Title:
The endocrine management of intractable masturbation after epilepsy surgery: A case report and literature review

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INTRODUCTION

We present the case of a man with intractable masturbation after epilepsy surgery effectively controlled by cyproterone acetate and haloperidol.

Case description

A 16-year-old man presented with a history of autism spectrum disorder with learning difficulties; and attention deficit hyperkinetic disorder.

Having been born at term following an uneventful pregnancy, he developed complex partial epilepsy aged sixteen weeks old occurring up to seventy times daily. He was initially optimized on oral anti-epileptic medication. However, his seizures became uncontrollable and age 12 he underwent a successful right temporal lobectomy and a radical amygdalo-hippocampectomy which significantly reduced his seizure frequency. However, he developed sexualized behaviour, characterized by incessant masturbation in any environment such that he was unable to interact with anyone outside his immediate family. Cognitive behavioural support proved unsuccessful. Aged 16, he was referred to the endocrinology and neuropsychology clinics.

Management

Physical examination showed incomplete pubertal development with Tanner stage 3 genitalia. There was no evidence his sexual behaviours was electro-neurophysiological in origins.

After neurodevelopmental and neuropsychiatric examination, it was felt that his behaviour may in part be androgen driven. His family consented to a trial of

cyproterone acetate 50mg once daily. Within two weeks the masturbation completely ceased and he was able to resume his social relationships.

His bone age at the start of treatment was reported as normal. The family were aware of the potential for the anti-androgenic effects on his bones but felt that given the clear benefits, cyproterone should be continued. The drug maintained excellent control for about three months and then the effect began to fade. In addition to the resurgence of the masturbation, he had developed recurrent symptoms of affective mood disorder. Low dose of haloperidol (3mg daily) was started for its prolactin mediated endocrine anti-libidinal side-effects and the dose of his cyproterone increased to 50mg in the morning and 25mg in the evening.

The combination of cyproterone and haloperidol successfully abolished the incessant masturbation in inappropriate settings. He now engages in socially appropriate masturbation in his room at a much reduced rate. However, his bone mineral density scan 4 years after starting his cyproterone showed T scores for his lumbar spine of -3.2 and of his left hip -2.9. Wrist X-rays assessing his bone age when repeated 5 years after starting his cyproterone showed epiphyseal fusion. Similarly, when plotted aged 14 years and 6 months, his height was just above the 0.4th centile and his height was on the 9th centile using standard UK charts for boys. Aged 17 years and 9 months his height and weight were both on the 9th centile. When last measured – aged 22 years and 10 months, his testosterone remained low at 1.1nmol/l (8.6-29.0).

DISCUSSION

We have described the successful treatment of a 12-year-old boy who developed hypersexualized conduct in the form of excessive masturbation following a right temporal lobe resection and right amygdalo-hippocampectomy for cortical dysplasia using a combination of cyproterone acetate and haloperidol.

It has been suggested that pharmacotherapy is unsuccessful in controlling seizures in 30-40% of patients with epilepsy (1). For those deemed suitable, surgery may be considered. However, hypersexuality has previously been reported as a recognised complication of epilepsy surgery (2-4). Such behaviours may be all consuming, leading to charges and convictions of sexual misconduct (3).

Temporal lobe resection is a common treatment for drug resistant epilepsy (1). However, it can be complicated by postoperative changes in sexual behaviour. Historically, findings of sexual dysfunction after bilateral anterior temporal lobectomy in male rhesus monkeys was first reported by Klüver and Bucy (3;5). Others have reported that the Klüver–Bucy syndrome is characterized by “excessive orality and hyperphagia, distractibility (especially for visual stimuli), hypersexuality and change in sexual preference, visual agnosia and loss of aggressive and fearful responses’’ (5;6).

This syndrome was first described in humans in 1955 by Ore et al following a temporal lobectomy (7). However, these reports are rare, with Ozmen et al described a 14-year-old girl who masturbated incessantly in inappropriate places after a left amygdalo-hippocampectomy (4).

In a study of personality changes following temporal lobectomy for epilepsy, more than half (15 out of 27) experienced increased sexual drive and potency (8). These postoperative changes in sexual behaviour or function have also led to abolition of pre-existing paraphilia as well as a decline in libido or sexual activity, with apparent restoration of “normal” sexual function (2). As with our patient, the onset of these changes in sexual behaviour occurred characteristically in the first three postoperative months (2).

Three potential mechanisms for the appearance of this hypersexual behaviour following temporal lobectomy have been proposed. The first is based on the notion that prior to surgery the uncontrolled epileptiform neuronal activity within the brain inhibits normal limbic function leading to preoperative hyposexuality. Following surgery, this removal of inhibitory signaling leads to a rebound in hypersexual behaviour (2;9). This would be consistent with the data from Braun et al, who describe the hemispheric contribution to libido (10). They suggest the intact right hemisphere inhibits libido and the left enhances it. They suggest that as a result of right temporal lobe resection or lesions that interfered with the normal functioning of the right temporal lobe would lead to hypersexuality and vice versa (10).

The second potential mechanism is that the lobe that is not operated on may have a degree of unrecognized pathological change which only comes to light with the removal of the more damaged lobe (2). The final proposed mechanism is that the physical expression of the hypersexual behaviour may be a normal part of the postoperative psychosocial adjustment (9). Given the disparate nature of the proposed mechanisms for developing hypersexual symptoms postoperatively

different treatment strategies have been tried. Ozmen et al successfully used 20mg citalopram in their case (4). Others have used a combination of quetiapine and sertraline with excellent results (3). As far as we are aware, the current case is the first report of the successful use of cyproterone acetate.

Patient consent

Informed written consent has been obtained from the patient's guardian for publication of this case report.

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