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Intraluminal esophageal teratoma in a neonate

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ABSTRACT

Teratomas are germ cell tumors containing one or more of the three embryonic layers (endoderm, mesoderm, and ectoderm). In neonates, the sacrococcygeal region is the most frequent site of origin, followed by the gonads and mediastinum. While most mediastinal teratomas are anterior, approximately 3–8% are found within the posterior mediastinum. Here we report the case of an infant who presented in the second week of life with frequent emesis followed by choking, dyspnea, and perioral cyanosis. An esophagram demonstrated a large pedunculated polypoid mass within the lumen of the esophagus. CT confirmed a 5 × 1.8 cm intraluminal mass with a stalk originating from the posterior wall of the esophagus at C6. Endoscopic retrieval was not possible due to its large size. The mass was operatively resected via a right-sided, 2 cm transverse cervical incision. An oblique esophagotomy was made; the polypoid mass was grasped and eviscerated using serial traction sutures. The mass was excised along with a rim of normal esophageal mucosa. Final pathology demonstrated a mature and solitary esophageal teratoma. To our knowledge, this is the first reported case of an intraluminal cervical esophageal teratoma, and the youngest patient in which a pedunculated esophageal polyp has been described.

Teratomas are the most common type of germ cell tumor, are composed of one or more of the three embryonic germ layers (endoderm, mesoderm, ectoderm), and contain tissue foreign to the anatomic site of origin. These benign tumors are categorized as mature or immature, depending on the presence of well-differentiated tissue. The most frequent site of origin is sacrococcygeal (45–60%), followed by tumors arising within the gonads (32%) and mediastinum (6%) [1]. Rarely, teratomas have been reported originating in the central nervous system, retroperitoneum, head, neck, stomach, liver, pericardium, and umbilical cord [2]. Teratomas arising from the posterior mediastinum are rare, representing 3–8% of all teratomas occurring within that compartment and less than 1% of all teratomas [3].

Large pedunculated polypoid lesions of the esophagus are a rare phenomenon. A review of the literature performed in 2006 identified only 110 cases of large (>5 cm) pedunculated polypoid lesions arising within the esophagus and hypopharynx. Of these, the most frequent histologic diagnoses were fibrovascular polyp (34%), fibroma (21%), lipoma (17%), and fibrolipoma (11%). Only 4 reported cases of polypoid malignancy within the esophagus were found [4]. A search of the literature revealed that the youngest patient ever reported to have had an esophageal polypoid lesion was a 6 week old infant with an esophageal hamartoma [5].

The following case report represents, to our knowledge, the first case of a teratoma arising from within the cervical esophagus, and the youngest patient in which a large pedunculated polypoid lesion of the esophagus has been described.

1. Case report

A two-week old girl presented with a one week history of coughing and respiratory distress after feedings. The patient was born to a healthy 27-year old G1P1 female by cesarean section for failure to progress at 39 weeks after a full term, uncomplicated pregnancy. The infant tolerated breast-feeding for the first week of life. However, she subsequently developed worsening non-bloody, non-bilious emesis after every feeding that was often followed by episodes of choking, coughing, respiratory difficulty and perioral

Abbreviations: CT, computed tomography; AFP, alpha fetoprotein; HCG, human chorionic gonadotropin; PDS, polydioxanone suture; GFAP, glial fibrillary acidic protein.

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cyanosis which improved with bulb suctioning. A modified barium swallow demonstrated passage of contrast material though the esophagus, although the cervical esophagus was narrowed and the esophageal body was dilated due to the presence of a large esophageal mass. Initial chest radiograph demonstrated compression and displacement of the trachea by an esophageal mass with normal aeration of bilateral lungs.

A formal esophagram demonstrated a pedunculated polypoid mass originating from the cervical esophagus, passing to the distal esophagus and ending just above the gastroesophageal junction (Fig. 1). CT confirmed a 5 × 1.8 cm solid intraluminal esophageal mass with a fibrous stalk originating from the posterior wall of the cervical esophagus at the level of the sixth cervical vertebral body (Fig. 2). Severe tracheal and bronchial compression was appreciated at the level of the carina.

A rigid esophagoscopy was initially performed to assess the mass and attempt endoscopic resection. However, the combination of the large diameter and long length of the mass prevented successful passage of the endoscopic snare and the procedure was terminated. The patient was taken to the operating room 48 h later for operative resection of the mass. A 2-cm horizontal incision was made on the right neck. The sternocleidomastoid muscle was identified and retracted laterally, and the carotid sheath, internal jugular vein and the common carotid artery were visualized and retracted laterally. The right recurrent laryngeal nerve was identified and preserved. A 2 cm segment of the cervical esophagus was exposed. The retroesophageal space was entered anterior to the prevertebral fascia. A right posterior lateral oblique esophagotomy was made. The polyp was identified, grasped and stay sutures were placed to aid retraction. The mass was exteriorized using gentle traction and serial traction sutures (Fig. 3). The stalk of the polyp, which was attached to the posterior esophagus, was resected along with a small rim of normal esophageal mucosa to obtain a negative resection margin. The muscular layer of the esophagus was not involved. The posterior esophageal mucosa was approximated with interrupted 5-0 PDS sutures and the esophagotomy was closed.

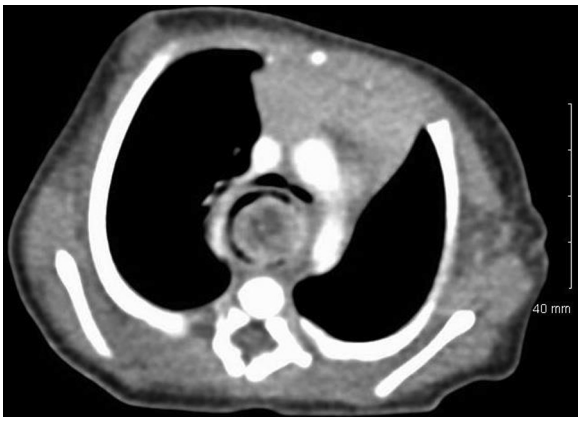

Fig. 2. Axial CT demonstrating an intraluminal esophageal mass compressing the trachea at the carina.

Final pathology demonstrated the mass to be a 4.5 cm esophageal teratoma, with cartilage, primitive gastrointestinal epithelium, multifocal cervical gastric heterotopia, smooth muscle, neuroglial tissue, adipose tissue, fibrovascular tissue, and inflammatory cells present (Fig. 4A–D). Immunohistochemistry stains were positive for synaptophysin, GFAP, desmin, S-100, and cytokeratin. Once the diagnosis had been confirmed as teratoma, serum alpha fetoprotein (AFP) was sent and found to be 1275, which was within normal limits for patient's age. Serum human chorionic gonadotropin (HCG) was less than 1.

The patient had an uneventful postoperative course. An esophagram on postoperative day seven demonstrated no leak, and the patient was started on a regular diet. She experienced dysphagia and required a gastrostomy tube for early enteral

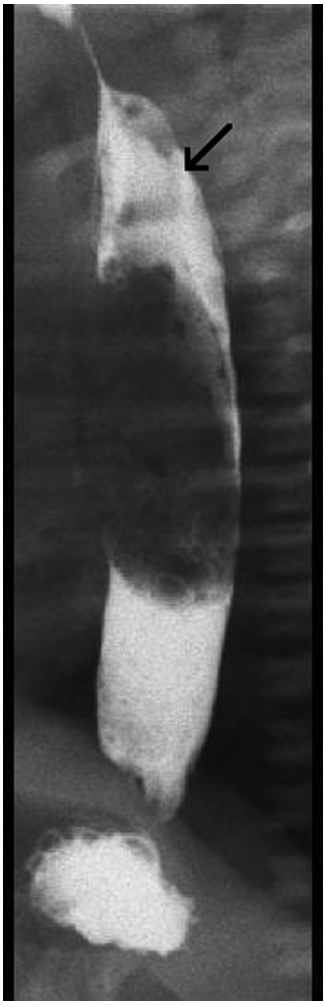

Fig. 1. Esophagram showing a pedunculated mass within the lumen of the esophagus connected by a thin stalk (arrow).

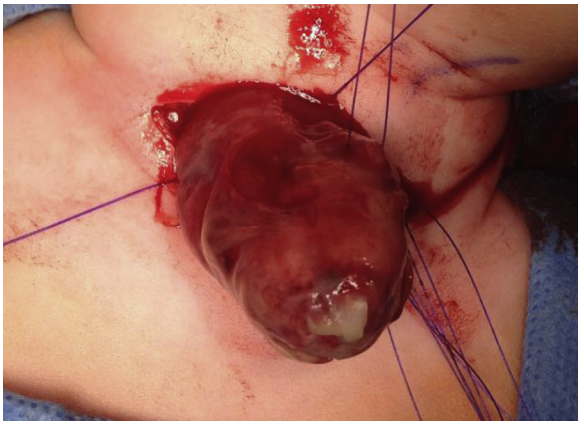

Fig. 3. Evisceration of the mass from the esophagus using serial traction sutures.