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ORIGINAL ARTICLE

A Review of Tracheal Bronchus in Universiti Kebangsaan Malaysia Medical Centre (UKMMC)

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Abstract Tracheal bronchus (TB) is a rare congenital anomaly described as a abnormal bronchus that originates directly from the trachea above the carina directed towards the upper lung lobe. We analysed all paediatric rigid endoscopies of the airway from January 2015 until August 2020 to determine the incidence and characteristic of TB. In total, 68 rigid endoscopic airway examination record from children aged 0 to 12 years were analyzed. Endoscopic examination was performed from supraglottic region to carina using a 0 degree Hopkins rod lens telescope. Patients with a TB were identified and the site of TB origin was noted. Data of the identified patients was reviewed for the presence of preoperative airway findings such as stridor, upper lobe pneumonia, other congenital anomalies, intraoperative findings and complications and postoperative general condition outcome. TB was detected in 8 (11.8%) of 68 airway endoscopic examinations. 6 children (75%) were syndromic. 5 patients (62.5%) has congenital malacic airway and 2 patients (25%) has congenital tracheal stenosis. All TB originated from the right lateral wall of the trachea. All children had stridor unrelated to TB as presentation and 4 (50%) of children had preoperative upper lobe pneumonia. Tracheal bronchus is not a rare finding and is highly associated with syndromes and other airway anomalies. Although children with TB can be asymptomatic, upper lobe pneumonia is a common presentation. TB should be included in the differential diagnosis in patients with recurrent right upper lobe pneumonia or collapse and patients with unexplained oxygenation problem during endotracheal intubation, particularly in children with syndromes or other congenital anomalies.

Keywords Tracheal bronchus · Rigid endoscopy · Paediatric · Upper lobe collapse

Introduction

Tracheal bronchus (TB) was first described by Kubik and Muntener [1] in 1785 as a right upper lobe bronchus originating from the trachea. True TB is a congenital anomaly in which a right upper lobe bronchus has its origin in the trachea rather than at the carina. Bronchus suis and "pig bronchus" are alternative names used because a TB is a normal entity in swine.

TB usually arises from the right lateral wall of the trachea within 2 cm of carina but can arise anywhere from cricoid cartilage to the carina. It can supply the entire upper lobe or just its apical segment TB has been reported to be have an increased risk of pneumonia, atelectasis, and ventilation difficulties. Diagnosis of TB can be made with rigid or flexible bronchoscopy in conjunction with imaging studies.

To date only a few studies exist regarding the incidence of TB in humans [2, 3] and to our knowledge this study will be the first done in Malaysia.

The aim of this study is to retrospectively review the associated comorbidities, clinical presentation, endoscopic finding and perioperative ventilation difficulties associated with TB.

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Materials and Methods

This is a retrospective study conducted at Universiti Kebangsaan Malaysia Medical Centre. All data were collected from an operating record of rigid airway endoscopies performed between January 2015 and August 2020 in children from birth until 12 years of age. The indications for endoscopic airway examination were airway obstruction, persistent lung collapse, failed multiple extubations as well as foreign body removal. Endoscopic examination was performed from supraglottic region to carina using a 0 degree Hopkins rod lens telescope. All rigid laryngotracheoscopies were performed by the second author.

In the analysis of the 68 endoscopic records of the trachea, patients with TB were identified and the site of origin of TB was documented. Preoperative data of the identified patients was reviewed to assess the presence of respiratory findings such as stridor, upper lobe pneumonia, and also the presence of associated congenital anomalies and/or syndromes. Intraoperative airway complications such ventilation difficulties, and/or oxygenation problems as well as postoperative respiratory complications were noted from the patient's anaesthesia and postanesthesia care unit records.

Results

A tracheal bronchus was detected in 8 (11.7%) of the 68 patients aged from birth to 12 years. The TB originated from the right side of the trachea in all 8 patients.

The clinical data of these 8 patients was shown in Table 1. The prevalence of TB was higher in males (87.5%) than in females. A total of 6/8 (75%) of patients with TB have syndromic association; 5 patients with Down Syndrome and 1 patient with CHARGE syndrome.

All children had stridor unrelated to TB as presentation but 4 children (50%) had concomittant preoperative chest radiograph finding of upper lobe pneumonia.

Three (37.5%) of TB patients had a chest CT scan for persistent lung infection before endoscopy with tracheal bronchus described in only 1 patient (33.3%).

All of our patients had right sided TB only in this study. Five children (62.5%) had associated congenital malacic airway, 3 children(37.5%) had acquired laryngotracheal stenosis and 2 children(25%) had congenital tracheal stenosis in the presence of TB.

None of the patients had complication postoperatively. All patients with TB could be discharged from the post anesthesia care unit as planned, without immediate respiratory complications that could be associated to the presence of an anomalous TB.

Discussion

Paediatric bronchoscopy has evolved in the last decades allowing more tracheobronchial anomalies such as tracheal bronchus to be detected. Despite the increasing number of bronchoscopies performed in children, either rigid or flexible, few data exist on the incidence and description of TB in children.

Our study evaluated the prevalence, location, and related clinical manifestations as well as intraoperative and postoperative complications of TB in young children. We report prevalence of TB as 11.7% in our study. To our knowledge this is the highest incidence rate in comparison to that reported in the literature (0.1–2%) [2–4]. The much higher prevalence of TB in males (87.5%) than in females is another new finding of this study. Previous study reported the prevalence of TB was higher in females than in males (72%) [4]. Our series also highlight the frequency of TB in children with syndromic association (75%) in which Down syndrome is the most common syndrome found in our series (62.5%). This correlates with the previous study which described that Down syndrome population tend to show a prevalence of ten to 20- fold more frequent than in general population [5].

High percentage of our patients (50%) had pre endoscopy upper lobe consolidation or atelectases. For this reason, a tracheal bronchus should be included in the differential diagnosis in a child with recurrent right upper lobe pneumonia or collapse, particularly in children with syndromes or other congenital anomalies. On the other hand, patients with a TB can be asymptomatic and only diagnosed incidentally by bronchoscopy or chest computed tomography for other respiratory diseases [4].

CT scan can represent as an alternative to bronchoschopy to diagnose and describe TB. Three dimensional reconstruction techniques allow a reliable evaluation of central airway diseases and morphological anomalies. In this study, a small number of patients had a CT scan before bronchoscopy for persistent respiratory symptoms. However, a TB was only correctly described in 33.3% of these patients. These results suggest that a systematic and careful CT scan image analysis is essential to correctly detect the presence of TB before bronchoscopy (Figs. 1, 2).

We observed only right sided tracheal bronchus in our population. Right TB is reported more commonly in the literature than left TB [6]. TB has been classified into 3 types in the current literature [7]. Type I is usually described to originate roughly at the junction of the middle and lower one third of the trachea. Type II is a small but distinct bronchus connected to the lower third of the trachea and type III is the TB arising from the tracheal wall

Table 1 Clinical data of 8 children with a tracheal bronchus (TB)

Patient	Age (months)	Sex	Associated congenital anomaly	Endoscopic indication	Pre endoscopy chest CT Scan	Intraoperative finding	Intraoperative complication	Postoperative complication
1	2	M	Down syndrome	Stridor	None	Laryngomalacia *Right TB*	None	None
2	22	M	None	Stridor with failed extubation Right lung pneumonia right hemithorax pleural effusion	Yes. No description of TB	Left vocal cord palsy Acquired tracheal stenosis *Right TB*	None	None
3	2	M	Down syndrome Duodenal Atresia	Stridor	None	Laryngomalacia Congenital lower tracheal stenosis *Right TB*	None	None
4	8	M	Down Syndrome Patent Ductus Arteriosus(PDA) Ventricular Septal Defect(VSD)	Stridor Failed multiple extubation Right upper lobe atelactasis	None	Acquired tracheal stenosis *Right TB*	None	None
5	3	F	CHARGE Syndrome Patent Ductus Arteriosus (PDA)	Stridor Right upper lobe pneumonia	None	Severe laryngomalacia *Right TB*	Intra operative desaturated during attempt to visualise the opening	None
6	1	M	Down syndrome Patent Ductus Arteriosus (PDA) Atrial Septal Defect(ASD)	Stridor Right upper lobe atelactasis	Yes. Presence of right TB	Laryngomalacia Bronchomalacia Congenital tracheal stenosis *Right TB*	None	None
7	1	M	Down Syndrome	Stridor with failed extubation Right upper lobe pneumonia	Yes. No description of TB	Laryngomalacia *Right TB*	None	None
8	108	M	None	Intubation injury with acquired subglottic stenosis	None	Acquired subglottic stenosis *Right TB*	None	None

almost at the level of the carina giving the appearance of trifurcated carina.

We are unable to classify TB in our study in view of no documentation on the level of TB origin available in our records. Nevertheless, the importance of identifying type TB in the literature cannot be overemphasized.

In type I TB, apart from the confusing appearance that can arise during a bronchoscopic foreign body removal, an endotracheal intubation of either into the true trachea or the TB would cause ventilation problem and pose a risk of hypoxia, pneumothorax, atelectasis as well as weaning difficulties [7–9]. It is of critical importance to confirm on the tube position in these circumstances using flexible fibreoptic scope. Type II and III TB are more likely to complicate patients undergoing cardiac surgery with one lung ventilation. An undiagnosed TB could lead to intubation or ventilation difficulty during the single lung ventilation attempt technique. It is advocated to perform a diagnostic bronchoscopy in all patients requiring single lung ventilation to avoid airway injury related to the blind insertion of a DLT (Double Lumen Tube). Where a diagnosis of TB is confirmed, the literature recommends the use of a normal endotracheal tube and a bronchial blocker rather than a DLT [7].

Other congenital airway anomalies often complicate tracheal bronchus as seen in this study. This include laryngotracheomalacia (62.5%) and congenital tracheal stenosis (25%). It is not surprising as other studies also

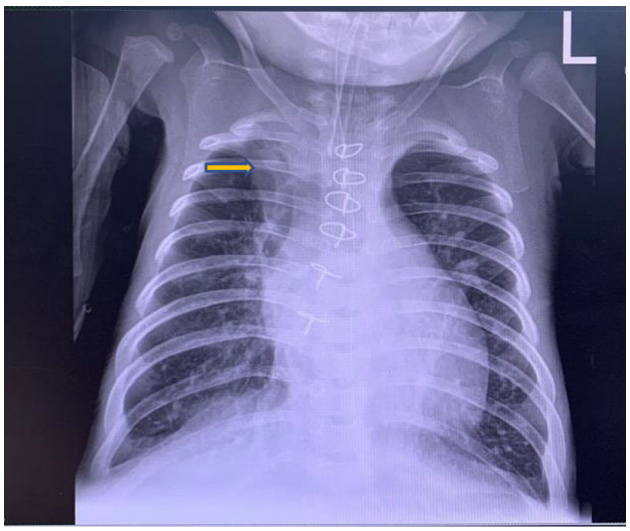

Fig. 1 Chest radiograph of a 8 month old Down syndrome child with persistent right upper lobe atelactasis

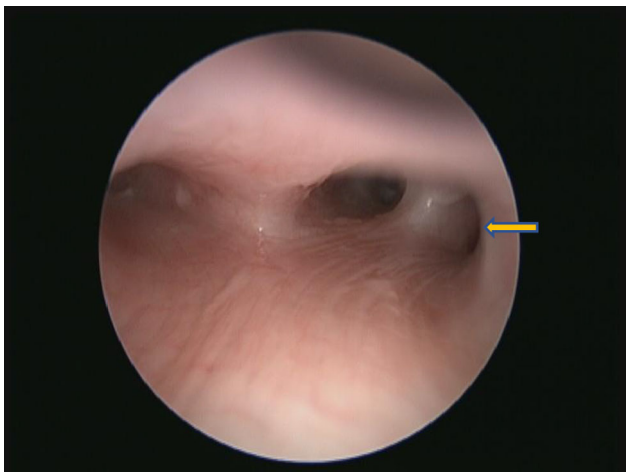

Fig. 2 Hopkins rod-lens telescope view at the true carina of the similar patient showing a tracheal bronchus arising from the right tracheal wall

found that besides the presence of tracheal bronchus many has other congenital airway anomalies including laryngomalacia, tracheomalacia, tracheal stenosis and trachea esophageal fistula [4, 10].

Despite all the intraoperative complications related to TB mentioned in the literature, our study showed only one intraoperative hypoxia during endoscopic examination procedure but it was temporary and brief episode which was not related to the TB but the endoscopic procedure itself.

Treatment of TB is based upon the severity of symptoms. Most patients with a TB can be managed conservatively. In the case of recurrent lung infections, surgical excision of the involved segment may become necessary [11].

In conclusion, tracheal bronchus is not a rare finding and is highly associated with syndromes and other airway anomalies. Although children with tracheal bronchus can be asymptomatic, upper lobe pneumonia is a common presentation. TB should be included in the differential diagnosis in patients with recurrent right upper lobe pneumonia or collapse and patients with unexplained oxygenation problem during endotracheal intubation, particularly in children with syndromes or other congenital anomalies. In special situation such as single lung ventilation cardiac surgery, the treating physician should be alerted regarding the presence of TB in order to prevent ventilation difficulty intraoperatively.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval Ethical committee permission was taken.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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