

Special issue • 2020 • vol.2 • 20-28 ESOPHAGEAL DUPLICATION: A CASE REPORT

Crivelli P.¹, Lampus M.L.², Sotgiu M.A.³, Conti M.² ¹AOU Sassari, Institute of Diagnostic Imaging 2, Italy ²Department of Clinical and Experimental Medicine, Institute of Diagnostic Imaging 2, University of Sassari, Italy ³Department of Biomedical Sciences, Medical School, University of Sassari, Sassari, Italy. Email: paocri2000@gmail.com

Abstract

Gastrointestinal tract duplications are rare congenital anomalies that can present diagnostic and therapeutic difficulties. Although they can occur anywhere from the mouth to the anus, they are commonly seen in to the ileum. We report imaging findings and present a case of esophageal duplication and a brief review the literature in order to contribute to increase the awareness of this rare disease.

Keywords: Gastrointestinal tract duplications, Rare congenital anomalies, Esophageal duplication, X-rays.

Introduction

Gastrointestinal tract duplications are rare congenital anomalies that can present diagnostic and therapeutic difficulties. Although they can occur anywhere from the mouth to the anus, they are commonly seen in to the ileum [1]. Intrathoracic foregut duplications, on the other hand, are even less common and present with a variety of symptoms, most commonly airway or esophageal obstruction. Esophageal cysts are most common and usually they are totally asymptomatic, they are a congenital malformation of the posterior primitive foregut and results from an aberration of the posterior division of the embryonic foregut at 3-4 weeks gestation.

We report imaging findings and present a case of esophageal duplication and a brief review the literature in order to contribute to increase the awareness of this rare disease.

Case presentation

A 28 year-old woman presented to the maternity hospital with contraction had a precipitous birth at 23th week of pregnancy. The female new born was conducted immediately in the unity of Intensive neonatal therapy for the cure. She has been intubated for mechanical ventilation and surfactant have been administered. She has suffered from left bleeding which hesitated intraventricular in hematoma and expansion of lateral ventricle. On the day she was born, supine thorax X-ray was performed to investigate lung conditions; this examination showed no respiratory distress, and no air in the abdomen (Figure 1). In the suspicion of esophageal atresia another x-ray was performed, with contrast enhancement (CE) which was administered by feeding tube (organe-iodate solution): imaging reveled that esophagus was contrasted in cranio-caudal sense, but also a contrast expansion in cervical-lateral area. (Figure 2). We decide to perform another x-ray, the day after, using another feeding tube which was replaced before the exam. Supine (Figure 3) and lateral (Figure 4) x-ray were performed before CE and only supine (Figure 5) scan after 20 minutes from CE administration.

Late acquisition revealed two parallels similar structures that originate both from pharynx to the stomach, located in the posterior mediastinum, with CE in the stomach. X-ray findings were suggestive for esophageal duplication. Furthermore, to excluding others gastro-intestinal tract anomalies, contrasted enhancement x-ray was performed after one week. CE was injected directly in the stomach (Figure 6) and we performed the acquisition after 40 minutes. X-ray showed regular opacification of the jejunum (Figure 7).

Discussion

Several hypothesis have been proposed to explain the origin of duplication in the gastrointestinal tract [2]. The split notochord theory postulated by Bentely and Smith is the most accepted explanation for foregut duplication. [1]. During the third week of intrauterine life the notochord fuses with the embryonic ectoderm and at about the fourth week, the endoderm separates from the notochord. Complete Esophageal duplication is a rare condition, usually it occurs in a partial form, as esophageal cist [3], a non -symptomatic form. Most foregut duplications are located in the thorax. Nonspecific symptoms of pain or discomfort often are secondary to their space-occupying effect mainly because enlargement occurring a relatively confined space (as with bronchogenic cysts and esophageal duplications in the posterior mediastinum) [4]. The rarity and variable presentation of this condition could lead to delay the diagnosis. In the abovementioned case the only sign that the newborn showed was no air in the abdomen, which first enables radiologist to suspect esophageal atresia. Esophageal atresia (EA) is the most common significant esophageal malformation, with an incidence of approximately 1 in 3500 live birth [5]. The diagnosis is clinico-radiological, mainly based on radiological findings on the x-ray imaging in an adequate scenario, as well as other gastro-duodenal condition [6, 7]. We would like to stress the importance of correct radiological management on the diagnosis of this rare conditions in premature child in order to start an appropriate pathway that may be decisive for the outcome.

References

- Akhter Nawaz, , Hilal Matta, , Alic Jacobsz, , Farhan Ansari, , Ghassan Al-Khouder, Ahmed Al-Salem. Intrathoracic foregut duplication cyst in neonates. Annals of Saudi Medecine, Volume 21, Issue 3-4, 2001.
- Rajeswarm R, Anupama C, La A et al. Case report: Antenatal MRI diagnosis of esophageal duplication cyst. Indian J Radiol Imaging. 2009 Feb; 19(1): 75–77.
- 3. D. Kolomainen, P. R. Hurley, S. R. Ebbs. Esophageal duplication cyst: case report and review of the literature. Dis Esophagus. 1998 Jan; 11(1):62-5.
- 4. Azzie, G., & Beasley, S. (2003). Diagnosis and treatment of foregut duplications. Seminars in Pediatric Surgery, 12(1), 46–54.
- El-Gohary, Y., Gittes, G. K., & Tovar, J. A. (2010). Congenital anomalies of the esophagus. Seminars in Pediatric Surgery, 19(3), 186–193.
- 6. Crivelli P, Carboni M, Montella RA, Amadu AM, Profili S, Conti M, Meloni GB. Gastroduodenal stenting: is still useful in the treatment of malignant obstruction? Radiol Med. 2017 Aug; 122(8):564-567.
- 7. Cuccurullo V, Cioce F, Sica A et al Gastroenteric diseases in the third millennium: A rational approach to optimal imaging technique and patient selection Recenti Progressi in Medicina 103(11):426-30



Figure 1. First x-ray showed no air in the abdomen.



Figure 2. Imaging shows esophageal cervical-duplication.



Figure 3. Supine scan before CE administration.



Figure 4. Lateral scan before CE administration.



Figure 5. Late acquisition after 20 minutes from CE administration.



Figure 6. Administration of CE directly into the stomach.



Figure 7. Late acquisition reveled normal anatomy of jejunum.