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Case report

Mature Gastric Teratoma in a Female Infant Associated with Malrotation: A Case Report

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SUMMARY

Introduction: Gastric teratoma is a extremely infrequent tumor and accounts for less than 1% of all teratomas among children population with male predominance.
Case history: Herewith is described a case of mature gastric teratoma in a three months-old female infant associated with malrotation, hypotonia, torticollis and fusion of labia minoria. Due to the abdominal distension, the child was admitted to the Paediatric Surgery Department. On physical examination, a large lump was palpable, and laboratory findings were normal. Computed tomography (CT) showed a major heterogeneous solid formation within the right hemi-abdomen, extending to pelvis and crossing the midline. The infant underwent surgery and complete excision of tumor originating from lesser curvature of the stomach was made. Tumor was weighing 600 gr and was 10 x 14 x 8 cm large. The histopathological examination finding pointed to mature gastric teratoma. The post-operative course and further follow-up were eventless.
Conclusion: As soon as the diagnosis is made, the surgery is of paramount importance, especially in cases of immature teratomas, where malignant alteration is possible.

Keywords: **mature gastric teratoma, infant**

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INTRODUCTION

Gastric teratoma is an extremely infrequent tumor and accounts for less than 1% of all teratomas among children population (1). It usually occurs in infants (94%), especially in neonates, having exophytic growth in > 60%; about 30% have endophytic growth, while mixed growths are rare (2, 3). Gastric teratomas differ from other teratomas, with male predominance (> 95%), non-association with congenital anomalies and syndromes, apart from one reported case of gastric teratoma combined with peritoneal gliomatosis and the Beckwithe Wiedemann syndrome (2). Herewith is presented a case of mature gastric teratoma in a three-month-old female infant associated with malrotation, torticollis and fusion of the labia minoria.

CASE HISTORY

A female infant aged three months from a non-consanguineous marriage and without any recorded complications during pregnancy, fully delivered, has been initially treated due to the muscle hypotonia and left-sided torticollis in other institution. Due to the abdominal distension, although tolerating breast feeds, the asymptomatic child was referred to Pediatric Surgery Department. On physical examination, a considerable solid mass that took possession of the right hemiabdomen crossing the midline was palpable. All the laboratory findings were within regular range, including alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin (β-hCG). X-ray was inconclusive; still, the

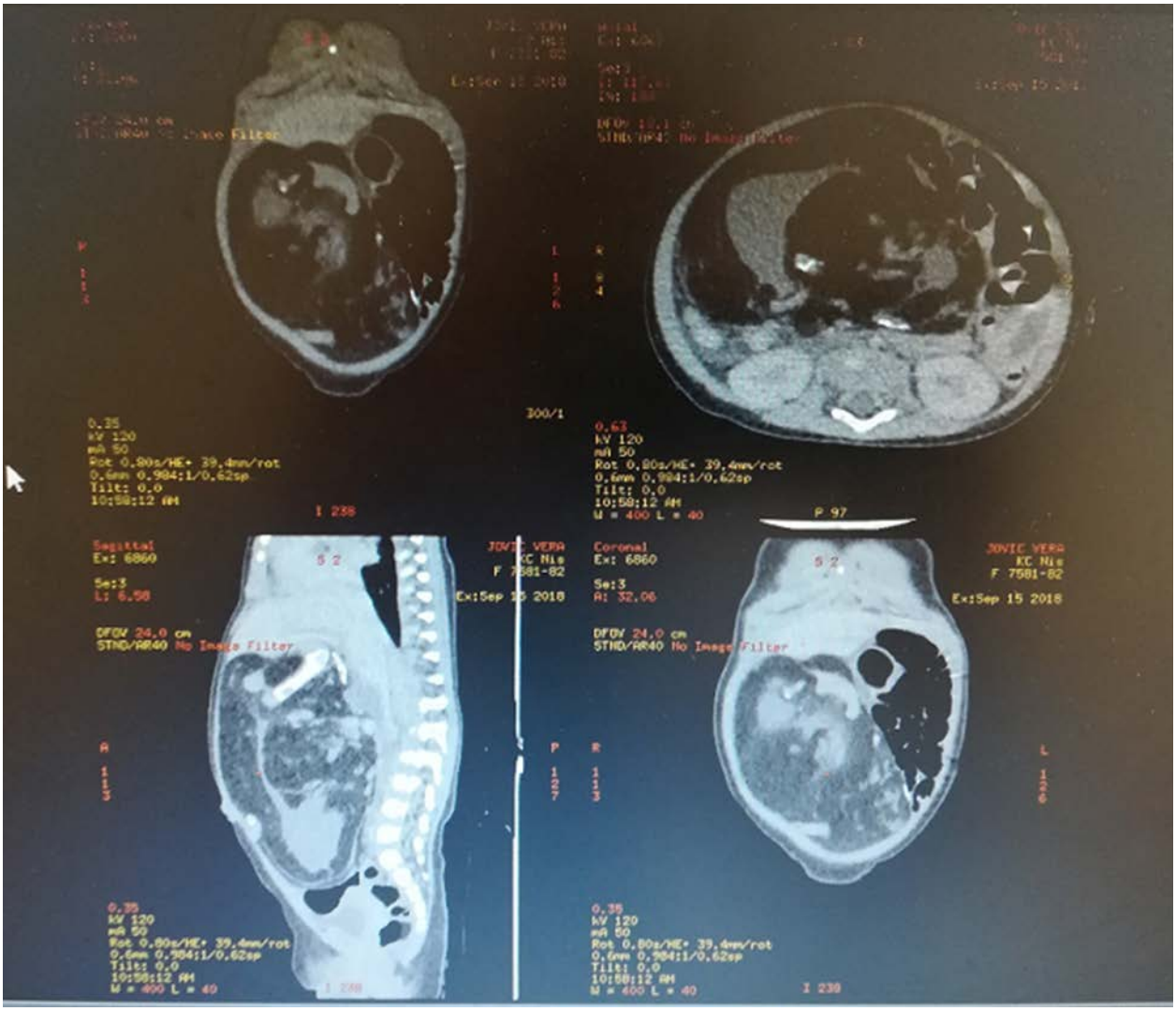

Figure 1. *Computed tomogrphy (CT) scans revealed a large tumor within the right hemiabdomen*

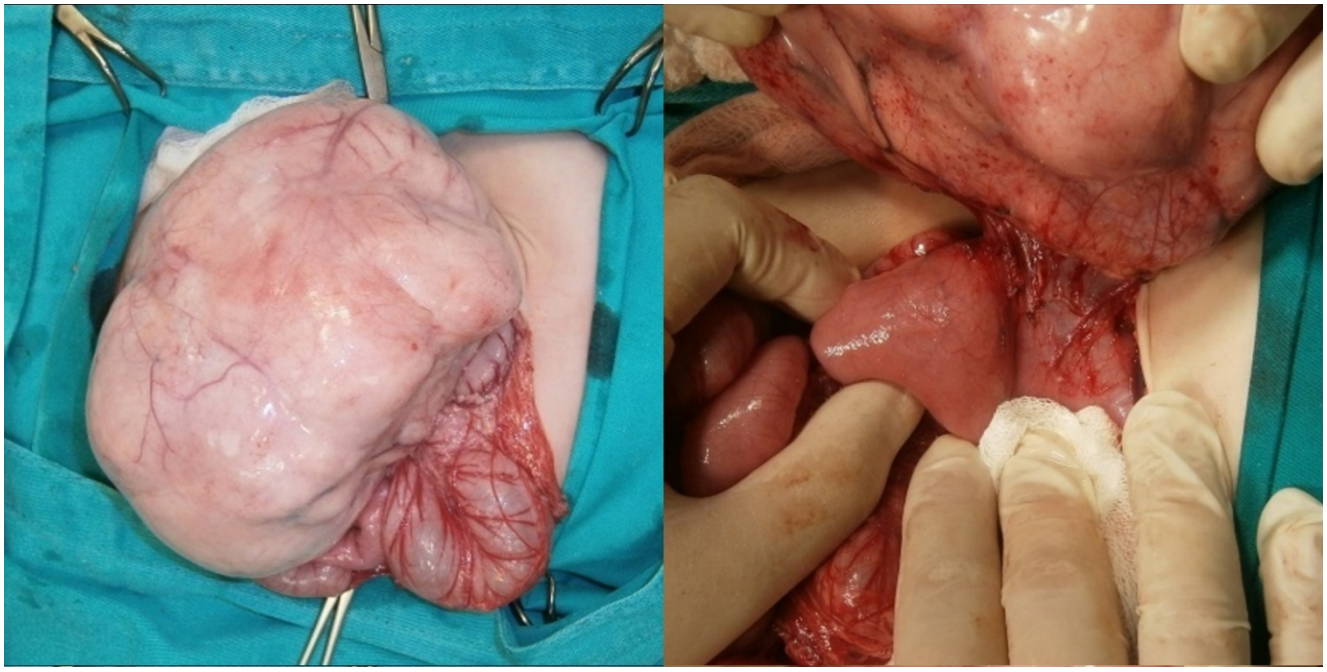

Figure 2. *Intraoperative findings of adherent gastric teratoma with exophytic growth on the lesser curvature of the stomach*

ultrasound examination revealed a massive firm complex tumorous formation. Computed tomography (CT) demonstrated a major heterogeneous solid tumor within the right hemi-abdomen extending to pelvis and crossing the midline, with dimension about 10 x 8 x 14 cm, with multiple calcifications, zones of necrosis and colliquation (Figure 1). The diagnosis of abdominal teratoma without certain site of origin was made preoperatively.

Exploratory laparotomy revealed a huge tumor which was completely extragastric, densely adherent to lesser curvature of the stomach (Figure 2); however, complete excision was performed. Additional intraoperative finding was malrotation, and due to this fact appendectomy was performed. Adhesions of labia minora were also present and separation was performed. The excised specimen measured 14 x 10 x 8 cm and weighed 600 gr. The postoperative course was with no complications and the infant was discharged on the 8th day after the surgery was performed. The histopathology was reported as mature teratoma.

DISCUSSION

Teratoma is the most iterative germ cell tumour in infancy and childhood with usual para-axial or midline presentation from the brain to the sacral area. Regarding its locations, the most frequent are sacrococcygeal teratomas (60 - 65%), gonadal (10 – 20 %), mediastinal (5 - 10%), presacral (5%) and rarely intracranial, retroperitoneal and cervical teratomas (4, 5). Gastric teratoma was first reported by Eusterman and Sentry in 1922 in a 31-year-old male and up to date closely 120 cases have been described in the English literature (6, 2).

The clinical findings vary depending on the size of the tumour, location and endogastric component. Gastric teratoma commonly presents as an palpable abdominal mass, abdominal distension, followed by anemia, gastrointestinal bleeding, haematemesis or malena, and vomiting (2, 3, 5). Respiratory difficulties such as multiple episodes of apnoea in newborns have been reported. Also, in neonates with large teratomas, premature labor or dystocia are possible (5). Unlike the majority of infants with gastric teratomas, this infant was female, and despite the large tumour it was completely asymptomatic. Abdominal distension and palpable abdominal mass with normal laboratory findings were reported. Mathur et al. reported in their study that teratomas originate from the greater curvature in 87.5% of cases, while in 12.5% derive from the lesser curvature (2). In this case, teratoma

originated from the lesser curvature.

It is very difficult to make an accurate diagnosis of gastric teratoma antenatally. Jeong et al. and Akram and al. both described prenatal ultrasound findings during the third trimester of pregnancy, which included an echogenic mass, bowel dilatation, intraperitoneal calcifications, ascites, hydroceles and polyhydramnios, mimicking meconium peritonitis (7, 8). Precise diagnosis is challenging in the neonatal period as differential diagnosis may be nephroblastoma, neuroblastoma, pancreatoblastoma and teratomas (8). In a 54-year retrospective study, Curtis identified other gastric tumors in children: gastric stromal tumors, lymphomas, adenocarcinomas, inflammatory myofibroblastic tumours, embryonal rhabdomyosarcoma, and hamartomas. The diagnosis of these tumours requires a higher index of suspicion (9). Preoperative diagnosis of teratoma with possible gastric origin was made after computed tomography was done.

In this case, a total excision was performed. However, Saleem reported other treatment modalities: excision with partial gastrectomy (54%), excision of a small part of serosa (mucosal sparing surgery) (23%), partial gastrectomy and limited transverse colectomy (15%), and total gastrectomy (8%) (10).

Histopathologically, depending on the differentiation of the neuroglial tissue, teratomas are divided into mature or immature (1). The most of gastric teratomas (61%) are identified as mature gastric teratomas which are benign. However, malignant alteration have been reported and for this reason, supervising of AFP and beta hCG in immature teratomas has an important role where chemotherapy is recommended (5, 10). Mature teratomas have an excellent prognosis due the fact that complete tumour excision with primary closure of gastric wall defect results in recurrence-free survival, and adjuvant chemotherapy and radiotherapy are not required (4).

CONCLUSION

Taking everything into consideration, it could be concluded that although being rare, gastric teratomas in neonates and infants are possible. As soon as the diagnosis is made, surgery is of paramount importance, especially in cases of immature teratomas, where malignant alteration is possible.

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Zreli gastrični teratom kod ženskog odojčeta sa malrotacijom creva: prikaz slučaja

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SAŽETAK

Uvod. Gastrični teratom je izuzetno redak tumor, koji je zastupljen u manje od 1% u pedijatrijskoj populaciji i češće se javlja kod dečaka.
Prikaz slučaja. Prikazan je zreli gastrični teratom kod tromesečnog ženskog odojčeta sa pridruženom a malrotacijom creva, hipotonijom muskulature, (fuzijom malih usmina, kao i pridruženim tortikolisom). Zbog abdominalne distenzije dete je hospitalizovano na Klinici za dečju hirurgiju i ortopediju. Fizikalnim pregledom detektovana je palpabilna tumorska masa u trbuhu, dok su laboratorijske analize bile u referentnim granicama. Kompjuterizovanom tomografijom (CT) verifikovana je velika, heterogena, solidna formacija u desnoj polovini trbuha, koja prelazi središnju liniju sa propagacijom do male karlice. Učinjena je hirurška intervencija sa kompletnom ekscizijom tumora, koji potiče od male kurvature želuca. Tumor je težio 600 gr, veličine 10 x 14 x 8 cm. Histopatološkim pregledom postavljena je dijagnoza zrelog gastričnog teratoma. Postoperativni tok i period praćenja bili su uredni.
Zaključak. Sprovođenje hirurške intervencije odmah po postavljanju dijagnoze je od izuzetnog značaja, zbog moguće maligne alteracije u slučaju nezrelog teratoma.

Ključne reči: **zreli gastrični teratom, odojče**