Gastric Teratoma—Unusual Cause of Neonatal Obstructive Jaundice: A Case Report

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Abstract The gastric teratoma is a very rare tumour that usually presents as an abdominal mass, gastrointestinal bleeding with/without features of gastric outlet obstruction. We report a rare case of immature gastric teratoma in a two months old female child who presented with abdominal lump and jaundice. The presentation was a diagnostic challenge and a dilemma, added by the fine needle aspiration cytology (FNAC) suggestive of small round cell tumor, favouring diagnosis of Wilms tumor over Neuroblastoma. Intra-operatively the tumor was found compressing upon the common bile duct (CBD). Final histopathological examination (HPE) revealed immature gastric teratoma Grade III. We wish to highlight this unusual presentation of gastric teratoma manifesting with obstructive jaundice and fallacy of FNAC in the diagnosis of teratomas.

Keywords Teratoma · Gastric · Immature teratoma

Introduction

Gastric teratoma is a rare tumor. First case was reported by Eusterman and Sentry in 1922 in a 31-year-old male [1]. A majority of the gastric teratomas occur in infants or neonates

with a striking male preponderance. Gastric teratomas generally present as a palpable abdominal mass. We report a rare case of immature gastric teratoma in a 2-month-old female infant presented with abdominal lump and jaundice. Presentation was a diagnostic challenge and dilemma, added by the fine needle aspiration cytology (FNAC) suggestive of small round cell tumor, favoring diagnosis of Wilms tumor over neuroblastoma.

Case Report

A 2-month-old female infant, a product of non-consanguineous marriage, uneventful pregnancy and full-term delivery by LSCS for non-progress of labor presented with palpable abdominal mass, vomiting, jaundice and poor acceptance of feed of 15 days duration (Fig. 2a). The mother noticed abdominal distension at birth but the infant remained asymptomatic. On physical examination, the infant was found to be malnourished, poorly built and *deeply icteric*. A large lump was palpable, occupying whole of the right side of abdomen and crossing the midline (Fig. 2a). Lab investigations showed hemoglobin 10.4 gm/dl, white blood cells 14,500/ml, platelets 78,000/ml, *total bilirubin 11.3 mg/dl, conjugated 9.8 mg/dl*, SGOT 176 U/l, SGPT 47 U/l and ALP 252 U/l. The renal parameters were within normal range.

Ultrasonography abdomen showed large, heterogeneous intra-abdominal mass showing calcification and increased vascularity on color Doppler (Fig. 1a). The mass appeared separate from the rest of the solid organs, suggesting neuroblastoma. Contrast enhanced computer tomography abdomen showed large, well circumscribed 12.8×10.3 cm heterogeneous mass, showing cystic changes with specks of calcification. The mass was crossing the midline, suggesting neuroblastoma. The rest of the abdominal viscera were unremarkable.

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Fig. 1 preoperative evaluation with (a) USG abdomen showing a large solid mass (arrows) with mixed echogenicity and focal cystic areas. (b) FNA cytology showing small round cells (black arrow) favouring Wilms tumor over Neuroblastoma. (400X LG stain)

FNA was performed and cytology was suggestive of a small round cell tumor, favoring Wilms tumor over neuroblastoma (Fig. 1b). Serum AFP was 54,000 ng/ml (normal range for this age, 46.5 ± 19.0 ng/ml) [2]. Other tumor markers were as follows: β -HCG 3.03 mIU/ml (<5.3 mIU/ml) and LDH 106 U/l (<450 U/l).

The infant was considered for neoadjuvant chemotherapy, but it was not given in view of deranged LFTs and lack of definitive tissue diagnosis, and the infant was planned for exploratory laparotomy. On exploration, there was $15 \times 20 \times 15$ cm sized mass arising from posterior wall of stomach and compressing the CBD. The tumor was adherent to the pancreatic neck and body and inferior surface of the liver. Total excision of the mass with distal pancreatectomy and Bilroth-I reconstruction was done (Fig. 2).

Final histopathological examination of resected specimen showed blastema and neuroepithelial tissue. Cartilage, adnexal structures and glands lined by both flattened and mucin secreting epithelium, lymphoid tissue, mesenchyma,

ectodermal tissue and nerve were noted (Fig. 3). The final diagnosis was gastric teratoma—immature type—grade 3.

Discussion

Gastric teratomas are found to be more commonly in male infants [3]. There are few reported cases of gastric teratoma in female infants [3]. Typical presenting symptoms are abdominal lump, distension and vomiting, but tumors with intramural extension causing gastrointestinal bleeding and gastric perforation have also been reported in literature [4]. The infant in this case presented with deep jaundice and lump abdomen, which is not a known feature of gastric teratoma.

Gastric teratomas generally arise from the greater curvature and posterior wall of the stomach [5]. Utsch et al. and Moriuchi et al. have reported cases of gastric teratoma originating from the lesser curvature [6]. In our case, tumor

Fig. 2 [Pre-operative & intra-operative pictures]- Large intra-abdominal mass occupying whole of abdomen (a) and same is confirmed after exploration (b). Tumour was arising from posterior and inferior wall of stomach, adjacent to pyloric antrum; depicted with Babcock forceps at 7'O clock (c) and was adherent to pancreatic neck and tail and was causing CBD compression (not in the photographs). Final tumour specimen with pyloric antrum at 6'O clock position (d)

Fig. 3 Final histological examination showing benign adenexa (a), cut section of nerve fibre (b), immature cartilage (c), tubules lined by flattened epithelium (d)

was originating from posterior wall of the stomach and was compressing upon the CBD.

Teratomas are embryonal neoplasms composed of derivatives of all three germinal cell layers. They can be classified into three types according to their histologic composition [7]. Mature teratoma consists of well-differentiated tissue, immature teratoma has varying degrees of immature fetal tissues and the malignant type contains at least one of the malignant germ cell elements. Immature teratomas are also graded from 1 to 3 by the amount of immature tissue contents, which are mainly neural elements, and by the degree of mitotic activity [8]. As a general approach, complete surgical excision is sufficient with grade 1 immature teratoma if AFP and β -HCG values are within normal age related ranges and there are no malignant germ cell elements [9].

Incomplete tumor resection, coccygeal or ovarian site and immaturity are known risk factors for both malignancy and relapse [9]. Although adjuvant therapies (i.e., chemotherapy or radiotherapy) are not recommended with completely resected grades 2 and 3 immature teratomas, optimal treatment in these groups is still controversial [10]. Marina et al. suggested that it seems safe to treat all patients with extra-gonadal immature teratomas by surgical excision followed by close observation, withholding chemotherapy until there is evidence of disease recurrence [10].

Elevated serum AFP levels may be the only alerting sign of the presence of malignant yolk sac component. Preoperative assessment of the tumor markers helps individualize treatment planning [7]. Evaluation of serum AFP levels in infants, especially in those younger than 8 months, is also an enigma because of wide physiologic variation [7]. It seems that the grading system is more useful in evaluating immature teratomas in adolescents and adults. In childhood, immature

teratomas have a better prognosis [7]. Although the value of AFP cannot be denied in the diagnosis and follow-up of germ cell tumors, in immature gastric teratoma it does not seem predictive for malignant potential. More case reports with well-defined histologic grading and AFP values will clarify the controversy in the therapeutic approach.

Conclusion

Authors wish to report a case of immature gastric teratoma with obstructive jaundice, which is a very unusual occurrence. Unusual presentation of an infant with lump abdomen and obstructive jaundice was a diagnostic challenge. Surgeons dealing with delicate patients should keep in mind this kind of rare presentation and fallacy of FNAC in the diagnosis. An accurate diagnosis of this tumor and differentiation from other common intra-abdominal neoplasms are essential in spite of its rarity. The role of AFP in diagnosis and adjuvant chemotherapy remains yet to be clearly defined.

Conflict of Interest None.

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