International Journal of Basic & Clinical Pharmacology
Anjana C et al. Int J Basic Clin Pharmacol. 2024 May;13(3):400-402
http://www.ijbcp.com
pISSN 2319-2003 | eISSN 2279-0780

DOI: https://dx.doi.org/10.18203/2319-2003.ijbcp20241002

Case Report

Video assisted thoracoscopy surgery a viable alternative for duplication cyst excision: a case report on foregut duplication cyst

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Received: 25 March 2024
Accepted: 11 April 2024

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ABSTRACT

Alimentary tract duplication is a relatively uncommon congenital abnormality. It can be found anywhere from the mouth to the anus and can be symptomatic or undetectable. Although congenital duplication can happen anywhere in the gastrointestinal tract, the ileum, oesophagus, and colon are the most common locations. The foregut, which includes the stomach, and first and second segments of the duodenum, is the source of one-third of all duplications. When foregut duplication involves the bronchial tree, respiratory symptoms are often present. Furthermore, excision through surgery is required for pathological examination of the resected specimen for definitive diagnosis. The most promising surgical approach for cyst excision has been thought to be open surgical resection through a posterolateral thoracotomy incision. However, video assisted thoracoscopy surgery has recently emerged as a viable surgical option for duplication cyst removal In paediatric surgery, video assisted thoracoscopy surgery has been established to facilitate the removal of mediastinal masses. However there is a limitation of research in the literature regarding the best surgical technique for oesophageal duplication cyst excision. In girls, foregut duplication is more common, especially in cases when bronchopulmonary involvement is present. While many duplications result in issues in early development, some are discovered by accident. When foregut duplication involves the bronchial tree, respiratory symptoms are often present. Haemoptysis and respiratory distress may be present in specific cases of the patient. Here, we described a 5-year-old male infant's case of foregut duplication with bronchial involvement and its management.

Keywords: Foregut duplication, Bronchial involvement, Lung, Video assisted thoracoscopy surgery, Oesophagus

INTRODUCTION

Alimentary tract duplication is a relatively uncommon congenital abnormality. It can be found anywhere from the mouth to the anus and can be symptomatic or undetectable. Although the majority of duplications are benign, the presence of ectopic gastric mucosa and the possibility of malignant degeneration are cause for concern.[1]

Although congenital duplication can happen anywhere in the gastrointestinal (GI) tract, the ileum, oesophagus, and colon are the most common locations. The foregut, which includes the stomach, and first and second segments of the duodenum, is the source of one-third of all duplications. An enteric duplication can be of varying sizes; it is usually solitary, spherical rather than tubular, and lined with alimentary canal mucosa.[2]

In girls, foregut duplication is more common, especially in cases when bronchopulmonary involvement is present. While many duplications result in issues in early development, some are discovered by accident. When foregut duplication involves the bronchial tree, respiratory symptoms are often present. Hemoptysis and respiratory distress may be present in specific cases of the patient.[3]

A frequently used method for evaluating and diagnosing duplication cysts is computed tomography, or CT. In

paediatric surgery, video assisted thoracoscopy surgery (VATS) has been established to facilitate the removal of mediastinal masses. But there is a limitation of research in the literature regarding the best surgical technique for oesophageal duplication cyst excision.[4] Here, we described a 5-year-old male infant's case of foregut duplication with bronchial involvement and its management.

CASE REPORT

A 5-month-old male baby was referred with small bilobed cystic lesion in the lower zone of right lung with mediastinal shifting. Baby had presented with breathing difficulty after birth and was diagnosed as pneumonia and treated from outside hospital following that a CT scan revealed mediastinal mass lesion which was suggestive of bronchogenic cyst. For further evaluation the baby was admitted in our hospital.

CT chest revealed two large ovoid cystic lesions with mild enhancing wall thickening in the right para vertebral region. These two cystic lesions were seen extending cranio-caudally from the level of cranial and extended inferiorly up to the distal end of esophagus. The possibilities suggested by the CT were the following: duplication cyst, type 1 pleuropulmonary blastoma and congenital pulmonary airway malformation (CPAM). In view of close relationship between esophagus and lower lobe of right lung the baby was diagnosed as foregut duplication cyst (FDC).

The surgery planned was VATS cyst excision and thoracic duct ligation. Preanesthetic follow up was done prior to the surgery and injection supacef was used for surgical prophylaxis and paracetamol for pain management. The baby was kept NPO from the night till the time of surgery. 1 unit of each PRBC and FFP was arranged as a precaution. The VATS was performed.

VATS procedure was done by 5 mm×3 ports placed, pleural queued with hook, cyst enucleated out after painted decompression using 16 g venflon. From oesophagus, separated using mono polar hook cautery, VATS findings showed large tense cystic lesions present along with esophagus 5×5 cm and 8×8 cm. Serosa continuous with esophagus non communicating. The excision biopsy from mediastinum showed consistent with a gastric duplication cyst. Hemostasis secured 20 fr ICD was placed. Strict monitoring of ICD was advised. Chyle (fat containing fluid) was predominantly draining through ICD. Thoracic duct injury chyle in the ICD was noticed after the procedure. Octreotide infusion was started along with Simyl MCT oil and medium chain TGL rich diet. Hence kept NPO and on octreotide to decrease the amount of chylothorax and continued on nutrition supplements as intravenous. Triglyceride levels were sent from the pleural fluid which showed predominantly a high value (784). On post op day 3 chyle was seen occasionally and on post op day 7 ICD was decreased to from 360 ml to approximately 10 ml. Octerotide was tapered and stopped as the following: 1 ml/hr followed by 0.7 ml/hr followed by 0.4 ml/hr. Planned for thoracic duct ligation+diaphragmatic fenestration/thoroctomy. Under sterile draping, right thoracotomy lung was retracted, mediastinal pleura was identified, dissected over the thoracic duct regional was taken care of nerves and the duct was ligated. Diaphragm fenestration was fenestrated and. The baby was extubated on post op day 1 hemodynamically stable maintained a 95% saturation at the room air and shifted to the ward. On post op day 3 chest X-ray was taken which showed normal and ICD was removed. A single fever spike was noted. Infectious diseases consultations advised to stop prolonged use of empiric antibiotics the fever spikes more likely to be post op systemic inflammatory response syndrome (SIRS). Blood cultures were advised to send and review with results. Cultures shows central associated blood stream infections (CLABSI) and the central line was removed as soon as possible. The baby was comfortable, afebrile, vitals were stable and was planned for discharged with oral fluconazole, paracetamol drops and multivitamin drops.

DISCUSSION

Embryologically, the foregut at the cranial end of the original gut gives rise to the pharynx, respiratory tract, oesophagus, stomach, and the first and proximal half of the second part of the duodenum.[5]

Parker introduced the concept of a duplication cyst during 1972. They stated duplicated the alimentary tract to create a cystic or spherical structure attached to a portion of the bowel, complete with a smooth muscle wall and mucous membrane similar to the alimentary canal. The presence of vertebral, spinal cord, and genitourinary malformations, as well as malrotation and intestinal atresia, indicated that alimentary tract duplications were the result of a complex process.[7]

Duplication cysts are usually connected by a smooth muscle wall and a blood supply. Size, location, type of duplication, and the presence of heterotopic mucosa are all associated with the symptoms. Alimentary tract duplications can cause a variety of symptoms such as abdominal distension, pain, obstruction, bleeding, respiratory compromise, or a painless mass. The majority (80%) present before the age of two years; prenatal ultrasonography can detect duplications as early as 16 weeks of gestation6. In our case, the patient was diagnosed at the antenatal scan done at the 23rd week of gestation.

After ileal duplication, oesophageal duplication is the most common type. The cervical portion accounts for 23% of all oesophageal duplications. They commonly manifest as enlarging neck masses or upper airway obstructive symptoms, but they can also be asymptomatic. Duplication cysts of the mid oesophagus account for 17% of all oesophageal duplications. Because of airway obstruction, they frequently present with respiratory distress.

The only effective treatment for oesophageal cysts is surgically removed. Furthermore, excision through surgery is required for pathological examination of the resected specimen for definitive diagnosis. The most promising surgical approach for cyst excision has been thought to be open surgical resection through a posterolateral thoracotomy incision. However, VATS has recently emerged as a viable surgical option for duplication cyst removal.[4,8]

The standard surgical approach for the excision of a foregut duplication cyst has been open surgical resection via a posterolateral thoracotomy. However, VATS is currently the preferred method. VATS has numerous advantages over open surgery, including reduced postoperative pain, earlier recovery, shorter hospital stays, and better cosmetic outcomes. In comparison to open thoracotomy, one of the major advantages is the much wider access to lesions, regardless of location. This is true even though computed tomography scans provide us with more or less reliable information about the location of the lesion. Another significant long-term advantage is the low risk of iatrogenic scoliosis, which has been reported to be as high as 22% in a series after thoracotomy.[9]

5 mm and 10 mm Hopkins rod-lens telescopes are used for VATS in children. Standard trocars (10, 5, and 3 mm) for laparoscopy can be used. Short trocars without valves, on the other hand, are specifically designed for VATS. Compared to hand instruments, these instruments can be inserted through an open incision and have a gentle "S" curve to conform to the shape of the thoracic cage. Newer robotic surgery methods for cyst excision have additionally been reported in the literature. VATS reduces postoperative pain, allows for earlier recovery and hospital discharge, and provides a better cosmetic outcome than open surgery. However, there are significant constraints in terms of availability, cost, and experience.[10]

CONCLUSION

Foregut duplication can have a variety of clinical presentations or can be asymptomatic and found incidentally. In our case, the major complication we faced was chylothorax after the VATS. Postoperative chylothorax remains a clinical challenge to the surgeon with substantial morbidity and risk of mortality. This was the major complication faced by our case along with electrolyte imbalances. Diaphragmatic fenestration as a mode of intervention is safe and efficacious in resolving prolonged chylothorax unresponsive to conservative measures. Diaphragmatic fenestrations are mainly used after cardiac surgeries. Here we use this method for a thoracoscopy surgery. There exists a need for further study to highlight the use of this procedure in other surgery cases.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Anjana C, Priya A, Yadav KAC. Video assisted thoracoscopy surgery a viable alternative for duplication cyst excision: a case report on foregut duplication cyst. Int J Basic Clin Pharmacol 2024;13:400-2.