Wydawnictwo UR 2024
ISSN 2544-1361 (online)
doi: 10.15584/ejcem.2024.3.31

*http://www.ejcem.ur.edu.pl*
*European Journal of Clinical and Experimental Medicine*
*Eur J Clin Exp Med 2024; 22 (3): 702–707*

**CASUISTIC PAPER**

# Benign endotracheal tumor (hamartoma) mimicking bronchial asthma

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**ABSTRACT**

**Introduction and aim**. The most common benign tumor of the lung is hamartoma. In many cases, it is a spherical tumor, located peripherally, often without clinical symptoms. Predominantly it is found accidentally during radiological examination. In some cases the tumor reaches a significant size in the lung parenchyma or in the lumen of the bronchi or trachea. Then, symptoms such as cough, dyspnea, wheezing, less commonly hemoptysis, and chest pain may occur. In addition, tumors located endobronchial or endotracheal may cause recurrent pneumonia or mimic obstructive diseases of the lower respiratory tract such as chronic obstructive pulmonary disease, or bronchial asthma. We present the case of a patient with an endotracheal tumor mimicking bronchial asthma.

**Description of the case**. A 53-year-old male was taking bronchodilators and inhaled steroids for several months. The baseline chest radiograph showed no abnormalities. Spirometry suggested an obstruction of respiratory flow in the central or upper airways. The lack of improvement after asthma treatment required an extension of the diagnosis. Computed tomography allowed accurate visualization of the tumor lesion of the trachea, which was significantly obstructing its lumen, and resection was carried out.

**Conclusion**. Our case demonstrates that tracheal tumors can present symptoms similar to respiratory tract diseases. In unresolved cases, spirometry and computed tomography are helpful in proper diagnosis.

**Keywords**. benign endotracheal splinter tumor (hamartoma), benign endotracheal tumor, endotracheal resection of tumor

## Introduction

Hamartoma is one of the most common benign tumor of the respiratory system. The incidence of the tumor in population is about 0.2%. It is 2–3 times more frequently found in men, and usually has character of a peripherally located lung tumor. In 10% of cases it may involve the bronchi, or trachea.[1] It accounts for approximately 8% of all lung tumors, 77% of which are benign.[2] The lesion may be composed of all mesenchymal tissue of lungs and airways, such as hyaline cartilage, adipose tissue, connective tissue, and smooth muscle.[3] If the lesions are peripheral, and small in size they do not cause complaints and are usually detected accidentally during imaging. However, if they reach a significant size, they may present with symptoms caused by compression of adjacent parts of the lungs, and bronchi such as coughing or hemoptysis. In case of endobronchial or endotracheal localization additional symptoms related to obstruction, such as dyspnea, wheezing, chest pain, or recurrent pneumonia may occur. On chest X-ray imaging, the lesion usually presents

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Received: 5.04.2024 / Revised: 18.05.2024 / Accepted: 27.05.2024 / Published: 30.09.2024

Kaziród T, Tokarski S, Kaznowska E, Rzeszutko-Grabowska M. Benign endotracheal tumor (hamartoma) mimicking bronchial asthma. *Eur J Clin Exp Med.* 2024;22(3):702–707. doi: 10.15584/ejcem.2024.3.31.

as a fairly well-defined, peripherally located, near circular or oval shadowing with uneven saturation, often with the presence of calcifications.

In some cases, calcifications in the lesion give a characteristic popcorn-like appearance. Usually the lesion is solitary, but less often multiple lesions are present. On chest computed tomography, the lesion is usually a tumor less than 2.5 cm in size with smooth or lobulated margins, with density characteristic of adipose tissue or calcifications.[4]

The prognosis of pulmonary hamartoma is good. Malignant changes have not been observed. The most common increase in lesion size is roughly 1–10 mm per year.[2] Treatment usually consists of removal of the lesion, lobectomy, or in extreme cases pneumonectomy. Endoscopic removal of the endotracheal or endobronchial lesion is also possible, together with treatment of pneumonia. In other cases, conservative treatment with observation of the lesion is done.

The primary symptoms of the bronchial asthma or chronic obstructive pulmonary disease are often nonspecific and can be considered as a variety of respiratory illnesses. This symptomatic resemblance can lead to diagnostic ambiguity, finally resulting in misdiagnoses or delayed diagnoses.

Spirometry can be used to assess lung functions, monitor the disease progression and response to treatment. Proper determination of the obstruction upper airways indicators in spirometry, can detect the changes suggesting larynx or trachea disorders. The findings in the mediastinum, including those in trachea, which are invisible in X-ray, are precisely evaluated by computer tomography.[5]

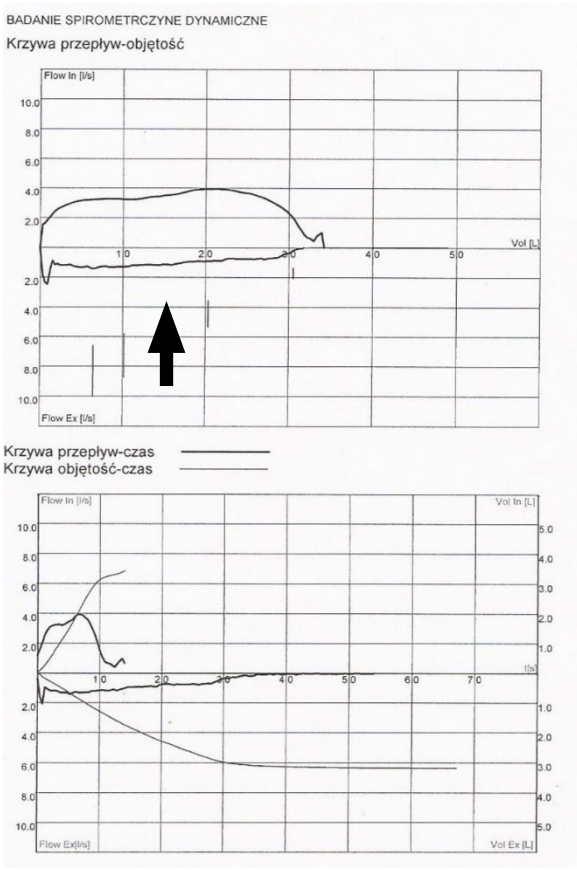
## Aim

The presented article indicates that spirometry and computed tomography are helpful in the diagnosis of lesions located in the trachea.

## Description of the case

A 53-year-old patient, non-smoker, was admitted to the department of pulmonology and allergology for extended diagnosis of dyspnea. He had been treated for 9 months for bronchial asthma without effect. Bronchodilators used included long-acting β mimetic in combination with an inhaled steroids and cholinolytics.

An anterior to posterior chest X-ray taken 10 months earlier showed an insignificantly enhanced bronchovascular pattern of the lungs, otherwise the image showed age-appropriate lungs and hilum, and free costophrenic angles. On admission to the clinic, the patient reported a feeling of dyspnea and wheezing on slight exertion, including when bending down and after eating. He denied coughing, hemoptysis or chest pain.




| Parametr | Jedn. | WN | WZ | %WN | LSR | P |
|---|---|---|---|---|---|---|
| FVC | l | 4.07 | 3.19 | 78.4 | -1.44 | 7.6 |
| FIVC | l | 4.07 | 3.44 | 84.3 | -1.05 | 15.1 |
| AT | l | | 0.24 | | | |
| AT/FIVC | | | 0.07 | | | |
| FEV 0.5 | l | 2.64 | 0.66 | 24.9 | -2.88 | 1.0 |
| FEV 1 | l | 3.28 | 1.29 | 39.3 | -3.91 | 1.0 |
| FEV 2 | l | 3.65 | 2.28 | 62.4 | -1.32 | 9.6 |
| FEV 3 | l | | 2.97 | | | |
| FEV 6 | l | | 3.17 | | | |
| FEV1/FVC | l/l | 0.78 | 0.40 | 52.0 | -5.20 | 1.0 |
| FEV1/FIVC | l/l | 0.78 | 0.38 | 48.3 | -5.60 | 1.0 |
| FEV1/FVCMAX | l/l | 0.78 | 0.38 | 48.3 | | |
| FEV1%FEV3 | % | | 43.44 | | | |
| FEV1%FEV6 | % | | 40.75 | | | |
| MEF 75 | l/s | 7.28 | 1.25 | 17.2 | -3.52 | 1.0 |
| MEF 50 | l/s | 4.45 | 1.16 | 26.1 | -2.49 | 1.0 |
| MEF 25 | l/s | 1.72 | 0.76 | 44.2 | -1.23 | 11.4 |
| MEF 25/75 | l/s | 3.72 | 1.04 | 27.9 | -2.58 | 1.0 |
| MEF 75/85 | l/s | | 1.33 | | | |
| PEF | l/s | 8.31 | 2.54 | 30.6 | -4.76 | 1.0 |
| PEF/MEF75 | | 1.14 | 2.03 | 177.5 | | |
| PIF | l/s | 7.88 | 3.95 | 50.2 | | |
| PEF/PIF | | 1.05 | 0.64 | 61.0 | | |
| MIF 50 | l/s | | 3.65 | | | |
| MEF50/MIF50 | | | 0.32 | | | |
| FIV 1 | l | | 3.07 | | | |
| FIV1/FIVC | | | 0.89 | | | |
| TFVC | s | | 3.39 | | | |
| TFIVC | s | | 1.41 | | | |
| TFVC/TFIVC | | | 2.41 | | | |
| AEX | l*l/s | 18.13 | 3.26 | 18.0 | | |
| AIN | l*l/s | | 11.56 | | | |
| AEX/AIN | | | 0.28 | | | |
| MVV | l/min | 131.32 | 51.60 | 39.3 | | |
| Ilość pom. zgodnych ERS ATS | | | 1 | | | |
| Plateau | T/N | TAK | TAK | | | |
| FET | s | 6.00 | 6.70 | | | |
| TPEF | s | 0.13 | 0.05 | | | |
| EV | l | 0.16 | 0.02 | | | |
| FVCQ | | | D | | | |
| FEV1Q | | | D | | | |
| FIVCQ | | | D | | | |

**Fig. 1.** Flow-volume curve shows significant expiration flattening – left upper part (black arrow), volume-time curve – left lower part, spirometry protocol – on the right

On physical examination, inspiratory and expiratory wheezes were noted. Functional tests showed: in spirometry, significant obstructive-type ventilation abnormalities ($FEV_1$ 1.29 l – 39.3% of the predicted value 3.28 liters, FVC 3.19 l – 78.4% of the predicted value 4.07 liters, $FEV_1$/FVC ratio: 0.40 – 52% of the predicted value 0.78, positive reversibility test – increase in $FEV_1$ by 300 ml (9%), FVC by 620 ml (15.1%)). $FEV_1$/PEF ratio 0.70 (0.63 after reversibility test) or $FEV_1$/PEF 8.46 ml/l/min ($FEV_1$ in ml, PEF in l/min) – indicates narrowing of the central or upper respiratory tract (Fig.1).

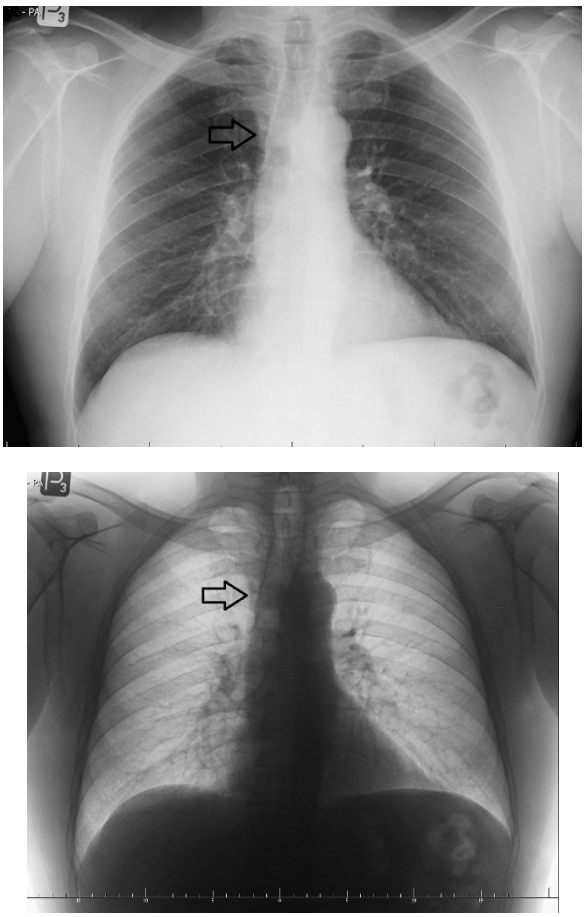


**Fig. 2.** Chest radiograph in antero-posterior projection, arrows point to a shadow in the distal 1/3 of the trachea

Body plethysmography volumetric parameters did not deviate from normal: TLC 94% of the predicted value, RV 97% of the predicted value. In contrast, a significant increase in respiratory resistance values was observed. Due to persistent inspiratory and expiratory wheezing on physical examination, suspected laryngeal stridor, the patient was consulted by otorhinolaryngologists, where no pathology was found in the larynx or the upper tracheal segment.

Chest radiograph in anterior to posterior projection showed no focal changes in either lung field, normal hilum was visualized, non-dilated mediastinal shadow, and free costophrenic angles. In the tracheal projection at the level of the aortic arch a near oval shadow was noted. A computed tomography scan showed a tumor 18 mm in diameter in the tracheal lumen, located 25 mm above the main branch, emerging from the lateral wall on the left side, smoothly circumscribed, homogeneous in structure and low in density corresponding to adipose tissue (approximately 115 IU). The lesion significantly constricted the tracheal lumen leaving 4 mm wide space around the tumor (Fig. 2 and Fig. 3). The patient was transferred to the Department of Thoracic Surgery, where endoscopic electro resection of the tracheal tumor with a metal loop was performed using a rigid bronchoscope. Subsequently, argon coagulation of the tracheal wall at the site of the severed pedicle was performed under bronchofiberoscope guidance, achieving full hemostasis. Two months after the procedure, a follow-up bronchofiberoscopy was performed, during which no recurrence of the tracheal tumor was noted.

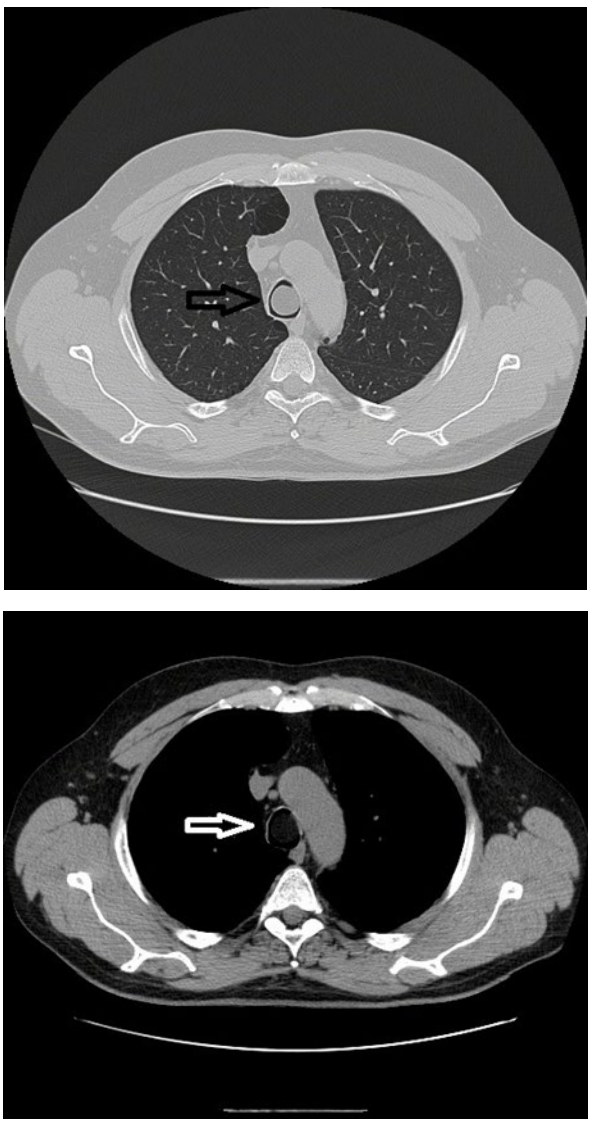


**Fig. 3.** CT scan of the thorax at the level of aortic arch: on the left – lung window, on the right – mediastinal window, the arrows indicate a lesion in the tracheal lumen

Macroscopically, the tracheal tumor presented as a partially encapsulated lesion measuring 20x19x20 mm with a base of excision of 14x12 mm, solid in cross-section, pale yellowish in color (Fig. 4).

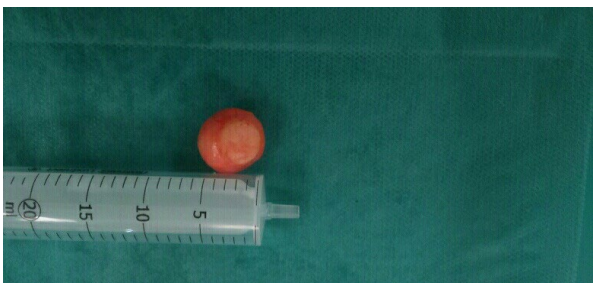


**Fig. 4.** Macroscopic image of the lesion

Histopathological examination revealed that the tumor composed predominantly of mature adipose tissue, fibro myxoid tissue, and pseudo glandular structures, arranged in disorganized manner. In some fields a lobular structure with the presence of clefts from the respiratory epithelium was present. On the lumen side of the bronchus, the tumor was covered by benign respiratory epithelium undergoing squamous metaplasia. Beneath the epithelium, a focal, chronic inflammatory infiltrate was visible. No cartilage elements or other tissues of mesenchymal origin were found. The tumor was removed in its entirety.

Microphotographs show mature adipose and fibro myxoid tissue overlying the respiratory epithelium. No cartilaginous tissue was visualized. The slice is from the endobronchial surface of the tumour. Slightly deeper, islands of benign bronchial glands with seromucous structure were found (Fig. 5–7).

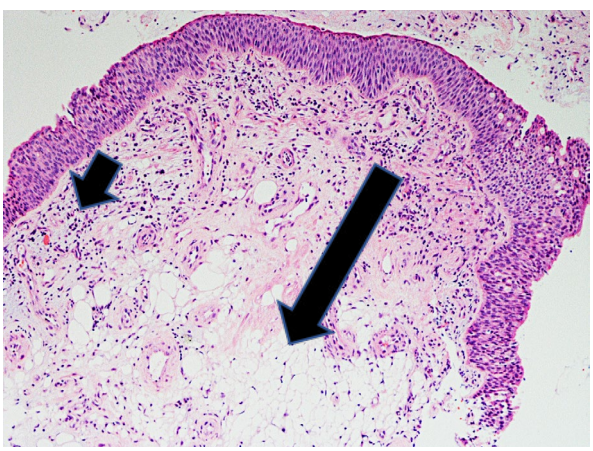


**Fig. 5.** Hamartoma composed of mature adipose tissue (large arrow) and fibro-myxoid tissue (small arrow) containing blood vessels with thickened walls, the nodule is covered by a benign, slightly proliferated squamous epithelium undergoing squamous metaplasia, beneath the epithelium, a chronic inflammatory infiltrate is visible (H&E stain, 200X)

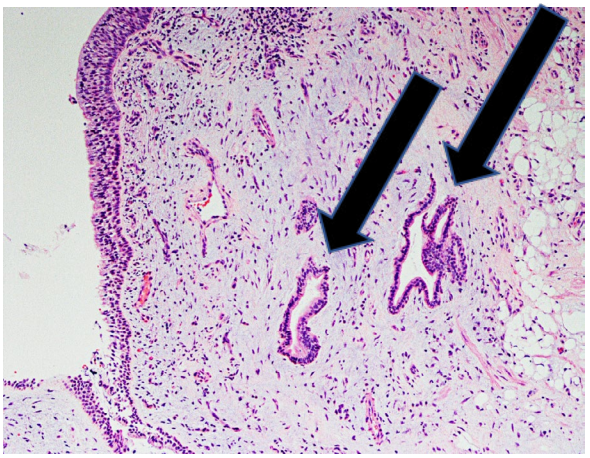


**Fig. 6.** Epithelial elements in the hamartoma tissue, forming pseudo glandular structures (arrows), fibro-myxoid tissue and islands of mature adipose tissue in the background (H&E stain, 100X)

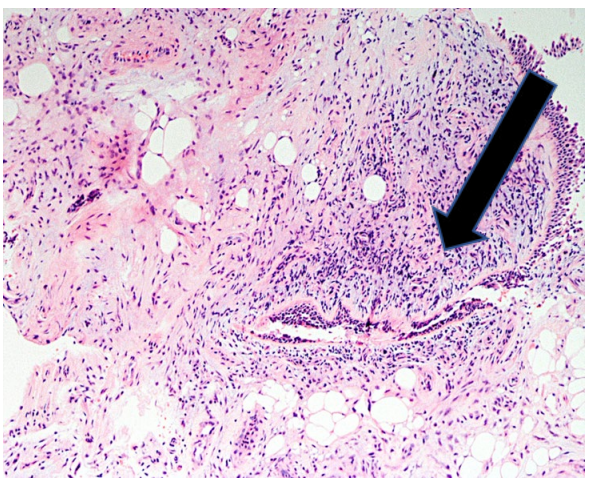


**Fig. 7.** Respiratory epithelial clefts separating the hamartoma tissue into lobules composed of mixed mesenchymal tissue (H&E stain, 100X)

## Discussion

Hamartomas of the respiratory tract are mainly found in the lung parenchyma, and are usually clinically silent. Small lesions do not require invasive treatment, and instead should be monitored for growth. When large, they may compress the adjacent lung tissue and bronchi causing obstruction. If the tumor grows inside the lumen of trachea or bronchi, the patient's complaints may mimic respiratory diseases such as bronchial asthma, chronic obstructive pulmonary disease, or recurrent pneumonia.[6-8] If they are small in size, the lesions may not be visible on a standard chest X-ray.

In the presented case the baseline chest X-ray did not describe pathology. The current thorough chest X-ray analysis indicated a discrete shading in the tracheal projection. Audible wheezing during auscultation of the patient lungs should take into account the possibility of central or upper airway obstruction. Entering

the above information on the referral to the radiologist has a significant impact on a more accurate assessment of the trachea in X-ray.

It may be helpful to mark the indicators: $FEV_1$/PEF (obstruction >0.48) or $FEV_1$/PEF ml/l/min (obstruction >8 ml/l/min), which suggest narrowing of the central or upper respiratory tract such as the main bronchi, trachea or larynx.[8] Chronic complaints such as cough, breathlessness, wheezing, stridor, hemoptysis, or recurrent pneumonia are a lead based on which the diagnosis should be expanded to include CT scan and/or bronchoscopy, which can detect even smaller lesions.[9]

In the presented case, the symptoms persisted for 10 months despite anti-asthmatic treatment. It is considered that the occurrence of symptoms for more than 2 months with optimal treatment of bronchial asthma should require an extension of the diagnosis, including, above all, computed tomography.

The prognosis of such finding is favorable, and usually involves endoscopic removal of the lesion. Resection of a benign lesion from the lung parenchyma or from the lumen of the airways is always advantageous, as it excludes the possibility for the lesion to become malignant. Patients with an unspecified lesion in the lung parenchyma or airways, who cannot be treated surgically, should have the lesion evaluated using positron emission tomography scan and a computed tomography for potential malignancy.[10]

Apart from causing an obstruction within the respiratory tract itself, hamartomas sprouting from the lung tissue can potentially cause issues in different organ systems, such as cardiovascular where the tumor can compress arteries or put pressure on the heart itself.[11] This case study is a good example of how important it is to not ignore recurrent clinical findings or chronic illnesses as they can have curable underlying causes.

## Conclusion

In the presented case there was a significant narrowing of the tracheal lumen. Inappropriate or delayed diagnosis could have led to serious complications and even death of the patient. Initially diagnosed with bronchial asthma, the patient was taking bronchodilators and inhaled steroids for several months.

Lack of improvement after the previous treatment, characteristic auscultatory changes in the form of inspiratory and expiratory wheezing and obstruction of the airways, especially the upper ones, require a revision of the current diagnosis. The use of the $FEV_1$/PEF index may indicate central or upper airway obstruction. Computed tomography allows proper assessment of the lungs and mediastinum. Invisible or poorly visible changes in chest X-ray should also be confirmed by it, as shown by the presented case.

## Declarations

***Funding***

This report received no funding.

***Author contributions***

Conceptualization, T.K. and M.R-G.; Methodology, S.T.; Software, T.K and E.K.; Validation, T.K., E.K. and S.T.; Formal Analysis, T.K. and S.T.; Investigation, T.K.; Resources, T.K. and E.K.; Data Curation, T.K.; Writing – Original Draft Preparation, T.K. and E.K.; Writing – Review & Editing, T.K. and E.K; Visualization, T.K.; Supervision, S.T. and M.R-G.

***Conflicts of interest***

The authors have no conflict of interest.

***Data availability***

Not applicable.

***Ethics approval***

The patient signed informed consent regarding publishing his data.

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