

Case Report

Somatoform Pain Disorder Presenting as “Atypical Facial Pain:” A Rare Presentation in a 13-year-old

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ABSTRACT

Children and adolescents often present to physicians and pediatricians with a range of medically unexplained symptoms, most common being headache, abdominal, and bone pains. These symptoms can be a manifestation of underlying depressive, anxiety or somatoform disorders, and sometimes the only symptom. Hence, it is important to recognize and manage these symptoms. Atypical facial pain (AFP) or atypical trigeminal neuralgia that has variably been described to be of psychological origin is considered to be rare in children. We describe the case of a 13-year-old adolescent girl who presented with AFP, who was finally diagnosed to have a somatoform disorder. We discuss the characteristics of AFP in the index case that justify the diagnosis. We also attempt to describe psychosocial factors related to such a presentation.

Key words: Adolescent, atypical facial pain, atypical trigeminal neuralgia, child, somatoform pain disorder

INTRODUCTION

Recurrent medically unexplained symptoms are quite common in children and adolescents presenting to primary care physicians as well as pediatricians.^[1-4] Children may present with a range of somatic symptoms, the most common being recurrent headaches, abdominal ache, and bone aches.^[4] These children are at risk of associated dysfunction, excessive and unwarranted investigations, and treatment and excessive utilization of health-care services.^[1,5] On the other hand, these somatic symptoms can be a manifestation of underlying depressive disorder, anxiety disorder or somatoform disorder,^[4] and most importantly can be the only manifestation. Therefore, it becomes all the more important to recognize and manage these in child

and adolescent patients. Facial pain with no known pathophysiological cause has been described variably as a medically unexplained symptom, a diagnosis of exclusion, a psychogenic cause of pain, and even as neuropathy.^[6,7] Atypical facial pain (AFP) or atypical trigeminal neuralgia has been described in adults and elderly and has been considered to be rare in children.^[8] With this brief background, we present a case of an adolescent girl who presented with atypical hemifacial pain, was initially diagnosed and treated for trigeminal neuralgia before being diagnosed as having somatoform disorder. We also highlight and discuss the web of psychosocial dynamics in children presenting with medically unexplained somatic symptoms.

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CASE REPORT

Miss. S was 13-year-old and lived in an extended family household in a semi-urban area in North India. Her father ran a family business while her mother was a homemaker and primary caregiver for the children. She had maintained well until 11 years of age when she started to complain of dull aching, nonradiating, mild to moderate pain in the right ear daily. She was given homemade remedies, but to no avail. She often cried and missed school due to the pain. Two months later, she had 3–4 episodes in school characterized by unresponsiveness with complete awareness of surroundings, each time for 4–5 min. Teachers advised her to take rest, and she stopped going to school altogether. Electroencephalogram (EEG) (no record available) done during this time reported “epileptiform discharges in left hemisphere” and computed tomography scan showed left temporal lobe granuloma. She was given carbamazepine 600 mg/day by a neurologist and parents were told about “epilepsy.” The episodes of unresponsiveness subsided, but an earache continued. Parents were quite concerned, especially as they were told that child had epilepsy. They, especially mother became more tolerant if she would be irritable or demanding, fearing it would worsen her pain. Pain usually occurred about half an hour before leaving for school. She often missed school. Soon school authorities complained of poor attendance. Parents blamed them of not cooperating, withdrew her from that school, and admitted her to a local school. Although there was some temporary relief, pain recurred following summer holidays when school reopened. She was seen by a pediatrician; trigeminal neuralgia was diagnosed. In addition to carbamazepine, pregabalin 75 mg was prescribed. Despite regular compliance for over 2 months, no improvement was seen; hence, she was referred for a psychiatry consultation.

When seen at our clinic, it was observed that though she complained of severe pain, she could be easily engaged in other topics. She had right-sided facial pain and tenderness involving right cheek, submandibular region, angles of mouth and jaw, but sparing lower pinna, side of the nose, lower eyelid, and forehead. In addition, the right occipital area which is innervated by maxillary and mandibular branches of the facial nerve was also involved. Overall, examination suggested that pain did not correspond to the sensory distribution of the trigeminal nerve. Further, history revealed that she reported more pain on school days and just before going to school. She often complained that my school is not as good as “... (another school).” She felt ashamed of going to her school. Miss. S was also temperamentally adamant and did not adapt easily. She often demanded for expensive toys and accessories. Grandmother who

avored male children over Miss. S would be extremely critical of her. On her part, she harbored extreme anger toward the grandmother. Her mother would handle her demands inconsistently and on several occasions fulfill these without the father’s knowledge. Mother thought that “grandma was being unfair to her child, so she should take care of her.” Repeat EEG showed no abnormality.

Diagnosis of “Persistent Somatoform Pain Disorder” was kept. Efforts were made toward building a rapport with her and improving her functionality. Amitriptyline 10 mg was started initially, and other drugs stopped. She was assured that the treating team believed in the genuineness of pain, but simultaneously, the role of focusing attention and distraction in pain perception was emphasized. She was able to relate to it and agreed to follow simple behavioral measures. Activity scheduling was done, and she was encouraged to continue with her normal activities despite pain. Her efforts were appreciated at every visit. Gradually, the focus was shifted on her coping and frustration tolerance. Parents were also counseled. Miss. S is on regular follow-up for the last 18 months, does not report pain, off drugs, and regular at school.

DISCUSSION

AFP refers to pain that does not follow anatomic pathways of cranial or peripheral nerves.^[9,10] It has no objective signs, and there is no obvious explanation for the cause of the pain. Investigations are normal, and there is a poor response to usual treatments for neuralgic pains.^[11,12] Also termed, persistent idiopathic facial pain, the pain is felt deep in the soft tissues or the bone, varying from burning or aching to severe throbbing, and is poorly localized. It is mostly found on one side of the face.^[12,13] In the index case, pain was hemifacial and of dull aching type. The pain did not correspond to the sensory distribution of the trigeminal nerve and did not respond to carbamazepine, the standard treatment for trigeminal neuralgia. Thus, the index case presented with what has been historically described as AFP. Of interest is that AFP is considered to typically affect middle-aged women and is rare in children.^[12] Our case demonstrates that though rare; similar symptoms can occur in children and adolescents, and clinicians must be aware of the same.

AFP has been variably viewed as being a medically unexplained symptom to being attributed to muscular activity such as bruxism or a centrally activated autonomic vascular disturbance, demyelination, or infectious causes.^[12-14] However, originally many authors had viewed this condition as a psychosomatic disease, with a psychogenic origin.^[15,16] Depression and anxiety

are also quite prevalent in these individuals.^[12] In the index case, a detailed history revealed the fluctuations in the pain intensity in relation to school hours. It tended to be worse during daytime, absent at night and during school holidays. A similar pattern has been described for other somatoform pain complaints in children.^[4] Moreover, the pain had begun and was probably being maintained in the context of the child's temperament and parental handling. In the light of this, AFP in the index child can be considered to be of psychogenic origin and provides some limited evidence for psychological causation being one of the etiologies of AFP. Furthermore, it adds to the literature, a rare presentation of somatoform pain disorder in children.

As mentioned earlier, AFP does not respond to standard treatments but is shown to respond to antidepressants, especially amitriptyline, dosstiepin, and fluoxetine.^[17-20] Our patient had pain relief after starting amitriptyline as well as holding supportive and exploratory sessions with her.

Besides presenting an uncommon manifestation of somatoform disorder in children, this case underscores the importance of a good history and examination.

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Conflicts of interest

There are no conflicts of interest.

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