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Recurrence of parasite in epigastric heteropagus

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ABSTRACT

Epigastric heteropagus (EH) refers to asymmetrically conjoined twins in whom an incomplete parasite is attached to the autosite's epigastric region from the xiphisternum to the umbilicus. We herein report an extremely rare case of EH in which unexpected rapid recurrence occurred 22 months after excision of the parasite. The recurrent mass comprised the intestinal wall with peristalsis-like movement, urogenital tissues, and hepatic tissue of the parasite. The recurrence was caused by a remnant of the parasite that extended from the autosite's xiphisternum to intra-abdominal cavity.

1. Introduction

Epigastric heteropagus (EH) is extremely rare. Only 71 cases of EH were reported in the English-language literature from 1946 to 2015. We previously described a male twin in a case of EH who had an omphalocele and double-outlet right ventricle [Sakaguchi 1]. At 22 months of age, his abdomen became rapidly distended due to recurrence of the parasite following surgical separation of the parasite in the neonatal period. Recurrence of the parasite in EH is exceedingly rare; only one case has been reported to date [2]. In the present report, we evaluate the pathogenesis of early recurrence in the autosite. We also discuss the importance of careful examination to identify any residual parasitic tissues in the abdominal cavity of the autosite at the time of excision of the parasite and closure of the omphalocele, which is highly associated with EH.

2. Case report

This case of EH was recently reported from our department in 2015 [Sakaguchi 1]. The parasite comprised a rudimentary head, and a lower limb was attached to the epigastrium of the male autosite, who had an omphalocele. Surgical separation of the parasite and silo placement for the omphalocele without opening of the abdominal cavity was performed at 4 days of age. Abdominal wall closure was performed at 10 days of age. The autosite underwent correction of a double-outlet right ventricle at 10 months of age [1].

An outpatient follow-up study performed at 18 months of age revealed no abnormal findings. At 22 months of age, he had developed a 2-month history of abdominal distension. However, his appetite was good and defecation was normal. Abdominal X-ray examination showed no signs of ileus (Fig. 1). We performed laboratory, ultrasonography, and computed tomography examinations to detect the cause of his abdominal distension. Tumor marker concentrations were not elevated (carcinoembryonic antigen, 1.3 ng/ml; beta human chorionic gonadotropin, 0.1 ng/ml; and alpha fetoprotein, 6.7 ng/ml). Ultrasonography revealed a mass resembling a luminal structure with peristaltic movement. Computed tomography showed that the mass extended from the anterior mediastinum to the abdominal cavity. The feeding vessel was thought to originate in the anterior mediastinum. The mass had no continuity with the intestinal canal (Fig. 2).

Resection of the mass was performed at 22 months of age. At the time of anesthetic induction, the luminal structure-like mass was moving slowly in his epigastric region. During laparotomy, the luminal structure was found to be adhered to the anterior surface of the liver. Because the luminal structure invaded the anterior mediastinum, repair of the diaphragm and pericardium was needed after the resection. No continuity between the luminal structure and intestine was found (Fig. 3).

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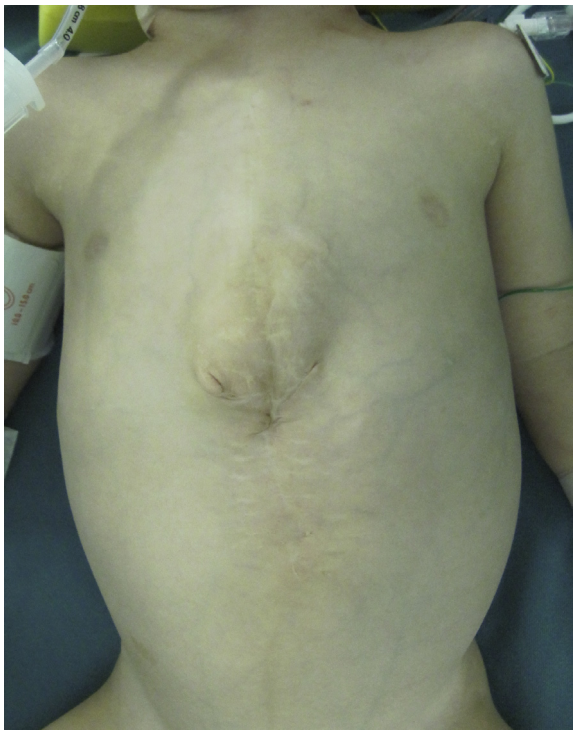

Fig. 1. Abdominal observation when he was 22 months old.

Pathologic examination of the specimen (Fig. 4A) revealed that the luminal structure comprised intestinal wall tissue with ganglion cells (Fig. 4B). The cystic lesion adhered to the anterior mediastinum was urogenital tissue (Fig. 4C). Hepatic tissue was also present, and immunohistochemical examination showed that it was anti-CK19 antibody-positive (Fig. 4D).

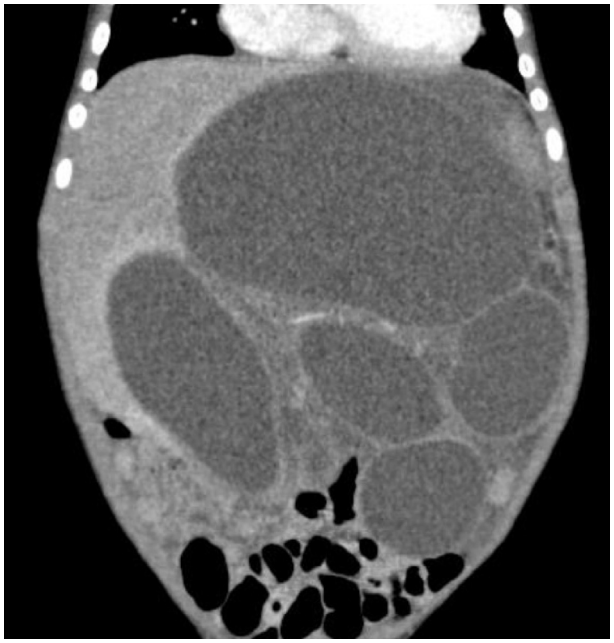

Fig. 2. CT showing a mimicking gut exists from anterior mediastinum to intraperitoneum.

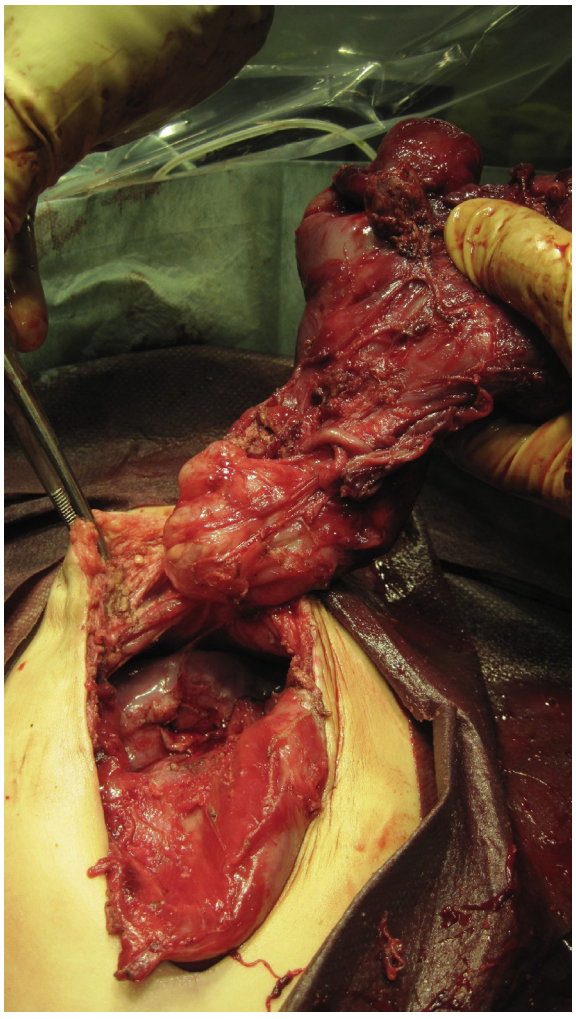

Fig. 3. The luminal structure invaded into the anterior mediastinum.

The patient was discharged on postoperative day 9. No recurrence had been detected by magnetic resonance imaging at the time of this writing.

3. Discussion

Seventy-one cases of EH were reported in the English-language literature from 1946 to 2015 except of stillbirth, abortion, and another site of parasite (Table 1). Recurrence of the parasite in cases of EH is very rare; the present case is only the second such report. The first case was reported by George et al. [2]; it was a recurrence of the case of EH reported by Surendran [12]. The recurrence interval was 17 years, and the parasitic remnant comprised intestinal and liver tissue and was located in the peritoneal cavity of the autosite. The recurrence interval in the present case was 22 months, and the parasitic remnant comprised intestinal, urinary tract, and liver tissue. Notably, the intestinal tissue extended from the autosite's xiphisternum to intra-abdominal cavity. In both cases, the recurrence occurred in the abdominal cavity of the autosite despite