

Sigh syndrome: Is it a sign of trouble?

Sigh syndrome, which involves irrepressible, persistent sighing, may be stressful for the patient, but it's benign

Practice recommendation

- 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.
- Ancillary testing and medication seem unnecessary; supporting reassurance appears sufficient, since the syndrome has a favorable outcome.

Abstract

Objective The goal of this study was to identify the characteristics and clinical course of patients presenting with considerable stress regarding irrepressible persistent sighing, and to determine whether any association exists between this syndrome and respiratory or other organic disease during the acute or follow-up period.

Study design We conducted a case series review of patients diagnosed with a defined symptom complex and gathered relevant data.

Population Forty patients who presented to 3 clinics in Israel met our 10 criteria for sigh syndrome: recurrent sighing (at least once a minute, for varying lengths of time throughout the day); otherwise shallow respiration; patient conviction that

deep breaths are obstructed; intensity of episodes provokes stress leading to consultation; no obvious trigger; episodes last a few days to several weeks; no interference with speech; sighing is absent during sleep; no correlation with physical activity or rest; and self-limited.

Outcomes measured We assessed demographic and health status information, as well as recent circumstances that could have served as triggers for the symptoms. We also performed systematic diagnoses of acute and chronic organic disease.

Results Physicians diagnosed "sigh syndrome" in 40 subjects (19 men [47.5%]), mean age 31.8 years, during the 3-year study period. All patients conformed to 10 sigh syndrome criteria. In 13 patients (32.5%), a significant traumatic event preceded onset of symptoms. Ten (25%) had previous anxiety or somatoform-related disorders. For 23 patients (57.5%), the sigh syndrome episode repeated itself after an initial episode. We found no association in any of the cases with any form of organic disease. Likewise, during the follow-up period (on average, 18 months), we did not observe the development of a specific organic disorder in any case.

Conclusions The "sigh syndrome" runs a benign course; it mainly demands the support and understanding of the treating physician to allay any patient concerns.

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In our clinical practices, we have repeatedly cared for patients who came into our clinics because of a worrisome irregular breathing pattern characterized by a deep inspiration, and followed by a noisy expiration. We have referred to the set of clinical signs that these patients present with as “sigh syndrome.”

We have long suspected that sigh syndrome is an underdiagnosed and self-limited condition that is often mistaken for a serious respiratory disorder. In our experience, this syndrome runs a benign course. However, we believed that this syndrome had characteristic and consistent features, and should not be considered a diagnosis of exclusion.

Thus, we undertook a study to observe a group of subjects with these features to judge whether this subjectively alarming symptom complex is in fact harmless, and whether it is appropriate to respond to it as we had, by taking a stress-alleviating approach alone.

What is sigh syndrome?

Patients with sigh syndrome exhibit a compulsion to perform single but repeated deep inspirations, accompanied by a sensation of difficulty in inhaling a sufficient quantity of air. Each inspiration is followed by a prolonged, sometimes noisy expiration—namely, a sigh. Observing such abnormal breaths and confirming that the patient feels a concomitant inability to fill his lungs to capacity is sufficient to make the diagnosis.

This breathing compulsion is irregular in nature: It may occur once a minute or several times a minute, and this breathing pattern may continue—on and off throughout the day—for a few days to several weeks. In our experience, it provokes significant anxiety in patients, prompting them to seek medical advice. It does not occur when the patient is asleep, and it is not triggered by physical activity.

Both patient and doctor may, at first, be convinced that the problem re-

flects a serious illness. The 10 features of sigh syndrome (**TABLE 1**) constitute a proposed definition. All of our study subjects exhibited these 10 features.

Sighing as an illness marker

Sighing has been described as one member of a group of signs exhibited by depressed or anxiety-ridden patients.¹ While Perin et al² were the first to point out the importance of distinguishing between sighing and respiratory disease, sighing per se has never been identified as a discrete illness marker.

A number of psychiatric disorders are already well known to incorporate breathing and chest symptoms along with widespread somatization. These include globus hystericus, neurocirculatory asthenia, and Tietze syndrome.³⁻⁶ As such, the acute pain of precordial catch syndrome stands out as an example of a distinguishable, clear-cut clinical state devoid of any apparent organic basis.^{7,8}

Methods

How we recruited the patients

Data was collected from 3 family practice clinics in Israel from February 2002 to February 2005. We requested that these practices contribute data of consecutive clinical cases presenting with the 10 set symptoms of sigh syndrome (**TABLE 1**).

Data collected included basic demographics, the circumstances of the onset of symptoms, concurrent medical conditions, and any associated symptoms. We assessed the patients' education level by asking questions pertaining to their years of schooling and college degrees. Patients rated their own economic status as being below, average, or above-average income.

The main outcome we examined was the clinical course of the sighing episodes during the ensuing months after their visit, in order to determine whether any patients developed a form of significant organic disease or a disorder that led to hospitalization.

FAST TRACK

In 13 of the 40 cases of sigh syndrome, the patient had experienced a recent, significant traumatic event

Results

40 cases that cut across the socioeconomic spectrum

Forty patients were recruited for this study. Nineteen (47.5%) were male; their ages ranged from 7 to 53 (mean, 31.8; standard deviation, 13.7). Two patients (5%) were Ashkenazi Jews; 34 (85%) were of North African ethnicity (Sephardic Jews); the other 4 were of varied Asian and European ethnicity. The number of patients with North African ethnicity overrepresents that of the population in their communities. Additionally, 3 members of this group were from the same family.

There was no predilection toward any specific education level or socioeconomic status. The occupations of the subjects were diverse.

Their clinical characteristics

The clinical characteristics of the 40 patients are presented in **TABLE 2**. The subjects' sighing began at various times of the day, without relationship to eating or any other activity, and disappeared during sleep. All patients reported the same feeling: that of an extra effort demanded to perform full inspiration. In many cases, adult patients were certain their complaints were a sign of cardiac or respiratory disease, and they were very concerned that there was some grave, underlying disorder.

History and examination failed to reveal evidence of any somatic findings related to breathing difficulty. Breathing rate was within normal limits in all cases.

The physicians' initial encounters with these patients led to further examination in many cases: electrocardiography, blood oxygen saturation, and complete blood counts. However, no abnormalities were found on any of these tests. Medications, if prescribed at all during these consultations, were usually given to alleviate insomnia or anxiety. Further ancillary investigations or referrals were not ordered.

The sole notable finding on physi-

TABLE 1

10 features of sigh syndrome

1. **Recurrent, forced deep inspiration** (one or more times per minute, continuing for varying lengths of time throughout the day), followed by a prolonged and often audible **sigh**
2. Otherwise **shallow respiration**
3. Patient believes that **each deep breath is obstructed in some way**
4. Intensity of episodes provokes **ample stress**, leading the patient to seek medical attention
5. **Spontaneous episodes**; usually **no obvious trigger** or provocation
6. Episodes last **a few days to several weeks**
7. **No interference with speech**; conversation is normal between the deep breaths and sighs
8. **Sighing is absent during sleep**
9. **There is no correlation with physical activity** or rest
10. **Self-limited**; responds well to reassurance.

cal examination was a typical murmur (which had already been diagnosed) in a young girl with a congenital atrial septal defect. This 7-year-old developed repeated episodes of sigh syndrome just before her annual visit to the pediatric cardiologist; her mother believed that the child was frightened by the thought of possible future surgery.

Traumatic events, anxiety disorders suggest stress as a cause

Thirteen patient histories disclosed a definite, recent, significant traumatic event that may have triggered the onset of sigh syndrome. Examples of 2 triggering events are the terrorist murder of several members of a neighbor's family, and a near-miss with a mortar shell. One subject—a teenager—had recently been left alone in the dark with 2 younger siblings during a power blackout; another young woman said that the sighing episodes began when she decided to get married.

Ten patients had previously diagnosed neurotic disorders, mainly generalized anxiety; these included 2 cases of somatization disorder and 1 case of posttraumatic stress disorder. This information from the patients' histories was documented in their medical files.

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FAST TRACK

The clustering of the syndrome in North African patients—and in one family—suggests that sigh syndrome may be a cultural or adopted expression of uneasiness

TABLE 2

**Clinical characteristics of the 40 patients studied—
 One third experienced a traumatic event**

CHARACTERISTIC	YES	NO
Smoker	2 (5%)	38 (95%)
History of anxiety or somatoform disorder	10 (25%)	30 (75%)
Taking prescription medication at time of diagnosis	4 (10%)	36 (90%)
Recurrence of sighing episodes during follow-up period	24 (60%)	16 (40%)
Trigger event (eg, exposure to traumatic event 1 month before presentation with syndrome)	13 (32.5%)	27 (67.5%)

FAST TRACK

Emphasize to your patient with sigh syndrome that further treatment is unnecessary

Episodes appear to be self-limited

In all patients, the episodes were self-limited. During the follow-up period, lasting an average of 18 months, none of the patients showed additional medical conditions—respiratory or otherwise—that could be linked to episodes of sigh syndrome. Recurrences of sighing episodes were reported by 24 subjects (60%) after the marker episode. One patient was diagnosed with carcinoma of the pancreas 2 years into follow-up, and later died.

Discussion

A benign, transient disorder

Aside from the solitary and unrelated death noted above, examination and follow-up in all 40 cases did not lead to an alternative diagnosis. Sigh syndrome thus seems to be an entirely benign and transient condition with no sequelae aside from possible recurrences.

Although the pathophysiology is unclear, our finding that 32.5% of patients had a recent traumatic incident strongly suggests a stress-related condition. Furthermore, 25% of the patients were already known to suffer from intermittent anxiety or somatoform disorders, although none were taking medications

for these conditions. This adds support to the assumption that a mind-body interaction is underpinning the disorder. Large-scale migration, recent war or terrorist acts, or natural disaster are likely to increase the chances that the average physician will see a patient with sigh syndrome.

A tendency towards North African/Sephardic ethnicity, rather than European ethnicity—in addition to the cluster of 3 cases belonging to the same family—suggests the presentation may be a subconscious cultural, learned, or adopted expression of uneasiness.

Making your evaluation: History and physical are enough

The diagnostic evaluation of sigh syndrome—consisting of careful history-taking and a thorough physical examination—should be sufficient to differentiate it from an array of organic diseases. A physical examination is imperative to exclude other causes for this breathing abnormality.

Ancillary testing is rarely, if ever, indicated. It can perhaps be justified only if the condition is accompanied by an additional (if serendipitous) finding such as the cardiac murmur in the 7-year-old girl noted earlier. Physicians sometimes perform unnecessary investigations, being reluctant to base their diagnoses solely on their clinical expertise.⁹ A patient may interpret this testing as uncertainty or begin to doubt the diagnosis, thus augmenting—rather than reducing—any anxiety.¹⁰ The additional burden of the costs and possible side effects compound the futility of testing indiscriminately.¹¹

Identifying these symptoms with the name “sigh syndrome,” and basing this diagnosis on the history and physical examination, stresses certainty and familiarity with the diagnosis.⁹ Not only does this reassure the patient, but it eases communication between professionals and forms a basis for research.

Management:**Reassure your patient**

Management of sigh syndrome consists largely of providing reassurances to your patient. You should emphasize that the condition is real, albeit benign, and that you understand the concern it causes.

Further treatment is unnecessary, aside perhaps from addressing any associated anxiety. A self-limiting (if sometimes recurrent) course can be confidently predicted, and follow-up visits can safely be left to the patient's own discretion. Since the major correlation with sigh syndrome seems to be stress and the experience of a recent traumatic event, you should always investigate these 2 possibilities when taking the history of a patient with suspected sigh syndrome. ■

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This work was inspired and developed by Arthur Furst, MD, who died following the submission of this manuscript. Dr Furst was a distinguished and dedicated family physician, a thorough researcher, and a renowned tutor. We dedicate this article to the fond memory of a true leader in the field of rural medicine, and an exceptionally funny and amicable colleague.

Disclosure

The authors reported no potential conflict of interest relevant to this article.

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