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Immature Gastric Teratoma – A rare paediatricintraabdominal neoplasm.

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Abstract: *We present a case report of a 4months old first born male child which was brought to our hospital with complaints of abdominal distension and mass in the upper abdomen causing feeding difficulty. Child was clinically found to have a firm non tender mass of about 10 x 8cms in the left upper quadrant of the abdomen which was clinically suspected to be Neuroblastoma. The child was subjected to ultrasound examination using 5-7Mhz Linear transducer in Philips HD11XE machine, which revealed a multicystic heterogeneous mass lesion of 10 x 8cms in the left hypochondrium, displacing the left kidney posteriorly and spleen inferiorly and crossing the midline showing significant peripheral colour uptake, possibility of Neuroblastoma. The child was then subject to CT scan of abdomen with contrast enhancement using 16slice Toshiba Activion scanner. The findings were a large, fairly well defined heterogeneous mass showing both solid and cystic areas showing significant internal and peripheral enhancement with areas of coarse amorphous calcifications. The mass was seen to erode the posterior wall of stomach and displacing the oral contrast within the stomach. The bowel loops were displaced inferiorly and towards the right, the left kidney posteriorly and the spleen inferiorly. No adjacent lymphadenopathy was seen. The child later underwent exploratory laparotomy and a large multicystic mass arising from postero-inferior wall of the stomach along its greater curvature was excised and stomach repaired. On histopathology it was proved to be an immature gastric teratoma containing mixed derivatives of all three germ cell layers.*

Keywords:*Immature, Gastric Teratoma, abdominal mass, neonate, male.*

I. Introduction

Teratomas are infrequent germ cell tumors of childhood. They may arise from any organ but the majorities are found in the ovary, testis, sacrococcygeal region and mediastinum. Gastric teratoma is rare, accounting for less than 1% of all teratomas.[1–3]The first case of gastric teratoma was reported in 1922 by Eustermann and Sentry and since then only 102 cases have been reported in the literature.[4]In 1979, Purvis et al reported the first case in a female infant; since then, five more cases have been added to theliterature.[5]In 1977, Moriuchi et al. reviewed 44 cases and found that 85% of the patients were below the age of one year and all were males.[6]More than 90% of teratomas arose along the greater curvature of the stomach and 100%were histologically benign. They usually present as a palpable mass in the epigastrium and left side of abdomen with abdominal distension. Mature gastric teratomas are even rare. They mostly present as exogastric growths, but can occur as a mix of exogastric as well as endogastric extension. They are classified into mature and immature teratomas based upon presence and differentiation of neuroglial tissue. Mature gastric teratomas are benign and have good prognosis after complete surgical excision.[7]

Teratomas are well-known neoplasms arising from totipotential cells with a capacity to differentiate along all three germ lines ectodermal, endodermal and mesodermal.[1] Gastric teratoma is a very rare condition comprising less than 1% of all childhood teratomas and less than 2% of all neonatal abdominal masses. The most common teratomas of childhood are sacrococcygeal (65%), gonadal (20%), mediastinal (10%), intracranial and cervical (5%). Unlike childhood teratomas in other locations, which have a female predominance of 68%, gastric teratomas occur overwhelmingly in males.[5]Gastric teratomasare, however, the most common teratoma of the gastrointestinal tract. The second most common site of origin is the oropharynx and tongue.[8] Pancreatic, mesenteric and small bowel teratomas have only been reported as single cases.[9,10]Gastric teratomas are large lesions which can cause premature labour, dystocia and respiratory difficulty because of their size.[11]A palpable mass is reported in 75% of patients and only a distended abdomen in 56%.Gastrointestinal hemorrhage is uncommon,except in those tumours with intramural growth, which usually arise posteriorly from the region of the greater curvature and grow exogastrically.[7]There has been at least one other case reported where the teratoma arose high on the stomach near the gastroesophageal junction.[11]Almost all gastric teratomas are benign. Excision is usually possible without gastric resection. Resection appears to have been curative in all cases.

II. Case Report

We present a case report of a 4months old first born male child which was brought to the paediatric department of our hospital with complaints of abdominal distension and a mass in the upper abdomen which was progressively enlarging in size since last 2 months. There was associated non projectile bilious vomiting since last 3 days. The child was however able to pass flatus and faeces.

On examination the child was moderately built and nourished with stable vitals. The abdomen of the child showed a fairly well defined mass occupying the left hypochondrium and epigastrium which was non tender, firm in consistency with restricted mobility. The mass was seen to move with respiration and was dull on percussion. The left kidney and spleen were not separately palpable. Bowel sounds appeared sluggish. Other systems were unremarkable.

Hemogram revealed an Hb of 8gm/dl, TC of 24350 cells/$mm^{3.}$N-40%, L-50% and M-10%. Peripheral smear revealed microcytic hypochromic anaemia with leucocytosis and thrombocytosis. Liver and Renal function tests were well within normal limits. C-reactive protein was elevated (392mg/L). Serum electrolytes were within normal limits. Alpha fetoprotein (AFP) was 21 IU. Clinically the child was suspected to have neuroblastoma arising from the left adrenal gland.

The child was referred to the Department of Radiology of Bapuji Hospital Davangere, and the following investigations were carried out.

Plain Abdominal Radiograph:

Plain radiograph of the abdomen showed a large, fairly well defined soft tissue mass arising in the left hypochondrium crossing the midline and reaching upto the epigastrium, obscuring the left renal shadow. The mass was seen to displace the air within the stomach upward sand the small bowel loops were seen to be displaced inferiorly and to the right. No definite areas of calcification noted within the mass.

Ultrasonography:

The child was subjected to real time grey scale ultrasound of abdomen using 5-7Mhz linear transducer in the Phillips HD11XE machine. Ultrasound revealed a large well defined, multicystic heterogeneous mass showing both solid and cystic areas measuring about 10 x 8cms, occupying the left hypochondrium and epigastrium, with areas of calcification showing post acoustic shadowing. The mass was seen to displace the bowel loops the right and the spleen inferiorly. The left kidney was seen compressed by the mass in the posterior aspect. Possibility of Neuroblastoma to be considered.

CECT Abdomen:

Contrast enhanced CT scan of the abdomen was performed with 16 slice Toshiba ActivionMultidetector Spiral CT scanner after administration of 7ml ultravist contrast agent along with oral and rectal contrast. Images were acquired with a pitch of 5mm and slice thickness of 3mm with tube potential of 80Kvp and tube current of 120mAs. The images were viewed using a WL of 40HU and WW of 400HU.

CT shows a large fairly well defined heterogeneous mass lesion showing both solid and cystic areas with significant internal and peripheral enhancement with interspersed areas of coarse amorphous calcifications and areas of fat density within it, arising from the posterior wall of stomach, measuring 87mm (L) x 91mm (W) x 110mm (H). The mass is seen to cross the midline displacing the bowel loops including transverse colon and stomach inferiorly and to the right. The mass is seen to abut the anterior abdominal wall. The left kidney is seen to be compressed posteriorly by the mass. The spleen was displaced posteriorly and inferiorly by the mass. No significant adjacent lymphadenopathy. Imaging features suggest possibility of Immature Gastric teratoma arising from the posterior wall of stomach and extending into the lesser sac.

The child underwent resection of the mass. Intraoperatively, the mass was seen to arise from the posterior wall of stomach and the diagnosis of Immature Gastric teratoma was confirmed. The specimen of the resected mass was sent for histopathological examination which revealed a neoplastic lesion composed of mixed derivatives of all the three germ layers, which include stratified squamous epithelium with adnexae, respiratory epithelium, intestinal epithelium and glial and cartilage forming elements. There were immature neuroepithelium with formation of true rossettes. These features were suggestive of Immature Gastric teratoma – Grade-2.

III. Discussion

The word teratoma is derived from Greek word "teratomas" meaning "monstrous growth". Generally, they are composed of tissue related to all the germinal layers. Gastric teratoma was initially described by Eustermann et al in 1922. After the first report in 1922, Margret et al reported a case of gastric teratoma in a one day old newborn and Gamanagatti et al published a case in a 2 year old boy.[12,13]Teratomas are well-known neoplasms arising from totipotential cells with a capacity to differentiate along all three germ lines ectodermal,

endodermal and mesodermal.[5]Gastric teratoma is a very rare condition comprising less than 1% of all childhood teratomas and less than 2% of all neonatal abdominal masses**.** [5]The most common teratomas of childhood are sacrococcygeal (65%), gonadal (20%), mediastinal (10%), intracranial and cervical (5%). Unlike childhood teratomas in other locations, which have a female predominance of 68%, gastric teratomas occur overwhelmingly in males.[5]Gastric teratomasare, however, the most common teratoma of the gastrointestinal tract. The second most common site of origin is the oropharynx and tongue**.**[8] Pancreatic, mesenteric and small bowel teratomas have only been reported as single cases.[9,10]

Gastric teratomas occur in neonates and infants, with approximately90% of cases reported in boys.[4,5,9] Cases in fewer than a dozen female children with gastric teratoma have been reported.[4,5] Gastric obstruction by the mass may produce polyhydramnios prenatally, premature labour or dystocia, or feeding problems after birth. Postnatal detection of the mass often is established with palpation, as is true for detection of many pediatric gastrointestinal tumors.[4,5] Gastrointestinal bleeding is a rare symptom of teratoma at presentation and may be caused by ulceration of the tumor.[4,14] In virtually all cases, gastric teratoma is an isolated finding and is not associated with other tumors or malformations.

The finding of an intragastric defect during an upper gastrointestinal series in a neonate evokes a differential diagnosis of intraluminal debris, such as a large organized blood clot or, less likely a lactobezoar, especially if these findings occur within the immediate perinatal period. With sonography, the diagnosis of a blood clot or a lactobezoar was excluded in two ways. Findings at sonography demonstrated that the mass was not mobile; a blood clot and a lactobezoar generally are. Findings at sonography also showed that the mass was complex, contained calcifications, and extended beyond the gastric lumen, and these characteristics are absent in patients with blood clots and lactobezoars. Gastric teratoma is an uncommon lesion, and it frequently projects into the lumen and includes calcifications in about 35%–60% of children.[9]

The differential diagnosis of a calcified mass in the left upper quadrant in a neonate would include mesoblasticnephroma, congenital neuroblastoma, and gastric teratoma. In this neonate, because the kidney was normal at sonography, mesoblasticnephroma was easily excluded. Neuroblastoma occurs more often in the neonate than in the older child. The uncommon cystic variant is more frequently seen in neonates, but even in this age group, neuroblastoma tends to be solid. In the neonate, liver involvement is a more common manifestation of neuroblastoma than is a focal abdominal mass. With CT, calcifications are seen in about 85% of neuroblastomas, but intragastric extension of a congenital neuroblastoma is exceedingly rare.[15]

The presence of solid, cystic, and calcified tissues in the left suprarenal location suggested two diagnoses: congenital neuroblastoma and gastric teratoma. The intragastric location of the mass increased the likelihood that the mass was a gastric teratoma. Majority of gastric teratomas are exogastric (>60%); endogastric growths are present in 30% of cases. Mixed exogastric and endogastric growths are rare.[16]In our case the main component of the mass was exogastric (90%) whereas a small proportion was endogastric which was detected by palpating the gastric lumen before its excision. In case of endogastric component there may be additional upper alimentary tract bleeding (hematemesis and melena), and pain abdomen.[1,7]Gastroscopy in case of endogastric component may aid in the preoperative diagnosis.

Patients with gastric teratomas, even those with malignant histologic features or exophytic extension into adjacent organs and tissues, have an excellent prognosis.[7-11]Abdominal radiograph, ultrasonography, CT/MRI, and endoscopy are important diagnostic tools. In most of the cases the preoperative diagnosis of gastric teratomas is difficult. However the CT- density value is very important. As most of the cystic teratoma contains fatty substances so CT density value is below '0'(zero)i.e. – (minus) in Hounsfield unit (HU). Surgical resection of the entire mass usually is curative; incomplete resection may result in recurrence. Additional therapies (ie, chemotherapy and radiation therapy) are not needed.

Diagnosis of teratoma is confidently made when the tumor contains derivatives of all three germ layers. Some pathologists will make the diagnosis of teratoma in tumors exhibiting only two germ layer derivatives, but such tumors are more correctly termed dermoids.[9] Tissues derived from the epithelium of teratomas frequently develop into the classically visualized elements of teeth, fat, and hair.The histopathology confirms the diagnosis and states about the maturity of the teratoma. A grading system, based on histopathological findings, divides the gastric teratoma in two main varieties viz. mature teratoma(grade 0) and immature teratoma (grade 1, 2, 3). Inmatureteratoma, mature and well differentiated tissues belonging to all the three germinal layers, is present. In case of immature teratoma, immature neuroectodermal tissue is usually found along with other germinal layer structures. In grade 1 immature teratoma the immature neuroectodermal tissue is confined to one site in a slide, where as in grade 2 and 3 the immature tissue is usually found in less than 4 and more than 4 fields per slide,respectively.[7,13] In our case the teratoma was assigned Grade- 2.

The monitoring of AFP and beta-HCG reflect the treatment response after excision and may be of significant value where chemotherapy is recommended(immature variety).[13.]

Total excision and primary closure of the gastric wall is the treatment of choice. Partial, subtotal and total gastrectomies have been performed as dictated by the extent of stomach involvement.[11]The prognosis following surgical excision of a gastric teratoma has been shown to be excellent.

IV. Figures

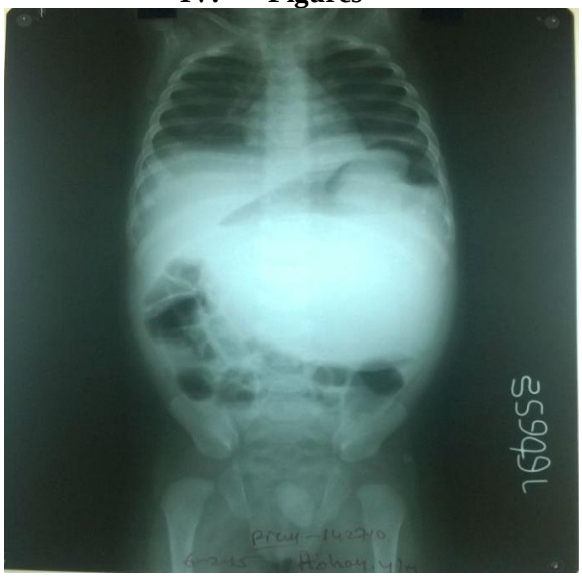


Fig 1: Supine abdominal radiograph of the 4 month old male child showing a large well defined soft tissue mass arising in the left hypochondrium crossing the midline and reaching upto the epigastrium, obscuring the left renal and splenic shadow and displacing the bowel loops towards the right side. The fundal gas shadow is seen in the superior aspect of the mass. No definite calcifications could be made out.

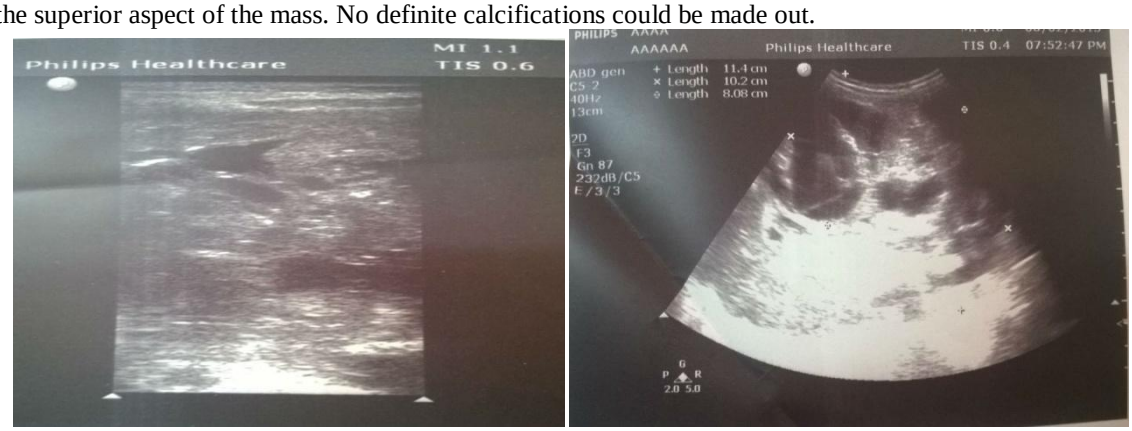
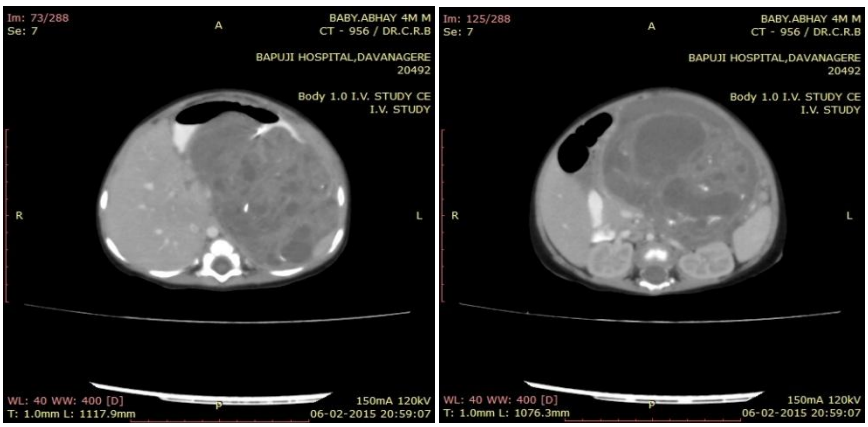


Fig 2:Grey scale ultrasound image of the abdomen showing a large well defined, multicystic heterogeneous mass showing both solid and cystic areas measuring about 10 x 8cms, occupying the left hypochondrium and epigastrium, with areas of calcification showing post acoustic shadowing



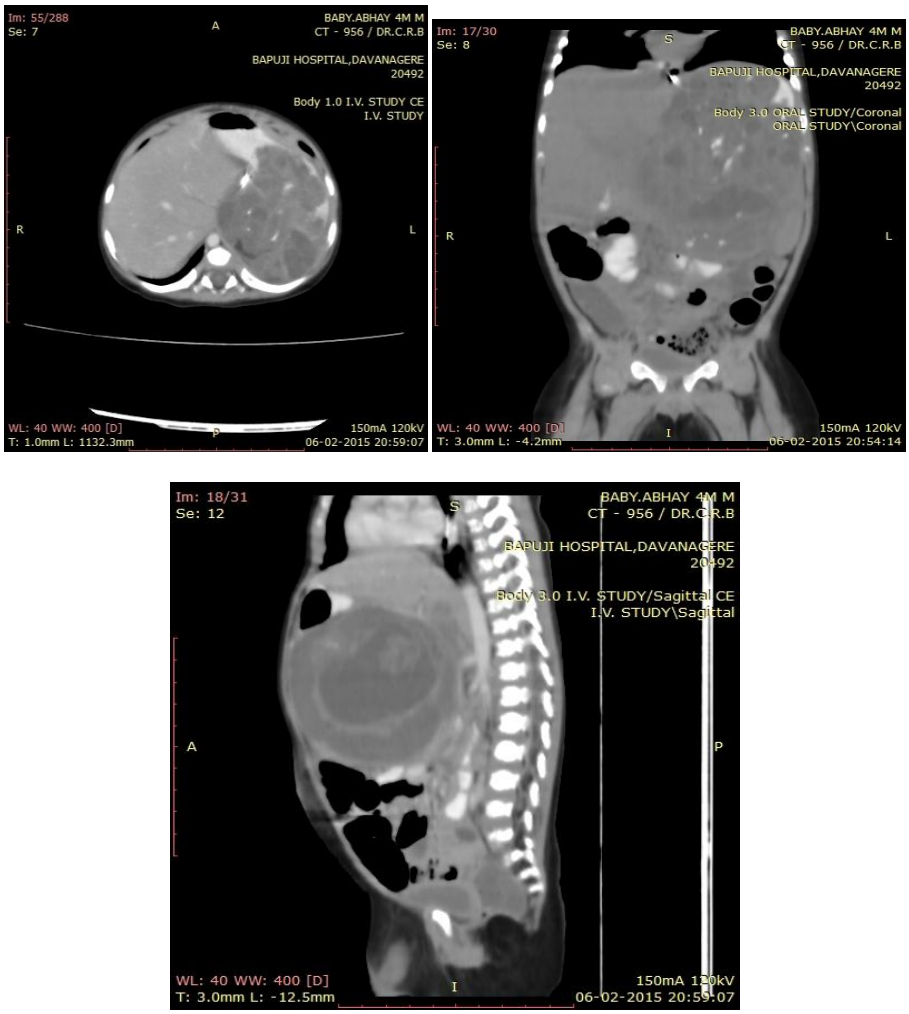


Fig 3: Multiplanar reformatted contrast enhanced CT scan of the abdomen showing a large fairly well defined heterogeneous mass lesion showing both solid and cystic areas with significant internal and peripheral enhancement with interspersed areas of coarse amorphous calcifications are areas of fat density within it, arising from the posterior wall of stomach, measuring 87mm (L) x 91mm(W) x 110mm (H). The mass is seen to cross the midline displacing the bowel loops including transverse colon and stomach inferiorly and to the right. The mass is seen to abut the anterior abdominal wall. The left kidney is seen to be compressed posteriorly by the mass. The spleen was displaced posteriorly and inferiorly by the mass.

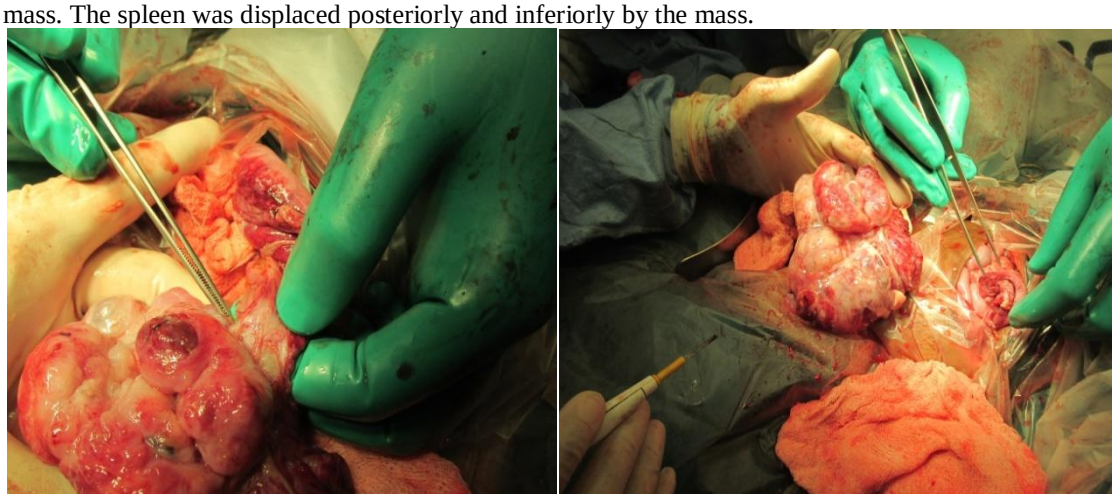


Fig: 4 Intraoperative images demonstrating a large fairly well defined heterogeneous mass lesion arising from the posterior wall of stomach occupying the lesser sac composed of both solid and cystic areas. The mass was fed by branches from the left gastric and splenic arteries. The lesion was removed en block and the posterior wall of stomach was repaired. The postoperative period of the child was uneventful.

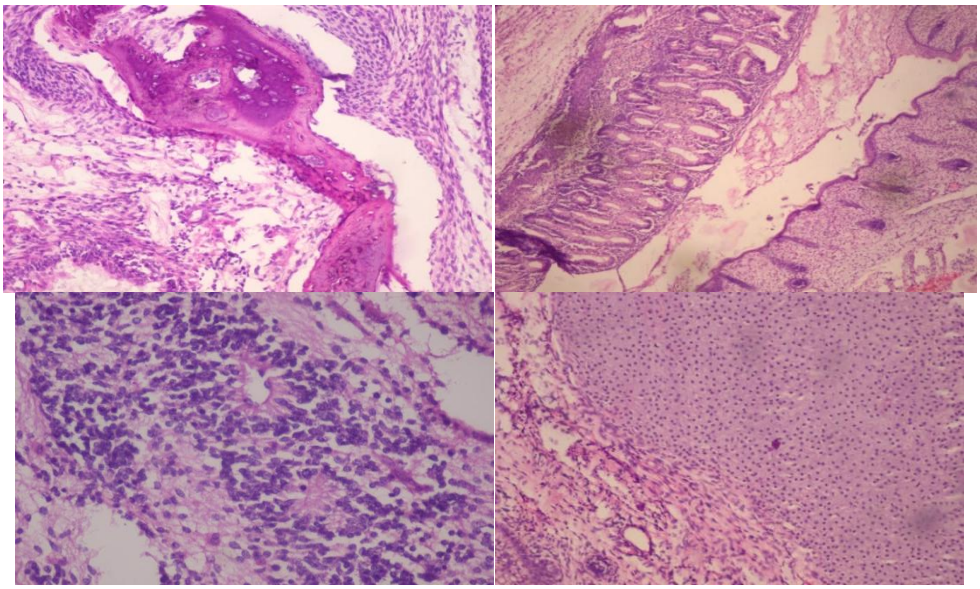


Fig:5 Histopathology H&E stained photomicrographs in 20X magnification from various parts of the resected tumour mass showing derivatives from all three germ layers confirming the diagnosis of immature teratoma. A. Demonstrating spicule of bone. B. Represents intestinal glands on left side and skin with appendaged on right side of slide. C. Demonstrates Immature neuroepithelium with pseudo rossets formation derived from primitive neuroectoderm. D. Shows a section of immature cartilage.

V. Conclusion

The diagnosis of immature gastric teratoma should be kept in mind in a neonate presenting with left upper quadrant calcified mass with or without features of upper gastrointestinal bleed. Ultrasound and CT helps to delineate the anatomical extend of the tumour and give a clue to the probable origin of the tumour. CT scans are superior to ultrasound in detecting areas of fat and calcifications within the mass which points towards the diagnosis. Histologically the tumour is characterised by presence of derivatives from all the three germ layers including neuro glial elements. Excellent prognosis is seen if complete surgical excision of the mass can be achieved. Recurrence rate although very rare have been reported.

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