Teratoma at the Esophagogastric Junction in a Neonate

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Received: July 07, 2014;
Initial review: August 04, 2014;
Accepted; October 08, 2014

Background: Teratoma at the esophagogastric junction is extremely rare. **Case Report:** A 26-day-old male neonate who presented with vomiting and melena. **Observation:** Investigations revealed a mass at the esophagogastric junction, which was excised and confirmed to be a teratoma. **Outcome:** A fistula at the lower end of the esophagus and an esophageal hiatal hernia were observed as complications. **Message:** A careful surgical approach is warranted for a teratoma in esophagogastric junction, to avoid postoperative complications.

Keywords: *Neonate, Teratoma, Vomiting.*

A teratoma containing hair, teeth, bone and, very rarely, more complex organs or processes may occur at any site [1], but its location at the esophagogastric junction is extremely rare. We report a newborn with immature teratoma of the esophagogastric junction.

CASE REPORT

A 26-day-old male neonate presented with intermittent vomiting of approximately two weeks duration, and malena for one day. The general physical and systemic examinations were unremarkable. An initial contrast-enhanced computed tomography (CECT) scan showed upper esophageal dilation, unclear lower esophagus, and a large tumor mass between the esophagus and gastric fundus. The upper gastrointestinal barium study (***Fig.* 1*a***) revealed a large tumor mass in the lower esophagus involving the cardia and gastric fundus. The laboratory tests were within normal limits, including liver function tests, lactate dehydrogenase, ferritin, carcino-embryonic antigen, and neuron-specific enolase. Alpha fetoprotein was elevated with 5284 ng/mL. A biopsy was taken from the margins of the tumor mass, and immuno-histochemistry revealed an immature teratoma (***Fig.* 2**).The tumor mass was surgically excised. The post-operative period was uneventful. The infant was discharged home after 2 weeks. He was breastfeeding well and there was no vomiting.

At the three-month follow-up, alpha fetoprotein levels had reduced to normal, 2.83 ng/mL. At follow-ups at 6 months and 1 year, the boy was in a better condition, accepting normal food and his growth and development were normal. An upper gastrointestinal barium study at 1 month after surgery revealed a fistula located at the lower end of the esophagus, fundus pulled up to the diaphragm, and an esophageal hiatal hernia (***Fig.* 1*b***). The infant is being followed closely and the decision to repair the fistula is pending.

DISCUSSION

Teratomas most often occur in a para-axial location, in the midline from the brain to the sacral area, or in the gonads. Teratoma at the esophagus and stomach is extremely rare in children, accounting for less than 1% of all teratomas. To date, only one such case has been reported in the English literature [2]. Bernat, *et al.* [3] reported seven cases of benign esophageal tumors treated in a hospital from 1972 to 1990, only one of them was finally diagnosed as mature teratoma. He reported complete cure in all patients after surgery [3].

Gastric teratoma is believed to arise from the pluripotent cells of the gastric visceral wall [4]. The majority is benign and immature. The site of gastric teratoma is variable, most commonly arising from the greater curvature and posterior wall. The clinical features of esophagogastric junction teratoma in the neonate of the present case appeared at three weeks of life, with vomiting and melena, and without abdominal distension, constipation, or respiratory distress.

The melena could be due to mucosal bleeding at the tumor site. The CECT scan suggested the possibility of a leiomyosarcoma or neuroblastoma. The final diagnosis of teratoma can only be confirmed by histopathological examination of tissue. The alpha fetoprotein levels reflect the treatment response after excision, and may be of significance when chemotherapy is recommended in immature teratomas. Complete excision of the teratoma carries a good prognosis.

Few post-operative complications were noted in the present case. An inadequately dissected fundus may pull it into the chest, which could increase the anastomotic

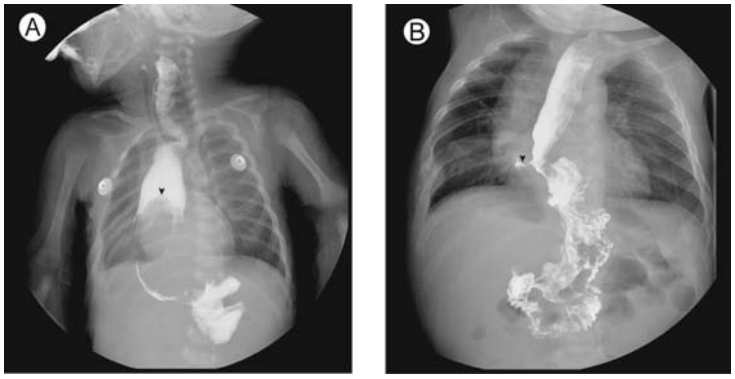

FIG. 1 *Upper gastrointestinal barium study showing (a) tumor mass in the lower esophagus involving the cardia and gastric fundus before surgery (arrows); (b) a fistula (arrows) located at the lower end of the esophagus, fundus being pulled up to the diaphragm, and an esophageal hiatal hernia at 1-month follow up.*

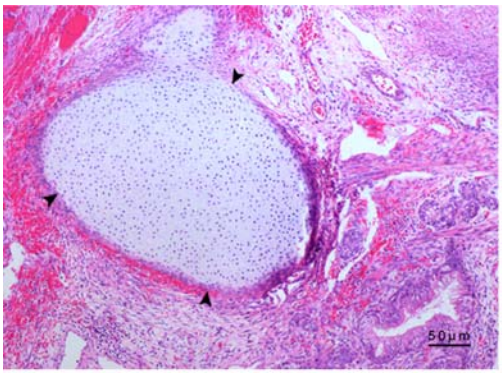

FIG. 2 *Immunohistochemistry reveals an immature teratoma (arrow shows the bone content). (See website for color image).*

tension between the esophagus and stomach, and lead to the postoperative fistula. On the other hand, the hiatal hernia could have been induced by an inadequately repaired esophageal hiatus. In addition, difficulties during surgery, especially exposing the mass, looking for a surgical entry point, and complete removal of the tumor may have added to the risk of postoperative complications.

While resecting such masses, full dissociation of the fundus and pulling it into the chest may reduce esophageal and gastric anastomotic tension and thereby avoid the occurrence of postoperative fistula.

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Erratum

In the article entitled "Newborn Screening for Congenital Hypothyroidism, Galactosemia and Biotinidase Deficiency in Uttar Pradesh, India" published in September 2014 issue of Indian Pediatrics on page nos. 701-705, following corrections are to be noted:

On page no. 704, under contributors: *"VG and KJ are both to be regarded as first authors"*

On page no. 704, funding section should be read as *"Department of Biotechnology, Government of India (BT/PR11395/SPD/24/335/2008) to Dr. Shubha Phadke."*

On page number 703, under discussion, first paragraph, 5th line, *"screened for hypothyroidism" should be read as "diagnosed to have CH"*. Full sentence should be read as "In a regional network catering to predominantly rural and low socioeconomic strata population, we were able to demonstrate more than 70% success in sampling, more than 80% success in recall and 90% success rate in follow-up of neonates diagnosed to have CH."

These corrections have already been carried out in the online version of the article at website of Indian Pediatrics *(www.indianpediatrics.net)*, but have not been carried out in the version of the article published at Springer website of the journal *(http://www.springer.com/medicine/pediatrics/journal/13312)*, as per individual publisher's policies.