Concurrent tracheal bronchus, tracheal stenosis, and pulmonary sequestration: A case report and mini-review of the literature

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Received June 17, 2025; Accepted September 8, 2025

DOI: 10.3892/wasj.2025.397

Abstract. Tracheal bronchus (TB) is an abnormal bronchus arising directly from the tracheal wall above the carina, supplying the upper lobes. This anomaly is occasionally associated with congenital tracheal stenosis (TS), with a reported incidence of 11.9-24.7% in affected patients. The simultaneous occurrence of TB, congenital TS and pulmonary sequestration (PS) in a single patient is extremely rare. The present study describes the case of a 4-year-old boy who presented with shortness of breath and stridor. He had a lifelong history of recurrent episodes, initially misdiagnosed as asthma. A clinical examination revealed tachypnea, audible stridor and fever (37.9˚C), with an elevated white blood cell count and C-reactive protein levels. A chest computed tomography scan demonstrated TB with severe TS, and the TB was supplying an additional lobar sequestration in the right lung. The patient was treated with oxygen, intravenous fluids, antibiotics and low-dose steroids; however, he was lost to follow-up before definitive surgical management. In addition, a literature review of articles from the PubMed and Google Scholar databases identified no previous reports of concurrent TB, congenital TS and PS in a single patient. However, six studies described 41 cases of TB associated with either congenital TS or PS. In summary, the coexistence of TB, congenital TS and PS is exceptionally rare, and may easily be misdiagnosed as common respiratory conditions, such as asthma. It is essential to consider such rare congenital anomalies in children presenting with persistent stridor or unexplained respiratory symptoms.

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Key words: tracheal bronchus, congenital tracheal stenosis, pulmonary sequestration, asthma, stridor, congenital anomaly

Introduction

The anatomy of the tracheobronchial tree has been thoroughly investigated over the past hundred years using anatomical dissections and bronchography. Various congenital abnormalities can affect the bronchi, lungs, and their vascular supply. These anomalies in bronchial anatomy can include atypical origins, missing branches, extra branches, and congenital diverticula (1). In contrast to the numerous variations found in lobar or segmental bronchial subdivisions, it is rare for abnormal bronchi to originate from the trachea or main bronchi. Significant bronchial abnormalities include the accessory cardiac and tracheal bronchus (TB) (2). TB is an abnormal bronchus that emerges directly from the lateral wall of the trachea above the carina, providing air to the upper lobes. First described by Sandifort in 1785, it is typically an asymptomatic anatomical variation that is often discovered incidentally during a chest computed tomography (CT) scan or bronchoscopy (3). The average incidence rate of TB is 0.1-2% in the general population. It is typically found on the right side of the trachea and is more common among males (4). Patients with congenital tracheal stenosis (TS) sometimes exhibit this abnormality, with the occurrence of TB in these patients varying from 11.9 to 24.7% (5). The congenital TS is a markedly rare malformation characterized by complete annular tracheal rings, which is in contrast to the typical C-shaped tracheal cartilages. It is estimated to affect ~1 in 64,500 live births, accounting for merely 0.3-1% of all laryngotracheal stenosis subtypes (6). However, accurately determining the incidence of congenital TS poses challenges, as numerous patients remain undiagnosed due to diagnostic uncertainties among practitioners and high mortality rates before definitive diagnosis (7). Another type of congenital pulmonary anomaly is pulmonary sequestration (PS), typically identified during childhood or adolescence. These non-functional masses of pulmonary tissue lack connection to the tracheobronchial tree

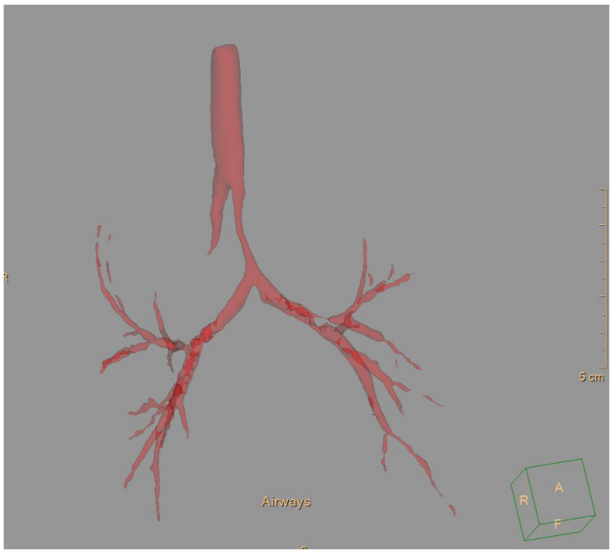

Figure 1. Computed tomography scan of the chest with 3D reconstruction of the tracheobronchial tree illustrating tracheal bronchus and tracheal stenosis starting just below the tracheal bronchus, extending to the carina.

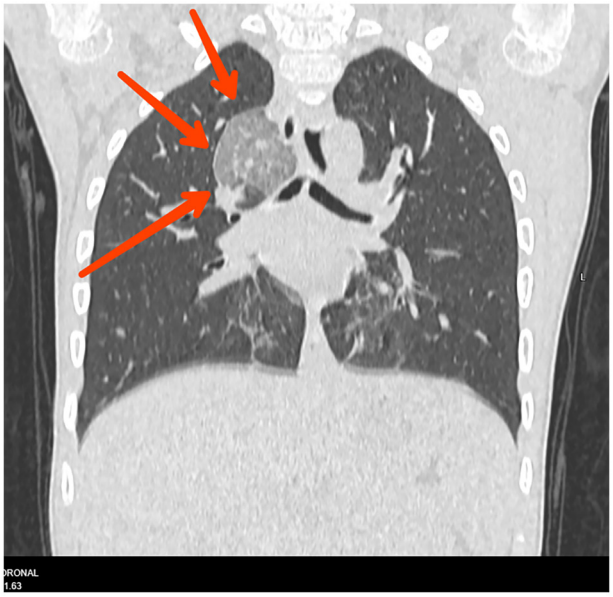

Figure 2. Computed tomography scan of the chest (coronal section) with intravenous contrast illustrating a round enhancing lesion (red arrows) on the right side of the chest above the hilum. Examining multiple sections revealed an arterial supply from the right subclavian artery and venous drainage to the superior vena cava, suggesting sequestration.

and are nourished by an abnormal systemic artery. Similar to tracheal bronchi, they commonly present no symptoms and are usually discovered incidentally (4,8). The PS is rare, accounting for ~1 to 6% of all congenital lung anomalies. It can often remain undetected during prenatal and early childhood (9). The present study describes an extremely rare case of simultaneous TB, congenital TS and PS in a single patient. It is organized according to the CaReL guidelines (10). All studies referenced herein were evaluated for eligibility (11).

Case report

Patient information. A 4-year-old boy presented to the Emergency Room at Smart Health Tower (Sulaymaniyah, Iraq) with shortness of breath and stridor. The patient had a history of frequent episodes of the same issue since birth and had been misdiagnosed with asthma. The management plan included inhaled corticosteroids as the primary treatment and short-acting beta-agonists, particularly during asthma attacks or severe symptoms.

Clinical findings. A clinical examination revealed tachypnea (respiratory rate, 30 cycles/min) and audible stridor, with a temperature of 37.9˚C. Hematological investigations revealed elevated white blood cell counts (13,000 cells/mm^3) and C-reactive protein levels (15 mg/dl).

Diagnostic approach. A subsequent chest CT scan revealed a TB and severe TS beginning just below the TB and extending to the carina. Notably, the TB was supplying additional lobar sequestration on the right side of the lung (Figs. 1-3).

Therapeutic interventions. The patient received resuscitation with oxygen therapy, intravenous (IV) fluids, IV antibiotics and a low dose of steroid (4 mg dexamethasone x2). Surgery was planned.

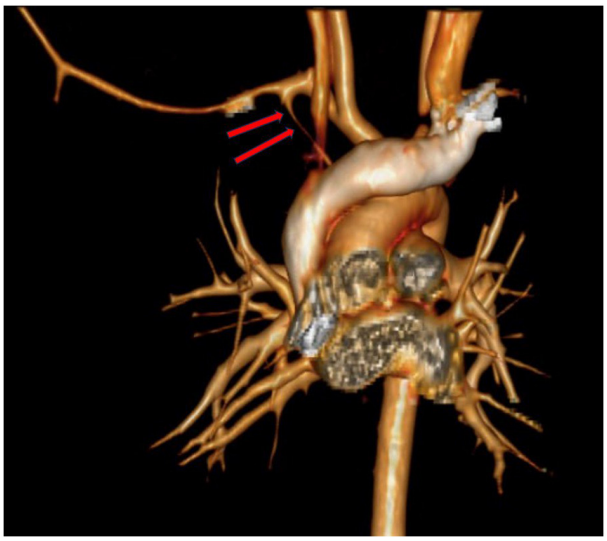

Figure 3. Computed tomography angiography of the chest illustrating an anomalous blood supply from the right subclavian artery to the pulmonary sequestration (red arrows).

Follow-up and outcomes. The patient was lost to follow-up, and as a result, the surgery was not performed (Table I).

Discussion

The typical development of the tracheobronchial tree in humans commences between the 24th and 26th days of gestation. It begins as a central protrusion on the front wall of the pharynx, forming at the lower end of the laryngotracheal groove. (3) Between the 28th and 30th weeks of gestation, the lung buds extend to form the primary bronchi. By the 36th day of gestation, all segmental bronchi are formed. TB is an

Table I. Chronological summary of clinical events.

Time point	Clinical event
Since birth	Recurrent respiratory distress and stridor.
Age 4 years	Emergency visit with acute respiratory symptoms.
Upon presentation	Chest CT scan performed, revealing TB, TS and intralobar PS.
Shortly afterward (on the same day of presentation)	Medical management was initiated; symptoms improved.
Post-discharge	The patient was lost to follow-up.

CT, computed tomography; TB, tracheal bronchus; TS, tracheal stenosis; PS, pulmonary sequestration.

abnormal anatomical development characterized by various bronchial anomalies originating from the trachea or main bronchi and directed toward the upper lung lobes (3).

Alescio *et al* (12) linked TB formation to an embryogenesis defect rather than a genetic abnormality. Their study demonstrated that transplanting bronchial mesenchyme into the tracheal epithelium led to TB (12).

Typically, TB manifests as a bronchus leading to the right upper lobe, although instances of left tracheal bronchi have been infrequently reported (13). Studies utilizing bronchography and bronchoscopy have reported a prevalence ranging from 0.1 to 2% for right TB, and 0.3 to 1% for left TB (2). The study by Dave *et al* (14) analyzed 1,021 rigid endoscopies of the trachea performed on children aged 0 to 6 years. The TB was identified in 11 cases. Of note, all detected TB cases originated from the right lateral wall of the trachea (14). In another study by Findik (1), an average of 12,648 adult patients underwent bronchoscopy in their clinic over a period of 10 years. Among these, 8 patients were diagnosed with right-sided TB (1). Accordingly, the present study found the TB on the right side of the trachea.

Of note, two primary types of TB have been identified: Supernumerary and displaced tracheal bronchi. The supernumerary type, considered relatively uncommon, is an accessory bronchus. Conversely, a displaced bronchus originates from an abnormal position and supplies one or more segments of the upper lobe, typically the apical segment (15). The type of TB in the patient in the present study was supernumerary.

The TB typically presents with either no or minimal clinical symptoms. However, recent case reports have highlighted associations with persistent atelectasis, hemoptysis, recurrent pulmonary infection, bronchiectasis necessitating surgery of the affected segment and instances of lung cancer (3,16,17). Additionally, patients may exhibit symptoms such as persistent cough, stridor and acute respiratory distress. These symptoms often arise from retained secretions in the narrow bronchus, with superadded infections as a contributing factor (3). The case presented herein suffered from shortness of breath and stridor from birth.

TB should be considered in patients with recurrent chest infections, persistent or chronic bronchitis and a persistent

Table II. Cases of tracheal bronchus associated with either congenital tracheal stenosis or pulmonary sequestration reported in the literature.

Authors, year or publication	Study design	No. of cases	Age	Sex (M/F)	Diagnostic method	Co-occurrence	Management	(Refs.)
Arslan *et al*, 2013	Case report	1	53 years	1/0	Bronchoscopy and CT scan	TB, PS and azygos lobe	Conservative treatment	(4)
Morita *et al*, 2016	Case series	13	4 months[a]	8/5	Bronchoscopy	TB and congenital TS	Surgery	(5)
Loh *et al*, 2015	Case report	1	66 years	0/1	CT scan and 3-D image	TB and congenital TS	N/A	(21)
Wang *et al*, 2015	Case series	24	20.6 months[a]	10/14	CT scan	TB, congenital TS and CHD	Surgery	(22)
Wong *et al*, 1998	Case report	1	2 years	1/0	Bronchoscopy	TB and congenital TS	Conservative treatment	(23)
Yamoto *et al*, 2020	Case report	1	37 weeks	1/0	CT scan	Bilateral TB and congenital TS	Surgery	(24)

[a]This value is presented as the mean. M, male; F, female; TB, tracheal bronchus; TS, tracheal stenosis; PS, pulmonary sequestration; CHD, congenital heart disease; N/A, not available.

cough. A chest X-ray may reveal an ectopic TB originating above the carina, although this is not always the case, and additional investigations are necessary to confirm the diagnosis (16). Bronchoscopy is considered an invasive method for diagnosing TB, as it can reveal the opening of the TB and other anomalies in the tracheobronchial tree. Non-invasive diagnostic methods include a chest CT scan, the preferred imaging modality for identifying this anomaly, as it can accurately delineate the tracheal and bronchial anatomy (17,18). In the case in the present study, a CT scan of the chest with 3D reconstruction of the tracheobronchial tree revealed the presence of TB and TS. The stenosis began just below the TB and extended down to the carina. However, a CT scan of the chest with intravenous contrast revealed sequestration on the right side of the chest, located above the hilum.

TB may be linked with various congenital anomalies, such as tracheal hypoplasia, TS, lobar emphysema, pulmonary cystic lesions, cardiovascular disorders (particularly cyanotic cardiac lesions), other bronchial tree abnormalities and Down syndrome (19,20). Arslan *et al* (4) reported TB with PS and an azygos lobe in a single case. Moreover, Morita *et al* (5) documented that out of 51 pediatric patients with congenital TS who underwent tracheal reconstruction at a single institution between January, 2006 and December, 2015, 14 patients had congenital TS associated with TB. In the present case report, the patient was unexpectedly diagnosed with TB associated with congenital TS and PS. To date, only six studies (4,5,21-24), including 41 cases of TB associated with congenital TS or PS, have been reported (Table II). To the best of our knowledge, the coexistence of these three anomalies in a single case has not yet been reported in the literature.

The PS occurs in the population at a rate of 0.15-1.7% and is more common in males than females. Typically, it is located in the inferior, posterior, and medial segments of the left half of the thoracic cavity. In 74% of cases, it is supplied by aberrant arteries originating from the thoracic aorta, and in 18.7% of cases, from the abdominal aorta (4). In the present case report, the PS was located on the right side of the chest of a 4-year-old male child above the hilum. The arterial supply came from the right subclavian artery, and the venous drainage was to the superior vena cava.

The co-occurrence of congenital TS with TB ranges from 11.9 to 24.7% of all congenital TS cases (5). The stenotic trachea is often located near the right upper lobe bronchus (RULB), and preserving the RULB complicates tracheal reconstruction for congenital TS when TB is involved.

The optimal surgical approaches for congenital TS with TB continues to be a matter of debate. This controversy primarily arises from the diverse forms of congenital TS associated with TB that have been documented (5). Congenital TS used to be associated with mortality rates as high as 50%. However, various surgical techniques have been developed to treat congenital TS, resulting in more acceptable operative mortality rates (7-13%). Slide tracheoplasty has been celebrated as a breakthrough in managing congenital TS, achieving significantly improved outcomes. For long-segment lesions, slide tracheoplasty has mostly replaced tracheal patching with costal cartilage or pericardium due to the high re-intervention rates associated with those methods. Conversely, end-to-end anastomoses are typically used for patients with short-segment congenital TS. Despite these advances, surgically managing TS with abnormal bronchial branching or involvement remains challenging (24). TB alone can often be observed when asymptomatic, whereas symptomatic cases with recurrent pneumonia typically necessitate resection of the affected lung segment (segmentectomy or lobectomy) (25). PS is definitely managed through surgical removal of the sequestered lobe or segment. Minimally invasive techniques, including video-assisted or robotic approaches, have been reported for PS, and uniportal segmentectomy can effectively preserve normal lung parenchyma during resection (26,27). In complex scenarios involving all three anomalies, a multidisciplinary approach is essential. For instance, airway reconstruction (slide tracheoplasty/bronchoplasty) may be performed in stages or combined with pulmonary resection to alleviate stenosis and excise the sequestration (4-6,8)

The case described in the present study illustrates the diagnostic challenges posed by rare tracheobronchial anomalies, particularly when they mimic more common conditions such as asthma. Early recognition through advanced imaging and prompt referral to specialized surgical centers are essential. The present case report emphasizes the importance of heightened clinical awareness to prevent misdiagnosis and delays in treatment. A limitation of the present case report is that the patient was lost to follow-up and was therefore unable to undergo further treatment.

In conclusion, the coexistence of TB, congenital TS and PS is exceptionally rare and may easily be misdiagnosed as common respiratory conditions such as asthma. It is essential to consider such rare congenital anomalies in children presenting with persistent stridor or unexplained respiratory symptoms.

Acknowledgements

Not applicable.

Funding

No funding was received.

Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

FHK and HKA were major contributors to the conception of the study, as well as to the literature search for related studies. SMA, KMH, HOK and BAM contributed to the clinical management of the patient, assisted in data acquisition and interpretation, and participated in the literature review and manuscript preparation. HAN and SHM contributed to the conception and design of the study, the literature review, the critical revision of the manuscript, and the processing of the table. SHT and RJR were the radiologists who performed the assessment of the case. FHK and HKA assisted in diagnosing the patient, contributed to the management of the patient, and participated in manuscript review. FHK and HAN confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was obtained from the patient's parent for this participation in the present study.

Patient consent for publication

Written informed consent was obtained from the patient's parent for the publication of this case report and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

References

1. Findik S: Tracheal bronchus in the adult population. J Bronchology Interv Pulmonol 18: 149-153, 2011.
2. Ghaye B, Szapiro D, Fanchamps JM and Dondelinger RF: Congenital bronchial abnormalities revisited. Radiographics 21: 105-119, 2001
3. Eddine HJ and Arab WA: Tracheal bronchus: Case report and literature review. SN Comprehensive Clinical Medicine 3: 399-403, 2021.
4. Arslan S, Sabancioğullari V and Akkurt İ: Concomitant occurrence of tracheal bronchus, pulmonary sequestration and azygos lobe. Turk Gogus Kalp Damar Cerrahisi Derg 21(2): 506-509, 2013.
5. Morita K, Yokoi A, Fukuzawa H, Hisamatsu C, Endo K, Okata Y, Tamaki A, Mishima Y, Oshima Y and Maeda K: Surgical intervention strategies for congenital tracheal stenosis associated with a tracheal bronchus based on the location of stenosis. Pediatr Surg Int 32: 915-919, 2016
6. Herrera P, Caldarone C, Forte V, Campisi P, Holtby H, Chait P, Chiu P, Cox P, Yoo SJ, Manson D and Kim PC: The current state of congenital tracheal stenosis. Pediatr Surg Int 23: 1033-1044, 2007.
7. Pellen G, Pandit C, Castro C, Robinson P, Seton C, Fitzgerald DA, Waters K and Cheng AT: Use of non-invasive ventilation in children with congenital tracheal stenosis. Int J Pediatr Otorhinolaryngol 127: 109672, 2019.
8. Alsumrain M and Ryu JH: Pulmonary sequestration in adults: A retrospective review of resected and unresected cases. BMC Pulm Med 18: 1-5, 2018
9. Walker CM, Wu CC, Gilman MD, Godwin JD II, Shepard JA and Abbott GF: The imaging spectrum of bronchopulmonary sequestration. Curr Probl Diagn Radiol 43: 100-114, 2014.
10. Prasad S, Nassar M, Azzam AY, José FG, Jamee M, Sliman RKA, Evola G, Mustafa AM, Abdullah HO, Abdalla BA, *et al*: CaReL guidelines: A consensus-based guidelines on case reports and literature review (CaReL). Barw Med J 2: 13-19, 2024.
11. Abdullah HO, Abdalla BA, Kakamad FH, Ahmed JO, Baba HO, Hassan MN, Bapir R, Rahim HM, Omar DA, Kakamad SH, *et al*: Predatory publishing lists: A review on the ongoing battle against fraudulent actions. Barw Med J 2: 26-30, 2024.
12. Alescio T and Cassini A: Induction in vitro of tracheal buds by pulmonary mesenchyme grafted on tracheal epithelium. J Exp Zool 150: 83-94, 1962.
13. O'Sullivan BP, Frassica JJ and Rayder SM: Tracheal bronchus: A cause of prolonged atelectasis in intubated children. Chest 113: 537-540, 1998.
14. Dave MH, Gerber A, Bailey M, Gysin C, Hoeve H, Hammer J, Nicolai T and Weiss M: The prevalence of tracheal bronchus in pediatric patients undergoing rigid bronchoscopy. J Bronchology Interv Pulmonol 21: 26-31, 2014.
15. Siegel MJ, Shackelford GD, Francis RS and McAlister WH: Tracheal bronchus. Radiology 130: 353-355, 1979.
16. Ikeno S, Mitsuhata H, Saito K, Hirabayashi Y, Akazawa S, Kasuda H and Shimizu R: Airway management for patients with a tracheal bronchus. Br J Anaesth 76: 573-575, 1996.
17. Ruchonnet-Metrailler I, Taam RA and de Blic J: Presence of tracheal bronchus in children undergoing flexible bronchoscopy. Respir Med 109: 846-850, 2015.
18. Manjunatha YC, Gupta AK and Gupta SK: Subacute combined degeneration of the spinal cord in a child. Indian J Pediatr 78: 240-241, 2011.
19. Aoun NY, Velez E, Kenney LA and Trayner EE: Tracheal bronchus. Respir Care 49: 1056-1058, 2004.
20. Reis JC, Martins CR, Otta LH, Tardelli MA and Amaral JL: Complication related to tracheal bronchus in infant: Case report. Rev Bras Anestesiol 56: 72-77, 2006 (In Portuguese).
21. Loh PS, Hashim SA, Aman RR, Lai LL and Chan L: A challenging case of tracheal bronchus with concurrent tracheal stenosis for one lung ventilation. Trends in Anaesthesia and Critical Care 5: 136-139, 2015.
22. Wang S, Zhang H, Zhu L, Zhen J, Liu J and Xu Z: Surgical management of congenital tracheal stenosis associated with tracheal bronchus and congenital heart disease. Eur J Cardiothorac Surg 49: 1201-1206, 2016.
23. Wong KS, Wang CR and Hsieh KH: Demonstration of tracheal bronchus associated with tracheal stenosis using direct coronal computed tomography. Pediatr Pulmonol 25: 133-135, 1998.
24. Yamoto M, Fukumoto K and Urushihara N: Tracheoplasty for congenital tracheal stenosis with bilateral tracheal bronchus. Ann Thorac Cardiovasc Surg 28: 159-162, 2022.
25. Gullapalli D, Tangutoori S and Ganti SS: Respiratory maze: An anatomical variant of tracheal bronchus. Respirol Case Rep 12: e01355, 2024.
26. Lin TH, Huang WL, Chang CC, Yen YT, Lai WW, Tseng YL and Chen YY: Uniportal video-assisted thoracoscopic surgery lobectomy and segmentectomy for pulmonary sequestration. J Thorac Dis 10: 3722, 2018.
27. Mezzetti M, Dell'Agnola CA, Bedoni M, Cappelli R, Fumagalli F and Panigalli T: Video-assisted thoracoscopic resection of pulmonary sequestration in an infant. Ann Thorac Surg 61: 1836-1838, 1996.