sex	Depressive disorder and suicide-related claims	Significantly more claims in CH group than control for depressive disorders and suicidal ideation

Crespi (2022)	Norway	Nationwide observational study	N= 1,891 CH	CH ICD-10 code on registry	N= 3,892,260: total adult Norway population in 2016. ORs adjusted for sex and gender	Diagnosis from Norwegian registry	Significantly more suicide attempts and diagnoses of depression in CH group compared to control population
Díaz-de-Terán (2021)	Spain	Cross-sectional study	N=28 ECH, 19 CCH	IHS Criteria	40 matched headache-free controls. Matched for sex, age and BMI	HAM-D	Significantly more depressive symptoms in CH group
Gesztelyi (2006)	Hungary	Cross-sectional study	N= 11 CH	Diagnosed by specialist, but does not state criteria applied	231 Migraine individuals. 176 TTH individuals. Age and gender differed significantly between the groups	BDI	Statistically significant difference in depressive symptoms among the different groups, with CH group having lowest levels of depressive symptoms
Gil-Martínez (2019)	Spain	Cross-sectional study	N= 12 ECH, 8 CCH	Diagnosed by a headache specialist, as defined by IHS criteria	16 headache free controls. Matched for sex, age, education, BMI, employment	BDI-II	Significantly more depressive symptoms in CH group

Gómez-Mayordomo (2020)	Spain	Cross-sectional study	N= 40 ECH	ICHD-III criteria	Forty age and sex matched headache-free men	HAM-D	Significantly more depressive symptoms in CH group
Işcan (2024)	Turkey	Cross-sectional study	N= 18	ICHD-III criteria	18 headache-free controls matched for age and gender	BDI	Significantly more depressive symptoms in CH group
Jorge (1999)	Argentina	Case-control study	N= 21 ECH	IHS Criteria	21 TTH individuals. Matched for age, sex and education	HAM-D	No significant difference in depression scores between head pain conditions.
Joshi (2017)	U.S.	Population-based Study	N= 75 CH	Searched registry for CH diagnosis	152 age and sex matched headache-free controls	Co-morbid disorders reported in patient's records using ICD	Patients with CH had significantly higher diagnoses of depression than control group
Jürgens (2011)	Germany	Multicentre, prospective study	N= 27 CCH, 26 ECHa, 22 ECHna	ICHD-II criteria	24 Migraine individuals and 31 head-ache free controls	The Mini-DIPS, a validated structured clinical interview	Depressive/suicidal symptoms more prevalent in CH patients compared to headache-free. Especially increased in CCH patients

Kim (2020)	South Korea	Cross-sectional study	N= 191 CH	ICHD-III criteria	63 headache-free controls. 36 Migraine controls. Age and sex matched	PHQ-9	Significantly more depressive symptoms in CH group compared to headache-free controls
Kim (2024)	South Korea	Cross-sectional study	N= 423 CH, 4% CCH	ICHD-III criteria	52 headache-free controls age and sex matched	PHQ-9	Significantly more depressive symptoms
Koo (2021)	U.S.	Observational, case-control study	N= 56 ECH, 44 CCH	ICHD-III criteria	135 headache-free controls. Matched for age, sex, race, income and marital status	SBQ-R. Self-report of lifetime Depression	Significantly more CH than control participants had lifetime passive and active suicidal ideation, plans for suicide, and lifetime depression.
Liang (2013)	Taiwan	Population based, follow-up, study	N= 673 CH (19 were CCH)	CH ICD-9 code in database	Age and sex matched controls. 2,692 individuals with Migraine and 2,692 headache-free controls	Depression diagnosis ICD - included only when coded by psychiatrists	Median 2.5-year follow-up. CH cohort had a greater risk for developing depression compared to the control cohort, but not Migraine cohort
Louter (2016)	Netherlands	Cross-sectional study	N= 462 CH	ICHD-II criteria	177 headache-free controls	HADS-D	Significantly more depressive symptoms for CH group

Mitsikosts (1999)	Greece	Cross-sectional study	N=10 ECH, 4 CCH	IHS Criteria	150 headache-free controls age and sex matched, 170 Migraine individuals, 263 TTH	HAM-D	Depressive symptoms higher in CH group compared to headache-free group, but lower than the Migraine and TTH groups
Torkamani (2015)	UK	Cross-sectional study	N= 11 CCH, 11 ECH	ICHD-II criteria	12 headache-free controls. Age matched to CH group, but significantly different gender ratio	BDI-II	Significantly more depressive symptoms in CH group compared to controls

Note. CH= Cluster Headache; CCH= Chronic Cluster Headache; ECH= Episodic Cluster Headache; ECHa= Episodic Cluster Headache Active Bout, ECHna= Episodic Cluster Headache Non-Active Bout, TTH= Tension Type Headache; ICHD= The International Classification of Headache Disorders; IHS= International Headache Society; ICD= International Classification of Diseases; PHQ-9= Patient Health Questionnaire; BDI = Beck Depression Inventory; HADS-D = Hospital Anxiety and Depression Scale; Depression subscale; HAM-D= Hamilton Depression Rating Scale; SBQ-R= Suicidal Behaviour Questionnaire Revised

Quality of Included Studies

The Newcastle Ottawa Scale was used to assess methodological quality and risk of bias of studies included. There was only one disagreement between reviewers in terms of ratings and together a joint decision was made. The quality of studies ranged from three to eight, out of a possible total score of eight (mode of six, 40%), see Table 2. One study was deemed low risk of bias, ten medium risk of bias, and nine high risk of bias. The most common limitation of studies was the lack of representativeness of the samples, so limiting generalisability of findings. Indeed, half of the studies included samples drawn from one specialist headache clinic. Another common weakness of studies was the cross-sectional nature of all but one study. A strength of the reviewed literature was that 70% of studies matched the CH and controls group on important characteristics, such as age and sex.

Table 2

Quality Assessment of the Included Studies using the Adapted Newcastle-Ottawa Quality Assessment Scale

Study Author	Selection			Comparability		Outcome		Total
	Representative	CH Diagnosis	Control Selection	CH Excluded in Control Group?	Matched	Measure Used	Timing of Test	Longitudinal	
Anagnostou, 2017		*		*		*	*		4
Ballesta, 2022		*	*		*	*	*		5
Chen, 2010	*	*	*			*	*		5
Choong, 2017	*	*	*	*	*		*		6
Crespi, 2022	*	*	*		*	*			5
Díaz-de-Terán, 2021		*	*	*	*	*	*		6
Gesztelyi, 2006		*	*			*	*		4
Gil-Martínez, 2019		*	*	*	*	*	*		6
Gómez-Mayordomo, 2020		*	*	*	*	*	*		6

Işcan, 2024		*	*	*	*	*	*		6
Jorge, 1999		*	*		*	*	*		5
Joshi, 2017	*	*	*	*	*	*			6
Jürgens, 2011		*	*	*					3
Kim, 2020	*	*	*	*	*	*	*		7
Kim, 2024	*	*	*	*	*	*	*		7
Koo, 2021	*			*	*	*	*		5
Liang, 2013	*	*	*	*	*	*	*	*	8
Louter, 2016	*	*	*	*		*	*		6
Torkamani, 2015		*	*	*		*	*		5
Mitsikosts, 1999		*	*	*	*	*	*		6

Note. Studies could be allocated a total of eight possible stars. Studies allocated eight stars were deemed low risk of bias, studies allocated six-seven stars were medium risk of bias, and studies with five stars or below were high risk of bias (Muka et al., 2020).

Quantitative Synthesis

Four meta-analyses were conducted with the available data in the R Metafor package.

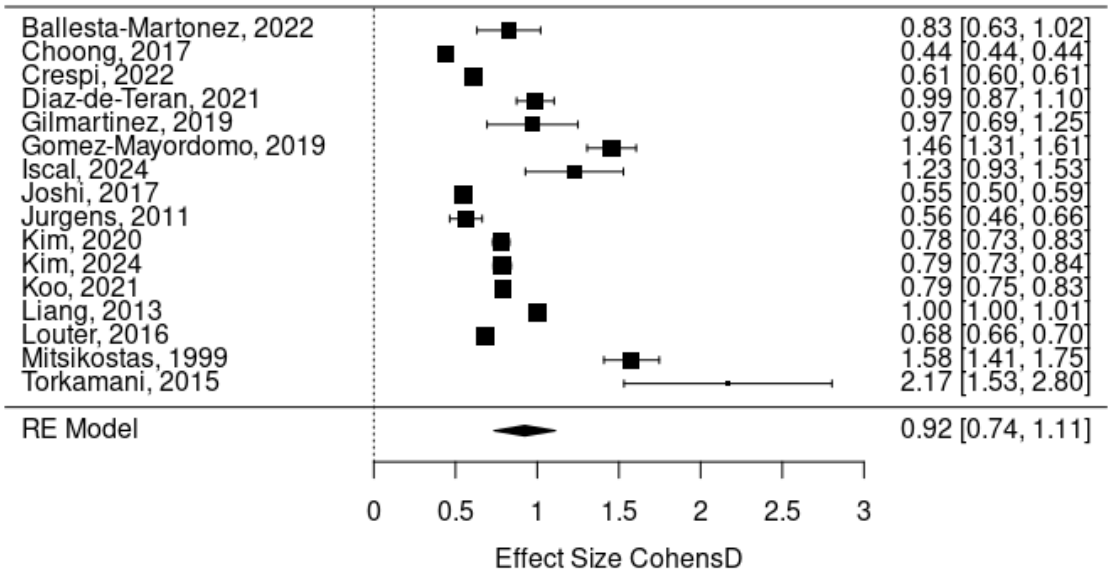
Cluster Headache vs Headache-Free Control Meta-Analysis

Depressive Symptoms.

A total of 16 studies reported data on depressive symptoms in individuals with CH and Headache-Free controls (comprising a total of 3,935,979 CH or headache-free individuals). Two studies (Jürgens et al., 2011; Torkamani et al., 2015) separated CH individuals into CCH and ECH. For these studies, the depression score was averaged. The levels of depression were significantly higher among individuals with CH compared to those without headaches and this was a large effect size (N= 16; SMD= 0.92; 95% CI, 0.74-1.11, p <.00001) (See Figure 3). Based on the Cochran's Q statistic ($\chi^2(15) = 162490.41$, $p < .0001$) and the I^2 statistic (100%) there was considerable heterogeneity between studies. The prediction interval ranged from 0.17-1.67.

Figure 3

Forest Plot for the Meta-analysis of Depression Levels Between Cluster Headache and Headache-Free Adults



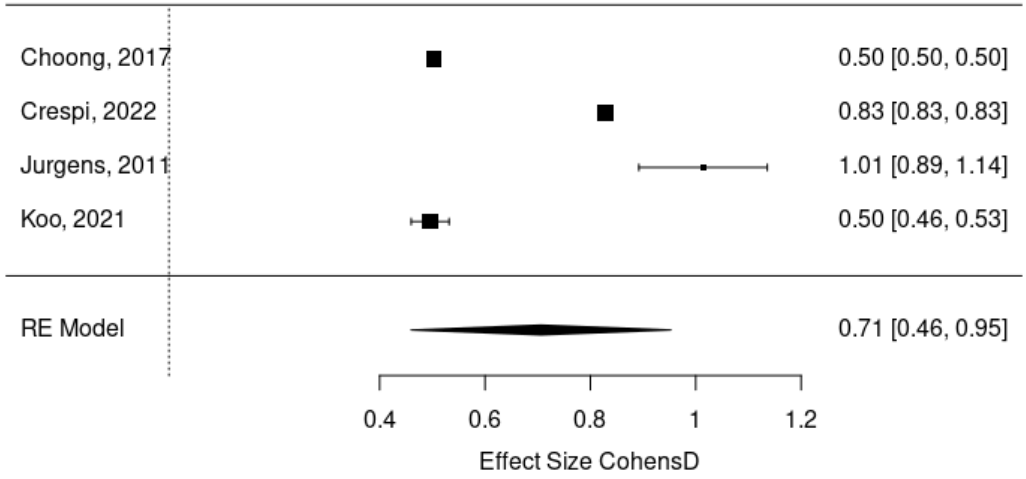
Note. Each study depicted with individual Cohen's d statistic (95% confidence intervals). RE Model = Random-effects model.

Suicidality.

Four of the 20 studies also reported data on suicidality symptoms, either attempts or ideation (comprising a total of 3,930,531 CH or headache-free individuals). One study (Jürgens et al., 2011) separated CH individuals into CCH and ECH. For this study, the suicidality score was averaged. Suicidality symptoms were significantly higher among individuals with CH compared to Headache-Free controls, with a large effect size (N=4; SMD= 0.71; 95% CI, 0.46-0.95, p <.0001) (See Figure 4). Based on the Cochran's Q statistic ($\chi^2(3) = 344508.59$, $p < .0001$) and the I^2 statistic (100%) there was considerable heterogeneity between studies. The prediction interval ranged from 0.16-1.25.

Figure 4

Forest Plot for the Meta-analysis of Suicidality Levels Between Cluster Headache and Headache-Free Adults



Note. Each study depicted with individual Cohen's d statistic (95% confidence intervals). RE Model = Random-effects model.

Cluster Headache vs Primary Headache Controls Meta-Analyses

Eight studies reported data relevant to the question regarding depressive symptoms in a CH versus another primary headache group. Only one of these studies (Jürgens et al., 2011) included suicidality symptoms. This study reported that suicidality was higher amongst all CH individuals compared to Migraine sufferers, particularly for CCH sufferers. As there was only one study, a meta-analysis was not possible for suicidality symptoms.

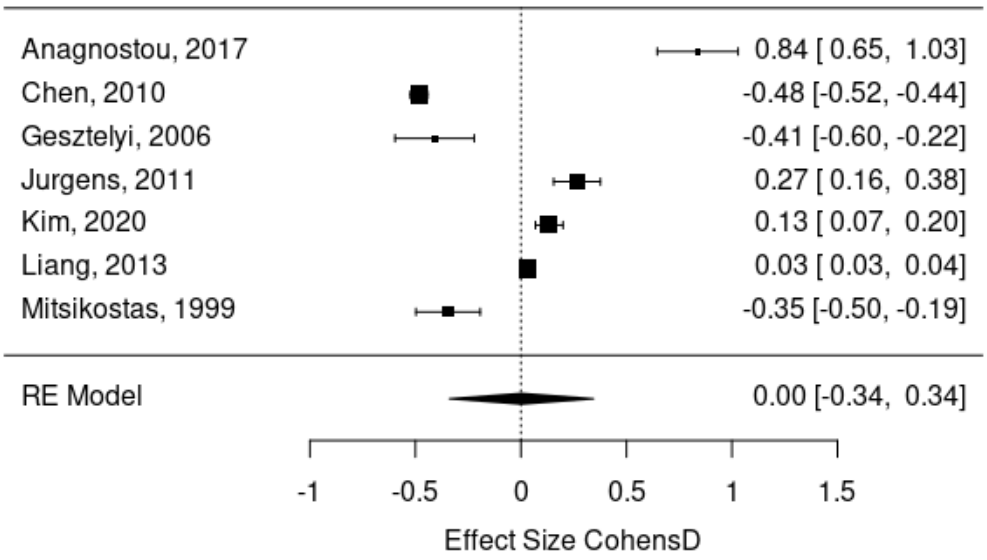
Migraine.

Seven studies compared Migraine and CH participants (comprising a total of 4,988 CH or Migraine individuals). There was no significant difference between levels of depression in the CH and Migraine group (N=7, SMD= 0.0027; 95% CI -0.34-0.34, p =0.97) (see Figure 5). Based on the Cochran's Q statistic ($\chi^2(6)= 734.94$, $p <0.0001$) and the I^2

statistic (99.62%) there was considerable heterogeneity between studies. The prediction interval ranged from -0.96-0.96.

Figure 5

Forest Plot for the Meta-analysis of Depression Levels Between Cluster Headache and Migraine Adults



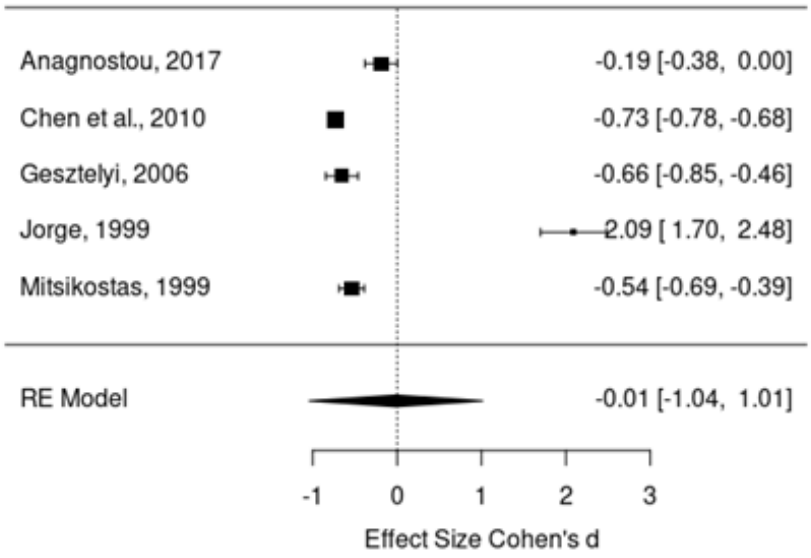
Note. Each study depicted with individual Cohen's d statistic (95% confidence intervals). RE Model = Random-effects model.

Tension-Type Headache.

Five studies reported rates of depression amongst individuals with CH and TTH (comprising a total of 816 CH or TTH individuals). There was no significant difference in levels of depression between the CH and TTH group (N=5, SMD= -0.01; 95% CI -1.04-1.01, p= 0.98) (see Figure 6). Based on the Cochran's Q statistic (χ^2 (4)= 222.55, p < 0.0001) and the I^2 statistic (99.58%) there was considerable heterogeneity between studies. The prediction interval ranged from -2.52-2.49.

Figure 6

Forest Plot for the Meta-analysis of Depression Levels Between Cluster Headache and Tension-Type Headache Adults



Note. Each study depicted with individual Cohen's d statistic (95% confidence intervals). RE Model = Random-effects model.

Sensitivity Analysis

Results of sensitivity analyses are reported in Table 3. On visual inspection of each forest plot, certain outliers were evident and so were removed for each meta-analysis and analysis was re-run. The outliers removed are detailed in Table 3. When outliers were removed, individuals with CH still had significantly higher rates of depression and suicidality than Headache-Free controls and these effects were moderate/large. Compared to individuals with Migraine, there remained no significant difference between rates of depression. However, after an outlier was removed, individuals with TTH had significantly higher levels of depression than CH, and this effect was moderate. For all meta-analyses, considerable heterogeneity remained.

Considerable heterogeneity was explored further through two further subgroup analysis. Analysis was re-run with 1) all high-risk-of-bias studies (≤ 5 stars on Newcastle Ottawa Scale) being removed and 2) only studies which used a continuous measure for depression/suicidality being included. These further analyses were only possible if more than two studies remained in the analysis. Therefore, it was only possible for depression meta-analyses for the Headache-Free and Migraine group. The heterogeneity could not be explained by risk-of-bias of the study or measure of depression, with variability remaining considerable ($I^2 > 98\%$) after sensitivity analyses. In all these sub-analyses, depression was significantly raised in individuals with CH compared to Headache-Free controls, and there was no significant difference between CH and Migraine individuals.

Table 3

Sensitivity Analyses

	N	Cohen's d	95% CI	I^2
Adjusted for Outliers Removed *(Studies removed)*				
Headache-Free Depression *(Torkamani et al., 2015)*	15	0.87*	0.70-1.03	100%[a]
Headache-Free Suicidality *(Jürgens et al., 2011)*	3	0.61*	0.39- 0.83	100%a
Migraine Depression *(Anagnostou et al., 2017)*	6	-0.13	-0.38-0.13	99.32%[a]
Tension-Type Depression *(Jorge et al., 1999)*	4	-0.54*	-0.77- -0.31	91.49%[a]
Adjusted for Study Bias				
Headache-Free Depression	11	0.94*	0.72-1.15	99.98%[a]
Migraine Depression	3	-0.05	-0.33-0.24	98.45%[a]
Adjusted for Measurement Type				
Headache-Free Depression	10	1.10*	0.85-1.35	98.99%[a]
Migraine Depression	5	-0.06	-0.54-0.42	99.28%[a]

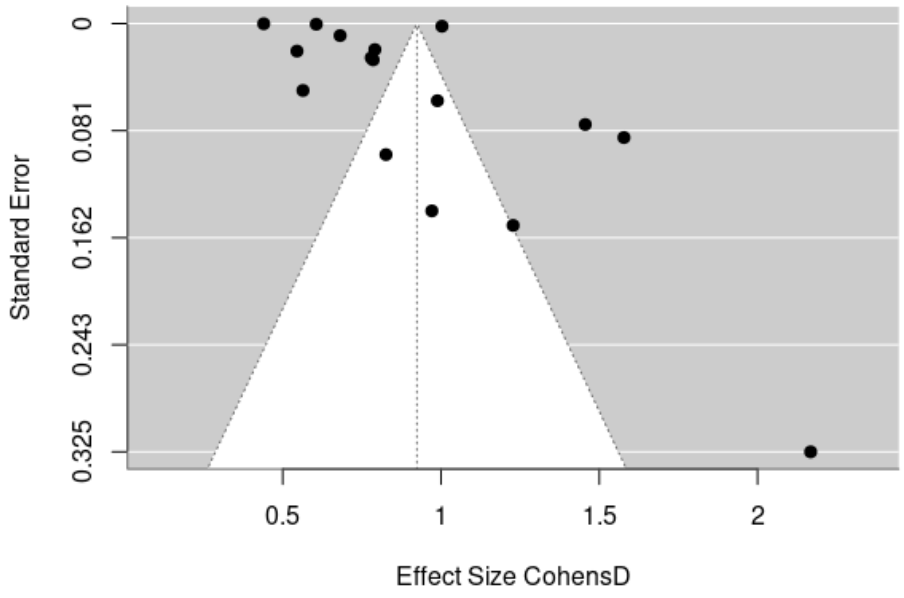
Note. N= number of studies included, *Significant at p<.0001, [a]-X-statistic is significant

Publication Bias

Publication bias was only reported for meta-analyses with 10 studies or more as per guidance (Mavridis & Salanti, 2014). Therefore, only the Headache-Free depression meta-analysis was assessed for publication bias. A visual inspection of a funnel plot indicated the distribution was asymmetrical, suggesting publication bias (see Figure 7). This was confirmed by a significant Egger's test ($z = 4.15$, $p < .0001$). To account for this bias, trim and fill was used, but no trimming was performed.

Figure 7

Funnel Plot for the Meta-analysis of Depression Levels Between Cluster Headache and Headache-Free Adults



Discussion

This meta-analysis explored the level of depressive symptoms and suicidality amongst individuals with Cluster Headache (CH) compared to controls without headaches and controls with other primary headache conditions: Migraine or TTH. Twenty papers were identified which were relevant to these questions and four meta-analyses were conducted.

Main Findings

Results of the meta-analyses demonstrated that adults with CH have more depressive and suicidality symptoms compared with headache-free adults and this effect size was large. This pattern remained significant throughout sensitivity analyses. However, there was considerable heterogeneity. To account for this, prediction intervals were considered (Hak et al., 2016). These intervals indicated that one can expect a diagnosis of CH to result in an increased level of psychopathology ranging from a trivial impact on depressive and suicidality symptoms in some individuals to a considerable one in others (Prediction intervals: Depression: 0.17-1.67; Suicidality: 0.16-1.25). Albeit the impact sometimes being trivial, the prediction intervals indicate that in 95% of future populations, there would be some increase in depression and suicidality symptoms among individuals with CH diagnosis compared to the general population with no head pain. Therefore, in relation to the first question of this review, authors would conclude that there is evidence that levels of depression and suicidality are elevated amongst CH individuals compared to individuals without headaches.

In terms of answering the question regarding psychopathology between CH individuals compared to individuals with Migraines and TTH, it was not possible to explore suicidality rates as there was insufficient data. In terms of depression, meta-analyses initially found that the difference in levels of depression was not significant between CH individuals and other primary headache individuals. For Migraine individuals, this remained a stable

finding throughout sensitivity analyses. However, for the TTH analysis, when an outlier was removed in sensitivity analysis, adults with CH had significantly lower levels of depression than adults with TTH, and this effect was moderate. Therefore, in answering the second question of the review, authors conclude that there is no evidence of a difference in levels of depression between CH and Migraine individuals, however TTH adults may experience more depression.

The findings in this review should not be overstated due to heterogeneity and publication bias. Furthermore, for some meta-analyses there was limited research meaning it was not possible to conduct sensitivity analysis or assess publication bias. However, the findings are in line with the majority of existing literature in the field, therefore adding weight to the robustness of findings. Indeed, in relation to depression, literature has cited that a CH diagnosis is associated with depression, however this has previously been based on small sample sizes, or research without a control group (Donnet et al., 2007; Gómez-Mayordomo, 2020; Grinberg et al., 2021; Mitsikostas & Thomas, 1999; Pohl et al., 2020). Furthermore, more broadly, it is well-recognised that chronic pain and depression have a bi-directional relationship. Indeed, reports have cited that up to 60% of chronic pain patients have co-morbid depression (Roughan et al. 2021). The association between chronic pain and depression is incompletely understood, however research points to a bio-psycho-social explanation. For example, studies have shown an overlap in neuroplasticity changes in the brain when one is depressed and living with chronic pain (Sheng et al., 2017). Furthermore, chronic pain has a hugely detrimental impact on individuals' lives; including difficulty with employment, social life, and engaging with pleasurable activities. Lewinsohn and colleagues' theory of depression (1974) suggests that when individuals do not engage with meaningful activities their mood reduces due to lack of positive reinforcement, which then results in a vicious cycle of engaging less with activities and so resulting lower mood (Mazzuchelli et al.,

2010). This mechanism may apply for chronic pain sufferers including CH individuals, particularly as the average age of onset is between 20 and 40 years (Ahmed et al., 2019), meaning these impacts come at a time when most individuals are at the most productive time of their lives for family, social life, and career (Rossi et al., 2018).

The finding that CH individuals seem to not have higher rates of depression than other primary headaches, and may have lower depression rates than individuals with TTH, is interesting considering the pain from CH is reported to be so much greater (Nesbitt & Goadsby, 2012). It has been hypothesised that the long pain-free periods which exist for ECH individuals may result in less impact on one's mental health than other headache disorders (Markley & Buse, 2006), and this could explain the finding. Furthermore, emotional disturbance and mental tension have been implicated as a risk factor and trigger for TTH and Migraine (Chowdhury, 2012; Fumal & Schoenen, 2006, NICE, 2022; Pescador & De Jesus, 2024; Shah et al., 2025) but generally are not cited as a risk factor for CH (Elbadawi et al., 2021). Therefore, perhaps depression is first and foremost for Migraine and TTH and results in some headache occurrence, rather than Migraine and TTH resulting in depression. As the majority of research included in the review was cross-sectional, conclusions about the direction of the relationship between depression and headache are not possible to draw, but this may explain the current systematic review findings.

The finding that CH individuals had higher suicidality than healthy controls is in line with most previous literature; with CH being referred to as "the suicide headache" (Wei & Goadsby, 2018). This systematic review did not measure suicidality at different times of the condition (i.e. in the moment of the attack, in the bout, or between bouts) and therefore conclusions cannot be made regarding the psychological processes and function behind the suicidality. A study by Ji Lee and colleagues (2019), not included in the review due to lack of control group, examined 175 CH individuals. Ji Lee and colleagues (2019) reported that rates

of suicidal ideation and behaviour were increased during an attack, but reduced throughout the bout, and were below levels in the general population during periods of remission for ECH sufferers. This drop could be due to euphoria related to being in a pain-free period and would suggest suicidality comes from wishing to stop the pain, but the current review cannot make conclusions regarding this. In relation to suicidality and other primary headaches, there was not enough research to conduct a meta-analysis. The one study which did report on suicidality amongst CH versus Migraine individuals reported higher suicidal ideation among CH individuals (Jürgens et al., 2011), however no firm conclusions can be made based upon one study. Therefore, whilst the review can conclude that CH is related to suicidality, it cannot be said that out of primary headache disorders, CH specifically, is "the suicide headache".

Methodological Limitations with Reviewed Literature

The systematic review identified a general lack of research measuring psychopathology amongst adults living with CH. This lack was particularly related to research comparing different primary headache conditions and measuring suicidality rates amongst CH individuals. The systematic review also highlighted various methodological weaknesses with existing literature. Indeed, 95% of studies were rated as medium or high risk of bias. A key limitation was that most of the research was cross-sectional in nature. Therefore, inferences regarding the direction of the relationship between headache diagnosis and psychopathology cannot be drawn. However, the one study (Liang et al., 2013) that did have a longitudinal design echoed the main finding of the meta-analysis; with CH individuals having an increased risk of developing depression compared to healthy controls but not compared to Migraine individuals. This is early evidence that the diagnosis of CH results in psychopathology. Another limitation of reviewed literature was that the representativeness of the samples were suboptimal, with half being drawn from one specialist headache clinic. This

limits generalisability of findings to non-specialist contexts. Furthermore, all the included research was conducted in the Northern Hemisphere making the results less applicable globally.

Another limitation of the majority of research included in the review was that researchers did not separate CH individuals based on whether they had CCH or ECH. The two papers that did report on depression and suicidality separately for ECH and CCH individuals (Jürgens et al., 2011; Torkamani et al., 2015) found that the level of depressive and suicidality symptoms were higher for CCH than ECH. Due to lack of literature, the current systematic review averaged their results, but authors wonder if chronic individuals' true psychopathological experience is being lost by most research in the field assuming CCH and ECH individuals are comparable. These nuances may also explain some of the heterogeneity.

Strengths and Limitations of the Review Process

In terms of evaluating the review process itself, a strength of this systematic review and meta-analysis was that it was the first, to authors' knowledge, reporting on the association between CH and depressive/suicidality symptoms. The review was based on pre-defined eligibility, quality of studies was considered, and various sensitivity analyses were conducted to test the robustness of effect sizes found. The meta-analysis had the benefit that much pre-existing research had small sample sizes and so pooling studies together increased the power of findings. Another strength was that the systematic review only included studies with a control group, meaning inferences regarding whether CH diagnosis is the factor influencing psychopathology, as opposed to another confounding variable, were able to be made. Further, studies were only included if participants' diagnosis of CH was confirmed by a specialist using internationally accepted criteria. This was important due to high rates of CH self-diagnosis (OUCH, 2024). Furthermore, studies were included from across the world,

assuming they could be accessed in English, from any time point. This flexible approach was taken to allow for an unbiased overview of the available literature.

However, there were several limitations with the review process. One limitation related to the unresolved heterogeneity in all meta-analyses. Authors wondered if this could be partly due to most studies not differentiating individuals based on whether they had ECH or CCH, despite these sub-types having distinct differences in terms of symptomology (Wei et al., 2021), and the two studies which did measure them separately reporting higher rates of psychopathology for CCH (Jürgens et al., 2011; Torkamani et al., 2015). Authors also wondered if heterogeneity could be due to differences in the way depression was measured (continuous questionnaire vs dichotomous techniques). However, sensitivity analysis in which studies were excluded if they used dichotomous techniques still highlighted considerable heterogeneity. Heterogeneity could also be partly due to the papers in the sample coming from Europe, the USA, and Asia. In these different parts of the world there are large variations in socio-cultural beliefs about mental health and access to healthcare (Dawkins et al., 2021). For example, there is a reported East-West divide, with stigma being higher amongst Asian (Eastern) countries, particularly for depression (Krendl et al., 2020). These cultural differences could impact reported levels of depression and suicidality amongst CH patients. Furthermore, these differences could impact whether appropriate psychological treatment is available and so the ongoing prevalence of psychopathology. A final factor to consider as a potential cause of heterogeneity was the high variability in sample sizes across the studies (sample size ranged from 34 to 3,892,260). All appropriate papers were included, irrespective of sample size, due to the limited research available, and hence desire to include as much research as possible. However, this inclusive approach could have increased heterogeneity. This relates to a statistical limitation related to the use of Cohen's d. This effect size was used as it is a commonly used standardised effect size and accounted for

variation of depression/suicidality measurements (Higgins et al., 2023). However, authors wonder whether Hedge's g would have been preferable as there were differences in the sample sizes between studies, and this is accounted for more with Hedge's g (Goulet-Pelletier & Cousineau, 2018). Notably, one of the meta-analyses was repeated using Hedge's g and there was very limited change in the meta-analysis output; indicating this statistical decision was unlikely to have had a large impact on the study's conclusions.

A final limitation of the current study was that when it came to suicidality, authors did not differentiate based on suicidal ideation and suicidal behaviours. This was due to the limited literature available. However, previous literature has indicated that there may be a difference between ideation and action for CH individuals (Ji Lee et al., 2019; Rozen & Fishman, 2012) and future research would benefit from making this distinction.

Clinical Implications and Suggestions for Future Research

The findings of this meta-analysis have important implications for clinical guidelines. It is recommended that healthcare professionals are made aware of the potential psychological impact of CH. This could be achieved through mandatory training for staff likely to come in contact with CH sufferers; namely GPs and clinicians in headache services. Findings could also be disseminated through media campaigns; for example, the results of this research will be published on the OUCH webpage. Professionals would then be able to keep psychopathology in mind when engaging with CH patients and consider whether liaison with, or referral to, services such as NHS Talking Therapies is required. Furthermore, current NICE guidelines for CH treatment (2022) are all pharmaceutical. These findings indicate that, like Migraine and TTH, perhaps there should be guidelines regarding psychological interventions to manage increased depression and suicidality for CH sufferers. Before such guidelines can be introduced, research exploring the psychological processes underlying the depression and suicidality is required.

A key finding of the systematic review is the scarce evidence regarding this topic; particularly comparing CH individuals to other primary headache conditions, measuring suicidality, and examining CCH and ECH sufferers separately. Future research is warranted to explore these gaps, potentially focused specifically on CCH individuals to explore their psychological profile. The fact CCH does not have any significant period of remission without pain means it can be hypothesised to be more psychologically damaging (Wei et al., 2021). Future research should also employ longitudinal designs to allow inferences to be made regarding the direction of the relationship between a CH diagnosis and depression and suicidality.

Conclusion

In this systematic review and meta-analysis, adults with CH had significantly higher levels of depressive symptoms and suicidality than individuals without headaches and this difference was large. Comparing adults with CH to adults with other primary headache conditions, there was no significant difference in depression levels compared to Migraine individuals. However, individuals with TTH may have higher levels of depression. High levels of heterogeneity mean caution should be taken when interpreting these results, however they do echo most previous research. These findings are important for clinical practice, suggesting individuals with CH should be considered for mental health support. Future research is warranted to explore what psychological support would be most helpful. It is worth considering whether individuals with chronic CH are particularly vulnerable to psychopathology.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Not applicable

Availability of data and materials

All data generated or analysed during this study are included in this published article and its supplementary information files. This information is currently in Appendix D.

Competing interests

The authors declare that they have no competing interests.

Funding

There was no grant funding to support this study, however it was conducted as part of the fulfilment of a Clinical Psychology Doctoral Programme funded by NHS England.

Authors' contributions

HW was the main author, with EN being primary supervisor and FG secondary supervisor. HW conducted all analysis, and had frequent research meetings with their supervisors. AF & KP were external reviewers and screened papers, extracted data, and quality assessed a certain percentage of papers.

Acknowledgements

See page five of the thesis portfolio.

References

Abbas, A., Hefnawy, M. T., & Negida, A. (2024). Meta-analysis accelerator: a comprehensive tool for statistical data conversion in systematic reviews with meta-analysis. *BMC Medical Research Methodology, 24*(1), 243. https://doi.org/10.1186/s12874-024-02356-6

Ahmed, F., Bahra, A., Tyagi, A. & Weatherby, S. (2019). *National headache management system for adults.* British Association for the Study of Headache. http://www.bash.org.uk

Amiri, S., Behnezhad, S., & Azad, E. (2019). Migraine headache and depression in adults: a systematic Review and Meta-analysis. Migräne und Depression bei Erwachsenen: Ein systematisches Review und Meta-Analyse. *Neuropsychiatrie : Klinik, Diagnostik, Therapie und Rehabilitation: Organ der Gesellschaft Osterreichischer Nervenarzte und Psychiater*, *33*(3), 131-140. https://doi.org/10.1007/s40211-018-0299-5

An, R., & Xiang, X. (2015). Smoking, heavy drinking, and depression among U.S. middle-aged and older adults. *Preventive Medicine, 81,* 295-302. https://doi.org/10.1016/j.ypmed.2015.09.026

Anagnostou, E., Vikelis, M., Tzavellas, E., Ghika, A., Kouzi, I., Evdokimidis, I., & Kararizou, E. (2017). Photophobia in primary headaches, in essential blepharospasm and in major depression. *The International Journal of Neuroscience*, *127*(8), 673-679. https://doi.org/10.1080/00207454.2016.1231185

Ballesta-Martínez, S., Del Pilar Navarro-Pérez, M., Bellosta-Diago, E., & Santos-Lasaosa, S. (2022). Theory of mind: a new perspective on cluster headache-a cross-sectional study.

Neurological Sciences: Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology, *43*(10), 6047-6051. https://doi.org/10.1007/s10072-022-06234-1

Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbauch, J. (1961). Beck Depression Inventory (BDI) [Database record]. *APA PsycTests.* https://doi.org/10.1037/t00741-000

Burish, M. J., Pearson, S. M., Shapiro, R. E., Zhang, W., & Schor, L. I. (2021). Cluster headache is one of the most intensely painful human conditions: Results from the International Cluster Headache Questionnaire. *Headache*, *61*(1), 117-124. https://doi.org/10.1111/head.14021

Chang, B. H., & Hoaglin, D. C. (2017). Meta-analysis of Odds Ratios: Current good practices. *Medical Care, 55*(4), 328-335. https://doi.org/10.1097/MLR.0000000000000696

Chen, P. K., Fuh, J. L., Chen, S. P., & Wang, S. J. (2010). Association between restless legs syndrome and migraine. *Journal of Neurology, Neurosurgery, and Psychiatry*, *81*(5), 524-528. https://doi.org/10.1136/jnnp.2009.191684

Choong, C. K., Ford, J. H., Nyhuis, A. W., Joshi, S. G., Robinson, R. L., Aurora, S. K., & Martinez, J. M. (2017). Clinical Characteristics and Treatment Patterns Among Patients Diagnosed With Cluster Headache in U.S. Healthcare Claims Data. *Headache: The Journal of Head and Face Pain*, *57*(9), 1359-1374. https://doi.org/10.1111/head.13127

Chowdhury D. (2012). Tension type headache. *Annals of Indian Academy of Neurology, 15*(Suppl 1), S83-S88. https://doi.org/10.4103/0972-2327.100023

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences, 2nd edition.* Lawrence Erlbaum Associates: Mahwah.

Crespi, J., Gulati, S., Salvesen, Ø., Bratbak, D.F., Dodick, D.W., Matharu, M.S. and Tronvik, E. (2022). Epidemiology of diagnosed cluster headache in Norway. *Cephalalgia Reports*, 5, 251581632210755. https://doi.org/10.1177/25158163221075569

Cuevas-Toro, A. M., Torrecillas, F. L., Medina, A. G., & Diaz-Bata-nero, M. C. (2008). Personality and coping strategies in fibromyalgia patients. *Behavioral Psychology, Revista Internacional ClínicaY de La Salud, 16,* 289-306.

Dekkers, O. M., Vandenbroucke, J. P., Cevallos, M., Renehan, A. G., Altman, D. G., & Egger, M. (2019). COSMOS-E: Guidance on conducting systematic reviews and meta-analyses of observational studies of etiology. *PLoS Medicine, 16*(2), e1002742. https://doi.org/10.1371/journal.pmed.1002742

Díaz-de-Terán, J., Sastre-Real, M., Lobato-Pérez, L., Navarro-Fernández, G., Elizagaray-García, I., & Gil-Martínez, A. (2021). Cluster headache, beyond the pain: a comparative cross-sectional study. *Neurological Sciences: Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology*, *42*(9), 3673-3680. https://doi.org/10.1007/s10072-020-04996-0

Donnet, A., Lanteri-Minet, M., Guegan-Massardier, E., Mick, G., Fabre, N., Géraud, G., Lucas, C., Navez, M., Valade, D., & Société Française d'Etude des Migraines et Céphalées (SFEMC) (2007). Chronic cluster headache: a French clinical descriptive study. *Journal of Neurology, Neurosurgery, and Psychiatry*, *78*(12), 1354-1358. https://doi.org/10.1136/jnnp.2006.112037

Elbadawi, A. S. A., Albalawi, A. F. A., Alghannami, A. K., Alsuhaymi, F. S., Alruwaili, A. M., Almaleki, F. A., Almutairi, M. F., Almubaddil, K. H., & Qashqari, M. I. (2021). Cluster headache and associated risk factors: A systemic review and meta-analysis. *Cureus, 13*(11), e19294. https://doi.org/10.7759/cureus.19294

Fischera, M., Marziniak, M., Gralow, I., & Evers, S. (2008). The incidence and prevalence of cluster headache: a meta-analysis of population-based studies. *Cephalalgia: an International Journal of Headache, 28*(6), 614-618. https://doi.org/10.1111/j.1468-2982.2008.01592.x

Fumal, A., & Schoenen, J. (2008). Tension-type headache: current research and clinical management. *The Lancet. Neurology, 7*(1), 70-83. https://doi.org/10.1016/S1474-4422(07)70325-3

Gambadauro, P., Carli, V., & Hadlaczky, G. (2019). Depressive symptoms among women with endometriosis: a systematic review and meta-analysis. *American Journal of Obstetrics and Gynecology*, *220*(3), 230-241. https://doi.org/10.1016/j.ajog.2018.11.123

Gatchel, R. J., Peng, Y. B., Peters, M. L., Fuchs, P. N., & Turk, D. C. (2007). The biopsychosocial approach to chronic pain: scientific advances and future directions. *Psychological Bulletin*, *133*(4), 581-624. https://doi.org/10.1037/0033-2909.133.4.581

Gesztelyi, G., & Bereczki, D. (2006). Determinants of disability in everyday activities differ in primary and cervicogenic headaches and in low back pain. *Psychiatry and Clinical Neurosciences*, *60*(3), 271-276. https://doi.org/10.1111/j.1440-1819.2006.01501.x

Gil-Martínez, A., Navarro-Fernández, G., Mangas-Guijarro, M. Á., & Díaz-de-Terán, J. (2019). Hyperalgesia and central sensitization signs in patients with cluster headache: a cross-sectional study. *Pain Medicine (Malden, Mass.)*, *20*(12), 2562-2570. https://doi.org/10.1093/pm/pnz070

Gómez-Mayordomo, V., Palacios-Ceña, M., Guerrero-Peral, Á., Fuensalida-Novo, S., Fernández-de-Las-Peñas, C., & Cuadrado, M. L. (2020). Widespread hypersensitivity to pressure pain in men with Cluster Headache during prolonged remission is not

related to the levels of depression and anxiety. *Pain Practice: the Official Journal of World Institute of Pain*, *20*(2), 147-153. https://doi.org/10.1111/papr.12839

Goulet-Pelletier, J.-C., & Cousineau, D. (2018). A review of effect sizes and their confidence intervals, Part 1: The Cohen's *d* family. *The Quantitative Methods for Psychology, 14*(4), 242-265. https://doi.org/10.20982/tqmp.14.4.p242

Grenard, J. L., Munjas, B. A., Adams, J. L., Suttorp, M., Maglione, M., McGlynn, E. A., & Gellad, W. F. (2011). Depression and medication adherence in the treatment of chronic diseases in the United States: a meta-analysis. *Journal of General Internal Medicine, 26*(10), 1175-1182. https://doi.org/10.1007/s11606-011-1704-y

Grinberg, A. S., Best, R. D., Min, K. M., Schindler, E. A. D., Koo, B. B., Sico, J. J., & Seng, E. K. (2021). Cluster Headache: Clinical Characteristics and Opportunities to Enhance Quality of Life. *Current Pain and Headache Reports*, *25*(10), 65. https://doi.org/10.1007/s11916-021-00979-8

Hak, T., Van Rhee, H. J., & Suurmond, R. (2016). How to interpret results of meta-analysis. (Version 1.3) Rotterdam, The Netherlands: Erasmus Rotterdam Institute of Management. www.erim.eur.nl/researchsupport/meta-essentials/downloads

Hamilton M. (1980). Rating depressive patients. *The Journal of Clinical Psychiatry, 41*(12 Pt 2), 21-24. https://pubmed.ncbi.nlm.nih.gov/7440521/

Hermesdorf, M., Berger, K., Baune, B. T., Wellmann, J., Ruscheweyh, R., & Wersching, H. (2016). Pain sensitivity in patients with major depression: differential effect of pain sensitivity measures, somatic cofactors, and disease characteristics. *The Journal of Pain, 17*(5), 606-616. https://doi.org/10.1016/j.jpain.2016.01.474

Higgins, J. P. T., Altman, D. G., Gøtzsche, P. C., Jüni, P., Moher, D., Oxman, A. D., Savovic, J., Schulz, K. F., Weeks, L., Sterne, J. A., Cochrane Bias Methods Group, & Cochrane Statistical Methods Group (2011). The Cochrane Collaboration's tool for assessing risk

of bias in randomised trials. *BMJ (Clinical research ed.)*, *343*, d5928. https://doi.org/10.1136/bmj.d5928

Higgins, J. P. T., Li, T., Deeks, J. J. (2023). Chapter 6: Choosing effect measures and computing estimates of effect. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). *Cochrane Handbook for Systematic Reviews of Interventions version 6.5. Cochrane, 2024.* Available from www.training.cochrane.org/handbook.

Higgins, J. P. T., Thomas, J., Chandler, J., Cumpston, M., Li, T., Page, M. J., Welch, V. A (editors). (2019). *Cochrane Handbook for Systematic Reviews of Interventions.* 2nd Edition. Chichester (UK): John Wiley & Sons.

Higgins, J. P. T., Thompson, S. G., & Spiegelhalter, D. J. (2009). A re-evaluation of random-effects meta-analysis. *Journal of the Royal Statistical Society. Series A, (Statistics in Society)*, *172*(1), 137-159. https://doi.org/10.1111/j.1467-985X.2008.00552.x

Higgins, J. P. T., & Thompson, S. G. (2002). Quantifying heterogeneity in a meta-analysis. Statistics in medicine, 21(11), 1539-1558. https://doi.org/10.1002/sim.1186

Horton, B. T., MacLean, A. R., & Craig, W. M. (1939). A new syndrome of vascular headache: results of treatment with histamine: preliminary report. *In Mayo Clinical Proceeding, 14,* 257-260).

Huedo-Medina, T. B., Sánchez-Meca, J., Marín-Martínez, F., & Botella, J. (2006). Assessing heterogeneity in meta-analysis: Q statistic or I2 index? *Psychological Methods*, *11*(2), 193-206. https://doi.org/10.1037/1082-989X.11.2.193

IntHout, J., Ioannidis, J. P. A., Rovers, M. M., & Goeman, J. J. (2016). Plea for routinely presenting prediction intervals in meta-analysis. *BMJ Open*, *6*(7), e010247. https://doi.org/10.1136/bmjopen-2015-010247

İşcan, D., & Dal, C. (2024). The relation of cluster headache to alexithymia, depression, and anxiety. La relación de la cefalea en racimos con la alexitimia, la depresión y la ansiedad. *Arquivos De Neuro-Psiquiatria*, *82*(11), 1-5. https://doi.org/10.1055/s-0044-1791516

Jang, B. N., Lee, H. J., Joo, J. H., Park, E. C., & Jang, S. I. (2020). Association between health behaviours and depression: findings from a national cross-sectional study in South Korea. *BMC Psychiatry, 20*(1), 238. https://doi.org/10.1186/s12888-020-02628-7

Ji Lee, M., Cho, S. J., Wook Park, J., Kyung Chu, M., Moon, H. S., Chung, P. W., Myun Chung, J., Sohn, J. H., Kim, B. K., Kim, B. S., Kim, S. K., Song, T. J., Choi, Y. J., Park, K. Y., Oh, K., Ahn, J. Y., Lee, K. S., Cho, S., & Chung, C. S. (2019). Increased suicidality in patients with CH. *Cephalalgia: an International Journal of Headache, 39*(10), 1249-1256. https://doi.org/10.1177/0333102419845660

Jorge, R. E., Leston, J. E., Arndt, S., & Robinson, R. G. (1999). Cluster headaches: association with anxiety disorders and memory deficits. *Neurology*, *53*(3), 543–547. https://doi.org/10.1212/wnl.53.3.543

Joshi, S., Rizzoli, P., & Loder, E. (2017). The comorbidity burden of patients with cluster headache: a population-based study. *The Journal of Headache and Pain*, *18*(1), 76. https://doi.org/10.1186/s10194-017-0785-3

Jürgens, T. P., Gaul, C., Lindwurm, A., Dresler, T., Paelecke-Habermann, Y., Schmidt-Wilcke, T., Lürding, R., Henkel, K., & Leinisch, E. (2011). Impairment in episodic and chronic cluster headache. *Cephalalgia: an International Journal of Headache*, *31*(6), 671–682. https://doi.org/10.1177/0333102410391489

Kalfas, M., Chisari, C., & Windgassen, S. (2022). Psychosocial factors associated with pain and health-related quality of life in Endometriosis: A systematic review. *European*

Journal of Pain (London, England), 26(9), 1827-1848. https://doi.org/10.1002/ejp.2006

Kessler, R. C., & Bromet, E. J. (2013). The epidemiology of depression across cultures. *Annual Review of Public Health*, *34*, 119-138. https://doi.org/10.1146/annurev-publhealth-031912-114409

Kim, B. S., Chung, P. W., Kim, B. K., Lee, M. J., Park, J. W., Chu, M. K., Ahn, J. Y., Bae, D. W., Song, T. J., Sohn, J. H., Oh, K., Kim, D., Kim, J. M., Kim, S. K., Choi, Y. J., Chung, J. M., Moon, H. S., Chung, C. S., Park, K. Y., & Cho, S. J. (2020). The impact of remission and coexisting migraine on anxiety and depression in cluster headache. *The Journal of Headache and Pain*, *21*(1), 58. https://doi.org/10.1186/s10194-020-01120-7

Kim, S. K., Chu, M. K., Kim, B. K., Chung, P. W., Moon, H. S., Lee, M. J., Choi, Y. J., Park, J. W., Kim, B. S., Song, T. J., Oh, K., Ahn, J. Y., Sohn, J. H., Lee, K. S., Park, K. Y., Chung, J. M., Chung, C. S., & Cho, S. J. (2024). An analysis of the determinants of the health-related quality of life in asian patients with cluster headaches during cluster periods using the time trade-off method. *Journal of Clinical Neurology (Seoul, Korea), 20*(1), 86-93. https://doi.org/10.3988/jcn.2022.0396

Koo, B. B., Bayoumi, A., Albanna, A., Abusuliman, M., Burrone, L., Sico, J. J., & Schindler, E. A. D. (2021). Demoralization predicts suicidality in patients with cluster headache. *The Journal of Headache and Pain*, *22*(1), 28. https://doi.org/10.1186/s10194-021-01241-7

Kroenke, K., Spitzer, R. L., & Williams, J. B. W. (2001). The PHQ-9: Validity of a brief depression severity measure. *Journal of General Internal Medicine, 16*(9), 606-613. https://doi.org/10.1046/j.1525-1497.2001.016009606.x2.

Landmark, L., Sunde, H. F., Fors, E. A., Kennair, L. E. O., Sayadian, A., Backelin, C., & Reme, S. E. (2024). Associations between pain intensity, psychosocial factors, and pain-related disability in 4285 patients with chronic pain. *Scientific Reports, 14*(1), 13477. https://doi.org/10.1038/s41598-024-64059-8

Lewinsohn, P. M. (1974). A behavioral approach to depression. In R. J. Friedman & M. M. Katz (Eds.), *The psychology of depression: Contemporary theory and research.* John Wiley & Sons.

Liang, J. F., Chen, Y. T., Fuh, J. L., Li, S. Y., Liu, C. J., Chen, T. J., Tang, C. H., & Wang, S. J. (2013). Cluster headache is associated with an increased risk of depression: a nationwide population-based cohort study. *Cephalalgia: an International Journal of Headache*, *33*(3), 182-189. https://doi.org/10.1177/0333102412469738

Lin, H. (2024). *Effect Size Converter.* MIT License. https://www.escal.site/

Lipsey, M. W., & Wilson, D. B. (2001). *Practical meta-analysis.* Sage Publications, Inc.

Long, R. J., Zhu, Y. S., & Wang, A. P. (2021). Cluster headache due to structural lesions: A systematic review of published cases. *World Journal of Clinical Cases, 9*(14), 3294-3307. https://doi.org/10.12998/wjcc.v9.i14.3294

Louter, M. A., Wilbrink, L. A., Haan, J., van Zwet, E. W., van Oosterhout, W. P., Zitman, F. G., Ferrari, M. D., & Terwindt, G. M. (2016). Cluster headache and depression. *Neurology*, *87*(18), 1899-1906. https://doi.org/10.1212/WNL.0000000000003282

Lund, N. L. T., Petersen, A. S., Fronczek, R., Tfelt-Hansen, J., Belin, A. C., Meisingset, T., Tronvik, E., Steinberg, A., Gaul, C., & Jensen, R. H. (2023). Current treatment options for cluster headache: limitations and the unmet need for better and specific treatments-a consensus article. *The Journal of Headache and Pain, 24*(1), 121. https://doi.org/10.1186/s10194-023-01660-8

Ma, L. L., Wang, Y. Y., Yang, Z. H., Huang, D., Weng, H., & Zeng, X. T. (2020). Methodological quality (risk of bias) assessment tools for primary and secondary medical studies: what are they and which is better? *Military Medical Research*, *7*(1), 7. https://doi.org/10.1186/s40779-020-00238-8

Markley, H. G., & Buse, D. C. (2006). Cluster headache: myths and the evidence. *Current Pain and Headache Reports*, *10*(2), 137-141. https://doi.org/10.1007/s11916-006-0025-z

Mazzucchelli, T. G., Kane, R. T., & Rees, C. S. (2010). Behavioral activation interventions for well-being: A meta-analysis. *The Journal of Positive Psychology, 5*(2), 105-121. https://doi.org/10.1080/17439760903569154

Mitsikostas, D., & Thomas, A. (1999). Comorbidity of headache and depressive disorders. *Cephalalgia*, *19*(4), 211-217. https://doi.org/10.1046/j.1468-2982.1999.019004211.x

Morgan, R. L., Whaley, P., Thayer, K. A., & Schünemann, H. J. (2018). Identifying the PECO: A framework for formulating good questions to explore the association of environmental and other exposures with health outcomes. *Environment International*, *121*(Pt 1), 1027-1031. https://doi.org/10.1016/j.envint.2018.07.015

Muka, T., Glisic, M., Milic, J., Verhoog, S., Bohlius, J., Bramer, W., Chowdhury, R., & Franco, O. H. (2020). A 24-step guide on how to design, conduct, and successfully publish a systematic review and meta-analysis in medical research. *European Journal of Epidemiology, 35*(1), 49-60. https://doi.org/10.1007/s10654-019-00576-5

Nesbitt, A. D., & Goadsby, P. J. (2012). Cluster Headache. *BMJ, 344,* e2407. https://doi.org/10.1136/bmj.e2407

NICE. (2022). *Cluster Headache.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/headache-cluster/management/management/

NICE. (2022). *Headache-Tension Type.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/headache-tension-type/

NICE. (2024). *Depression.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/depression/

NICE. (2022). *Depression in Adults: Treatment and Management.* The National Institute for Health and Care Excellence. https://www.nice.org.uk/guidance/ng222/chapter/recommendations#treatment-manuals

NICE. (2024). *Migraines.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/migraine/management/adults/

Osman, A., Bagge, C. L., Gutierrez, P. M., Konick, L. C., Kopper, B. A., & Barrios, F. X. (2001). The Suicidal Behaviors Questionnaire-Revised (SBQ-R): validation with clinical and nonclinical samples. Assessment, 8(4), 443–454. https://doi.org/10.1177/107319110100800409 OUCH. (2024). *Cluster Headache.* https://ouchuk.org/

Page, M. J., McKenzie, J. E., Bossuyt, P. M., Boutron, I., Hoffmann, T. C., Mulrow, C. D., Shamseer, L., Tetzlaff, J. M., & Moher, D. (2021). Updating guidance for reporting systematic reviews: development of the PRISMA 2020 statement. *Journal of Clinical Epidemiology, 134,* 103-112. https://doi.org/10.1016/j.jclinepi.2021.02.003

Paul, M., & Leeflang, M. M. (2021). Reporting of systematic reviews and meta-analysis of observational studies. *Clinical Microbiology and Infection: the Official Publication of the European Society of Clinical Microbiology and Infectious Diseases, 27*(3), 311-314. https://doi.org/10.1016/j.cmi.2020.11.006

Pescador, R, M. A., & De Jesus, O. (2024). *Migraine Headache.* StatPearls Publishing.

Pohl, H., Gantenbein, A. R., Sandor, P. S., Schoenen, J., & Andrée, C. (2020). Interictal burden of cluster headache: results of the EUROLIGHT cluster headache project, an

internet-based, cross-sectional study of people with cluster headache. *Headache*, *60*(2), 360-369. https://doi.org/10.1111/head.13711

Qin, L., Song, P., Li, X. *et al.* Tension-Type Headache Management: A Systematic Review and Network Meta-analysis of Complementary and Alternative Medicine. *Pain Therapy 13*, 691-717 (2024). https://doi.org/10.1007/s40122-024-00600-x

R Core Team. (2019). *R: a language and environment for statistical computing.* Vienna R Foundation for Statistical Computing. https://www.R-project.org.

Robbins, M. S., Starling, A. J., Pringsheim, T. M., Becker, W. J., & Schwedt, T. J. (2016). Treatment of Cluster Headache: The American Headache Society Evidence-Based Guidelines. *Headache*, *56*(7), 1093-1106. https://doi.org/10.1111/head.12866

Rossi, P., Little, P., De La Torre, E. R., & Palmaro, A. (2018). If you want to understand what it really means to live with CH, imagine... fostering empathy through European patients' own stories of their experiences. *Functional Neurology, 33*(1), 57-59. https://doi.org/10.11138/fneur/2018.33.1.057

Roughan, W. H., Campos, A. I., García-Marín, L. M., Cuéllar-Partida, G., Lupton, M. K., Hickie, I. B., Medland, S. E., Wray, N. R., Byrne, E. M., Ngo, T. T., Martin, N. G., & Rentería, M. E. (2021). Comorbid chronic pain and depression: Shared risk factors and differential antidepressant effectiveness. *Frontiers in Psychiatry, 12,* 643609. https://doi.org/10.3389/fpsyt.2021.643609

Rozen, T. D., & Fishman, R. S. (2012). Cluster headache in the United States of America: demographics, clinical characteristics, triggers, suicidality, and personal burden. *Headache, 52*(1), 99-113. https://doi.org/10.1111/j.1526-4610.2011.02028.x

Sadeghi, M. (2025). Public bias and funnel plot. In M, Ghajarzadeh,., J, Hopkins., N, Rezaei, S, Hanaei, S. (Eds). Systematic Review and Meta-Analysis. (1st edition). Elsevier Inc

Schünemann, H. J., Vist, G. E., Higgins, J. P. T., Santesso, N., Deeks, J. J., Glasziou, P., Akl, E. A., Guyatt, G. H. (2023). Chapter 15: Interpreting results and drawing conclusions. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). *Cochrane Handbook for Systematic Reviews of Interventions version 6.5.* Cochrane, 2024. Available from www.training.cochrane.org/handbook.

Shah, N., Asuncion, R. M. D., & Hameed, S. (2024). *Muscle Contraction Tension Headache.* In StatPearls. StatPearls Publishing.

Sheng, J., Liu, S., Wang, Y., Cui, R., & Zhang, X. (2017). The Link between Depression and Chronic Pain: Neural Mechanisms in the Brain. Neural plasticity, 2017, 9724371. https://doi.org/10.1155/2017/9724371

Søborg, M. K., Jensen, R. H., Barloese, M., & Petersen, A. S. (2024). Biomarkers in cluster headache: A systematic review. *Headache*, *64*(1), 98-116. https://doi.org/10.1111/head.14641

Stoll, C. R. T., Izadi, S., Fowler, S., Green, P., Suls, J., & Colditz, G. A. (2019). The value> of a second reviewer for study selection in systematic reviews. *Research Synthesis Methods*, *10*(4), 539-545. https://doi.org/10.1002/jrsm.1369

Torkamani, M., Ernst, L., Cheung, L. S., Lambru, G., Matharu, M., & Jahanshahi, M. (2015). The neuropsychology of cluster headache: cognition, mood, disability, and quality of life of patients with chronic and episodic cluster headache. *Headache*, *55*(2), 287-300. https://doi.org/10.1111/head.12486

Waung, M. W., Taylor, A., Qualmann, K. J., & Burish, M. J. (2020). Family History of Cluster Headache: A Systematic Review. *JAMA neurology*, *77*(7), 887-896. https://doi.org/10.1001/jamaneurol.2020.0682

Wei, D. Y., & Goadsby, P. J. (2021). Cluster headache pathophysiology - insights from current and emerging treatments. *Nature Reviews. Neurology, 17*(5), 308-324. https://doi.org/10.1038/s41582-021-00477-w

Wells, G.A., Wells, G., Shea, B., Shea, B., O'Connell, D., Peterson, J., Welch, Losos, M., Tugwell, P., Ga, S.W., Zello, G.A., & Petersen, J.A. (2014). *The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomised studies in meta-analyses.* Ottawa Hospital Research Institute https://www.ohri.ca/programs/clinical_epidemiology/oxford.asp

World Health Organization, WHO. (2021). *Depression.* World Health Organization. https://www.who.int/news-room/fact-sheets/detail/depression

Zigmond, A. S., & Snaith, R. P. (1983). Hospital Anxiety and Depression Scale (HADS) [Database record]. *APA PsycTests.* https://doi.org/10.1037/t03589-000

Chapter Three: Bridging Chapter

The systematic review, presented in Chapter Two, indicates that the condition, Cluster Headache (CH), is related to depressive symptoms and suicidality. As well as depression and suicidality, some research has suggested CH is associated with other psychopathologies, such as anxiety (Grinberg et al., 2021). Currently, NICE guidelines for CH treatment (2022) are all pharmaceutical. Review findings from Chapter Two indicate that perhaps there should be guidelines regarding psychological interventions to support individuals with CH. There is very little understanding of the psychological processes related to the CH experience and the relationship with psychopathology. To inform treatment guidelines for CH, further understanding of the psychological processes involved is required.

Literature which currently exists regarding psychopathology and CH, hypothesises reasons for the relationship. For example, a review proposed that loss of independence and social connection due to pain could result in depressive symptoms, acute pain during an attack could result in suicidality, and anticipation of future attacks could result in anxiety (Grinberg et al., 2021). However, to our knowledge, no research has directly explored the psychological processes. This is important, because psychopathology can be treated via different processes of change based on what factors are found to maintain and perpetuate distress. For example, for depression, various therapies are recommended and effective for different individuals (O'Driscoll et al., 2023). Traditional Cognitive Behavioural Therapy (CBT) brings about change through changing thoughts and maladaptive behaviours (Beck et al., 1979), Acceptance Commitment Therapy (ACT) brings about change through lessening the impact that one's thoughts and emotions have on them (Hayes & Pierson, 2005), counselling brings about change by altering individuals' emotional meaning of experiences (Roth et al., 2009), and Interpersonal Psychotherapy (IPT) helps to manage psychopathology through improving relationships with others (Cuijpers et al., 2011). To understand which

therapies may be effective for CH individuals, it is important to understand the psychological aspects of the CH experience and what maintains distress e.g., particular cognitions, behaviours, interpersonal relationships. Chapter Four will take a qualitative exploratory approach to understand the psychological processes involved in living with CH.

Chapter Four: "If We Had Blood Pouring Out of Our Eyeballs, People Would Notice"; a Qualitative Exploration into the Psychological Experience of Cluster Headache

Submitted to the International Journal of Clinical Practice (come back with revisions)

Word Count: 8,621

Miss Helena Whitley (She/Her), University of East Anglia, Trainee Clinical Psychologist

Mr Wayne Nolan, Patient and Public Involvement Representative

Dr Fergus Gracey, University of East Anglia

Dr Elisabeth Norton, University of East Anglia

Abstract

Background

The primary headache disorder, Cluster Headache (CH), is known as one of the most painful conditions a human can experience. This study aimed to explore the psychological experience of living with CH, both during acute headache attacks and between attacks.

Methods

Semi-structured interviews were conducted with 13 adults with episodic or chronic CH. Interviews were carried out via Microsoft Teams or the telephone. Qualitative interview data was analysed using Reflective Thematic Analysis (RTA) through a critical realist lens. Participants also completed the Pain Self Efficacy Questionnaire and Pain Catastrophising Scale.

Results

RTA resulted in five themes emerging related to the psychological experience of CH: "Darkness", "Battling", "Shifting", "Control", and "Despair". Psychological experiences during the acute attack appeared distinct from experiences of the condition between attacks. Whether individuals had chronic or episodic CH, and how long they had lived with the condition, influenced the psychological experience.

Conclusion

Living with CH is a complex and challenging, psychological experience. Further research to identify how psychological processes may maintain distress and exacerbate pain could help establish a psychological model of CH. Such a model could, in turn, enable exploration of psychological techniques to support individuals during and between CH attacks. Authors also call for research to explore how to increase visibility of CH.

Introduction

Cluster Headache (CH) is cited as one of the most painful conditions known to humankind (Burish et al., 2021). The condition involves painful 'attacks' which last between five and 180 minutes and then pain-free periods (IHS, 2018). In the more common, episodic form of CH (ECH), individuals experience a bout of attacks and then a period of remission lasting at least three months (Ahmed et al., 2019; IHS, 2018). The less common, chronic form (CCH) involves no significant period of remission. CH has been associated with suicidality, reduced quality of life, depression, and anxiety (D'Amico et al., 2020; Freeman et al., 2022; Ji Lee et al., 2019; Rossi et al., 2018; Wei & Goadsby, 2021). Despite these associated psychological aspects, National Institute for Health and Care Excellence (NICE) guidelines for treatment in the United Kingdom (UK) are all related to medical interventions, and individuals with CH are rarely offered mental health support (Grinberg et al., 2021; NICE, 2022).

Lack of treatment guidelines for CH focused on psychological approaches likely stems from a lack of related research; as NICE guidelines only advise when there is a strong evidence base (NICE, 2024). Researchers have hypothesised the potential benefits of Cognitive Behavioural Therapy (CBT), Acceptance and Commitment Therapy (ACT), and relaxation, for managing CH (Grinberg et al., 2021). However, limited research has empirically explored such hypotheses (Grinberg et al., 2021). The few studies conducted examined progressive muscle relaxation, bio-feedback, and cognitive behavioural stress management (Benson et al., 1974; Blanchard et al., 1982; Grinberg et al., 2021; Jensen et al., 2010; Sargent et al., 1973). However, these studies were uncontrolled, had a small sample of CH participants, and did not distinguish which aspect of treatment had an effect, if one was reported (Grinberg et al., 2021; Schenck & Andrasik, 2019). Two previous qualitative interview studies in Spain (Palacios-Ceña et al., 2016) and the UK (Andre & Cavers, 2021)

have been conducted related to CH, focusing on the general experiences of living with the condition. Authors used phenomenological (Palacios-Ceña et al., 2016) and thematic (Andre & Cavers, 2021) analysis to highlight that CH sufferers felt misunderstood and dominated by the headache, dreaded future attacks, were dissatisfied with pharmacological treatments, and struggled to control the pain by any means.

The present study aimed to build on prior qualitative research by focusing on the psychological experience of CH i.e., the cognitive, affective, and behavioural aspects. Whilst no research exists related to these factors regarding CH, a large body of literature demonstrates pain is influenced by psychological factors (Day et al., 2018; Gorczyca et al., 2013; Klonowski et al., 2022; Sánchez-Rodríguez, 2020; Schenck & Andrasik, 2019). For example, self-efficacy is an individual's perceived ability to cope with their pain and low self-efficacy has been related to worse pain management (Bandura, 1977; Jensen et al., 2018). Similarly, pain catastrophising is an exaggerated and negative belief about one's pain experience and has been related to poorer adjustment (Quartana et al., 2009). Behaviorally, both over and under exertion are associated with higher levels of pain (Hasenbring et al., 2009). Finally, in terms of affect, anxiety and depression have been shown to increase pain in a bi-directional manner (Wei et al., 2016). These psychological facets, whilst not studied in CH, have been explored in Migraine and Tension-Type Headache (TTH) individuals, and low self-efficacy, pain catastrophising, activity avoidance, and anxiety, have been related to longer and worse head pain (González et al., 2022; Hee et al., 2024; Yu & Tan, 2024).

The present study aimed to explore the psychological factors related specifically to CH. The study explored factors present both during the acute attack, and in the period between attacks, as these phases are markedly different (Fischera et al., 2008). Authors hoped that the knowledge gathered from this research would be a step towards understanding which psychological treatments, if any, are best placed to support the CH community.

Research Question:

1. What psychological aspects are important in the experience of an acute CH attack?
2. What psychological aspects are important in the experience of CH between attacks?

Methods

The study was reported in accordance with the Consolidated Criteria for Reporting Qualitative Studies (COREQ) 32-item checklist (Tong et al., 2007; See Appendix E).

Design

The primary aim was to achieve a greater understanding of the unique human experience of CH. A largely qualitative approach was adopted, allowing exploratory enquiry of an area of new interest (Agius, 2013; Lim, 2024). The study involved semi-structured interviews to collect the qualitative data and Reflexive Thematic Analysis (RTA) to analyse this data. A critical realist lens was applied throughout RTA, meaning the researchers assumed there is an objective truth; however, knowledge is socially constructed and context-bound (Schiller, 2016). This ontological stance meant that the researchers were reflective that their own beliefs and experiences influenced their understanding of the research findings (Koopmans & Schiller, 2022). Furthermore, the researchers were aware that each participant's context influenced their experience, meaning the researchers were seeking complex and nuanced subjective variations of an objective truth. A critical realist lens was helpful for this study, as this perspective values and acknowledges both the biological reality of the CH condition, and the subjective psychological experience of individual sufferers.

Participants also completed two questionnaires measuring self-efficacy and pain catastrophising. These concepts were measured as they have been associated with health-related disability (Bandura, 1977; Jensen et al., 2018; Quartana et al., 2009). Initially authors planned to analyse this quantitative data alongside the interview transcripts; for example, comparing qualitative experiences to quantitative scores. However, once the interviews had

been conducted, researchers reflected that integrating the rich qualitative data with the questionnaire data would diminish the power of the interviewees' accounts. Therefore, it was decided that questionnaire data would instead simply help contextualise the participants.

Patient and Public Involvement (PPI)

This research topic was proposed by an individual who suffered from CCH and had no contact with mental health professionals throughout their five-year NHS treatment journey, despite feeling this was essential. NHS planning documents highlight that involving PPI representatives in research, empowers patients and promotes improved care (Care Quality Commission, 2018; NHS England, 2023; NIHR, 2015). Service user involvement felt particularly important for this study, as CH sufferers report feeling misunderstood by those who do not experience the condition (Andre & Cavers, 2020). The PPI representative (WN) attended every research meeting and was involved in decisions regarding study methods, and data analysis. Whilst the PPI representative's opinions were considered crucial, the final decision resided with the traditional research team. The PPI level of involvement was therefore considered as "contribution" (Sweeney & Morgan, 2009). When reporting on PPI involvement, GRIPP 2 short form guidelines were followed (Staniszewska et al., 2017); See Appendix F.

Participants

To be included, individuals had to be over 18 years of age, living in the UK, English speaking, and diagnosed with ECH or CCH by a healthcare professional. Participants had to provide their GP surgery contact details, so that the researchers could follow up regarding risk concerns. Individuals were excluded if they lived with comorbid facial or head pain.

Participants were recruited using opportunity sampling. The study was advertised on the webpage of the charity Organisation for the Understanding of Cluster Headache (OUCH). OUCH is a UK charity advocating for CH individuals (OUCH, 2024). Interested individuals

were asked to contact the primary researcher (HW) using the contact details on the advertisement. The desired sample size was 15 participants, which was in line with relevant literature (Andre & Cavers, 2021; Palacio-Cena et al., 2016). A first-come-first served approach was taken, with those who expressed interest first being screened. After 15 participants were selected, the OUCH advertisement was removed. If individuals emailed after the required sample had been reached, they were informed by email that recruitment had finished. See Appendix G for the study advertisement.

Materials

An interview schedule was produced by the research team, which included the PPI representative (WN) and a clinical psychologist who previously worked in pain management services (EN). Feedback was also received from OUCH. See full details of this process in the Additional Methods Chapter (Chapter Five) and Appendix H for the interview schedule.

Participants also completed the 13-item Pain Catastrophising Scale (PCS: Sullivan et al., 1995) and the Pain Self-Efficacy Questionnaire (PSEQ; Nicholas, 2007) (See Appendix I). These scales are psychometrically sound (British Pain Society, 2019). For the PCS, high scores (> 30) indicate clinically significant levels of catastrophising (Sullivan et al., 1995), and for the PSEQ, low scores (<20) indicate low self-efficacy (Tonkin, 2008).

Procedure

Individuals who contacted the primary researcher from the OUCH advertisement were emailed a study information sheet and initial consent form (See Appendix J). If still interested, individuals were asked to email the researcher a completed initial consent form which gave permission for the researcher to telephone them. The researcher telephoned the consenting participants to discuss the research, determine suitability, and collect demographic information. If eligible, participants were then given a final consent form to complete (See Appendix K) and interviews were organised.

Participants were given a choice of how interviews could be conducted: Microsoft Teams video, telephone, or in person. Semi-structured interviews were conducted solely by the primary researcher (HW). After interviews, the researcher emailed over the questionnaires. After questionnaires were returned, the interviewer emailed a debrief form (See Appendix L) and a £10 Love2Shop voucher.

The audio recordings of the interviews were automatically transcribed by Microsoft Teams. The primary researcher (HW) reviewed and edited transcripts for accuracy to ensure verbatim accounts. Participants were sent a copy of their transcript, and they were given two weeks to edit/redact/withdraw consent. One participant edited their transcript, and this version was included instead of the original.

Data Analysis

RTA was used to analyse interview data, following the six-step approach created by Braun and Clarke (2013; 2019; 2020; 2022). A critical realist perspective was embraced for this analysis (Bhaskar, 1998, 2008). The data was analysed inductively to allow the data to catalyse discovery. The primary researcher analysed all of the data. As researcher bias is inevitable (Braun and Clarke, 2013), reflexivity was employed throughout to foster a transparent and self-aware analysis, and all decisions were made collaboratively with the wider research team. The PPI representative's voice (WN) was given power throughout this process, with the researchers also being conscious not to allow WN's personal CH experience to override other participants' voices. The Eight Big Tent Criteria (Tracy, 2010) was used when assessing this piece of research: (1) worthy topic, (2) rich rigor, (3) sincerity, (4) credibility, (5) resonance, (6) significant contribution, (7) ethics, and (8) meaningful coherence.

Ethics

This study was approved by the Faculty of Medicine and Health Sciences Ethics Committee at the University of East Anglia, after one set of amendments (ID: ETH2324-0070, see Appendix M). All participants provided written informed consent prior to participation. Participants were given the opportunity to withdraw their consent after reading their transcript. Confidentiality of data was assured through removal of any identifiable information. See full ethical considerations in Chapter Five.

Results

Participants

There was significant interest in the study, with over 100 individuals responding to the advertisement. The first responding 15 participants were contacted, and 13 completed consent forms and so had interviews arranged. After the 13 interviews, it was determined that the data contained sufficient information power, with the interviews having strong quality of dialogue, and the participants'characteristics being highly specific to the study aim (Malterud et al., 2016). Therefore, no further interviewees were sought.

Interviews took place by telephone (N= 8) or Microsoft Teams (N= 5). The mean length of the interview was 68 minutes, all occurred in one sitting, and only the interviewer and participant were present. Of the 13 included individuals, 69% were male, and nine had ECH and four CCH. The age of participants ranged from between 33 and 76 years old. Length of time living with CH ranged from 8 to 40 years.

Questionnaires

11 out of the 13 participants completed the two questionnaires. 64% of participants scored above the cut-off (> 30) for clinically significant pain catastrophising (mean score of 30.70). 72% of participants scored below the threshold (<20) for low self-efficacy (mean

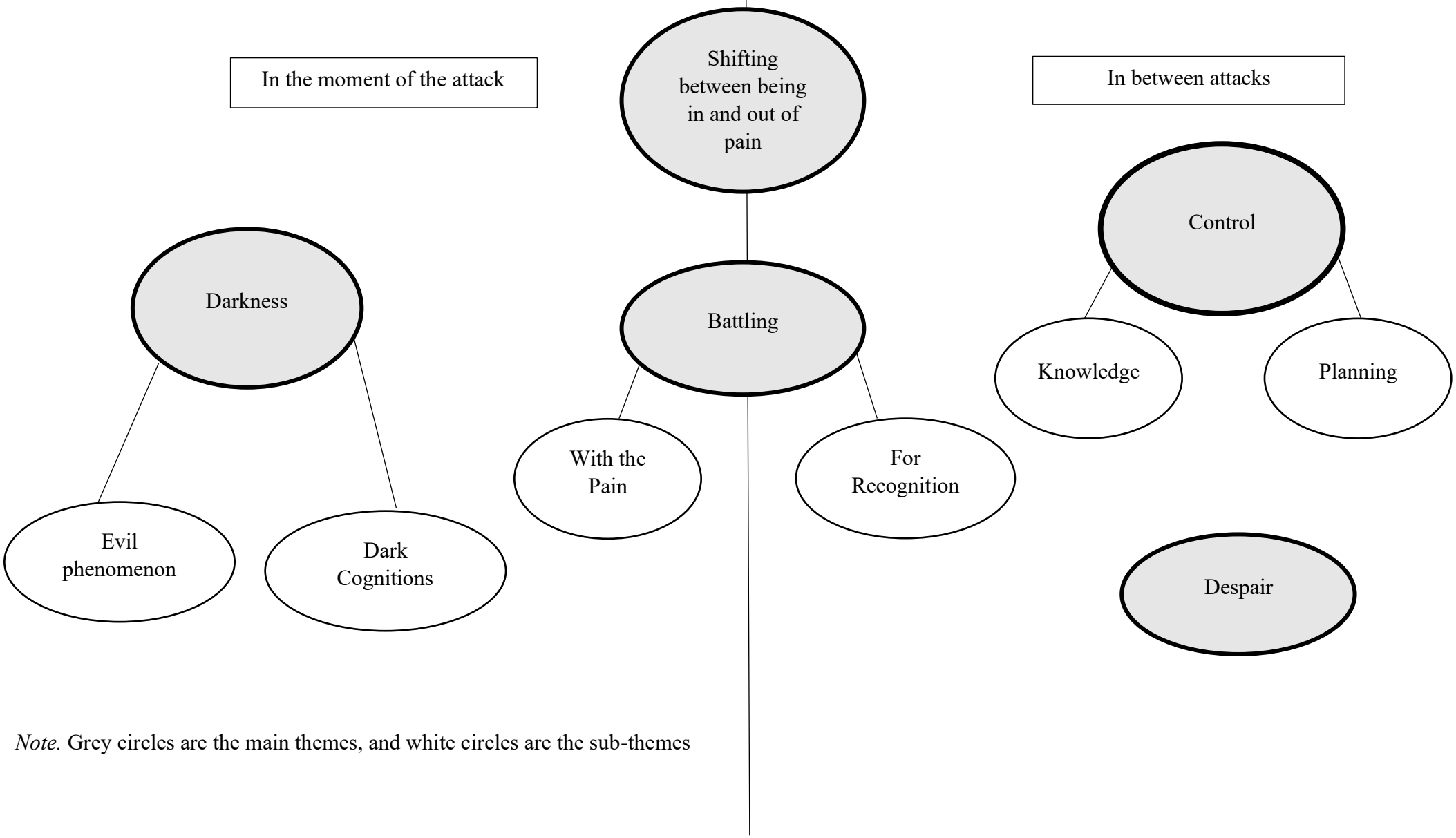
score of 15), indicating the majority of participants had low self-belief in managing pain. See Appendix N for individual participant scores.

Reflexive Thematic Analysis

Five themes were identified in relation to psychological aspects of CH, 1) Darkness, 2) Battling, 3) Shifting, 4) Control, and 5) Despair. The themes were separated into 1) when participants were in the moment of the attack of pain, and 2) in-between attacks, as these stages felt distinct. Theme One (Darkness) related to the experience of the attack of pain and comprised of two sub-themes; 1) the horror-like quality of the painful attack, and 2) participants negative thoughts throughout the attack, including suicidal ideation and self-blaming thoughts. Theme Two (Battling) was also comprised of two sub-themes: 1) fighting against the pain in the moment of the attack, for example through pacing and hitting one's head, and 2) fighting for visibility and acknowledgement for the condition outside of the attack; both from healthcare professionals and the public. Theme Three (Shifting) related to the dramatic 'shift' between being in and out of an acute painful attack and between being in and out of a bout period for episodic sufferers. Theme Four and Five related to experiences between attacks. Theme Four (Control) was separated into the sub-themes 'Knowledge' and 'Planning' and referred to participants gaining a sense of control over their condition through receiving a diagnosis, understanding the science behind the pain, and planning their daily life to account for the attacks. Finally, Theme Five (Despair) related to the sense of hopelessness, depression, and exhaustion which was evoked by participants' accounts of living with CH. This was particularly true for CCH individuals. See the Thematic Analysis map in Figure 8 for themes and sub-themes.

Figure 8

Thematic Analysis Map



Note. Grey circles are the main themes, and white circles are the sub-themes

Each of the themes are detailed below alongside quotes. Removal of text is denoted by […], (text) explains context of quotes. Next to each quote is the participant's number, gender, CH sub-type, and age.

Theme One: Darkness: In the Moment of the Attack

Evil Phenomenon.

The theme "Darkness" relates to an almost other-worldly, evil, horror quality which the researchers felt was evoked from many participants' accounts of when they were in a CH attack. Participants often struggled to articulate their experience, finding it difficult to put into words something so different from their usual, pain-free, state.

> You can't even describe the pain when you're not having it [...] I think of it now and again, as the monster. I also think of it as every foul word you can think [...] when it's there, I want it to go away and it's horrible and evil. (P7, M, Chronic).

Participants spoke about their CH attacks as being a separate entity with ill intentions against them. It could be that CH sufferers externalised their pain in this way to separate the 'evil' entity from themselves, so reducing self-blame and maintaining a coherent sense of self. It is interesting to consider whether the dark interpretation of CH relates to pain catastrophising, which was raised in 64% of study participants' results.

> It's very dark as well. You just think. It's it's like. It's er I don't wanna. This sounds very odd. It's like. It's like. It's out to get ya. Like a really. Like it's coming for ya […] You'd go through your. You know that expression, "the dark night of the soul?" (P12, M, Episodic).

The hesitancy in the way P12 spoke perhaps indicates awkwardness with sharing their experience, or even fear, due to the otherworldly, strange quality.

Several participants also spoke about their experience in spiritual terms, referring to God and punishment.

> This has gone into the family vocabulary now, so now a Cluster Headache is an attack of being smited by God. (P13, F, Episodic).

CH attacks often occur at night, which may have contributed to the dark and other-worldly associations. The research team reflected that this theme felt very lonely and was the most powerful theme.

Dark Cognitions.

Within the theme of "Darkness" was also the sub-theme "Dark Cognitions" within the moment of the attack. Individuals reported negative, blaming, thoughts about themselves. Again, note the spiritual lens.

> My mind goes to some quite dark places. So like during the cluster headaches you, you end up thinking like all kinds of ridiculous things. Like you know, "is this my fault? Is this happening? Is this being sent by God to make me a more understanding (healthcare professional)? (P4, F, Episodic)

Some participants spoke about suicidal thoughts throughout the attack to stop the pain.

> If there was a pill that would kill me, I would take it […] because I can't bear it. (P10, F, Chronic).

Given CH is nick-named "the suicide headache", these experiences are perhaps unsurprising. For all individuals who spoke about suicidal ideation, this was isolated to the moment of the attack and related to stopping the pain. For example, P10 clearly stated "I do not wish to die, I have a lovely life". This process of cognitions shifting when in and out of attacks was a common experience; with individuals sharing that it was hard to think logically when in extreme pain.

> In that intensity of pain […] It's it's a different state of being in terms of how you think. You literally have such a thin layer of consciousness […] it's almost like you're drowning. (P5, M, Episodic).

Theme Two: Battling

Battling with the Pain: In the Moment of the Attack.

This theme relates to how individuals cope with the "Darkness" during attacks. Some participants described fighting against the pain throughout an attack, through pacing, throwing objects, and hitting their head. This is an experience previously reported on, with such behaviours being used as a diagnostic marker to differentiate CH from other primary headache disorders.

> I like turn into a different person. It is. Quite frightening because. I can't talk. You know. I I get very, very frustrated. Extremely agitated [...] I just wanna. Hit my head against the wall sometimes. (P10, F, Chronic).

> If you try and read while you're having it (an attack), you end up throwing the book across the room [...] I've punched erm a bedpost. (P4, F, Episodic).

Whilst battling with the pain was a common experience, several interviewees reported changing their psychological response over time, moving from fighting the pain to "grinning and bearing it" (P11, M, Episodic & P6, M, Episodic). Several mentioned they have mantras they repeat to themselves. P6, who had lived with CH for several decades , said:

> You don't get complacent to it all, but you kind of looking and think I've had a million of these now I can deal with these. (P6, M, Episodic).

These changes from battling against the pain towards trying to be with the pain, may be indicative of sufferers coming to a place of acceptance for their condition.

Several participants took this further, speaking about trying to relax throughout an attack and breathing deeply. They reported realising, over years, that becoming agitated was likely "feeding" (P12, M, Episodic) the pain. Several theorised that increased heart rate and becoming tense couldn't be helping.

> I try to stay calm […] probably a decade or so ago […] I just kind of concluded that like, you know, kind pacing and getting agitated and stuff, you know, kind of surely that can't be helping. And I used to. I of used to like hit back and I hit my head. (P2, M, Episodic).

P5 expanded further on this, describing how they tried to separate from the pain and in so doing tried to ride the attack out:

> In an attack I've I've worked myself into position where I can distinguish the two. I can feel the pain in my head, right? But […] I am not the pain […] For the longest time, I was the pain and I would be on the floor. I would be banging my head against the wall trying to relieve it […] But in my latest attack […] I could try and dissociate from the physical pain. (P5, M, Episodic).

P13 also spoke about a different approach to battling the pain. They used humour when referring to the pain to make it easier to cope with. This humour involved referring to the condition in dramatic terms.

> Making something of a joke of it and some of my friends will use it. You are not going to let it beat you. It's like erm people making fun of Hitler during the war, takes the horror out of it out. (P13, F, Episodic).

Battling for Recognition: In-Between Attacks.

Outside of the moment of pain, participants described a battle related to seeking recognition and support for the condition.

> You have to do a lot of battling at a time when you're not in the mood to be battling. (P3, M, Episodic).

Many said the battle for support was due to lack of awareness and understanding of the condition; particularly from GPs. Furthermore, participants felt that the name "Cluster Headache" undermined and diminished the severity of the condition, with the public likening CH to other headaches or Migraine. This experience was often accompanied by emotions of embarrassment and frustration.

> I hate the word cluster headache. I just hate it. I hate the phrase. I hate saying it. Because it undermines what it is [...] It makes you feel. Stupid because (chuckles) you know. People just think "oh she's always off with a headache". (P10, F, Chronic).

Several participants felt that the lack of physical symptoms meant the public could not understand the level of distress.

> It's the visual representation. If you know, if we had blood pouring out of our eyeballs, people would notice. (P2, M, Episodic).

Again, this quote uses dark, horror-like language to describe the experience, linking it to the "Darkness" theme. Such evocative language to describe the CH experience could be used as a vehicle to share the invisible experience of pain, so others can try to understand and empathise.

The researchers reflected that the sense of battling for recognition was also evident by the project being proposed by an individual with CCH and through the huge response to the study advertisement. There was a sense of almost desperation to have ones' experiences heard.

Interestingly, P4 had a different experience, feeling their experience was always understood and validated. They were a medical professional and said that because CH is known as a very painful condition in the medical world, they had never been doubted. This participant also expressed that the fact CH is more common in males, means that the severity of the pain is believed. This is an example of how one's context, in this case, perceived patriarchal hierarchy, can alter experience.

> I'm quite lucky. Because uh, it's a condition that mainly affects men [...] So all the medical textbooks say [...]"it's the most painful condition known to man" [...] Whereas medical conditions that mainly affect female the medical textbooks traditionally say things like "this is probably all psychological, and there's no point in trying to fix it" [...] So everybody's very sympathetic. (P4, F, Episodic).

However, P4, went on to share that there is a lack of attention from a mental health lens; something echoed by other participants. P4 explained that the shared understanding that CH is "the suicide headache" means that when people voice this, it doesn't evoke the appropriate response of support.

> If you say to a (healthcare professional) "my COPD is so bad I wish I was dead" […] it's a prompt for everybody to be like "oh my gosh, do we need to do a mental health services" […] erm but if you say "my cluster headaches are so bad I wish I was dead" everyone's like "oh yeah classic textbook presentation" and it doesn't, and I feel like it just stays as like a diagnosis thing. (P4, F, Episodic).

Theme Three: Shifting

This theme relates to the jarring experience of shifting from being in and out of pain. Participants described how, with no warning, they could go from being completely pain-free, to being in incredible pain. Individuals reported that this made planning difficult, resulting in anxiety about upcoming events. Furthermore, several participants commented that the sudden, dramatic onset, meant that their pain was often not believed.

> I can be absolutely fine one minute. Talking to you normally and then suddenly bang I'll be in absolute agony. Which is really difficult because erm people seem to think you're making it up. (P10, F, Chronic).

The Shifting theme also relates to episodic sufferers' experience of being in and out of a bout season. Participants described almost a change in who they were as a person and their identity; a healthy 'normal' person, to someone incapacitated by illness.

> Most of the the year. It's like that plain, clear skies. Beautiful, right? […]. But then during my cluster season, it's like you hear thunder, [...] you see the clouds forming and and then the storm hits you […] and then that will be it for six weeks. (P5, M, Episodic).

Note the metaphorical language used to try and describe the invisible experience. Episodic sufferers often spoke about the feeling of freeness once out of the bout, not remembering the pain, and living their life with no impingements.

> When you realize that you're out of them, it's like, you know, happy times. Let's let's go out. Let's celebrate […] I'm a happy go lucky chap out of my bouts erm (1 second pause) I'm I'm fully pain free. (P6, M, Episodic).

P2 expanded further, expressing that out of bout season they were in denial of their pain, making it easier to focus on engaging with life.

> Between the bouts I'm definitely in denial about them. So actually I would kind of forget about it if that makes sense and which is which is kind of a good thing. So I've never, I've never not planned something in case I get them. (P2, M, Episodic).

However, P3 did not share these experiences, stating that between bouts they were focused on the next one.

> It'd be nice to feel like the weights lifted off your shoulders, but it doesn't because you're more you're already thinking "when's the next bout"? […] it doesn't sort of leave you. (P3, M, Episodic).

Furthermore, several participants spoke about the catching up they had to do between bouts/attacks. For example, P4 was a month post their last bout:

> I'm kind of trying to pick up the pieces a little bit about my life. (P4, F, Episodic).

Of course, CCH sufferers did not have the experience of being out of bout season. Therefore, the condition was always there, which came with emotions of tiredness and a sense of despair; which relates to Theme Five.

> You can just feel like it's just one long. Horrible. Road of. Pain and. You know, feeling ill all the time. (P10, F, Chronic).

Interestingly, P7 took a different stance to the chronicity of their headaches, stating they felt episodic sufferers had a worse experience due to increased uncertainty and unpredictability in pain patterns.

> It's even worse for them (episodic sufferers), I think, because erm because their life gets completely interrupted. Where mine is mine doesn't start because because this is part of it. Tonight I'm almost certainly gonna wake up a few times (in pain). (P7, M, Chronic).

The researcher reflected that the psychological experience of ECH and CCH felt distinct, with uncertainty being exacerbated for the former, and certainty but also perhaps despair exacerbated for the latter.

Theme Four: Control: In-between Attacks

Knowledge.

All participants spoke about the defining moment they received their CH diagnosis and understood what was happening to them. This diagnosis often took years due to perceived failings from the NHS. It was common for individuals to believe they were suffering from a

life-threatening illness prior to diagnosis. This knowledge seemed to help them feel less concerned by the condition and allowed them to start taking control of helping themselves.

> When they first started I was very very concerned and I thought I was probably having a brain tumour […] Once I'd sort of started seeing more specialists and things, they really were reassuring. Erm. And. Over over the years I've gotten to accept it is it is what it is. (P11, M, Episodic).

All participants were incredibly knowledgeable about CH. Participants reflected that this knowledge was obtained through having lived with the condition for so long, and from self-learning, rather than the information being provided. Individuals seemed to always be trying to learn more about the condition and how to manage it.

> I know it so well (because it's happened) thousands of thousands of thousands of times. (P8, F, Chronic).

> Invariably erm my Google search history changes quite drastically when I'm in a bout cause you constantly trying to search for answers. (P3, M, Episodic).

Having a physiological understanding about the condition seemingly helped participants detach/separate from the dark, evil quality of the condition identified by Theme One "Darkness", and made it easier to manage the pain.

> It's probably something to do with some chemical some some hormones being released which regulate the body clock […] so I'm quite happy having this chemical

> enemy if you like […] rather than some entity which is making me feel bad because it doesn't like me or he wants to punish me. (P7, M, Chronic).

Furthermore, this medical understanding helped participants feel more in control of their experience through lifestyle changes.

> It (the physiological understanding) takes away that sort of. Doom and gloom and it it it. If something's happening in my body. If something's happening in my brain and that it's my pituitary gland. My hypothalamus somethings going on. You know, I think I've. I've I've kind of. Like with the amount of supplements I'm taking and and the why I'm healthily eating. Cut out sugar cut out carbs and stuff. And I'm I'm getting a lot of fat I'm feeding my head basically. (P12, M, Episodic).

Planning.

As well as becoming knowledgeable about the condition, participants spoke extensively about the medication and lifestyle changes they had tried over time. They referred to routines they had developed which helped them plan for future attacks and feel calmer and more in control.

> I've got my own. Little. Kind of routine that I get into when it's happening […] That's that's worked for me because I don't panic. (P12, M, Episodic).

All participants verbalised a complete reliance on routines to manage the pain. Throughout the interviews, participants would often keep coming back to speak about these pain management strategies, particularly medication, even if the interviewer tried to move the

conversation on. This likely highlighted the clear salience of these strategies for people. This reliance on pain management routines may have related to low self-efficacy; indeed, 72% of the current sample scored below the level indicative of low self-efficacy

> The thoughts of being out somewhere and starting with an attack and not having any access to (medication) I can't. That that I I can't. It's a bit like being caught out in the middle of town with no clothes on. (P1, M, Episodic).

It was also common for individuals to plan upcoming events around their headaches.

> We didn't book a wedding in (a certain month) because I had got cluster headaches in (this month). (P4, F, Episodic).

However, others took an approach of trying to not plan around the CH, to not let the condition consume their life. This was particularly true for individuals who had lived with the condition for many years. Furthermore, CCH sufferers, who did not have remission periods, spoke about having no choice but to continue with life events.

> If I let it (cluster headache) be front and centre with everything […] I would literally never go anywhere or do anything, and I can't do that and I wouldn't do that to my family and friends because it's not fair on them […] It is always in the back of my mind, I worry, you know, Christmas, birthdays. (P10, F, Chronic).

> What's the choice? You lie down and die. Or you go to bed permanently, or you just carry on and have as much of a life as you can. (P8, F, Chronic).

Theme Five: Despair

The final theme identified was despair, meaning hopelessness, depression, exhaustion, and negative thoughts. This related to feeling distressed that life was interrupted and being physically drained by repeated intense pain. This theme was particularly present for CCH sufferers, which perhaps is understandable considering they do not have the respite from pain that episodic sufferers experience.

> Why wouldn't you be depressed? Your life is impacted. You're in pain. You can't do the things you used to do. You're continually brought up against your limit. These these are these are things that make people feel miserable. (P8, F, Chronic).

> It does make you feel extremely drained as well. Like afterwards. I'm just like a wreck because I just feel like someone has zapped all of the energy out of me. (P10, F, Chronic).

> You just don't see an end to- you don’t see light at the end of the tunnel sometimes. (P6, M, Episodic).

Some participants spoke about the negative thoughts they had about themselves throughout bout season.

P12 stated that physical pain caused “deep emotional pain”, dwelling on the past and thinking “terrible things about yourself”. (P12, M, Episodic).

Several participants spoke about their thoughts becoming less negative over time. P5, who had had CH for many years said:

> 10 years ago. Uh, there's a lot of fear […] Because I didn't know what it was […] I thought like a tumour, I thought cancer […] There was a lot of woe is me […] the world is horrible to me. It's not fair. (P5, M, Episodic).

P5 said that now having a "label" and knowing "I'm not about to die, even if it feels like that" meant that now "whatever, negative thoughts I feel are all about relieving that pain" rather than negative thinking. The importance of a diagnosis is something which is echoed in many chronic pain conditions, and indeed health conditions more broadly. Moreover, this relates to the theme of "Control".

Similarly, P4 stated that they now tried to challenge irrational thoughts. However, they spoke about the buildup of fatigue over the course of the bout making this balanced thinking progressively more challenging. This would be an important consideration when thinking about psychological interventions for CH sufferers. It may be that cognitive treatment throughout a bout may be difficult to engage with, and instead psychological interventions outside of bout season would be preferential. Of course, this would not be possible for CCH individuals.

> I try and do that (challenge thoughts) but obviously I get worse erm, the worse the pain is and the less sleep I've had. (P4, F, Episodic)

Despite individuals feeling hopeless, there was a general sense of individuals not allowing themselves to be beaten by their pain. Furthermore, there was a clear wish to share this hope with other sufferers. The interviewer reflected that they felt these emotions throughout the interview, with there being moments they felt despairing as the participants spoke of their condition, however the interviewer always felt hopeful by the end of the meetings. They also felt a strong desire to advocate for CH sufferers; which links to the 'Battling' theme.

> It's a nightmare. This condition […] it's really, really awful. It's not well understood. The treatments are limited […] but don't give up […] would be my message. To anyone who's starting on this nightmare […] Don't give up. You know you will find some way to manage it. (P8, F, Chronic).

Discussion

This qualitative study aimed to explore the psychological experience of CH. Reflective Thematic Analysis (RTA) identified themes and (subthemes): Darkness (Evil Phenomenon and Dark Cognitions), Battling (With the Pain and for Recognition), Shifting, Control (Knowledge and Planning), and Despair. The themes existed within specific phases: 1) in the moment of the pain attack and 2) between attacks. All participants reported the jarring shift between being in pain and pain-free. For ECH sufferers this pain-free period could last for a substantial time; and many reflected feeling like a 'normal' person in this period. As CCH sufferers did not have such periods, the psychological experience felt distinct between the sub-types. Uncertainty felt exacerbated for ECH, whereas certainty, but also despair, felt heightened for CCH.

Main Findings in the Context of Previous Literature

To the authors' knowledge, this is the first study to identify the dark quality of the experience of CH. Previous research has noted that individuals describe the intensity of CH pain using vivid language (Nesbitt & Goadsby, 2012; Palacios-Ceña et al., 2016) but has not commented on the almost other-worldly, evil atmosphere. Using detailed stories and metaphors, such as "the monster", to describe pain, is noted in chronic pain literature as a vehicle to share the invisible experience of pain and promote understanding and empathy(Clarke et al., 2012; Johnson et al., 2023; Nortvedt & Engelsrud, 2014). As well as using such language to convey the experience, one participant in the present study suggested they used this language in a humorous way to make the experience less difficult. Use of humour to support positive reappraisal of chronic pain has been cited as effective previously for other pain conditions (Cuevas-Toro et al., 2008; Fritz et al., 2017; Pérez-Aranda et al., 2019).

Within the theme of "Darkness" was also "Dark Cognitions", including suicidal ideation. Several participants referred to suicidal thoughts throughout the attack to stop the pain. This finding is perhaps unsurprising given CH is known as "the suicide headache" (Wei & Goadsby, 2021) and research has reported higher rates of suicidality amongst CH individuals compared to non-headache controls (Choong et al., 2017; Crespi et al., 2022; Jürgens et al., 2011; Koo et al., 2021). Current study participants were clear that outside of attacks they did not have suicidal thoughts. This experience is comparable to findings by Ji and Lee (2019) who explored 175 CH patients and reported suicidality throughout an attack which reduced between attacks. It is interesting to consider whether the reputation of CH being associated with suicidality results in healthcare professionals viewing this as a diagnostic marker rather than catalysing mental health support; an idea speculated by one of the current study participants. This links to the theme of "Battling for Recognition".

The "Battling with the Pain" theme related to participants becoming behaviourally agitated throughout attacks. The agitated behaviour arguably relates to the fear-anxiety-avoidance model of chronic pain (Vlaeyen & Linton, 2000). This model proposes that when one perceives pain, their appraisal of this pain influences their behaviour and physiological response. If one catastrophises their pain, such as thinking the condition is a monster/evil (Darkness Theme), this can result in physiological arousal and escape/defensive behaviours. Arousal results in increased heart rate and muscle tension, which counterintuitively can exacerbate pain (Asmundson et al., 2004; Flor et al., 1992; Norton & Asmundson, 2003). Some interviewees spoke about, over the years, learning that battling was not helping and so tried to relax instead. Interestingly, all the participants who tried to relax had scores on the pain catastrophising questionnaire below the threshold, whereas most participants were above the threshold. It may be that less catastrophic interpretations of their pain allowed them to respond in a way which was more adaptive i.e. not battling the pain.

The theme "Battling" also included "Battling for Recognition" which involved feeling misunderstood by those around them and let down by the NHS. These experiences paralleled with the two previous qualitative studies which explored CH experiences (Andre & Cavers, 2021; Palacios-Ceña et al., 2016). Participants from these previous studies reported feeling their pain was doubted by people in their lives, and that there were often strained relationships with clinicians due to perceived failures in diagnosis and treatment provision. Indeed, one participant in the Andre and colleagues' study (2021) referred to the process of dealing with GPs as "battles" (p. 422). More broadly, the experience of feeling one's pain level is questioned (Glenton, 2003; Goffman, 1963; Newton et al., 2013) and being let down by healthcare (Hadi et al., 2017) is commonly reported by individuals with various chronic pain conditions. This results in both actual and perceived stigma, which has been associated with feelings of hopelessness, guilt, and embarrassment (Newton et al., 2013); all affective

experiences noted by the participants in the present study. Participants in both the current study and the Andre and Cavers (2021) study voiced disliking the term "Cluster Headache" as it undermined the severity of the condition.

Theme Three related to "Shifting" between being in and out of pain. Sufferers depicted having almost two lives and identities. This experience of shifting is seen in many chronic diseases (Paterson, 2001). When one suffers a chronic illness, the presence of illness can be inconsistent with one's previous healthy identity so causing identity-discrepancy and psychological distress (Higgins, 1987). Illness identity is how much one's illness is incorporated into their overall identity (Charmaz, 1995) and four responses have been proposed: 1) Engulfment, when illness dominates oneself, 2) Rejection, when one denies their illness, 3) Acceptance, where one willingly incorporates their illness into overall identity, and 4) Enrichment, where one learns from their illness (Oris et al., 2016). Acceptance and Enrichment are viewed as more adaptive, as identity coherence results in better self-management (Oris et al., 2018; Peters & Brown, 2022). Individuals in the current study, who had been living with CH for many years, spoke about changing the way they responded to their condition; going from a position of battling against the pain, being hyper-vigilant to triggers, and avoiding social events, to trying to relax with the pain and continue engaging with life. This could be likened to reaching a place of Acceptance or Enrichment.

The theme of "Control" through knowledge and planning reported in the present study draws parallels with the previous qualitative UK CH study (Andre & Cavers, 2021). Andre and Cavers (2021) highlighted the huge moment participants received a diagnosis, and how this catalysed a process of self-learning about CH. Participants in both the present study and the study by Andre and Cavers (2021) seemed to develop well-established routines, often involving medication, to manage the pain. Both studies' participants appeared dependent upon these routines, perhaps indicating low belief regarding their ability to cope with pain.

Low self-efficacy may be an important aspect in the theme of "Control", with the majority scoring below the level indicative of low self-efficacy on the PSEQ (Tonkin, 2008).

In both the current study and the previous UK study (Andre & Cavers, 2021) some participants spoke about planning their lives around CH. This involved not planning important events in bout season and avoiding triggers. However, it was common that such hyper-vigilance restricted individuals' lives and participants voiced the conflict of trying to plan for CH whilst not allowing it to dominate. This aligns with the fear-anxiety-avoidance model (Vlaeyen & Linton, 2000) which proposed that fear of pain and resulting reduction in activity to try and reduce pain, exacerbates pain through deconditioning and reduced engagement with life (Ruscheweyh et al., 2019). Research has tested this model amongst headache patients. A qualitative study of Migraine sufferers reported they struggled to find the balance between managing pain and engaging with life (Varkey et al., 2013) and exaggerated avoidance of headache triggers has been found to be maladaptive (Martin & MacLeod, 2009). Successful headache treatment involves gradually re-introducing activities to increase desensitisation and quality of life (Martin et al., 2014). Perhaps CH sufferers have learnt over time to do this independently.

Finally, the theme "Despair" has been echoed by previous literature. Both previous qualitative studies exploring CH detailed experiences of depression and life stopping throughout bout seasons (Andre & Cavers, 2021; Palacios-Ceña et al., 2016). CH individuals experiencing depression is also something echoed more widely by literature, with research reporting higher rates of depression in CH adults compared to non-headache controls (Choong et al., 2017; Crespi et al., 2022; Kim et al., 2024). To authors' knowledge, this is the first study to identify that this despair may have been more pronounced for CCH individuals.

Strengths and Limitations

The Eight Big Tent Criteria was used to assess this piece of work (Tracy, 2010). The *worthiness* of the topic was indicated by this research being suggested by a PPI representative and by the huge interest in the study.

In terms of *rigor*, the sample size was considered appropriate. The research team reflected on the strong quality of dialogue within the interview transcripts and so concluded that the data contained sufficient information power to meet the study aims (Malterud et al., 2016). Interviews were conducted either using telephone or video technology. Whilst there were concerns the lack of face-to-face interaction may diminish rapport and reduce the *rigor* and quality of information gathered (Saarijärvi & Bratt, 2021), authors felt a good rapport was established with all interviewees; evidenced by the long, rich, interviews.

The use of OUCH for recruitment meant that the sample may have been biased towards digitally literature individuals, so limiting transferability and *resonance* of the findings. However, the demographics of the sample, in terms of gender and type of CH, reflected the CH epidemiological profile (Wei et al., 2018). Further, utilising the nationwide charity, meant the sample was from across the UK. The findings were concordant with pre-existing literature conducted in both similar and different contexts (UK and Spain; Andre & Cavers, 2021; Palacios-Ceña et al., 2016) perhaps suggesting findings are transferable across settings. The way findings interconnect and develop pre-existing literature supports the *meaningful coherence* of the research.

Researchers took a critical realist approach, assuming the qualitative process is subjective. To account for this and maintain *sincerity*, the primary researcher (HW) kept a diary of their thought processes and reflected with the research team. Furthermore, the research process was transparent, with an example of the RTA process being available in Appendix O. The research followed appropriate *ethical* protocols.

Authors aimed to increase *credibility* by displaying extensive interview quotes, hence providing thick description, and giving voice to participants (Geertz, 1973). Sharing quotes also allowed for the powerfulness of participants' accounts to be shared, increasing the work's *resonance*. Furthermore, the researcher focused on achieving multivocality, by paying attention to the full range of interviewees' viewpoints, and incorporating reflections from the PPI representative.

To authors' knowledge, this is the first research to explore the psychological processes present for CH individuals, both throughout an attack and between, and to highlight the distinct nature of CCH and ECH. Additionally, this research gave the PPI member an opportunity to gain something positive from living with CH (see PPI reflections in Chapter Five). Therefore, it can be argued that this study *significantly contributed* to the field.

Implications and Future Directions

This research emphasised the need for increased visibility of CH, both within the healthcare world to improve diagnosis and treatment, and within the public generally to increase empathy and understanding. Authors recommend research to explore how greater awareness can be achieved; be that through changing the name "Cluster Headache" or through public health campaigns. Furthermore, currently individuals seem to become knowledgeable about the condition through independent discovery. Having a consultation post-diagnosis where one is informed about the condition and appropriate treatment, is recommended to support individuals.

This study identified the complex psychological experience related to CH, which can be separated distinctly into the acute attack and between attacks. Authors identified that participants described a dark, evil atmosphere throughout an attack. This theme felt powerful and almost superordinate. Future research could employ Interpretative Phenomenological

Analysis which examines individual experience in detail and is useful for emotionally laden, elusive, phenomenon (Smith & Osborn, 205).

Authors identified that once sufferers became familiar with CH, their psychological response changed, and their distress often reduced. Participants reported trying to relax through the pain, not avoiding social events, reduced negative thoughts about CH, and feeling more in control through knowledge and planning. Participants had not received psychological input, and so these were learnings they developed independently. This could suggest that fighting against pain, avoidance, potent negative catastrophic cognitions, and lack of control may maintain distress in the CH experience. Authors call for further exploration into the nature of these factors and the extent to which specific factors are associated with maintaining suffering.

Speculative Discussion

Once a clear formulation of CH is defined, there is potential to develop psychological treatments. These treatments could involve adapting existing evidence-based approaches; as factors identified by this research are easily recognisable from existing evidence-based psychological models. Indeed, the idea of stopping battling against pain has similarities to psychological flexibility in ACT (Hayes & Pierson, 2005) and remaining calm through attacks has similarities to distress tolerance grounding techniques in Dialectical Behavioural Therapy (DBT; Linehan, 1993). Further, engaging with life, not avoiding situations, and changing one's thoughts, has parallels with behavioral activation, exposure therapy, and cognitive restructuring in CBT (Beck et al., 1979; Beck et al., 2005). Finally, participants' reported change in perspective over time is comparable to accepting illness into one's overall identity (Oris et al., 2016), something encouraged in ACT through 'self as context' (Crombez et al., 2003; Hayes et al., 1999).

Psychological experiences appear different in the acute attack, between attacks, and between bouts for episodic sufferers. Therefore, when developing a treatment, timing will be an important consideration. For example, it has been speculated that psychological support could be offered in headache free periods, when one is not exhausted and drained, in preparation for attacks (Schenck & Andrasik, 2019). Importantly, CCH sufferers do not have remission periods, so timing would need to be individualised. Research exploring CCH and ECH separately is recommended.

Conclusion

This study demonstrated the huge burden placed on those living with the primary headache condition, CH. The complex psychological experience was evidenced by five themes emerging from RTA: Darkness, Battling, Shifting, Control, and Despair. These themes were separated into distinct phases: 1) the moment of the painful attack, and 2) between attacks. Experiences seemed to be different for ECH and CCH. Authors call for greater attention to the nature of the psychological aspects of CH, and how certain psychological processes may maintain distress. Following this, authors recommend exploration into how psychological approaches and the social context around individuals, can improve the lives of those with CH.

Ethical Approval and Consent to Participate

This study was approved by the Faculty of Medicine and Health Sciences Ethics Committee at the University of East Anglia, after one set of amendments (ID: ETH2324-0070, see Appendix M).

Consent for publication

Participants completed consent forms for their transcriptions to be included in data analysis and presented in this paper. The data has been anonymised.

Availability of data and materials

Full transcriptions are not publicly available to protect participant confidentiality.

Competing interests

The authors declare that they have no competing interests.

Funding

There was no grant funding to support this study, however it was conducted as part of the fulfilment of a Clinical Psychology Doctoral Programme funded by NHS England.

Authors' contributions

HW was the main author, with EN being primary supervisor and FG secondary supervisor. WN was the PPI representative. HW conducted all interviews and analysis but had frequent research meetings with the other members of the team to reflect on the thematic analysis process.

Acknowledgements

See page five of the thesis portfolio.

References

Agius, S. J. (2013). Qualitative research: Its value and applicability. *Psychiatrist, 37*(6), 204-206. https://doi.org/10.1192/pb.bp.113.042770

Ahmed, F., Bahra, A., Tyagi, A. & Weatherby, S. (2019) *National headache management system for adults.* British Association for the Study of Headache. http://www.bash.org.uk

Andre, L., & Cavers, D. (2021). 'A cry in the dark': a qualitative exploration of living with cluster headache. *British Journal of Pain, 15*(4), 420-428. https://doi.org/10.1177/2049463720976695

Asghari, A., & Nicholas, M. K. (2001). Pain self-efficacy beliefs and pain behaviour. A prospective study. *Pain, 94*(1), 85-100. https://doi.org/10.1016/S0304-3959(01)00344-X

Asmundson, G., Vlaeyen, J., & Crombez, G. (2004). *Understanding and treating fear of pain.* In Oxford University Press eBooks. https://doi.org/10.1093/oso/9780198525141.001.0001

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioral change. *Psychological Review, 84*(2), 191-215. https://doi.org/10.1037/0033-295X.84.2.191

Beck, A. T., Emery, G., & Greenberg, R. L. (2005). *Anxiety disorders and phobias: A cognitive perspective.* Basic Books/Hachette Book Group.

Beck, A. T., Rush, A., J., Shaw, B. F. (1979). *Cognitive therapy of depression.* New York: Guilford Press.

Benson, H., Klemchuk, H. P., & Graham, J. R. (1974). The usefulness of the relaxation response in the therapy of headache. *Headache, 14*(1), 49-52. https://doi.org/10.1111/j.1526-4610.1974.hed1401049.x

Bhaskar R. (2008). *A realist theory of science*. Routledge.

Bhaskar, R. (1998). Philosophy and scientific realism. In Archer M. S. (Ed.), *Critical realism: Essential readings.* Routledge.

Blanchard, E. B., Andrasik, F., Jurish, S. E., & Teders, S. J. (1982). The treatment of cluster headache with relaxation and thermal biofeedback. *Biofeedback and Self-regulation, 7*(2), 185-191. https://doi.org/10.1007/BF00998782

Braun, V., Clarke, V. (2013). *Successful Qualitative Research: A Practical Guide for Beginners.* Sage Publications, Thousand Oaks

Braun, V., & Clarke, V. (2019). Reflecting on reflexive thematic analysis. Qua*litative Research in Sport, Exercise and Health, 11,* 589-597. https://doi.org/10.1080/2159676X.2019.1628806

Braun, V., & Clarke, V. (2020). One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology, 18*(3), 328-352. https://doi.org/10.1080/14780887.2020.1769238

Braun V., Clarke V. (2022). *Thematic analysis: A practical guide.* London.

Braun, V., & Clarke, V. (2022). Toward good practice in thematic analysis: Avoiding common problems and be(com)ing a *knowing* researcher. *International Journal of Transgender Health*, *24*(1), 1-6. https://doi.org/10.1080/26895269.2022.2129597

British Pain Society. (2019). *Outcome Measures.* https://www.britishpainsociety.org/static/uploads/resources/files/Outcome_Measures_January_2019.pdf

Burish, M. J., Pearson, S. M., Shapiro, R. E., Zhang, W., & Schor, L. I. (2021). Cluster headache is one of the most intensely painful human conditions: Results from the

International Cluster Headache Questionnaire. *Headache*, *61*(1), 117-124. https://doi.org/10.1111/head.14021

Burke, P. J., & Stets, J. E. (2009). *Identity theory.* Oxford, UK: Oxford University Press.

Buzzi, M. G., Pellegrino, M. G., & Bellantonio, P. (1995). Causes and mechanisms of primary headaches: toward a bio-behavioral model. Italian Journal of Neurological Sciences, 16(9), 15- 19. https://doi.org/10.1007/BF02333240

Care Quality Commission. (2018). Quality Improvements in Hospital Trusts. https://www.cqc.org.uk/sites/default/files/20180911_QI_hospitals_FINAL.pdf

Charmaz, K. (1995). The body, identity, and self: adapting to impairment. *The Sociological Quarterly, 36*(4), 657-680. https://doi.org/10.1111/j.1533-8525.1995.tb00459.x

Clarke, A., Anthony, G., Gray, D., Jones, D., McNamee, P., Schofield, P., Smith, B. H., & Martin, D. (2012). "I feel so stupid because I can't give a proper answer…" How older adults describe chronic pain: a qualitative study. *BMC Geriatrics*, *12*, 78. https://doi.org/10.1186/1471-2318-12-78

Crombez, G., Morley, S., McCracken, L., Sensky, T., & Pincus, T. (2003). Self, identity, and acceptance in chronic pain. *Proceedings of the 10th World Congress on Pain*, 24, 651-659. https://www.researchgate.net/publication/230596566_Self_identity_and_acceptance_in_chronic_pain

Cuevas-Toro, A. M., Torrecillas, F. L., Medina, A. G., & Diaz-Bata-nero, M. C. (2008). Personality and coping strategies in fibromyalgia patients. *Behavioral Psychology: Revista Internacional ClínicaY de La Salud, 16,* 289-306.

D'Amico, D., Raggi, A., Grazzi, L., & Lambru, G. (2020). Disability, quality of life, and socioeconomic burden of CH: A critical review of current evidence and future perspectives. *Headache, 60*(4), 809-818. https://doi.org/10.1111/head.13784

Day, M. A., Ward, L. C., Thorn, B. E., Lang, C. P., Newton-John, T. R. O., Ehde, D. M., & Jensen, M. P. (2018). The Pain-Related Cognitive Processes Questionnaire: Development and Validation. Pain Medicine, 19(2), 269-283. https://doi.org/10.1093/pm/pnx010

Fischera, M., Marziniak, M., Gralow, I., & Evers, S. (2008). The incidence and prevalence of cluster headache: a meta-analysis of population-based studies. *Cephalalgia: an International Journal of Headache, 28*(6), 614-618. https://doi.org/10.1111/j.1468-2982.2008.01592.x

Flor, H., Birbaumer, N., Schugens, M. M., & Lutzenberger, W. (1992). Symptom-specific psychophysiological responses in chronic pain patients. *Psychophysiology, 29*(4), 452-460. https://doi.org/10.1111/j.1469-8986.1992.tb01718.x

Freeman, E., Adair, M., Beeler, D., Casper, R., Herman, M., Reeves, D., & Reinsch, S. (2022). Patient identified burden and unmet needs in patients with CH: An evidence-based qualitative literature review. *Cephalalgia Reports, 5,* 1. https://doi.org/10.1177/25158163221096866

Fritz, H. L., Russek, L. N., & Dillon, M. M. (2017). Humor use moderates the relation of stressful life events with psychological distress. *Personality & Social Psychology Bulletin, 43*(6), 845-859. https://doi.org/10.1177/0146167217699583

Geertz, C. (1973). *The interpretation of cultures: Selected essays.* New York: Basic Books.

Glenton C. (2003). Chronic back pain sufferers-striving for the sick role. *Social Science & Medicine (1982), 57*(11), 2243-2252. https://doi.org/10.1016/s0277-9536(03)00130-8

Goffman, E. (1963). *Stigma: notes on the management of spoiled identity.* London: Penguin Books.

González de la Flor, Á., García Pérez de Sevilla, G., Domíngez Balmaseda, D., Martín Vera, D., Montero Martínez, M., & Del Blanco Muñiz, J. Á. (2022). Relationship between self-efficacy and headache impact, anxiety, and physical activity levels in patients with chronic tension-type headache: an observational study. *Behavioural Neurology, 6,* 8387249. https://doi.org/10.1155/2022/8387249

Gorczyca, R., Filip, R., & Walczak, E. (2013). Psychological aspects of pain. *Annals of Agricultural and Environmental Medicine (AAEM), 1*, 23-27. https://www.researchgate.net/publication/263778347_Psychological_Aspects_of_Pain

Grinberg, A. S., Best, R. D., Min, K. M., Schindler, E. A. D., Koo, B. B., Sico, J. J., & Seng, E. K. (2021). Cluster Headache: Clinical Characteristics and Opportunities to Enhance Quality of Life. *Current Pain and Headache Reports*, *25*(10), 65. https://doi.org/10.1007/s11916-021-00979-8

Hadi, M. A., Alldred, D. P., Briggs, M., Marczewski, K., & Closs, S. J. (2017). 'Treated as a number, not treated as a person': a qualitative exploration of the perceived barriers to effective pain management of patients with chronic pain. *BMJ Open*, *7*(6), e016454. https://doi.org/10.1136/bmjopen-2017-016454

Hasenbring, M. I., Hallner, D., & Rusu, A. C. (2009). Fear-avoidance- and endurance-related responses to pain: development and validation of the Avoidance-Endurance Questionnaire (AEQ). *European Journal of Pain (London, England), 13*(6), 620-628. https://doi.org/10.1016/j.ejpain.2008.11.001

Hayes, S.C., Pierson, H. (2005). Acceptance and Commitment Therapy. In: Freeman, A., Felgoise, S.H., Nezu, C.M., Nezu, A.M., Reinecke, M.A. (eds) *Encyclopedia of Cognitive Behavior Therapy.* Springer, Boston, MA. https://doi.org/10.1007/0-306-48581-8_1

Hayes, S. C., Strosahl, K. D., & Wilson, K. G. (1999). *Acceptance and commitment therapy: An experiential approach to behavior change.* Guilford Press.

Headache Classification Committee of the International Headache Society (IHS). (2018). The International Classification of Headache Disorders, 3rd edition. *Cephalalgia: an International Journal of Headache, 38*(1), 1-211. https://doi.org/10.1177/0333102417738202

Hee, S. W., Patel, S., Sandhu, H., Matharu, M. S., Underwood, M., & CHESS Team (2024). Does pain self-efficacy predict, moderate or mediate outcomes in people with chronic headache; an exploratory analysis of the CHESS trial. *The journal of Headache and Pain, 25*(1), 77. https://doi.org/10.1186/s10194-024-01768-5

Hennink, M., & Kaiser, B. N. (2022). Sample sizes for saturation in qualitative research: A systematic review of empirical tests. *Social Science & Medicine (1982)*, *292*, 114523. https://doi.org/10.1016/j.socscimed.2021.114523

Higgins, E. T. (1987). Self-discrepancy: A theory relating self and affect. *Psychological Review, 94*(3), 319-340. https://doi.org/10.1037/0033-295X.94.3.319

Jensen, M. P., Thorn, B. E., Carmody, J., Keefe, F. J., & Burns, J. W. (2018). The role of cognitive content and cognitive processes in chronic pain: An important distinction? *The Clinical Journal of Pain, 34*(5), 391-401. https://doi.org/10.1097/AJP.0000000000000559

Jensen, R., Zeeberg, P., Dehlendorff, C., & Olesen, J. (2010). Predictors of outcome of the treatment programme in a multidisciplinary headache centre. *Cephalalgia: an International Journal of Headache, 30*(10), 1214-1224. https://doi.org/10.1177/0333102410361403

Ji Lee, M., Cho, S. J., Wook Park, J., Kyung Chu, M., Moon, H. S., Chung, P. W., Myun Chung, J., Sohn, J. H., Kim, B. K., Kim, B. S., Kim, S. K., Song, T. J., Choi, Y. J., Park, K. Y., Oh, K., Ahn, J. Y., Lee, K. S., Cho, S., & Chung, C. S. (2019). Increased suicidality in patients with CH. *Cephalalgia: an International Journal of Headache, 39*(10), 1249-1256. https://doi.org/10.1177/0333102419845660

Johnson, M. I., Hudson, M., & Ryan, C. G. (2023). Perspectives on the insidious nature of pain metaphor: we literally need to change our metaphors. *Frontiers in Pain Research (Lausanne, Switzerland)*, *4*, 1224139. https://doi.org/10.3389/fpain.2023.1224139

Klonowski, T., Kropp, P., Straube, A., & Ruscheweyh, R. (2022). Psychological factors associated with headache frequency, intensity, and headache-related disability in migraine patients. *Neurological Sciences: Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology, 43*(2), 1255-1266. https://doi.org/10.1007/s10072-021-05453-2

Koopmans, E., & Schiller, D. C. (2022). Understanding causation in healthcare: an introduction to critical realism. *Qualitative Health Research, 32*(8-9), 1207-1214. https://doi.org/10.1177/10497323221105737

Lim, W. M. (2024). What is qualitative research? An overview and guidelines. *Australasian Marketing Journal, 0*(0). https://doi.org/10.1177/14413582241264619

Malterud, K., Siersma, V. D., & Guassora, A. D. (2016). Sample size in qualitative interview studies: guided by information power. *Qualitative Health Research*, *26*(13), 1753-1760. https://doi.org/10.1177/1049732315617444

Martin, P. R., & MacLeod, C. (2009). Behavioral management of headache triggers: Avoidance of triggers is an inadequate strategy. *Clinical Psychology Review*, *29*(6), 483-495. https://doi.org/10.1016/j.cpr.2009.05.002

Martin, P. R., Reece, J., Callan, M., MacLeod, C., Kaur, A., Gregg, K., & Goadsby, P. J. (2014). Behavioral management of the triggers of recurrent headache: a randomized controlled trial. *Behaviour Research and Therapy*, *61*, 1-11. https://doi.org/10.1016/j.brat.2014.07.002

Newton, B. J., Southall, J. L., Raphael, J. H., Ashford, R. L., & LeMarchand, K. (2013). A narrative review of the impact of disbelief in chronic pain. *Pain Management Nursing: Official Journal of the American Society of Pain Management Nurses, 14*(3), 161-171. https://doi.org/10.1016/j.pmn.2010.09.001

NHS England. (2023). *Co-production: An Introduction.* NHS England. https://www.england.nhs.uk/long-read/co-production-an-introduction/

NICE. (2022). Cluster Headache. The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/headache-cluster/management/management/

NICE. (2022). *Headache-Tension Type.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/headache-tension-type/

NICE. (2024). *Migraines.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/migraine/management/adults/

NICE. (2024). NICE processes and methods. https://www.nice.org.uk/process/pmg20/chapter/introduction

Nicholas, M. K. (2007). The pain self-efficacy questionnaire: Taking pain into account. *European Journal of Pain (London, England), 11*(2), 153-163. https://doi.org/10.1016/j.ejpain.2005.12.008

Norton, P. J., & Asmundson, G. J. G. (2003). Amending the fear-avoidance model of chronic pain: What is the role of physiological arousal? *Behavior Therapy, 34*(1), 17-30. https://doi.org/10.1016/S0005-7894(03)80019-9

Nortvedt, F., & Engelsrud, G. (2014). "Imprisoned" in pain: analyzing personal experiences of phantom pain. *Medicine, Health Care, and Philosophy*, *17*(4), 599-608. https://doi.org/10.1007/s11019-014-9555-z

O'Brien, B. C., Harris, I. B., Beckman, T. J., Reed, D. A., & Cook, D. A. (2014). Standards for reporting qualitative research: a synthesis of recommendations. *Academic Medicine: Journal of the Association of American Medical Colleges*, *89*(9), 1245-1251. https://doi.org/10.1097/ACM.0000000000000388

Oris, L., Luyckx, K., Rassart, J., Goubert, L., Goossens, E., Apers, S., Arat, S., Vandenberghe, J., Westhovens, R., & Moons, P. (2018). Illness identity in adults with a chronic illness. *Journal of Clinical Psychology in Medical Settings, 25*(4), 429-440. https://doi.org/10.1007/s10880-018-9552-0

Oris, L., Rassart, J., Prikken, S., Verschueren, M., Goubert, L., Moons, P., Berg, C. A., Weets, I., & Luyckx, K. (2016). Illness identity in adolescents and emerging adults with Type 1 Diabetes: Introducing the Illness Identity Questionnaire. *Diabetes Care, 39*(5), 757-763. https://doi.org/10.2337/dc15-2559

OUCH. (2024). *Cluster Headache.* https://ouchuk.org/

Palacios-Ceña, D., Talavera, B., López-Ruiz, P., Gutiérrez-Viedma, Á., Palacios-Ceña, M., Arias, J. A., Fernández-de-Las-Peñas, C., & Cuadrado, M. L. (2016). Living with cluster headache: a qualitative study of patients' perspectives. *Headache, 56*(7), 1171–1182. https://doi.org/10.1111/head.12886

Paterson B. L. (2001). The shifting perspectives model of chronic illness. *Journal of Nursing Scholarship: an Official Publication of Sigma Theta Tau International Honor Society of Nursing, 33*(1), 21-26. https://doi.org/10.1111/j.1547-5069.2001.00021.x

Peters, L. A., & Brown, E. M. (2022). The relationship between illness identity and the self-management of Inflammatory Bowel Disease. *British Journal of Health Psychology, 27*(3), 956-970. https://doi.org/10.1111/bjhp.12584

Quartana, P. J., Campbell, C. M., & Edwards, R. R. (2009). Pain catastrophizing: a critical review. *Expert Review of Neurotherapeutics, 9*(5), 745-758. https://doi.org/10.1586/ern.09.34

Pérez-Aranda, A., Hofmann, J., Feliu-Soler, A., Ramírez-Maestre, C., Andrés-Rodríguez, L., Ruch, W., & Luciano, J. V. (2019). Laughing away the pain: A narrative review of humour, sense of humour and pain. European journal of pain (London, England), 23(2), 220–233. https://doi.org/10.1002/ejp.1309

Rossi, P., Little, P., De La Torre, E. R., & Palmaro, A. (2018). If you want to understand what it really means to live with CH, imagine... fostering empathy through European patients' own stories of their experiences. *Functional Neurology, 33*(1), 57-59. https://doi.org/10.11138/fneur/2018.33.1.057

Ruscheweyh, R., Pereira, D., Hasenbring, M. I., & Straube, A. (2019). Pain-related avoidance and endurance behaviour in migraine: an observational study. The Journal of Headache and Pain, 20(1), 9. https://doi.org/10.1186/s10194-019-0962-7

Saarijärvi, M., & Bratt, E. L. (2021). When face-to-face interviews are not possible: tips and tricks for video, telephone, online chat, and email interviews in qualitative research. *European Journal of Cardiovascular Nursing*, *20*(4), 392–396. https://doi.org/10.1093/eurjcn/zvab038

Sánchez-Rodríguez, E., Aragonès, E., Jensen, M. P., Tomé-Pires, C., Rambla, C., López-Cortacans, G., & Miró, J. (2020). The role of pain-related cognitions in the relationship between pain severity, depression, and pain interference in a sample of primary care

patients with both chronic pain and depression. Pain Medicine, 21(10), 2200-2211. https://doi.org/10.1093/pm/pnz363

Sargent, J. D., Walters, E. D., & Green, E. E. (1973). Psychosomatic self-regulation of migraine headaches. *Seminars in Psychiatry, 5*(4), 415-428. https://psycnet.apa.org/record/1974-23529-001

Schenck, L. A., & Andrasik, F. (2019). Behavioral and psychological aspects of cluster headache: an overview. *Neurological sciences: official journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology*, *40*(Suppl 1), 3-7. https://doi.org/10.1007/s10072-019-03831-5

Schiller C. J. (2016). Critical realism in nursing: An emerging approach. *Nursing Philosophy*, *17*(2), 88-102. https://doi.org/10.1111/nup.12107

Smith, J. A., & Osborn, M. (2015). Interpretative phenomenological analysis as a useful methodology for research on the lived experience of pain. *British Journal of Pain, 9*(1), 41-42. https://doi.org/10.1177/2049463714541642

Staniszewska, S., Brett, J., Simera, I., Seers, K., Mockford, C., Goodlad, S., Altman, D. G., Moher, D., Barber, R., Denegri, S., Entwistle, A., Littlejohns, P., Morris, C., Suleman, R., Thomas, V., & Tysall, C. (2017). GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research. *BMJ (Clinical research ed.), 358,* j3453. https://doi.org/10.1136/bmj.j3453

Sullivan, M. J. L., Bishop, S. R., & Pivik, J. (1995). The pain catastrophizing scale: development and validation. *Psycholological Assessment, 7*(4), 524-32. https://doi.org/10.1037/1040-3590.7.4.524

Sweeney, A., & Morgan, L. (2009). The levels and stages of service user/survivor involvement in research. *In Handbook of Service User Involvement in Mental Health Research* (pp. 25-35). Wiley Blackwell. https://doi.org/10.1002/9780470743157.ch3

The National Institute for Health Research [NIHR]. (2015). *Going the extra mile: improving the Nation's health and wellbeing through public involvement in research.* England: Department of Health. https://www.nihr.ac.uk/documents/about-us/our-contribution-to-research/how-we-involve-patients-carers-and-the-public/Going-the-Extra-Mile.pdf

Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care: Journal of the International Society for Quality in Health Care, 19*(6), 349-357. https://doi.org/10.1093/intqhc/mzm042

Tonkin L. (2008). The Pain Self-Efficacy Questionnaire. *The Australian Journal of Physiotherapy*, *54*(1), 77. https://doi.org/10.1016/s0004-9514(08)70073-4

Torres, N. D., Newman, A. K., Day, M. A., Chan, J. F., Friedly, J. L., & Jensen, M. P. (2024). The Moderating Effects of Cognitive Processes on Pain-related Outcomes. *The Journal of Pain, 25*(7), 104483. https://doi.org/10.1016/j.jpain.2024.01.348

Tracy, S. J. (2010). Qualitative quality: Eight "big-tent" criteria for excellent qualitative research. *Qualitative Inquiry*, *16*(10), 837-851. https://doi.org/10.1177/1077800410383121

Varkey, E., Linde, M., & Henoch, I. (2013). "It's a balance between letting it influence life completely and not letting it influence life at all"--a qualitative study of migraine prevention from patients' perspective. *Disability and Rehabilitation*, *35*(10), 835-844. https://doi.org/10.3109/09638288.2012.709304

Vlaeyen, J. W. S., & Linton, S. J. (2000). Fear-avoidance and its consequences in chronic musculoskeletal pain: a state of the art. *Pain*, *85*(3), 317-332. https://doi.org/10.1016/S0304-3959(99)00242-0

Wei, C. B., Jia, J. P., Wang, F., Zhou, A. H., Zuo, X. M., & Chu, C. B. (2016). Overlap between headache, depression, and anxiety in general neurological clinics: A cross-

sectional study. *Chinese Medical Journal, 129*(12), 1394-1399. https://doi.org/10.4103/0366-6999.183410 g

Wei, D. Y., & Goadsby, P. J. (2021). Cluster headache pathophysiology - insights from current and emerging treatments. *Nature Reviews. Neurology, 17*(5), 308-324. https://doi.org/10.1038/s41582-021-00477-w

Wei, D. Y., Yuan Ong, J. J., & Goadsby, P. J. (2018). Cluster Headache: Epidemiology, pathophysiology, clinical features, and diagnosis. Annals of Indian Academy of *Neurology, 21*(Suppl 1), S3-S8. https://doi.org/10.4103/aian.AIAN_349_17

Yu, X., & Tan, G. (2024). Role of psychological factors in migraine. *Cureus,16*(12), e75858. https://doi.org/10.7759/cureus.75858

Chapter Five: Additional Methods Chapter

This chapter includes an in-depth description of the methodological process used in the empirical paper; specifically, the philosophical positioning, the development of the interview schedule, the questionnaires used, the thematic analysis conducted, further ethical considerations, and a detailed thematic map with example codes.

Epistemology and Ontology

In terms of philosophical positioning, the primary researcher (HW) took a pragmatic stance (Creswell & PlanoClark, 2007). The reason for this was because the design, whilst predominately qualitative, had a quantitative component, with the questionnaires. A mixed-methods design can result in philosophical issues, with qualitative work generally being associated with constructivism and subjectivity and quantitative research with empiricism and objectivist perspectives (Ma, 2012). Pragmatism assumes that of primary importance is the research question, and so the underlying philosophical view is secondary. In this approach, both qualitative and quantitative approaches can be accepted, assuming the methods are justified (Fishman, 1999; Tashakkori and Teddlie, 1998). The researcher does not inquire to seek a truth independent of human experience but instead aims to create shared meanings through complementary qualitative and quantitative approaches which can account for each other's shortcomings (Maxcy, 2003; Morgan, 2007; Shannon-Baker, 2016).

When considering the qualitative data, a critical realist approach was applied (Bhaskar, 1998, 2008). This approach has a long history of being used in Reflective Thematic Analysis (RTA; Braun & Clarke, 2022). The researcher assumes that there is an objective truth that exists, however knowledge is socially constructed and subjective; meaning our understanding of phenomena is fluid and dependent on the context (Schiller, 2016). To account for this, the researcher (HW) kept a reflective journal and discussed ideas with the

research team. When reporting the quantitative data, objectivity was assumed (Cisneros-Puebla, 2007; Losantos et al., 2016).

Materials

Interview Schedule

A draft interview schedule was created by the research team, which included the PPI representative (WN), and a clinical psychologist who previously worked in pain management services (EN). The charity OUCH gave feedback on the interview schedule and adjustments were made based on this feedback. As shown in Appendix H, the interview started with a 'settling in question' to support the participant to feel at ease (Britten, 1995). After this, there was a 'grand tour' question which was a broad question about the research topic which aimed to facilitate the participant talking about their experience (DeJonckheere & Vaughn, 2019). The interview schedule then became more specific, enquiring about participants' thoughts, behaviours, and feelings related to their pain, and their views about what could improve their pain management. This interview schedule was used flexibly, with the open-ended questions provided in the schedule being used to create a dialogue which resulted in other questions emerging (DiCicco-Bloom & Crabtree, 2006). The semi-structured nature also meant that the questions were modified by the interviewer when required to fit the context (DeJonckheere & Vaughn, 2019). This flexibility was important to ensure deep exploration into the research topic, which was not constricted by a strict set of questions.

Questionnaires

Supplementary to the interview, participants completed the 13-item Pain Catastrophising Scale (PCS: Sullivan et al., 1995) and the Pain Self-Efficacy Questionnaire (PSEQ; Nicholas, 2007). These scales are both psychometrically sound (Asghari & Nicholas, 2001; British Pain Society, 2019). Indeed, both have been found to have high internal consistency: Pain Catastrophising Scale (Cronbach's alpha 0.82-0.98; British Pain Society,

2019) and Pain Self-Efficacy Questionnaire (Cronbach's alpha 0.92; Asghari & Nicholas, 2001; Nicholas, 2007). Both questionnaires have also been found to have validity through high correlations to related concepts (Darnall et al., 2017; Dubé et al., 2021; Osman et al., 1997; Osman et al., 2000; Sullivan et al, 1995; British Pain Society, 2019; Van Damme et al., 2002).

Qualitative Data Analysis

Reflexive Thematic Analysis (RTA) was used to analyse the interview data. This method was chosen as it is a usable, straight-forward method to draw out common themes in a transparent way (Braun & Clarke, 2006; 2013; 2019; 2020; 2022). As there is limited research in the area it was decided it was more appropriate to explore themes across participants rather than individual experiences. The six-step approach created by Braun and Clarke (2006; 2013; 2019; 2020; 2022) was employed; familiarisation of data, coding, generation of themes, reviewing themes, naming themes, and writing up. The data was analysed inductively to allow the data to drive discovery. This was important as this analysis aimed to explore people's subjective experience and so it was not desirable that pre-conceived theories direct the results. The primary researcher (HW) coded the data. To account for unavoidable research bias (Braun and Clarke, 2012), the primary researcher kept a reflective journal throughout the process and reflected with others in the research team, most importantly the PPI representative. The primary researcher was mindful about their own personal characteristics which could influence the interview process and data analysis. Namely, the fact they didn't experience headaches, that they were coming from a psychological background, and that they were a young female.

Interviews were automatically transcribed by Microsoft Teams and the primary researcher edited them to ensure accuracy. Data analysis involved initially cleaning the data, through removing identifiable information and allocating each transcript a participant

number. This process, along with editing the transcripts, aided in familiarisation with the data. Transcripts were then transferred into NVivo14 (Lumivero, 2023). The primary researcher then actively listened to each recording again, whilst keeping analytic memos using the 'memos' feature of NVivo14. Salient thoughts/feelings were reflected upon in supervision. The primary researcher then went through each transcript line by line coding sections; a code is a word or succinct sentence that captures the data's core message (Byrne, 2022). Coding initially remained close to the data, and both semantic and latent coding occurred. After each interview was coded, all initial codes were extracted from NVivo14 into Microsoft Word where they were clustered together into groups of similar concepts/experiences (see Appendix O). This process was completed collaboratively with the research team (EN, FG, WN). The primary researcher and PPI representative (WN) also met individually to discuss the clusters.

Through a process of distillation, the clustered codes were allocated into potential sub-themes which were named based on the contents of the codes. These sub-themes were then examined and categorised into themes. These themes and sub-themes were then transferred back into NVivo14 and reviewed in relation to the whole dataset. The primary researcher checked whether the themes identified captured the data, helped to answer the research question, whether the theme had enough meaningful data to support it, and whether it was coherent (Byrne, 2022). After further reviewing and changing, five themes emerged. When naming themes, Braun and Clarke's advice (2013; 2020) that theme names should be evocative and creative, whilst demonstrating the important aspect of the theme, was kept in mind.

Ethical Considerations

Informed consent was gained for study participation through the procedure detailed in Chapter Four and participants' withdrawal rights were made explicit (see consent forms in

Appendix J and K). In terms of confidentiality, all identifiable information, such as name and date of birth, were removed from the study data. Researchers aimed to ensure that enough detail was removed from qualitative accounts to make it unidentifiable. However, as individuals offered their personal accounts, and the CH community is small, it was not possible to ensure complete confidentiality. This was clearly stated on the information sheet. This was also why participants were given the opportunity to read over their transcript before it was included in data analysis. Of course, even if qualitative accounts were removed, the researcher remembered the information, and this may have influenced the study results inadvertently. Again, this was made clear to potential participants prior to study participation.

Participants were not deceived throughout the research process. However, the interview focused on difficult experiences, which could have resulted in distress. The interviewer (HW), who was a trainee clinical psychologist, used their clinical skills to manage this. As there is thought to be a high rate of suicidal ideation in this group (Rossi et al., 2018), the researcher was aware of holding risk in mind throughout the study. The research team also had the contact details of all participants' General Practitioners to ensure participant safety. Fortunately, this was not deemed necessary. At the end of active participation, participants were emailed a debrief form which included various support details (See Appendix L).

The University of East Anglia's (UEA) data protection regulations were followed throughout the study. Email addresses, for individuals who expressed interest in the study after the sample size had been reached, were deleted. All the interview, questionnaire, and participant data collected was secured safely on a password protected computer on a UEA OneDrive. Identifiable information was kept separate to interview/questionnaire data. Only the direct research team had access to this data. The PPI representative was not granted

access. This was deemed appropriate as the CH community is small, and as they are actively involved in the community, the chance of them recognising a participant was considered too high. Interview recordings were deleted as soon as transcripts were completed and verified by participants. Participants were given the option of receiving a copy of the published thesis when complete, and all opted for this. Therefore, their contact details were stored securely on a locked OneDrive folder after the interview process so the published research could be sent to them.

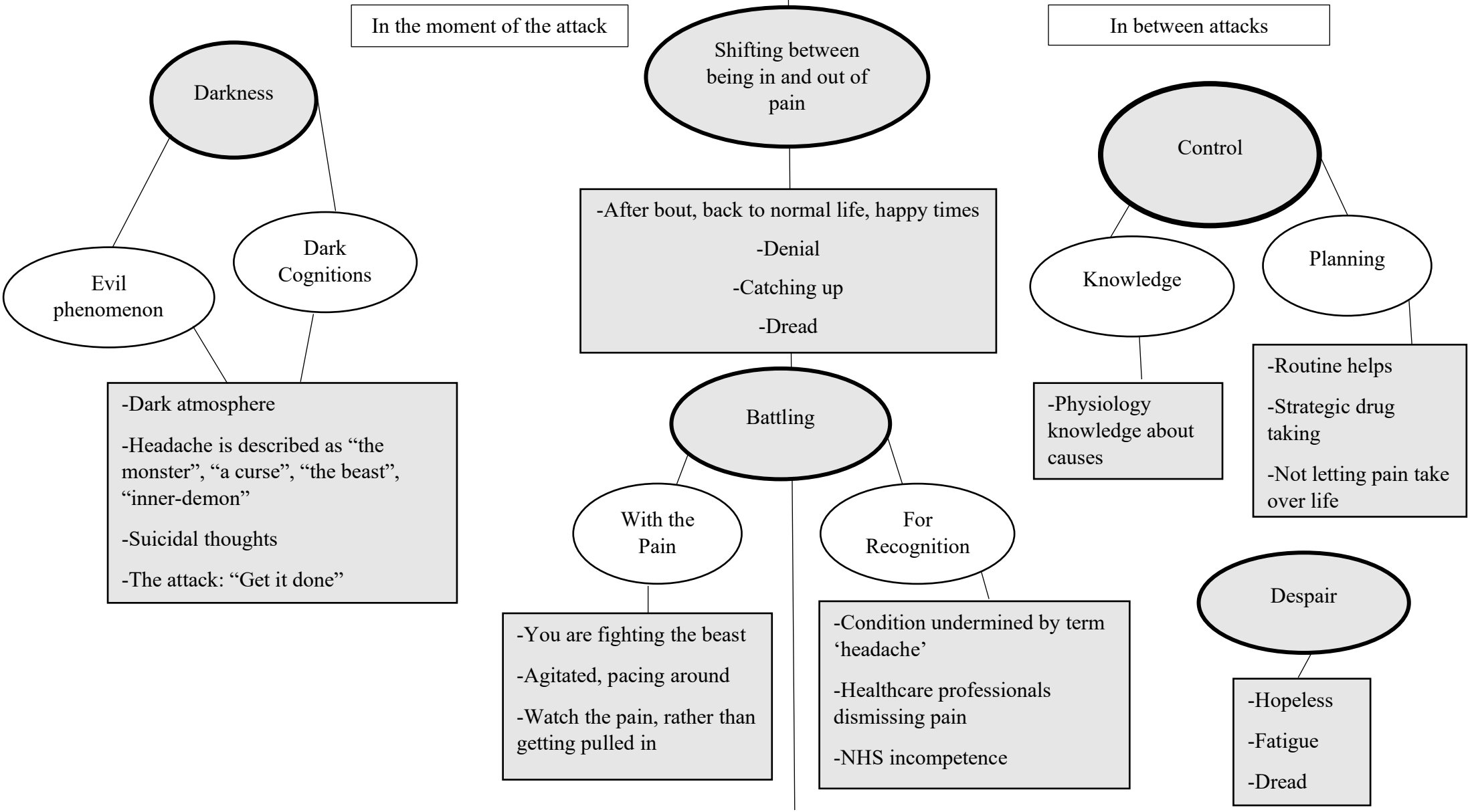
It was deemed appropriate to offer payment, because the BPS Code of Ethics (2023) states that payment should be offered when participants are giving up substantial time; which was the case. £10 was chosen due to budget constraints and consideration of not wishing to offer too high an amount which could incentivise individuals to participate.

Thematic Map

See in Figure 9, a thematic map with example codes for each theme and sub-theme.

Figure 9

Thematic Analysis Map with Codes



Note. Grey circles are the main themes, white circles are the sub-themes, grey boxes are example codes

Reflection from the PPI Representative

"I feel that having the opportunity to initiate and be involved in the research of the Psychological effects of Cluster Headaches, has given me the opportunity to use my experience of Chronic Cluster Headaches, where life literally stops due to pain, thorough exhaustion, and where my mental health fell to the deepest of lows, to a positive effect. In a way, my involvement gives the whole harrowing experience a positivity which I would not otherwise have had, as well as taking the fight to the condition, rather than being on the receiving end". (Wayne Nolan, 19th February 2025).

Chapter Six: General Discussion and Critical Evaluation

This final chapter will summarise the findings of the whole thesis portfolio; bringing together the systematic review and empirical paper and critically evaluating the piece of work. Recommendations for clinical practice and ongoing research will then be presented.

Overall Findings

The purpose of this thesis was to gain a greater understanding into psychological aspects of CH. The systematic review and meta-analysis explored depression and suicidality symptoms in CH individuals compared to non-headache controls and individuals with other primary headache conditions. The review identified 20 studies relevant to this question, and four meta-analyses were conducted. The findings from these meta-analyses were that CH had higher levels of depressive symptoms and suicidality than individuals without headaches, and the effect sizes were large. These findings remained stable through sensitivity analyses. Compared to adults with Migraines and Tension-Type Headaches (TTH), CH adults showed no significant difference in depression symptoms. This remained a stable finding when sensitivity analysis was conducted for the Migraine meta-analysis, but when an outlier was removed for the TTH meta-analysis, TTH individuals had significantly higher rates of depression than CH individuals and this effect was moderate. These results could be argued to provide evidence that depression and suicidality are raised in CH individuals compared to non-headache individuals but not compared to those with other primary headache conditions, and TTH individuals may have higher depression symptoms. However, results should not be overstated due to high rates of heterogeneity, publication bias, and for some of the meta-analyses the small number of studies involved.

The empirical paper resulted in five themes emerging as psychological factors relevant to the CH experience: “Darkness”, “Battling”, “Shifting”, “Control”, and “Despair”. The study findings could be argued to support the findings of the systematic review, with

several participants indicating that throughout an attack of pain they had suicidal thoughts, and many mentioning feelings of hopelessness, despair, and depression. Importantly, for the interviewees, suicidal ideation was only present throughout the acute attack, and there were no suicidal behaviours reported. Further, despair and depression seemed to mainly occur for CCH sufferers or when ECH individuals were in a bout of attacks; indeed, ECH sufferers often reported being happy and free out of bout season. Furthermore, as the course of the condition progressed, many interviewees seemed to adopt a more optimistic perspective. Such a change in perspective seemed to occur due to aspects such as gaining a diagnosis, a routine to manage the attacks, and the knowledge that they are safe. Having these qualitative findings adds depth to the quantitative findings reported in the systematic review, highlighting the benefits of both study methods. The interview study allowed for additional level of detail regarding subtle varied aspects of the psychological response dependent on context such as; in or out of the painful attack, being in a bout or not, duration of condition, and whether one had ECH or CCH.

Strengths and Limitations

Strengths of the overall project are that, to the authors' knowledge, this portfolio was the first to systematically review research focused on rates of depression and suicidality amongst CH adults and to explore the psychological factors of the experience qualitatively. This makes it a novel, arguably worthy, thesis, especially as it was proposed by an individual with lived experience.

The PPI aspect of the study is a key strength of this thesis portfolio. The PPI representative was an active member in every research meeting, with multi-directional learning continually occurring between academic researchers and the PPI member. Input from a PPI representative was invaluable to ensure the research met the needs of the CH community (UKIR, 2022). As the primary researcher came from a psychologically-minded

lens, but without personal experience of head pain, it felt important to have the PPI representative to provide personal accounts and prevent the researcher from solely viewing the research data with psychological theory in mind. PPI is not a simple process; with it often being tokenistic in nature (Soklaridis et al., 2024). The primary researcher was mindful of this and so tried to make the research meetings feel psychologically safe, encouraging genuine collaboration. Overall, all authors felt that 'contribution' level PPI was achieved (Sweeney & Morgan, 2009) and viewed it as the most valuable part of the research process.

Whilst a huge strength of the project, the involvement of a PPI representative throughout did come with challenges and involved navigating power relations (Soklaridis et al., 2024), which the primary author (HW) reflected on with the PPI representative (WN) and their research supervisors (EN and FG). For example, throughout the thesis process the primary author raised the issue of the PPI representative being paid for their time and this was arranged to ensure their significant time was reimbursed. A further consideration was that having a PPI representative did add time to the research process. This was because multi-directional learning meant that meetings were often longer to allow opportunities for explanation from both parties. This was particularly true for meetings related to the systematic review, which included statistical concepts. As the PPI was not directly involved with the systematic review, the research team reflected subsequently on whether the PPI should have attended these meetings. A final consideration relates to communication outside of meetings via emails. The PPI's passion for the area, which was so helpful, did also result in frequent lengthy emails, which were time-consuming for the primary researcher to respond to. Reflecting back, authors feel that research involving a PPI representative should involve the creation of a contract in the first research supervision where all parties discuss their expectations of the PPI's role. This could include the time commitment of the PPI, which

meetings the PPI would attend, and an arrangement of appropriate contact outside of research meetings. See the PPI's reflections of the experience in Chapter Five.

Another strength of this thesis is that research attempted to distinguish between individuals with CCH and ECH which is unlike most research in the field. This is important, as both the systematic review and qualitative analysis identified that the sub-types may involve different psychological aspects. Indeed, the two studies included in the systematic review which reported data on individuals with ECH and CCH separately, reported rates of depression and suicidality were higher for CCH (Jürgens et al., 2011; Torkamani et al., 2015). Jürgens and colleagues (2011) reported the odds ratio of having depression in the CCH group versus non-headache group was 5.21:1, whereas for ECH versus non-headache controls the odds ratio was only 1.50:1. This study also compared CH individuals to individuals with Migraine and reported that individuals with CCH had over three times more odds of developing depression, whereas ECH individuals had only 0.90 increased odds of developing depression compared to individuals with Migraine. Similarly, the interview study detailed in Chapter Four, highlighted that individuals with CCH did not have periods of being pain-free and so had less opportunities to get back to engaging with their lives, unlike ECH individuals. This was associated with feelings of despair amongst CCH individuals. As research most frequently combines data on episodic and chronic CH, authors wonder whether the true negative experience for chronic sufferers is being under-estimated. CCH sufferers may struggle more than other primary headache conditions; however, this was not evidenced by the current systematic review.

Despite thesis portfolio strengths, the piece of work did have limitations. Firstly, both the systematic review and the empirical paper were focused on participants in the northern hemisphere. The limited research in countries south of the equator is something previously highlighted (Hoffman & May, 2018) and this thesis was not able to counter this. These results

therefore cannot necessarily be applied globally, as how pain is perceived, expressed, and treated within a society varies significantly depending on cultural beliefs and social norms (Okolo et al., 2024; Yoshikawa, et. al., 2020). Some research has even found pain thresholds to be influenced by cultural beliefs (Mathur et al., 2022). The importance of cultural beliefs came through the data in the present interview study. For example, several participants referred to religion in terms of a Christian God and one participant reflected on the influence of CH being seen as a male disorder in their perceived patriarchal society. These culturally based ideas/beliefs could have impacted participants' distress levels and so results cannot necessarily be applied to different cultures.

Another weakness of the thesis portfolio was that it presented largely cross-sectional research. The systematic review only included one longitudinal study, meaning it was difficult to establish causal relationships between headache diagnosis and depression/suicidality (Wang & Cheng, 2020). The interview study was also cross-sectional. Interviewees did refer to their change in experience over time, offering some insights into the temporal experience of CH, but this was all reported at one time-point; meaning recall bias may have impacted results (Althubaiti, 2016).

Clinical Implications

This thesis has highlighted the psychological aspects of the CH experience. Both the systematic review and empirical paper highlighted the fact that the condition can be associated with suicidal ideation during the attack and feelings of depression, despair, and hopelessness throughout bout seasons. Therefore, it could be suggested that those with a diagnosis of CH should routinely be screened for depression and undertake risk assessments. At the present time, there is limited access to psychological support embedded within headache services. Therefore, whilst screening is desirable, a more realistic clinical implication could involve training to healthcare professionals in headache services to make

them aware of the psychological impact of CH. Clinicians would then be able to consider whether referral to existing services, such as Talking Therapies, may be appropriate.

The empirical paper identified the complex psychological experience which accompanies the CH condition, and highlighted potential processes maintaining distress which could be targets for psychological treatment. Processes identified as potentially maintaining distress were highlighted by capturing how participants who had had CH for many years gradually changed their response to the condition to make it easier to live alongside. Specifically, they reported trying to relax throughout an episode of pain, adapting their cognitions to being less negative towards the condition, and trying to feel in control through becoming knowledgeable about the condition and planning social events, whilst trying to continue engaging with life. Authors proposed that this could suggest that fighting against the pain, avoidance, negative cognitions, and lack of control may maintain distress of the CH experience. Further research should explore these psychological processes in detail, with the view to then conducting rigorous research, such as randomised controlled trials (RCTs), focused on psychological treatments for CH. Once the evidence-base is established, NICE guidelines could be adapted to include a psycho-behavioural element. Making such changes would bring the NICE guidelines in line with those for other primary headache conditions (NICE, 2022; 2024). The reason for lack of such recommendations currently is likely due to the lack of research in the area; with no previous systematic review being conducted to bring CH literature together. Also, authors wonder whether the reflections of one of the interviewees in the empirical study could offer an explanation. Participant 4, who was a healthcare professional, reflected that the condition is seen as a male condition in the medical world. A recent review of chronic pain reported that men are generally seen as being stoic and brave in relation to pain, whereas females are viewed as being more emotional and sensitive to pain (Samulowitz et al., 2018). Therefore, perhaps the association between CH

being a male condition could have resulted in management all being focused on medical approaches. It is hoped this thesis has highlighted the need for a psychological lens on this condition; so, supporting the "battle" towards individuals getting the support they need.

Next Steps

This study has provided information regarding what processes may be salient in the CH experience. Going forwards, authors encourage research to determine the relative contributions of these processes and what contextual factors may influence them. Once identified, psychological techniques to support individuals with CH can be explored. For example, studies could trial a tailored course of traditional Cognitive Behavioural Therapy (CBT) with Acceptance Commitment Therapy (ACT), and Dialectical Behavioural Therapy (DBT) components. Authors propose these specific therapies as potential options due to them targeting processes mentioned above as potentially maintaining distress for CH individuals, Timing of treatment should be explored to ascertain the most appropriate time for treatment e.g., for ECH sufferers, in bout season or out of bout season. Research should differentiate between CCH and ECH individuals and explore their psychological experience separately. To enhance the robustness of research findings, longitudinal research is warranted to explore the direction of the relationship between CH and psychopathology. Furthermore, authors encourage research to be conducted in the southern hemisphere to understand the experience of the condition from a greater variety of cultures. Finally, and perhaps most importantly, authors would encourage research going forward to include PPI throughout. This is encouraged, because it allows for authentic and important research to be carried out, in the most appropriate way. When conducting PPI authors encourage use of a contract in the initial research meeting to ensure this involvement meets the needs of all research members.

Speculative Discussion

Research must be conducted to assess the appropriateness of psychological treatment, however certain techniques will now be speculated upon. These speculations are based on psychological processes identified by this thesis portfolio to potentially be maintaining distress, predominantly from the empirical interview study, as well as pre-existing literature. From these identified psychological processes, potential mechanisms of change will be discussed. Authors propose that treatment should perhaps be separated into in-the-moment of pain and between attacks. This is because when in an acute attack, individuals have limited cognitive capacity, and so strategies which do not require such skills are likely preferential; whereas between attacks capacity is available and so strategies requiring more cognitive abilities are an option. These hypotheses should be explored by future research.

A common theme in the moment of pain related to participants viewing the CH as a dark, separate entity to themselves; calling it terms such as 'the monster'. The use of dark language in the present study is not a new phenomenon. Vera (2012) reported on metaphorical language used in medicine in England between the 14th and 16th century, with illness often being described as living with ill intentions, and physicians "fighting" against it. This is related to the psychological technique of externalisation, which has been proposed to empower an individual and blame a separate part of them for their illness, so allowing maintenance of a coherent identity (Achenbach et al., 2016; Demjén et al., 2016). Such externalisation techniques have been used in both mental and physical illnesses (Achenbach et al., 2016; Demjén et al., 2016). Whilst research is warranted, this could suggest that following CH diagnosis, individuals can be supported to create their own external understanding of their condition to transform how they relate to the pain and reduce distress.

One consideration with the proposed externalisation strategy comes from a body of research highlighting the issue with talking of an illness as an enemy. Using warmongering, catastrophic, language can result in a feeling of civil war against oneself and subsequently

being in a state of fight or flight, and psychological distress (Johnson et al., 2023). Bury (1991) determined that within a context, illness comes with different symbolic connotations and imagery. Within the UK context, CH is colloquially known as “the worst pain known to man” and "the suicide headache” (Burish et al., 2021; Grinberg et al., 2021). Perhaps this has unhelpfully perpetuated catastrophic, dark, ways of thinking about CH. Terminology which is less catastrophic such as “navigating” rather than “fighting” the pain and viewing pain as a companion, has been advocated, to try to get to a place of “re-embodiment” (Williams, 1996b). The suggestion of changing the imagery associated with the acute pain attack has similarities to imagery re-scripting and art therapy which have been used when working with trauma to make experiences more cohesive and less distressing (Brown et al., 2023; Wang et al., 2025). Authors reflect that CH individuals insightfully alluded to these critical sentiments. For example, Participant 7 in the empirical paper stated about the CH pain “I don't hate the monster. Erm because all I'm doing is hating myself”. Overall, externalising CH may be helpful for individuals to maintain a coherent sense of self but creating a neutral or positive image is perhaps favourable.

The theme “Battling” related to fighting against the pain through becoming agitated and pacing around. The qualitative study identified various ways in which CH sufferers tried to manage their condition over time. This included several participants in the interview study who referred to no longer battling with their pain throughout an attack, but instead trying to relax, saying mantras to keep calm, breathing deeply, and separating themselves from the pain. These concepts are arguably similar to aspects of Dialectical Behavioural Therapy (DBT) which was originally created for individuals with emotion dysregulation in crisis (Linehan, 1993). DBT involves teaching distress tolerance grounding techniques for moments of crisis/dissociation, such as being mindful and paired breathing (Linehan, 2014). Whilst there is no evidence regarding these techniques in CH patients, research related to pain

management in childbirth could apply, as the pain levels have been indicated to be comparable (Burish et al., 2021). A systematic review (Wang et al., 2024), focused on non-pharmacological approaches to managing pain throughout labour, reported that techniques such as relaxation, breathing, and mindfulness were effective strategies, with the mindfulness strategy showing a large effect. Techniques from Acceptance Commitment Therapy (ACT) may also be relevant to consider. ACT is a well-established treatment within chronic pain (Feliu-Soler et al., 2018). This modality proposes the concept of psychological flexibility, which is a willingness to continue acting in accordance with one's values, whilst being open and aware of difficult thoughts and feelings (Hayes & Pierson, 2005). Psychological flexibility is made up of six interrelated cognitive processes. One such process is *acceptance,* which is a willingness to sit with difficult feelings and not fight against them (McCracken & Morley, 2014). This resonates with participants' experience of not battling with the pain. Another ACT process is *self as observer* which involves viewing oneself as separate from difficult experiences (McCracken & Morley, 2014), which again some interview participants referred to. Taken together, this could suggest psychological techniques similar to DBT and ACT could be explored as a potential avenue for treatment during a CH attack; with individuals being trained in these strategies prior to the attack.

Another theme related to "Shifting" and complex identity struggles. A psychological technique which could be beneficial relates to illness identity. Bury (1982) described chronic illness as a "biographical disruption" in which one's normal daily activities are impacted, one's self-narratives are changed, and one's bodily states are brought into focus. Adjusting to chronic illness in general can be a psychologically challenging experience with individuals often becoming engulfed by the illness, or rejecting it (Oris et al., 2016). A more adaptive approach is to come to accept the illness, whilst not losing one's identity outside of illness, and trying to maintain a coherent self (Bury, 1991). The empirical paper identified that over

time individuals often seemed to come to a place of acceptance. If individuals could be supported to incorporate their illness identity earlier, they may adhere to treatment regimes more consistently and benefit from a coherent sense of self. Furthermore, research focused on individuals with chronic heart conditions reported that coming to accept illness within one's identity was related to reduced depression (Van Bulck et al., 2019). As depression was identified in the systematic review (Chapter Two) to be heightened in CH individuals, this is a further reason to support CH individuals to accept their illness identity. Authors wonder about the identity shift differences between episodic and chronic CH individuals; when the former return to 'normal' life between bouts, and the latter live with the condition continually. Some individuals with ECH spoke of their life stopping throughout bout season (like engulfment?) and denying their CH out of bout season (like rejection?) It would be interesting to explore whether the process of acceptance/enrichment takes longer for episodic sufferers.

The empirical paper also identified that interviewees sought to feel more in control of their experience. This came from them trying to become knowledgeable about their condition, to increase self-efficacy. Several participants also sought control through avoiding situations which could trigger pain. However, over time many learnt to try and find a balance between planning for attacks whilst not letting the condition stop them engage in important aspects of their lives. Participants in the interview study also referred to the despair and ruminative thinking they experienced. Again, some participants referred to the change over time to thinking more positively and less catastrophically, albeit this being difficult when drained and tired after multiple attacks. All these psychological changes could be compared to traditional Cognitive Behavioural Techniques (CBT) such as psychoeducation, behavioural activation, exposure therapy, and cognitive restructuring (Beck & Emery, 2005). Participants

in this study came to these psychological changes over years of living with the condition. Perhaps if these skills could be learnt earlier, following diagnosis, this could reduce suffering.

The role for CBT for CH individuals may be supported by existing systematic reviews which have showed that CBT can reduce disability for patients with chronic pain (İnce, 2020), including Migraine patients (Bae et al., 2021). As stated by interviewees in the empirical paper, CH is not comparable to Migraines and therefore results cannot necessarily be extended. However, Migraine studies have found that concepts such as pain catastrophising and low self-efficacy can result in behavioural avoidance, and increased disability (Yu & Tan, 2024), so there may be similarities in the psychological processes maintaining distress in both primary headache conditions. Furthermore, a small feasibility study which explored CBT in patients with Trigeminal Neuralgia, a condition similar to CH in symptomology, found that CBT improved self-efficacy, reduced pain catastrophising, and increased engagement in life (Daniel et al., 2021). This tailored CBT programme included psychoeducation regarding pain mechanisms of Trigeminal Neuralgia, mindfulness-based stress reduction, values-based goal setting, pacing, working with unhelpful thoughts, managing distressing emotions, and improving communication skills (Daniel et al., 2021). These cognitive and behavioural targets are similar to the psychological aspects reported by the current thesis portfolio to be relevant for CH, and so could provide preliminary evidence for the proposal of such tailored CBT for CH.

Finally, the theme “Battling” also included a desire for more recognition and support from others. As well as working with healthcare professionals to raise awareness, authors propose assertiveness work with CH individuals to support them to get their needs met. This has similarities to DBT which includes lessons around interpersonal assertiveness skills (Linehan, 1993). Furthermore, the small feasibility study which explored CBT in patients

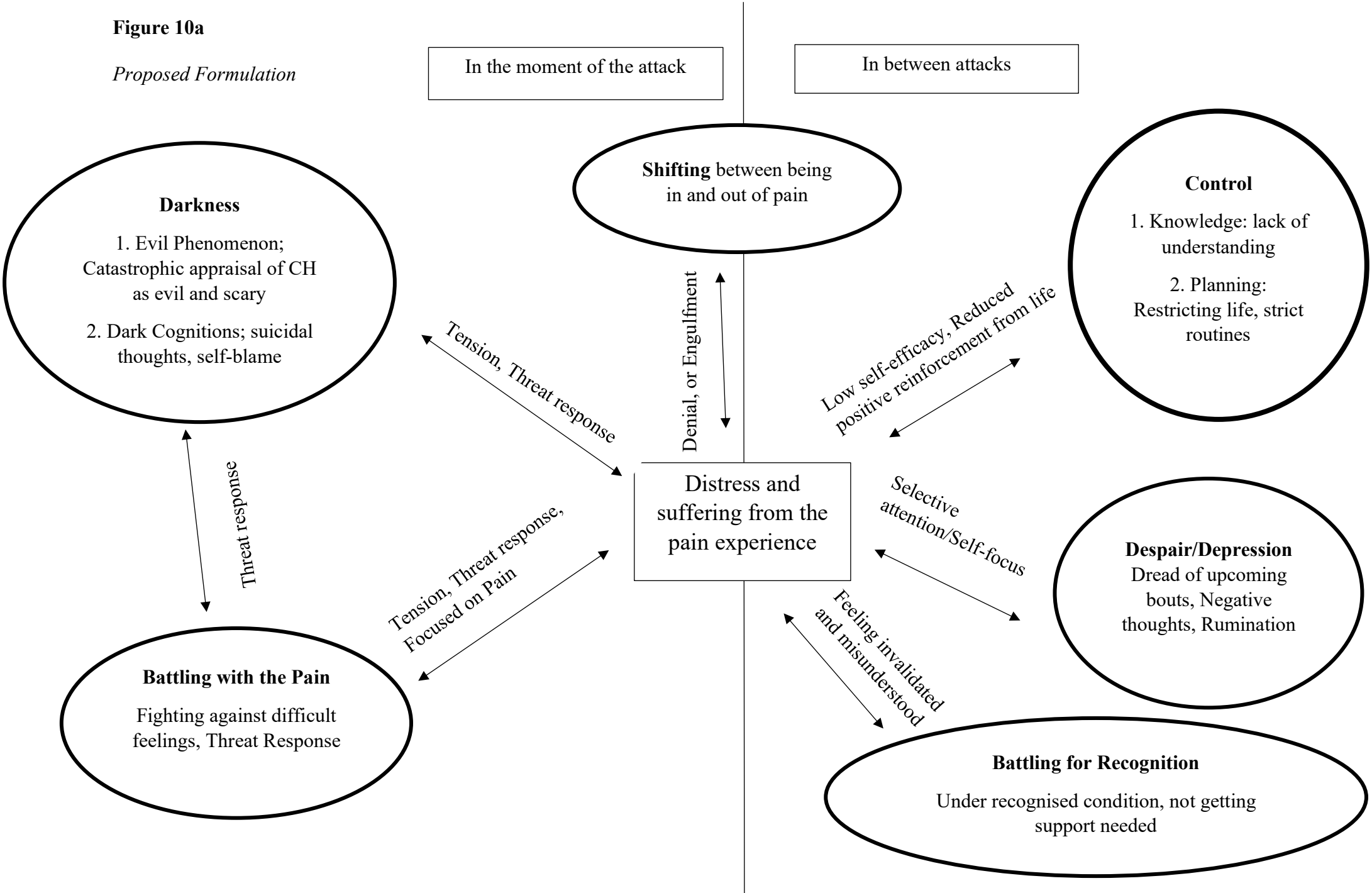
with Trigeminal Neuralgia, included developing communication skills for when speaking with healthcare providers and significant others (Daniel et al., 2021).

Proposed Formulation

See Figure 10a below for a formulation which has been created drawing together the ideas from the thesis portfolio, particularly the empirical paper, into an integrative model. The psychological processes which may be maintaining distress are detailed by the arrows.

Figure 10a

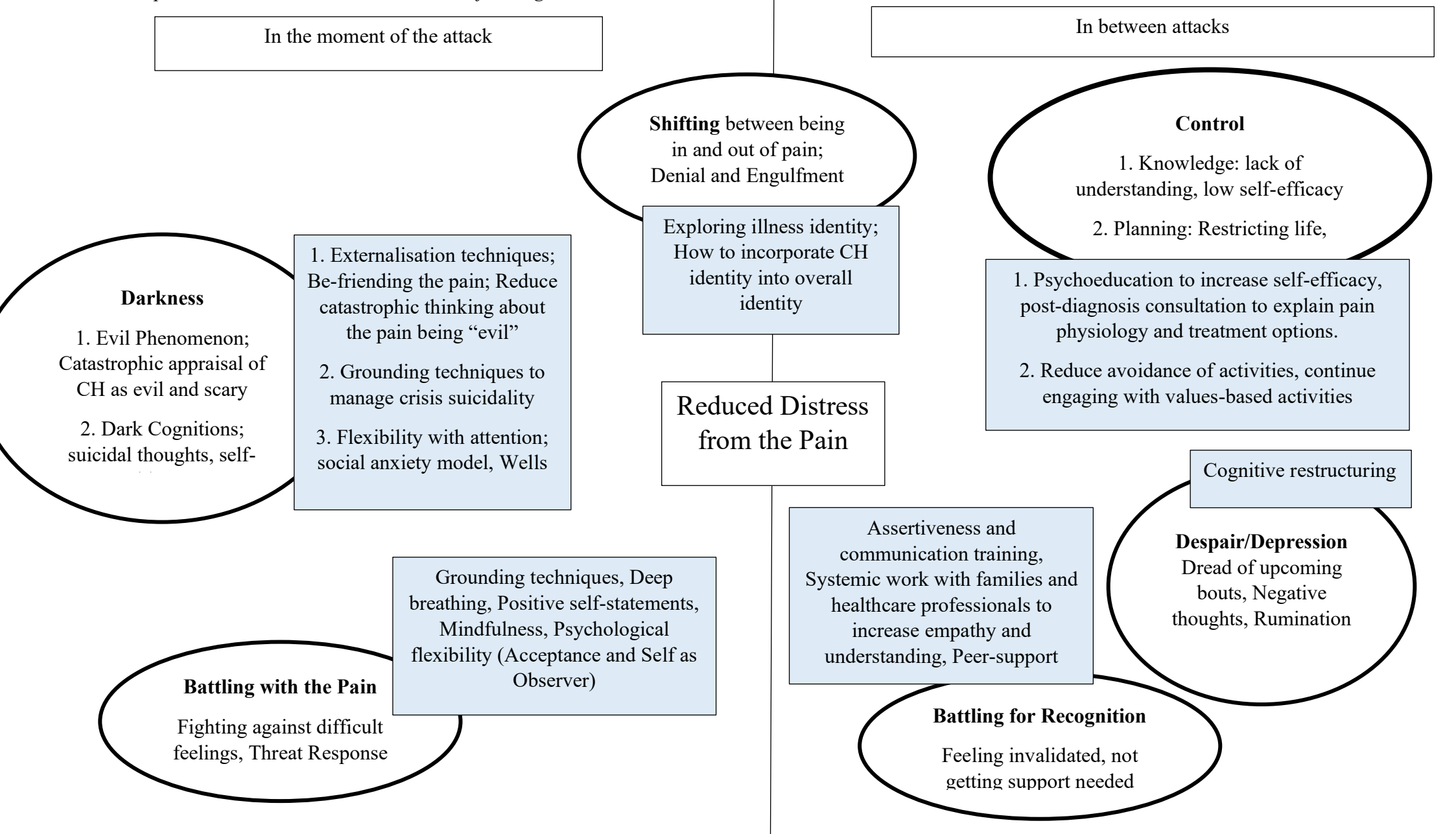
Proposed Formulation



See Figure 10b below for proposed mechanisms of change to break the feedback loops and reduce distress and suffering from CH pain.

Figure 10b

Proposed Formulation with Mechanisms of Change



For all potential psychological interventions, timing the treatment is likely an important consideration. Throughout a period of frequent bouts, individuals report feeling tired and drained, perhaps making therapy more difficult. The authors support speculations that psychological support could involve treatment in a head-ache free period in preparation for attacks (Schenck & Andrasik, 2019). However, authors wonder about motivation outside of bout season when individuals may try to catch up on life and deny their condition. Furthermore, if they have therapy and then no bout for a long period, their skills may decay before practical application. As bout seasons are often predictable, Schenck and Andrasik (2019) proposed that individuals contact healthcare providers a short time before their bout is due, to receive an intensive course. Throughout the bout, the clinician could then remain "on call" to support with implementation of techniques. This is an interesting idea, and whilst expensive and likely difficult to adopt in the NHS, worth exploring further. It holds resonance to DBT for emotion dysregulation which has an intensive program and then on-call clinicians, which has been found to be effective and is offered on the NHS (Linehan, 1993; NICE, 2009). Of course, chronic sufferers do not have the luxury of long pain-free periods and so this may be trickier, with treatment timing needing to be individualised. This provides further reasons to consider ECH and CCH sufferers as separate. **Conclusion**

This thesis portfolio explored the psychological aspects relevant to the experience of CH. A systematic review and meta-analysis of 20 studies concluded that individuals with CH have higher rates of depression and suicidality than individuals without head pain. Depression rates were largely comparable to those with other primary headaches, and individuals with TTH may have higher levels of depression than CH individuals. However, these conclusions should not be over-stated due to high heterogeneity, publication bias, and the risk-of-bias of much reviewed research. A reflexive thematic analysis of interview transcripts from 13 individuals with CH resulted in five themes emerging: "Darkness",

"Battling", "Shifting", "Control", and "Despair". From these themes, authors proposed that psychological processes, such as catastrophic interpretations of the pain, fighting against the pain, incoherent sense of self, low self-efficacy, rumination, avoidance of activities, and lack of recognition from others, may exacerbate distress for CH individuals. Psychological strategies such as positive externalisation, grounding techniques, psychological flexibility, identity work, psychoeducation, cognitive restructuring, behavioural activation, and communication training, may be helpful. Authors propose different techniques may be helpful at different points of the CH condition i.e. either within or between an attack. Both the systematic review and empirical study highlighted the distinct difference between chronic and episodic forms of CH and so a key implication from this thesis portfolio is that the two subtypes should be researched separately going forwards. Research should explore these ideas using PPI-directed research with the view to supporting individuals with CH in a more multidisciplinary way. Overall, this thesis portfolio has proposed novel insights into the psychological aspects of CH and offers a foundation for future research to explore how psychological approaches could improve the lives of those with CH.

Portfolio References

Abbas, A., Hefnawy, M. T., & Negida, A. (2024). Meta-analysis accelerator: a comprehensive tool for statistical data conversion in systematic reviews with meta-analysis. *BMC Medical Research Methodology, 24*(1), 243. https://doi.org/10.1186/s12874-024-02356-6

Achenbach, T. M., Ivanova, M. Y., Rescorla, L. A., Turner, L. V., & Althoff, R. R. (2016). Internalizing/externalizing problems: review and recommendations for clinical and research applications. *Journal of the American Academy of Child and Adolescent Psychiatry, 55*(8), 647-656. https://doi.org/10.1016/j.jaac.2016.05.012

Agius, S. J. (2013). Qualitative research: Its value and applicability. *Psychiatrist, 37*(6), 204-206. https://doi.org/10.1192/pb.bp.113.042770

Ahmed, F., Bahra, A., Tyagi, A. & Weatherby, S. (2019) *National headache management system for adults.* British Association for the Study of Headache. http://www.bash.org.uk

Althubaiti A. (2016). Information bias in health research: definition, pitfalls, and adjustment methods. *Journal of Multidisciplinary Healthcare*, *9*, 211-217. https://doi.org/10.2147/JMDH.S104807

Amiri, S., Behnezhad, S., & Azad, E. (2019). Migraine headache and depression in adults: a systematic Review and Meta-analysis. Migräne und Depression bei Erwachsenen: Ein systematisches Review und Meta-Analyse. *Neuropsychiatrie : Klinik, Diagnostik, Therapie und Rehabilitation: Organ der Gesellschaft Osterreichischer Nervenarzte und Psychiater*, *33*(3), 131-140. https://doi.org/10.1007/s40211-018-0299-5

An, R., & Xiang, X. (2015). Smoking, heavy drinking, and depression among U.S. middle-aged and older adults. *Preventive Medicine, 81,* 295-302. https://doi.org/10.1016/j.ypmed.2015.09.026

Anagnostou, E., Vikelis, M., Tzavellas, E., Ghika, A., Kouzi, I., Evdokimidis, I., & Kararizou, E. (2017). Photophobia in primary headaches, in essential blepharospasm and in major depression. *The International Journal of Neuroscience*, *127*(8), 673-679. https://doi.org/10.1080/00207454.2016.1231185

Andersson, M., Persson, M., & Kjellgren, A. (2017). Psychoactive substances as a last resort-a qualitative study of self-treatment of migraine and CHs. *Harm Reduction Journal, 14*(1), 60. https://doi.org/10.1186/s12954-017-0186-6

Andre, L., & Cavers, D. (2021). 'A cry in the dark': a qualitative exploration of living with cluster headache. *British Journal of Pain, 15*(4), 420-428. https://doi.org/10.1177/2049463720976695

Asghari, A., & Nicholas, M. K. (2001). Pain self-efficacy beliefs and pain behaviour. A prospective study. *Pain, 94*(1), 85-100. https://doi.org/10.1016/S0304-3959(01)00344-X

Asmundson, G., Vlaeyen, J., & Crombez, G. (2004). *Understanding and treating fear of pain.* In Oxford University Press eBooks. https://doi.org/10.1093/oso/9780198525141.001.0001

Bae, J. Y., Sung, H. K., Kwon, N. Y., Go, H. Y., Kim, T. J., Shin, S. M., & Lee, S. (2021). Cognitive behavioral therapy for migraine headache: a systematic review and meta-analysis. *Medicina (Kaunas, Lithuania), 58*(1), 44. https://doi.org/10.3390/medicina58010044

Ballesta-Martínez, S., Del Pilar Navarro-Pérez, M., Bellosta-Diago, E., & Santos-Lasaosa, S. (2022). Theory of mind: a new perspective on cluster headache-a cross-sectional study.

Neurological Sciences: Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology, *43*(10), 6047-6051. https://doi.org/10.1007/s10072-022-06234-1

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioral change. *Psychological Review, 84*(2), 191-215. https://doi.org/10.1037/0033-295X.84.2.191

Beck, A. T., Emery, G., & Greenberg, R. L. (2005). *Anxiety disorders and phobias: A cognitive perspective.* Basic Books/Hachette Book Group.

Beck, A. T., Rush, A., J., Shaw, B. F. (1979). *Cognitive therapy of depression.* New York: Guilford Press.

Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbauch, J. (1961). Beck Depression Inventory (BDI) [Database record]. *APA PsycTests.* https://doi.org/10.1037/t00741-000

Belin, A. C., & Barloese, M. C. (2023). The genetics and chronobiology of cluster headache. *Cephalalgia: an International Journal of Headache, 43*(10), 3331024231208126. https://doi.org/10.1177/03331024231208126

Benson, H., Klemchuk, H. P., & Graham, J. R. (1974). The usefulness of the relaxation response in the therapy of headache. *Headache, 14*(1), 49-52. https://doi.org/10.1111/j.1526-4610.1974.hed1401049.x

Bhaskar R. (2008). *A realist theory of science*. Routledge.

Bhaskar, R. (1998). Philosophy and scientific realism. In Archer M. S. (Ed.), *Critical realism: Essential readings.* Routledge.

Blanchard, E. B., Andrasik, F., Jurish, S. E., & Teders, S. J. (1982). The treatment of cluster headache with relaxation and thermal biofeedback. *Biofeedback and Self-regulation, 7*(2), 185-191. https://doi.org/10.1007/BF00998782

Braun, V., & Clarke, V. (2012). Thematic analysis. In H. Cooper, P. M. Camic, D. L. Long, A. T. Panter, D. Rindskopf, & K. J. Sher (Eds.), *APA handbook of research methods in psychology, Vol. 2. Research designs: Quantitative, qualitative, neuropsychological, and biological* (pp. 57-71). American Psychological Association. https://doi.org/10.1037/13620-004

Braun, V., Clarke, V. (2013). *Successful Qualitative Research: A Practical Guide for Beginners.* Sage Publications, Thousand Oaks

Braun, V., & Clarke, V. (2019). Reflecting on reflexive thematic analysis. *Qualitative Research in Sport, Exercise and Health, 11*, 589 - 597. https://doi.org/10.1080/2159676X.2019.1628806

Braun, V., & Clarke, V. (2020). One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology, 18,* 328-352. https://doi.org/10.1080/14780887.2020.1769238

Braun V., Clarke V. (2022). *Thematic analysis: A practical guide.* London.

Braun, V., & Clarke, V. (2022). Toward good practice in thematic analysis: Avoiding common problems and be(com)ing a *knowing* researcher. *International Journal of Transgender Health*, *24*(1), 1-6. https://doi.org/10.1080/26895269.2022.2129597

British Medical Journal, BMJ. (2024). *Cluster Headache.* https://bestpractice.bmj.com/topics/en-gb/11

British Pain Society. (2019). *Outcome Measures.* https://www.britishpainsociety.org/static/uploads/resources/files/Outcome_Measures_January_2019.pdf

British Psychological Society (BPS). (2023). *Ethics queries.* https://www.bps.org.uk/ethics-queries

Britten N. (1995). Qualitative interviews in medical research. *BMJ (Clinical research ed.), 311*(6999), 251-253. https://doi.org/10.1136/bmj.311.6999.251

Brown, G., Salter, C., Parker, E., Murray, H., Looney, K., El Leithy, S., Medin, E., Novakova, B., & Wheatley, J. (2023). Imagery re-scripting for PTSD: session content and its relation to symptom improvement. *Behavioural and Cognitive Psychotherapy, 51*(1), 1-10. https://doi.org/10.1017/S1352465822000479

Burish, M. J., Pearson, S. M., Shapiro, R. E., Zhang, W., & Schor, L. I. (2021). Cluster headache is one of the most intensely painful human conditions: Results from the International Cluster Headache Questionnaire. *Headache*, *61*(1), 117-124. https://doi.org/10.1111/head.14021

Burke, P. J., & Stets, J. E. (2009). *Identity theory.* Oxford, UK: Oxford University Press.

Bury, M. (1991). The sociology of chronic illness a review of research and prospects. *Sociology of Health & Illness, 13,* 451-468. https://doi.org/10.1111/j.1467-9566.1991.tb00522.x

Bury, M. (1982). Chronic illness as biographical disruption. *Sociology of Health & Illness*, *4*(2), 167-182. https://doi.org/10.1111/1467-9566.ep11339939

Buzzi, M. G., Pellegrino, M. G., & Bellantonio, P. (1995). Causes and mechanisms of primary headaches: toward a bio-behavioral model. Italian Journal of Neurological Sciences, 16(9), 15- 19. https://doi.org/10.1007/BF02333240

Byrne, D. (2022). A worked example of Braun and Clarke's approach to reflexive thematic analysis. *Quality and Quantity, 56,* 1391-1412. https://doi.org/10.1007/s11135-021-01182-y

Chang, B. H., & Hoaglin, D. C. (2017). Meta-analysis of Odds Ratios: Current good practices. *Medical Care, 55*(4), 328-335. https://doi.org/10.1097/MLR.0000000000000696

Charmaz, K. (1995). The body, identity, and self: adapting to impairment. *The Sociological Quarterly, 36*(4), 657-680. https://doi.org/10.1111/j.1533-8525.1995.tb00459.x

Chen, P. K., Fuh, J. L., Chen, S. P., & Wang, S. J. (2010). Association between restless legs syndrome and migraine. *Journal of Neurology, Neurosurgery, and Psychiatry*, *81*(5), 524–528. https://doi.org/10.1136/jnnp.2009.191684

Choong, C. K., Ford, J. H., Nyhuis, A. W., Joshi, S. G., Robinson, R. L., Aurora, S. K., & Martinez, J. M. (2017). Clinical Characteristics and Treatment Patterns Among Patients Diagnosed With Cluster Headache in U.S. Healthcare Claims Data. *Headache: The Journal of Head and Face Pain*, *57*(9), 1359-1374. https://doi.org/10.1111/head.13127

Chowdhury D. (2012). Tension type headache. *Annals of Indian Academy of Neurology, 15*(Suppl 1), S83-S88. https://doi.org/10.4103/0972-2327.100023

Cisneros-Puebla, C. A. (2007). Los rostros deconstructivo y reconstructivo de la construcción social. Kenneth Gergen en conversación con César A. *Cisneros-Puebla. Introducción de Robert B. Faux [83 párrafos]. Forum Qualitative Sozialforschung / Forum: Qualitative Social Research, 9*(1)

Clarke, A., Anthony, G., Gray, D., Jones, D., McNamee, P., Schofield, P., Smith, B. H., & Martin, D. (2012). "I feel so stupid because I can't give a proper answer…" How older adults describe chronic pain: a qualitative study. *BMC Geriatrics*, *12*, 78. https://doi.org/10.1186/1471-2318-12-78

Clinical Knowledge Summarises, CKS. (2024). *Headache- Cluster.* National Institute of Care Excellence (NICE). https://cks.nice.org.uk/topics/headache-cluster/background-information/prevalence/

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences, 2nd edition.* Lawrence Erlbaum Associates: Mahwah.

Crespi, J., Gulati, S., Salvesen, Ø., Bratbak, D.F., Dodick, D.W., Matharu, M.S. and Tronvik, E. (2022). Epidemiology of diagnosed cluster headache in Norway. *Cephalalgia Reports*, 5, 251581632210755. https://doi.org/10.1177/25158163221075569

Creswell, J.W., & Plano Clark, V.L. (2007). Designing and conducting mixed methods research.Thousand Oaks, CA: Sage

Cronin, S., Hardiman, O., & Traynor, B. J. (2007). Ethnic variation in the incidence of ALS: a systematic review. *Neurology, 68*(13), 1002-1007. https://doi.org/10.1212/01.wnl.0000258551.96893.6f

Cuevas-Toro, A. M., Torrecillas, F. L., Medina, A. G., & Diaz-Bata-nero, M. C. (2008). Personality and coping strategies in fibromyalgia patients. *Behavioral Psychology: Revista Internacional ClínicaY de La Salud, 16,* 289-306.

Cuijpers, P., Geraedts, A. S., van Oppen, P., Andersson, G., Markowitz, J. C., & van Straten, A. (2011). Interpersonal psychotherapy for depression: a meta-analysis. *The American Journal of Psychiatry, 168*(6), 581-592. https://doi.org/10.1176/appi.ajp.2010.10101411

Crombez, G., Morley, S., McCracken, L., Sensky, T., & Pincus, T. (2003). Self, identity, and acceptance in chronic pain. *Proceedings of the 10th World Congress on Pain*, 24, 651-659. https://www.researchgate.net/publication/230596566_Self_identity_and_acceptance_in_chronic_pain

D'Amico, D., Raggi, A., Grazzi, L., & Lambru, G. (2020). Disability, quality of life, and socioeconomic burden of CH: A critical review of current evidence and future perspectives. *Headache, 60*(4), 809-818. https://doi.org/10.1111/head.13784

Daniel, H. C., Poole, J. J., Klein, H., Huang, C., & Zakrzewska, J. M. (2021). Cognitive behavioural therapy for patients with trigeminal neuralgia: a feasibility study. *Journal of Oral & Facial Pain and Headache, 35*(1), 30-34. https://doi.org/10.11607/ofph.2664

Darnall, B. D., Sturgeon, J. A., Cook, K. F., Taub, C. J., Roy, A., Burns, J. W., Sullivan, M., & Mackey, S. C. (2017). Development and validation of a daily pain catastrophizing scale. *The Journal of Pain, 18*(9), 1139-1149. https://doi.org/10.1016/j.jpain.2017.05.003

Day, M. A., Ward, L. C., Thorn, B. E., Lang, C. P., Newton-John, T. R. O., Ehde, D. M., & Jensen, M. P. (2018). The Pain-Related Cognitive Processes Questionnaire: Development and Validation. Pain Medicine, 19(2), 269-283. https://doi.org/10.1093/pm/pnx010

DeJonckheere, M., & Vaughn, L. M. (2019). Semistructured interviewing in primary care research: a balance of relationship and rigour. *Family Medicine and Community Health, 7*(2), e000057. https://doi.org/10.1136/fmch-2018-000057

Dekkers, O. M., Vandenbroucke, J. P., Cevallos, M., Renehan, A. G., Altman, D. G., & Egger, M. (2019). COSMOS-E: Guidance on conducting systematic reviews and meta-analyses of observational studies of etiology. *PLoS Medicine, 16*(2), e1002742. https://doi.org/10.1371/journal.pmed.1002742

Delaruelle, Z., Ivanova, T. A., Khan, S., Negro, A., Ornello, R., Raffaelli, B., Terrin, A., Mitsikostas, D. D., Reuter, U., & European Headache Federation School of Advanced Studies (EHF-SAS). (2018). Male and female sex hormones in primary headaches. *The Journal of Headache and Pain, 19*(1), 117. https://doi.org/10.1186/s10194-018-0922-7

Demjén, Z. (2016). Laughing at cancer: Humour, empowerment, solidarity and coping online. *Journal of Pragmatics, 101*, 18-30. https://doi.org/10.1016/j.pragma.2016.05.010

Díaz-de-Terán, J., Sastre-Real, M., Lobato-Pérez, L., Navarro-Fernández, G., Elizagaray-García, I., & Gil-Martínez, A. (2021). Cluster headache, beyond the pain: a comparative cross-sectional study. *Neurological Sciences: Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology*, *42*(9), 3673-3680. https://doi.org/10.1007/s10072-020-04996-0

Dicicco-Bloom, B., & Crabtree, B. F. (2006). The qualitative research interview. *Medical Education, 40*(4), 314-321. https://doi.org/10.1111/j.1365-2929.2006.02418.x

Donnet, A., Lanteri-Minet, M., Guegan-Massardier, E., Mick, G., Fabre, N., Géraud, G., Lucas, C., Navez, M., Valade, D., & Société Française d'Etude des Migraines et Céphalées (SFEMC) (2007). Chronic cluster headache: a French clinical descriptive study. *Journal of Neurology, Neurosurgery, and Psychiatry*, *78*(12), 1354-1358. https://doi.org/10.1136/jnnp.2006.112037

Dubé, M. O., Langevin, P., & Roy, J. S. (2021). Measurement properties of the Pain Self-Efficacy Questionnaire in populations with musculoskeletal disorders: a systematic review. *Pain Reports, 6*(4), e972. https://doi.org/10.1097/PR9.0000000000000972

Ekbom, K., Svensson, D. A., Träff, H., & Waldenlind, E. (2002). Age at onset and sex ratio in cluster headache: observations over three decades. *Cephalalgia: an International Journal of Headache, 22*(2), 94-100. https://doi.org/10.1046/j.1468-2982.2002.00318.x

Elbadawi, A. S. A., Albalawi, A. F. A., Alghannami, A. K., Alsuhaymi, F. S., Alruwaili, A. M., Almaleki, F. A., Almutairi, M. F., Almubaddil, K. H., & Qashqari, M. I. (2021).

Cluster Headache and Associated Risk Factors: A Systemic Review and Meta-Analysis. *Cureus, 13*(11), e19294. https://doi.org/10.7759/cureus.19294

Evers, S., Frese, A., Majewski, A., Albrecht, O., & Husstedt, I. W. (2002). Age of onset in cluster headache: the clinical spectrum (three case reports). *Cephalalgia: an International Journal of Headache, 22*(2), 160-162. https://doi.org/10.1046/j.1468-2982.2002.00320.x

Feliu-Soler, A., Montesinos, F., Gutiérrez-Martínez, O., Scott, W., McCracken, L. M., & Luciano, J. V. (2018). Current status of acceptance and commitment therapy for chronic pain: a narrative review. *Journal of Pain Research, 11,* 2145-2159. https://doi.org/10.2147/JPR.S144631

Fischera, M., Marziniak, M., Gralow, I., & Evers, S. (2008). The incidence and prevalence of cluster headache: a meta-analysis of population-based studies. *Cephalalgia: an International Journal of Headache, 28*(6), 614-618. https://doi.org/10.1111/j.1468-2982.2008.01592.x

Fishman, D. B. (1999). *The case for pragmatic psychology.* New York University Press.

Flor, H., Birbaumer, N., Schugens, M. M., & Lutzenberger, W. (1992). Symptom-specific psychophysiological responses in chronic pain patients. *Psychophysiology, 29*(4), 452-460. https://doi.org/10.1111/j.1469-8986.1992.tb01718.x

Freeman, E., Adair, M., Beeler, D., Casper, R., Herman, M., Reeves, D., & Reinsch, S. (2022). Patient identified burden and unmet needs in patients with CH: An evidence-based qualitative literature review. *Cephalalgia Reports, 5,* 1. https://doi.org/10.1177/25158163221096866

Fumal, A., & Schoenen, J. (2008). Tension-type headache: current research and clinical management. *The Lancet. Neurology, 7*(1), 70-83. https://doi.org/10.1016/S1474-4422(07)70325-3

Gambadauro, P., Carli, V., & Hadlaczky, G. (2019). Depressive symptoms among women with endometriosis: a systematic review and meta-analysis. *American Journal of Obstetrics and Gynecology*, *220*(3), 230-241. https://doi.org/10.1016/j.ajog.2018.11.123

Gatchel, R. J., Peng, Y. B., Peters, M. L., Fuchs, P. N., & Turk, D. C. (2007). The biopsychosocial approach to chronic pain: scientific advances and future directions. *Psychological Bulletin*, *133*(4), 581-624. https://doi.org/10.1037/0033-2909.133.4.581

Geertz, C. (1973). *The interpretation of cultures: Selected essays.* New York: Basic Books.

Gesztelyi, G., & Bereczki, D. (2006). Determinants of disability in everyday activities differ in primary and cervicogenic headaches and in low back pain. *Psychiatry and Clinical Neurosciences*, *60*(3), 271-276. https://doi.org/10.1111/j.1440-1819.2006.01501.x

Gil-Martínez, A., Navarro-Fernández, G., Mangas-Guijarro, M. Á., & Díaz-de-Terán, J. (2019). Hyperalgesia and central sensitization signs in patients with cluster headache: a cross-sectional study. *Pain Medicine (Malden, Mass.)*, *20*(12), 2562-2570. https://doi.org/10.1093/pm/pnz070

Given, L. M. (2008). *Narrative inquiry.* In *The SAGE Encyclopedia of Qualitative Research Methods* (Vol. 0, pp. 542-544). SAGE Publications, Inc., https://doi.org/10.4135/9781412963909

Glenton C. (2003). Chronic back pain sufferers-striving for the sick role. *Social Science & Medicine (1982), 57*(11), 2243-2252. https://doi.org/10.1016/s0277-9536(03)00130-8

Goffman, E. (1963). *Stigma: notes on the management of spoiled identity.* London: Penguin Books.

Gómez-Mayordomo, V., Palacios-Ceña, M., Guerrero-Peral, Á., Fuensalida-Novo, S., Fernández-de-Las-Peñas, C., & Cuadrado, M. L. (2020). Widespread hypersensitivity to pressure pain in men with Cluster Headache during prolonged remission is not related to the levels of depression and anxiety. *Pain Practice: the Official Journal of World Institute of Pain*, *20*(2), 147-153. https://doi.org/10.1111/papr.12839

González de la Flor, Á., García Pérez de Sevilla, G., Domíngez Balmaseda, D., Martín Vera, D., Montero Martínez, M., & Del Blanco Muñiz, J. Á. (2022). Relationship between self-efficacy and headache impact, anxiety, and physical activity levels in patients with chronic tension-type headache: an observational study. *Behavioural Neurology, 6,* 8387249. https://doi.org/10.1155/2022/8387249

Gorczyca, R., Filip, R., & Walczak, E. (2013). Psychological aspects of pain. *Annals of Agricultural and Environmental Medicine (AAEM), 1*, 23-27. https://www.researchgate.net/publication/263778347_Psychological_Aspects_of_Pain

Goulet-Pelletier, J.-C., & Cousineau, D. (2018). A review of effect sizes and their confidence intervals, Part 1: The Cohen's *d* family. *The Quantitative Methods for Psychology, 14*(4), 242-265. https://doi.org/10.20982/tqmp.14.4.p242

Grenard, J. L., Munjas, B. A., Adams, J. L., Suttorp, M., Maglione, M., McGlynn, E. A., & Gellad, W. F. (2011). Depression and medication adherence in the treatment of chronic diseases in the United States: a meta-analysis. *Journal of General Internal Medicine, 26*(10), 1175-1182. https://doi.org/10.1007/s11606-011-1704-y

Grinberg, A. S., Best, R. D., Min, K. M., Schindler, E. A. D., Koo, B. B., Sico, J. J., & Seng, E. K. (2021). Cluster Headache: Clinical Characteristics and Opportunities to Enhance Quality of Life. *Current Pain and Headache Reports*, *25*(10), 65. https://doi.org/10.1007/s11916-021-00979-8

Hadi, M. A., Alldred, D. P., Briggs, M., Marczewski, K., & Closs, S. J. (2017). 'Treated as a number, not treated as a person': a qualitative exploration of the perceived barriers to effective pain management of patients with chronic pain. *BMJ Open*, *7*(6), e016454. https://doi.org/10.1136/bmjopen-2017-016454

Hak, T., Van Rhee, H. J., & Suurmond, R. (2016). How to interpret results of meta-analysis. (Version 1.3) Rotterdam, The Netherlands: Erasmus Rotterdam Institute of Management. www.erim.eur.nl/researchsupport/meta-essentials/downloads

Halahakoon, D. C., Kieslich, K., O'Driscoll, C., Nair, A., Lewis, G., & Roiser, J. P. (2020). Reward-processing behavior in depressed participants relative to healthy volunteers: a systematic review and meta-analysis. *JAMA Psychiatry, 77*(12), 1286-1295. https://doi.org/10.1001/jamapsychiatry.2020.2139

Hamilton M. (1980). Rating depressive patients. *The Journal of Clinical Psychiatry, 41*(12 Pt 2), 21-24. https://pubmed.ncbi.nlm.nih.gov/7440521/

Hasenbring, M. I., Hallner, D., & Rusu, A. C. (2009). Fear-avoidance- and endurance-related responses to pain: development and validation of the Avoidance-Endurance Questionnaire (AEQ). *European Journal of Pain (London, England), 13*(6), 620-628. https://doi.org/10.1016/j.ejpain.2008.11.001

Hayes, S.C., Pierson, H. (2005). Acceptance and Commitment Therapy. In: Freeman, A., Felgoise, S.H., Nezu, C.M., Nezu, A.M., Reinecke, M.A. (eds) *Encyclopedia of Cognitive Behavior Therapy.* Springer, Boston, MA. https://doi.org/10.1007/0-306-48581-8_1

Hayes, S. C., Strosahl, K. D., & Wilson, K. G. (1999). *Acceptance and commitment therapy: An experiential approach to behavior change.* Guilford Press.

Headache Classification Committee of the International Headache Society (IHS) (2013). The International Classification of Headache Disorders, 3rd edition (beta version). *Cephalalgia: an International Journal of Headache, 33*(9), 629-808. https://doi.org/10.1177/0333102413485658

Headache Classification Committee of the International Headache Society (IHS). (2018). The International Classification of Headache Disorders, 3rd edition. *Cephalalgia: an International Journal of Headache, 38*(1), 1-211. https://doi.org/10.1177/0333102417738202

Hee, S. W., Patel, S., Sandhu, H., Matharu, M. S., Underwood, M., & CHESS Team (2024). Does pain self-efficacy predict, moderate or mediate outcomes in people with chronic headache; an exploratory analysis of the CHESS trial. *The journal of Headache and Pain, 25*(1), 77. https://doi.org/10.1186/s10194-024-01768-5

Hennink, M., & Kaiser, B. N. (2022). Sample sizes for saturation in qualitative research: A systematic review of empirical tests. *Social Science & Medicine (1982)*, *292*, 114523. https://doi.org/10.1016/j.socscimed.2021.114523

Hermesdorf, M., Berger, K., Baune, B. T., Wellmann, J., Ruscheweyh, R., & Wersching, H. (2016). Pain sensitivity in patients with major depression: differential effect of pain sensitivity measures, somatic cofactors, and disease characteristics. *The Journal of Pain, 17*(5), 606-616. https://doi.org/10.1016/j.jpain.2016.01.474

Higgins, E. T. (1987). Self-discrepancy: A theory relating self and affect. *Psychological Review, 94*(3), 319-340. https://doi.org/10.1037/0033-295X.94.3.319

Higgins, J. P. T., Li, T., Deeks, J. J. (2023). Chapter 6: Choosing effect measures and computing estimates of effect. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). *Cochrane Handbook for Systematic Reviews of*

Interventions version 6.5. Cochrane, 2024. Available from www.training.cochrane.org/handbook.

Higgins, J. P. T., Thomas, J., Chandler, J., Cumpston, M., Li, T., Page, M. J., Welch, V. A (editors). (2019). *Cochrane Handbook for Systematic Reviews of Interventions.* 2nd Edition. Chichester (UK): John Wiley & Sons.

Higgins, J. P. T., Altman, D. G., Gøtzsche, P. C., Jüni, P., Moher, D., Oxman, A. D., Savovic, J., Schulz, K. F., Weeks, L., Sterne, J. A., Cochrane Bias Methods Group, & Cochrane Statistical Methods Group (2011). The Cochrane Collaboration's tool for assessing risk of bias in randomised trials. *BMJ (Clinical research ed.)*, *343*, d5928. https://doi.org/10.1136/bmj.d5928

Higgins, J. P. T., Thompson, S. G., & Spiegelhalter, D. J. (2009). A re-evaluation of random-effects meta-analysis. *Journal of the Royal Statistical Society. Series A, (Statistics in Society)*, *172*(1), 137-159. https://doi.org/10.1111/j.1467-985X.2008.00552.x

Higgins, J. P. T., & Thompson, S. G. (2002). Quantifying heterogeneity in a meta-analysis. Statistics in medicine, 21(11), 1539-1558. https://doi.org/10.1002/sim.1186

Hoffmann, J., & May, A. (2018). Diagnosis, pathophysiology, and management of cluster headache. *The Lancet. Neurology, 17*(1), 75-83. https://doi.org/10.1016/S1474-4422(17)30405-2

Horton, B. T., MacLean, A. R., & Craig, W. M. (1939). A new syndrome of vascular headache: results of treatment with histamine: preliminary report. *In Mayo Clinical Proceeding, 14,* 257-260).

Huedo-Medina, T. B., Sánchez-Meca, J., Marín-Martínez, F., & Botella, J. (2006). Assessing heterogeneity in meta-analysis: Q statistic or I2 index? *Psychological Methods*, *11*(2), 193-206. https://doi.org/10.1037/1082-989X.11.2.193

IASP Subcommittee on Taxonomy. (1979). Pain terms: a list with definitions and notes on usage. Recommended by the IASP Subcommittee on Taxonomy. *Pain, 6,*249-52. https://pubmed.ncbi.nlm.nih.gov/460932/

İnce, B. (2020). Systematic review of the comparative effectiveness of cognitive-behavioural therapies for chronic pain. *Journal of Cognitive Behavioral Psychotherapy and Research, 9*(3), 248-259. https://doi.org/10.5455/JCBPR.64324

International Association for the Study of Pain. (2020). *IASP Revises Its Definition of Pain for the First Time Since 1979*. https://www.iasp-pain.org/wp-content/uploads/2022/04/revised-definition-flysheet_R2-1-1-1.pdf

IntHout, J., Ioannidis, J. P. A., Rovers, M. M., & Goeman, J. J. (2016). Plea for routinely presenting prediction intervals in meta-analysis. *BMJ Open*, *6*(7), e010247. https://doi.org/10.1136/bmjopen-2015-010247

İşcan, D., & Dal, C. (2024). The relation of cluster headache to alexithymia, depression, and anxiety. La relación de la cefalea en racimos con la alexitimia, la depresión y la ansiedad. *Arquivos De Neuro-Psiquiatria*, *82*(11), 1-5. https://doi.org/10.1055/s-0044-1791516

Jang, B. N., Lee, H. J., Joo, J. H., Park, E. C., & Jang, S. I. (2020). Association between health behaviours and depression: findings from a national cross-sectional study in South Korea. *BMC Psychiatry, 20*(1), 238. https://doi.org/10.1186/s12888-020-02628-7

Jensen, M. P., Thorn, B. E., Carmody, J., Keefe, F. J., & Burns, J. W. (2018). The role of cognitive content and cognitive processes in chronic pain: An important distinction? *The Clinical Journal of Pain, 34*(5), 391-401. https://doi.org/10.1097/AJP.0000000000000559

Jensen, R., Zeeberg, P., Dehlendorff, C., & Olesen, J. (2010). Predictors of outcome of the treatment programme in a multidisciplinary headache centre. *Cephalalgia: an International Journal of Headache, 30*(10), 1214-1224. https://doi.org/10.1177/0333102410361403

Ji Lee, M., Cho, S. J., Wook Park, J., Kyung Chu, M., Moon, H. S., Chung, P. W., Myun Chung, J., Sohn, J. H., Kim, B. K., Kim, B. S., Kim, S. K., Song, T. J., Choi, Y. J., Park, K. Y., Oh, K., Ahn, J. Y., Lee, K. S., Cho, S., & Chung, C. S. (2019). Increased suicidality in patients with CH. *Cephalalgia: an International Journal of Headache, 39*(10), 1249-1256. https://doi.org/10.1177/0333102419845660

Johnson, M. I., Hudson, M., & Ryan, C. G. (2023). Perspectives on the insidious nature of pain metaphor: we literally need to change our metaphors. *Frontiers in Pain Research (Lausanne, Switzerland)*, *4*, 1224139. https://doi.org/10.3389/fpain.2023.1224139

Jorge, R. E., Leston, J. E., Arndt, S., & Robinson, R. G. (1999). Cluster headaches: association with anxiety disorders and memory deficits. *Neurology*, *53*(3), 543–547. https://doi.org/10.1212/wnl.53.3.543

Joshi, S., Rizzoli, P., & Loder, E. (2017). The comorbidity burden of patients with cluster headache: a population-based study. *The Journal of Headache and Pain*, *18*(1), 76. https://doi.org/10.1186/s10194-017-0785-3

Jürgens, T. P., Gaul, C., Lindwurm, A., Dresler, T., Paelecke-Habermann, Y., Schmidt-Wilcke, T., Lürding, R., Henkel, K., & Leinisch, E. (2011). Impairment in episodic and chronic cluster headache. *Cephalalgia: an International Journal of Headache*, *31*(6), 671–682. https://doi.org/10.1177/0333102410391489

Kalfas, M., Chisari, C., & Windgassen, S. (2022). Psychosocial factors associated with pain and health-related quality of life in Endometriosis: A systematic review. *European*

Journal of Pain (London, England), 26(9), 1827-1848. https://doi.org/10.1002/ejp.2006

Kessler, R. C., & Bromet, E. J. (2013). The epidemiology of depression across cultures. *Annual Review of Public Health*, *34*, 119-138. https://doi.org/10.1146/annurev-publhealth-031912-114409

Kim, B. S., Chung, P. W., Kim, B. K., Lee, M. J., Park, J. W., Chu, M. K., Ahn, J. Y., Bae, D. W., Song, T. J., Sohn, J. H., Oh, K., Kim, D., Kim, J. M., Kim, S. K., Choi, Y. J., Chung, J. M., Moon, H. S., Chung, C. S., Park, K. Y., & Cho, S. J. (2020). The impact of remission and coexisting migraine on anxiety and depression in cluster headache. *The Journal of Headache and Pain*, *21*(1), 58. https://doi.org/10.1186/s10194-020-01120-7

Kim, S. K., Chu, M. K., Kim, B. K., Chung, P. W., Moon, H. S., Lee, M. J., Choi, Y. J., Park, J. W., Kim, B. S., Song, T. J., Oh, K., Ahn, J. Y., Sohn, J. H., Lee, K. S., Park, K. Y., Chung, J. M., Chung, C. S., & Cho, S. J. (2024). An analysis of the determinants of the health-related quality of life in asian patients with cluster headaches during cluster periods using the time trade-off method. *Journal of Clinical Neurology (Seoul, Korea), 20*(1), 86-93. https://doi.org/10.3988/jcn.2022.0396

Klonowski, T., Kropp, P., Straube, A., & Ruscheweyh, R. (2022). Psychological factors associated with headache frequency, intensity, and headache-related disability in migraine patients. *Neurological Sciences: Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology, 43*(2), 1255-1266. https://doi.org/10.1007/s10072-021-05453-2

Koo, B. B., Bayoumi, A., Albanna, A., Abusuliman, M., Burrone, L., Sico, J. J., & Schindler, E. A. D. (2021). Demoralization predicts suicidality in patients with cluster headache.

The Journal of Headache and Pain, *22*(1), 28. https://doi.org/10.1186/s10194-021-01241-7

Koopmans, E., & Schiller, D. C. (2022). Understanding causation in healthcare: an introduction to critical realism. *Qualitative Health Research, 32*(8-9), 1207-1214. https://doi.org/10.1177/10497323221105737

Kroenke, K., Spitzer, R. L., & Williams, J. B. W. (2001). The PHQ-9: Validity of a brief depression severity measure. *Journal of General Internal Medicine, 16*(9), 606-613. https://doi.org/10.1046/j.1525-1497.2001.016009606.x2.

Landmark, L., Sunde, H. F., Fors, E. A., Kennair, L. E. O., Sayadian, A., Backelin, C., & Reme, S. E. (2024). Associations between pain intensity, psychosocial factors, and pain-related disability in 4285 patients with chronic pain. *Scientific Reports, 14*(1), 13477. https://doi.org/10.1038/s41598-024-64059-8

Lewinsohn, P. M. (1974). A behavioral approach to depression. In R. J. Friedman & M. M. Katz (Eds.), *The psychology of depression: Contemporary theory and research.* John Wiley & Sons.

Liang, J. F., Chen, Y. T., Fuh, J. L., Li, S. Y., Liu, C. J., Chen, T. J., Tang, C. H., & Wang, S. J. (2013). Cluster headache is associated with an increased risk of depression: a nationwide population-based cohort study. *Cephalalgia: an International Journal of Headache*, *33*(3), 182-189. https://doi.org/10.1177/0333102412469738

Lim, W. M. (2024). What is qualitative research? An overview and guidelines. *Australasian Marketing Journal, 0*(0). https://doi.org/10.1177/14413582241264619

Linehan, M. M. (1993). *Cognitive behavioral treatment of borderline personality disorder.* New York: Guilford Press.

Linehan, M. M. (2014). DBT Skills Training Handouts and Worksheets, Second Edition. Guilford Publications.

Lin, H. (2024). *Effect Size Converter.* MIT License. https://www.escal.site/

Lipsey, M. W., & Wilson, D. B. (2001). *Practical meta-analysis.* Sage Publications, Inc.

Long, R. J., Zhu, Y. S., & Wang, A. P. (2021). Cluster headache due to structural lesions: A systematic review of published cases. *World Journal of Clinical Cases, 9*(14), 3294-3307. https://doi.org/10.12998/wjcc.v9.i14.3294

Losantos, M., Montoya, T.M., Exeni, S., & Paz, L. (2016). Applying social constructionist epistemology to research in psychology. *International Journal of Collaborative-Dialogic Practices, 6*(1), 29-42. https://iicc.ucb.edu.bo/wp-content/uploads/2017/09/aplying-social-constructionist-epistemology-to-research-in-psychology.pdf

Louter, M. A., Wilbrink, L. A., Haan, J., van Zwet, E. W., van Oosterhout, W. P., Zitman, F. G., Ferrari, M. D., & Terwindt, G. M. (2016). Cluster headache and depression. *Neurology*, *87*(18), 1899-1906. https://doi.org/10.1212/WNL.0000000000003282

Lumivero (2023) NVivo (Version 14) www.lumivero.com

Lund, N. L. T., Petersen, A. S., Fronczek, R., Tfelt-Hansen, J., Belin, A. C., Meisingset, T., Tronvik, E., Steinberg, A., Gaul, C., & Jensen, R. H. (2023). Current treatment options for cluster headache: limitations and the unmet need for better and specific treatments-a consensus article. *The Journal of Headache and Pain, 24*(1), 121. https://doi.org/10.1186/s10194-023-01660-8

Ma, L. L., Wang, Y. Y., Yang, Z. H., Huang, D., Weng, H., & Zeng, X. T. (2020). Methodological quality (risk of bias) assessment tools for primary and secondary medical studies: what are they and which is better? *Military Medical Research*, *7*(1), 7. https://doi.org/10.1186/s40779-020-00238-8

Ma, L. (2012). Some philosophical considerations in using mixed methods in library and information science research. *Journal of the Association for Information Science and Technology, 63,* 1859- 1867. https://doi.org/10.1002/asi.22711

Majumdar, A., Ahmed, M. A., & Benton, S. (2009). Cluster headache in children-experience from a specialist headache clinic. *European Journal of Paediatric Neurology: EJPN: Official Journal of the European Paediatric Neurology Society, 13*(6), 524-529. https://doi.org/10.1016/j.ejpn.2008.11.002

Malterud, K., Siersma, V. D., & Guassora, A. D. (2016). Sample size in qualitative interview studies: guided by information power. *Qualitative Health Research*, *26*(13), 1753-1760. https://doi.org/10.1177/1049732315617444

Manzoni, G. C., Taga, A., Russo, M., & Torelli, P. (2016). Age of onset of episodic and chronic cluster headache - a review of a large case series from a single headache centre. *The Journal of Headache and Pain, 17,* 44. https://doi.org/10.1186/s10194-016-0626-9

Markley, H. G., & Buse, D. C. (2006). Cluster headache: myths and the evidence. *Current Pain and Headache Reports*, *10*(2), 137-141. https://doi.org/10.1007/s11916-006-0025-z

Martin, P. R., & MacLeod, C. (2009). Behavioral management of headache triggers: Avoidance of triggers is an inadequate strategy. *Clinical Psychology Review*, *29*(6), 483-495. https://doi.org/10.1016/j.cpr.2009.05.002

Martin, P. R., Reece, J., Callan, M., MacLeod, C., Kaur, A., Gregg, K., & Goadsby, P. J. (2014). Behavioral management of the triggers of recurrent headache: a randomized controlled trial. *Behaviour Research and Therapy*, *61*, 1-11. https://doi.org/10.1016/j.brat.2014.07.002

Mathur, V. A., Trost, Z., Ezenwa, M. O., Sturgeon, J. A., & Hood, A. M. (2022). Mechanisms of injustice: what we (do not) know about racialized disparities in pain. *Pain, 163*(6), 999-1005. https://doi.org/10.1097/j.pain.0000000000002528

Maxcy, S.J. (2003). *Pragmatic threads in mixed methods research in the social science. Handbook of Mixed Methods Research in Social and Behavioural Research.* London, Sage, pp.51-87.

May, A. (2013). Diagnosis and clinical features of trigemino-autonomic headaches. *Headache, 53*(9), 1470-1478. https://doi.org/10.1111/head.12213

McCracken, L. M., & Morley, S. (2014). The psychological flexibility model: a basis for integration and progress in psychological approaches to chronic pain management. *The Journal of Pain, 15*(3), 221-234. https://doi.org/10.1016/j.jpain.2013.10.014

Mitsikostas, D., & Thomas, A. (1999). Comorbidity of headache and depressive disorders. *Cephalalgia*, *19*(4), 211-217. https://doi.org/10.1046/j.1468-2982.1999.019004211.x

Morgan, D. (2007). Paradigms lost and pragmatism regained: Methodological implications of combining qualitative and quantitative methods. *Journal of Mixed Methods Research, 1*(1), 48- 76. https://doi.org/10.1177/2345678906292462

Morgan, R. L., Whaley, P., Thayer, K. A., & Schünemann, H. J. (2018). Identifying the PECO: A framework for formulating good questions to explore the association of environmental and other exposures with health outcomes. *Environment International*, *121*(Pt 1), 1027-1031. https://doi.org/10.1016/j.envint.2018.07.015

Muka, T., Glisic, M., Milic, J., Verhoog, S., Bohlius, J., Bramer, W., Chowdhury, R., & Franco, O. H. (2020). A 24-step guide on how to design, conduct, and successfully

publish a systematic review and meta-analysis in medical research. *European Journal of Epidemiology, 35*(1), 49-60. https://doi.org/10.1007/s10654-019-00576-5

Mazzucchelli, T. G., Kane, R. T., & Rees, C. S. (2010). Behavioral activation interventions for well-being: A meta-analysis. *The Journal of Positive Psychology, 5*(2), 105-121. https://doi.org/10.1080/17439760903569154

Nesbitt, A. D., & Goadsby, P. J. (2012). Cluster Headache. *BMJ, 344,* e2407. https://doi.org/10.1136/bmj.e2407

Newton, B. J., Southall, J. L., Raphael, J. H., Ashford, R. L., & LeMarchand, K. (2013). A narrative review of the impact of disbelief in chronic pain. *Pain Management Nursing: Official Journal of the American Society of Pain Management Nurses, 14*(3), 161-171. https://doi.org/10.1016/j.pmn.2010.09.001

NHS England. (2023). *Co-production: An Introduction.* NHS England. https://www.england.nhs.uk/long-read/co-production-an-introduction/

NICE. (2009). Borderline personality disorder: Treatment and Management. NICE Clinical Guidelines, No. 78. London: National Institute for Health and Clinical Excellence. https://www.nice.org.uk/guidance/cg78/chapter/1-guidance

NICE. (2022). *Cluster Headache.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/headache-cluster/management/management/

NICE. (2022). *Headache-Tension Type.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/headache-tension-type/

NICE. (2024). *Depression.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/depression/

NICE. (2022). *Depression in Adults: Treatment and Management.* The National Institute for Health and Care Excellence. https://www.nice.org.uk/guidance/ng222/chapter/recommendations#treatment-manuals

NICE. (2024). *Migraines.* The National Institute for Health and Care Excellence. https://cks.nice.org.uk/topics/migraine/management/adults/

NICE. (2024). NICE processes and methods. https://www.nice.org.uk/process/pmg20/chapter/introduction

Nicholas, M. K. (2007). The pain self-efficacy questionnaire: Taking pain into account. *European Journal of Pain (London, England), 11*(2), 153-163. https://doi.org/10.1016/j.ejpain.2005.12.008

Norton, P. J., & Asmundson, G. J. G. (2003). Amending the fear-avoidance model of chronic pain: What is the role of physiological arousal? *Behavior Therapy, 34*(1), 17-30. https://doi.org/10.1016/S0005-7894(03)80019-9

Nortvedt, F., & Engelsrud, G. (2014). "Imprisoned" in pain: analyzing personal experiences of phantom pain. *Medicine, Health Care, and Philosophy*, *17*(4), 599-608. https://doi.org/10.1007/s11019-014-9555-z

O'Brien, B. C., Harris, I. B., Beckman, T. J., Reed, D. A., & Cook, D. A. (2014). Standards for reporting qualitative research: a synthesis of recommendations. *Academic Medicine: Journal of the Association of American Medical Colleges*, *89*(9), 1245-1251. https://doi.org/10.1097/ACM.0000000000000388

O'Driscoll, C., Buckman, J. E. J., Saunders, R., Ellard, S., Naqvi, S. A., Singh, S., Wheatley, J., & Pilling, S. (2023). Symptom-specific effects of counselling for depression compared to cognitive-behavioural therapy. *BMJ Mental Health, 26*(1), e300621. https://doi.org/10.1136/bmjment-2022-300621

Okolo, C. A., Babawarun, O., & Olorunsogo, T. O. (2024). Cross-cultural perspectives on pain: a comprehensive review of anthropological research. *International Journal of Applied Research in Social Sciences, 6*(3). https://doi.org/10.51594/ijarss.v6i3.888

Oris, L., Luyckx, K., Rassart, J., Goubert, L., Goossens, E., Apers, S., Arat, S., Vandenberghe, J., Westhovens, R., & Moons, P. (2018). Illness identity in adults with a chronic illness. *Journal of Clinical Psychology in Medical Settings, 25*(4), 429-440. https://doi.org/10.1007/s10880-018-9552-0

Oris, L., Rassart, J., Prikken, S., Verschueren, M., Goubert, L., Moons, P., Berg, C. A., Weets, I., & Luyckx, K. (2016). Illness identity in adolescents and emerging adults with Type 1 Diabetes: Introducing the Illness Identity Questionnaire. *Diabetes Care, 39*(5), 757-763. https://doi.org/10.2337/dc15-2559

Osman, A., Bagge, C. L., Gutierrez, P. M., Konick, L. C., Kopper, B. A., & Barrios, F. X. (2001). The Suicidal Behaviors Questionnaire-Revised (SBQ-R): validation with clinical and nonclinical samples. *Assessment, 8(*4), 443–454. https://doi.org/10.1177/107319110100800409

Osman, A., Barrios, F. X., Gutierrez, P. M., Kopper, B. A., Merrifield, T., & Grittmann, L. (2000). The Pain Catastrophizing Scale: Further psychometric evaluation with adult samples. *Journal of Behavioral Medicine, 23*(4), 351-365. https://doi.org/10.1023/A:1005548801037

Osman, A., Barrios, F. X., Kopper, B. A., Hauptmann, W., Jones, J., & O'Neill, E. (1997). Factor structure, reliability, and validity of the Pain Catastrophizing Scale. *Journal of Behavioral Medicine, 20*(6), 589-605. https://doi.org/10.1023/a:1025570508954

OUCH. (2024). *Cluster Headache.* https://ouchuk.org/

Page, M. J., McKenzie, J. E., Bossuyt, P. M., Boutron, I., Hoffmann, T. C., Mulrow, C. D., Shamseer, L., Tetzlaff, J. M., & Moher, D. (2021). Updating guidance for reporting

systematic reviews: development of the PRISMA 2020 statement. *Journal of Clinical Epidemiology, 134,* 103-112. https://doi.org/10.1016/j.jclinepi.2021.02.003

Palacios-Ceña, D., Talavera, B., López-Ruiz, P., Gutiérrez-Viedma, Á., Palacios-Ceña, M., Arias, J. A., Fernández-de-Las-Peñas, C., & Cuadrado, M. L. (2016). Living with cluster headache: a qualitative study of patients' perspectives. *Headache, 56*(7), 1171–1182. https://doi.org/10.1111/head.12886

Paterson B. L. (2001). The shifting perspectives model of chronic illness. *Journal of Nursing Scholarship: an Official Publication of Sigma Theta Tau International Honor Society of Nursing, 33*(1), 21-26. https://doi.org/10.1111/j.1547-5069.2001.00021.x

Paul, M., & Leeflang, M. M. (2021). Reporting of systematic reviews and meta-analysis of observational studies. *Clinical Microbiology and Infection: the Official Publication of the European Society of Clinical Microbiology and Infectious Diseases, 27*(3), 311-314. https://doi.org/10.1016/j.cmi.2020.11.006

Peng, K. P., & Burish, M. J. (2023). Management of cluster headache: Treatments and their mechanisms. *Cephalalgia: an International Journal of Headache, 43*(8), 3331024231196808. https://doi.org/10.1177/03331024231196808

Pérez-Aranda, A., Hofmann, J., Feliu-Soler, A., Ramírez-Maestre, C., Andrés-Rodríguez, L., Ruch, W., & Luciano, J. V. (2019). Laughing away the pain: A narrative review of humour, sense of humour and pain. *European Journal of Pain (London, England)*, *23*(2), 220-233. https://doi.org/10.1002/ejp.1309

Pescador, R, M. A., & De Jesus, O. (2024). *Migraine Headache.* StatPearls Publishing.

Peters, L. A., & Brown, E. M. (2022). The relationship between illness identity and the self-management of Inflammatory Bowel Disease. *British Journal of Health Psychology, 27*(3), 956-970. https://doi.org/10.1111/bjhp.12584

Pohl, H., Gantenbein, A. R., Sandor, P. S., Schoenen, J., & Andrée, C. (2020). Interictal burden of cluster headache: results of the EUROLIGHT cluster headache project, an internet-based, cross-sectional study of people with cluster headache. *Headache*, *60*(2), 360-369. https://doi.org/10.1111/head.13711

Qin, L., Song, P., Li, X. *et al.* Tension-Type Headache Management: A Systematic Review and Network Meta-analysis of Complementary and Alternative Medicine. *Pain Therapy 13*, 691-717 (2024). https://doi.org/10.1007/s40122-024-00600-x

Quartana, P. J., Campbell, C. M., & Edwards, R. R. (2009). Pain catastrophizing: a critical review. *Expert Review of Neurotherapeutics, 9*(5), 745-758. https://doi.org/10.1586/ern.09.34

R Core Team. (2019). *R: a language and environment for statistical computing.* Vienna R Foundation for Statistical Computing. https://www.R-project.org.

Raja, S. N., Carr, D. B., Cohen, M., Finnerup, N. B., Flor, H., Gibson, S., Keefe, F. J., Mogil, J. S., Ringkamp, M., Sluka, K. A., Song, X. J., Stevens, B., Sullivan, M. D., Tutelman, P. R., Ushida, T., & Vader, K. (2020). The revised International Association for the Study of Pain definition of pain: concepts, challenges, and compromises. *Pain, 161*(9), 1976-1982. https://doi.org/10.1097/j.pain.0000000000001939

Robbins, M. S., Starling, A. J., Pringsheim, T. M., Becker, W. J., & Schwedt, T. J. (2016). Treatment of Cluster Headache: The American Headache Society Evidence-Based Guidelines. *Headache*, *56*(7), 1093-1106. https://doi.org/10.1111/head.12866

Rossi, P., Little, P., De La Torre, E. R., & Palmaro, A. (2018). If you want to understand what it really means to live with CH, imagine... fostering empathy through European patients' own stories of their experiences. *Functional Neurology, 33*(1), 57-59. https://doi.org/10.11138/fneur/2018.33.1.057

Roth, A, D., Hill, A., Pilling. S. (2009). *The competences required to deliver effective humanistic psychological therapies.* University College London

Roughan, W. H., Campos, A. I., García-Marín, L. M., Cuéllar-Partida, G., Lupton, M. K., Hickie, I. B., Medland, S. E., Wray, N. R., Byrne, E. M., Ngo, T. T., Martin, N. G., & Rentería, M. E. (2021). Comorbid chronic pain and depression: Shared risk factors and differential antidepressant effectiveness. *Frontiers in Psychiatry, 12*, 643609. https://doi.org/10.3389/fpsyt.2021.643609

Rozen, T. D., & Fishman, R. S. (2012). Cluster headache in the United States of America: demographics, clinical characteristics, triggers, suicidality, and personal burden. *Headache, 52*(1), 99-113. https://doi.org/10.1111/j.1526-4610.2011.02028.x

Ruscheweyh, R., Pereira, D., Hasenbring, M. I., & Straube, A. (2019). Pain-related avoidance and endurance behaviour in migraine: an observational study. The Journal of Headache and Pain, 20(1), 9. https://doi.org/10.1186/s10194-019-0962-7

Russell M. B. (2004). Epidemiology and genetics of cluster headache. *The Lancet. Neurology, 3*(5), 279-283. https://doi.org/10.1016/S1474-4422(04)00735-5

Saarijärvi, M., & Bratt, E. L. (2021). When face-to-face interviews are not possible: tips and tricks for video, telephone, online chat, and email interviews in qualitative research. *European Journal of Cardiovascular Nursing*, *20*(4), 392–396. https://doi.org/10.1093/eurjcn/zvab038

Sadeghi, M. (2025). Public bias and funnel plot. In M, Ghajarzadeh,., J, Hopkins., N, Rezaei, S, Hanaei, S. (Eds). Systematic Review and Meta-Analysis. (1st edition). Elsevier Inc

Samulowitz, A., Gremyr, I., Eriksson, E., & Hensing, G. (2018). "Brave Men" and "Emotional Women": A Theory-Guided Literature Review on Gender Bias in Health

Care and Gendered Norms towards Patients with Chronic Pain. *Pain Research & Management*, 6358624. https://doi.org/10.1155/2018/6358624

Sánchez-Rodríguez, E., Aragonès, E., Jensen, M. P., Tomé-Pires, C., Rambla, C., López-Cortacans, G., & Miró, J. (2020). The role of pain-related cognitions in the relationship between pain severity, depression, and pain interference in a sample of primary care patients with both chronic pain and depression. Pain Medicine, 21(10), 2200-2211. https://doi.org/10.1093/pm/pnz363

Sargent, J. D., Walters, E. D., & Green, E. E. (1973). Psychosomatic self-regulation of migraine headaches. *Seminars in Psychiatry, 5*(4), 415-428. https://psycnet.apa.org/record/1974-23529-001

Schenck, L. A., & Andrasik, F. (2019). Behavioral and psychological aspects of cluster headache: an overview. *Neurological sciences: official journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology*, *40*(Suppl 1), 3-7. https://doi.org/10.1007/s10072-019-03831-5

Schiller C. J. (2016). Critical realism in nursing: An emerging approach. *Nursing Philosophy*, *17*(2), 88-102. https://doi.org/10.1111/nup.12107

Schindler, E. A. D., & Burish, M. J. (2022). Recent advances in the diagnosis and management of cluster headache. *BMJ (Clinical research ed.), 376*, e059577. https://doi.org/10.1136/bmj-2020-059577

Schünemann, H. J., Vist, G. E., Higgins, J. P. T., Santesso, N., Deeks, J. J., Glasziou, P., Akl, E. A., Guyatt, G. H. (2023). Chapter 15: Interpreting results and drawing conclusions. In: Higgins JPT, Thomas J, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (editors). *Cochrane Handbook for Systematic Reviews of Interventions version 6.5.* Cochrane, 2024. Available from www.training.cochrane.org/handbook.

Shah, N., Asuncion, R. M. D., & Hameed, S. (2024). *Muscle Contraction Tension Headache.* In StatPearls. StatPearls Publishing.

Shannon-Baker, P. (2016). Making paradigms meaningful in mixed methods research. *Journal of Mixed Methods Research, 10,* 319-334

Sheng, J., Liu, S., Wang, Y., Cui, R., & Zhang, X. (2017). The Link between Depression and Chronic Pain: Neural Mechanisms in the Brain. Neural plasticity, 2017, 9724371. https://doi.org/10.1155/2017/9724371

Smith, J. A., & Osborn, M. (2015). Interpretative phenomenological analysis as a useful methodology for research on the lived experience of pain. *British Journal of Pain, 9*(1), 41-42. https://doi.org/10.1177/2049463714541642

Søborg, M. K., Jensen, R. H., Barloese, M., & Petersen, A. S. (2024). Biomarkers in cluster headache: A systematic review. *Headache*, *64*(1), 98-116. https://doi.org/10.1111/head.14641

Soklaridis, S., Harris, H., Shier, R., Rovet, J., Black, G., Bellissimo, G., Gruszecki, S., Lin, E., & Di Giandomenico, A. (2024). A balancing act: navigating the nuances of co-production in mental health research. *Research Involvement and Engagement, 10*(1), 30. https://doi.org/10.1186/s40900-024-00561-7

Staniszewska, S., Brett, J., Simera, I., Seers, K., Mockford, C., Goodlad, S., Altman, D. G., Moher, D., Barber, R., Denegri, S., Entwistle, A., Littlejohns, P., Morris, C., Suleman, R., Thomas, V., & Tysall, C. (2017). GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research. *BMJ (Clinical research ed.), 358,* j3453. https://doi.org/10.1136/bmj.j3453

Stoll, C. R. T., Izadi, S., Fowler, S., Green, P., Suls, J., & Colditz, G. A. (2019). The value> of a second reviewer for study selection in systematic reviews. *Research Synthesis Methods*, *10*(4), 539-545. https://doi.org/10.1002/jrsm.1369

Sullivan, M. J. L., Bishop, S. R., & Pivik, J. (1995). The pain catastrophizing scale: development and validation. *Psycholological Assessment, 7*(4), 524-32. https://doi.org/10.1037/1040-3590.7.4.524

Sweeney, A., & Morgan, L. (2009). The levels and stages of service user/survivor involvement in research. *In Handbook of Service User Involvement in Mental Health Research* (pp. 25-35). Wiley Blackwell. https://doi.org/10.1002/9780470743157.ch3

Tashakkori, A., Teddlie, C. (1998). *Mixed methodology: combining qualitative and quantitative approaches.* London, Sage.

The National Institute for Health Research [NIHR]. (2015). *Going the extra mile: improving the Nation's health and wellbeing through public involvement in research.* England: Department of Health. https://www.nihr.ac.uk/documents/about-us/our-contribution-to-research/how-we-involve-patients-carers-and-the-public/Going-the-Extra-Mile.pdf

Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care: Journal of the International Society for Quality in Health Care, 19*(6), 349-357. https://doi.org/10.1093/intqhc/mzm042

Tonkin L. (2008). The Pain Self-Efficacy Questionnaire. *The Australian Journal of Physiotherapy*, *54*(1), 77. https://doi.org/10.1016/s0004-9514(08)70073-4

Torkamani, M., Ernst, L., Cheung, L. S., Lambru, G., Matharu, M., & Jahanshahi, M. (2015). The neuropsychology of cluster headache: cognition, mood, disability, and quality of life of patients with chronic and episodic cluster headache. *Headache*, *55*(2), 287-300. https://doi.org/10.1111/head.12486

Torres, N. D., Newman, A. K., Day, M. A., Chan, J. F., Friedly, J. L., & Jensen, M. P. (2024). The Moderating Effects of Cognitive Processes on Pain-related Outcomes. *The Journal of Pain, 25*(7), 104483. https://doi.org/10.1016/j.jpain.2024.01.348

Tracy, S. J. (2010). Qualitative quality: Eight “big-tent” criteria for excellent qualitative research. *Qualitative Inquiry*, *16*(10), 837-851. https://doi.org/10.1177/1077800410383121

Vadivelu, N., Kai, A. M., Kodumudi, G., Babayan, K., Fontes, M., & Burg, M. M. (2017). Pain and Psychology-A Reciprocal Relationship. *Ochsner Journal, 17*(2), 173-180.

Van Bulck, L., Luyckx, K., Goossens, E., Oris, L., & Moons, P. (2019). Illness identity: Capturing the influence of illness on the person's sense of self. *European Journal of Cardiovascular Nursing, 18*(1), 4-6. https://doi.org/10.1177/1474515118811960

Van Damme, S., Crombez, G., Bijttebier, P., Goubert, L., & Van Houdenhove, B. (2002). A confirmatory factor analysis of the Pain Catastrophizing Scale: Invariant factor structure across clinical and non-clinical populations. *Pain, 96*(3), 319-324. https://doi.org/10.1016/S0304-3959(01)00463-8

Varkey, E., Linde, M., & Henoch, I. (2013). "It's a balance between letting it influence life completely and not letting it influence life at all"--a qualitative study of migraine prevention from patients' perspective. *Disability and Rehabilitation*, *35*(10), 835-844. https://doi.org/10.3109/09638288.2012.709304

Vera, J. E. D. (2012). When pain is not a place: Pain and its metaphors in late middle English medical texts. *Onomázein Revista De Lingüística Filología Y Traducción*, *26*, 279-308. https://doi.org/10.7764/onomazein.26.10

Vlaeyen, J. W. S., & Linton, S. J. (2000). Fear-avoidance and its consequences in chronic musculoskeletal pain: a state of the art. *Pain*, *85*(3), 317-332. https://doi.org/10.1016/S0304-3959(99)00242-0

Wang, J., Zhang, B., Yahaya, R., & Abdullah, A. B. (2025). Colors of the mind: a meta-analysis of creative arts therapy as an approach for post-traumatic stress disorder intervention. *BMC Psychology, 13*(1), 32. https://doi.org/10.1186/s40359-025-02361-4

Wang, R., Lu, J., & Chow, K. M. (2024). Effectiveness of mind-body interventions in labour pain management during normal delivery: A systematic review and meta-analysis. *International Journal of Nursing Studies, 158,* 104858. https://doi.org/10.1016/j.ijnurstu.2024.104858

Wang, X., & Cheng, Z. (2020). Cross-Sectional Studies: Strengths, Weaknesses, and Recommendations. Chest, 158(1S), S65–S71. https://doi.org/10.1016/j.chest.2020.03.012

Waung, M. W., Taylor, A., Qualmann, K. J., & Burish, M. J. (2020). Family History of Cluster Headache: A Systematic Review. *JAMA neurology*, *77*(7), 887-896. https://doi.org/10.1001/jamaneurol.2020.0682

Wei, C. B., Jia, J. P., Wang, F., Zhou, A. H., Zuo, X. M., & Chu, C. B. (2016). Overlap between headache, depression, and anxiety in general neurological clinics: A cross-sectional study. *Chinese Medical Journal, 129*(12), 1394-1399. https://doi.org/10.4103/0366-6999.183410 g

Wei, D. Y., & Goadsby, P. J. (2021). Cluster headache pathophysiology - insights from current and emerging treatments. *Nature Reviews. Neurology, 17*(5), 308-324. https://doi.org/10.1038/s41582-021-00477-w

Wei, D. Y., Khalil, M., & Goadsby, P. J. (2019). Managing cluster headache. *Practical Neurology, 19*(6), 521-528. https://doi.org/10.1136/practneurol-2018-002124

Wei, D. Y., Yuan Ong, J. J., & Goadsby, P. J. (2018). Cluster Headache: Epidemiology, pathophysiology, clinical features, and diagnosis. Annals of Indian Academy of *Neurology, 21*(Suppl 1), S3-S8. https://doi.org/10.4103/aian.AIAN_349_17

Wells, G.A., Wells, G., Shea, B., Shea, B., O'Connell, D., Peterson, J., Welch, Losos, M., Tugwell, P., Ga, S.W., Zello, G.A., & Petersen, J.A. (2014). *The Newcastle-Ottawa*

Scale (NOS) for assessing the quality of nonrandomised studies in meta-analyses. Ottawa Hospital Research Institute https://www.ohri.ca/programs/clinical_epidemiology/oxford.asp

Williams, S. J. (1996). The vicissitudes of embodiment across the chronic illness trajectory. *Body & Society, 2*(2), 23-47. https://doi.org/10.1177/1357034X96002002002

World Health Organization, WHO. (2021). *Depression.* World Health Organization. https://www.who.int/news-room/fact-sheets/detail/depression

Yoshikawa, K., Brady, B., Perry, M. A., & Devan, H. (2020). Sociocultural factors influencing physiotherapy management in culturally and linguistically diverse people with persistent pain: a scoping review. *Physiotherapy, 107,* 292-305. https://doi.org/10.1016/j.physio.2019.08.002

Yu, X., & Tan, G. (2024). Role of psychological factors in migraine. *Cureus,16*(12), e75858. https://doi.org/10.7759/cureus.75858

Zigmond, A. S., & Snaith, R. P. (1983). Hospital Anxiety and Depression Scale (HADS) [Database record]. *APA PsycTests.* https://doi.org/10.1037/t03589-000

Portfolio Appendices

Appendix A

PRISMA Checklist

Section and Topic	Item	Checklist item	Location where item is reported
TITLE			
Title	1	Identify the report as a systematic review.	Pg 14
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Pg 15
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Pg 20
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Pg 20
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Pg 21
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Pg 22 & Pg 26
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Pg 22 & Pg 179
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 22 & Pg 23
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Pg 23
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in	Pg 23

		each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	Pg 23
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Pg 24, 180
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	Pg 24, 25
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Pg 22, 24, 25
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Pg 23, 24, 25
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Pg 25
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Pg 25
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	Pg 25
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	Pg 25
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	Pg 25
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	

RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Pg 26, 27
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	Pg 27
Study characteristics	17	Cite each included study and present its characteristics.	Pg 30
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	Pg 35, 36
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	Pg 190, 192
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	Pg 38-44
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	Pg 38-44
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	Pg 38-44
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	Pg 42-43
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	Pg 44
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Pg 45-48
	23b	Discuss any limitations of the evidence included in the review.	Pg 48
	23c	Discuss any limitations of the review processes used.	Pg 49

	23d	Discuss implications of the results for practice, policy, and future research.	Pg 50-51
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	Pg 20
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	Pg 20
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	Pg 20 and 21
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	Pg 52
Competing interests	26	Declare any competing interests of review authors.	Pg 52
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	Pg 52

Appendix B

Full Electronic Search Terms for Systematic Review

Term One	Combined With	Term Two
"Cluster Headache OR "Trigeminal Autonomic Cephalalgia"	AND	"Depression" OR "Suicide" OR "Suicidal Ideation"

Appendix C

Adapted Newcastle-Ottawa Quality Assessment Scale

Selection (Max 4 Stars)

1. Representativeness of the Cluster Headache sample:

a) No apparent concerns regarding the representativeness of the sample with regard to the average person with Cluster Headache in the population *

b) Selected group of users (e.g., one headache unit), or small sample size, meaning there was selection bias and a threat to representativeness.

c) No description of the derivation of the sample.

2. Ascertainment of Cluster Headache Diagnosis

a) Diagnosis of CH confirmed by registered diagnosis according to ICD criteria, or by an experienced clinician*

b) Self-report

c) No description

3. Selection of the non-Cluster Headache group

a) Drawn from the same population as the Cluster Headache sample *

b) Drawn from a different source

c) No description of the derivation of the non-Cluster Headache group

4. Exclusion of Cluster Headache in non-Cluster Headache sample

a) Controls were defined clearly as having no current or past Cluster Headache pain *

b) Not description

Comparability (Max 1 Star)

1. Comparability of groups on the basis of the design or analysis. Important factors either controlled for, excluded from all groups, or considered as main grouping variables.

a) Study controlled for age and gender/sex *

Outcome (Max 3 Stars)

1. Assessment of depression/depressive symptoms/suicidal ideation/suicidal behaviour

a) Validated questionnaire/Diagnostic interview or registered diagnosis *

b) Self-report/No description/non-validated questionnaires

2. Timing of depression/suicidality stated? Longitudinal study design/follow-up?

a) Timing of depression/suicidality stated*

b) Longitudinal study design/follow-up*

c) No information regarding timing of depression

d) Cross-sectional design

Note. Adaptations supported by comparable research (Gambadauro et al., 2019; Halahakoon et al., 2020)

Appendix D: Additional Systematic Review Data

Table 1*: Detailed Table of Study Characteristics of Systematic Review Papers*

First author, study year	Country	Study Type	Cluster Headache Group	CH Diagnosis	Control Group	Depressive /Suicidality Measurement	Relevant Findings
Anagnostou ,2017	Greece	Cross-sectional study	N= 4 CCH, 17 ECH (1 female, 20 males). Mean age: 39.0 +/- 9.9	ICHD-II	27 Migraines individuals (100% women, Mean age: 34.6 +/- 12.2). 20 TTH individuals (13 women, 7 men; Mean age: 49.1 +/- 11.5)	HAM-D	Significantly more depressive symptoms in CH and TTH group than Migraine group
Ballesta-Martínez, 2022	Spain	Cross-sectional study	N= 31 ECH (100% male). Mean age (SD): 51.8 (9.8)	ICHD-III	20 headache-free controls; age-, gender-, and education-matched. 100% male. Mean age (SD): 47.4 (9.3)	HADS-D	Significantly more depressive symptoms in CH group compared to control group
Chen et al, 2010	Taiwan	Cross-sectional study	N=51 CH (11 female, 40	ICHD-II	772 Migraine individuals (83.4% females; Mean age,	HADS	No significant difference in

			male). Mean age: 38.46 +/- 12.6		42.1 +/- 13.3). 218 TTH individuals (67% female; Mean age: 50.6 +/- 16.2		depression scores between head pain conditions
Choong, 2017	U.S	Observational retrospective database study	N= 7,589 CH. (57.4% male). 73.2% 35-64 years old	CH ICD-9 code in database	30,341 headache-free controls, matched for age and sex	Depressive disorder and suicide-related claims	Significantly more claims in CH group than control for depressive disorders and suicidal ideation
Crespi, 2022	Norway	Nationwide observational study	N= 1,891 CH (59.5% were male). Median age 42 (interquartile range 20)	CH ICD-10 code on registry	N= 3,892,260 - total population in Norway over 18 in year 2016 was used to calculate ORs adjusted for sex and gender	Diagnosis from Norwegian registry	Significantly more suicide attempts and diagnoses of depression in CH group compared to control population
Díaz-de-Terán, 2021	Spain	Cross-sectional study	N=28 ECH, 19 CCH (35 men, 12	IHS Criteria	40 matched headache-free controls (23 men, 17 women; mean age:	HAM-D	Significantly more depressive

			women). Mean age: 46 +/- 11.3		43.9 +/- 15.5). Matched for sex, age and BMI		symptoms in CH group
Gesztelyi, 2006	Hungary	Cross-sectional study	N= 11 CH (8 men, 3 women). Mean age: 36 +/- 8	Diagnosed by neurologist, but does not state criteria applied	231 Migraine individuals (36 men and 195 women; age mean 37 + /- 10), 176 TTH individuals (48 men, 128 women; age: 39 +/- 15). Age and gender differed significantly between the groups	BDI	Statistically significant difference in depressive symptoms among the different groups, with CH group having lowest levels of depressive symptoms
Gil-Martínez, 2019	Spain	Cross-sectional study	N= 12 ECH, 8 CCH (17 men and 3 women). Median age: 37.5 (IQR: 31.25-52.5)	Diagnosed by a headache specialist, as defined by IHS criteria	16 matched headache free controls (13 men, 3 women; median age: 37 (IQR: 29-51). Matched for sex, age, education, BMI, employment	BDI-II	Significantly more depressive symptoms in CH group
Gómez-Mayordomo, 2020	Spain	Cross-sectional study	N= 40 ECH (100%).	ICHD-III criteria	Forty age and sex matched headache-free	HAM-D	Significantly more depressive

			Mean age (SD): 42(5)		men. Mean age (SD): 41 (4)		symptoms in CH group
Işcan, 2024	Turkey	Cross-sectional study	N= 18 (2 women, 16 men). Mean age of 34.78 +/- 8.981	ICHD-III criteria	18 headache-free controls matched for age and gender. 2 females, 16 men; mean age of 35.28 +/- 9.138)	BDI	Significantly more depressive symptoms in CH group
Jorge, 1999	Argentina	Case-control study	N= 21 ECH (19 males, 2 females). Mean age (SD): 39.2 (8.7)	IHS Criteria	21 TTH individuals (19 men, 2 women; mean age (SD): 36.5 (12.1). Matched for age, sex and education	HAM-D	No significant difference in depression scores between head pain conditions.
Joshi, 2017	U.S.	Population-based Study	N= 75 CH (80% male). Mean age of 43.4 (range: 20-74)	Searched registry for diagnosis of CH from one of nine headache specialists	152 age and sex matched healthy controls without CH	Co-morbid disorders reported in patient's records using ICD	Patients with CH had significantly higher diagnoses of depression than control group
Jürgens, 2011	Germany	Multicentre, prospective study	N= 27 CCH (M/F ratio	ICHD-II criteria	24 Migraine individuals (mean age: 37.4, M/F ratio: 1:3.8) and 31	The Mini-DIPS, a validated	Depressive symptoms and suicidal

			4.4:1). Mean age: 42.10. N= 26 ECH in active period (M/F ratio: 5.5:1). Mean age: 41.30. N= 22 ECH outside active period (M/F ratio: 1.8:1). Mean age: 40.60		head-ache free controls (mean age: 38.4, M/F ratio: 1.1:1)	structured clinical interview for the diagnosis of psychiatric disorders	tendencies more prevalent in headache patients compared to headache-free individuals. Especially increased in CCH patients
Kim, 2020	South Korea	Cross-sectional study	N= 191 CH (13.6% female). Age = 38.3 +/-11.1.	ICHD-III criteria	63 headache-free controls (age: 37.6 +/-10.2; 11 female (17.5%). 36 Migraine controls (age: 34.8+/-7.4, 6 females (16.7%). Age and sex matched	PHQ-9	Significantly more depressive symptoms in CH group compared to headache-free controls
Kim, 2024	South Korea	Cross-sectional study	N= 423 CH, 4% CCH (81.3%	ICHD-III criteria	52 headache-free controls age and sex matched. Age: 36.2 +/-8.8. 43 male (82.7%)	PHQ-9	Significantly more depressive symptoms

			were male). Age: 37.8 +/- 9.6.				
Koo, 2021	U.S.	Observational, case-control study	N= 56 ECH, 44 CCH (53% males). Mean age (SD): 45.5 +/- 12.3	ICHD-III criteria	135 headache-free controls 57% males. Mean age (SD): 46.5 +/- 16.6. Matched for age, sex, race, income and marital status	Suicidal Behaviour Questionnaire revised. The Brief Lifetime Depression Scale, Self report of History of Depression	Significantly more CH than control participants had lifetime passive and active suicidal ideation, plans for suicide, and lifetime depression. No significant difference between groups in terms of self-report for depression and suicide attempts
Liang, 2013	Taiwan	Population based, follow-up, study	N= 673 CH, 19 were CCH	CH ICD-9 code in database	Age and sex matched controls. 2,692 individuals with	Depression diagnosis ICD -	After the median 2.5-year follow-

			(79.95% male). Age (IQR): 35.8 (29.4-43.8).		Migraine (Age (IQR): 36.0 (29.6-44.1), 79.9% male) and 2,692 headache-free controls (Age (IQR): 35.8 (29.3-43.7))	included only when coded by psychiatrists	up duration, the CH cohort had a greater risk for developing depression compared to the control cohort, but not to the Migraine cohort
Louter, 2016	Netherlands	Cross-sectional study	N= 462 CH (73.4% male). Mean age: 49.2 +/- 11.3	ICHD-II criteria	177 headache-free controls (44.6% male; Mean age: 46.4 +/- 14.3)	HADS-D	Significantly more depressive symptoms
Mitsikosts, 1999	Greece	Cross-sectional study	N=10 ECH, 4 CCH (91% male). Mean age: 35.	IHS Criteria	150 headache-free controls age and sex matched (63% women, mean age; 33 +/- 4), 170 Migraine individuals (30% men, mean age: 35), 263 TTH (40% men, mean age: 35)	HAM-D	Depressive symptoms higher in CH group compared to headache-free group, but lower than the

							Migraine and TTH groups
Torkamani, 2015	UK	Cross-sectional study	N= 11 CCH. Mean age: 49.18 +/- 11.02. N= 11 ECH. Mean age: 40.82 +/-1 15.11.	ICHD-II criteria	12 headache-free controls (mean age: 53.17 +/- 16.25, 33% male and 67% female). Age matched to CH group, but significantly different gender ratio	BDI-II	Significantly more depressive symptoms in CH group compared to controls

Note. CH= Cluster Headache; CCH= Chronic Cluster Headache; ECH= Episodic Cluster Headache; TTH= Tension Type Headache; ICHD= The International Classification of Headache Disorders; IHS= International Headache Society; ICD= International Classification of Diseases; PHQ-9= Patient Health Questionnaire; BDI = Beck Depression Inventory; HADS-D = Hospital Anxiety and Depression Scale; Depression subscale; HAM-D= Hamilton Depression Rating Scale

Systematic Review: Individual Study Statistics

Table 2: *Meta-analysis for depression levels in CH adults versus healthy controls. Individual study means, standard deviations and effect size estimates.*

Study	N1	M1	SD1	N2	M2	SD2	dval	OR
Ballesta-Martonez, 2022	31	3.9	4.1	20	1.2	1	0.83	
Choong, 2017	7589			30341			0.44	2.22
Crespi, 2022	1891			3,892,260			0.61	3.0
Diaz-de-Teran, 2021	47	6.66	4.98	40	2.65	2.57	0.99	
Gilmartinez, 2019	20	7.75	6.58	16	2.17	4.47	0.97	
Gomez-Mayordomo, 2019	40	2.6	1.1	40	1.2	0.8	1.46	
Iscal, 2024	18	10.78	6.88	18	4.73	1.11	1.23	
Joshi, 2017	75			152			0.55	2.69*
Jurgens, 2011	75			31			0.56	2.78*
Kim, 2020	191	7.4	6.1	63	3.1	3.1	0.78	
Kim, 2024	423	7.33	6.69	52	2.33	2.29	0.79	
Koo, 2021	100			135			0.79	4.20*
Liang, 2013	673			2692			1.003	6.1*
Mitsikostas, 1999	14	11.5	6.6	150	5.7	3.3	1.58	
Louter, 2016	462	5.3	4.3	177	2.6	2.9	0.68	
Torkamani, 2015	22	21.02	8.94	12	4.92	2.811	2.17	

Note. N1= sample size of CH group, M1 & SD1= Mean and standard deviation for depression levels in CH group. N2= sample size of non-headache group, M2 & SD2= mean and standard deviation for depression levels in non-headache group. dval= cohen's d. OR: odds ratio for when means and standard deviation were not available. *When OR was not provided, it was calculated from proportion data provided.

Table 3: *Meta-analysis for suicidality levels in CH adults versus healthy controls. Individual study means, standard deviations and effect size estimates.*

Study	N1	M1	SD1	N2	M2	SD2	dval	OR
Choong, 2017	7589			30341			0.50	2.49
Crespi, 2022	1891			3,892,260			0.83	4.5
Jurgens, 2011	75			31			1.01	6.29*
Koo, 2021	100	6.1	3.1	135	4.7	2.6	0.50	

Note. N1= sample size of CH group, M1 & SD1= Mean and standard deviation for suicidality levels in CH group. N2= sample size of non-headache group, M2 & SD2= mean and standard deviation for depression levels in non-headache group. dval= cohen's d. OR: odds ratio for when means and standard deviation were not available.*When OR was not provided, it was calculated from proportion data provided.

Table 4*: Meta-analysis for depression levels in CH adults versus Migraine controls.*

Individual study means, standard deviations and effect size estimates.

Study	N1	M1	SD1	N2	M2	SD2	dval	OR
Anagnostou, 2017	21	16.76	10.91	27	9.43	6.63	0.84	
Chen, 2010	51	13		772	16.7	7.9	-0.48	
Gesztelyi, 2006	11	5.67	3.39	231	8.67	7.46	-0.41	
Jurgens, 2011	75			24			0.27	1.62*
Kim, 2020	191	7.4	6.1	36	6.6	5.2	0.13	
Liang, 2013	673			2692			0.032	1.06*
Mitsikostas, 1999	14	11.5	6.6	170	14.1	7.6	-0.35	

Note. N1= sample size of CH group, M1 & SD1= Mean and standard deviation for depression levels in CH group. N2= sample size of Migraine group, M2 & SD2= mean and standard deviation for depression levels in Migraine group. dval= cohen's d. OR: odds ratio for when means and standard deviation not available. *When OR was not provided, it was calculated from proportion data provided.

Table 5: *Meta-analysis for depression levels in CH adults versus Tension-Type Headache controls. Individual study means, standard deviations and effect size estimates.*

Study	N1	M1	SD1	N2	M2	SD2	dval
Anagnostou, 2017	21	16.76	10.91	20	18.6	8.37	-0.19
Chen et al., 2010	51	13		218	15.1	3.2	-0.73
Gesztelyi, 2006	11	5.67	3.39	176	12.83	11.21	-0.66
Jorge, 1999	21	8.6	0.9	21	6.5	1.1	2.09
Mitsikostas, 1999	14	11.5	6.6	263	14.9	6.3	-0.54

Note. N1= sample size of CH group, M1 & SD1= Mean and standard deviation for depression levels in CH group. N2= sample size of Tension-Type Headache group, M2 & SD2= mean and standard deviation for depression levels in Tension-Type Headache group. dval= cohen's d.

Appendix E

Consolidated Criteria for Reporting Qualitative Studies COREQ-32

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. International Journal for Quality in Health Care. 2007. Volume 19, Number 6: pp. 349-357

Item No	Guide Questions/Description	Reported on Page:
Domain 1: Research team and reflexivity		
Personal Characteristics		
1. Interviewer/ facilitator	Which author/s conducted the interview or focus group?	Pg 75
2. Credentials	What were the researcher's credentials? E.g., PhD, MD	Pg 69
3. Occupation	What was their occupation at the time of the study?	Pg 69
4. Gender	Was the researcher male or female?	Pg 69
5. Experience and training	What experience or training did the researcher have?	Pg 69
Relationship with participants		
6. Relationship established	Was a relationship established prior to study commencement?	Appendix J
7. Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research?	Appendix J
8. Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Pg 119
Domain 2: study design		
Theoretical framework		
9. Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	Pg 73 & 117
Participant selection		
10. Sampling	How were participants selected? e.g., purposive, convenience, consecutive, snowball	Pg 74
11. Method of approach	How were participants approached? e.g., face-to-face, telephone, mail, email	Pg 74
12. Sample size	How many participants were in the study?	Pg 77

13. Non-participation Setting	How many people refused to participate or dropped out? Reasons?	Pg 77
14. Setting of data collection	Where was the data collected? e.g., home, clinic, workplace	Pg 77
15. Presence of nonparticipants	Was anyone else present besides the participants and researchers?	Pg 77
16. Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Pg 77
Data collection		
17. Interview guide	Were questions, prompts, and guides provided by the authors? Was it pilot tested?	Pg 75 & 198
18. Repeat interviews	Were repeat interviews carried out? If yes, how many?	N/A
19. Audio/visual recording	Did the research use audio or visual recording to collect the data?	Pg 77
20. Field notes	Were field notes made during and/or after the interview or focus group?	N/A
21. Duration	What was the duration of the interviews or focus group?	Pg 74
22. Data saturation	Was data saturation discussed?	Pg 78-information power
23. Transcripts returned	Were transcripts returned to participants for comment and/or correction?	Pg 76
Domain 3: analysis and findings		
Data analysis		
24. Number of data coders	How many data coders coded the data?	Pg 76, 119
25. Description of the coding tree	Did the authors provide a description of the coding tree?	Pg 123
26. Derivation of themes	Were themes identified in advance or derived from the data?	Pg 76 (inductive analysis)
27. Software	What software, if applicable, was used to manage the data?	Pg 119-120
28. Participant checking	Did participants provide feedback on the findings?	No, but PPI did.
Reporting		
29. Quotations presented	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g., participant number	Pg 80-94

30. Data and findings consistent	Was there consistency between the data presented and the findings?	Pg 94-98
31. Clarity of major themes	Were major themes clearly presented in the findings?	Pg 79, 123
32. Clarity of minor themes	Is there a description of diverse cases or a discussion of minor themes?	Pg 80-94

Appendix F

GRIPP 2 Short Form for PPI

Section and Topic		Reported on page No
1. Aim	Report the aim of PPI in the study	73/74
2. Methods	Provide a clear description of the methods used for PPI in the study	73/74/75/76
3. Study Results	Outcomes—Report the results of PPI in the study, including both positive and negative outcomes	99, 100, 124
4. Discussion and Conclusions	Outcomes—Comment on the extent to which PPI influenced the study overall. Describe positive and negative effects	99, 100, 124
5. Reflections/Critical Perspective	Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	124, 126, 127
PPI= Patient and Public Involvement		

Appendix G

Study Advertisement

Exploring Psychological Factors Involved in Cluster Headache Pain

Often called 'Suicide Headache' due to the severity of the pain; this condition can result in poor mental health, loss of employment, loss of relationships, and reduced quality of life.

Are you living with Cluster Headache?

Help us find out more about how psychology services could better support you.

The study will involve an **interview**, and the completion of three **questionnaires.** Location: online, at UEA, or at your home.

You will receive a £10 Love2Shop Voucher!

To take part you must:

- Have been diagnosed by a medical professional as a Chronic, or Episodic sufferer currently experiencing Cluster Attacks (i.e., not in remission).
- Not have any other facial / head pain.
- Be a UK resident, 18+ years, Fluent in English.
- Provide the details of your GP so researchers can keep you safe.

Interested?

This study is doctoral research at the University of East Anglia. Contact h.whitley@uea.ac.uk to find out more.

Appendix H

Interview Schedule

Settling in Question.

- Thank you for coming today. I have got lots of things I would like to ask you, but I would love to hear a bit about yourself.

Grand Tour Question.

- As you know, the focus of this research is cluster headaches. Can you tell me about what the experience is like for you?

Possible Follow-Up Questions: can you give an analogy?

Cognitions about the Pain.

- I am curious, what do you think about your cluster headaches?

Possible Follow-Up: what thoughts do you have whilst experiencing the acute attack?

Possible Follow-up: what thoughts do you have between the attacks?

- What do you think other people think about the pain?

Possible Follow-up: differentiate between friends/family and healthcare professionals

- Do you think you perceive yourself as capable of managing the pain/recovering?
- What are your expectations for your recovery?

Feelings about the pain.

- I am curious, what do you think about your cluster headaches?

Possible Follow-Up: what feelings do you have whilst experiencing the acute attack?

Possible Follow-up: what feelings do you have between the attacks?

Behavioural Response and Impact of the Pain.

- How do your cluster headaches affect you at the time of the attack?

Possible Follow-up: What do you do in response to the pain?

- How do you think your pain impacts your life?

Possible Follow-up: family, work, and social life?

Coping Strategies/Treatment.

- Has anything helped you with your cluster headaches?

Potential Follow-up: Consider medication, surgery, psychological and mental health support, dentistry, charities, participant's own actions (e.g., distraction, relaxation)

- Has anything not helped you with your cluster headaches?
- We want to know how the NHS can help people with cluster headaches. What do you think would help you?

Potential Follow-Up: Differentiate between treatment for the acute attack and period between attacks

Summary/Debrief.

- Thank you for answering all my questions. Is there anything else you think I should know?

Appendix I

Questionnaires Collected in Empirical Paper (Chapter Four)

Pain Catastrophising Scale (Sullivan, 1995, 2001, 2004, 2006, 2009)

ID Identifier:

Everyone experiences painful situations at some point in their lives. Such experiences may include headaches, tooth pain, joint or muscle pain. People are often exposed to situations that may cause pain such as illness, injury, dental procedures, or surgery.

INSTRUCTIONS We are interested in the types of thoughts and feelings that you have when you are in pain. Listed below are thirteen statements describing different thoughts and feelings that may be associated with pain. Using the following scale, please indicate the degree to which you have these thoughts and feelings when you are experiencing pain.

Rating:	Not at all (0)	To a slight degree (1)	To a moderate degree (2)	To a great degree (3)	All the time (4)
When I'm in pain....	0	1	2	3	4
I worry all the time about whether the pain will end	0	1	2	3	4
I feel I can't go on	0	1	2	3	4
It's terrible and I think it's never going to get better	0	1	2	3	4
It's awful and I feel that I overwhelms me	0	1	2	3	4
I feel I can't stand it anymore	0	1	2	3	4
I become afraid that the pain will get worse	0	1	2	3	4

I keep thinking of other painful events	0	1	2	3	4
I anxiously want the pain to go away	0	1	2	3	4
I can't seem to get it out of my mind	0	1	2	3	4
I keep thinking about how much it hurts	0	1	2	3	4
I keep thinking about how badly I want the pain to stop	0	1	2	3	4
There's nothing I can do to reduce the intensity of the pain	0	1	2	3	4
I wonder whether something serious may happen	0	1	2	3	4

Pain Self Efficacy Questionnaire (PSEQ) (Nicholas, 1989)

ID Identifier:

Please rate how confident you are that you can do the following things at present, despite the pain. To indicate your answer, mark one of the options on the scale under each item, from "not at all confident" to "completely confident".

	Not at all confident (0)	(1)	(2)	(3)	(4)	(5)	Completely confident (6)
I can enjoy things, despite the pain.	0	1	2	3	4	5	6
I can do most of the household chores (e.g., tidying up, washing	0	1	2	3	4	5	6

dishes, etc.), despite the pain							
I can socialise with my friends or family members as often as I used to do, despite the pain.	0	1	2	3	4	5	6
I can cope with my pain in most situations.	0	1	2	3	4	5	6
I can do some form of work, despite the pain. ('work' includes housework, paid and unpaid work)	0	1	2	3	4	5	6
I can still do many of the things I enjoy doing, such as hobbies or leisure activity, despite pain	0	1	2	3	4	5	6
I can cope with my pain without medication	0	1	2	3	4	5	6
I can still accomplish most of my goals in life, despite the pain.	0	1	2	3	4	5	6
I can live a normal lifestyle, despite the pain	0	1	2	3	4	5	6
I can gradually become more active, despite the pain.	0	1	2	3	4	5	6

Appendix J

Participant Information Sheet and Initial Consent Form

Participant Information Sheet.

Exploring Psychological Factors Involved in Cluster Headache Pain; a Qualitatively Driven Mixed-methods Study.

December 2023, Version Number 2

Introduction

You are invited to take part in a research study being run by the clinical psychology department at the University of East Anglia. This study hopes to explore cluster headache pain, and the experience for individuals living with this condition. The aim is that with more understanding of cluster headache, mental health services in the NHS can be improved to best meet the needs of this patient group. The study will involve interviewing individuals currently living with cluster headaches and asking them to complete several questionnaires about their experience. Before deciding whether you would like to apply to take part, please read the below information about what the study will entail.

If you have further questions, please feel free to contact the lead researcher using the contact details at the bottom of this sheet. Thank you for your interest in this study.

The Researchers

My name is Helena Whitley. I am a Trainee Clinical Psychologist at the University of East Anglia (UEA) and am doing this research as part of my Doctoral qualification. I am co-producing this research with Wayne Nolan, who is a gentleman who is a chronic Cluster Headache sufferer. His insight into the research is invaluable to ensure the research is best meeting the needs of individuals with cluster headache. The research is being supervised by Dr Elisabeth Norton (Clinical Psychologist).

What is the Purpose of the Research?

Cluster headache pain is known to be one of the most painful conditions experienced by humans; anecdotally more painful than childbirth and a gunshot wound. The condition results in recurring headache 'attacks' which can occur daily and last between minutes to hours. The condition results in reduced quality of life, loss of employment, difficulty in maintaining social relationships and is associated with anxiety and depression. This condition is generally treated through medication. There is an absence of evidence available surrounding other treatment options for this condition and so an absence of other services set up specifically for this group. This study hopes to explore whether psychology services could support this group of patients.

Who is being Invited to Participate?

We are looking for 15 adults who are currently living with cluster headache to take part in our study. The cluster headache type can be either episodic or chronic. Due to the study involving an interview, we are looking for individuals who can speak English fluently. To ensure the study is specific to cluster headache pain, if you are also experiencing other head or facial pain, unfortunately you will

not be eligible to take part. To keep you safe throughout the research, it is also a requirement of the study that we have your GP details. We will only contact your GP if we feel you are not able to keep yourself safe. This is something we would discuss with you before making contact.

Should I take part?

Participation in this study is completely voluntary. If you wish to take part, and fill in the attached consent form, the lead researcher (Helena Whitley) will email you to arrange a short phone call to ensure you are eligible, and if so, organise your participation in the study.

Feel free to take your time in considering if you wish to take part. You can contact the lead researcher to ask any questions before making your decision (h.whitley@uea.ac.uk). Also, you may find it helpful to speak to someone independent of the study; for example, a friend, family member, or healthcare professional involved in your care.

What will happen if I take part?

If you indicate that you are interested in taking part by sending back the attached initial consent form, you will be called to discuss the study. You will have the opportunity to ask any questions you have. The researcher will also ask some brief questions about your cluster headache pain to ensure you are eligible to take part. If deemed eligible, you will then be asked you to fill out a form giving consent to take part in the study. This consent form will be sent out via email for you to return. If you are not eligible to take part, the researcher will delete your email address and contact details to protect your confidentiality.

After consent is given, the lead researcher (Helena Whitley) will arrange with yourself whether it would be better for the interview to take place in person at your home, over the telephone, or over video. The research team will make themselves flexible to your needs, in terms of when would work best for you in relation to your cluster headache pain patterns. The interview can either take place over one session, or over multiple; again, depending upon what works best for you. If completed in one go, the interview should take around 45 minutes to one hour. The interview will be audio-recorded. In the interview session the researcher will also ask some brief demographic questions to contextualise the study results. They will ask your age, gender, employment status, type of cluster headache, and geographical location in the UK.

As well as the interview part of the study, you will be asked to complete three questionnaires related to your experience of cluster headache. It should take around 15 minutes to complete these questionnaires. These questionnaires will be completed at a separate time to the interview to account for fatigue from the interview process. Again, we will be flexible to your needs as to whether this is completed in person or over the phone/video.

After the interview/questionnaires, the researcher will discuss if you would like to be signposted to any support services.

Can I stop taking part if I change my mind?

Before the study is carried out, you can choose to withdraw yourself at any point. You can also decide part-way through the process to discontinue. After the interview/questionnaires are complete, the research team will transcribe the interview and send a copy to yourself. From this point, you will have 14 days to withdraw permission for your data to be used, or to state that you wish certain sections of the interview to be removed/changed. Importantly, if you do withdraw your information after the interviews have been completed, the researcher will naturally be able to remember the information you told them, and so whilst direct information can be excluded, this may inadvertently influence the results.

Will my taking part in this study be anonymous and kept confidential?

All the interview and questionnaire data collected throughout the study will be secured safely on a password protected computer. Only the research team will have access to this data. This data will not be linked to your identifiable information, and instead you will have a participant ID number. On a document separate to the data, your name and ID number will be linked. Therefore, if you do wish to withdraw, we can then link your data at this point and remove it from the study. All identifiable information about yourself will be removed when writing up the study, and so the interview transcript and questionnaire data will not be linked to yourself. We will do our utmost to remove enough details in the report to not allow quotes to be identifiable to yourself. Importantly, the cluster headache community is small, and so this cannot be guaranteed. This is why you will have the opportunity to read the transcript before the report is written up to check you are happy with the content. As stated above, you will have 14 days to let us know if you would like anything to be changed in the transcripts. After this point we will begin analysis of the transcriptions, and so it will be more difficult to remove your content as it will not be linked directly to yourself. This is why there is this 14-day deadline.

Once the study is complete, all research data will be stored and archived at the University of East Anglia for 10 years and will then be deleted. This information will all be deleted from the UEA OneDrive once transferred to the archive. This does not include the audio-files of your interview however, which will be deleted as soon as the transcriptions are complete and will not be stored in the archives. Only transcriptions of your interviews, which you have checked and edited, will be stored in the archives. The researcher will keep a document of your email addresses on the UEA OneDrive for up to two years after the study finishes, so they can contact you with the published research. After you have received this publication, your contact details will be deleted.

What will happen to the results of the research study?

The information collected through this study will be written up as a doctoral thesis and will then be submitted for publication in an appropriate journal. Participants will not be identified in the thesis or published work. As stated above, you will be given the option to receive a copy of this publication when it is complete.

What are the possible disadvantages and risks of taking part?

The interview will involve talking about your experience of cluster headache. This may bring up distressing topics. Also, you may find the process of completing the interview and questionnaires tiring. You are free to withdraw from the study at any time, and we can also pause the interview and restart at another point to account for this. If you feel distressed throughout the study, or after, we will also signpost you to various support services. For example, your General Practitioner (GP), or mental health charities such as the Samaritans who are available 24 hours a day for support (116 123, https://samaritans.org/). As part of the agreement to take part in the study, we ask that you give us your GP details. Therefore, if we feel you are vulnerable and in need of support, we will make contact for you. We will let you know if we are going to contact your GP.

Importantly, if we are concerned about your safety, we will contact your GP even if you do not give permission at the time. This is for your own safety, and we ask at the beginning of the study for you to consent to this.

What are the potential benefits of taking part?

You may find it validating to have the opportunity to speak about your experience. Furthermore, you will be contributing to important research into cluster headache, and so be part of the move to improving the treatments available for the cluster headache community.

To thank you for your time, you would receive a £10 Love2Shop gift card. You will receive this as soon as you have completed the interview and completed the questionnaires, even if you then choose to withdraw your consent.

What if there is a problem?

If you have any questions about the study, feel free to email the lead researcher and they will try to answer your questions (h.whitley@uea.ac.uk). Alternatively, you can contact their supervisor Elisabeth (Elisabeth.norton@uea.ac.uk). If you would like to make a formal complaint throughout the study, you can contact the Clinical Psychology department team (clinpsyd@uea.ac.uk).

Are you interested?

If still interested after reading this form, please fill in the attached consent form. Helena Whitley will then contact you to ensure you are eligible to take part and ask any further questions you may have.

Please contact Helena Whitley as soon as possible as we will recruit on a first-come-first-served basis. After we have got 15 participants, we will therefore stop recruitment. If you contact the researcher when recruitment is over, they will delete your email to protect your confidentiality.

Miss Helena Whitley

Doctoral Programme in Clinical Psychology,

Department of Psychological Sciences

Norwich Medical School

University of East Anglia

h.whitley@uea.ac.uk

Dr Elisabeth Norton

Doctoral Programme in Clinical Psychology,

Department of Psychological Sciences

Norwich Medical School

University of East Anglia

Elisabeth.norton@uea.ac.uk

University of East Anglia

Initial Consent Form

Title of Project: Psychological factors and cluster headache pain; a qualitatively driven mixed-methods study

December 2023, Version Number 1

Name of Researchers: Miss Helena Whitley and Dr Elisabeth Norton

Please put your initials in each box to show your consent.

I confirm that I have read the information sheet for the above study and am interested in taking part. ☐

I consent to be contacted by the researcher, Helena Whitley, by email and phone to discuss the study, and check I am eligible to take part. ☐

I understand that my phone number and email address will be stored securely and only available to Helena Whitley, the lead researcher. ☐

I understand that if I decide to not take part/I am not eligible to take part, my number and email address will be deleted immediately. ☐

By providing my phone number and email address below I am consenting to be contacted by Helena Whitley, the lead researcher. ☐

Phone number…….

Email Address…….

Name of Participant	Date	Signature
Name of Researcher	Date	Signature
Helena Whitley	January 2024	HEJWhitley

Appendix K

Final Consent Form

Consent Form

Title of Project: Psychological factors and cluster headache pain; a qualitatively driven mixed-methods study

December 2023, Version Number 2

Name of Researchers: Miss Helena Whitley and Dr Elisabeth Norton

Please place your initials in each box if you consent to the below statements.

I confirm that I have read the information sheet for the above study. I have had the opportunity to consider the information, ask questions, and have had these answered satisfactorily. ☐

I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, and without any consequences. ☐

I understand that the information about me collected in this study, whilst anonymised, will be used in a written report which will be published in a relevant journal. ☐

I understand that the information collected about me will not be anonymised to the researchers on the study as they will be carrying out the interview and completing the questionnaires with me. ☐

I agree that if I disclose anything to the researcher which suggests I am a danger to myself or others, my GP can be contacted, and in an emergency an ambulance called. ☐

I understand that whilst I can withdraw my information at any time, the researcher will not be able to forget the information I provide, and this may inadvertently influence the study results. ☐

I agree to take part in the above study. ☐

My GP Contact Details

Name of Participant	Date	Signature
Name of Researcher	Date	Signature
Helena Whitley	*11/12/2023*	*HEJWhitley*

Appendix L

Participant Debrief Form

Debrief Form

The Psychological Factors Involved in the Experience of Cluster Headache Pain; a Qualitatively Driven Mixed-Methods Study
2024

Thank you for your participation in our research study. This study, conducted by the University of East Anglia (UEA), aims to explore the experience of cluster headache pain, and so deepen our understanding of the condition. The hope is that through understanding more about the experience of pain in this headache condition, we can better understand how mental health services in the UK can be improved to support this group. You have completed an interview and various questionnaires around the topic of your experience of cluster headache. This data will now be analysed and used to meet the above aims.

What happens now?

Your active part in the research study has now come to an end. As a gesture of our appreciation for your time, you will now be given a £10 Love2Shop Voucher.

The research team will now transcribe your interviews. Once the interviews have been transcribed, we will send you a transcript of your interview. Please look over the transcription, and let the researchers know if you would like to remove/change anything. You will have ***two weeks (14 days) to contact us, and if we do not hear from you, we will assume you are happy for all the information to be included without edit.*** Once the interviews have been transcribed, the audio recording will be deleted.

After analysing the data, the lead researcher will write up the findings as part of their Doctoral qualification. This will involve writing a doctoral thesis and then submitting it to an appropriate journal. You will not be identified in the thesis or published work.

If you consented to receive the results of the study when you signed the consent form, you will receive the final report by email. If you did not consent before, but would now like the results, please let the researcher know now.

Once the study is complete, research data will be stored and archived in the University of East Anglia archives for 10 years. Once transferred to the archives, the research data will all be deleted from the researcher's work laptop. This is normal practice. This does not include the audio-files of your interview however, which will be deleted as soon as the transcriptions are complete. For the researcher to contact you with the published research document, they will keep your email address on a secure password protected document for up to two years, to allow time for publication. Once you have received the published work, your email address will be deleted.

All identifiable information about yourself will be removed when writing up the study, and the interview transcript and questionnaire data will not be linked to yourself. We will do our utmost to remove enough details in the report to not allow quotes to be identifiable to yourself. Importantly, the cluster headache community is small, and so this cannot be guaranteed. This is why you will have the

opportunity to read the transcript before the report is written up to check you are happy with the content.

Support Services

We are aware that speaking about your difficult experience may be challenging for you. If you would like to discuss this, please reach out to one of the following:

1. If you feel you need to speak to someone in an emergency, please dial 999. If you are with the researcher when reading this, let them know and they can call 999 with you.

2. If you feel you are in crisis, experiencing feelings of despair or are suicidal, contact the Samaritans on 116 123. Or for urgent help, call NHS 111. These services are available for free 24 hours a day.

3. Your GP

4. If you would like support with your wellbeing, you can contact your local Talking Therapies Service. They offer one-to-one therapy and counselling. They also run various workshops, including workshops on managing chronic pain.

2. Contact the advice line at the charity OUCH. You can access this by calling their advice line on 0800 6696 824, emailing info@ouchuk.org or by looking on their website: https://ouchuk.org/

If you have any questions about receiving support, please ask Helena Whitley after the interview. Alternatively, you can email them on h.whitley@uea.ac.uk. They will try to direct you to what is most helpful.

If you have questions
The main researcher conducting this study is Helena Whitley, a trainee Clinical Psychologist, at the University of East Anglia. Please ask any questions you have now. If you have questions later, you may contact Helena Whitley on h.whitley@uea.ac.uk. Alternatively, you can contact their supervisor Dr Elisabeth Norton on (Elisabeth.norton@uea.ac.uk). If you have any questions or concerns or want to make a compliant, you can contact the Clinical Psychology department team (clinpsyd@uea.ac.uk).

Thank you again for your participation; it is hugely valued by the research team and by the whole Cluster Headache Community.

Dr Elisabeth Norton

Doctoral Programme in Clinical Psychology, Department of Psychological Sciences. Norwich Medical School

University of East Anglia

Elisabeth.Norton@uea.ac.uk

Miss Helena Whitley

Doctoral Programme in Clinical Psychology, Department of Psychological Sciences, Norwich Medical School

University of East Anglia

h.whitley@uea.ac.uk

Appendix M

Ethical Approval from the Faculty of Medicine and Health Sciences Ethics Committee at the University of East Anglia

University of East Anglia
Norwich Research Park
Norwich. NR4 7TJ

Email: ethicsmonitor@uea.ac.uk
Web: www.uea.ac.uk

Study title: Exploring Psychological Factors Involved in Cluster Headache Pain; a Qualitatively Driven Mixed-methods Study

Application ID: ETH2324-0070

Dear Helena,

Your application was considered on 3rd November 2023 by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee).

The decision is: **approved**.

You are therefore able to start your project subject to any other necessary approvals being given.

If your study involves NHS staff and facilities, you will require Health Research Authority (HRA) governance approval before you can start this project (even though you did not require NHS-REC ethics approval). Please consult the HRA webpage about the application required, which is submitted through the IRAS system.

This approval will expire on **1st June 2025**.

Please note that your project is granted ethics approval only for the length of time identified above. Any extension to a project must obtain ethics approval by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) before continuing.

It is a requirement of this ethics approval that you should report any adverse events which occur during your project to the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) as soon as possible. An adverse event is one which was not anticipated in the research design, and which could potentially cause risk or harm to the participants or the researcher, or which reveals potential risks in the treatment under evaluation. For research involving animals, it may be the unintended death of an animal after trapping or carrying out a procedure.

Any amendments to your submitted project in terms of design, sample, data collection, focus etc. should be notified to the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) in advance to ensure ethical compliance. If the amendments are substantial a new application may be required.

Approval by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) should not be taken as evidence that your study is compliant with the UK General Data Protection Regulation (UK GDPR) and the Data Protection Act 2018. If you need guidance on how to make your study UK GDPR compliant, please contact the UEA Data Protection Officer (dataprotection@uea.ac.uk).

Please can you send your report once your project is completed to the FMH S-REC (fmh.ethics@uea.ac.uk).

I would like to wish you every success with your project.

On behalf of the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee)

Yours sincerely,

Dr Paul Linsley

Appendix N

Participants' Scores on Pain Catastrophising Scale (PCS) and Pain Self-Efficacy Questionnaire (PSEQ) from Empirical Paper (Chapter Four).

Participant	PCS	PSEQ
1	39	34
2	30	3
3	45	10
4	36	4
5	25	3
6	41	0
8	16	14
9	13	48
10	35	3
11	16	36
13	42	10
Mean score (range of scores)	30.7 (0-52)	15 (0-60)

Appendix O

Example of Thematic Analysis Process for the Darkness Theme

Stage One: Quotes (in brackets are codes drawn from quotes)

>Participant 12: "It was like someone had a curse on you […] You know that expression, "the dark night of the soul?" (It's like someone had cursed you) **(The dark night of the soul)**

>Participant 12: This sounds very odd. It's like (1 second pause). It's like (1 second pause) it's out to get ya. Like a really. Like it's coming for ya […] like you're having a bad dream or a nightmare […] There's this atmosphere. **(Dark atmosphere) (It's like the headache is out to get you, coming for you)**

>Participant 13: "A cluster headache is an attack of being smited [...] it is the enemy and you always need to be looking out for it to try and stop it, but often it defeats you." **(Cluster headache is an attack of being smited) (enemy)**

>Participant 7: But the night times are the ones which are just night after night after night. So it it's a different psychology because I'm in bed when these are happening or I go to bed when these are happening, you know, go in my bedroom that's it […] Shut the house down all the lights off. Erm. So. The the nights I, I've never say, "I didn't look forward to going to bed because they'll be a cluster coming" I just couldn't think like that. It's just I needed to go to bed and hope to goodness tonight would be one of them where I didn't wake up in a horror show with **(Night-time different psychology) (Horror), (Monster), (Shut the house down, all the lights off)**

>Participant 6: The beast comes and you're fighting the beast and it's trying to take control. Yeah it's it's an inner demon I suppose **(The beast comes and you are fighting the beast) (Inner-demon)**

>Participant 8: because in my long history with this damned curse. And that's what it's like. It's like being cursed, I think [...] I absolutely hate it [...] it's like having an enemy that attacks you **(damned curse) (enemy)**

>Participant 7: It comes on whether I like it or not. It's got a mind of its own. **(CH has a mind of it's own)**

>Participant 11: That's like a punishment **(Punishment)**

>Participant 12: “But I'm getting up 3 or 4 times a night […] You're not. I'm not getting good sleep (1 second pause) having to get up quietly so I don't know wake my wife up. Creep around the house **(Creep around the house)**

Stage Two: Example Codes for Darkness Theme

-Dark atmosphere, call it shadow, grey fuzz, smothering, descends on hemisphere of your head

-Dark night of the soul

-It's like the headache is out to get you, coming for you

-Different state of being

-You feel alone

-The monster

-Horror

-Room on own when in attack

-Most at night-time, and it's a different psychology, go into bedroom, and know that's it, shut down the house, turn the lights off

-Some people call it 'the beast'

-When it's there, I want it to go away, it's horrible and evil

-The beast comes and you are fighting the beast

-Inner-demon

Mind goes to dark places in acute phase, is this my fault, sent by God, lesson

If I don't wake up in the morning, won't have the pain

Please kill me, I can't bear this anymore, if pill to kill would take it

What why why would this happen

Please just kill me because I can't bear it anymore

Suicidal thoughts

If I don't wake up in the morning, won't have the pain

Please kill me, I can't bear this anymore, if pill to kill would take it

Cluster headache is an attack of being smited, humour, takes horror out of it

Call it monster, or many other foul words

8 months of really hellish time with up to six attacks a day

Doctor said must have upset a witch

Damned curse

Night-time different psychology

Painting, it blew me away, most powerful thing I have seen, sums up cluster headaches

Some people call it 'the beast'

Whatever negative thoughts I have are about releiving the pain

In attack, thinking how difficult the pain is and where you will be proded next

In attack, you feel a bit sorry for yourself, and think 'why me' 'why have i got these' 'why can't someone else suffer', selfish thoughts

Oh my gosh, what if this never get's better

Pain triggers emotional pain about pain, disturbs you, feeling can trigger dwelling on difficult times in life, childhood

Why me thoughts

Try to challenge the thought, 'this is a thought'

In attack everything is going through my head about why it might not be getting better

Clock watch throughout attack, it should have stopped by now. why is it not stopping

Creep around house, be quiet, not disturb wife asleep

Want to be in a darkened room

In bedroom, dark, cold ideal scenario

In darkened room with oxygen next to bed

In attack, lie down, curtains closed

All could do was sit in a darkened room and wait for them to pass

Normally I'm in a relaxed position, close eyes, darken room, jog feet up and down

You feel alone

Most at night time, and it's a different psychology, go into bedroom, and know that's it, shut down the house, turn the lights off

Absolutely hate it, like having enemy that attacks you, your own brain. You can understand why I thought I was mad

CH has a mind of it’s own

Creep around house, be quiet, not disturb wife asleep, triggered relaxing

Dark atmosphere, call it shadow, grey fuzz, smothering, descends on hemisphere of your head

Dark night of the soul

Caged animal, pace up and down, claw at my skin

Hear a thunding clap on clear skies and then clouds form and then storm within 40 minutes

I don't hate the monster, because if I do I'm hating myself

Some people call it 'the beast'

When it's there, I want it to go away, it's horrible and evil

The beast comes and you are fighting the beast

The pain is something and it's got hold of you

Sort of pain where you can't get away from it

It was like someone had cursed you

Punishment

It is an enemy, often defeats you

When feel niggle, 5 minutes until reaches fever pitch, so you need your oxygen, too late for tablet

It is part of you, it's not an alien, I call it the monster, it's a bit of me which hurts, I don't know if it knows it's a monster or not, entity in itself, positive thinking attitude

It is stupid scary pain

It's like the headache is out to get you, coming for you

You feel like your body doesn't belong to you.

You have to give way to it, pacify it, how dress, behave, walk talk. Even though it controls you, you have to control it

In that intensity of pain. Something about your cognition switches. Different state of being

Stage Three: Clustered Codes into Similar Concepts

1. Dark Atmosphere

-Dark atmosphere, call it shadow, grey fuzz, smothering, descends on hemisphere of your head

-Dark night of the soul

-It's like the headache is out to get you, coming for you

-Different state of being

-Some people call it 'the beast'

-When it's there, I want it to go away, it's horrible and evil

-The beast comes and you are fighing the beast, and it's trying to take control, inner-demon, issue with fighting and talking to yourself

2. Dark Thoughts

Mind goes to dark places in acute phase, is this my fault, sent by God, lesson

If I don't wake up in the morning, won't have the pain

Please kill me, I can't bear this anymore, if pill to kill would take it

What why why would this happen

Whatever negative thoughts I have are about releiving the pain

In attack, thinking how difficult the pain is and where you will be proded next

In attack, you feel a bit sorry for yourself, and think 'why me' 'why have i got these' 'why can't someone else suffer', selfish thoughts

Oh my gosh, what if this never get's better

Pain triggers emotional pain about pain, disturbs you, feeling can trigger dwelling on difficult times in life, childhood

Why me thoughts

Try to challenge the thought, 'this is a thought'

In attack everything is going through my head about why it might not be getting better

Clock watch throughout attack, it should have stopped by now. why is it not stopping

3. Physically Dark

Creep around house, be quiet, not disturb wife asleep

Want to be in a darkened room

In bedroom, dark, cold ideal scenario

In darkened room with oxygen next to bed

In attack, lie down, curtains closed

All could do was sit in a darkened room and wait for them to pass

Normally I'm in a relaxed position, close eyes, darken room, jog feet up and down

You feel alone

Most at night time, and it's a different psychology, go into bedroom, and know that's it, shut down the house, turn the lights off

Shut the house down, all the lights off

4. Description like Enemy

Absolutely hate it, like having enemy that attacks you, your own brain. You can understand why I thought I was mad

Creep around house, be quiet, not disturb wife asleep, triggered relaxing

Dark atmosphere, call it shadow, grey fuzz, smothering, descends on hemisphere of your head

Dark night of the soul

CH has a mind of it's own

Caged animal, pace up and down, claw at my skin

Hear a thunding clap on clear skies and then clouds form and then storm within 40 minutes

I don't hate the monster, because if I do I'm hating myself

Some people call it 'the beast'

When it's there, I want it to go away, it's horrible and evil

The beast comes and you are fighing the beast, and it's trying to take control, inner-demon, issue with fighting and talking to yourself

The pain is something and it's got hold of you

Sort of pain where you can't get away from it

It was like someone had cursed you

Punishment

It is an enemy, often defeats you

When feel niggle, 5 minutes until reaches fever pitch, so you need your oxygen, too late for tablet

It is part of you, it's not an alien, I call it the monster, it's a bit of me which hurts, I don't know if it knows it's a monster or not, entity in itself, positive thinking attitude

It is stupid scary pain

It's like the headache is out to get you, coming for you

You feel like your body doesn't belong to you.

You have to give way to it, pacify it, how dress, behave, walk talk. Even though it controls you, you have to control it

In that intensity of pain. Something about your cognition switches. Different state of being

<u>5. Physically dark</u>

Want to be in a darkened room

In bedroom, dark, cold ideal scenario

In darkened room with oxygen next to bed

In attack, lie down, curtains closed

All could do was sit in a darkened room and wait for them to pass

Mind goes to dark places in acute phase, is this my fault, sent by God, lesson

Normally I'm in a relaxed position, close eyes, darken room, jog feet up and down

6. Very lonely illness

You feel alone

Room on own when in attack

Most at night time, and it's a different psychology, go into bedroom, and know that's it, shut down the house, turn the lights off

7. Suicidal in Moment

Please just kill me because I can't bear it anymore

Suicidal thoughts

If I don't wake up in the morning, won't have the pain

Please kill me, I can't bear this anymore, if pill to kill would take it

8. Externalisation/humour/language to explain the CH

Cluster headache is an attack of being smited, humour, takes horror out of it

Call it monster, or many other foul words

8 months of really hellish time with up to six attacks a day

Doctor said must have upset a witch

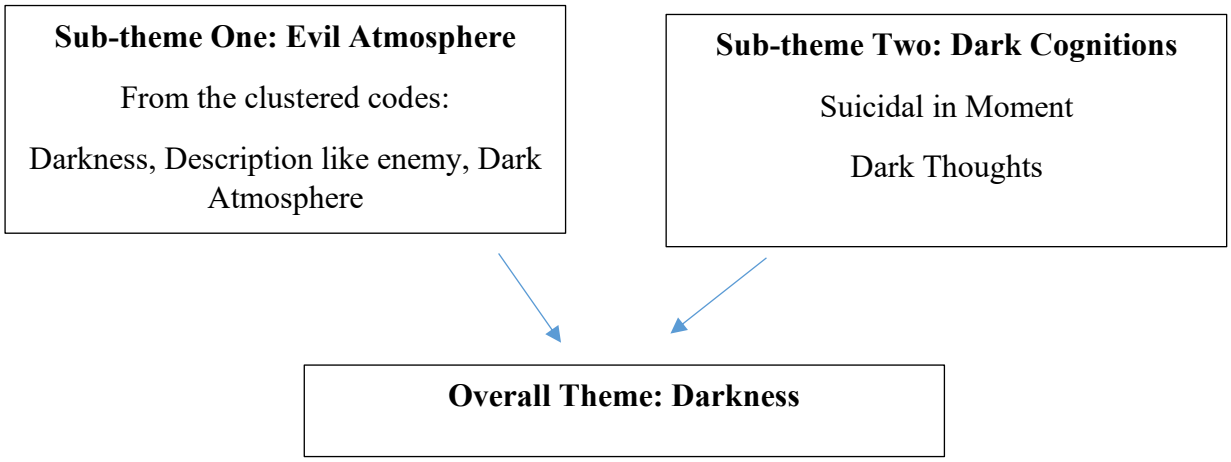
Damned curse

Painting, it blew me away, most powerful thing I have seen, sums up cluster headaches

Some people call it 'the beast'

Psychological intervention to help people find phrase or visual, to help weather storm

Stage 4: Sub-Themes, from clustered codes



Reflections

"This theme feels powerful, and emotive" (HW)

"I feel a real sense of responsibility to make sure the participants' experiences are highlighted correctly" (HW)

"Perhaps the fact the clusters arrive at night, is why it feels so dark" (HW)

"Is this a superordinate theme?"

"I feel drawn to the 'darkness of the soul' quote

Appendix P

Author guidelines for the Journal of Headache and Pain

https://thejournalofheadacheandpain.biomedcentral.com/submission-guidelines/preparing-your-manuscript/research

Aims and scope

The Journal of Headache and Pain is a peer-reviewed open access journal published under the brand BMC, part of Springer Nature. It is specifically dedicated to researchers involved in all aspects of headache and related pain syndromes, including epidemiology, public health, basic science, translational medicine, clinical trials and real-world data.
With a multidisciplinary perspective, *The Journal of Headache and Pain* covers headache medicine and related pain syndromes in all medical disciplines and particularly encourages clinical, translational and basic science submissions in the areas of pain management, genetics, neurology and internal medicine. The journal publishes research articles, reviews, letters to the Editor as well as consensus articles and guidelines, which promote best practice in the management of patients with headaches and related pain.

Preparing main manuscript text

Quick points:

Use double line spacing

Include line and page numbering

Use SI units: Please ensure that all special characters used are embedded in the text, otherwise they will be lost during conversion to PDF

Do not use page breaks in your manuscript

File formats

The following word processor file formats are acceptable for the main manuscript document:

Microsoft word (DOC, DOCX)

Rich text format (RTF)

TeX/LaTeX

Please note: editable files are required for processing in production. If your manuscript contains any non-editable files (such as PDFs) you will be required to re-submit an editable file when you submit your revised manuscript, or after editorial acceptance in case no revision is necessary.

Criteria

Research articles should report on original primary research.

The Journal of Headache and Pain strongly encourages that all datasets on which the conclusions of the paper rely should be available to readers. We encourage authors to ensure that their datasets are either deposited in publicly available repositories (where available and appropriate) or presented in the main manuscript or additional supporting files whenever possible. Please see Springer Nature's information on recommended repositories.
The Journal of Headache and Pain **strongly encourages** authors to submit a **visual abstract** along with their manuscripts.
A visual abstract is a figure that clearly and succinctly conveys the main message of your research (paper). The goal of a visual abstract is to attract readers' attention to the article and encourage them to read the whole paper, but also promote interdisciplinary scholarship and help readers quickly identify which papers are most relevant to their research interests. This will appear underneath to the Abstract on the website.
TECHNICAL SPECIFICATIONS: The visual abstract image should be 920x300 pixels and a maximum of 150KB, and should be submitted with your contribution as a separate file in a JPG, PNG, or SVG electronic format. Please name the picture file "Visual Abstract".

KEEP IT SIMPLE: Emphasize the new findings, highlight one process or make one point clear, use text sparingly and simple labels. Be also aware that effective use of color can enhance the graphical abstract both aesthetically and by directing the reader's attention to focal points of interest.
PROFESSIONALLY PRODUCED VISUAL ABSTRACTS: High quality and affordable visual abstracts are available through services provided at Springer Nature. As an author submitting at *The Journal of Headache and Pain* you are entitled to a 20% discount on this service. Click here to find out more about the service, and your discount will be automatically applied when using this link.
Preparing your manuscript

The information below details the section headings that you should include in your manuscript and what information should be within each section.

Please note that your manuscript must include a 'Declarations' section including all of the subheadings (please see below for more information).

Title page

The title page should:

- present a title that includes, if appropriate, the study design e.g.:
  - "A versus B in the treatment of C: a randomized controlled trial", "X is a risk factor for Y: a case control study", "What is the impact of factor X on subject Y: A systematic review"
  - or for non-clinical or non-research studies a description of what the article reports

- list the full names and institutional addresses for all authors
  - if a collaboration group should be listed as an author, please list the Group name as an author. If you would like the names of the individual members of the Group to be searchable through their individual PubMed records, please include this information in the "Acknowledgements" section in accordance with the instructions below
  - Large Language Models (LLMs), such as ChatGPT, do not currently satisfy our authorship criteria. Notably an attribution of authorship carries with it accountability for the work, which cannot be effectively applied to LLMs. Use of an LLM should be properly documented in the Methods section (and if a Methods section is not available, in a suitable alternative part) of the manuscript.
- indicate the corresponding author

Abstract

The Abstract should not exceed 350 words. Please minimize the use of abbreviations and do not cite references in the abstract. Reports of randomized controlled trials should follow the CONSORT extension for abstracts. The abstract must include the following separate sections:

- **Background:** the context and purpose of the study
- **Methods:** how the study was performed and statistical tests used
- **Results:** the main findings
- **Conclusions:** brief summary and potential implications
- **Trial registration:** If your article reports the results of a health care intervention on human participants, it must be registered in an appropriate registry and the registration number and date of registration should be stated in this section. If it was not registered prospectively (before enrollment of the first participant), you should include the words 'retrospectively registered'. See our editorial policies for more information on trial registration

Keywords

Three to ten keywords representing the main content of the article.

Background

The Background section should explain the background to the study, its aims, a summary of the existing literature and why this study was necessary or its contribution to the field.

Methods

The methods section should include:

- the aim, design and setting of the study
- the characteristics of participants or description of materials
- a clear description of all processes, interventions and comparisons. Generic drug names should generally be used. When proprietary brands are used in research, include the brand names in parentheses
- the type of statistical analysis used, including a power calculation if appropriate

Results

This should include the findings of the study including, if appropriate, results of statistical analysis which must be included either in the text or as tables and figures.

Discussion

This section should discuss the implications of the findings in context of existing research and highlight limitations of the study.

Conclusions

This should state clearly the main conclusions and provide an explanation of the importance and relevance of the study reported.

List of abbreviations

If abbreviations are used in the text they should be defined in the text at first use, and a list of abbreviations should be provided.

Declarations

All manuscripts must contain the following sections under the heading 'Declarations':

- Ethics approval and consent to participate
- Consent for publication
- Availability of data and materials
- Competing interests
- Funding
- Authors' contributions
- Acknowledgements
- Authors' information (optional)

Please see below for details on the information to be included in these sections.

If any of the sections are not relevant to your manuscript, please include the heading and write 'Not applicable' for that section.

Ethics approval and consent to participate

Manuscripts reporting studies involving human participants, human data or human tissue must:

- include a statement on ethics approval and consent (even where the need for approval was waived)
- include the name of the ethics committee that approved the study and the committee's reference number if appropriate

Studies involving animals must include a statement on ethics approval and for experimental studies involving client-owned animals, authors must also include a statement on informed consent from the client or owner.

See our editorial policies for more information.
If your manuscript does not report on or involve the use of any animal or human data or tissue, please state "Not applicable" in this section.

Consent for publication

If your manuscript contains any individual person's data in any form (including any individual details, images or videos), consent for publication must be obtained from that person, or in the case of children, their parent or legal guardian. All presentations of case reports must have consent for publication.

You can use your institutional consent form or our consent form if you prefer. You should not send the form to us on submission, but we may request to see a copy at any stage (including after publication).
See our editorial policies for more information on consent for publication.
If your manuscript does not contain data from any individual person, please state "Not applicable" in this section.

Availability of data and materials

All manuscripts must include an 'Availability of data and materials' statement. Data availability statements should include information on where data supporting the results reported in the article can be found including, where applicable, hyperlinks to publicly archived datasets analysed or generated during the study. By data we mean the minimal dataset that would be necessary to interpret, replicate and build upon the findings reported in the article. We recognise it is not always possible to share research data publicly, for instance when individual privacy could be compromised, and in such instances data availability should still be stated in the manuscript along with any conditions for access.

Authors are also encouraged to preserve search strings on searchRxiv https://searchrxiv.org/, an archive to support researchers to report, store and share their searches consistently and to enable them to review and re-use existing searches. searchRxiv enables researchers to obtain a digital object identifier (DOI) for their search, allowing it to be cited.
Data availability statements can take one of the following forms (or a combination of more than one if required for multiple datasets):

- The datasets generated and/or analysed during the current study are available in the [NAME] repository, [PERSISTENT WEB LINK TO DATASETS]
- The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.
- All data generated or analysed during this study are included in this published article [and its supplementary information files].
- The datasets generated and/or analysed during the current study are not publicly available due [REASON WHY DATA ARE NOT PUBLIC] but are available from the corresponding author on reasonable request.
- Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.
- The data that support the findings of this study are available from [third party name] but restrictions apply to the availability of these data, which were used under license for the

current study, and so are not publicly available. Data are however available from the authors upon reasonable request and with permission of [third party name].

- Not applicable. If your manuscript does not contain any data, please state 'Not applicable' in this section.

More examples of template data availability statements, which include examples of openly available and restricted access datasets, are available here.
BioMed Central strongly encourages the citation of any publicly available data on which the conclusions of the paper rely in the manuscript. Data citations should include a persistent identifier (such as a DOI) and should ideally be included in the reference list. Citations of datasets, when they appear in the reference list, should include the minimum information recommended by DataCite and follow journal style. Dataset identifiers including DOIs should be expressed as full URLs. For example:

Hao Z, AghaKouchak A, Nakhjiri N, Farahmand A. Global integrated drought monitoring and prediction system (GIDMaPS) data sets. figshare.
2014. http://dx.doi.org/10.6084/m9.figshare.853801
With the corresponding text in the Availability of data and materials statement:

The datasets generated during and/or analysed during the current study are available in the [NAME] repository, [PERSISTENT WEB LINK TO DATASETS].[Reference number]
If you wish to co-submit a data note describing your data to be published in *BMC Research Notes*, you can do so by visiting our submission portal. Data notes support open data and help authors to comply with funder policies on data sharing. Co-published data notes will be linked to the research article the data support (example).

Competing interests

All financial and non-financial competing interests must be declared in this section.

See our editorial policies for a full explanation of competing interests. If you are unsure whether you or any of your co-authors have a competing interest please contact the editorial office.
Please use the authors initials to refer to each authors' competing interests in this section.

If you do not have any competing interests, please state "The authors declare that they have no competing interests" in this section.

Funding

All sources of funding for the research reported should be declared. If the funder has a specific role in the conceptualization, design, data collection, analysis, decision to publish, or preparation of the manuscript, this should be declared.

Authors' contributions

The individual contributions of authors to the manuscript should be specified in this section. Guidance and criteria for authorship can be found in our editorial policies.

Please use initials to refer to each author's contribution in this section, for example: "FC analyzed and interpreted the patient data regarding the hematological disease and the transplant. RH performed the histological examination of the kidney, and was a major contributor in writing the manuscript. All authors read and approved the final manuscript."

Acknowledgements

Please acknowledge anyone who contributed towards the article who does not meet the criteria for authorship including anyone who provided professional writing services or materials.

Authors should obtain permission to acknowledge from all those mentioned in the Acknowledgements section.

See our editorial policies for a full explanation of acknowledgements and authorship criteria. If you do not have anyone to acknowledge, please write "Not applicable" in this section.

Group authorship (for manuscripts involving a collaboration group): if you would like the names of the individual members of a collaboration Group to be searchable through their individual PubMed records, please ensure that the title of the collaboration Group is included on the title page and in the submission system and also include collaborating author names as the last paragraph of the “Acknowledgements” section. Please add authors in the format First Name, Middle initial(s) (optional), Last Name. You can add institution or country information for each author if you wish, but this should be consistent across all authors.

Please note that individual names may not be present in the PubMed record at the time a published article is initially included in PubMed as it takes PubMed additional time to code this information.

Authors' information

This section is optional.

You may choose to use this section to include any relevant information about the author(s) that may aid the reader's interpretation of the article, and understand the standpoint of the author(s). This may include details about the authors' qualifications, current positions they hold at institutions or societies, or any other relevant background information. Please refer to authors using their initials. Note this section should not be used to describe any competing interests.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Figures, tables and additional files

See General formatting guidelines for information on how to format figures, tables and additional files.
Submit manuscript