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Esophageal duplication cyst in newborn



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Esophageal duplications are the second cause of posterior mediastinal mass in children, after neuronal tumors, with a prevalence of approximately 1/22.500 live births.¹ In newborns, the most common presenting symptoms are respiratory distress secondary to airways compression, dysphagia, and stridor. However, the majority of esophageal duplications are diagnosed incidentally in asymptomatic patients.¹ When diagnosis occurs prenatally, instrumental follow-up is recommended due to the risk of compression-induced hydrops and esophageal obstruction with polyhydramnios.¹

We reported a full-term male neonate presenting with a prenatal diagnosis of esophageal duplication. Prenatal ultrasound revealed a mediastinal cyst, which was both T1hypointense and T2-hyperintense on MRI, well-defined, and partially lobulated at the subcarinal level of the left paraesophageal region (Fig. 1). The patient was born via vaginal delivery without perinatal complications. He was asymptomatic having a 2920 g birth weight. While esophagram showed an intramural lesion, narrowing the lumen, in the

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anterior part of distal esophagus (Fig. S1), ultrasonography confirmed the septated submucosal anechoic cyst. At 6 days of age, the cyst was endoscopically fenestrated, with whitish viscous fluid drained into the esophageal lumen. The procedure was performed under general anesthesia, through the use of gastroscope 5 mm (GIF-XP180N, Olympus) and biopsy forceps 1.8 mm without cautery in order to reduce the risk of esophageal perforation. Additionally, ultrasound showed complete cyst emptying and therefore a gastric tube was placed. Antibiotic and antifungal prophylaxis was then administered. After 24 h, oral feeding was tolerated while gastric tube was removed. Consequently, he was discharged after 12 days. Four months later, esophagoscopy was performed showing normal result. Subsequently, the postoperative course was uneventful in a 30-month follow-up. Histology confirmed the diagnosis of esophageal duplication cyst.

Elective resection of esophageal duplications cyst is usually recommended not only due to potential complications (i.e., esophageal obstruction, dysphagia, airway compression, infections, ulceration-bleeding-perforation due to heterotopic gastric mucosa)¹ but also to lower complication rate of surgical treatment in asymptomatic rather than symptomatic patients. Endoscopic fenestration is minimally invasive.^{2,3} Taken into account the risk of postoperative esophageal stenosis, endoscopic fenestration

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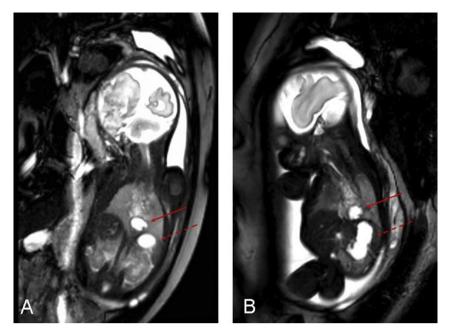


Figure 1 Prenatal magnetic resonance imaging of the esophageal duplication cyst. Coronal (A) and sagittal (B) T2-weighted MRI scans showing, at the subcarinal level of the left paraesophageal region, a well-defined, partially lobulated lesion with a hyperintense signal, measuring approximately $18 \times 13 \times 12$ mm and corresponding to the fluid-filled duplication cyst (solid arrow) with the subdiaphragmatic stomach (dotted arrow).

was therefore considered the best option in the present case in order to avoid the surgical resection through mediastinal approach. Nonetheless, potential complications of endoscopic fenestration are iatrogenic perforation and, due to the incomplete excision, relapse and/or malignant transformation, which has been anecdotally reported. In the pediatric literature, very few cases of endoscopic fenestration of esophageal cystic duplications are described,² and, to our knowledge, the present case is the youngest reported performed without cautery. Hence, further studies are needed to establish adequate timing for subsequent endoscopic/radiological surveillance and also to evaluate medium- and long-term outcomes.

Conflict of Interest

None.

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- Ethical approval: all procedures performed in this study were in accordance with the ethical standards of the Institutional and National Research Committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.
- Informed consent was obtained from all individual participants (children's parents) included in the study.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.pedneo.2019.08.007.

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