Original Research :

Sigh syndrome in children: a prospective study.

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Abstract:

Objective: Aim of this study was to assess the clinical spectrum & outcome of children with sigh syndrome. **Material & methods:** This prospective study enrolled paediatric patients with sighing dyspnoea presented to paediatric outpatient department but free from apparent cardiopulmonary diseases. On the basis of clinical examination, including psychological evaluation, those who conform to the criteria of sigh syndrome were included in the study. **Results:** Health status, demographic factors and recent circumstance(s) that could have served as triggers for the symptoms. Significant stress-related condition or trigger event were noted in 60.8% children. Academic stress was an important factor to be noted. No major psychological abnormality was noted in any child. With standard treatment of reassurance, breath-holding exercise, relaxation therapy and addressing the triggering factors, all improved within few weeks time. Recurrence was noted in 13% cases. **Conclusion:** The "Sigh syndrome" runs a benign course, mainly demanding support and understanding of treating physician to allay patient's concerns, if any. Possible presence of psychological stress should be evaluated in each case.

Key words:

Sigh syndrome, Eupneic breath, Dyspnoea, Psychological stress.

Introduction:

Sigh syndrome is a genuine medical diagnosis with distinct criteria, conferring significant stress for those affected. Despite outward signs of an abnormal breathing pattern, this symptomatology is unrelated to any respiratory or organic pathology[1,2]. However, it is a significant concern for the parents. They are always worried because of irregular breathing pattern of their child and doctors are unable to find a cause of this health problem. The study was conducted to identify the characteristics, clinical course and associated mental stress or behavioral abnormality if any in the child.

Material and methods:

This prospective study was conducted in a tertiary care teaching hospital from January to December 2015. Children with sighing dyspnoea presenting to the pediatric outpatient department who had no apparent cardiopulmonary diseases were prospectively enrolled. This study was approved by the hospital's ethical committee. The diagnostic evaluation of sigh syndrome included careful history-taking and thorough physical examination. Unnecessary investigations were avoided. Those who conformed to criteria (Table 1) of sigh syndrome were included in the study[1,2.] Psychological evaluation was done in every patient. Child's functioning at home, within the family, at school, and with peers was noted. Their clinical course was evaluated. Standard treatment protocol was followed in each patient. Patients with organic heart or lung diseases were excluded from the study. Apart from basic demographic data, circumstances of the onset of symptoms, concurrent medical conditions, schooling and socioeconomic status of the family has also been assessed. Clinical course for six months has been

observed.

Criteria for diagnosis of sigh syndrome[1,2] Recurrent, forced deep inspiration followed by a prolonged and often audible sigh.

Feeling of incomplete breath in spite of sighing.

1. Otherwise shallow respiration.
2. Spontaneous episodes; usually no obvious trigger or provocation.
3. Episodes occur more when alone or at rest.
4. Episodes last a few days to several weeks.
5. No interference with speech; conversation normal between deep breaths and sighs
6. Sighing absent during sleep.
7. There is no correlation with physical activity or rest.

Results:

Sigh syndrome was diagnosed in 23 children with mean age of 8.9 yrs (+ 1.8 yrs). 12 (52.1%) were girls and 11 (47.9%) were boy. 10 (43.4%) had grade II and 7 (30.4%) had grade I malnutrition. As per modified Kuppswamy scale 5 (21.7%) were in lower socio economic, 15 (65.2%) were in middle and 3 (13%) were in higher socioeconomic status [3].

All 23 patients conformed to the all ten criteria of sigh syndrome. History and examination failed to reveal evidence of any somatic findings related to breathing difficulty. Average breathing rate was within normal limits in all cases. Deep sighing respiration was absent during play, sleep or when mind distracted by any means. There was no diurnal variation. There was no evidence of any interference with speech. Sixteen out of twenty three children, included in the study, had already been seen by more than one doctors and been investigated for cardio respiratory diseases. Ten of the sixteen children were getting bronchodilator therapy including three with steroid inhalation therapy. All these sixteen children had been investigated with X- ray chest and ECG but did not reveal any abnormality. Spirometry was performed in 10 patients and was normal in all. Significant traumatic event or stress-related condition or trigger event were noted in 14 (60.8%) children. Sibling rivalry was present in 5 of the 4 children. Academic stress was in 7 of 14 children in the form of poor performance and fear of punishment from a particular teacher or parents. There was history of death of near relative was present in one patient. In one child, father had transferable job. Parents noted that symptoms of sighing dyspnoea developed in their child after change of residence to the new place. No major psychological abnormality was noted in any patients. There was history of bed-wetting in 2 children. Signs of anxiety were noted in 4 children. There was history of occasional angry outburst & defiant behavior with frequent disobeying of parents in 4 of the 23 children. 5 (21.7%) children were having mild anemia with peripheral blood picture suggestive of iron deficiency anemia.

With standard treatment of reassurance, breath holding exercise and relaxation therapy, treatment of anemia, anxiolytic medicine for short period of one week and by addressing the triggering factors, there was reduction of about 50% sighing spells within two weeks. Sighing spells stopped within four weeks in 17 (73.9%) of 23 children. Sighing stopped in all children within 14 weeks.

There was recurrence of sighing spells in three (13%) children after absence of sighing spells for two to three weeks. During the follow-up period (on an average, six months), we did not observe, development of any specific organic disorder in any case.

Discussion:

The sigh is a deep augmented breath with distinct neurobiological, physiological, and psychological properties that distinguish it from a normal eupneic breath [4]. A sigh consisted of a slow deep inspiration followed by a slow expiration. It is usually audible and may be accompanied by

obvious movement of the chest, shoulders and head. It is a physiological response to a period of shallow breathing and expands the lungs to full capacity in order to prevent the lungs' air sacs collapsing.

Every individual normally sighs involuntarily 6-8 times per hour 5. This helps keep open the alveoli that are not frequently used during quiet breathing. Many people will sigh when tired or emotional. Occasional emotional sigh is very common, usually described as "Sigh of relief" or "Sigh of sorrow" [6, 7]. However, when it is recurrent and troublesome, it qualifies for the diagnosis of "compulsive sighing" or "sigh syndrome" [1, 2].

In sighing dyspnoea, the child feels the need to repeatedly take a deep breath and feels they cannot get enough air in with normal breathing. The inspiration can be quite exaggerated and is often staccato or shuddering in nature, rather than a smooth movement. However the overall respiratory rate remains the same as has been found in our study also [2, 8]. There are usually no other symptoms and the child is well. Examination and lung function was normal in all children in our study population as was found in study by Wong KS et al 8.

How common this is in children is unknown. It is important to make the correct diagnosis, which is often one of exclusion. A careful history is the key and allowing enough time is important. Diagnosis of sigh syndrome is mainly clinical 1. In our study, we found that such children had been seen by many doctors before seeing an experienced specialist who is familiar with the relevant condition. Awareness of pediatricians and other attending physicians about this condition is very important to avoid unnecessary investigations.

The parents are often extremely worried, and sometimes the child is as well. The child can usually demonstrate the symptom when asked, and will often do it more often when it is being discussed in clinic.

Misdiagnosis is common and the children are often on inappropriate medications, including oral steroids, and these must be stopped. We found 43% of our patients were on inappropriate medications. Usually the symptoms can be managed quite easily with explanation and reassurance. Sometimes physiotherapists are needed (for breathing training and relaxation therapy), especially with types of dysfunctional breathing.

Like other studies also, the current study revealed presence of some form of psychological stress in a significant number among children having sigh syndrome [2, 4]. Presence of underlying emotional traumatic event should be probed in all diagnosed as sigh syndrome. In more severe cases, input from a psychologist may be sought. It is always appropriate to refer these patients for specific management, including psychotherapy. This would ensure preventing recurrences.

Conclusion:

Apart from possible recurrence, sigh syndrome seems to be an entirely benign and transient condition with no squeal. As most of these children are likely to be seen first by paediatrician or family physician, it is essential for these physicians to be aware of this condition to avoid unnecessary investigations. Possible presence of any psychological stress should be evaluated in every patient.

Contribution of individual authors:

Dr. Subinay Mandal has investigated all the cases and has prepared the manuscript. He will act as guarantor. Dr Satyaki Dey, Dr Meghdeep Mukhopadhyay & Dr Sumita Kundu have helped in the clinical examination and also in compiling the data.

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