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Sleep-related laryngospasm

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ABSTRACT: The term "sleep-related laryngospasm" refers to episodic, abrupt interruption of sleep accompanied by feelings of acute suffocation followed by stridor. The condition is included in the diagnostic and coding manual of the American Sleep Disorders Association (ASDA), but there are few references in the peer-reviewed literature.

Our description of the distinct clinical picture associated with this condition is based on an analysis of the histories of a series of 10 patients. The patients and their families gave precise, uniform accounts of the dramatic attacks. Diagnostic work-up included pulmonary and gastroenterological assessment.

All patients reported sudden awakening from sleep due to feelings of acute suffocation, accompanied by intense fear. Apnoea lasting 5–45 s was followed by stridor. Breathing returned to normal within minutes. Patients were left exhausted by the attacks. Nine of our 10 patients had evidence of gastro-oesophageal reflux and six responded to antireflux therapy.

We conclude that the nocturnal choking attacks (and the occasional daytime attacks experienced by some of the patients) are caused by laryngospasm. The pathogenesis of the apparent underlying laryngeal irritability is unknown. The condition may be related to a gastro-oesophageal reflux.

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The anaesthesiological, laryngological and pneumological literature contains reports of laryngospasm occurring secondary to procedures involving manipulation of the larynx. However, very little has been published on the subject of spontaneous laryngospasm during sleep and its devastating effects on sleep quality and patients' overall quality of life.

In 1987, a patient referred to us for assessment of suspected obstructive sleep apnoea syndrome (OSAS) reported suffering repeated, brief, nocturnal choking attacks. In the years that followed, we encountered several more patients who complained of sleep disturbance due to the same cause. Their distressing experiences all followed the same pattern, and the words and gestures they used to describe the attacks strongly suggested an aetiology involving mechanical obstruction of the upper airway.

On the basis of a case series, we describe the clinical characteristics of this by no means rare disorder, for which the designation "sleep-related laryngospasm" is used, and present arguments supporting the conclusion that spontaneous laryngospasm is indeed the triggering factor.

Methods

One of the authors (AK) began to compile this series, made up of patients seen at Kantonsspital St. Gallen (St. Gallen cantonal hospital) between 1987 and 1994, because of the consistent pattern of similarities that emerged in their case histories. The hospital, a referral centre for a population of some 500,000 people in northeast Switzerland, is a 1,000 bed facility that offers pneumology, gastroenterology and otolaryngology services, among others.

The patients of this series underwent gastroscopy, oesophageal pH recording and laryngoscopy, as indicated. To rule out pulmonary disease, radiographs of the thorax in two planes and spirometry were performed. In autumn 1994, all 10 patients were interviewed by telephone to determine the subsequent course of their disorder. If necessary, additional information was obtained from their general practitioners.

Results

Characteristics of the 10 patients and their attacks are summarized in table 1. It is notable that nine of the 10 patients were male. No particular occupational category was strongly represented. Five patients experienced occasional daytime attacks in addition to their nocturnal attacks.

On one occasion, one of the patients (No. 1) suffered an attack that ended with an episode of syncope. Two other patients (Nos. 5 and 6) suffered severe, prolonged laryngospasm during attempts to pass a gastroscope and a pH probe, respectively. Patient No. 3 suffered prolonged laryngospasm upon extubation after anaesthesia. Laryngoscopy, carried out in five patients after careful preparation with local anaesthetic and performed between attacks, revealed discrete patches of redness of the

Table 1. – Characteristics of patients and their attacks of sleep-related laryngospasm

Pt No.	Age	Cyanosis	Estimated length of attacks	Frequency per month	Period from first attack to diagnosis	Daytime attacks	Evidence for gastro-oesophageal reflux			Response to antireflux therapy	Follow-up
							Symptoms	24 h pH	Endoscopy		
			s								months
1	47	-	10–30	0.5	3	+	+	+	-	Good	84
2	40	+	90	1	20	+	+	+	-	Poor	60
3	71	?	180–300	2	24	-	+	-	-	Good	22
4	55	+	120–180	1	2	+	+	ND	-	Poor	37
5	46	+	10	2	2	-	+	+	-	Good	16
6	21	-	30	2	0.5	+	-	-	-	Good	6
7	16	-	60	12	0.4	-	ND	ND	ND	ND	6
8	61	-	5–120	8	1	-	+	ND	+	Fair	21
9	56	-	10	8	3	+	?	ND	ND	Good	12
10	68	-	10–20	1	2	-	+	ND	-	Good	14

All subjects, except number 4, were male. All subjects had stridor. 24 h pH: 24 h pH recording; ND: not done.

laryngeal mucosa in one patient. The vocal cords were not erythematous and showed normal mobility in all five patients. Only one patient (No. 5) was a current smoker. Six patients regularly consumed alcohol, one of them (No. 5) to excess. Five were snorers, but only one (No. 8) complained of excessive daytime sleepiness. In this last patient, a diagnosis of OSAS was confirmed by polysomnography.

Nine of the 10 patients underwent spirometry between attacks, and nine expiratory and eight inspiratory flow-volume curves were available for evaluation. Three patients had mild obstructive defects, with forced expiratory volume in one second/forced vital capacity (FEV_1/FVC) ratios of 67, 67 and 68%. The descending limb of the expiratory portion of the flow-volume loop showed an undulating course in all nine patients, while the inspiratory curve showed a transient mid-inspiratory decrease in flow in one patient (No. 2).

There was direct and/or indirect (response to antireflux therapy) evidence of gastro-oesophageal reflux in nine of the 10 patients (table 1). Nine patients received treatment for gastro-oesophageal reflux, which was very successful in four patients and moderately so in two more. Patient No. 3's attacks did not resolve completely until he raised the head of his bed by means of a mattress wedge. In the other two cases, antireflux treatment produced no improvement despite demonstrable gastro-oesophageal reflux. In one 16 yr old patient (No. 7), who experienced just one series of nightly attacks over a 14 day period, resolution of the symptoms coincided with the start of erroneously prescribed antiasthma treatment.

Discussion

We report a case series of 10 patients who suffered repeated nocturnal choking attacks. Such attacks are traumatic experiences that provoke intense fear, and patients who suffer frequent attacks are afraid to go to sleep at night. This anxiety, which can also affect other family members, severely impairs quality of life. It becomes a dominant feature of patients' lives, leading to frequent medical consultation.

The attacks, which patients or relatives described using strikingly similar words and gestures, have the following characteristic features: 1) The patient suddenly wakes up, unable to breathe. When questioned, patients specify that they are unable to breathe either in or out, with the period of complete blockage lasting an estimated 5–45 s. Asked where the blockage occurs, they point to the larynx, take the thyroid cartilage between the thumb and index finger, tighten an imaginary cord around the neck or make a throat-cutting gesture. One patient described the feeling as being "as if a lid had closed on top of my windpipe"; 2) The patient immediately sits up straight, jumps out of bed or rushes in panic to the bathroom or a window; 3) The attack is accompanied by acute fear of suffocation. Desperate respiratory manoeuvres produce little or no airflow; 4) The short phase of total respiratory blockage is usually followed by stridor. This, in turn, resolves within a few minutes and breathing returns to normal; 5) An attack leaves the patient completely exhausted and wet with perspiration; 6) After an attack most patients go straight back to sleep, but some try to stay awake, fearful of a recurrence.

In many of the cases reviewed here, the referring physician thought the attacks had a psychogenic basis. However, we became convinced that we were dealing with somatic disease: the patients' nocturnal apprehension and fear of going to bed was quite understandable when it was realized that they were quite literally "waking in fright" to episodes of suffocation.

In our opinion, these attacks are caused by laryngospasm. The complete or almost complete obstruction of breathing, the observation by family members of patients fighting for air with forced but unsuccessful respiratory manoeuvres, and the ensuing stridor are consistent with this interpretation. In addition, the swift and total reversibility of the attacks and the fact that patients indicate the larynx as the site of obstruction point to this pathophysiology. The infrequent, erratic and almost exclusively nocturnal occurrence and short duration of the attacks make them inaccessible to medical observation. However, two of the patients (Nos. 5 and 6) provided direct evidence that laryngospasm was the cause. In these patients, attempts to pass a gastroscope and a pH probe, respectively, provoked prolonged and potentially dangerous laryngospasm. Both described these events as identical to their nocturnal attacks. In another patient (No. 3), extubation after general anaesthesia induced prolonged laryngospasm, necessitating reintubation.

While the medium-term prognosis for these patients was good, this case series is too small to permit generalization. Apart from the two cases of iatrogenic laryngospasm described above, the experiences of two other

patients from the series show that serious consequences cannot be ruled out. Patient No. 2 suffered an attack while swimming in deep water, and, subsequently, swam only in places where he could stand with his head out of water. Patient No. 9 suffered repeated attacks while driving on the motorway, which usually forced him to stop in the breakdown lane. On one occasion, however, an attack occurred while he was driving through a tunnel, where he was unable to stop; a series of forced Müller and Valsalva manoeuvres led to violent vomiting, and he had great difficulty in keeping the car under control. Finally, the episode of syncope experienced by the present index patient (No. 1) is also a reminder that a real risk of injury exists.

One patient in this series (No. 8) had OSAS; this patient also had severe reflux oesophagitis and spent the nights sitting in a chair to prevent attacks. Only when treatment with omeprazole had stopped the attacks, was the patient able to lie supine. Most references to nocturnal choking attacks are found in the literature on parasomnias, often in review articles [1], but the condition gets only brief mention, and no sources are given. A textbook chapter [2] on obstructive sleep apnoea briefly mentions 22 patients, who seem comparable in every respect to the 10 described here. In an abstract, GUILLEMINAULT and MILES [3] refer to five patients with identical symptoms, all of whom had gastro-oesophageal reflux. They reported that the attacks could sometimes be suppressed by coughing, as did patient No. 9 of the present series. Another four patients are described in a second abstract [4].

In 1995, ALOE and THORPY [5] published the first case series describing this condition in a peer-reviewed paper. They coined the term "sleep-related laryngospasm" which was adopted by the authors of the American Sleep Disorders Association (ASDA)'s diagnostic and coding manual [6]. The attacks suffered by their seven patients resembled those reported here in every respect. They recorded an attack in one patient by polysomnography during stage 3 sleep, supporting the reports by the patients of the present series that the attacks started during sleep, and the report by the wife of patient No. 5 that his attacks started with short coughs and inspiratory stridor while he was still asleep. BORTOLOTTI [7] postulated reflex central apnoea due to gastro-oesophageal reflux as the cause in two patients with nocturnal choking attacks, but in both there was also evidence of laryngeal involvement. Detailed clinical descriptions of laryngospasm-related attacks are found in paediatrics, but only in one case is an argument made for gastro-oesophageal reflux as the triggering factor [8]. CAMPBELL and PIERCE [9] describe a diurnal syndrome similar to that presented here. It seems that the syndrome we describe here and that of CAMPBELL and PIERCE [9] could be nocturnal and diurnal variants of the same disorder.

The differential diagnosis should include nocturnal hyperventilation, nocturnal bronchial asthma, nocturnal coughing fits, epilepsy manifesting as laryngospasm [10], primary and secondary parasomnias, and the sleep choking syndrome [6].

The clinical picture of sleep-related laryngospasm is distinct from that seen in vocal cord dysfunction [11, 12]. In vocal cord dysfunction, patients present with dyspnoea and laryngeal wheezing and attacks can readily be witnessed. Unlike patients with vocal cord dysfunction, who frequently have a history of conversion disorder, the patients in the present series had no history of psychiatric disorder. Also, individuals with vocal cord dysfunction are often members of the paramedical professions but this was not the case with any of the patients presented here. Finally, nine of the 10 patients presented were males, whereas vocal cord dysfunction affects mainly females.

Diagnosis of sleep-related laryngospasm relies on a careful and detailed history and the exclusion of other conditions. It is sometimes helpful to have patients or their relatives act out an attack or imitate the sounds that are heard.

The patients described awoke because of laryngospasm. The pathogenesis of this parasomnia is unexplored and is likely to be complex. Our observations suggest that gastro-oesophageal reflux may play a role. Postnasal drip and minor psychological disturbances may be other factors [5] that contribute to the laryngeal irritability. Treatment aims at relieving underlying causes, if identifiable.

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