

## **Pediatric Severe OSAHS with a Funnel Chest**

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### **ABSTRACT**

The primary etiopathology of pediatric obstructive sleep apnea hypopnea syndrome (OSAHS) includes tonsil or adenoid hypertrophy. Severe OSAHS contributes to or aggravates thoracic deformity, which is rarely reported. In the current report, a child was admitted to the hospital with over four-year history of snoring during sleep, and two-day-old aggravation with severely depressed sternum during inhalation. Clinical examination indicated tonsil and adenoid hypertrophy, and polysomnography revealed OSAHS. The symptoms of OSAHS and severe inhalation-related sternum depression disappeared rapidly after tonsillectomy. Our findings indicated that OSAHS were the major causes underlying funnel chest in children. The rarity of the incidence may result in missed diagnosis or misdiagnosis. Polysomnography was recommended for the child diagnosed with funnel chest accompanied by upper airway stenosis.

**Keywords:** Obstructive sleep apnea syndrome; funnel chest; polysomnography

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## **INTRODUCTION**

OSAHS adversely affect the development of multiple systems in children, resulting in growth retardation, metabolic abnormality, increased blood viscosity, hypertension and altered heart structure (1). Funnel chest is a common thoracic deformity in children with an incidence of about 1/300 (2). Pediatric OSAHS is caused predominantly by tonsil or adenoid hypertrophy. Although rare, it results in thoracic deformity. In the current report, a pediatric case of severe OSAHS accompanied with funnel chest is reviewed.

## **CASE REPORT**

The five-year-old patient, of Chinese Han descent, was admitted to our hospital on September 9, 2014 with ‘a four-year history of snoring, and shortness of breath for two days’. Nasal congestion, oral breathing during sleep, snoring, shortness of breath, stuffy nose aggravated after cold, obvious nocturnal snoring accompanied with shortness of breath, and occasional earache were observed four years ago without apparent reason. The child was afflicted with cold a week ago and aggravated nasal congestion, nocturnal snoring accompanied with shortness of breath. The symptoms were further aggravated two days before admission.

The thoracic deformity and depressed sternum were found by his parents. The child was admitted as an out-patient with the diagnosis of OSAHS. The physical examination showed that the child was 120 cm high and weighed 21 kg, with an adenoid face and funnel-shaped thorax. The lower segment of the sternum was connected to the costal cartilage measuring  $10 \times 8 \times 2$  cm. The frequency of respiration was increased and the inhalation-induced thoracic depression was overt (Figure 1). The lung and heart auscultation was normal.

The outward appearance of nose showed no abnormality; however, the nasal septum was deviated with infratubinal congestion and plenty of purulent nasal discharge, without any neoplasm. Pharyngeal cavity showed stenosis with bilateral grade 3 swelling of amygdala, without acute hyperemia. No pus or pus membrane appeared on the surface of tonsil and no uvular hypertrophy was seen either. There was no sagging of soft palate or tongue hypertrophy. Ear drum was complete though the boundaries were not clear. Laboratory data showed that the white blood cell count was  $16.8 \times 10^9/L$  with 80% neutrophils. Trace elements in the blood showed normal levels of calcium levels at 2.01 / L; blood phosphorus, 1.57 / L; and serum 25-hydroxyl vitamin D3, 55 ng/L. The physical examination revealed adenoid hypertrophy during nasal endoscopy and occupied 3/4 of posterior nostril. Sinusitis and adenoid hypertrophy were revealed using three-dimensional CT. The lower fragment of sternum was depressed inward though there was no obvious cardiac compression, and the thoracic index of HI (4) was 3.4. The binaural tympanic pressure was increased by acoustic immittance testing. Other tests such as ECG and echocardiogram were normal. Pulmonary function testing showed normal pulmonary ventilation and diffusion function. The sleep apnea hypoventilation index (AHI) was 34.8 with polysomnography and the lowest oxygen saturation was 67% (Figure 2). The diagnosis was severe OSAHS accompanied with severe nocturnal hypoxemia and funnel chest (moderate) (3).

Proactive symptomatic treatment was carried out by nasal resin twice daily, mometasone furoate nasal spray twice daily, and cefazolin sodium intravenous injection twice daily at a dose of 1.0 g. Noninvasive positive pressure ventilator-assisted breathing was performed at night during sleep. The symptoms of stuffy nose improved, without purulent nasal mucus, cough, or sore throat; however, sleep apnea still occurred at night. Blood cell count returned to normal, and bilateral tonsillectomy and adenoidectomy under general anesthesia and nasal endoscopy were

performed on September 15, 2014. The surgical process was uneventful, the intraoperative bleeding was round 5 mL and the dissected tonsil and adenoid tissue was approximately 3 cm × 2.5 cm × 2.0 cm in size.

The dissected tissue was pathologically confirmed as chronic tonsillitis adenoiditis. The consciousness of child was restored after operation, with no active nasopharyngeal bleeding. The symptoms of snoring and shortness of breath vanished three days after operation, and the inhalation-induced thoracic depression was also greatly improved. The postoperative blood cell counts were normal. No stuffy or running nose, sore throat, or ear-ache were observed a month after operation during follow-up. The nasal pharynx was smooth during endoscopic examination and the posterior nasal was patent. The thoracic cavity was still funnel-shaped; however, there was no inhalation-induced depression. The AHI was 2.0 and the lowest blood oxygen was 90% by polysomnography (Figure 3). The OSAHS was cured, and the funnel chest was corrected surgically. The follow-up review one year later indicated that the child had good sleep without nasal congestion, oral breathing, snoring or breathlessness at night. The funnel chest was not corrected due to the cost involved, and the severity of funnel chest was unchanged.

## **DISCUSSION**

Funnel chest is a common deformity of the thoracic wall. The funnel-shaped deformity is formed by the sternum, rib cartilage and ribs, which are suppressed towards the spine, and apparently inherited as a dominant trait (4). The inward depression of sternum suppressed the major organs in the thoracic cavity, which resulted in reduced tolerance to activity due to chronic pulmonary and cardiac suppression. The depressed sternum suppressed the heart and lung severely, leading

to severe consequences (5). The pathogenesis of funnel chest was unclear, and probably associated with poor development of diaphragm, rib and rib cartilage (6). It was also believed to be related to the overgrowth of rib, which was bent backwards and resulted in a funnel-shaped chest (7). However, Tatsuo Nakaoka *et al* believed that the pathogenesis of funnel chest was complicated, and involved multiple factors but not rib overgrowth (8). It was suggested by D. Jaroszewski that the incidence of funnel chest was significantly increased (6.6%) in children with laryngeal cartilage softening syndrome (9). It was further suggested by Daniel Schaerer *et al* that the laryngeal cartilage softening resulted in stenosis and obstruction of multiple airway passages, which increased the negative pressure in the chest and caused thoracic deformity. Therefore, the laryngeal cartilage softening syndrome was closely associated with funnel chest (10). Funnel chest was believed to be a sequela of OSAHS by M.B. Pringle *et al*, who suggested preoperative polysomnography in children with severe OSAHS, and postoperative medical care (11).

In the current report, preoperative polysomnography indicated that sleep latency was significantly prolonged and sleep was fragmented with increased frequency of arousal and decreased slow-wave sleep pattern. Dynamic video monitoring showed difficult respiration nocturnally, and inhalation-induced depression was observed during sleep apnea. These findings indicated that the funnel chest formation might be due to upper airway stenosis and obstruction, which increased the negative thoracic pressure. The long-term negative thoracic pressure induced an inward, funnel-shaped rib and sternum depression, especially when the rib and sternum were still in the developmental stage.

The therapeutic effect of tonsillectomy and adenoidectomy in pediatric OSAHS ranged between 52.9% and 100.0%, with an average of 82.9% (12). We suggested that upper airway stenosis release should be performed initially in order to effectively stop the development of

funnel chest (10). In the current report, the inhalation-induced thoracic depression disappeared after tonsillectomy and adenoidectomy. The AHI was restored to normal by polysomnography one month after operation. The severity of funnel chest remained unchanged one year later compared with preoperative level.

The current case study suggested that OSAHS is the main cause of funnel chest. Due to the rarity and ignorance of the syndrome, patients were usually admitted to the departments of Pediatrics, ENT, or thoracic surgery, with missed diagnosis or misdiagnosis. We suggest that polysomnography should be performed in children with funnel chest accompanied with upper airway stenosis, to exclude OSAHS and facilitate accurate diagnosis and treatment.

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#### **AUTHORS' NOTE**

All authors declare that they have no any conflict of interests.

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Fig. 1: Funnel chest. The funnel-shaped thoracic cavity; the lower segment of sternum was associated with rib cartilage forming a depression deformity, measuring approximately  $10 \times 8 \times 2$  cm (indicated with red arrow).

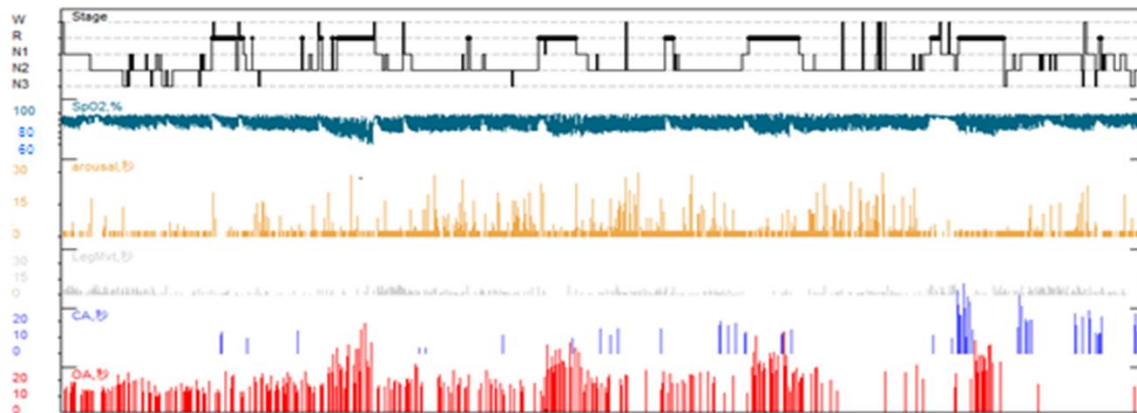


Fig. 2: Preoperative status chart of polysomnography.

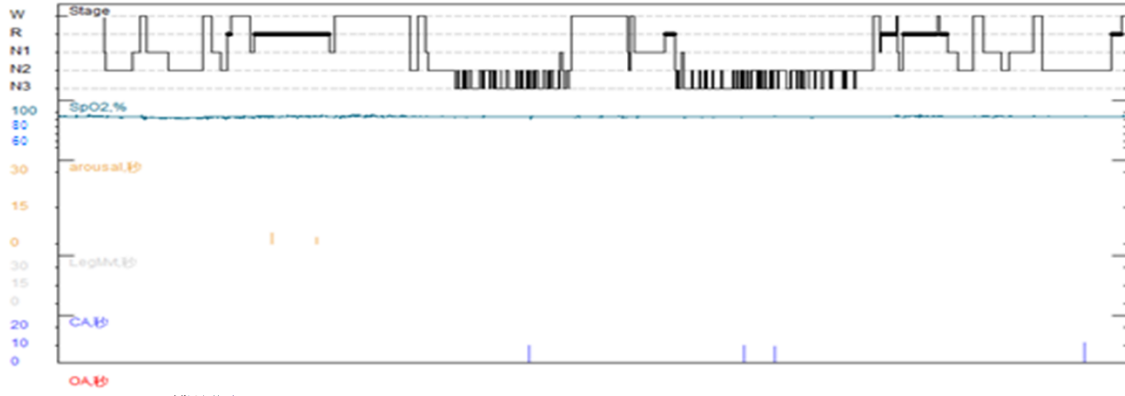


Fig. 3: Postoperative outcomes of polysomnography.