Resection of esophageal duplication cyst in a 6-month child: a surgical challenge by video-assisted thoracoscopic surgery VATS

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Introduction and importance: Esophageal duplication cysts (EDCs) are rare congenital entities, with an incidence of 1:100,000\textsuperscript{[1,2]}. EDCs are categorized into 3 types: cystic, diverticular, and tubular\textsuperscript{[3]}. Moreover, the majority of patients are asymptomatic, and lesions are usually found incidentally on imaging. However, in the case of large esophageal cysts, dysphagia, retrosternal pain, and respiratory distress have been reported\textsuperscript{[4]}. Because of the potential of cyst rupture, infection, and bleeding, surgical resection is recommended at the time of finding both in symptomatic and asymptomatic patients\textsuperscript{[1,5,6]}. Previously, these cysts were removed via open surgery, but more recently, intrathoracic EDCs have been removed via robotic or laparoscopic procedures\textsuperscript{[6,7]}. Since the early 1990s, adults and children have undergone video-assisted thoracoscopic surgery (VATS) for the removal of mediastinal masses\textsuperscript{[8]}. Thoracoscopic in children is effective and safe\textsuperscript{[9]}. The treatment options vary based on the size and site of the cyst, which includes thoracoscopic VATS, endoscopic submucosal dissection, and thoracotomy. The main advantages of VATS are shorter hospital stay, patient comfort, decreased perioperative complications, and postoperative neath chest tube drainage\textsuperscript{[10,11]}. This study aimed to report an extremely rare case of EDC in a 6-month child. The cyst extended from the left side of the neck, below the hyoid bone, posterior to the esophagus, to the right hemi thorax’s para cardiac. The cyst was removed by VATS. To the best of our knowledge, this is the first challenge surgery ECDs in Iraq to treat EDCs in a 6-month by the VATS technique. The current paper has been written in the line with SCARE guidelines\textsuperscript{[12]}. Esophageal duplication cysts (EDCs) are rare congenital entities that occur at 3–4 weeks of gestation following failure to vacuolize the embryonic foregut, with an incidence of 1:100,000\textsuperscript{[1,2]}. EDCs are categorized into 3 types: cystic, diverticular, and tubular\textsuperscript{[3]}. Moreover, the majority of patients are asymptomatic, and lesions are usually found incidentally on imaging. However, in the case of large esophageal cysts, dysphagia, retrosternal pain, and respiratory distress have been reported\textsuperscript{[4]}. Because of the potential of cyst rupture, infection, and bleeding, surgical resection is recommended at the time of finding both in symptomatic and asymptomatic patients\textsuperscript{[1,5,6]}. Previously, these cysts were removed via open surgery, but more recently, intrathoracic EDCs have been removed via robotic or laparoscopic procedures\textsuperscript{[6,7]}. Since the early 1990s, adults and children have undergone video-assisted thoracoscopic surgery (VATS) for the removal of mediastinal masses\textsuperscript{[8]}. Thoracoscopic in children is effective and safe\textsuperscript{[9]}. The treatment options vary based on the size and site of the cyst, which includes thoracoscopic VATS, endoscopic submucosal dissection, and thoracotomy. The main advantages of VATS are shorter hospital stay, patient comfort, decreased perioperative complications, and postoperative neath chest tube drainage\textsuperscript{[10,11]}. This study aimed to report an extremely rare case of EDC in a 6-month child. The cyst extended from the left side of the neck, below the hyoid bone, posterior to the esophagus, to the right hemi thorax’s para cardiac. The cyst was removed by VATS. To the best of our knowledge, this is the first challenge surgery ECDs in Iraq to treat EDCs in a 6-month by the VATS technique. The current paper has been written in the line with SCARE guidelines\textsuperscript{[12]}.
Case presentation

Patient information

A 6-month-child male was admitted to a special Kurdistan hospital in Rania Kurdistan-Region, Iraq presented with shortness of breath, stridor associated with cough, repeated vomiting, and fever. Previously, she has been admitted twice to a pediatric hospital with dyspnea, cough, continuous stridor, chest retraction by dysphagia, and repeated vomiting. The pediatricians in Ranya maternity and pediatric teaching hospital consulted the thoracic department because he did not respond to medical treatment and rapidly deteriorated his condition.

Clinical findings and diagnostic assessment

On examination, the patients’ respiratory rate was 62 beats per minute (bpm) (reference range: 30–60 bpm), body temperature was 37.2°C (reference range: 36.2–37.5°C), blood pressure was 77/54 mm Hg (reference range: 65–90/45–65 mm Hg), and pulse rate was 110 bpm (reference range: 100–160 bpm) followed by chest retraction, inspiratory stridor, wheezy, chest, and SPO2 79% ambient air. Furthermore, laboratory test results are within normal limits. Chest x-ray showed well-defined soft tissue density on the right upper and middle lobe with normal lung field (Fig. 1), and computed tomography (CT) scan revealed that the cystic lesion measuring 7 × 3 cm extended from the thyroid cartilage downward to the posterior mediastinum and toward the right side of the chest causing compression of the trachea, esophagus, and right mainstem bronchus (Fig. 2). Finally, the patient was diagnosed with an EDC by histopathologic examination.

Therapeutic intervention

The patient underwent surgery under general anesthesia by uniportal VATS through the right fourth intercostal space was performed, and there was a giant cyst measuring 7.8 × 4 cm in size filled with milky white fluid. It was para-cardiac and connected to the upper and middle part of the esophagus; the cyst had a connection with the esophageal lumen. The cyst was resected, and an esophageal tear occurred because of the wall of the esophagus as part of the cyst. Nasogastric (Ryles) tube was put intraoperatively until 5 days postoperative, and removed on day 6, nothing has been taken per mouth (NPO) till 7 days postoperatively. He was on fluid, analgesic. Double antibiotic intravenously. The chest tube was kept in situ for 4 days, and then removed. The patient was observed for 5 more days and was discharged on day 11 postoperative. The surgery was successfully done without complications postoperatively.

Follow-up

The patient was discharged to the surgical department on day 11. The patient was healthy and asymptomatic last seen 5 months after surgery, and his SPO2 was 97% without oxygen.

Discussion

EDCs are rare congenital entities. The incidence estimated is 0.0122% live births with male prevalence. Approximately 80% is symptomatic and diagnosed in early childhood[13]. However, the diagnosis of EDCs remains a challenge due to the rarity of this condition, and difficult differential diagnoses such as bronchogenic cysts, pericardial cysts, mature cystic teratomas, neurogenic tumors, and congenital cystic adenomatoid malformations[14]. In addition, these cysts may have the same appearance by x-ray, ultrasonography, and even CT scan[15]. The most sensitive and accurate tool for making a diagnosis is magnetic resonance imaging. Moreover, our case was diagnosed by a CT scan of the chest and revealed that a cystic lesion measuring 7 × 3 cm extended from the thyroid cartilage downward to the posterior mediastinum and toward the right side of the chest causing compression of the trachea, esophagus, and right mainstem bronchus. Therefore, to confirm the clinical diagnosis a histologic investigation is ultimately required. There are 3 major characteristics: (1) it is lined with a squamous, cuboidal, or ciliated epithelium, (2) it is covered by 2 muscle layers, and (3) must be attached to or within the esophageal wall[16]. In this patient, the same criteria have appeared on histopathologic examination. The chest pain, cough, stridor, and feeding difficulties may occur depending on the size, location of EDC.

Nowadays, surgical excision is the standard approach for treating of EDCs at the time of finding both in symptomatic and asymptomatic patients[17]. Because of the risk of complications such as bleeding and aspiration[18]. Particularly, VATS is a less invasive procedure, effective, safe, and has a good clinical outcome compared with open surgical resection. Thoracoscopic, robotic-assisted surgery of mediastinal tumors and primary motility disorders has been increasingly experienced[19,20]. To remove these benign cysts, thoracoscopic surgery should be considered the gold standard[21]. The thoracoscopic VATS approach has many advantages, such as shorter hospital stay, patient comfort, decreased perioperative complications, and postoperative need chest tube drainage[19,21,22]. This study demonstrates that elective thoracoscopic VATS is safe, effective, minimal skin scaring, low morbidity rate, and is the gold standard option to treatment EDCs in children. In addition, pseudodiverticulum can occur if the muscles have not fully approximated and vagal nerve injury may occur if they are not preserved[23]. Recurrence is rare, particularly if the whole cyst has been removed[23]. The overall complication of the VATS approach is
Complication includes persistent leak air, pneumonia, deep venous thrombosis, vagus nerve paralysis, wound infection, and esophageal leak or pseudodiverticulum\(^2\). Our case has a great outcome similar to Petrović et al\(^3\) study.

We have performed a successful VATS resection of ECD in a 6-child male with excellent tolerance to this surgical technique. The postoperative period was uneventful, and the child was discharged within 11 days after the operation. Five months of follow-up revealed no symptoms and recurrence. Further studies are required to establish standard guideline management, intervention, and follow-up of duplication cysts in children.

**Conclusion**

EDC is an exceedingly rare entity. VATS is a less invasive procedure, effective, safe, and has a good clinical outcome compared with open surgical resection. We conclude that EDC should be maintained in the differential diagnosis of unexplained stridor, wheeze, and recurrent chest infection in children. Surgical resection is recommended at the time of finding, both in symptomatic and asymptomatic patients. This operation is technically possible, outcomes in minimal chest wall trauma, and has an effective outcome in young children.

**Ethical approval**

Ethical approval has been given by the ethics committee of our faculty.

**Sources of funding**

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

**Author contribution**

R.A.E.: conception and design, execution, analysis and interpretation of data, involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript. S.K.A.: conception and design, execution, analysis and interpretation of data, involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript. D.H.P.: involved in drafting the article, revised it critically for important intellectual content, read, interpretation of data and approved the final version of the manuscript. C.P.A.: involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript. S.A.R.: involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript. A.A.K.: involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript. S.B.M.: involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript. P.K.M.-A.: involved in drafting the article, revised it critically for important intellectual content, read and approved the final version of the manuscript.
Conflicts of interest disclosure

The authors declare that they have no financial conflict of interest with regard to the content of this report.

Research registration unique identifying number (UIN)

None.

Guarantor

Dr Rawand A. Essa, and Registered Nurse Sirwan K. Ahmed: accept full responsibility for the work and conduct of the study, had access to the data, and controlled the decision to publish.

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