Respiratory Medicine (2015) 109, 846–850

Available online at www.sciencedirect.com

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

Presence of tracheal bronchus in children undergoing flexible bronchoscopy

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Received 5 February 2015; accepted 11 April 2015
Available online 20 April 2015

KEYWORDS
Flexible bronchoscopy;
Tracheal bronchus;
Lung development

Summary
Background and objectives: Tracheal bronchus (TB) is a rare congenital malformation of the lung tree with a bronchus originating from the trachea. Only a small number of publications have analyzed the frequency and diagnostic procedure of TB in children, based on a restricted sample of patients. In the present study, we analyze and discuss new aspects of prevalence, clinical presentation and associated malformations of TB based on a large pediatric cohort.
Methods: Data from 5970 children having a flexible bronchoscopy for investigation of respiratory symptoms were selected. We analyzed the anesthesic management, the presence of associated malformations, and all tracheobronchial anomalies observed during the endoscopic procedure.
Results: Fifty-seven cases of tracheal bronchus were identified (0.9%). In the majority of them, tracheal bronchus was a fortuitous discovery without clear clinical relevance. Statistical analysis revealed that the majority of TB originated from the middle and lower one third of the trachea (56%). 61.5% of patients had associated anomalies such as syndromic association (21%), cardiac malformations (19.2%) or tracheal stenosis (14%). Only 38.5% of children had no associated anomalies.

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http://dx.doi.org/10.1016/j.rmed.2015.04.005

Conclusions: Tracheal bronchus is a rare morphological anomaly of the tracheobronchial tree. Most often TB is associated with other birth defects such as another tracheo-bronchial tree malformation, vascular abnormality, congenital heart malformation or in the context of a syndromic pattern. A relationship between respiratory symptoms and the presence of TB is very rare and selective treatment is infrequent.

Introduction

In humans, normal tracheobronchial development begins between 24 and 26 days of gestation as a median bulge of the ventral wall of the pharynx, which develops at the caudal end of the laryngotracheal groove. Between days 28 and 30, the lung buds have elongated into primary bronchi. All segmental bronchi are formed at day 36. Within the same period, the vascular supply develops from branches of the splanchnic plexus to define the pulmonary artery. Tracheal bronchus is known as an anatomically abnormal evolution with a wide range of bronchial anomalies originating from the trachea or the main bronchi and directed to the upper lobes of the lung. The evolution of pediatric bronchoscopy in the last decades has allowed detecting more tracheobronchial anomalies such as tracheal bronchus. Tracheal bronchus is reported in 0.1—2% of the general population [1]. The term tracheal bronchus (TB) is a term applied to any airway, which arises from the trachea above the level of the carina. Two different classifications are found in the literature. The first one describes three different types of TB based on the level of connection in the trachea (type I, II or III) [2]. The second classification, which is a modified nomenclature of Boyden [3] and Kubik [4] differentiates aberrant bronchi directed to the upper lobes. This second classification depends on the pulmonary artery and presence of displaced bronchus or supernumerary bronchus.

In adults, some studies have reported that patients having TB had symptoms such as persistent or recurrent atelectasis, localized recurrent pneumonia, congenital stridor, wheezing or hemoptysis [5—7]. An adult case report described also an endobrochial squamous cell carcinoma associated with TB [8].

Using these two classifications, the objective of this paper is to describe the anatomical characteristics of a large pediatric cohort of patients (n = 5970) having flexible bronchoscopy for respiratory symptoms, and to analyze the associated symptoms, or malformations.

Methods

Data collection

The current study was conducted at Necker University Hospital (Paris, France). All data were collected from an endoscopic database including bronchoscopies performed between January 2000 and September 2013 at the pulmonology unit. Data were collected in a prospective and a retrospective manner. Criteria of inclusion for analysis were an endoscopy performed in children between birth and 16 years of age. Exclusion criteria included endoscopy performed by a surgeon or otorhinolaryngology team. Preoperative data such as endoscopic indication, radiological exams and associated malformations were collected. We also documented the anesthesic management with the type of sedation. Finally, we collected all anomalies observed during endoscopic procedure at the laryngeal, tracheal and bronchial levels.

Statistical analysis

Quantitative variables are presented as median and interquartile range (IQR), corresponding to the difference between the third (75% of the distribution) and first (25% of the distribution) quartiles.

Results

Out of the 5970 pulmonary flexible bronchoscopies, 57 cases of TB were described (0.95%). Ninety percent of procedures were performed under general anesthesia and nine percent under conscious sedation.

Median age of patients diagnosed with tracheal bronchus was 22 months (CI 1—110 months) with a majority of males (54.3%) (Table 1). The most frequent indications for the procedure were recurrent bronchopneumonia (45.6%) and difficult to treat asthma (21%). Other indications are listed in Fig. 1.

61.5% of children with TB had an associated malformation (Table 2) with mainly cardiac malformations (19.2%: Tetralogy of Fallot, ventricular septal defect, aortic arch anomalies, great vessels transposition, pulmonary vascular malformations), syndromic association (21%: three patients with Down syndrome and two with CHARGE syndrome), eight cases of congenital tracheal stenosis (14%) and three

Table 1 Clinical characteristics of children with tracheal bronchus (n = 57).

	N or median	CI or %
Tracheal bronchus	57	0.95
Mean age (months)	22	(1—110)
Sex male	31	54.3
Sedation		
General anesthesia	52	91
Conscious sedation	5	8.7