Seizure 1995; **4**: 147–149

CASE REPORT

Pseudoseizure manifestations in two preschool age children

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We report two patients with epilepsy with pseudoseizures at age 6 years. Both presented with intractable staring spells. Pseudoseizures were provoked and aborted by suggestion, leading to the diagnosis. In both patients, evidence of a neuropsychological disturbance was later found and psychotherapy started. Monitoring of intractable staring episodes is recommended prior to escalating antiepileptic drug levels or resorting to polytherapy. In addition, differentiation from other non-epileptic phenomena is necessary to initiate proper therapy.

Key words: pseudoseizures; children.

INTRODUCTION

The distinction between epileptic vs non-epileptic seizures can be a challenging differential diagnosis. The more subtle the behaviour, the more difficult the distinction. In addition, with children, lack of a clear description often complicates the task. Pseudoseizures (PS) have been described mainly in adults and teenagers. But even in these age groups, typical clinical descriptions vary[1–5]. Studies addressing PS in children and adolescents rarely include patients younger than 10 years[6] and specific behavioural features are not delineated. In young children, staring episodes may be dismissed as a behavioural disorder. The youngest age with documented 'psychogenic' seizures is in eight-year-old children[7]. Here, we report two patients with PS, diagnosed at age 6, characterized by prolonged staring. Both were of borderline normal intelligence to mildly retarded, and also had documented epileptic events. The differential diagnosis included, in addition to seizures, abnormal behaviour of the mentally retarded[8,9] and paroxysmal non-epileptic phenomena[10]. Reproducibility by suggestion, as well as evidence for underlying psychopathology, confirmed our diagnosis. We propose that staring at this age can be a sign of psychogenic seizures. Proper diagnosis requires video-EEG monitoring and is important for starting proper therapeutic interventions.

CASE REPORTS

Patient 1 is a six-year-old girl. Her seizures started at age five years with episodes of sudden quietness, staring and unresponsiveness lasting 30 seconds to a minute. A generalized tonic–clonic seizure was witnessed once. The diagnosis of primarily generalized epilepsy was made and the patient treated with ethosuximide. Persistent staring spells were noted by the parents. These episodes differed from the initial ones in that they lasted up to three minutes. An ambulatory EEG was performed and showed left frontal inter-ictal spikes with bifrontal spread ictally. Valproic acid, the com-

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bination of valproate and carbamazepine, and phenytoin were tried with only partial improvement. The patient was referred to our service for further investigation. Monitoring showed no epileptiform activity on the EEG inter-ictally and none of the recorded typical events showed epileptiform changes using the 10–20 International system of electrode placement with additional electrodes placed at Fz, Cz, Pz, T1 and T2. Presentation of unpleasant stimulation (tickling or noxious) was performed and consistently aborted the spells. Suggestion induced and arrested episodes of staring. Neuropsychological evaluation showed left frontal dysfunction with language processing difficulties. No evidence of underlying family dysfunction or history of sexual abuse could be elicited, but evidence for secondary gain was exposed. The spells were successful in taking attention away from the newborn sister. Psychotherapy was started and the patient is now seizure free 9 months later on valproic acid monotherapy.

Patient 2 is a 6-year-old right handed boy with a perinatal history complicated by sepsis and a neonatal stroke which left him with a mild left hemiparesis and mild global delay. He presented at age four years with a mixed seizure disorder characterized by tonic and atonic seizures once or twice a week. EEG showed a right posterior quadrant spike focus. He was started on carbamazepine and did relatively well. However, within a month, he developed a new type of spell characterized by staring and unresponsiveness, lasting from 30 seconds to a few minutes, occurring daily and increasing in frequency despite a switch to valproic acid. The patient was admitted for long term monitoring to better characterize his seizure type. Absence-like seizures, lasting up to five minutes, did not correlate with electrographic changes using the 10–20 International system of electrode placement with additional electrodes placed at Fz, Cz, Pz, T1 and T2. Events could be provoked by suggestion and aborted by unpleasant stimulation. No other type of event was recorded. The inter-ictal EEG confirmed the presence of epileptiform activity over the right posterior quadrant. Neuropsychiatric evaluation revealed an underlying family problem due to resentment towards the stepfather. This problem led to homicidal ideations towards the mother and coincided with the onset of the non-epileptic episodes. Therapy was started and is ongoing. The 'blacking out' events persist.

DISCUSSION

Staring episodes are a common cause of monitoring in our epilepsy unit. Between 1988 and 1992, 146 patients with staring spells with or without other seizure types, were admitted for video-EEG monitoring. Age ranged from 5 months to 43 years. In eight patients (5%), a diagnosis of PS was made. This compares to the incidence of 4% of PS in all patients monitored in our unit over the same period of time[11]. Other than the above two patients, all PS characterized by staring were in teenagers. Differential diagnosis includes absence seizures, complex partial seizures, normal behaviour of concentration or drowsiness, and abnormal behaviour of the mentally retarded[12–14]. The latter is often the hardest to differentiate. In this population, medical history, inter-ictal EEG and clinical description by caregivers have been shown to lead to inaccurate diagnosis[8]. Proper diagnosis requires video-EEG[8,9,14]. In his series, Holmes *et al* found staring to be a complaint in eight of 38 patients, three of which were of non-epileptic origin[9]. Donat and Wright reported seven of 31 retarded children presenting with staring episodes as their non-epileptic events.[15]

Staring episodes of non-epileptic origin in children can be classified as of behavioural origin[14]. The latter are a sign of an underlying attention or concentration deficit. In contrast to psychogenic seizures, they are not a source of secondary gain nor a sign of underlying distress. The differential diagnosis is significant and must be entertained in children as young as 6 years with staring episodes.

In both children reported, similarities in their clinical presentation led us to the diagnosis of pseudoseizures, despite their young age. Both had epileptic and non-epileptic seizures. In adults and children, up to 20% of seizure patients have PS, while 13 to 70% of PS patients have epileptic seizures[1–3,16]. The higher percentages are found in children[6].

Here, staring spells lasting up to several minutes, with no associated automatism or aura, make the diagnosis of either generalized or partial epilepsy unlikely[17]. Behavioural staring is also usually brief[14]. Typically, PS last longer than epileptic seizures although there is an overlap[2,18].

Both children had evidence of neuropsychological disorders. This is recognized as a risk factor in the adult population[16] but underestimated in children[19]. It can take the form of

a typical conversion syndrome or more subtly manifest as an abnormal child–parent relationship, rarely as 'Munchausen by Proxy'.

Finally, events were induced and/or terminated by suggestion or stimulation of the patient, which is a strong marker for pseudoseizures[20]. Pseudoseizures, in this clinical setting, must be differentiated from behavioural staring episodes. The latter represent signs of an underlying attention/concentration disorder, are typically brief and should not be suggestible. Here, the length and the ability to induce/abort the clinical events led to the proper final diagnosis, unmasking underlying neuropsychological disturbances.

In conclusion, we believe that pseudoseizures can be seen as early as 6 years of age. In this age group, staring can be the only manifestation of the PS. PS should be differentiated from abnormal behaviour by the duration of the staring events and by interacting with the child. Inducing and arresting an attack is very important to confirm the diagnosis. Proper neuropsychological intervention is useful in preventing fixation of symptoms and is the first step towards a cure.

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