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CLINICAL REPORT

Approaching Saber-Sheath Trachea in a Patient with Bilateral Vocal Cord Abductor Palsy and Post Tracheostomy Tracheal Stenosis

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Abstract
Human trachea has variable shapes and the most common are circular and oval-shaped. Other shapes are rare to find and usually related to some diseases. Such as the Saber-sheath shaped trachea, which was commonly attributed to patients with chronic obstructive pulmonary disease (COPD). This case is about a patient who was detected to have dual pathology, bilateral vocal cord abductor palsy, and post-tracheostomy complicated with tracheal stenosis for 40 years. Symptoms worsened in the last three years until he was admitted to the ward. However, upon managing this case with multilevel airway obstruction, we have found out that this patient who did not have COPD, has a Saber-sheath trachea shape. This finding may indicate that the shape is not limited to a certain disease only.

Keywords Trachea abnormalities · Saber-sheath · Multilevel airway obstruction · Bilateral vocal cord palsy

Introduction

Bilateral vocal cord abductor palsy is a life-threatening condition that requires emergent surgical management to secure the airway which is tracheostomy. Despite its purpose to secure the airway, the tracheostomy itself may cause another level of airway stenosis. The stenosis can be at the subglottic or tracheal region. Hence, here we report such case of a gentleman who has withstood for 40 years of symptomatic bilateral vocal cord abductor palsy and tracheal stenosis secondary to tracheostomy for prolonged ventilation (multilevel obstruction). A saber-sheath-shaped trachea structure was identified during the second tracheostomy while managing the current upper airway issues.

Case Report

A 74-year-old gentleman presented to the Emergency Department with a one-week history of cough and difficulty in breathing. He was admitted to the medical ward and treated for community-acquired pneumonia. He has a history of severe traumatic brain injury 40 years ago necessitating the insertion of ventriculoperitoneal shunt and tracheostomy due to prolonged ventilation. The tracheostomy tube was decannulated one year later. Presently, he has been experiencing hoarseness since the tracheostomy tube decannulation and has worsened over the past three years. He never had aspiration symptoms and the swallowing function remains unaffected. Upon examination, the patient was tachypnoeic despite on oxygen supplementation via face mask with the presence of hoarseness and biphasic stridor.

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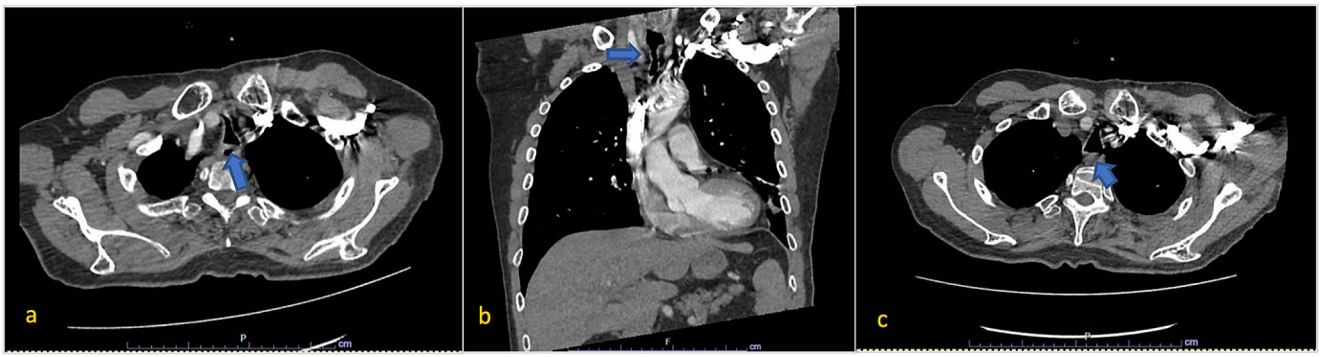

Fig. 1 (a): Axial view of the stenotic segment (blue arrow) of the trachea at the level of 1st thoracic vertebra, (b): Coronal view of the CT scan showing the stenotic segments at the level of 1st thoracic vertebra (blue arrow), (c): Axial view of the CT scan showing the saber-sheath shape trachea at the level of 2nd thoracic vertebra (blue arrow)

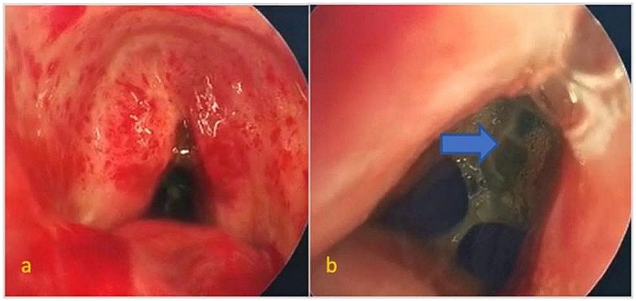

Fig. 2 (a): "A"-shaped tracheal stenosis at the level of 1st thoracic vertebra, (b): Tracheal stenosis ends at the level of the 2nd thoracic vertebra at the suprastomal region. Tracheostomy tube marked with blue arrow. Noted persistence triangular shaped or saber-sheath appearance of the trachea

Indirect laryngoscopy showed bilateral vocal cord abductor palsy with a narrow-slit airway. Chest radiograph revealed a central trachea with a small consolidation over the left middle lobe suggestive of infection. A tracheostomy procedure was performed under local anaesthesia to secure the airway.

Intraoperative findings revealed a collapsed trachea distal to the first tracheal ring. An incision was made between the first and second tracheal rings; however, insertion of the smallest available cuffed tracheostomy tube, a size 6.0 mm internal diameter (ID), was not possible. Consequently, a 5.5 mm ID endotracheal tube was temporarily placed.

A computed tomography (CT) scan of the neck revealed a stenotic segment of the trachea at the infraclavicular level, corresponding to the first to third thoracic vertebrae. (Fig. 1**)**. The trachea below the stenotic segment has a "saber-sheath" shape with a ratio of sagittal/coronal length is more than 1.5 **(**Fig. 1**)**. The patient was subsequently scheduled for airway evaluation under anaesthesia and tracheostoma revision. There was presence of subglottic stenosis with Cotton-Myer grade I (about 30% stenosed lumen) over supra stomal region. The stenotic segment of the trachea was characterized by mature scar tissue along the lateral walls, as depicted in **(**Fig. 2**).** Distal to the stenosis, the tracheal morphology appeared triangular, extending to the carina without evidence of malacic segments. The tracheostoma was widened, facilitating the placement of a size 8.0 tracheostomy tube.

Six month post-operative, the patient is comfortable on room air and has improved effort tolerance. The patient currently using a double-lumen fenestrated tracheostomy tube and can vocalize.

Discussion

A normative range of the normal population for coronal and sagittal diameters of the trachea are, respectively, in men aged 20–79 are 13–25 mm and 13–27 mm; in women, they are 10–21 mm and 10–23 mm, respectively. Deviation from these numbers indicates pathologic tracheal air column widening or narrowing. The relationship between tracheal caliber and body weight or height was not statistically significant [1, 2].

There are few documented variations in tracheal morphology. Adult trachea exhibits a range of shapes, with some maintaining a circular configuration rather than adopting an ovoid form [3]. Changes in intraluminal pressure significantly cause an impact on the trachea's morphology. Coughing, breathing, or ventilation may be to blame for this. With aging, the cross-sectional shape can significantly change, especially if chronic obstructive pulmonary disease (COPD) is present. Tracheal cartilage softening or "malacia" is another presentation that related to the disease. The "saber-sheath" trachea is an acquired condition resulting in narrowed lateral diameter and enlarged anteroposterior diameter of trachea [3]. There is narrowing of the intrathoracic trachea, characterized by an internal coronal diameter that is two-thirds or less of the sagittal diameter, measured at a level 1 cm above the aortic arch [3–5]. These changes may result to various degrees of obstruction on coughing and expiration. Other unusual forms reported to occur together

with tracheal diseases such as tracheopathia, osteoplastica, and tracheobronchomegaly (Mounier-Kuhn syndrome) [2].

Our patient has no signs of COPD as he is neither a smoker nor has a history of recurrent admission throughout his 40 years for any lung problems. He was able to communicate well under room air despite having hoarseness. His condition with bilateral vocal cord abductor palsy and tracheal stenosis causing multilevel chronic upper airway obstruction may contribute to the increase in his intrathoracic pressure. The anteroposterior diameter of the chest expands as a result of pulmonary hyperinflation, which also causes the trachea's sagittal dimension to lengthen [6, 7]. The cartilaginous rings resist such elongation due to their "U" shape, which causes too much pressure on both lateral sides. The cartilage becomes weakened, degenerated, and calcified as a result of this tension, particularly in the anterior region [3].

Conclusion

This patient who had bilateral vocal cord palsy due to traumatic brain injury and tracheal stenosis secondary to old tracheostomy developed a Saber-sheath trachea due to the multilevel obstruction. This condition is not specifically attributed to patients with COPD only. Surgeons need to be aware of this clinical entity as it may cause a difficult tracheostomy during an emergency airway procedure.

Declarations

Conflicts of Interest The authors have no conflicts of interest to declare relevant to this article's content.

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