*An incidental finding of a double-lumen trachea in human – a case report.*

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Short title: Double-lumen trachea in human – case report.

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Abstract

The aim of this case report is to present an incidental finding of a firm tracheal septum in a 61-year-old woman. This is an extremely rare finding and it is crucial to share our experience in management of patient with aforementioned post-intubation complication.

The patient was admitted to the hospital with mild dyspnea and preliminary diagnosis of a tracheal subglottic stenosis. During microlaryngoscopy, just below the subglottic stenosis, a firm, vertical symphysis (septum), forming a double-lumen trachea was found. There was no record of any previous difficulties with intubation. A CT scan performed after the microlaryngoscopy revealed an airway branch arising from the trachea at the level of thyroid gland and joining its lumen below.

Radiologic and endoscopic findings in the presented case hardly resemble the conditions described in the literature, as the discovered septum does not have a pseudomembranous nature nor does it form a tracheal bronchus. Therefore, the finding is thought to be an unusual complication of multiple intubations in the past.

Keywords: tracheal septum, microlaryngoscopy, tracheal stenosis

**Introduction**

The review of literature describing anomalies of tracheobronchial anatomy estimates the prevalence of congenital malformations to be 1-12% among the population of patients undergoing diagnostic endoscopic procedures in intrathoracic airways [1]. However, the majority of anomalies of tracheobronchial anatomy are caused by previous intubation [2]. In this article, we present a case of a double-lumen trachea. This is an extremely rare complication of intubation and it is crucial to share our experience in management of patient with aforementioned tracheal malformation. Especially due to the fact that radiologic and endoscopic findings in this case are unusual for acquired, post-intubation complication.

**Case report**

We present a case of 61-year –old female with an incidentally found tracheal defect of double-lumen trachea. Mircolaryngoscopy performed because of patient's subglottic stenosis revealed a firm vertical tracheal septum forming double-lumen trachea. The patient was admitted to the hospital with mild dyspnea associated with an upper respiratory tract infection (URTI) and preliminary diagnosis of a tracheal subglottic stenosis visualized in laryngoscopy (the narrowest part measuring 7,5mm). During surgery, just under subglottic stenosis, a firm, vertical symphysis (septum), forming a double-lumen trachea was found in microlaryngoscopy (Fig 1.). The stiffness of the septum prevented the tracheoscope from passing through. Patient's history however, does not include previous difficulties with intubation (during nefrectomy, laparoscopy or hysteroscopy). The patient suffered also from an episode of tracheitis and pneumonia earlier that year. A CT scan performed after the endoscopic examination, revealed an airway branch with a 7mm diameter arising from the trachea at the level of thyroid gland and joining its lumen 20mm below (Fig 2). “insert Fig 1 and 2. Here”

The mechanical balloon dilatation of tracheal stenosis located above the septum was performed leaving the tracheal septum intact. As the patient's general condition after the procedure was stable and she did not experience difficulties in breathing, the patient was released home and instructed to return for a follow-up in the outpatient setting. After 6 months a bronchofiberoscopy was performed showing good result of previous tracheal stenosis dilatation. The tracheal anomaly was still present and unchanged compared to previous examination.

Fig 1. Double-lumen trachea visualized during laryngoscopy

Fig 2. CT scan showing an airway branch with a 7mm diameter arising from the trachea at the level of thyroid gland (1A) and joining its lumen 20mm below (1C)

**Disscussion**

A review of the literature on such anomalies suggests 1) tracheal bronchus, 2) obstructive pseudomembranes (as a complication of endotracheal intubation or infections), 3) pleural junctional line that could give a false impression of a septum in diagnostic imaging or 4) tracheal secretions.

Congenital defects of the lower airways are mostly clinically relevant, in which case are usually diagnosed and managed early in life, yet they may also not manifest clinically and therefore become fortuitous findings. [3-5] They can also be associated with other syndromes. Tracheal bronchus represents one of the congenital anomalies that rarely bears any clinical relevance [6], but is not an uncommon finding, with 0,1-5% incidence during bronchoscopy [7]. Tracheal bronchi form as a result of abnormal evolution of the lung buds in the gestational period, arise mostly from the middle or lower parts of trachea [6] and are directed towards the upper lobes [8]. As it can also be associated with tracheal stenosis, a tracheal bronchus could seem a plausible differential diagnosis of our patient's malformation, however in this case the airway branch leaves the trachea only to join its lumen 2cm below.

Preoperatively, one of the most probable causes of tracheal stenosis in the patient was presumably a stenosis secondary to endotracheal intubation. The patient underwent three independent surgeries (nephrectomy, laparoscopy, hysteroscopy) in the past, under general anesthesia with the use of endotracheal tubes. Despite the increasing awareness of the possible complications of incorrect or prolonged intubation among intensive care teams, the incidence of tracheal stenosis with this etiology remains high [9]. A cuff pressure over 30mmHg can lead to mucosal ischemia and consequently damage in cartilages structure. If present, it may eventually cause fibrosis and circumferential stenosis of the trachea. [10] Most commonly observed sign of this finding is dyspnea, the severity of which depends on the stenosis level. Mild cases of subglottic stenosis may not require surgical treatment. In severe cases, surgical treatment is needed and surgical procedures might be categorized into open or endoscopic neck surgery with resection of stenotic fragment or endoscopic mechanical dilatation by balloons or rigid instruments.

The other common tracheal stenosis causes in adults are an infectious or inflammatory background. [11] This could be considered in this case as patient was treated for tracheitis in the past. However, both aforementioned causes were excluded intraoperatively, as the observed picture of firm septum dividing the trachea lumen did not morphologically resemble these suspected entities.

Pseudomembrane is also a likely explanation for a septum formed in the trachea. It could either have an idiopathic or post-infectious etiology. As a complication of endotracheal intubation, it is rare and could be severe - it may induce a life-threatening obstruction of the airways [12] and may present itself as atelectasis, respiratory infection [13,14] or post-extubation stridor [15]. Its appearance is explained by the mucosal ischemia caused by the intubation tube, resulting in overproduction of growth factors [16] or secretions [13]. Most of them however, are overlying the tracheal mucous membrane, slowly causing obstruction, instead of presenting as a vertical septum in the trachea.

Radiologic and endoscopic findings in the presented case do not resemble any condition described in the literature. Therefore, a congenital defect with minor influence on the patient's clinical condition theory cannot be rejected. However, patient’s history of uncomplicated previous intubations and recent tracheitis act strongly against this thesis. Thus, we hypothesized that in this case the most probable explanation of this lesion’s ethiology is an uncommon presentation of complication secondary to endotracheal intubation. It is crucial to perform regularly a bronchofiberoscopy and a thorough observation of this particular patient. So far, we assume the tracheal septum is clinically irrelevant, however it may predispose to dyspnea or inflammation. Although this anomaly is a rare finding in humans, it is important for clinicians to be aware of its presence. This case report suggests that double-lumen trachea is not a life threatening condition and it can be managed with “watch and wait” approach.

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Conflict of interest:

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References:

1. McDonough RJ., Niven AS., Havenstrite KA. Congenital pulmonary airway malformation: a case report and review of the literature. Respiratory Care. 2012;57(2):302–306.
2. Wain JC. Postintubation Tracheal Stenosis. Semin Thorac Cardiovasc Surg. 2009;21(3):284–9.
3. Graça A, Lucas M, Pereira L, Barreto C, Lobo L, Gonçalves M et al. (2004) Sliding tracheoplasty: a good choice for the treatment of congenital tracheal stenosis. Acta Med Port. 17(3):247-252.
4. Oliveira-Santos JA, Pereira-da-Silva L, Clington A, Serelha M. (2004) Neonatal bronchoscopy: a retrospective analysis of 67 cases and a review of their indications. Acta Med Port. 17(5):341-348.
5. Gonlugur U., Efeoglu T., Kaptanoglu M., Akkurt I. (2005). Major anatomical variations of the tracheobronchial tree: bronchoscopic observation. Anatomical Science International, 80(2), 111–5.
6. Zach MS., Eber E. (2001). Adult outcome of congenital lower respiratory tract malformations. Thorax, 56(1), 65–72.
7. Ruchonnet-Metrailler I., Abou Taam R., de Blic J. (2015). Presence of tracheal bronchus in children undergoing flexible bronchoscopy. Respiratory Medicine, 109(7), 846–850.
8. Setty SP., Michaels AJ. (2000). Tracheal bronchus: case presentation, literature review, and discussion. The Journal of Trauma, 49(5), 943–5.
9. Donatelli J, Gupta A, Santhosh R, Hazelton TR, Nallamshetty L, Macias A, et al. To breathe or not to breathe: a review of artificial airway placement and related complications. Emerg Radiol. 2014;22(2):171–9.
10. De S., De S. (2008). Post intubation tracheal stenosis. Indian Journal of Critical Care Medicine : Peer-Reviewed, Official Publication of Indian Society of Critical Care Medicine, 12(4), 194–7.
11. Abdala FA. Benign Tracheal Stenosis. Chest Surgery. Berlin Heidelberg:Springer; 2015. 31–40.
12. Desir A., Ghaye B. (2009). Congenital Abnormalities of Intrathoracic Airways. Radiologic Clinics of North America, 47(2), 203–225.
13. Sehgal I. S., Dhooria S., Bal A., Aggarwal AN., Behera D., Agarwal R. (2016). Obstructive Fibrinous Tracheal Pseudomembrane After Endotracheal Intubation. Respiratory Care, 61(9), 1260–1266.
14. Arenas-de Larriva M. del S., Cosano-Povedano J., Cosano-Povedano A. (2014). Inflammatory Tracheal Pseudomembrane. Archivos de Bronconeumología (English Edition), 50(7), 307–308.
15. Crespo-Lessmann A., Torrego-Fernández A. (2013). Obstructive Inflammatory Tracheal Pseudomembrane. Archivos de Bronconeumología (English Edition), 49(9), 402–404.
16. Fiorelli A., Vicidomini G., Messina G., Santini M. (2011). Spontaneous expectoration of an obstructive fibrinous tracheal pseudomembrane after tracheal intubation. European Journal of Cardio-Thoracic Surgery, 40(1), 261–263.