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Caso Clínico

Tracheocele: an unusual cause of dysphonia

Traqueocelo: una causa rara de disfonía

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Abstract

Tracheocele, also known as tracheal diverticulum, is a herniation of the tracheal mucous membrane and it is rarely reported in the literature. Its estimated prevalence is about 1%. It may be a congenital defect or an acquired lesion. Traumas, high pressure injuries, long lasting tracheostomy, obstructive tracheal diseases, recurrent infections of the mucous glands of the trachea with subsequent ductal obstruction and dilatation may play a role at the etiology. It affects preferably the right posterior aspect of the trachea. The best diagnostic procedures are endoscopic examination and computed tomography. The management of tracheocele is primarily conservative, but surgical intervention may be indicated for symptomatic cases. Herein, we present a case report of a 57 year old male presenting with slow progressive dysphonia and chronic cough for one year. Flexible laryngoscopy revealed a severely paretic right true vocal cord. Computed tomography scan revealed a tracheocele compressing the recurrent laryngeal nerve. The patient went surgery with complete removal of the lesion. Afterwards, the patient attended 3 month of voice therapy with complete recovery of the dysphony.

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Keywords: Tracheocele, tracheal diverticulum, dysphonia.

Resumen

El traqueocele, también conocido como divertículo traqueal, es una herniación de la mucosa traqueal y rara vez se informa en la literatura. Su prevalencia estimada es de alrededor del 1%. Puede ser un defecto congénito o una lesión adquirida. Traumas, lesiones de alta presión, traqueotomía de larga duración, enfermedades traqueales obstructivas, infecciones recurrentes de las glándulas mucosas de la tráquea con posterior obstrucción ductal y dilatación pueden desempeñar un papel en la etiología. Afecta preferentemente el aspecto posterior derecho de la tráquea. Los mejores procedimientos diagnósticos son el examen endoscópico y la tomografía computarizada. El tratamiento de la traqueocele es principalmente conservador, pero la intervención quirúrgica puede estar indicada para los casos sintomáticos. En este trabajo se presenta un caso clínico de un hombre de 57 años de edad con disfonía progresiva lenta y tos crónica por un año. La laringoscopia flexible reveló una cuerda vocal parética. La tomografía computarizada reveló un traqueocele que comprime el nervio laríngeo recurrente. El paciente fue operado con la extirpación completa de la lesión. Posteriormente, el paciente atendió 3 meses de terapia de voz con recuperación completa de la disfonía.

Palabras Clave: Traqueocelo, divertículo traqueal, disfonía.

Introduction

Tracheocele is also referred to as tracheal pouch, tracheal diverticula and tracheogenic cyst. A tracheocele is a rarely encountered entity. Fewer than 30 cases have been reported in world literature ¹. When symptomatic the most related complaints are chronic cough, intermittent dysphagia and recurrent bronchopulmonary infections^{1,2}.

Case Report

Herein, we present a case report of a 57 year old carpenter referred to our clinic with a history of with slowly progressive dysphonia and chronic cough for one year. There was no history of vocal abuse, respiratory infection or evening rise of temperature. There was no history of any surgery or trauma. There was no history of smoking, alcohol or drug abuse. He reported no dysphagia or odynophagia. Flexible laryngoscopy revealed a severely paretic right true vocal cord with glottic air escape. Computed tomography scan revealed a tracheocele measuring 24 x 23 x 20 mm compressing the right recurrent laryngeal nerve (figure 1) at cervicothoracic junction level. Flexible bronchoscopy was performed to identify the site of origin of the tracheocele and to determine if difficulty would be encountered endotracheal intubation; no problems were encountered and a single lesion was confirmed. Functional respiratory tests were normal.

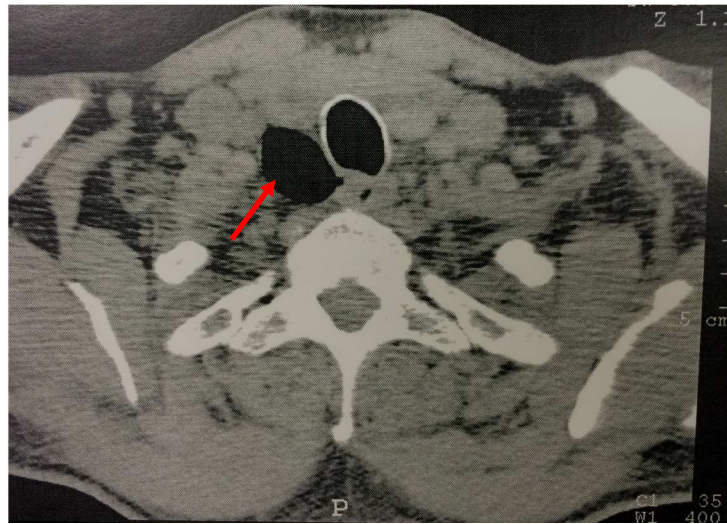


Figure 1: right-sided tracheocele measuring 24 x 23 x 20 mm (red arrow).

We performed an elective surgical resection of the lesion with the patient under general anaesthesia after. A transverse cervical incision was made 2 finger breadths above the sternal notch, extending from the right anterior border of the sternocleidomastoid to 2 cm off the midline on the left. By subplatysmal dissection, the sternocleidomastoid muscle was retracted laterally and strap muscles medially. The thyroid gland was exposed. The lesion was identified after increasing intracheal pressure by mechanical ventilation by the anaesthetist. The sac was then dissected free from surrounding tissue, paying special attention to the right recurrent nerve which was lying stretched on the medial surface of the lesion. Surgical clips were used to close the tracheocele neck. The remaining tissues were closed with 3-0 Vycril. 2 g of cefoxitin was used prophylactically. A Redivac® drain system was left for 2 days. The patient was discharged at day 3 post-operatively. There were no interurrences during the recovery time. Post operative CT revealed complete removal of the lesion. (figure 2). The anatomo-pathologic exam confirmed the nature of the lesion: a cavitary empty sac lined up by respiratory epithelium. After 3 month the patient had full recovery of his voice after voice therapy.



Figure 2: Post-operative CT showing full removal of the lesion (white arrow marks surgical clips).

Discussion

Tracheocele is more common at the right posterolateral aspect of the upper trachea at the level T1-T3, due to the lack of protection of the carotid arch and esophagus on that side². This tracheal lesion is characterized by the presence of a single cystic pouch filled with air or a mixture of liquid and air¹. The most widely used classification divides this entity into congenital or acquired. Some authors consider classify air cysts of tracheal origin into tracheogenic cysts (with muscle and cartilage in the wall and of embryonic origin), tracheoceles, and diverticula (both lacking a muscular frame). The latter two are differentiated by number or by size, with tracheoceles being of one single formation or else larger than 2 cm and diverticula being multiple formations or smaller than 2 cm^{1,3-5}. Traumas, high pressure injuries, long lasting tracheostomy, obstructive tracheal diseases, recurrent infections of the mucous glands of the trachea with subsequent ductal obstruction and dilatation may play a role at the etiology of acquired tracheoceles². This type of lesion can also occur in cases of Chronic Obstructive Pulmonary Disease, tracheobronchomegaly (Mounier-Kuhn Disease) or Cystic Fibrosis^{2,3,4}. Congenital tracheoceles are thought to be malformed supernumerary branches of the trachea³. Clinically, most tracheoceles are asymptomatic and do not require intervention. For symptomatic tracheocele, various methods of treatment have been reported in the literature, including surgical resection, fulguration, and conservative medical management of symptoms (antibiotics, mucolytic agents, and physiotherapy). Resection may be performed transcervically, avoiding the need for a thoracotomy, or endoscopically with laser surgery or electrocautery⁴⁻⁶. Options vary depending on the patient's general condition and symptoms, preferring endoscopic management in children with small lesions^{2,6}.

These swellings have to be differentiated from lymphangioma, branchial cyst, venectasis and pulmonary bulla by clinical examination and radiological investigations⁷.

Tracheoceles, although a rare etiology of vocal fold paralysis, should be considered as a differential diagnosis, especially in patients with no tumour risk factors.

Conflict declaration: The authors declare no conflicts.

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