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Gastric Teratoma in a Neonate: A Rare Case Report

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Abstract: ***Introduction: Gastric teratoma in neonatal period, accounts for less than 1 % of all teratomas with less than 200 cases being reported in literature. It is an extragonadal germ cell tumors composed of all the 3 layers – ectoderm, mesoderm, endoderm. Stomach is an unusual site, with presentation seen as early as infancy. Case Presentation: Day of life 9, full term, male child was referred with chief complaint of gross abdominal distention gradually increasing from birth, with a visible epigastric lump and left inguinal hernia. It was a huge mass occupying epigastric and left hypochondriac region measuring 10*8*5 cm, well defined, variegated surface, moving with respiration. CT scan reported a solid, cystic mass (10.5 × 7.8 × 6.2cm), occupying mainly left upper quadrant, containing septations with coarse calcifications. The diagnosis of intrabadominal mass was established, considering the differentials of mesenteric cyst, teratoma, gastric lymphoma. The tumor was en-bloc resected, along with the part of the gastric wall. Postoperative course was uneventful. On histo-pathological examination, it was an immature grade 2 teratoma and complete remission was seen by en-masse resection. Conclusion: Gastric teratoma is a rare neoplasm, with resection being it's, the-line of management, owing to its benign course. Given the rarity of this case, this case report underscores the importance of considering it in its differentials.***

Keywords: neonate, gastric teratoma, case report

1. Introduction

Gastric teratoma is a rare paediatric extra-gonadal germ cell tumor, especially in the neonatal period. It accounts for less than 1% of all teratomas in infants and children, with only around one hundred and two cases reported in the literature till date. The most common site of teratomas in the neonatal age group is in the sacro coccygeal region, followed by the gonadal region, while the stomach is the rarest site. Despite being rare, it's the most common site for gastrointestinal teratomas, with male pre-ponderance [1].

We present a case of a neonatal gastric teratoma, who underwent a complete en bloc resection. Our case is an addition to the few gastric teratomas reported in thw world literature.

This manuscript was prepared following CARE guidelines.

2. Case Presentation

A full-term-day of life nine-male child, born out of normal vaginal delivery, referred from peripheral district hospital in view of progressive abdominal distention with feed intolerance, with intermittent episodes of non-projectile vomiting, with a left inguinal hernia since birth. The distention was seen gradually progressing till the present day. No bowel bladder complaints reported by parents, and no history of per rectal bleed.

On examination – neonate was alert, with a heart rate of 128 beats / minute, respiratory rate of 35breaths /minute, afebrile, oxyen saturation of 98%.

On inspection – grossly distended globular shaped abdomen was seen, with a a vague lump was visible in the epigastric region. Palpatory findings revealed a grossly distended abdomen with a firm, well defined, irregular mass extendig from epigastric till left hypochondriac region. (Figure -1).

Hematological investigations were suggestive of an haemoglobin of 14.5g/dL and total leucocyte count of 15000/cm ^{3}. Liver and renal function test were within normal limits. X-ray abdomen showed a mass effect with displacement of bowel gas shadowcdown wards, with diffuse calcifications in the left hypochondriac region. Ultrasonography was suggestive of a multicystic mass with mixed echogenicity, without and free fluid in the peritoneal cavity

CT was suggestive of solid cystic heterogenous lesion occupying the upper quadrant of the abdomen measuring 10*8*7 cm, extending from left hypochondriac reaching till umbilicus, with intralesional calcifications present, thereby raising a suspicion of gastric teratoma (Fig.1). The organ of origin of the lesion could not be traced (Figure-2inclusive). Oncological markers like alpha-fetoprotein, beta hcg and lactate dehydrogenase were sent, in which alpha fetoprotein was 40, 000 IU/L, B-HCG <2ng/dl, LDH – 122 ng/dl (within normal range)

The decision for surgical intervention with an exploratory laparotomy was done.

The abdomen was explored by a midline incision, and intra-operatively, a heterogeneous, multi-cystic mass occupying the center of the abdomen was noted. On dissecting further, its origin was traced to the lesser curvature of the stomach, which was an exo-gastric growth. The tumor mass was resected en-bloc along with a part of lesser curvature of the stomach as a margin, thus leaving a rent of about 2 cm in the stomach. It was closed in a double layer using vicryl 4-0 and silk 4-0 sutures.

The resected specimen was a fleshy mass measuring approximately 10*7*6 cm cm. Grossly, it had a variegated red-gray cut surface with areas of cartilage and calcifications, consistent with a teratoma (Figure 5). No obvious gross invasion of the neighbouring organs was seen; the margins were complete.

The post-operative course was uneventful, and feeding was started on post-operative day 5. The patient was discharged on post-operative day 7.

3. Discussion

Most of the teratomas upto 60%seen in sacrococcygeal region, followed by mediatstinal (11.7%), with 10 %in gonadal and 5 %in retroperitoneal, with less than 5 %accounting for cervical, cranial and less than 1 %being gastric. Owing to the paediatric age group especially in neonate, infants and toddlers – extragonadal germ cell tumors are seen unlike gonadal involvement in adults [2].

Tracing back to history, the first report of gastric teratoma was by Eustermann and Sentry in 1922. They have a male predilection and lesser curvature being the commonest site, or the posterior wall [3].

Gastric teratomas are generally observed as a exo phytic growth (>60%), endophytic (30%), with a very few presenting as mixed thereby leading to a varied set of presentations.

The most common form of presentation is a palpable abdominal lump with progressive distention from day of life one. A few neonates may also experience feed intolerance owing to gastric compression secondary to a large, rapidly growing tumor. In a few cases, these tumors have known to cause gastrointestinal bleeding in cases of endophytic growth or respiratory compromise due to diaphragmatic elevation [4]

On imaging its usually a heterogenous mass, with solid and /or cystic components with calcification / fat may or may not be seen. The other differentials include a lymphangioma, neuroblastoma or a benign cystic lesion, thus posing a difficulty in diagnosis. Definitive diagnosis always rests on surgical excision with a histopathological confirmation [4].

In the above case, we faced the diagnostic challenge initially as imaging was inconclusive. Based on the clinical findings, differentials of mesoblastic nephroma, massive neuroblastoma, retroperitoneal teratoma, and meconium pseudocyst owing to the cystic nature of the lesion, could not be ruled out completely.

The differential diagnosis for a neonatal abdominal mass is broad, including lymphatic malformations, neuroblastoma, and other germ cell tumors [4].

Similarly, Li and Wei reported a neonatal gastric teratoma misdiagnosed as a hemorrhagic lymphangioma due to its predominantly cystic nature [5].

Also, in this case, no clear gastric bubble was identified. Lack of a gastric bubble may not help differentiate a gastric teratoma from a meconium pseudocyst, given that meconium pseudocysts can demonstrate mass effect on normal fetal structures and theoretically make identification of a gastric bubble difficult.

In the setting of a multicystic complex mass in the left upper quadrant, one should attempt to identify a stomach bubble because these tumors can exhibit endogastric or exogastric growth [6]

Ultimately, a definitive diagnosis requires histopathological examination.

Thus histopathologically, there are two hypothesis regarding the origin of germ cell tumors, which include Tealium's theory, wherein tumors arising from primordial germ cells migrate in an aberrant fashion thus undergoing malignant transformation, thereby delineating the proclivity for midline location of these tumors. Alternative embryonal theory states that extragonadal germ cell tumors arise from abnormally migrated pluripotent embryonic cells.

Among gastric teratomas, mature ones composed of fully differentiated tissue elements are more common, and only a dozen (approximately) cases of immature gastric teratomas have been described to date. Grading of immature teratoma may not always envisage recurrence as immaturity does not correlate with malignant potential [7].

The presence of rarely described elements like immature renal and pulmonary tissue also does not alter the prognosis.

However, on analysis of cases presenting with recurrences, a yolk sac component was identified as a risk factor with instances of peritoneal gliomatosis [8].

The yolk sac component in a tumor can also be determined by serum alpha-fetoprotein levels. Though alpha feto protein levels are high in neonate, age adjusted levels are found to

be significantly higher when the tumor has a yolk sac component, which also warrants recurrence even after complete surgical excision. Serial estimation of serum alpha fetoprotein levels and its immunostaining can also be of aid in such cases [8].

In our case, it was a Grade II immature teratoma, which lacked a yolk sac or other malignant components, portending a favorable prognosis.

Long-term outcomes are excellent when complete resection is achieved. Recurrences are rare but have been documented in immature cases, often within two years of surgery; hence, vigilant follow-up with serial alpha feto protein levels and imaging is advised every three months for the first year, then biannually for two years, and annually thereafter for up to five to seven years [9].

Lifelong surveillance may be considered, given reports of late relapse decades after initial resection

4. Conclusion

Any neonate presenting with an upper abdominal lump with solid and cystic components on imaging, along with fat and calcification, gastric teratoma should be considered as a differential diagnosis.

Its benign course should be taken into consideration, even with immature components present in the tumor, thus surgical resection is the only best possible management for these malignant-looking tumors.

Statements

1) "Informed consent was obtained from the parents"
2) "All authors attest that they meet the current ICMJE criteria for Authorship.

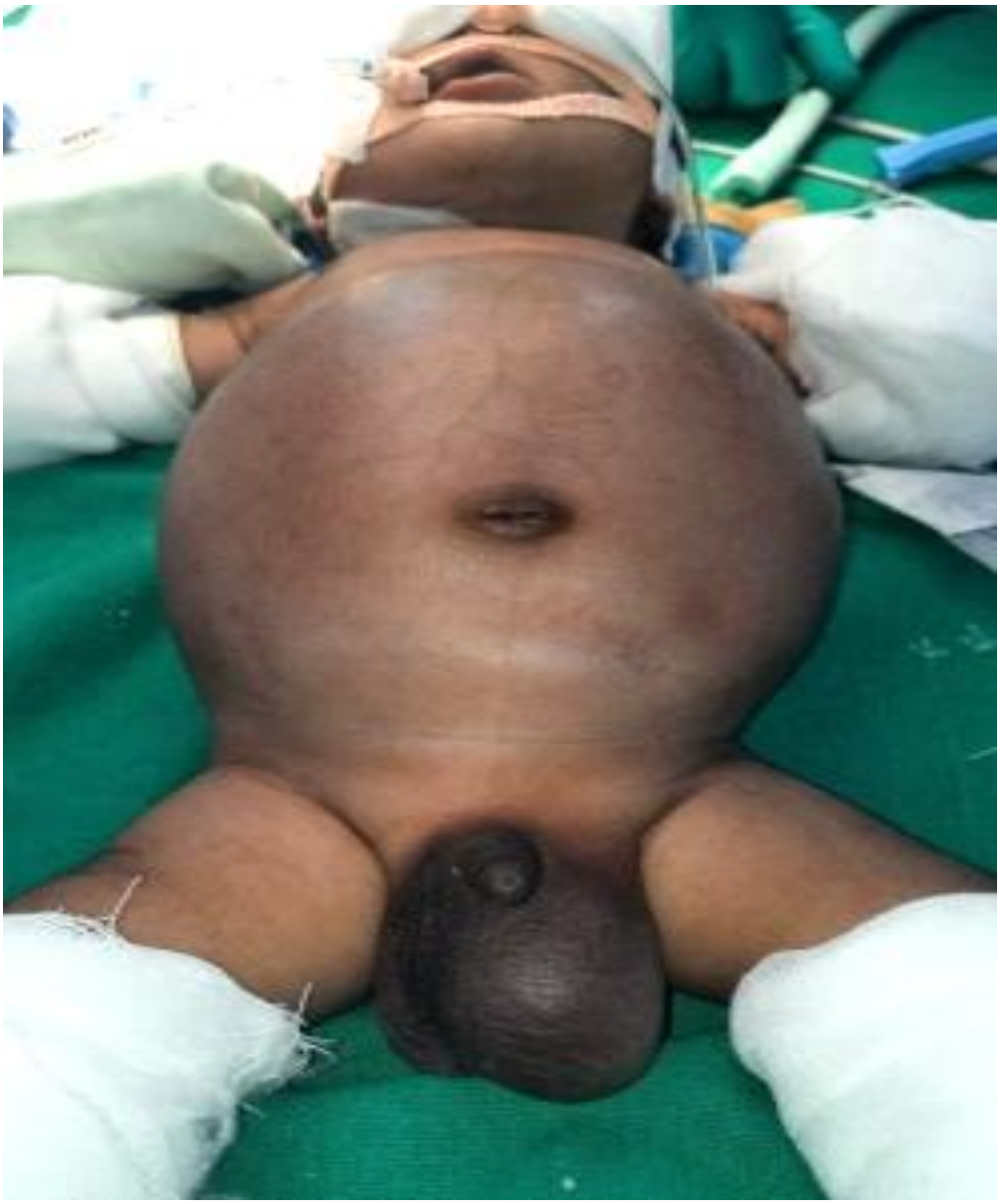

Figure 1: Clinical Image – gross abdominal distention with a left inguinal hernia

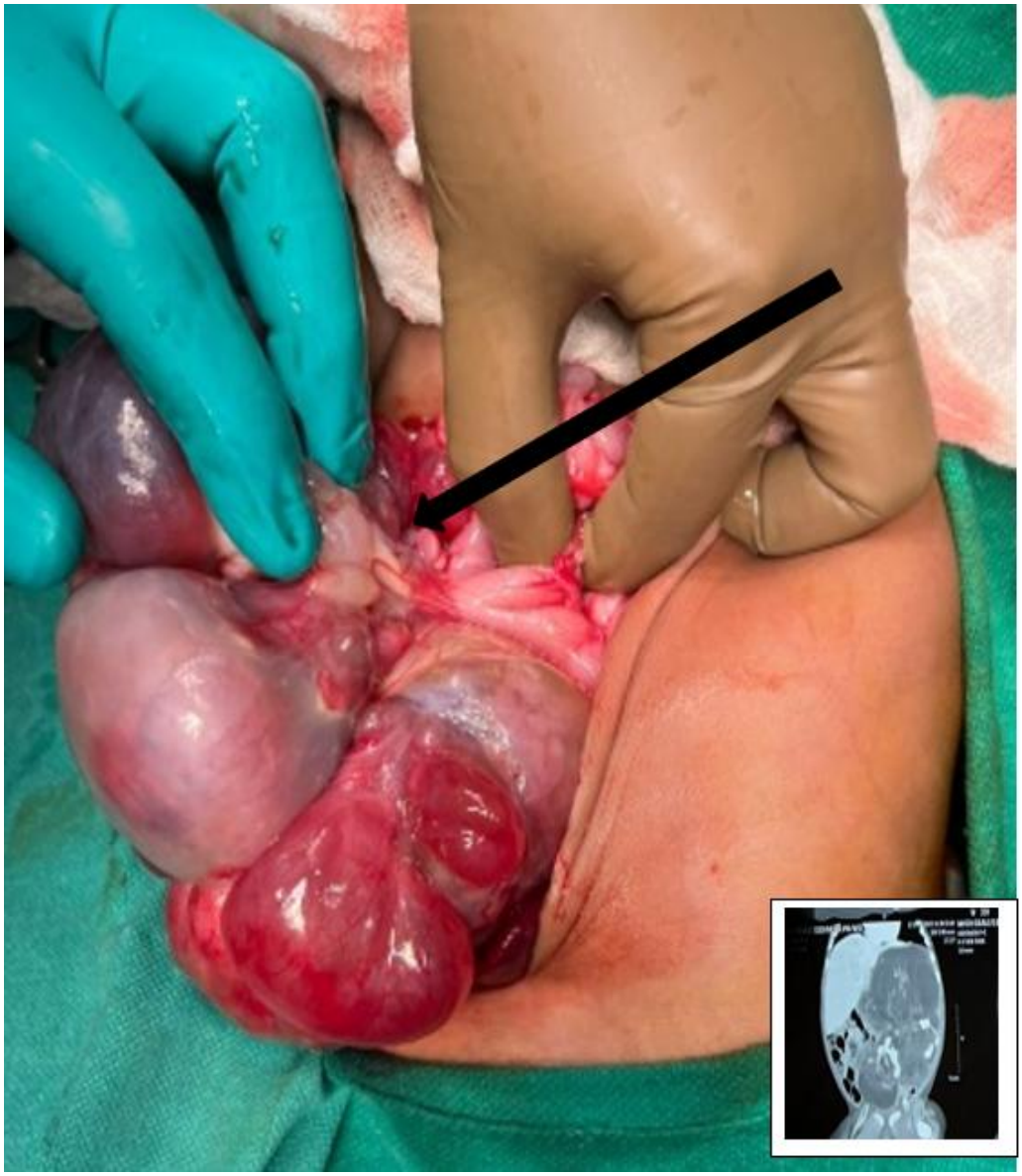

Figure 2: Intra operative photograph showing a large multicystic mass arising from posteroinferior wall along its lesser curvature of the stomach (arrow mark) with an inclusive picture of CT scan – showing a lasrge heterogenous mass with solid cystic components with calcifications.

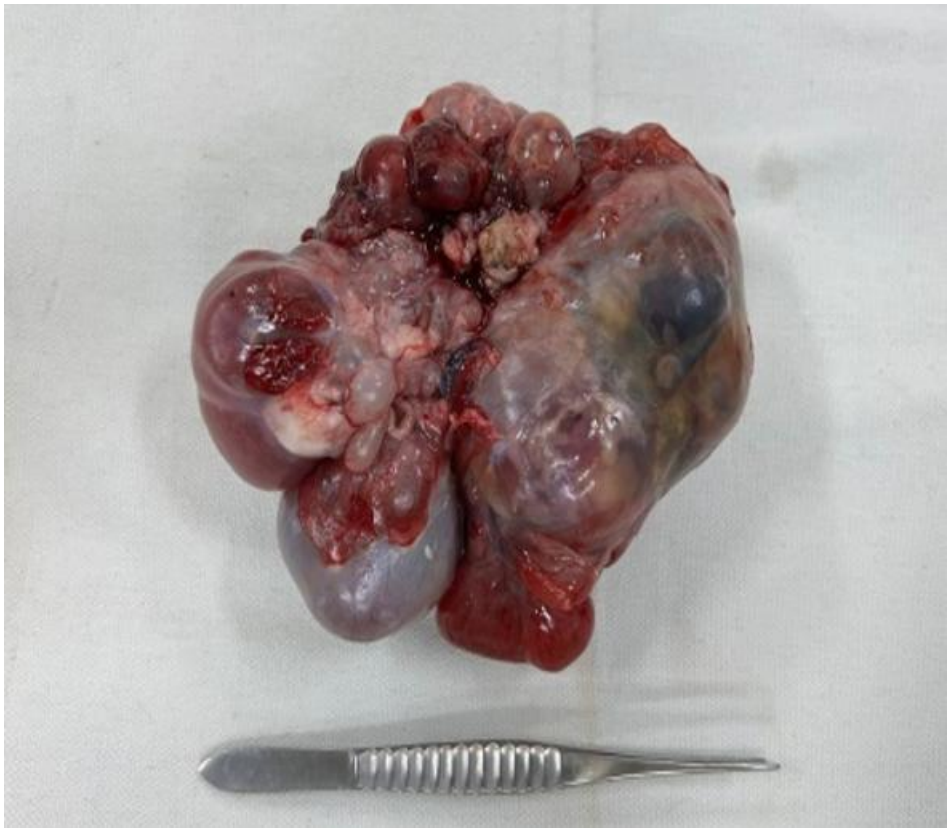

Figure 3: Resected specimen of of the mass showing a Variegated appearance, multicystic in nature

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