

ORIGINAL ARTICLE

C. Merry · W. Spurbeck · T. E. Lobe

Resection of foregut-derived duplications by minimal-access surgery

Accepted: 21 December 1998

Abstract Eight children underwent minimal-access surgery (MAS) for duplications of foregut derivatives. The efficacy and safety of this approach are reviewed. The seven patients with mediastinal lesions had video-assisted thoracoscopic resection. One lesion presented as a subdiaphragmatic esophageal diverticulum, which was excised laparoscopically. Between March 1991 and October 1997, eight children were treated. Mean age was 27 months and mean weight was 11.4 kg. Mean operating time was 106 min, and mean postoperative hospital stay was 4.5 days (median = 2 days). Persistent air leaks occurred in two patients who had centrally-located bronchogenic cysts. One of these, who had undergone subtotal excision with laser photoablation of the remaining cyst mucosa, developed a recurrence that was excised at thoracotomy. We conclude that esophageal and bronchogenic cysts and duplications may be safely excised by MAS in children, with excellent cosmetic and functional outcome. Two technical points are noted: (1) a thoracostomy tube is required for central mediastinal lesions; and (2) complete excision is required to prevent recurrence.

Key words Thoracoscopy · Mediastinal cysts · Bronchogenic cysts · Esophageal duplications · Minimal-access surgery

Introduction

Developmental disorders of ventral budding of the lung primordium from the embryonic foregut may present as

cystic duplications or diverticuli of the bronchus or esophagus. Open resection by thoracotomy has been the only definitive treatment for these lesions in the past, but in view of their benign histology, a less-invasive approach is desirable. Growing experience and improved instrumentation for small patients have broadened the repertoire of procedures that can be accomplished by pediatric minimal-access surgery (MAS). Small children present unique problems in terms of size and anesthetic requirements for the MAS team.

Materials and methods

Seven mediastinal lesions were treated by video-assisted thoracoscopic excision. Single-lung ventilation with collapse and retraction of the ipsilateral lung was employed in all cases to give adequate surgical exposure. This was achieved where possible using selective left bronchial intubation with a double-lumen endotracheal tube. Smaller children presented a greater technical challenge, and a combination of endotracheal intubation and selective bronchial blockade using a Fogarty embolectomy catheter was employed. A pediatric fiberoptic flexible bronchoscope was useful in these maneuvers. Single-lung anesthesia was well tolerated.

Patients were positioned for thoracoscopy in the lateral position and rolled forward to facilitate access to the posterior mediastinum. A “bean bag” (Olympic Vac Pac, Olympic Medical, Seattle) was useful for holding small children in this position. All port sites were injected with lidocaine 1% with epinephrine 1:10,000 prior to incision, both to reduce blood loss and for postoperative analgesia. A 10-mm, 0° end-viewing telescope (Circon/ACMI, Stamford, CT) was introduced first. A three-chip camera (Circon/ACMI, Stamford) was used with two screens positioned so that the operating surgeon and assistants had a clear view. After incising the pleura, the surgeon’s finger was introduced to break any adhesions prior to carefully introducing the telescope and port under vision. The lung was collapsed by controlled CO₂ pneumothorax, insufflated via the camera port, and the lesion was visualized to plan the position of the operating port sites. Two or more 5-mm instrument ports were then introduced under endoscopic vision. The lesion was grasped with forceps and dissected from surrounding structures by a combination of sharp and blunt dissection using hook cautery and dissecting forceps. Feeding vessels could usually be cauterized; if not, endoscopic clips were applied. If the cyst remained intact, it was punctured and aspirated with a percutaneously-introduced needle to facilitate removal through a port site.

C. Merry (✉)¹ · W. Spurbeck · T. E. Lobe
University of Tennessee, Section of Pediatric Surgery,
777 Washington Avenue, Suite P220,
LeBonheur Children’s Medical Center,
Memphis, Tennessee, 38105, USA

Corresponding address:

¹Children’s Research centre,
Our Lady’s Hospital for Sick Children,
Crumlin, Dublin 12, Ireland

Patient 1 had a cyst that was closely adherent to the esophagus. To avoid esophageal injury subtotal excision was performed, with attempted ablation of the remaining mucosa with an Nd-YAG laser. A thoracostomy drain was inserted in all thoracoscopy patients except patient 6. Prior to removing the camera, the lung was re-expanded under endoscopic vision and the operative site inspected for air leaks by instillation of saline and positive-pressure ventilation. Wounds were closed with subcuticular absorbable sutures.

Patient 4 had an upper-abdominal, paraesophageal mass. Laparoscopy was performed utilizing a 10-mm camera introduced at the umbilicus, with 5-mm operating ports in the right and left upper quadrants. The lesion was found to be an esophageal diverticulum, which was removed by firing a linear stapler across its base, flush with the esophagus. Patient 5 had a bronchogenic cyst that was closely applied to, and had a common wall with, the right main bronchus. The bronchus was opened on removing the cyst, and was repaired by a continuous endoscopic suture.

Results

Eight children (five boys) were referred to our service from March 1991 to October 1997 for management of mediastinal cystic lesions in seven patients or an upper-abdominal parasophageal mass in one. They were 3.5 months to 8 years of age (mean 27 months) and weighed between 5.2 and 30 kg (mean 11.4 kg). Clinical details are given in Table 1.

Operating time ranged from 57 to 163 min (mean 106 min). Bleeding was minimal in all procedures and no patient required transfusion. A definitive cure was achieved in six of the eight patients with no complication, all of whom went home within 3 days of surgery. Narcotic requirements were low, most patients requiring no more than two doses.

Complications occurred in two patients. One bronchogenic cyst recurred early in the series; this was the only cyst that had not been completely excised, but had undergone subtotal excision and mucosal photoablation as described above. This patient also had a

postoperative air leak, and underwent open thoracotomy and removal of the recurrent cyst 1 year later. One other patient who did not have a chest tube placed at operation had a post operative air leak with pneumothorax, pneumomediastinum, and surgical emphysema. Two chest drains were inserted and the problem resolved. The patient was discharged on day 10.

The patients were admitted to the hospital on the morning of surgery, and the postoperative hospital stay was between 1 and 12 days (mean 4.5, median 2 days). Except for the two patients with postoperative air leaks, all patients were mobilized early and went home within 2 days of the operation. The cosmetic results of the surgery were excellent. Histologic diagnoses were bronchogenic cyst (4), mediastinal esophageal duplication (2), and, in the subdiaphragmatic esophageal diverticulum, tracheobronchial chondroepithelial remnant (1).

Discussion

Ventral budding of the lung primordia from the foregut occurs at 3 to 4 weeks of gestation. Aberrations in this process during this or subsequent stages of development may result in duplications of the esophagus or bronchi. Simple esophageal or bronchial duplication cysts are found most commonly in the mediastinum [1, 2]. Histologically, they may be differentiated by the presence of cartilage in the wall of a bronchial cyst and the presence of two well-defined muscle layers in an esophageal duplication [3]. The common embryologic origin of these structures is reflected by the fact that the lining epithelium of both lesions may be ciliated respiratory epithelium. The only subdiaphragmatic lesion in this series was interesting, because although anatomically it was an esophageal diverticulum, histologic examination showed cartilage in the wall, therefore classifying it as a bronchogenic remnant.

Table 1 Clinical characteristics of eight pediatric patients with foregut duplications treated by minimal-access surgery (L left, R right, RUL right upper lobe)

Patient	Age	Symptoms	Diagnosis	Operation	Notes
1	5 months	Pneumonia	Bronchogenic cyst, L Paratracheal	Thoracoscopic subtotal excision, laser to base	Aberrant RUL bronchus noted. Pneumothorax, cyst recurred
2	3.5 years	Pneumonia	Bronchogenic cyst	Thoracoscopic resection	
3	5 months	Recurrent pneumonia	R esophageal duplication	Thoracoscopic resection	
4	4 months	Dysphagia	Subdiaphragmatic esophageal diverticulum	Laparoscopic resection	Tracheo-bronchial chondroepithelial remnant
5	8 years	Pneumonia	Bronchogenic cyst, R main bronchus	Thoracoscopic resection	Endoscopic suture repair of bronchus
6	8 months	Recurrent pneumonia Croupy cough, "asthma"	Bronchogenic cyst, L main bronchus	Thoracoscopic resection	Air leak, chest tube 10 days
7	16 months	Pneumonia	Bronchogenic cyst, R main bronchus	Thoracoscopic resection	
8	3.5 months	Pneumonia, stridor	Subcarinal bronchogenic cyst	Thoracoscopic resection	

These cysts are histologically benign, and some controversy exists regarding the need to excise asymptomatic cysts in adult patients. Complete excision is the preferred treatment in children because of the high risk of obstructive respiratory problems, and also because these cysts do not regress spontaneously and occupy space destined for growing respiratory tissue [4]. Malignant transformation, although rare, has been described in children and adults [4]. All of our patients had symptoms directly related to the presence of the duplications, which is consistent with previous reports that show that the prevalence of symptoms is greatest among young infants with these lesions [5, 6].

Our experience demonstrates the risks of incomplete removal of mediastinal duplication cysts. In one patient early in the experience we did not achieve complete excision, but instead left a portion of the cyst attached to the esophagus. The mucosa appeared to have been completely obliterated by the Nd-YAG laser. Successful treatment by subtotal resection of bronchogenic cysts with ablation of the remaining cyst mucosa by silver nitrate [7] or the argon-beam coagulator (6) has been described by other authors. In our patient, however, the cyst recurred within 1 year. Computed tomography-guided aspiration of these lesions with or without injection of sclerosants has been described in adult patients [8, 9]. No long-term results are available, and this approach is probably not suitable for healthy pediatric patients because of the long-term uncertainty regarding recurrence, which is to be expected given the propensity of these lesions to recur if the lining mucosa is not excised.

Reports of thoroscopic removal of mediastinal cysts in adults [7, 10, 11] and children [12] have been limited to one or two cases. Our growing experience with pediatric MAS has allowed us to extend the use of this technology to younger infants. Refinements in technique such as the use of bronchial blockers in children too small to accept double-lumen endobronchial tubes [13], and the ongoing development of smaller endoscopes and instruments, have improved safety and surgical exposure.

The proximity of central bronchogenic cysts to the bronchial tree was emphasized by this experience. Two patients developed postoperative air leaks, presumed to be from the trachea or bronchus. We therefore recom-

mend chest-tube drainage in all cases of central bronchogenic cysts. This is in accordance with previously reported experience with pediatric bronchogenic cysts [5].

We conclude from this experience that many simple foregut duplications may be excised safely by MAS in children. Two technical points are noted: (1) central mediastinal lesions require chest-tube drainage postoperatively; and (2) complete excision is required to avoid recurrence.

References

1. Phillipart AI, Farmer DL (1998) Benign mediastinal cysts and tumors In: O'Neill JA, Rowe MI, Grosfeld JL, Fonkalsrud EW, Coran AG (eds) *Pediatric surgery*. Mosby-Yearbook, St. Louis, pp 839–851
2. Harmon CM, Coran AG (1998) Congenital anomalies of esophagus In: O'Neill JA, Rowe MI, Grosfeld JL, Fonkalsrud EW, Coran AG (eds) *Pediatric surgery*. Mosby-Yearbook, St. Louis, pp 941–967
3. Harvell JD, Macho JR, Klein HZ (1996) Isolated intra-abdominal esophageal cyst. *Am J Surg Pathol* 20: 476–479
4. Bolton JWR, Shahian DM (1992) Asymptomatic bronchogenic cysts: what is the best management? *Ann Thorac Surg* 53: 1134–1137
5. Di Lorenzo M, Collin PP, Vaillancourt R, et al (1989) Bronchogenic cysts. *J Pediatr Surg* 24: 988–991
6. Ribet ME, Copin MC, Gosselin B (1995) Bronchogenic cysts of the mediastinum. *J Thorac Cardiovasc Surg* 109: 1003–1010
7. Lewis RJ, Caccavale RJ, Sisler GE (1992) Imaged thoroscopic surgery: a new thoracic technique for resection of mediastinal cysts. *Ann Thorac Surg* 53: 318–320
8. Schwartz AR, Fishman EK, Wang KP (1986) Diagnosis and treatment of a bronchogenic cyst using transbronchial needle aspiration. *Thorax* 41: 326–327
9. Zimmer WD, Kamida CB, Mc Gough PF, et al (1986) Mediastinal duplication cyst. Percutaneous aspiration and cystography for diagnosis and treatment. *Chest* 90: 772–773
10. Watson DI, Britten-Jones R (1995) Thoroscopic excision of bronchogenic cyst of the esophagus. *Surg Endosc* 9: 824–825
11. Schwartz CD, Puschmann R, Echmayr J, et al (1995) Videoendoscopic procedures in thoracic surgery: technical aspects and report of removal of a mediastinal cyst. *Surg Laparosc Endosc* 5: 94–99
12. Rodgers BM, Harman PK, Johnson AM (1986) Bronchopulmonary foregut malformations. *Ann Surg* 203: 517–524
13. Shah RS, Varela PJ, Merry CM (1997) Selective ventilation using a Fogarty balloon catheter as a bronchial blocker: an essential technique for pediatric thoroscopic surgery. *Pediatr Endosurg Innov Techn* 1: 147–150