



# Foregut duplications in the superior mediastinum: beware of a common wall with the tracheo-bronchial tree

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## Abstract

**Background** Foregut duplication cysts (FD) with a common muscular wall with the oesophagus are well recognised. Our case series highlights the existence of a common wall between superior mediastinal FD and the tracheo-bronchial tree.

**Materials and methods** Over the last 15 years, we have thoroscopically resected 41 FD. Five cases were identified to have a common wall with the tracheo-bronchial tree at operation. The clinical history, radiology, findings at operation and pathology were evaluated to highlight learning points.

**Results** Five superior mediastinal cysts with a common wall were identified, with two antenatally and three postnatally diagnosed. All three postnatal cases and one antenatal case presented with symptoms of respiratory compromise and stridor. Only one neonate was relatively asymptomatic before resection. CT similarities in all cases were: separation of trachea and oesophagus by the cyst, oesophageal deviation to the right or compression and compression of trachea/bronchus. Thoroscopically, two cysts were resected without injury to the airway, while three had inadvertent tracheo-bronchial injury. Repair of the tracheal injury was possible in one case thoroscopically, while two cases required conversions, as adequate oxygenation could not be maintained despite on-table endotracheal tube advancement beyond the injury and thoroscopic manoeuvres. All cases had excellent outcomes at follow-up (median 25 months, range 4–39 months) with resolution of symptoms and no recurrences.

**Conclusions** Our report highlights the existence of a common wall between the superior mediastinal FD and the tracheo-bronchial tree. Thoroscopic resections are feasible including repair of inadvertent airway injury, provided adequate oxygenation can be maintained.

**Keywords** Foregut duplication · Superior mediastinal cyst · Airway common wall · Bronchogenic cyst · Thoroscopic

## Introduction

Foregut duplication cysts (FD) are an uncommon congenital anomaly that results from disordered formation of the embryonic foregut. They commonly present with either an antenatal diagnosis, or postnatal respiratory or swallowing difficulties. The presence of a common wall between FD and oesophagus is well documented [1], but the existence of a common wall with an airway is not as well recognised and

is rarely reported in the literature. We examined our cohort of FD to investigate the prevalence of this and to help raise awareness in surgeons looking after these conditions.

## Materials and methods

We evaluated 5 superior mediastinal FD cases that had a common wall with the tracheo-bronchial tree amongst 41 cases resected in our institute thoroscopically. We retrospectively reviewed the case notes for clinical presentation, pathology, radiology and operative findings.

All patients underwent preoperative investigations including chest X-ray, ECHO, CT chest with IV contrast and upper gastrointestinal contrast study.

Thoroscopic approach was via a right or left side depending on the preoperative imaging. Typically, three

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trocars (5 mm) were used in a lateral position, one for the 5 mm optical camera and two for instruments. Dissection was performed close to the cyst and gradually separating the FD from its surrounding mediastinal structures. The monopolar diathermy hook was judiciously used to reduce the risk of thermal damage and conduction to the mediastinal viscera. As the common wall was encountered, sharp dissection was undertaken under vision remaining close to the cyst. During dissection if the cyst was found to be tense, it was aspirated to avoid its rupture. Rupture of the cyst tends to make the dissection more difficult as the tissue planes are less easily defined.

In uncomplicated cases, a postoperative chest drain was not inserted, while in cases in which an airway injury occurred a chest drain was left in situ. Uncomplicated patients were managed on the surgical ward with appropriate postoperative analgesia, while cases with tracheal injury were managed in our intensive care unit for 2–3 days. Postoperative X-rays were performed to demonstrate full expansion of the lung and, once the subsequent postoperative days demonstrated no air leak from the repaired site, the chest drains were removed. The children were monitored for a further 2–3 days for delayed air leakage before discharge and followed up.

## Results

A total of five cases were identified at operation with a common wall between the superior mediastinal cyst and the tracheobronchial tree. Two cases were identified antenatally with the finding of a cystic mediastinal structure, of which one was symptomatic at birth with noisy breathing and acute life-threatening events. Three cases were diagnosed postnatally (age range 6 months – 12 years) due to clinical symptoms including wheezing, stridor, cough, and respiratory infection. Antenatal history was unavailable for this group. All patients were female. (Table 1).

One patient from the antenatal group had a concurrent left congenital pulmonary airway malformation (CPAM), and postoperatively was found to have left bronchomalacia. Otherwise, there were no significant comorbidities in the patient group.

Common features were identified on the preoperative imaging. The chest X-rays uniformly showed mediastinal widening (Fig. 1). CT thorax with IV contrast was performed in all cases and had a number of common features (Fig. 2). All showed a posterior cystic structure with compression of the adjacent airway. The cyst was located between, and with significant separation of, the oesophagus and trachea. Each patient's images showed displacement or narrowing of an adjacent airway—uniformly the trachea was displaced anteriorly and compressed, with one patient also showing deviation of the carina to the right. Two patients also had compression of the left main bronchus. In three cases, the oesophagus was deviated to the right side.

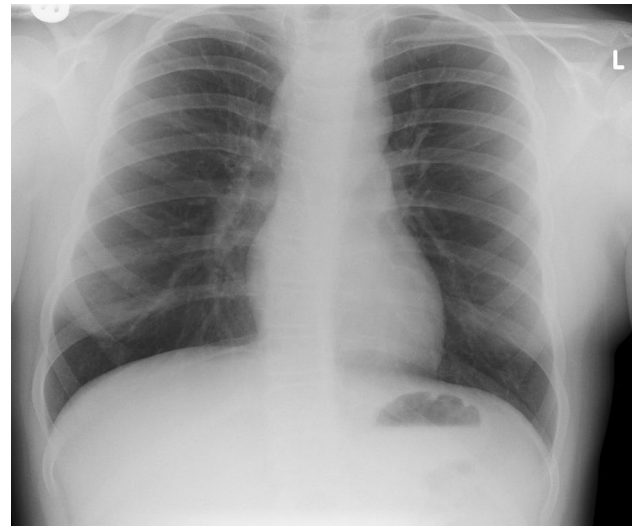


Fig. 1 Mediastinal widening evident on CXR

**Table 1** Patient demographics and outcomes

Patient	Gender	Age at diagnosis	Symptoms	Age at operation	Operation	Airway injury	Follow-up (months)
1	Female	Antenatal	N/A	1 week	Right thoracoscopic	No	27
2	Female	5 years	(LRTI, Left PTHX year prior to diagnosis) 1 year wheezing and chronic cough	5 years	Right thoracoscopic converted to thoracotomy	Yes	38
3	Female	Antenatal	Noisy breathing, ALTEs	4 weeks	Left thoracoscopic (concurrent CPAM)	Yes	16
4	Female	12 years	SOBOE, wheezing, cough	12 years	Left thoracoscopic	No	4
5	Female	6 months	Stridor, recurrent croup/bronchiolitis	7 months	Right thoracoscopic converted to thoracotomy	Yes	39

LRTI lower respiratory tract infection, PTHX pneumothorax, ALTE apparent life-threatening event, SOBOE shortness of breath on exertion

**Fig. 2** a–c CT images showing separation of trachea and oesophagus, compression of trachea and left main bronchus, and displacement of the airways



**Fig. 3** Fluoroscopic swallow showing deviation of the oesophagus to the right



Two patients also had a preoperative fluoroscopic contrast swallow performed. (Fig. 3)[Fig. 3 Fluoroscopic swallow showing deviation of oesophagus to right.] Both these confirmed deviation of the oesophagus to the right, with no communication between the cyst and oesophagus evident.

Age at operation was 1–4 weeks for the antenatally diagnosed group and 7 months to 12 years in the postnatal group. Two surgeons were involved, with four performed by the senior surgeon. Three cases were performed via right thoracoscopy, and two via left—one of these patients had a concurrent left CPAM that was resected at the same time.

In all cases, the FD was easily dissected from the oesophagus but densely adherent to an airway, usually the posterior wall of the trachea. In three cases, an injury was made in an airway—two in the posterior wall of the trachea and one in the left main bronchus. These were all due to dissection of the cyst off the airway at the site of a common wall and did not always occur at the site of maximal compression on preoperative imaging. One case occurred in the antenatally diagnosed group which was able to be repaired thoracoscopically—this was the injury to the left main bronchus. The opened bronchus was repaired with a continuous 5/0 poly-lactin suture with no obvious compromise to the calibre of the airway. The other two occurred in the postnatally diagnosed group and both required conversion to thoracotomy. In both cases, intraoperative manoeuvres were attempted to reduce air leak from the injuries and to prevent conversion to thoracotomy. These included advancement of endotracheal tube to below the level of the injury, intubation of the left

main bronchus, and allowing intermittent expansion of the operative-side lung.

The two other patients had uneventful thoracoscopic excision where the cyst could be dissected off the posterior wall of the trachea without injury. There was nothing specific to differentiate these two cases preoperatively from the three cases where injury occurred.

Operating time (OT) ranged from 83 to 300 min (mean 191.8) and length of stay (LOS) ranged from 3 to 42 days. Patient 3 required 42 days of admission because of requiring a prolonged admission with CPAP in the intensive care due to bronchomalacia of the left main bronchus and vocal cord palsy. This patient had concurrent FD and CPAM resection and thus skewed both the OT and LOS. The OT for this patient was 218 min. Excluding this patient, the mean LOS is 5 days.

Histology on all five specimens confirmed FD containing respiratory type epithelium. Cartilage was present in two specimens. All specimens were complete resections. This compares with the total cohort of 41 patients, of which 19 (46.3%) had respiratory epithelium, 12 (29.3%) had squamous epithelium, 6 (14.6%) had mixed squamous and respiratory, 3 (7.3%) had gastric mucosa and 1 (2.4%) was a neuroenteric cyst.

The median follow-up was 24.8 months and the range was 4–39 months. Follow-up in four cases was uneventful and patients were completely symptom free. Patient 3, as stated above, required prolonged admission for ventilation issues—at a recent follow-up (16 months) this patient is also symptom free and thriving with no evidence of ongoing vocal cord palsy clinically.

All follow-up chest X rays have been normal in appearance with complete resolution of mediastinal widening.

## Discussion

This case series highlights the rare, but important association of a common wall between superior mediastinal FD and adjacent airways. To our knowledge, this represents the largest series reported in the literature.

The common findings preoperatively were all in the imaging. Chest X-rays have limited predictive value except that they all had mediastinal widening which warranted further imaging for the diagnosis and operative planning. Computed tomography of the chest with intravenous contrast is essential to define the pathology and anatomical relationships of the FD. The common findings here were of a superior mediastinal cyst causing deviation of the trachea, narrowed airway lumen, and separation of the trachea and oesophagus. Luminal narrowing most commonly involved the posterior distal trachea and/or the left main bronchus. Importantly, the narrowed site was not always

the site of common wall. Fluoroscopy, when performed, showed oesophageal deviation to the right and lack of communication between the cyst and oesophagus. None of these features are diagnostic of a common wall with an airway, but as they were features in all the cases their presence should raise suspicion and necessary caution during resection.

Our study showed that thoracoscopic resection in these patients is feasible. While we had three airway injuries, one of these was repaired successfully thoracoscopically. In the other two cases, the anaesthetist could not maintain oxygenation and required repeated inflation of lung. Airway manoeuvres failed to control air leak from the injured site. Continuing to repair the airway injury and complete resection of the FD could not have been performed safely due to poor visibility and therefore conversion to thoracotomy was required. The patient that had the thoracoscopic repair was the most recent patient in this series and may be related to resection of the CPAM of the left upper lobe and the ability of the anaesthetist to maintain adequate oxygenation despite the small air leak from the defect.

There is scarce literature describing a common wall with an airway in FD, especially in the paediatric population. Michel et al. reported their experience with thoracoscopy in mediastinal cysts. They found three cases with a common wall with a bronchus and subsequently left the common wall in situ, while acknowledging the risk of recurrence [2]. Recurrence was also reported by Nobuhara et al.'s study which showed that marsupialisation had a high recurrence rate [1]. Our approach is to attempt complete resection to avoid this risk. With the improvement in thoracoscopic surgery experience and instrumentation in recent years, we believe that a complete resection is feasible. Airway injuries can be managed successfully either thoracoscopically or via open conversion.

Bratu et al. reported their series of FD in paediatric patients, comparing thoracotomy with thoracoscopic resection [3]. They included 39 patients of which 3 had a tracheal injury. It is not evident from the paper whether this corresponds to the presence of a common wall with the airway, as this is not mentioned. Tölg et al. reported their series of nine patients undergoing thoracotomy or thoracoscopy for bronchogenic cysts [4]. One of five thoracoscopic procedures required conversion due to difficult dissection of a bronchial lesion. It is unclear how many of these had common walls with an airway. Operating times in this series were shorter than ours.

Merry et al. note that two patients had postoperative air leaks following FD resection, and presume this was from trachea or bronchus injury, with resolution of air leak using an intercostal catheter [5]. However our experience of major airway air leak is that repair is required rather than simple drainage, making this presumption unlikely. Equally, it is

uncertain whether the air leaks were directly associated with the lesion or from collateral damage.

Martinod et al. describe an adult series of 20 patients resected thoroscopically [6]. In this, they mention one injury to the trachea, and once again, while assumed, it is uncertain if this represents a common wall.

With appropriate suspicion preoperatively based on our findings, the surgeon and anaesthetist can plan for the possibility of airway injury. In our current climate of improving non-technical skills and communication between teams, this is invaluable and may be the difference between completing the case thoroscopically rather than converting to thoracotomy. However, the authors advocate for conversion to thoracotomy if adequate ventilation cannot be maintained.

## Conclusion

The superior mediastinal foregut cysts situated between trachea and oesophagus can have a common wall with the tracheo-bronchial tree. While the risk of tracheo-bronchial injury cannot be predicted with certainty, radiological markers such as deviation of the trachea, narrowed airway lumen, and separation of the trachea and oesophagus should heighten the surgeon's awareness of this possibility. Bronchial injuries can be repaired thoroscopically provided adequate oxygenation can be maintained.

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## Compliance with ethical standards

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

**Ethical Approval** All procedures performed in studies involving human participants were in accordance with the ethical standards of the institution and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. For this type of study formal informed consent is not required.

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