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Imaging in Gluteal teratoma: a rare site of extragonadal teratoma

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Abstract*: Extragonadal teratomas are rarely encountered in infants and children. These tumours are reported in retroperitoneum, floor of the mouth, mediastinum, cranio-facial region or even in the solid organs of the body. Most of the gluteal teratomas reported in the literature are in fact, lateralized sacrococcygeal teratomas. Imaging always plays an important role in the localization & characterization of these masses as clinical presentation often nonspecific. We are reporting a case of fetiform teratoma in the gluteal region in a 6 year old girl where CEMR showed a well encapsulated lesion containing well differentiated small & large bowel loops within .The tumor did not have any connection with the sacrum or coccyx and was successfully removed. Histology showed well differentiated bowel as well as fetiform structures.*
Key words*: Teratoma; extragonadal teratoma; fetiform.*

I. Introduction

Extragonadal teratomas are rare tumours and usually present during infancy or early childhood. These tumours may arise in unusual locations and have characteristic fetiform pattern .Imaging is always very important for a complete surgical excision which is curative, as because their location near important organs may make complete excision very challenging. This report entails imaging findings of a fetiform teratoma in the gluteal region that followed by successful surgical excision, which clinically appeared to be a lipoma in a young girl. Case report: A 6 year old girl was referred to the department with a swelling in her left buttock, which was present since birth. The swelling was gradually increasing in size along with pain and discomfort while seating (Figure 1). The tumor was bulky and soft on palpation and could be separately felt from the sacrum and coccyx.Contrast enhanced MR scan (CEMR) showed a large well encapsulated lobulated T1 (fig :3)and T2 hyperintense lesion which is suppressed on fat suppressed sequences (T2 FS) was seen deep to the gluteal muscle extending up to the left side of pelvic cavity in the saggital & coronal planes (fig :4). The lesion was displacing the urinary bladder, uterus and ovaries upwards into the false pelvis and the rectum and sigmoid colon (fig: 2) to right side along with widening of the pelvic outlet (fig :5). Small and large bowel loops are seen within the lipomatous lesion (fig: 2). Two STIR hyperintense cystic components noted within the lesion largest measuring 0.9cmx1.2cm .Post contrast study did not reveal any abnormal enhancing component within the lesion.GRE sequences shows blooming signifying calcifications. The lesion was in close approximation with the sacrum .But no connection with the Sacrum was seen. Sacrum & Coccyx were normal. No free fluid is seen in peritoneal cavity.

Serum alpha-fetoprotein level was normal. On exploration, well developed colon and small bowel loops were seen along with small cysts, bones and muscle elements (Figure 6). The tumor was completely excised along with its sac; redundant skin and subcutaneous tissues were excised. The patient recovered well and was discharged on day 6. The histology showed structure of normal colon and small bowel with areas of fetiform structure in soft tissues. Derivatives of endoderm, mesoderm and ectoderm were identified. Fetiform structure also contained mature respiratory epithelium with rudimentary cartilage and gut mucosa. The patient has been on follow up for 16 months. At last follow up, the scar was healthy and there was no evidence of recurrence.

II. Discussion

Extragonadal teratomas of childhood are rare tumours and may occur in unusual locations such as, retroperitoneum, mediastinum, cranio-facial region or even in the solid organs (liver, kidney). These tumours often present with acute symptoms and may have some distinctive features such as an unusual location, and a "fetiform" histotype of the lesion. Imaging always important for localization & characterization of these masses i.e. to demonstrate their extension or involvement of neighboring organs or tissues to guide surgical resection [9, 12]. Ultrasonography (USG),Computed Tomography (CT) & Magnetic Resonance Imaging (MRI) are done alone or in combination to get anatomic delineation & depiction of various tissues types i.e. fat ,fluid, calcification ect [9,11].Some tumours are detected on antenatal ultrasound and treatment in such cases can be initiated early to prevent complications. Prenatal Ultrasonography (USG) can demonstrate mass & polyhydramnios. The presence of fetal hydrops with ascites, pleural effusion & skin edema indicates poor outcome especially in presence of placentomegaly. Large tumours that are solid initially may develop anechoic

areas signifying hemorrhage within .Presence of hemorrhage & necrosis in a smaller lesion suggests malignant change [9, 10]. Tumours occurring in unusual sites may cause problem because of their anatomical location and size. An emergency surgery may sometimes be needed because of mass effect of the tumour.

CT & MRI are excellent methods of investigation for defining the extent of the sacrococcygeal mass. In case of a pelvic tumor, extension through the sacrosciatic notch & into the buttock can be detected as in our case.MRI is better for the detection of extension into the spinal canal & to assess the pelvic floor musculature [9]. Cohen et al reported MRI is superior than CT in predicting tumor histology It can differentiate soft tissue from fat ,hemorrhage, fluid & calcification .It is better than CT in defining tumor size & extent of the lesion .Above all it uses no radiation [13].

Bhalla et al reported 267 cases of teratomas, of which 10 cases occurred at rare sites, such as retroperitoneal space (3 cases), floor of the mouth (3) and one each in the kidney, mediastinum, thyroid gland and the urinary bladder [1]. 2 of these tumours were solid. All these 10 tumours had foci of immature elements.Paradies et al reported 4 cases of teratomas at unusual sites in 2 neonates, 1 infant and one 4 year old child [2]. 2 neonates had tumours detected on antenatal ultrasonography and 3 patients presented with acute symptoms requiring emergency surgery. 2 neonates had tumours in temporozygomatic region and abdomen respectively. The infant had a tumour arising from the liver and the older child had a retroperitoneal tumour. All these tumours had mature elements on histology.

Lange described a benign cystic teratoma in the gluteal region [3]. Carro et al used MRI imaging to differentiate a gluteal teratoma from more common tumours in this site arising from fat or fibrous tissue [4].Fetiform teratoma is a distinct entity with highly organized differentiation but without visceral organ differentiation. However, Kuno et al described a fetiform teratoma with both a highly developed axial skeleton and organs that included a brain, eyelike structure, trachea, thyroid gland, blood vessels, gut, and phallus like structure. Remarkably, skeletal muscle has never been documented in a teratoma[5]. In the present case, well developed colon and small bowel loops were noted inside the teratoma, however, the tumour had no connection with the sacrum or coccyx. Most of the gluteal teratomas reported in the literature are, in fact sacrococcygeal teratomas presenting in a lateral location. Sood et al reported an 11 year old girl who presented with a fetiform gluteal mass attached to the sacrum by a fibrous band [6]. Gajbhiye et al also reported a 4 month old girl with anorectal malformation, who had a gluteal mass [7]. The mass was excised and found to be a sacrococcygeal teratoma. Jan et al described a case of lateralized cystic sacrococcygeal teratoma mimicking gluteal abscess [8].Conclusion :Children with extragonadal tumours may present with complications necessitating urgent surgery. In elective cases, it is ideal to have estimation of serum alpha-fetoprotein level and proper imaging studies to know about the characteristics and extent of the tumour. Serial section of the excised mass is important, especially in older children to detect any immature neuroepithelial elements or elements of Yolk sac tumour.

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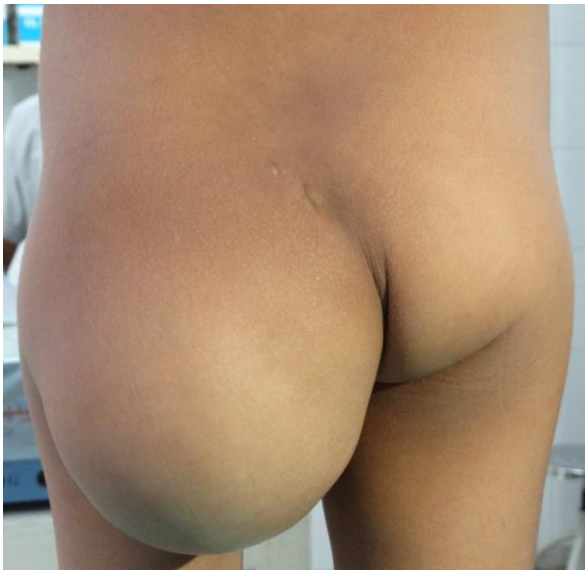
Legends:
Figure 1: Mass in the left gluteal region
Figure 2 : CE MRI T1 post contrast saggital sequence showing encapsulated mass with bowel loops (black open arrow) and fat containing tissues (white triangle) displacing Rectum &Urinary bladder (open arrow) .
Figure 3& 4 : PD FS saggital image showing Fat & well developed colon segment in the gluteal region .
Fig:5 : CE MRI T1 post contrast axial image showing encapsulated fat containing mass in the pelvic outlet.
Fig:6 : Surgical photograph of mass with well developed colonic segment .



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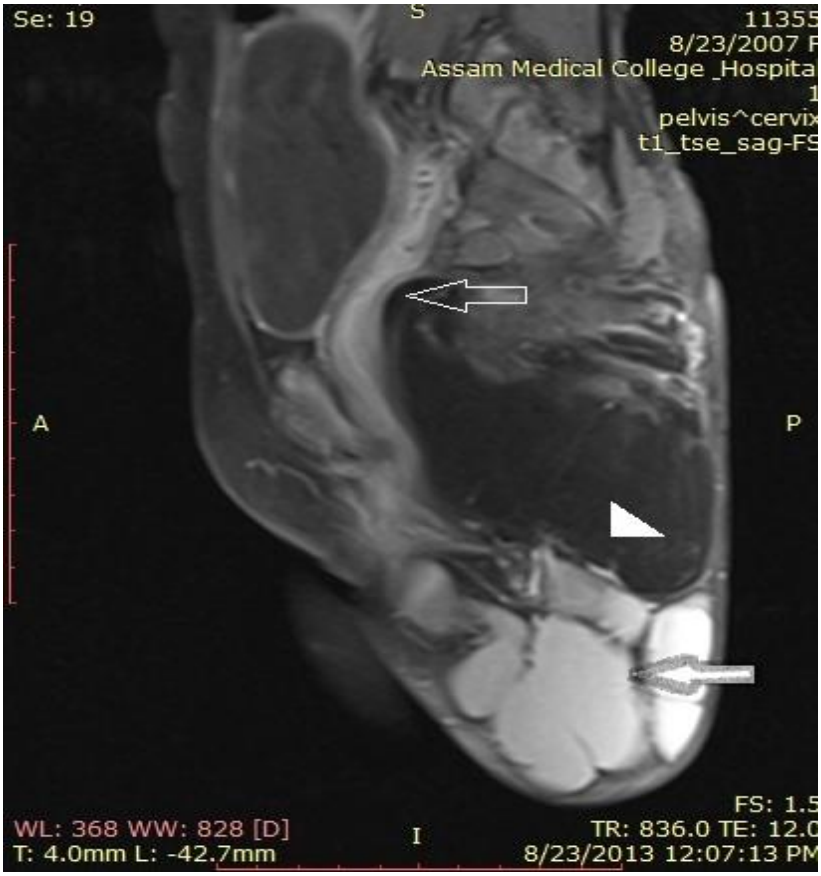

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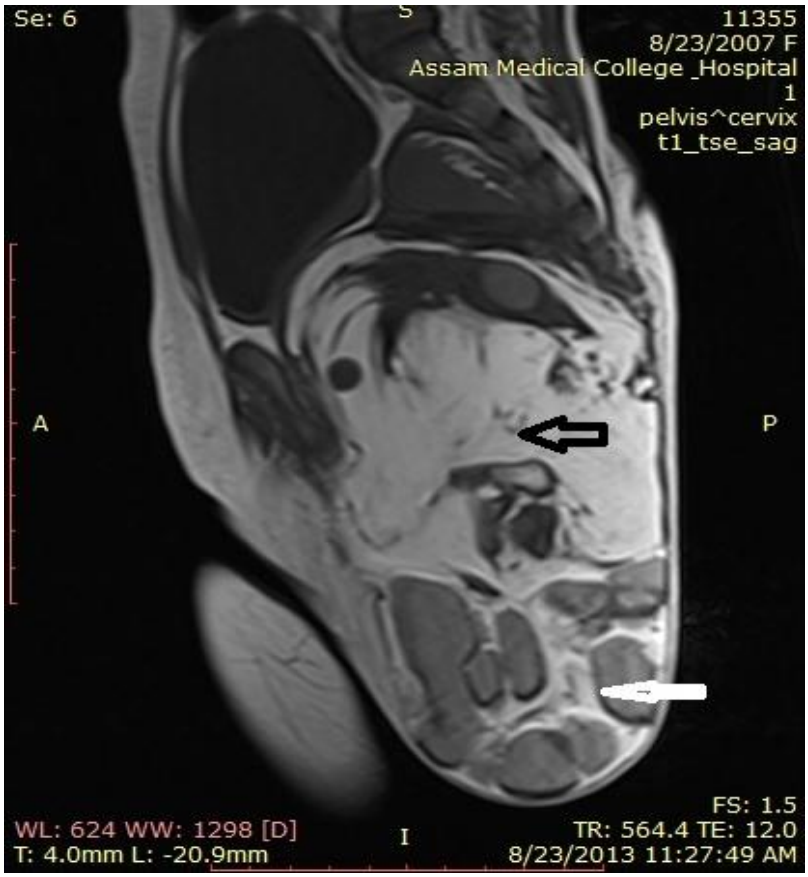
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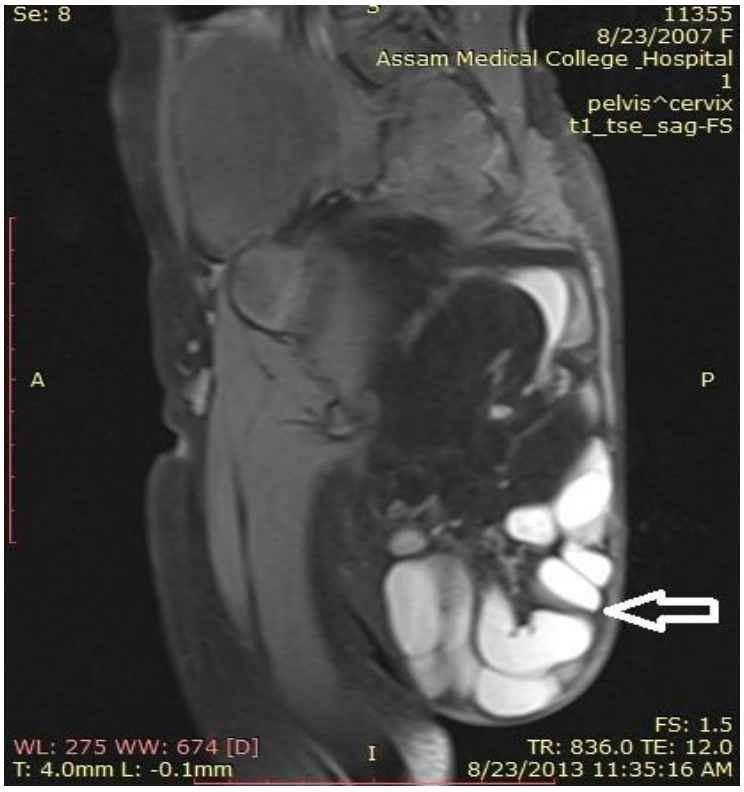
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Fig :4

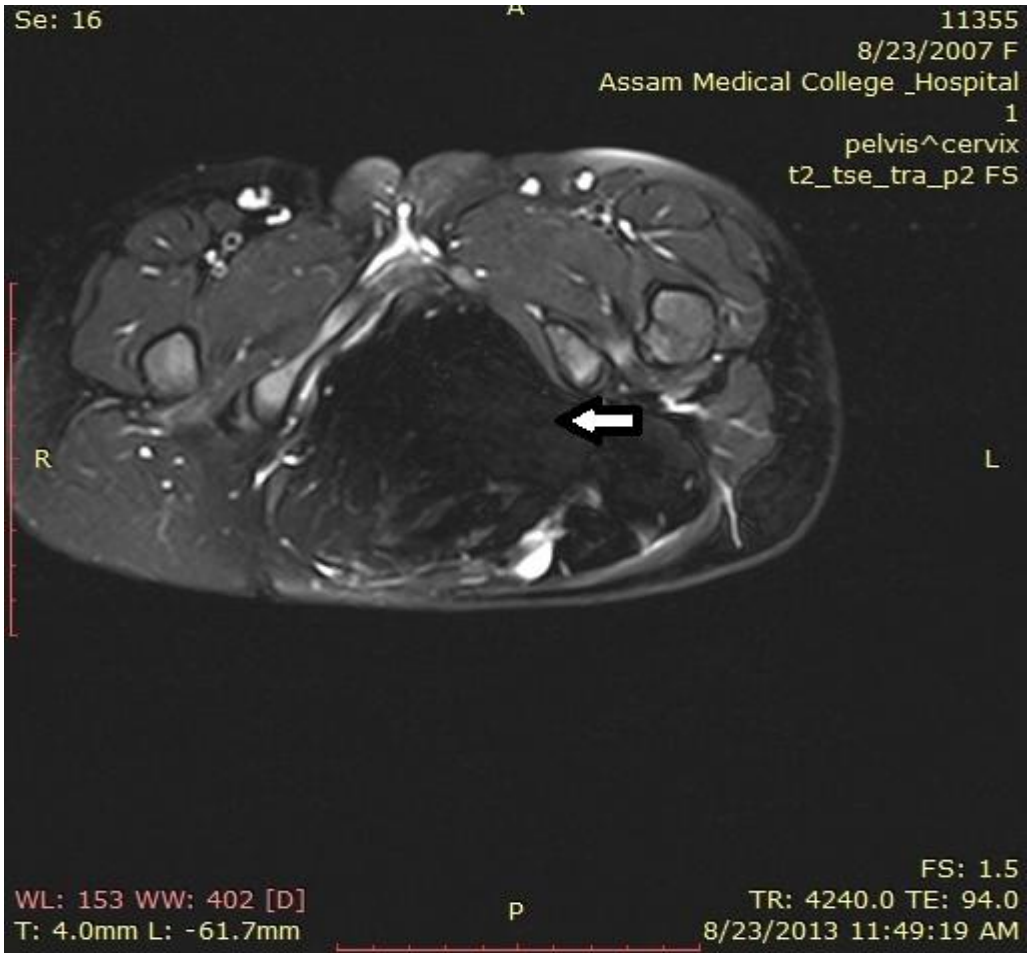
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Fig :5

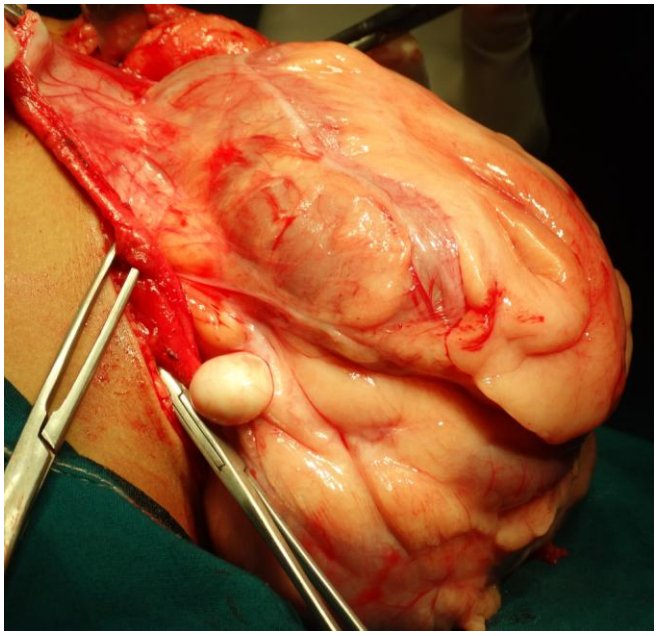

Fig :6