A Case Report of Trachea Diverticulum—Divergent Symptoms of an Uncommon Pathology

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Abstract
The tracheocele or tracheal diverticulum is a cavity secondary to a congenital or acquired weakness of the tracheal wall. It is constituted as a rare clinical entity with very few cases published in the literature and given its low symptomatology, its diagnosis is difficult. We present the case of a 63-year-old patient with odynophagia and right cervicalgia, with no significant personal history, and with a diagnosis of cervical abscess, was taken to an exploratory cervical surgical procedure, finding a right tracheal diverticulum intraoperatively. Complete surgical resection of the lesion was performed, without complications and remission and with satisfactory postoperative evolution.

Keywords: Trachea; Tracheocele; Diverticulum; Diagnosis

Introduction
The tracheocele or tracheal diverticulum is a cavity secondary to a congenital or acquired weakness of the tracheal wall. It is constituted as a rare clinical entity with very few cases published in the literature and with a low symptomatology, the diagnosis is very difficult. It is often diagnosed incidentally and, due to its rarity, there is no standard treatment. It is a benign entity but has the potential to cause specific symptoms or be asymptomatic [1]. We report a case of a 63-year-old patient with an initial diagnosis of right cervical abscess and subsequent intraoperative finding of right tracheal diverticulum managed by surgery by the multidisciplinary surgery group (thoracic and head and neck services). We completely resected the lesion, evidencing remission of uncomplicated symptoms.

Clinical Case
We present the case of a 63-year-old, non-smoker with one week of odynophagia and right cervicalgia, associated with signs of local and systemic infection. Complementary imaging studies were taken. Chest x-ray was normal. A CT scan shows an image compatible with a retropharyngeal abscess. It was evaluated by the head and neck surgery service that perform right cervical abscess drainage by a puncture. They recollected a simple of transparent fluid without any smell, approximately 5 cc, and sent for studies, with negative results. So, they do not use any antibiotic treatment.

Due to the persistence of systemic symptoms (tachycardia and elevated leucocytes), we decided to take another CT scan, finding an image with radiolucency in the right cervical base related to the posterior wall of the trachea without showing communications or pathways (Figure 1). Given the characteristics of the lesion, thoracic surgery evaluation was requested with a surgical treatment.

It was carried out to an exploratory cervicotomy. A right tracheal diverticulum adhered to deep planes was demonstrated and the total resection of the lesion was performed. We had no need to remove tracheal rings. The following neurophysiological monitoring of the right recurrent laryngeal nerve was performed, reducing the risk of the lesion of the structure; then we used an intraoperative bronchoscopy to transilluminate the trachea, without identifying trajectory or communication with the diverticulum (Figure 2). Hemostasis is checked and Blake drain is placed in the surgical place. The postoperative result was excellent with the removal of drains and verification of adequate phonation.
The specimen was sent to pathology and histological analysis revealed fibrotic and inflammatory changes with numerous diverticulums that is the reason why due to the absence of predisposing personal antecedents and the histological findings, the diagnosis of congenital tracheal diverticulum was confirmed. The patient had a normal hospital stay receiving intravenous antibiotic treatment, and his symptoms disappeared without further complications.

**Discussion**

A tracheal diverticulum is a rare entity that is part of the differential diagnosis of paratracheal cysts [1]. It was first described by Rotkiansky in 1838, with poor bibliography in the world literature [2]. A report of the largest series of 64 cases, carried out by Goo et al. [3], appears in up to 1% of autopsies [4], they described that may be single or multiple [5] and Buterbaugh and Erly [6] estimated that they can occur in approximately 3.7% of the population.

They are usually found between the T1 and T2 thoracic vertebrae [3]. Most of them are asymptomatic. If they present clinical symptoms, they are usually respiratory (chronic cough, dyspnoea, stridor) and because they can act as reservoirs of secretions, they could develop chronic secondary infections in the tracheobronchial tree [7]. In some cases, it may be associated with alterations in lung function and obstructive pulmonary disease [8], mainly emphysema [9]. Only one case related to alpha-1 antitrypsin deficiency [10]. In other cases, digestive symptoms, recurrent nerve palsy, foreign body sensation and/or extrinsic airway compression had been described [11].

While the precise etiology is unknown, they have been associated with obstructive lung disease, inflammation, tracheal mucus impaction and increased intrathoracic pressures as possible causes. Also, it might be postulated that conditions associated with chronic cough, such as cystic fibrosis, could predispose the disease [12].

They have been classified into two types according to their origin, which may be congenital and acquired, based on their differences in histological type characteristics and according to their location. The former may have a component of normal tracheal elements such as muscle or cartilage in their thick wall and may be filled with mucosal contents [13,14]. They are identified as a consequence of an abnormal division of the primitive pulmonary organ and its location approximately distant from 4 to 5 centimeters of the vocal cords or may be above the carina, usually on the right side. The acquired ones can appear in any tracheal level, as a consequence of the increase of the intratracheal pressure and with a thin wall, are very susceptible to the herniation of the mucosa (which is composed of respiratory epithelium), with more frequency of the right side.

Acquired and congenital tracheal diverticula are often asymptomatic and conservative care is an adequate treatment. However, surgical treatment has been reported to be effective and safe for symptomatic tracheal diverticula to prevent the recurrence [15]. Another difference was the size of the communication port with the trachea, which is larger in the acquired forms and narrower in the congenital forms [13], even with difficult identification during surgery [16]. In addition, they have been classified into cysts (with muscle and cartilage in the wall of embryonic origin), tracheoceles and diverticula (without muscular frame and differentiated by number and size) [17]. Diverticulum commonly displaces the esophagus. For this reason, we can find clinical symptoms and patients have to be studied with chest X-rays, fiberoptic bronchoscopies and CT scans (which are not always able to identify tracheal communication) [9].

However, finding air bubbles, communication or contiguity of the tissues with empyema or a subphrenic abscess, as well as the clinical, can identify complications such as infection [17], which has been reported recently, requiring emergency surgical intervention [19,20]. Due to the description, most of them are an incidental finding for that reason, it is necessary to take a high-resolution tomography, which provides a fundamental aid to identify the characteristics of the diverticulum.

Differential diagnosis should include oesophageal disease, pharyngoecele, Zenker’s diverticulum, laryngoecele, apical pulmonary hernia, or apical ampuellae. In our case, the patient had no history of previous respiratory disease or procedures for surgical manipulation of the trachea. The communication with the trachea was not identified in the tomography and without history for predisposition or the lesion, we can argue the congenital origin [22].

In general, the postoperative complications are exceptional and could appear with recurrent compression, laryngeal paralysis [23], pneumomediastinum [2] and dyspnoea in a lactant patient [24].

Given the non-specific symptoms, the therapeutic options vary from the conservative treatment with respiratory physiotherapy, mucolytic agents, and antibiotic therapy until the surgical management that must evaluate the approach and the general condition of the patient, for the realization of the total resection of the lesion. This has been reserved for larger diverticula for aesthetic purposes [16], pediatric cases with severe respiratory symptoms [24] or more symptomatic forms with frequent superinfections [22].

Similarly, endoscopic models have been described, using cauteryization with laser or electrocoagulation, allowing the eradication of symptoms.

In summary, tracheal diverticula is a rare disease that may present a variety of symptoms and complications that require immediate surgical intervention (25), to provide favorable postoperative results, without long-term complications.
Conclusions

A tracheal diverticulum is a rare, benign entity. Frequently localized on the right side, with two possible etiologies known with a divergent presentation that must be studied and treated correctly with a personalized and specialized surgery groups to obtain satisfactory results in long-term.

Conflict of Interest

Author’s don’t have conflict of interest.

References


