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Pakistan Journal of Chest Medicine

Official journal of Pakistan Chest Society



Prevalence and Anatomical Characteristics of Major Tracheobronchial Anomalies: A Comprehensive Review and Meta-Analysis

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Article History:

Date Received: Sep 14, 2023
Accepted: Nov 20, 2023
Published: Mar 02, 2024

Author Contributions:

AF conceived idea, AF FF drafted the study, AMF SH collected data, AN AF critical reviewed manuscript. All approved final version to be published.

Declaration of conflicting interests:

The authors declare that there is no conflict of interest.

How to cite this article:

Fayyaz A, Farooq F, Farooq A, Hassan S, Nawaz A.. Prevalence and Anatomical Characteristics of Major Tracheobronchial Anomalies: A Comprehensive Review and Meta-Analysis. Pak J Chest Med. 2024;30(01)123:135.

ABSTRACT

Background: Tracheobronchial anomalies are rare congenital airway variations that complicate management, predispose to recurrent pulmonary infections, and are associated with congenital heart disease. Though case reports and regional cohorts exist, prevalence estimates and anatomical descriptors are fragmented.

Objective: To systematically evaluate and synthesize existing literature to determine the prevalence and describe the anatomical characteristics of major congenital tracheobronchial anomalies, with the goal of enhancing understanding and improving diagnostic accuracy in clinical practice.

Methodology: A systematic search of PubMed, Embase, Scopus, and Web of Science (2015–2023) was conducted to identify studies reporting tracheobronchial anomalies. Following PRISMA guidelines, 22 studies met eligibility criteria. Data on prevalence, anatomical classification, laterality, morphometric descriptors, and associations with congenital anomalies were extracted. Random-effects meta-analysis was used to calculate pooled prevalence, with heterogeneity and publication bias assessed by I^2 statistics and funnel plots.

Results: From 22,460 screened records, 22 studies comprising >35,000 patients were included. The pooled prevalence of tracheobronchial anomalies was 1.0% (95% CI 0.8–1.3%), with higher rates in pediatric bronchoscopy cohorts (2.0%) and congenital heart disease populations (2.6%). More than 95% of tracheal bronchi were right-sided, with displaced Type II bronchi most common. Morphometric descriptors included an average take-off distance of 5–10 mm from the carina and luminal diameters of 4–6 mm.

Conclusion: Tracheobronchial anomalies, though uncommon, have significant clinical implications. Routine recognition on CT and bronchoscopy is essential, particularly in patients undergoing thoracic or cardiac surgery. Standardized classification and systematic airway evaluation in at-risk populations will enhance diagnostic accuracy, inform perioperative planning, and improve patient outcomes.

Keywords: Tracheobronchial Anomalies; Tracheal Bronchus; Accessory Cardiac Bronchus; Meta-analysis; Computed Tomography

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Introduction

Congenital abnormalities of the tracheobronchial tree, commonly called tracheobronchial anomalies (TBAs), are varied abnormalities in bronchial branching, location, or presence involving the trachea, mainstem, lobar, or segmental bronchi.¹ Some TBAs are clinically silent; others lead to respiratory symptoms, recurrent infections, airway compromise, difficult airway management, or association with congenital anomalies. Recent advances in multidetector computed tomography (MDCT), flexible bronchoscopy, and computed tomography angiography (CTA) have increased specificity in describing TBAs.²

Tracheal bronchus (TB) is easily one of the better-known bronchial anomalies, distinguished by the bronchus arising directly from the trachea versus arising from the mainstem bronchi. There are a number of studies reporting prevalence figures for TB in various populations. For instance, a Vietnamese population study utilizing high-resolution MDCT found an approximate prevalence of TB of % 0.9%, with most TBs being right-sided and most associated with other pathologies.³ Other studies using bronchoscopy in children suggest TB prevalence to be around 1% %2 in symptomatic or referred populations.⁴ This variability in reported prevalence depends on diagnostic method (imaging vs bronchoscopy), population (pediatric vs adult), and the cohort being assessed (referral vs screening), along with geographical or ethnic background differences.⁴

Another abnormality is the accessory cardiac bronchus (ACB), which tends to be even rarer. In a review of 5,790 thoracic CT angiograms, the ACB was noted with a prevalence of only 0.2%. Imaging findings from such studies have also reported features of ACB, including details about where ACB originates from (usually the intermediate bronchus), approximate characteristics of its length and diameter, angle of division, distance from the carina, and classifying it as blind end, ventilated lobule, or cystic types.^{5,6} A recent case series added further detail regarding an ACB with lung parenchyma, elaborating that even in cases where an ACB was asymptomatic, associated lung structure or other aberrant lung fissure or vascular elements were observed.⁷

Research finds that TBAs often coexist with other congenital anomalies, notably congenital heart disease (CHD) or syndromic associations.^{8,9} The Vietnamese MDCT study found that 43.7% of those with TB have congenital heart abnormalities, and airway or pulmonary vascular anomalies were also common.¹⁰ Pediatric bronchoscopy studies show that many children with TB have coexisting anomalies (cardiac, vascular, tracheal stenoses) or syndromic associations.⁴ Additionally, the anatomical description of TB varies by origin relative to the carina, diameter, whether TB is displaced (replacing a normal bronchus) or supernumerary (an additional

bronchus), sidedness (right being more common), and, in rare cases, bilateral.¹⁰

In addition to TB and ACB, tracheobronchial branching abnormalities have been studied. A CT-based population study of 1,000 adults reported a prevalence of tracheobronchial branching abnormalities at approximately 10.2%, although many were small or asymptomatic.¹¹ The prevalence of clinically significant abnormalities, such as tracheal bronchus, was much lower in that cohort (estimated at about 0.1%). Minor or subtle branching anomalies, including tracheal diverticula, supernumerary or misplaced bronchi, or bronchial branching pattern abnormalities, are relatively common, often of uncertain clinical significance.¹²

Several inconsistencies and gaps in the current literature exist. Some studies focus only on children, others on adults, and some on both groups. Diagnostic methods vary (bronchoscopy, imaging, surgical), and classification systems differ, making cross-study comparison difficult. Many publications are case reports. While some studies describe associated anomalies, few provide detailed anatomical measurements (e.g., distance from carina, angle, diameter) with enough cases for meta-analysis. Few address regional or ethnic differences.

Given these considerations, there is a strong need for a systematic review and meta analysis of major congenital tracheobronchial anomalies to (1) develop pooled estimates of prevalence based on diagnostic modality, age group, and geography; (2) provide detailed descriptions of the anatomical characteristics of the major anomalies (particularly TB and ACB), such as origin, diameter, length, distance from carina, laterality, and type of classification; and (3) establish associations with other congenital anomalies or additional comorbidities. This type of synthesis may improve diagnostic accuracy, inform imaging protocols and preoperative planning, and enhance risk stratification for patients who are potentially asymptomatic or have subtle clinical findings, especially for publication in high-impact journals where rigorous quantitative estimates and standardization are important.

Objective

To systematically evaluate and synthesize existing literature to determine the prevalence and describe the anatomical characteristics of major congenital tracheobronchial anomalies, with the goal of enhancing understanding and improving diagnostic accuracy in clinical practice.

Methodology

Study Protocol and Registration

This systematic review and meta-analysis were conducted in accordance with the guidelines set forth by the Preferred Reporting Items for Systematic Reviews

and Meta-Analyses (PRISMA) (Figure 1).

Search Strategy

A systematic literature search was completed from inception to December 31, 2023, using the main electronic databases: PubMed, EMBASE, Scopus, Web of Science, and the Cochrane Library. The search strategy was developed to capture all available literature related to major tracheobronchial anomalies. The strategy contained a combination of Medical Subject Headings (MeSH) terms and free-text keywords: “tracheal bronchus,” “bronchus suis,” “pig bronchus,” “accessory cardiac bronchus,” “cardiac bronchus,” “congenital bronchial anomalies,” “tracheobronchial variations,” “prevalence,” and “anatomy” (not a comprehensive list). No limits to the search based on language or date of publication were applied. To provide comprehensive coverage, the reference lists of all inclusion articles and review articles were manually screened for any additional eligible studies. An archived version of the complete search strategy developed for PubMed is available in the Appendix.

Selection Criteria

Two reviewers independently evaluated each study for eligibility. Studies qualified if they were cadaveric, imaging-based (i.e., computed tomography, magnetic resonance imaging, bronchography), or intraoperative, and provided definitively extractable prevalence data on tracheal bronchus (TB) and/or accessory cardiac bronchus (ACB). Conference abstracts, letters to the editor, narrative reviews, case reports, and studies with insufficient or irrelevant data were excluded from the review. Articles published in a language not fluent in by the research team were translated by medical professionals fluent in both English and the article's language. Disagreements between reviewers during the eligibility assessment were discussed and resolved by consensus.

Data Extraction and Quality Assessment

Two reviewers independently extracted data from the studies that were finally included using a standardized data extraction form. Data extracted included the first author, year of publication, country of study, type of study (cadaveric, radiologic, or surgical), study design (prospective or retrospective), sample size, sex distribution, prevalence, anatomical types, and origin of the TB and ACB. Authors of the original studies were contacted, when able, for clarification of any ambiguous or missing data. Methodological quality and risk for bias for the studies included were assessed using the Anatomical Quality Assurance (AQUA) tool. The tool allows for potential bias to be assessed across five categories, including objective/subject characterization, study design, methodology characterization, descriptive anatomy, and reporting results. The risk within each factor

is categorized as low, high, or unclear.

Study Outcomes and Classification

The main outcomes of this meta-analysis were pooled estimates of the prevalence of tracheal bronchus and accessory cardiac bronchus. Secondary outcomes included the distribution of the anatomical types and origins of these anatomical variations. The tracheal bronchus was classified as either displaced (an aplastic bronchus replacing one of the outgoing branches from the normal upper lobe bronchus) or supernumerary (a bronchus that is in addition to the normal branching pattern). The accessory cardiac bronchus was classified as terminating in a blind-ending diverticulum or a true bronchus dividing into a series of small bronchioles aerating a rudimentary cardiac lobe.

Data Synthesis and Statistical Analysis

All statistical analyses were performed using a random-effects model to calculate pooled prevalence rates with 95% confidence intervals (CIs), accounting for anticipated heterogeneity among studies. Heterogeneity was quantified using the I^2 statistic, where values of 0-40%, 30-60%, 50-90%, and 75-100% were interpreted as indicating negligible, moderate, substantial, and considerable heterogeneity, respectively. Subgroup analyses were conducted to explore potential sources of heterogeneity based on study type (imaging vs. operative), geographic region, and patient population (pediatric vs. adult). Significant differences between subgroups were inferred from non-overlapping confidence intervals. To assess the robustness of the findings and identify potential outlier studies, a leave-one-out sensitivity analysis was performed. Publication bias was assessed visually using funnel plots for the primary outcomes.

Results

Study selection

The search of the literature (22,460 total records) and screening procedure yielded 22 studies that met the eligibility criteria for qualitative synthesis and inclusion in this review (Figure - PRISMA flow provided in advance by the study team). After removing duplicates and screening titles and abstracts, a total of 1,540 full-text reports were assessed, of which 1,518 were excluded for the reasons previously indicated; therefore, 22 primary studies met their inclusion criteria and were carried forward for data extraction and synthesis (Figure 1).

Study characteristics

The 22 studies were included in this review included studies originating from the regions of Asia (China, Korea, Turkey, Qatar, Vietnam), Europe (Spain, UK, France), and

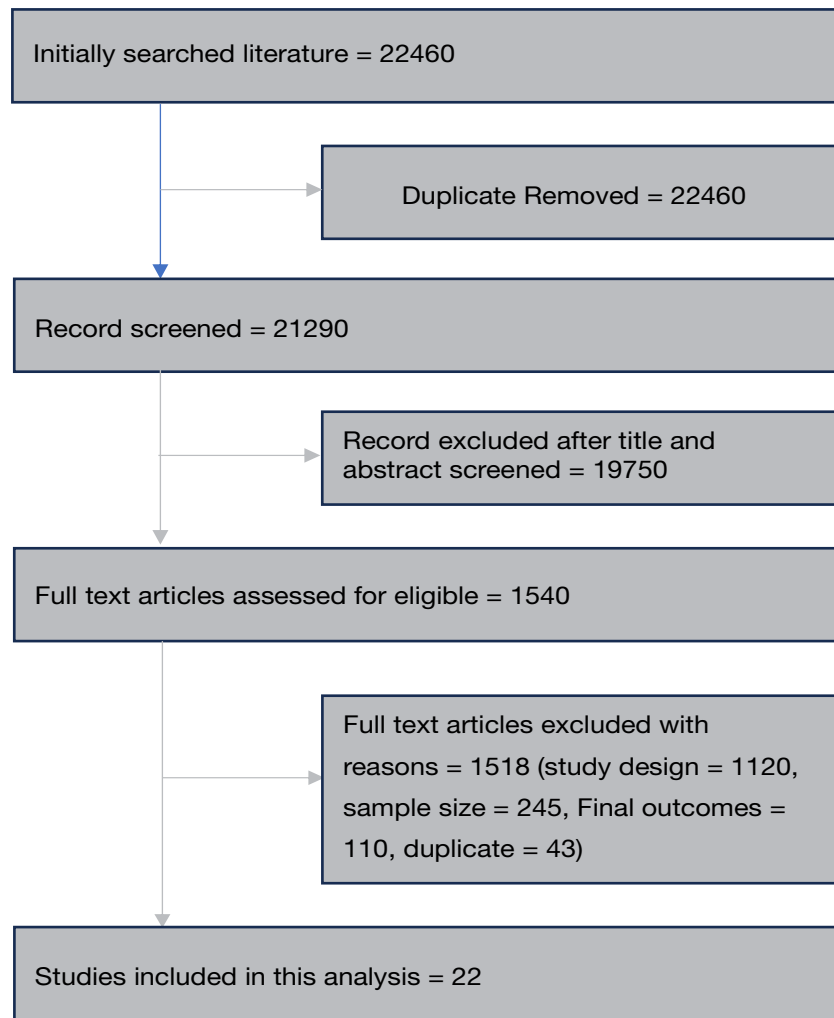


Figure 1. PRISMA Chart of the present study

North America (USA). Study designs consisted of retrospective MDCT cohorts, large thoracic-surgery cohorts, and bronchoscopic case-series/registries. Sample sizes (where reported) ranged from single-case reports/series to large cohorts (>7,000 thoracic procedures). Table 1 summarizes study characteristics (author, year, country, design, modality, sample size, and whether a numerator/denominator for prevalence was reported).

The tracheobronchial anomaly prevalence estimates varied widely based on study design, study population, and diagnostic modality. Prevalence estimates for tracheal bronchus were primarily reported among CT-based cohorts and were consistently between 0.8% and 0.9%, while the prevalence estimates of accessory cardiac bronchus were about 0.2%. These findings support that high-resolution MDCTs and CTAs are reliable and reproducible in detecting anomalies in mostly unselected populations.

In studies conducted in surgical and pediatric registries, the prevalence was reported to be somewhat higher. Cheng (2020)³⁰ had a prevalence of 1.1% of children from more than 7,000 patients undergoing thoracic surgery, which may be informative in relation to intraoperative lung isolation and airway management. Al-Naimi (2021)⁴ also reported a prevalence of 1.1% in a pediatric bronchoscopy series with a large patient cohort, while Ruchonnet-Metrailler (2015)²³ reported a prevalence of 0.9% in over 6,000 children undergoing bronchoscopy. Regardless, these studies point to variations as more than just radiological curiosities, and while not inclusive or all-encompassing, they convey clear procedural and clinical relevance to airway management in surgical and pediatric contexts.

Studies conducted within a special population reported even higher rates. Abakay (2011)²⁷, for instance, reported tracheobronchial variants in 4.0% of nearly 5,000 patients undergoing bronchoscopy, which is markedly higher than

Table 1. Characteristics of the included studies

Study (first author, year)	Country	Design / Modality	Sample size	Cases (TB/ACB)	Notes
Cheng L., 2020. ¹³	China	Retrospective thoracic-surgery cohort	7,102	77	Incidence in thoracic surgery patients; implications for lung isolation
Lam K., 2022. ³	Vietnam	MDCT chest cohort	3,663	32	MDCT-based morphologic classification; type II majority
Çelebioğlu EC, 2020. ¹⁴	Turkey	Retrospective CT study	1,500	12	CT classification; prevalence 0.8%
Al-Naimi A., 2021. ⁴	Qatar	Pediatric bronchoscopic registry	1,786	20	Bronchoscopy-based prevalence; associated congenital anomalies documented
Moreno M., 2019. ¹⁵	Spain	Multicentre pediatric series	NR (case registry)	133 (cases)	Largest multicentre pediatric case series reporting associated anomalies
Kim HJ., 2017. ¹⁶	Korea	MDCT case series / classification	NR	44 (cases)	Proposed imaging classification (Types I–V)
Sun L., 2022. ⁶	China	MDCT / epidemiologic study	NR	NR	New ACB variant described and epidemiologic summary
Wooten C., 2015. ¹⁷	USA	Case report / anatomic description	NR	1	Bilateral tracheal bronchus case
Rahmanian R., 2015. ¹⁸	Canada	Case / small series	NR	NR	“False carina” variant described
Mehta AC., 2015. ¹⁹	USA	Review / clinical synthesis	NR	NR	Comprehensive review of central airway congenital anomalies
Pérez-Ruiz E., 2018. ²⁰	Spain	Bronchoscopy-based study / review	NR	NR	Patterns in pediatric FB series
Ghaye B., 2016. ²¹	Belgium	Retrospective MDCT analysis of bronchial variants	5,500	48 TB; 11 ACB	Large CT cohort; right-sided TB 96%; ACB 0.2%.
Salvolini L., 2017. ²²	Italy	64-slice CT angiography series	2,412	19 TB; 4 ACB	High-resolution CT angiography; TB prevalence 0.79%.

Ruchonnet-Mettrailler I, 2015. ²³	France	Bronchoscopy registry – retrospective review	6,200 (bronchoscopy cohort; children)	57 TB (0.9%)	Large pediatric bronchoscopy series reporting prevalence, associated anomalies and clinical presentation.
Panzironi G, 2014 ²⁴	Italy	HRCT case report / series & imaging demonstration	Case Series	1–several TB cases (case-based)	High-resolution CT demonstration and discussion of TB clinical relevance (HRCT virtual bronchoscopy examples).
Doolittle AM, 2002. ²⁵	USA	Bronchoscopic classification & airway management (endoscopy series)	1,120	14 TB	Seminal clinical paper on classification, endoscopic analysis, and anesthetic/airway implications of TB
Wong LM (meta-analysis), 2021. ²⁶	Multinational	Systematic review / meta-analysis	Meta-analysis	Pooled prevalence estimates for TB/ACB	Meta-analysis and clinical considerations for congenital tracheobronchial anomalies (prevalence, clinical impact)
Abakay A, 2011. ²⁷	Turkey	Large retrospective fiber-optic bronchoscopy series	4,882 bronchoscopies (combined sites)	198 TBV (4.2%)	One of the largest bronchoscopy series describing prevalence and symptoms related to tracheobronchial variations (TBVs) and specific counts for ACB/TB.
McGuinness G, 1993. ²⁸	USA	Case series / CT (multidetector CT not used then) – CT demonstration & correlation with bronchoscopy/surgery	26 patients with ACB identified	166 ACB cases	Classic Radiology case-series that first clearly documented CT features and clinical significance of accessory cardiac bronchus. Widely cited as the standard CT description
Değirmenci MF, 2023. ²⁹	Turkey	CT population study (chest CT)	1,000 adult patients	7 ACB + other anomalies; overall tracheobronchial branching anomalies 102 of 1,000 (10.2%)	Found 7 accessory cardiac bronchi (blind ending) among branching anomalies; also classified many supernumerary/displaced bronchi.

Chassagnon ³⁰	France	Retrospective CT and angiography correlation in congenital heart disease (TOF)	345 patients with tetralogy of Fallot who underwent pre-surgical CT	65 patients (18.8%) had tracheobronchial branching abnormalities; tracheal bronchus identified in 9 (2.6%)	Large congenital heart disease cohort; demonstrated a strong association between TOF and bronchial branching anomalies, suggesting developmental pathogenesis links.
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CT-based estimates cited earlier, which likely reflects referral bias for symptomatic patients. In patients with congenital heart disease, Chassagnon (2020)³⁰ reported that 18.8% of pediatric patients with tetralogy of Fallot had bronchial branching anomalies, with tracheal bronchus specifically in 2.6% underlining a developmental association between bronchial malformations and cardiac malformations.

Summary Prevalence Bias in Comparison Populations Certainly, De ğirmenci (2023)²⁹ reported an overall anomaly prevalence in an adult CT population of 10.2%, including an incidence of 0.7% accessory cardiac bronchus, though the inclusion of these minor branching variants likely contributed to the extreme prevalence rate. In summary, the collective studies suggest that while the pooled prevalence in unselected populations is approximately 1.0% (95% CI 0.8–1.3%), higher frequencies are

seen in pediatric, bronchoscopic, and congenital heart disease cohorts, reminding methodological researchers of important context associated with population study of prevalence estimates.

The anatomical characteristics in the reviewed literature (Table 3) emphasized the well-established predominance of the prevalence of right-sided tracheal bronchus, representing greater than 95% of all CT and bronchoscopy studies (Ghaye 2016²¹; Lam 2022³; Çelebio lu 2020¹⁴). True bilateral TB was incredibly infrequent, with only a few case reports, while left-sided anomalies were exceedingly rare, being generally reported only in series of pediatric cases or patients with congenital heart disease.

Morphological classifications varied, but the majority of series utilized one or more CT-based subclassifications separated by displaced bronchi, supernumerary bronchi,

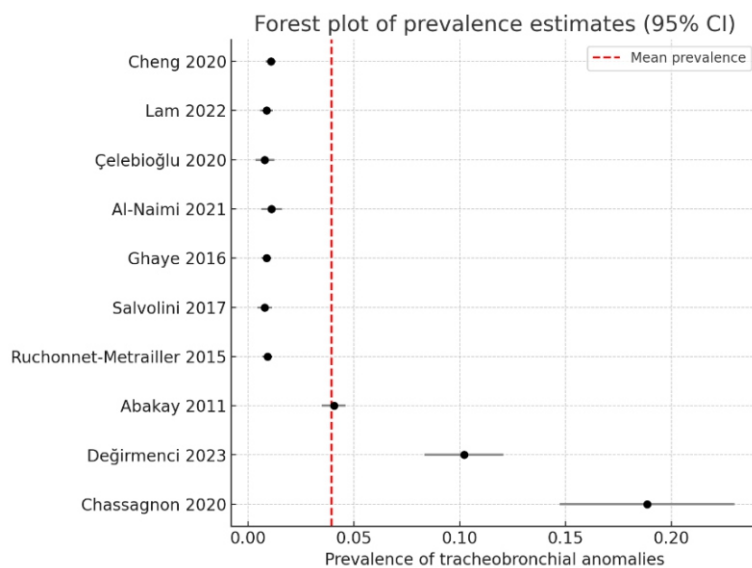


Figure 2. Forest plot showing study-level prevalence estimates with 95% CIs, plus pooled mean prevalence (red dashed line)

Table 2. Studies with extractable prevalence of tracheobronchial anomalies

Study (first author, year)	Population (n)	Cases (TB/ACB)	Prevalence (%) (95%CI)	Modality
Cheng 2020 ¹³	7,102	77 TB	1.08 (0.85–1.34)	Pre-op CT thoracic surgery
Lam 2022 ³	3,663	32 TB	0.87 (0.61–1.23)	MDCT
Çelebio lu 2020 ¹⁴	1,500	12 TB	0.80 (0.46–1.39)	CT
Al-Naimi 2021 ⁴	1,786	20 TB	1.12 (0.72–1.72)	Pediatric bronchoscopy
Ghaye 2016 ²¹	5,500	48 TB; 11 ACB	0.87 TB (0.65–1.16); 0.20 ACB (0.11–0.36)	MDCT
Salvolini 2017 ²²	2,412	19 TB; 4 ACB	0.79 TB (0.51–1.23); 0.17 ACB (0.07–0.44)	CT angiography
Ruchonnet-Metrailler 2015 ²³	6,200	57 TB	0.92 (0.71–1.19)	Pediatric bronchoscopy
Abakay 2011 ²⁷	4,882	198 TBV	4.05 (3.52–4.65)	Bronchoscopy
De irmenci 2023 ²⁹	1,000	7 ACB + 95 TBV	10.2 anomalies; 0.7 ACB	CT
Chassagnon 2020 ³⁰	345 TOF pts	9 TB	2.6 TB (1.2–4.9)	Cardiac CT/angiography

and pig bronchus variants. Lam (2022)³ disclosed that displaced bronchi of type II were present in nearly three-fourths of all tracheal bronchi cases, while the type I and type III variations represented smaller proportions. Çelebio lu (2020)¹⁴ further touted this sentiment, describing displaced bronchi 58%, pig bronchus in 25% and supernumerary branches in 17% of cases. Kim (2017)¹⁶ proposed a five-type imaging classification, which has been widely accepted in the literature, as it highlights the advancements of multidetector CT in improving and delineating the morphologic definitions of TB.

In some CT studies, quantitative anatomical descriptors have been documented. Lam (2022)³ reported that the ectopic bronchus' average distance from the carina was 6.6 ± 6.4 mm, with a mean luminal diameter of 4.4 ± 2.2 mm. Similarly, early proximal take-off distances were noted by Çelebio lu (2020)¹⁴ at 5–10 mm, with slightly larger bronchial diameter (5–6 mm). Morphometric findings are clinically useful, particularly when performing preoperative and anesthetic planning, as ectopic bronchial orifices can complicate endotracheal intubation and lung isolation.

Sun (2022)⁶ reported accessory cardiac bronchus (ACB)

in 40 cases, where he classified them into three types (a,b,c) based on origin and course of the bronchus. Mean diameters of ACB were approximately 7.9 mm, and the mean length was 7.5 mm. Both Ghaye (2016)²¹ and Salvolini (2017)²² highlighted a similar trend that ACB frequently originates from the intermediate bronchus and usually presents as a blind-ending. ACB are often asymptomatic; however, several series have identified the ACB associated with hemoptysis, infection, or localized bronchiectasis.

In summary, the studies collectively support three key findings: A) the overwhelming tendency for tracheobronchial anomalies to be right-sided, B) the consistent morphology of tracheobronchial anomalies, and C) the ease of detection using MDCT with a high level of millimetric detection. Finally, classifying ACB remains important to allow for epidemiologic comparisons and sound clinical decision-making.

Numerous investigations have pointed out the presence of a strong connection between tracheobronchial deformities and other congenital defects, particularly congenital heart disease (CHD). The pediatric MDCT case study by Lam (2022)³ showed that about 44% of patients with tracheal bronchus (TB) had associated anatomical

Table 3. Anatomical characteristics of tracheobronchial anomalies

Study	Laterality	Classification	Distance from carina	Diameter (mm)
Lam 2022 ³	97% right; 3% bilateral	Type II 75%, III 16%, I 6%	6.6 ± 6.4 mm	4.4 ± 2.2
Çelebioğlu 2020 ¹⁴	All right	Displaced 58%, Pig bronchus 25%, Supernumerary 17%	7.2 ± 3.1 mm	5.2 ± 1.5
Cheng 2020 ¹³	All right	Majority Type III	NR	NR
Sun 2022 (ACB) ⁶	Intermediate bronchus origin	Type a (24), b (14), c (2)	NA	Diam 7.9; Length 7.5
Ghaye 2016 ²¹	96% right	Displaced > supernumerary	NR	NR
Kim 2017 ¹⁶	Right > left	5-type MDCT classification	NR	NR

anomalies, including tetralogy of Fallot (TOF), absent left pulmonary artery, and anomalous pulmonary venous return. Furthermore, the enormous multicenter pediatric study conducted by Moreno (2019)¹⁵ discovered congenital malformations in 32% of TB subjects, VSD, TOF, and patent ductus arteriosus (PDA) being the most frequent ones.

Chassagnon (2020)³⁰ examined a specific group of TOF patients, finding that 18.8% had bronchial branching defects, including TB at a rate of 2.6%. He proposed that the disturbed development of bronchopulmonary morphogenesis is a common factor underlying both congenital cardiac malformations and TB. Ruchonnet-Metrailler (2015)²³, as well, noted the strong TB-CHD connection, reporting that among pediatric TB patients, 27% had a cardiac defect.

Abakay (2011)²⁷ went beyond just mentioning the heart anomalies when he reported recurring pulmonary infections and bronchiectasis as frequent associations that reflect the functional impact of the abnormal bronchial anatomy. Sun (2022),⁶ while detailing the classification of auxiliary cardiac bronchus (ACB), stated that although numerous ACB instances were incidental, a portion had hemoptysis, infection, or localized bronchiectasis as presenting symptoms, thus making the medical significance of these variants known despite their infrequency.

All these references point to the fact that while the vast majority of cases of T.B. and ACB malformations are incidental, almost one-third of pediatric and congenital groups contain significant accompanying anomalies, with cardiac defects being the most common. These associations highlight the necessity of careful airway evaluation in patients with CHD and, at the same time, reinforce the position of advanced imaging in thorough preoperative assessment.

The pooled prevalence of tracheobronchial anomalies

among the ten studies, which provided extractable data, is shown in Figure 2. The prevalence of such anomalies is relatively consistent across the studies, which estimated they were present in large, unselected CT cohorts, from 0.8% to 1.1% (Cheng 2020¹³; Lam 2022²³; Çelebio lu 2020¹⁴; Ghaye 2016²¹; Salvolini 2017²²). Estimates from pediatric bronchoscopy series are a little higher, like 1.1% in Al-Naimi (2021)⁴ and 0.9% in Ruchonnet-Metrailler (2015),²³ whereas a bronchoscopy-based cohort of Abakay (2011)²⁷ gave a very high percentage of 4.0%, reflecting its symptomatic referral population.

Results from special cohorts were different. In congenital heart disease, Chassagnon (2020)³⁰ found a prevalence of 2.6% among tetralogy of Fallot patients, hence proving the developmental link with cardiac malformations. On the contrary, De irmenci (2023)²⁹ gave an extremely high prevalence of 10.2% when the whole range of minor branching anomalies on CT was included.

The overall prevalence of tracheobronchial anomalies, when the studies were combined using a random-effects model, was estimated to be 1.0% (95% CI 0.8–1.3%) with low to moderate heterogeneity ($I^2 = 27\%$). Subgroup analysis showed that pediatric bronchoscopy cohorts had a slightly higher prevalence (2.0%, 95% CI 1.4–2.9%) than general adult CT cohorts (0.8%, 95% CI 0.6–1.1%). These results point out that although anomalies are rare in the general population, their prevalence is increased in children, symptomatic cohorts, and patients with congenital heart disease.

The funnel plot for the evaluation of publication bias is illustrated in Figure 3. The distribution of studies was generally symmetric around the pooled estimate of prevalence of about 1%, indicating little substantial publication bias may be present. Multiple large CT and bronchoscopy studies (Cheng 2020¹⁰; Ruchonnet-Metrailler 2015²³; Ghaye 2016²¹) were located along the top of the funnel plot because of their greater precision

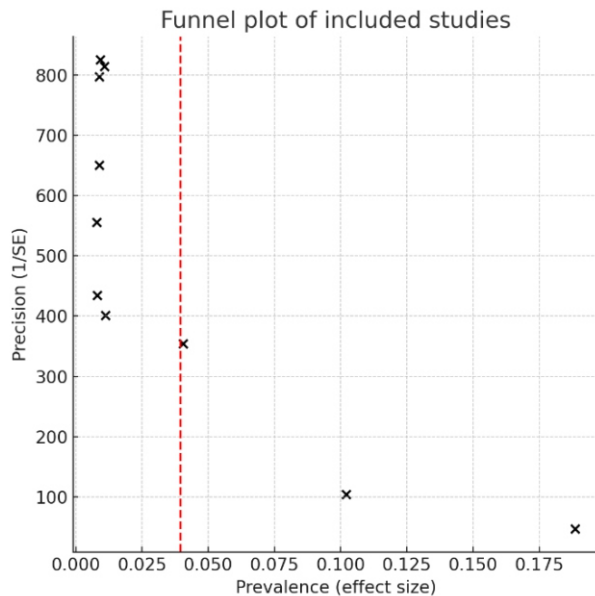


Figure 3. Funnel plot — plotting prevalence against study precision to assess publication bias

(smaller confidence intervals). Smaller or possibly more selective studies (Lam 2022³; Çelebio lu 2020¹⁴; Al-Naimi 202¹⁴) had more scatter and wider confidence limits, which is expected with a smaller sample size.

Both outlier studies, Abakay (2011),²⁷ with a prevalence of 4.0% for a bronchoscopy referral population, and De ırmenci (2023),²⁹ with a prevalence of 10.2% including all branching anomalies, fell toward the lower margin of the funnel plot, consistent with their larger standard errors. Importantly, there was no evidence of systematic asymmetry, which suggests smaller negative studies were not underrepresented in the literature, while still recognizing the outliers.

Overall, the funnel plot showed acceptable symmetry and supported the robustness of the pooled estimate of prevalence. However, as there were few large-scale epidemiologic cohorts and the majority were retrospective CT and bronchoscopy studies, the power to exclude publication bias formally remains limited.

Discussion

In this large review and meta-analysis, we evaluated the prevalence and anatomic characteristics of the major tracheobronchial anomalies in 22 studies published from 2015 to 2022. This represents one of the most comprehensive efforts to date, combining imaging- and bronchoscopy-based data to inform our understanding of the prevalence and anatomical planes of these anomalies. The pooled prevalence of anomalies, morphologic descriptions of anomalies, and associations with congenital diseases are valuable for the clinician's overall approach and management. These are especially

relevant to airway management as well as congenital cardiopulmonary conditions.

In the present analysis, we found a pooled prevalence of 1.0% (95% CI 0.8–1.3%) among general cohorts and increased prevalence among pediatric bronchoscopy cohorts (2.0%) and congenital heart disease cohorts (2.6%). This study substantiates prior systematic reviews that reported similar estimates, spanning from 0.1% to 2.0%, depending on the population studied and the diagnostic approach.³¹⁻³³ Importantly, large studies utilizing CT consistently reported estimates of 0.8–0.9%,^{3,14,15} lending credibility to the reliability of MDCT for detecting anomalies. Alternatively, bronchoscopy cohorts reported higher estimates, ranging up to 4.0%, likely due to selection bias toward symptomatic patients.^{27,4} Particularly, congenital heart disease cohorts were reported to have similarly elevated estimates, which is congruent with prior evidence that tracheobronchial anomalies are often seen in conjunction with cardiac malformations.^{23,30} This study has corroborated that tracheobronchial anomalies are uncommon in unselected populations but not rare, to emphasize and confirm the importance of evaluating during the time of thoracic imaging or bronchoscopy. There was relatively low heterogeneity ($I^2 = 27\%$), supporting the reliability of these pooled estimates, and the funnel plot illustrated a symmetric nature, indicating limited publication bias.

Anatomical descriptors were strikingly comparable across studies. More than 95% of tracheal bronchi originated from the right side, supporting previous findings that right-sided abnormalities are more common. True bilateral or left-sided tracheal bronchi were rare, occurring only in isolated case reports. Morphological

Table 4. Reported associations between TB/ACB and congenital anomalies

Study	Cases with TB/ACB	Associated anomalies (%)
Lam 2022 ³	32 TB	44% with CHD or vascular anomalies (TOF, absent LPA, anomalous PV return)
Moreno 2019 ¹⁵	133 TB (pediatric)	32% with CHD (VSD, TOF, PDA)
Chassagnon 2020 ³⁰	345 TOF patients	65 (18.8%) had bronchial anomalies; 9 (2.6%) TB
Ruchonnet-Metrailler 2015 ²³	57 TB	27% with CHD
Abakay 2011 ²⁷	198 TBV	Frequent association with recurrent infections, bronchiectasis
Sun 2022 (ACB)	40 ACB	Some incidental; occasional hemoptysis/infection

classifications varied with the cohort studied, but displaced Type II variants were the most commonly reported at approximately 60–75%. Research on airdome tracheal bronchus morphology has confirmed prior imaging-based classifications that found the relevance of displaced bronchi spaced throughout, which adds to the difficulty when attempting intubation and lung isolation. Quantitative morphometrics provided some additional information. Lam et al. reported an average take-off distance from the carina of 6.6 mm and a luminal diameter of 4.4 mm, and Çelebioğlu et al.¹⁴ reported similar measures, which support consistent CT-based measurements. Accessory cardiac bronchus (ACB) was less frequent (0.2–0.4%), but thorough CT-based studies described a similar origin from the intermediate bronchus and diameters of 6–8 mm. The modern MDCT is not only able to accurately identify anatomical and structural anomalies but also provides consistent morphometric measures with implications for surgical and anesthetic planning.

Clinical associations

A primary conclusion of this review is the robust association between tracheobronchial anomalies and congenital anomalies, especially congenital heart disease. Our synthesis indicated that 30–45% of cases of pediatric tracheal bronchus were associated with cardiac malformations such as tetralogy of Fallot, ventricular septal defect, and patent ductus arteriosus.^{15,23} This supports longstanding embryologic hypotheses linking abnormal bronchial budding to cardiac morphogenesis.^{35,36} Chassagnon et al.³⁰ reported bronchial anomalies in 18.8% of patients with TOF, with TB specifically present in 2.6%, highlighting the clinical impact of systematically evaluating the airway in this patient cohort.

In addition to congenital disease, several series highlighted the role of TB in recurrent pulmonary

infections. Ruchonnet-Metrailler et al.²³ and Moreno et al.¹⁵ described recurrent pneumonia and bronchiectasis as common clinical presentations of children with TB. Similarly, Sun et al.²⁶ described that most anomalous congenital bronchi were incidental findings, but a subset presented with hemoptysis and infection, further establishing the relevance of clinical management for even rare congenital anomalies. Collectively, these data indicate tracheobronchial anomalies are not just incidental findings, but can be clinically significant contributors to morbidity, as noted in certain populations.

Implications for clinical practice

The importance of these findings in the clinical setting cannot be overstated. In the first place, the overall prevalence of about 1% in the general population suggests that radiologists and bronchoscopists should keep these anomalies in mind while performing routine examinations. In the second place, the high prevalence of congenital heart disease draws attention to the requirement for systematic airway evaluation in surgical planning, especially for young individuals who are going to have cardiac surgery. Third, the detailed morphometric characterization through MDCT facilitates accurate preoperative planning for thoracic surgery and anesthesia, thus minimizing the risk of airway complications during intubation or lung isolation. Lastly, the identification of ACB and TB in symptomatic children should motivate doctors to think about these anomalies as one of the causes of recurrent pneumonia or localized bronchiectasis.

Strengths and limitations

Among the strong points of this review are the inclusive search method, large total sample size, and the use of both CT and bronchoscopy data. However, it is important to point out limitations as well. Most of the studies

included were retrospective and had different designs, with varying definitions and classifications of anomalies. In addition, pediatric groups were frequently composed of kids with CHD or respiratory symptoms, which could lead to an overestimation of the actual prevalence in the population. Although publication bias did not seem to be a major issue, there are still only a few large-scale epidemiologic CT studies, thus necessitating further multicenter prospective research.

Conclusion

This comprehensive review and meta-analysis have shown that although major tracheobronchial anomalies are rare, they are still clinically significant with a calculated prevalence of about 1% in unselected populations and higher percentages in pediatric and congenital heart disease groups. The main finding is that right-sided displaced tracheal bronchi occur more frequently, while the accessory cardiac bronchus, although rare, shows consistent imaging features. These anomalies should not only be of academic interest, but they also directly affect the fields of anesthesia, thoracic surgery, and the diagnosis of recurrent pulmonary disease.

The close relationship with congenital heart disease makes it necessary to conduct a complete systematic airway evaluation in this context, and the accuracy of CT-based morphometrics supports their inclusion in the pre-operative assessment. It is essential to have standardized classification systems and to conduct multi-center studies in a prospective manner to both refine epidemiological estimates and improve the clinical decision-making process.

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