Intractable sneezing: is it always psychogenic?

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Intractable sneezing is not a commonly encountered disorder. The clinical presentation of such patients may cause the physician to undertake several tests before making a diagnosis. Are the performed analytic tests a waste of the health service funds or are they really sufficient? In this case, after undergoing several tests to rule out potential causes of the prevailing symptoms, psychiatric evaluation revealed a triggering event that contributed to the diagnosis of psychogenic sneezing. This specific case demonstrates why the diagnosis of psychogenic sneezing is difficult. An overwhelming number of such patients may be linked to a psychogenic cause; nevertheless, organic lesions or causes should be carefully excluded. Addressing the psychosocial issues through psychotherapy and coincidental use of locally administered anesthesia assisted in resolving the symptoms.

Key words: intractable sneezing, conversion disorder, topical nasal anesthesia, psychogenic, adolescents.

Intractable paroxysmal sneezing has been described and reported as an unusual disorder. Typical episodes of sneezing, manifested as 'pseudo-sneezes', occur with atypical frequency and rhythm, with eyes open, and are usually absent during sleep or when the patient is alone. This can be construed as a manifestation of conversion disorder. Since 1949, when it was first reported by Shilkret[1], an overwhelming majority of these cases have been reported to have a psychogenic origin, primarily affecting female adolescents. Approximately 25% of the reported cases resolve without any form of treatment, except counseling of the patient and family[1]. We present here a case of intractable sneezing who tested negative for any organic lesion and for which a specific provoking psychosocial event was uncovered; hence, the diagnosis of conversion disorder was considered. A resolving response to local administration of pantocaine during rhinoscopic examination prior to psychiatric interview aroused our interest about the nature of the disorder. We discuss herein the diagnostic difficulties and whether the use of local anesthetic had any effect on the treatment of the symptoms.

Case Report

An 11-year-old female patient was seen in the adolescent outpatient clinic for the evaluation of intractable sneezing. Her sneezing episodes dated back two weeks and she had been examined by primary care physicians, who had administered antibiotics, sympathomimetic agents and systemic antihistaminics, but the medications had failed to resolve the complaints. Her sneezing was manifested by a repetitive, head-bobbing snort, occurring every 5-8 seconds, and prevailing for 10 days. Of note, the complaint only persisted while she was awake. There was no nasal congestion, rhinorrhea, nasal pruritus, eye irritation, wheezing, dyspnea, cough, eczema, or dermatitis. The patient had no prior history of any disease, and the physical examination revealed no signs of a respiratory infection or nasal foreign body. A neurologic evaluation was also reported to be normal. Her laboratory work-up was normal, dermal prick test showed no allergic predisposition, and her rhinoscopic evaluation was normal with minimal nasal mucosal inflammation. She was administered topical nasal anesthesia (pantocaine) prior to

the rhinoscopic examination, but the sneezing persisted even during the examination. On completion of her ear, nose and throat evaluation approximately two hours after the local anesthetic administration, the interval between the sneezing paroxysms started extending to intervals of over a couple of minutes. Due to the partial termination of sneezing paroxysms with the topical nasal anesthesia, a cranial magnetic resonance imaging (MRI) was performed to rule out an organic lesion affecting the olfactory tract or cortex, which was reported as normal. A psychiatric interview with the patient and her mother was performed at the child and adolescent psychiatry clinic. The psychiatric examination revealed that she was a cooperative child but had immature, narcissistic and dependent features. She had difficulties in the expression of emotions. Just prior to the onset of symptoms, she had problems with her friends at school and did not want to go to school. Her mother was anxious and overprotective. The patient was diagnosed as having a conversion disorder. She and her mother were informed about the unconscious nature of the illness, and the relationship problems experienced between the patient and her friends were identified. On evaluation the following day, the sneezing had reduced significantly, her physical examination was normal and on examination by the 3rd day, the sneezing had completely resolved.

Discussion

Sneezing or sternutation is usually the physiologic response to nasal irritation. The reflex follows a very complicated pathway requiring coordinated communication between the trigeminal and nervus terminalis afferent impulses as well as from the glottic and inspiratory muscles in the efferent limb[3]. The physiologic reflex mechanism of sneezing has been reviewed[4-6] and stimuli that can induce sneezing have been mentioned; however, intractable paroxysms of sneezing are unrelated to these usual causes[7]. In the case of intractable repetitive paroxysms of sneezing, there is an "aborted" or "pseudo-sneeze" kind of action with little or no aerosolization of nasal mucous secretions[8]. The sneezing episodes generally prevail while individuals are awake, and they sneeze with their eyes open[9]. The evaluation of such patients requires a thorough history, physical examination and laboratory investigation aimed at identifying an organic cause to the problem, such as in allergic reactions, infectious causes, local nasal factors, central nervous system disease, and reflex, vasomotor, factitious, and psychogenic factors[10]. Of the several cases mentioned in the literature, an overwhelming number have been related to psychogenic origin, one due to immunoglobulin (Ig)E-mediated triethanolamine sensitivity[11], one due to a seizure disorder and one due to tuberculous cervical lymphadenitis[6].

Medically unexplained somatic symptoms in children are a major challenge in pediatric clinics. They increase health costs, and also cause the children to be exposed to unnecessary medical investigations. The presence of physical symptoms that cannot be explained by a known neurological or medical disorder is not adequate for the diagnosis of conversion disorder. There are many medically unexplained organic disorders in the literature. The essential requirement for the diagnosis of conversion disorder is the presence of unconscious psychological factors that are associated with the initiation of the symptoms[12]. Psychosomatic disorders most often affect organs regulated by the autonomic nervous system, whereas in conversion disorders, emotional problems and conflicts find expression in voluntarily controlled or sense-perception organs, and usually exhibit an acute onset. There may, however, be a crossover between these two groups[13]. Psychosocial factors are believed to have a more important influence in the etiology of conversion disorders than neurobiological factors. Various theories such as psychoanalytic theory, learning theory, family systems theory, and expression of emotions theory have been suggested in the etiology of conversion disorder. Furthermore, personality characteristics of the children, family factors, child-rearing models, stressful life events, cultural factors, stigmatization, and attitudes of the physicians play important roles in the development of conversion disorder[14].

In our case, the clinical presentation, physical examination, laboratory work-up, and rhinoscopic analysis revealed no signs of an organic disorder. However, even though it appeared to be yet another case of a psychogenic disorder, the fact that the

sneezing persisted during the rhinoscopic examination under topical nasal anesthesia and resolved shortly after did not rule out the possibility of an olfactory nervous stimulation. The meticulous psychiatric evaluation of the patient and confrontation with the parents disclosed events that could potentially be triggering stressors prior to the commencement of symptoms. Our case study fit the criteria for psychogenic sneezing; hence, psychotherapy sessions were started immediately. The patient was an immature and dependent girl. She was insecure and had difficulties in expressing her emotions. Somatic symptoms are the body language or a communicative tool of children who have difficulties in expressing emotions verbally.

Although the outcome of conversion disorder in children is reported to be favorable, there is a high prevalence of psychiatric morbidity even after recovery of conversion symptoms[15]. It is known that many conversion symptoms recover with minimal intervention or even spontaneously in a few days or weeks. The evaluation of a possible physical disorder and reassurance that the symptoms do not indicate a serious underlying disease can alone be therapeutic in some cases. Giving information about the nature of the disorder, helping the patient to gain awareness about factors initiating the symptoms and trying to understand the meaning of the somatic symptom by the patient and the family are the milestones in the treatment of conversion disorder. On the other hand, there are more serious conversion symptoms with very poor prognosis.

Medically unexplained physical symptoms usually carry diagnostic difficulties for the physicians[14]. The most important factors that increase these diagnostic difficulties are the possibility of an underlying physical illness and the uncertainty encountered as to how far the investigations for physical causes should go. It was determined that in some somatization patients, organic pathologies were revealed during follow-up. Paradoxically, it is known that repetitive and advanced investigations for any organic etiology in conversion disorder may increase the anxiety and doubts in the family and thus prolong the duration of the illness.

One must not assume that every case of paroxysmal sneezing is of psychogenic origin[16]. Due to the nature of such a disorder, these patients frequently undergo medical evaluations before a psychogenic cause is even considered. We still feel that there could have been an organic factor that we failed to trace. The role of coincidental use of topical nasal anesthetic (pantocaine) in resolving the symptoms has to be considered. We believe that routine use of topical nasal anesthesia together with other treatment modalities in such patients may be in order. Nevertheless, we must not forget that somatic symptoms are the body language of children. The primary goal in the treatment of conversion disorder is to analyze and resolve the psychological conflict rather than to remove the physical symptoms. For that reason, a collaborative effort, referral for psychiatric evaluation, and if necessary, treatment are required in many cases.

REFERENCES

1. Shilkret HH. Psychogenic sneezing and yawning. Psychosom Med 1949; 11: 127-128.
2. Fochtmann LJ. Intractable sneezing as a conversion symptom. Psychosomatics 1995; 36: 103–112.
3. Hotaling AJ, Park AH, Chow JM. A case of intractable sneezing: "a pleasure sent from God?" Int J Pediatr Otorhinolaryngology 1994; 30: 63-70.
4. Brubacher A. The physiology of sneezing. JAMA 1919; 73: 585-587.
5. Kofman O. Paroxysmal sneezing. Can Med Assoc J 1964; 91: 154-157.
6. Co S. Intractable sneezing: case report and literature review. Arch Neurol 1979; 36: 111-112.
7. Bergman GE, Hiner LB. Psychogenic intractable sneezing in children, a case report. J Pediatr 1984; 105: 496-498.
8. Elkins M, Milstein JJ. Hypnotherapy of pseudo-sneezing: a case report. Am J Clin Hypn 1962; 4: 273-275.
9. Keating MU, O'Connell EJ, Sachs MI. Intractable paroxysmal sneezing in an adolescent. Ann Allergy 1989; 62: 429-431.
10. Gopalan P, Browning ST. Intractable paroxysmal sneezing. J Laryngol Otol 2002; 116: 958-959.
11. Herman JJ. Intractable sneezing due to IgE-mediated triethanolamine sensitivity. J Allergy Clin Immunol 1983; 71: 339-344.
12. ICD-10. The ICD-10 Classification of Mental and Behavioural Disorders. Diagnostic Criteria for Research. Geneva: World Health Organization; 1993.

13. Shenker IR, Nussbaum M, Abramson AL, Ebin E. Intractable paroxysmal sneezing: a conversion reaction of adolescence. Int J Pediatr Otolaryngol 1979; 1: 171-175.

14. Pehlivantürk B, Ünal F. Cultural and clinical aspects of conversion disorder with special reference to Turkish children and adolescents. In: ME Garralda, JP Raynoud (eds). Culture and Conflict in Child and Adolescent Mental Health. IACAPAP Book Series. Maryland: Jason Aronson, Rowman & Littlefield Publishers, Inc.; 2008: 133-156.

15. Pehlivantürk B, Ünal F. Conversion disorder in children and adolescents: a four-year follow-up study. J Psychosomatic Res 2002; 52: 187-191.

16. Shapiro RS. Paroxysmal sneezing in children: two new cases. J Otolaryngol 1992; 21: 437-438.