Stool withholding presenting as a cause of non-epileptic seizures

Case report

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Determining clinically whether or not a child with abnormal movements is having seizures is rarely easy. Several non-epileptic causes of apparent seizures are well described, especially gastro-oesophageal reflux. Stool withholding is very common in childhood, and can start very soon after birth. Although it can cause major issues for the child and family, it is rarely considered by health care professionals to be more than a nuisance. A major feature of this is pain related to defaecation. Further, in order to enhance withholding and thus prevent defaecation, a child may adopt positions and facial features that can mimic seizures. This paper presents four children (two males, two females) who presented, between the ages of 3 and 13 months, over a 2-year period to a general paediatric clinic (held in hospitals in Watford and Hemel Hempstead, Hertfordshire, UK). All received multiple investigations before accurate diagnosis. This highlights the fact that stool withholding should be added to the differential diagnosis of seizures in infancy.

Seizures can be difficult to diagnose in infants as they may present in a variety of subtle ways. The possibilities of either over-diagnosing or missing epileptic seizures is very real, both with undesirable consequences.

Identifying non-epileptic causes of seizures can prevent parental distress, over-investigation, and unnecessary treatment. Sandifer's syndrome, in which gastro-oesophageal reflux and oesophagitis cause posturing mimicking seizures, is well known. Less well recognized is the fact that stool withholding, which is also common in children of all ages, can cause transient changes in posture and behaviour that may be interpreted as seizures. Presented here are four such children who all attended a general paediatric general paediatric clinic (held in hospitals in Watford and Hemel Hempstead, Hertfordshire, UK), over a 2-year period, which suggest that investigation of infantile seizures should contain a voiding history. All investigations were carried out with informed parental consent.

Case 1

A 4-month-old white European female presented with presumed seizures. Although these had started at 2 weeks of age, they were becoming increasingly frequent and now occurred many times a day. The mother described two types of abnormal movement in her child. In the first she would flex her legs and push forward her upper body. Associated with this, she would become red and appear vacant, the whole episode lasting a few seconds. The other movements were similar to this but included deviation of the head to the right and right-sided twitching of her limbs.

Her birth history was unremarkable and her development was recorded as normal. There was nothing else of note in the history.

Initial blood tests and brain computed tomography (CT) were normal, but on observation she had repeated episodes of these movements. Metabolic investigations were undertaken and an urgent electroencephalogram (EEG) ordered.

At this stage a history of stool withholding was ascertained, with her passing a hard stool every 3 days. Laxatives were started before the EEG.

The EEG was normal, so she was discharged on laxatives. A few weeks later, at review, the movements had stopped. As the stool had softened, her mother decided to stop the laxatives.

Four months later she was referred by her general practitioner (GP) as the abnormal movements had returned. The GP felt that further investigations should be made. Although the history of stool withholding had also re-emerged, the mother was aware of the GP's concerns so a further EEG was ordered which was, as before, normal. Again, proper treatment of stool withholding aborted the symptoms.

At 22 months she remains well and off her medication.

Case 2

A white European male was referred at 1 year of age with a putative diagnosis of 'petit mal'. He was said to have vacant episodes with staring lasting for 30 seconds. He was not distractible during these. He was also head banging and suffering from night terrors. His past history was unremarkable and his development normal. An EEG showed no abnormality, and his symptoms appeared to become less marked.

He presented 18 months later with proctalgia and stool withholding. In retrospect, it became clear that this had been present at initial presentation but that the history had not been obtained. He remains well, on maintenance laxatives.

Case 3

A 3-month-old black African male presented as an emergency. Three weeks earlier he was said to have had a convulsion a few days after his first set of immunizations. During this, his eyes rolled and there was shaking of all four limbs. He received no treatment and was well until 3 days before admission, when these symptoms returned. The episodes were described as 'his eyes roll forward with fists clenched and facial grimacing, his legs straighten, and at the end he lets out a scream'.

At this stage, simple metabolic tests were performed and an EEG arranged, which were all normal. On further questioning there was a suggestion of stool withholding and laxatives were started. His mother, however, stopped these. He presented a few months later with more reported seizures, which were noted to be more marked in the few days after immunizations. Again, an EEG was performed which was normal and a history of stool withholding was obtained. Laxatives were recommenced and prevented further 'seizures'.

Case 4

A 13-month-old white European twin was referred for vacant episodes and jaw shaking. She was reported to become blank and unresponsive, with spontaneous recovery. Initially there was some jaw tremor but increasingly her whole body would shake.

She was developing normally, but may have had some breath-holding attacks. An EEG was normal.

On further questioning she appeared to be stool withholding, and again the abnormal movements disappeared as this improved.

Discussion

Constipation in childhood is most commonly due to stool withholding, which can start in very early infancy. Even very young infants have pain awareness and memory, and will try to avoid painful stimuli. If passing stool is painful, the child will try to avoid the pain: the only way of doing this is by stopping the passage of stool, by holding on. The more the child holds on, the harder the stool will become, so that when it does finally pass it is even more painful. This serves to reinforce the child's fear of defaecation and strengthens the impulse to hold on. This escalates into a vicious cycle, leading to the development of abnormal anal reflexes because of fear and pain. However, holding on is never easy, especially as time progresses. The amount of concentration and physical effort required increases as the stool builds up. The effort and technique of holding on can lead to changes in posture and appearance; further, the infant's behavioural response to pain includes many features that may mimic seizures (Henry et al. 2004). Children are often reported to be straining to pass stool, but further questioning usually identifies that the children are adopting positions that are helping them to withhold rather than pass.

Frequently a history of stool withholding will start from early infancy. An obvious trigger for this process may be mild dehydration due to poor feeding, sometimes because of neuromuscular disorders or a febrile illness. This will cause the stool to contain less water and hence become hard and painful to pass.

The association between stool withholding and seizure-like activity has been reported twice previously, in older children. These presented to specialist gastroenterology and neurology departments. The children were 4 and 5 years old respectively (see Fernando del Rosario et al. 1998, Loddenkemper et al. 2003). Both displayed typical features of stool withholding. Many of the episodes were followed by failed withholding, i.e. the passage of a small amount of stool, which seems to have been interpreted as seizure-associated incontinence. From the children described above it would seem that stool withholding mimicking seizures is more common than is recognized and can present even in the youngest age group. This may be overlooked, as stool withholding is often not considered in early infancy.

There is an increasing list of causes of non-epileptic seizures in infants, which, although usually benign, often warrant and improve with treatment. Of these, oesophagitis secondary to gastro-oesophageal reflux is undoubtedly the most common. Benign paroxysmal torticollis (Drigo et al. 2000) is rare, self-limiting, and presents with episodes of torticollis that are transient, although often prolonged. A small study recently suggested that this was a form of migraine (Giffin et al. 2002).

Gratification 'disorder' (infantile masturbation; Nechay et al. 2004) is well recognized. To the untrained or unsuspecting it may show all the hallmarks of an epileptic seizure, including a presumed postictal phase. The straps in car seats appear to be designed to facilitate this activity – especially for females – and are often a notable feature of the history. Again, this can occur in extremely young children who partake more for visceral than sexual pleasure. The correct management can be difficult. As the activity is harmless the general view is that children should not develop negative feelings towards their genitalia, otherwise problems may be caused in later life. Therefore, the usual advice is to either ignore it or provide gentle distraction. Nevertheless, it can provide intense parental embarrassment – especially in social situations. It has even been suggested that stool withholding may be a form of anal masturbation (Aruffo et al. 2000), although this is very much a minority view.

Abnormal movements may be secondary to the withholding of hard stools and may resolve when the latter are changed into soft easy stools. Yet again, one concludes with the ancient wisdom that an accurate history may prevent unnecessary anxiety, investigation, and treatment.

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Book Review

Magnetic Resonance Imaging in Epilepsy: Neuroimaging Techniques (**2nd edn**)
By Ruben Kuzniecky and Graeme D Jackson
Wisconsin, USA: Elsevier Inc., 2004, pp 442, £95.00, US$149.95
ISBN 0 12 431152 0 (Hardback)

Graeme Jackson and Ruben Kuzniecky have brought together an impressive group of contributors with expertize in clinical epilepsy and imaging sciences. They have produced an important textbook for paediatricians, neurologists, and radiologists.

It has been suggested by the International League against Epilepsy's Commission on Neuroimaging that all people with epilepsy should undergo magnetic resonance imaging (MRI) unless they have a definite electroclinical diagnosis of idiopathic generalized epilepsy or of benign childhood epilepsy with centrotemporal spikes. Therefore, it is essential that all clinicians who deal with epilepsy have an understanding of the role of MRI in the management of their patients.

After a short introduction to epilepsy, there is a section on the principles of MRI and a detailed chapter on brain anatomy. Subsequent chapters outline the clinical, pathophysiological, and aetiological aspects of temporal and extratemporal epilepsies. The section on temporal lobe epilepsies is heavily biased towards mesial temporal sclerosis with detailed information on how MRI has been invaluable in defining its spectrum and its implications on presurgical evaluation. The section also covers methodological and interpretative aspects of quantitative techniques such as hippocampal volumetry and T2 relaxometry. This is followed by a useful section on the pathogenesis of mesial temporal sclerosis. The rest of the chapter on temporal lobe epilepsies and the subsequent chapters on extratemporal epilepsies deal with the structural substrates for these epilepsies, with many example images of a variety of structural abnormalities.

The next part of the book is devoted to newer MRI techniques and application of these techniques to epilepsy. There continues to be a significant proportion of patients with clinical and electrophysiological evidence for extratemporal focal epilepsies who have normal MRI on visual assessment. Therefore, techniques that can identify structural abnormalities in these patients will potentially revolutionize their presurgical evaluation. The section on structural analysis covers techniques, such as shape analysis, curvilinear multiplanar reformatting, and voxel-based morphometry; although these show promise, none is yet used in mainstream presurgical evaluation. Functional MRI has a role in the elucidation of areas of eloquent cortex and in the characterization of areas of interictal discharges. The former is becoming increasingly important in presurgical evaluation and may ultimately remove the need for more invasive investigations. Diffusion and perfusion imaging interrogate different physiological processes to those described above. These methods have found more applicability in the investigation of patients with stroke, but there are ongoing studies in patients with epilepsy.

The technically complex chapter on magnetic resonance spectroscopy assumes a reasonable background in analytical magnetic resonance techniques and biochemical knowledge for a clinical audience, but is supported by a useful appraisal of the clinical literature on the potential applications of this technique.

Finally, this book is completed with chapters on radioisotope imaging and magneto-encephalography which are important parts of neuroimaging in epilepsy, particularly at specialist referral centres.

However, it is unfortunate that there is so little emphasis on the investigation of children. There are no data on the age-related changes in volumes, relaxation times, and diffusion parameters. For those investigating children, age-related changes need to be taken into account in order to avoid misinterpretation of images. In addition, there is no advice on the role of MRI in common paediatric epilepsies such as infantile spasms or Lennox-Gastaut syndrome. Nevertheless, this book should be in the library of every MRI unit that scans patients with epilepsy.

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