Radiology Case Reports 17 (2022) 2052–2057

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

A rare case of unilateral vocal cord paralysis: neurovascular conflict due to an aberrant bronchial artery detected at computed tomography ☆

Maurizio CÈ, BA, MD[a,], Francesco Bombaci, MD[b], Silvana SDAO, MD[b], Sara Marziali, MD[a], Giovanni Irmici, MD[a], Sara Boemi, MD[a], Marco Cervelli, MD[a], Michaela Cellina, MD[b]*

[a] *Postgraduate School of Diagnostic and Interventional Radiology, University of Milan, Via Festa del Perdono, 7, 20122, Milan, Italy*

[b] *Radiology Department, ASST Fatebenefratelli Sacco, piazza Principessa Clotilde 3, 20121, Milan, Italy*

ARTICLE INFO

Article history:
Received 29 December 2021
Revised 9 March 2022
Accepted 9 March 2022

Keywords:
Bronchial artery
Anatomical variants
Neurovascular conflict
Vocal cord paralysis
Dysphonia

ABSTRACT

We report the case of a 29-year-old patient without medical history presenting with dysphonia associated with left unilateral vocal cord paralysis. The patient underwent a contrast-enhanced computed tomography with an angiographic arterial phase of the head, neck and chest, and the only significant finding was the presence of a large, aberrant right bronchial artery originating directly from the aortic arch, where the recurrent left laryngeal nerve loops. After excluding alternative etiologies, the hypothesis of neurovascular conflict between this vessel and the recurrent left laryngeal nerve was formulated. To the best of our knowledge, this is the first case reported in the literature. Thanks to its high spatial resolution, contrast-enhanced computed tomography is the examination of choice for the study of anatomical variants and should be included in the routine work-up of patients presenting with unilateral vocal cord paralysis.

Introduction

Dysphonia is the impairment of voice production; this term is sometimes used interchangeably with hoarseness, a symptom of altered voice quality. It has a lifetime prevalence of 30% and can affect patients of all ages and genders, with an increased prevalence in patients with voice misuse or overuse [1]. Pathophysiology is characterized by impaired vocal fold oscillations and can occur secondary to damage to the vocal

☆ Competing Interests: The authors have declared that no competing interests exist.

* *Corresponding author.*
E-mail address: mauriziocе.md1@gmail.com (M. CÈ).

https://doi.org/10.1016/j.radcr.2022.03.033

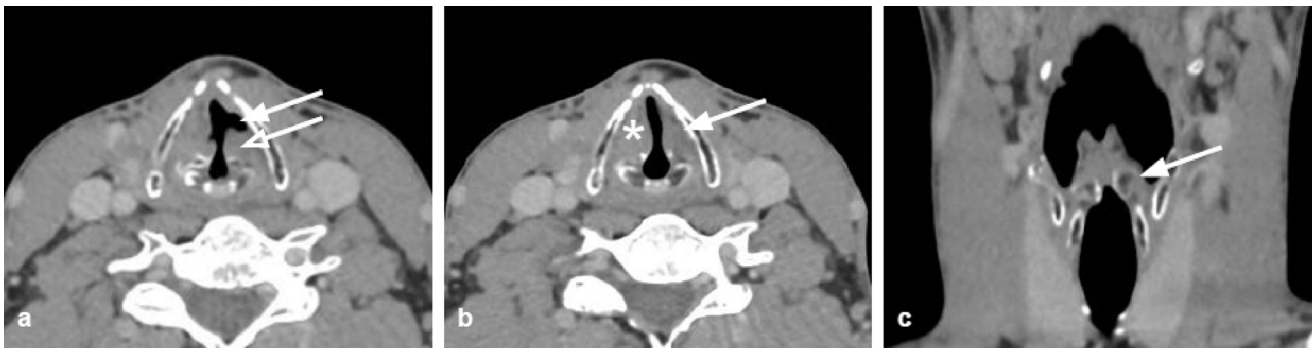

Fig. 1 – (A) Medialization and thickening of the left aryepiglottic fold (empty arrow). Note the "mushroom sign" resulting from the combination of the medialization of the left posterior cord margin (empty arrow) and dilatation of the left laryngeal ventricle (arrow). (B) Abduction of the left vocal cord during breath-hold imaging (arrow), with compensatory medial bowing of the contralateral vocal cord (asterisk). (C) Medial and cranial deviation of the left arytenoid cartilage.

folds (benign or malignant) or laryngeal nerves [1]. In the latter case, the patients may present at the clinical examination with a unilateral paralysis of the vocal cord [2].

The closure of the vocal cords is regulated by the recurrent laryngeal nerves, which innervates all the muscles of the larynx (excluding the cricothyroid muscle) and in particular the posterior cricoarytenoid muscle, the only muscle with the function of opening the vocal cords [3]. Recurrent laryngeal nerves arise from the tenth cranial nerve (C.N. X, Vagus) and have a long course throughout the neck and thorax, descending alongside the trachea. The left nerve loops under the arch of the aorta while the right under the subclavian artery before returning to the larynx. There can be significant variation in anatomy, with a small proportion of the population having a non-recurrent laryngeal nerve [3].

Any lesion throughout the course of the nerve, from the brainstem to the inferior border of the nerve, can result in the absence of cord movement and dysphonia. Etiologies include iatrogenic surgical lesions, neck or chest trauma, malignant tumors, stroke, and other neurological diseases (myasthenia gravis, multiple sclerosis), vascular, inflammatory, and infectious conditions [4]. Idiopathic paralysis disease should only be diagnosed when all other causes have been ruled out.

In light of their anatomical path, imaging represents a crucial step in the diagnostic process to search for causes of nerve damage not detectable on clinical examination.

Thanks to its fast acquisition time, high resolution, and multiplanar reconstructions availability, multidetector contrast-enhanced computed tomography (CECT) scanning from the brainstem superiorly, to the mediastinum, inferiorly, is the technique of choice for the full-length course assessment of the recurrent laryngeal nerve [1].

We present the case of a young male patient who presented to our attention for a left recurrent laryngeal nerve palsy. The CECT demonstrated the presence of a right aberrant bronchial artery running through the aorticopulmonary window and resulting in a neurovascular conflict.

To the best of our knowledge, this is the first case reported in the literature.

Case report

A young 29-year-old male patient presented to the medical examination due to dysphonia which had arisen for about 2 months. No significant medical history was reported. The patient was referred to the Otolaryngologist for laryngoscopy, which evaluated both the laryngeal structure and function. The fibroscopic examination revealed fixity of the left hemilarynx in the paramedian position, with modest movements of the ipsilateral arytenoid cap. Right motility was normal. For the rest, no pathological findings in the nasopharynx, tongue, and pyriform sinuses. Breathing space was conserved. At the narrow-band imaging, there were no pathological vascularization areas. At the videostroboscopy with pulsed light a normal cordial mucous wave was present.

The patient was suggested a 1-week follow-up which confirmed the clinical picture. It was therefore suggested to perform an ultrasound of the neck and a CECT to search for possible causes of unilateral paralysis of the vocal cord not detectable at the clinical examination. Ultrasound of the neck was negative for suspicious expansive lesions affecting the thyroid or parathyroids [5].

After acquiring informed consent, the CT examination was performed on a 64 multislice CT (General Electric, Boston, MA) using automatic tube current modulation. The acquisition parameters, according to our Institution protocol, were as follows: reference tube current 250 mAs, tube voltage 120 kV, rotation time 0,5 sec, collimation 128 × 0.625 mm. Contrast medium concentration (120 ml of Ultravist 370 mg I / mL, Bayer, Leverkusen, GE) was injected intravenously at 4,8 mL/s and bolus tracking technique with the ROI placed in the ascending aorta was used to synchronize the start of the acquisition with contrast bolus arrival and obtain an angiographic arterial phase to detect possible vascular anomalies. Venous phase was also acquired. The image dataset was then transferred for analysis and post-processing.

No pathological findings were reported in the brain. A post-contrast study of the neck was performed under normal conditions and during phonation too. Abduction of the left vo-

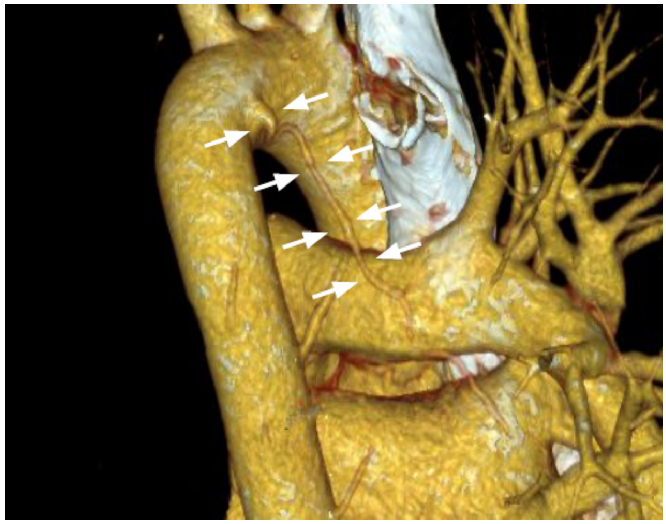

Fig 2 – 3D reconstruction highlighting the presence of a heterotopic bronchial artery, originating from the right posterolateral wall of the aortic arch, dilated at the proximal tract (arrows).

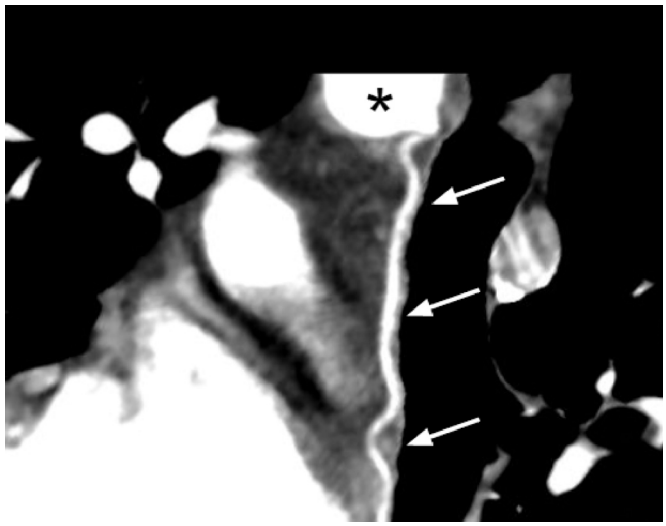

Fig. 3 – Curvilinear multiplanar reconstruction showing the course of the anomalous bronchial artery (arrows), originating from the aortic arch (asterisk).

cal cord was observed (Fig. 1). No structural alterations of the larynx or areas of altered post-contrast enhancement were found.

Chest evaluation didn't show focal pulmonary opacities or suspicious nodules. No significant ilo-mediastinal lymphadenopathies were found and no pleural effusion was present either.

The thyroid and thymus showed size within the normal range and homogeneous density.

As the only noteworthy relief, CECT revealed the presence of an aberrant bronchial artery (an anatomic variant), which originated from the right posterolateral wall of the aortic arch; this bronchial artery showed an ectatic appearance at the origin (about 3.5 mm), resulting in a partial obliteration of the adipose space at the tracheoesophageal groove (Figs. 2-6). The left bronchial artery showed a regular course.

In the absence of other imaging or instrumental findings that could justify the symptom, the hypothesis of neurovascular conflict between the left recurrent laryngeal nerve loop and the aberrant right bronchial artery was posed.

Spontaneous resolution of the symptoms occurred within 1 month. The follow-up endoscopy showed complete resolution of the vocal cord palsy. Clinical follow-up is still ongoing.

Discussion

The concept of "neurovascular conflict" generally applies in cranial neuropathies, the most common are trigeminal neuralgia and hemifacial spasm. It indicates a pathophysiological phenomenon in which a vascular structure determines me-

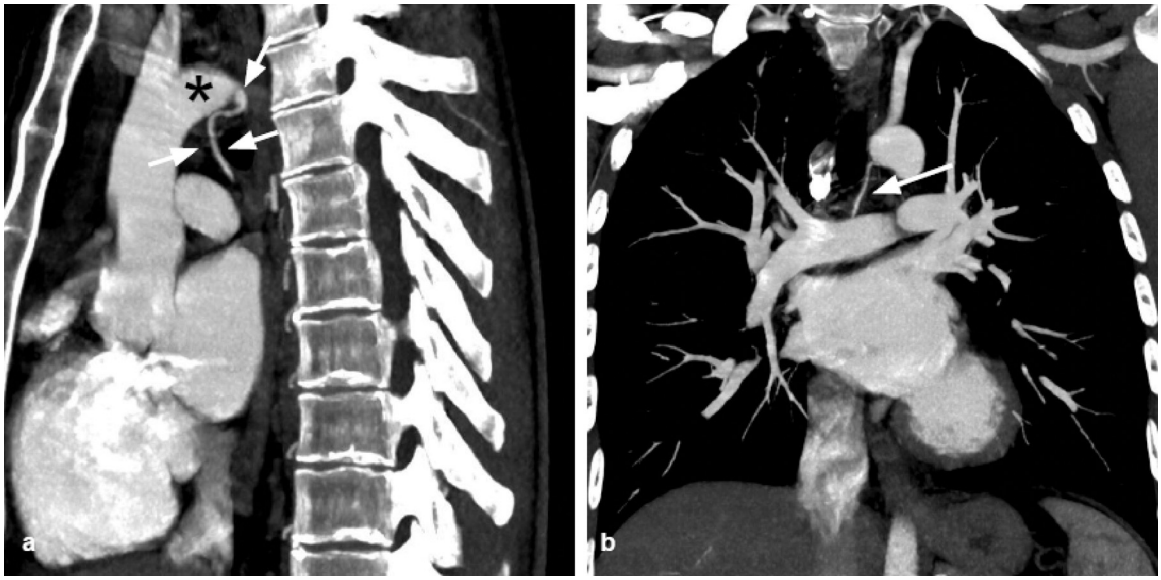

Fig. 4 – Maximum Intensity Projection Reconstructions on the sagittal (A) and coronal (B) plane demonstrate the course of the proximal tract of the aberrant bronchial artery (arrows), originating from the aortic arch (asterisk).

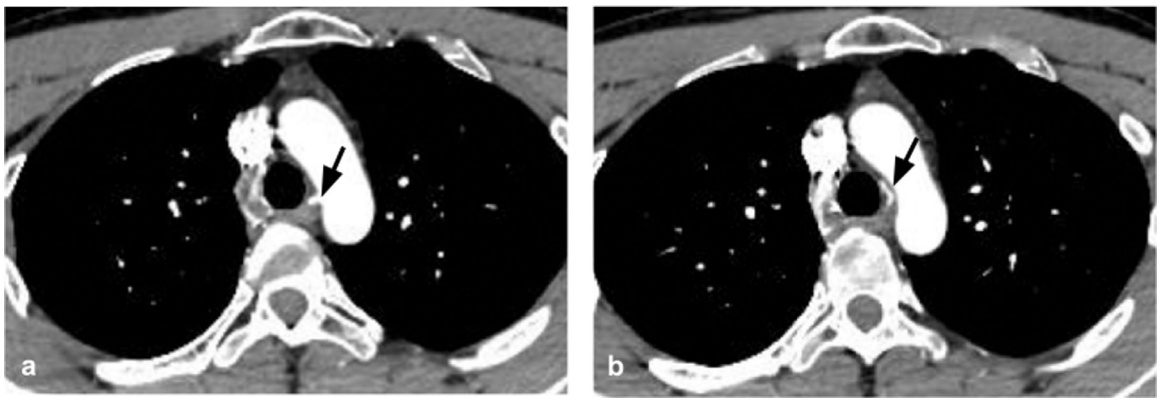

Fig. 5 – Axial CT showing the course of the aberrant bronchial artery in the tracheoesophageal groove (A) and medial to the aortic arch (B) (black arrows).

chanical irritation/compression of a nerve due to its proximity. Neurovascular conflicts present with neuropathic symptoms such as numbness and tingling, burning and/or pain, even very severe, and functional impairment [6].

At the base of this phenomenon, there are congenital or acquired anatomical conditions that lead to neurovascular contact, even if not all cases of neurovascular contact are symptomatic. In our case, the neurovascular conflict between the left recurrent laryngeal nerve and the right bronchial artery, which showed an anatomical variant.

Normal anatomy involves 2 bronchial arteries, 1 for the right lung and 2 for the left lung [7], but many variants in number and origin have been identified, with important therapeutic clinical implications [8–10]. Bronchial arteries provide blood supply to the bronchial structures, the interstitial connective tissue of the lung, the visceral pleura, the wall of the pulmonary vessels, the bronchial lymph nodes, and also distributes some branches to the esophagus and pericardium.

Two left bronchial arteries usually arise directly from the anteromedial thoracic aorta. The right bronchial artery usually has a common origin with a posterior intercostal artery called the intercostobronchial trunk and arises from the right anteromedial aspect of the thoracic aorta [3]. Running behind the bronchus, it follows its path, dividing with it into the bronchial branches [11].

In our case, the right bronchial artery presents an anatomical variant, originating from the right anterolateral wall of the aortic arch and running first in the tracheoesophageal groove, then medial to the infero-medial wall of the aortic arch. Accordingly, our anatomical imaging-based observations allow us to conclude that the neurovascular conflict with the left recurrent laryngeal nerve is the cause of dysphonia.

Another extremely rare cause of neurovascular conflict with the recurrent laryngeal nerve is represented by the common bronchial artery trunk (ACBAT) [12]. This unusual anatomical variant consists of a common bronchial artery that

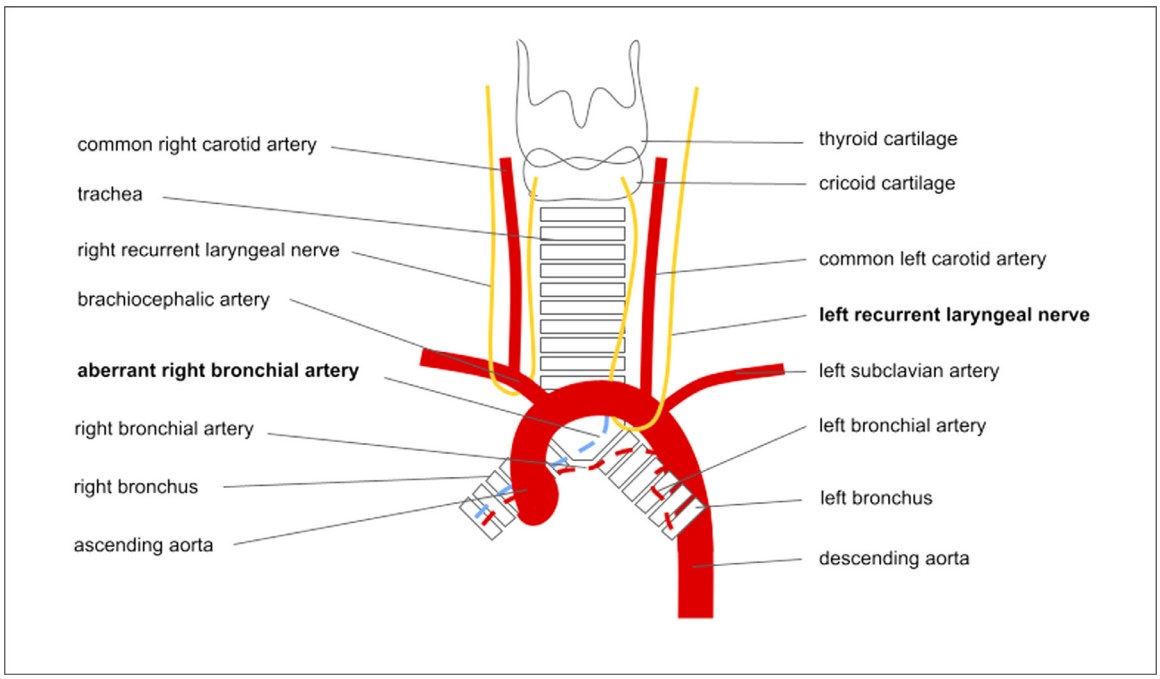

Fig. 6 – The figure represents the course of the right and left recurrent laryngeal nerves, of the bronchial arteries (in red, most common presentation) and of the aberrant right bronchial artery identified in our patient (in blue). The aberrant artery runs where the left recurrent laryngeal nerve loops below the aortic arch.

is usually tortuous and dilated and can determine compression of the laryngeal recurrent nerve with consequent vocal cord palsy [12].

Computed Tomography is considered the gold-standard technique for anatomical variants assessment [13,14] and its execution with the appropriate craniocaudal extension represents the first approach for the study of recurrent laryngeal nerve palsy [15].

CECT signs of recurrent laryngeal palsy at the neck include the ipsilateral piriform sinus dilatation, the medial rotation and thickening of the aryepiglottic fold, and ipsilateral laryngeal ventricle dilatation [14]. When the palsy is caused by any disorder in the proximal course of the nerve, indirect signs on CT consist of dilatation of the ipsilateral oropharynx, atrophic thinning of the pharyngeal constrictor muscles, and uvular displacement away from the causing lesion's side: if these signs are recognizable, a brain magnetic resonance is recommended [15].

The most common mediastinal causes are traumas with direct or deceleration nerve injury, lung cancer, lymphoma, nodal metastases, esophageal cancer, neurogenic neoplasms, thyroid and thymic neoplasms, pulmonary embolism, and aortic dissection; uncommon etiologies consist of sarcoidosis, amyloidosis, tuberculosis [15].

According to the age of our patient, and his silent clinical history, lymphoma, thymic and thyroid carcinomas could be considered the main differential diagnosis.

The execution of the angiographic phase has been decisive for the detection of the vascular anatomical variant causing the symptomatology.

Conclusion

Recurrent laryngeal nerve paralysis may be caused by several different entities, including vascular anomalies, as in our case.

CECT allows the assessment of the entire course of the vagus and recurrent laryngeal nerve and can be considered the first-line diagnostic tool in the differential diagnosis of patients with dysphonia and suspected nerve involvement.

Radiologists should be aware of the possibilities of uncommon causes of recurrent laryngeal nerve paralysis related to vascular anatomical variation and include an angiographic phase in their clinical CT protocols.

Patient consent

The patient has given written informed consent to the execution of the contrast enhanced computed tomography and the use of his data anonymously for scientific purposes.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2022.03.033.

REFERENCES

[1] Stachler RJ, Francis DO, Schwartz SR, Damask CC, Digoy GP, Krouse HJ, et al. Clinical practice guideline: hoarseness (Dysphonia) (Update) executive summary. Otolaryngol Head Neck Surg 2018;158(3):409–26 Erratum in: Otolaryngol Head Neck Surg. 2018 Aug;159(2):403. PMID: 29494316. doi:10.1177/0194599817751031.

[2] Williamson AJ, Shermetaro C. Unilateral vocal cord paralysis. [Updated 2021 Aug 11]. StatPearls [Internet], Treasure Island (FL): StatPearls Publishing; 2021. Available from: https://www.ncbi.nlm.nih.gov/books/NBK535420/ .

[3] Standring S. Chapter 54 - Pleura, lungs, trachea and bronchi. In: Gray's Anatomy. (2015).

[4] Cantarella G, Dejonckere P, Galli A, Ciabatta A, Gaffuri M, Pignataro L, et al. A retrospective evaluation of the etiology of unilateral vocal fold paralysis over the last 25 years. Eur Arch Otorhinolaryngol 2017;274(1):347–53. doi:10.1007/s00405-016-4225-9.

[5] Floridi C, Cellina M, Buccimazza G, Arrichiello A, Sacrini A, Arrigoni F, et al. Ultrasound imaging classifications of thyroid nodules for malignancy risk stratification and clinical management: state of the art. Gland Surg 2019;8(Suppl 3):S233–44. doi:10.21037/gs.2019.07.01.

[6] Haller S, Etienne L, Kövari E, Varoquaux AD, Urbach H, Becker M. Imaging of neurovascular compression syndromes: trigeminal neuralgia, hemifacial spasm, vestibular paroxysmia, and glossopharyngeal neuralgia. AJNR Am J Neuroradiol 2016;37(8):1384–92. doi:10.3174/ajnr.A4683.

[7] Walker CM, Rosado-de-Christenson ML, Martínez-Jiménez S, Kunin JR, Wible BC. Bronchial arteries: anatomy, function, hypertrophy, and anomalies. Radiographics 2015;35(1):32–49. doi:10.1148/rg.351140089.

[8] Jie B, Sun XW, Yu D, Jiang S. An aberrant left bronchial artery originating from the proximal ascending aorta. Cardiovasc Intervent Radiol 2015;38(6):1632–4. doi:10.1007/s00270-015-1061-z.

[9] Choi WS, Kim MU, Kim HC, Yoon CJ, Lee JH. Variations of bronchial artery origin in 600 patients: Systematic analysis with multidetector computed tomography and digital subtraction angiography. Medicine (Baltimore) 2021;100(22):e26001. doi:10.1097/MD.0000000000026001.

[10] Hartmann IJ, Remy-Jardin M, Menchini L, Teisseire A, Khalil C, Remy J. Ectopic origin of bronchial arteries: assessment with multidetector helical CT angiography. Eur Radiol 2007;17(8):1943–53. doi:10.1007/s00330-006-0576-8.

[11] Monroe EJ, Pierce DB, Ingraham CR, Johnson GE, Shivaram GM, Valji K. An interventionalist's guide to hemoptysis in cystic fibrosis. Radiographics 2018;38(2):624–41. doi:10.1148/rg.2018170122.

[12] Eid N, Otsuki Y. Anomalous bronchial arteries with surgically important relationships to abnormal recurrent laryngeal nerve and thoracic duct. Clin Anat 2010;23(8):897–8. doi:10.1002/ca.20932.

[13] Cappella A, Gibelli D, Cellina M, Mazzarelli D, Oliva AG, De Angelis D, et al. Three-dimensional analysis of sphenoid sinus uniqueness for assessing personal identification: a novel method based on 3D-3D superimposition. Int J Legal Med 2019;133(6):1895–901. doi:10.1007/s00414-019-02139-5.

[14] Cellina M, Meglio LD, Marziali S, Cè M, Oliva G, Visconti A, et al. A rare case of congenital atresia of the left main coronary artery. Radiol Case Rep 2021;17(1):232–4. doi:10.1016/j.radcr.2021.10.012.

[15] Paquette CM, Manos DC, Psooy BJ. Unilateral vocal cord paralysis: a review of CT findings, mediastinal causes, and the course of the recurrent laryngeal nerves. Radiographics 2012;32(3):721–40. doi:10.1148/rg.323115129.