



CASE REPORT

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Somatic cough syndrome: a report of two cases and review of literature



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Abstract

Background: Somatic cough syndrome is a somatization disorder that usually presents in children and which is often diagnosed late after an extensive search for organic causes. The condition was redefined by the DSM-5 criteria and grouped with the other conditions under the umbrella of somatoform disorders. Nonetheless, clinical presentation is heterogeneous, and etiology is still not clearly defined. Several management approaches have been proposed, but treatment remains essentially nonpharmacological.

Case presentation: We report two cases of somatic cough syndrome that were diagnosed after 4 years and 3 months respectively, and document the successful treatment of both cases using behavioral therapy.

Discussion: Behavioral therapy is the most reportedly successful nonpharmacological treatment in somatic cough syndrome, and it was successful with both our cases. The condition poses a significant burden over the function and quality of life in patients and their caregivers. The heterogeneous nature of this syndrome necessitates a high index of suspicion on part of both the physician and the psychiatrist in order to secure a timely diagnosis and mitigate the impact of this condition.

Keywords: Psychophysiological disorders, Somatoform disorders, Cough

Introduction

Somatic cough syndrome is a rare and often missed cause of chronic cough that is usually diagnosed after prolonged and unyielding tests and empirical treatments. It affects both sexes roughly equally and occurs mainly in children and adolescents, with most cases clustered between the ages of 8 and 14 years [1–3]. The condition may in a portion of cases be triggered by an upper respiratory tract infection or social stress [2]. It usually lasts weeks to months, but cases of somatic cough syndrome lasting years have been reported [2].

Clinically, this syndrome presents as a loud, repetitive, and persistent cough with a honking or barking quality, which disturbs patients' lives substantially but subsides once they are asleep [4]. A striking lack of distress "la

belle indifference" towards the cough despite its incessant and intrusive nature may suggest the diagnosis early in some cases [5]. Other cases may be associated with comorbid anxiety or depression [6].

The prevalence of somatic cough syndrome remains undefined due to rarity and heterogeneity of studies and case reports on the subject, but it is estimated to affect between 3 and 10% of children with chronic cough due to unknown causes [5]. Also, a systematic review and meta-analysis in 2016 reported an estimated prevalence of 3.02% in Chinese inpatients with chronic cough [1].

Several attempts were made to explain the condition from psychodynamic or behavioral perspectives, but etiology remains open for debate. Different treatment approaches have been proposed. Suggestion therapy, hypnosis, or counseling and reassurance are the most commonly used and reportedly successful interventions. Meanwhile, pharmacological treatment is generally perceived as ineffective [2].

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Case 1

A 14-year-old girl presented to the ENT clinic complaining from a 4-year history of chronic cough. It was described as abrupt episodes of harsh, non-productive cough with a barking quality, which lasted few seconds and recurred several times per minute for most of her waking hours. Speech was paused during these episodes although there were no signs of respiratory distress, and the cough subsided completely during sleep. There were no other associated signs such as fever, sore throat, frequent throat clearing, sensation of postnasal drip, rhinorrhea, or wheezing, and there was no personal or family history of atopy. She did not recall suffering stressful events or an upper respiratory tract infection prior to the onset of the cough 4 years ago. Over the protracted course of her symptoms, the patient had repetitively undergone extensive and lengthy investigations by doctors at different hospitals, and she was diagnosed with different conditions including a labeling with asthma for 2 years. However, she failed to show any improvement despite treatments that included bronchodilators, oral and inhalation steroids, antitussive medications, and repeated courses of antibiotics. Different investigations were performed following her presentation to our tertiary hospital, but the investigating team was unable to arrive at a definitive diagnosis; hence, they consulted psychiatry.

Upon psychiatric assessment, the cough was notably audible from outside the interview room, and the barking quality could be picked up easily, but the patient showed no signs of distress and there were no expectorations. She appeared indifferent to her symptoms contrary to her concerned mother. The patient denied any premonitory tension or change in frequency during periods of stress. She had no personal history of psychiatric illness. There was no history of motor or vocal tics, obsessional thoughts, or nocturnal enuresis. The patient also denied any history of school avoidance, social anxiety, punitive parenting, or sexual abuse and asserted that she has good relations with friends and teachers. She regretted having to miss classes due to recurrent admissions and felt sorry about having her academic record deteriorating. She was the eldest of her siblings and described herself as a lovely and responsible sister. Birth and developmental history were normal. Family history was also negative for psychiatric illness, tic disorders, or similar conditions. Mental state examination revealed a happy, confident, well-dressed girl with proper hygiene. Her speech was coherent and relevant, with good volume and tone. She smiled most of the time and described her mood as good. Affect was appropriate, without any formal thought or content disorder. The cough was not distractible during the interview. Neurological examination was unremarkable.

The patient was diagnosed as a probable case of somatic cough syndrome. Suggestion therapy was discussed as an inpatient since she lived in a rural area roughly 100 km away from the hospital, and she and her mother were willing to participate.

Suggestion therapy began with education about somatic cough syndrome. The patient was assured that she was physically healthy and had no serious conditions and that her cough will gradually disappear. She was then instructed to train herself to suppress the cough for brief periods of time about 1 min each, which she was told could be helped by using a sip of water to soothe her throat and help suppress the urge. Affirmative head nods and verbal praise were forms of positive reinforcement offered by the mother and the psychiatrist as the patient held back on the urge to cough for progressively longer periods. The next step was autosuggestion where the patient learned to do these behavioral tricks at home with help from her mother when needed. The patient was showing improvement on the next day and reported complete resolution of her cough on follow-up at the clinic after 1 month. This remarkable response was sustained 6 months later, at which time she was back to her previous highly achieving school record.

Case 2

An 8-year-old girl presented to the psychiatry clinic following a 2-month history of cough that occurred continuously throughout the day and disappeared during sleep. It was in the form of bursts of barking cough that lasted few seconds and recurred several times per minute, disturbing her speech but not yielding any expectorations. The cough was of enough frequency and severity to cause abdominal pain on several occasions. There was no associated fever, rhinorrhea, or shortness of breath, and while no aggravating factors were identified, the cough was temporarily stopped during reciting verses from the Quran or with singing. There were several visits to the hospital due to these symptoms, with clinical, laboratory, and radiological tests unyielding of a diagnosis. Her symptoms also failed to improve with inhalers and antibiotics before she was referred to psychiatry.

The psychiatric interview was interrupted by bouts of barking cough, but the patient was not distressed and the cough was not productive of expectorations. During the interview, it was illustrated that there was no history of motor or vocal tics, obsessional thoughts, or nocturnal enuresis. The patient denied having recent stressful events or upper respiratory tract infections preceding cough onset. She also denied any previous history of school refusal, sibling rivalry, punitive parenting, sexual abuse, disturbed sleep, or decreased appetite. Her medical and surgical history was unremarkable. She was the

product of a planned pregnancy, with an uneventful delivery and no developmental delay. Family history was unremarkable for psychiatric illness, tic disorders, or similar cough symptoms. She belonged to a stable and supportive family and was the third of four children. Her mother described her as the brightest among her siblings and remarked positively about her daughter's school performance and social skills and attitude. During her assessment at the clinic, the girl was able to give the full history of her condition and express her feelings about it. She also described eloquently the disturbances to her life brought about by these new symptoms. However, unlike case 1, cough improved temporarily with distraction. It also disappeared while reciting verses from the Quran and with singing. The patient seemed confident. Her affect was appropriate and speech was coherent, with normal tone and speed despite interruption by coughing episodes. There were no abnormalities in her thought content or form. Neurological examination was unremarkable.

The patient was eager to rid of her symptoms and resume her daily activities. A child psychiatrist reassured the patient and her parents that her symptoms were common among children and that different treatment modalities can potentially relieve her condition. After thorough discussion, a shared decision was made to try suggestion therapy. The patient was instructed to recite verses from the Quran for 5-min sessions through which the cough disappeared. Positive reinforcement was offered at the end of each session. As the patient learned to hold the cough for progressively longer periods after each recital, the sessions were spaced out gradually until after 3 days her cough was gone completely. This dramatic response was sustained at subsequent follow-up visits up to 4 months later.

Discussion

A systematic review by Haydour et al. in 2014 demonstrated that many clinical characteristics frequently attributed to somatic cough syndrome were not sufficiently specific [2]. Although a barking quality was the most reported cough feature, it was noted only in 8 out of 18 of the studies reviewed by Haydour et al. Also, the differential diagnosis of cough with a barking quality includes several organic diseases [5]. Cough disappearance during sleep, which is a feature considered by many to be a hallmark of somatic cough syndrome [4], was not reported in 5% of the patients included in the review. Both our cases presented with a barking cough quality, and their cough subsided during sleep. Distractibility was demonstrated in the second case only. In addition, triggers such as an upper respiratory tract infection or social stress preceding the onset of chronic cough in somatic cough syndrome were reported in

about half of the studies only, with varying frequencies of each trigger between individual studies. Neither trigger was present in our cases. Owing to this lack of specific clinical signs and investigations, Somatic cough syndrome is generally regarded as a retrospective diagnosis that is supported by improvement with psychiatric therapy after organic conditions—including rare ones—have been ruled out [2, 5].

The Diagnostic Statistical Manual-5 criteria “DSM-5” criteria in 2013 grouped somatic cough syndrome with the other somatoform disorders under common criteria, which necessitated the presence of all of the following: (a) one or more somatic symptoms that are distressing or result in significant disruption of daily life; (b) excessive thoughts, feelings, or behaviors related to the somatic symptoms (manifested by one or more of the following: disproportionate and persistent thoughts about the seriousness of symptoms, high level of anxiety about symptoms, or excessive time and energy devoted for the symptoms); and (c) persistence of symptoms [6]. Our two cases met the DSM-5 criteria, but they differed in how the second criterion was fulfilled; while the second case was anxious about her symptoms, the first one appeared to be indifferent towards the cough although she ended up devoting significant time and resources to solve the problem.

The DSM-5 criteria also replaced the outdated nomenclature of psychogenic or habitual cough with the term somatic cough syndrome. This was in part because the old terms were felt to be inaccurate and not reflecting the different aspects of the condition, and also to accommodate new findings on functional imaging studies which demonstrated cerebral correlates for disorders previously thought to be of a pure psychogenic nature [6, 7]. The new DSM-5 nomenclature and diagnostic criteria were subsequently endorsed by the American College of Chest Physicians updated recommendations on the condition [6, 8].

Psychodynamic and behavioral theorists tried to explain the etiology underlying the vicious cycle in somatic cough syndrome. Bernstein described the cough as a “bark out” protest against a dominating mother [9], while Lindenbaum and Clark reflected that the cough fits were a way for some children to express hidden anger and dissatisfaction [10]. Behaviorists regarded somatic cough as a learned behavior that can be altered through reinforcement and extinction of the undesired behavior [11]. Behavioral modification therapy shaped by this explanation was successful in many cases including ours, where a sustained resolution of symptoms was achieved with behavioral therapy, and where positive reinforcement offered by the family was essential for recovery. On a different note, some cases of somatic cough syndrome were associated with other psychiatric

comorbidities such as depression and anxiety [6, 11, 12], and the finding that cough was controlled with successful management of these associated comorbidities suggests that it may merely be a symptom of the comorbidity in at least some of these cases rather than an independent diagnosis.

Pharmacological and nonpharmacological modalities were tried for treatment of somatic cough syndrome, with the latter perceived to be more efficacious although randomized head to head comparisons between modalities remain lacking [2, 6]. Suggestion therapy is a non-pharmacological modality that is reported to be most successful; a resolution of symptoms was noted in 96% of cases ($n = 52$) who received this intervention in the studies reviewed by Haydour et al. [2]. This approach generally starts with patient education and reassurance, followed by specific exercises to suppress the undesired behavior and psychoeducational counseling to maintain the gains on the long run [4, 13]. Our patients demonstrated a prompt and sustained response with this technique despite a lag of 4 years between onset and treatment in the first case.

The other nonpharmacological treatments include hypnosis, which had a response rate (resolution or improvement combined) of 83% in patients included in the review by Haydour et al., as well as counseling and reassurance which resulted in symptomatic improvement in 93% of patients [2]. However, limited inferences can be made from these percentages as they were extracted from heterogeneous, retrospective, and nonrandomized studies and case series. On the other hand, Haydour et al. noted a general lack of benefit from pharmacological interventions. Also, no drugs have been approved for managing somatic cough syndrome, and they are generally reserved for cases with other psychiatric comorbidities [2, 4, 11].

Chronic cough due to somatic cough syndrome poses a significant impact over the function and quality of life in patients and their caregivers [2, 4, 12, 14, 15]. Recurrent hospital visits over time may jeopardize the job security of caregivers or adult patients. They also disrupt social activities and school routines. For instance, the patient in our first case reported a deterioration by over 15% in her school grades during the 4-year course of her illness. Also, periods of missed school attendance extending up to 6 months were registered in eight of the 18 studies reviewed by Haydour et al. which sought to document the impact of cough. These findings underscore the importance of early detection and treatment in order to limit the burden of somatic cough syndrome.

Conclusion

Our cases are a demonstration of the diverse presentations of somatic cough syndrome and its burden on

patients and their caregivers. A high index of suspicion on part of both the physician and the psychiatrist is needed in order to secure a timely diagnosis and mitigate this impact on daily life.

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Authors' contributions

MB and AA participated in patient management and follow-up, data collection, literature review, writing of the initial draft, and preparation of the final manuscript and figures. BA contributed to literature review, writing of the initial draft, and preparation and revision of the final manuscript. AA is the corresponding author. The authors read and approved the final manuscript.

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Consent for publication

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Competing interests

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