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Case report / Olgu sunumu

AN UNUSUAL UNILATERAL GLUTEAL MASS IN A CASE OF ANORECTAL MALFORMATION

Gluteal kitle şeklinde kendini gösteren sıradışı bir anorektal malformasyon olgusu

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ABSTRACT

A 4 months old female baby presented with a mass at left gluteal region associated with rectovestibular fistula type of anorectal malformation (ARM). Magnetic resonance imaging (MRI) revealed the mass to be sacrococcygeal teratoma (SCT). She was undergone pelvic colostomy at day 2 of her life for rectovestibular fistula. The mass was excised during PSARP. ARM with SCT is rare. Appearance of SCT with an atypical presentation (as a Gluteal mass) at an atypical age (infancy) is further rare.

Key words: Anorectal malformation, gluteal mass, teratoma.

ÖZET

Anorektal malformasyonun rektovestibuler fistül tipi ile gluteal bölgede teratom birlikteliğiyle kendini gösteren 4 aylık bir kız çocuğu olgusu sunuldu. Yapılan magnetik rezonans (MR) ile ayrıca sakrokoksigeal teratom varlığı saptandı. Doğumunun 2. gününde fistül nedeniyle pelvik kolostomi yapıldı. Kitle çıkarıldı.Teratom ve anomalinin birlikteliği oldukça nadir bir durum olup herhangi bir yaşta da saptanabilir.

Anahtar kelimeler: Anorektal malformasyon, gluteal kitle, teratom.

INTRODUCTION

Anorectal malformation (ARM) with sacrococcygeal teratoma (SCT) is a rare. Late presentation of SCT with ARM is further rare and till date no such case found in available literature. Here we present a case which we have treated successfully.

CASE

Four months old female baby presented to our department with a history of a progressively increasing lump over left gluteal region from 3 months of age (Figure 1). The lump was 8x6 cm in size with firm in consistency. The lump was appeared to be adherent to underlying structure though overlying skin was free. The baby was previously brought in emergency department at day 2 of her life with abdominal distension and absent anal opening. On examination, a tiny opening was found at vestibule through which scanty amount of meconium was passed only once. Ultrasound of abdomen showed distended rectal pouch was situated >2 cm from the anal point. Her gluteal muscles were well developed without any other anomaly. Spine was normal. Echocardiography revealed no abnormality. She was diagnosed as a case of Recto-vestibular fistula (Figure 2). Staged operation was planned and pelvic colostomy was done on the same day.

An USG of the gluteal lump showed a variegated mass with a probable diagnosis of SCT. MRI showed the mass at gluteal region consistent with teratoma with an attachment to coccyx (Figure 3) without any vertebral deformity. A preoperative serum alpha-fetoprotein (AFP) level was 35 ng/ml (normal ≤ 6-8 ng/ml). Report of complete hemo-

gram, serum urea/creatinine, serum electrolytes were within normal limits.

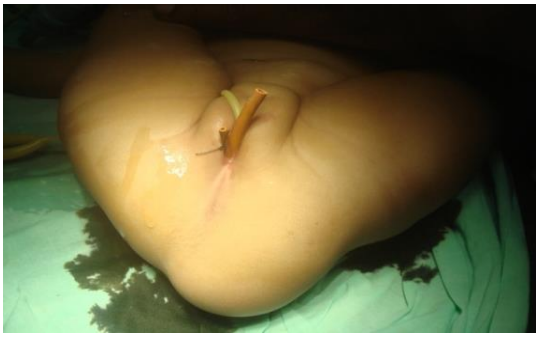

Figure 1: Anorectal malformation with gluteal mass.

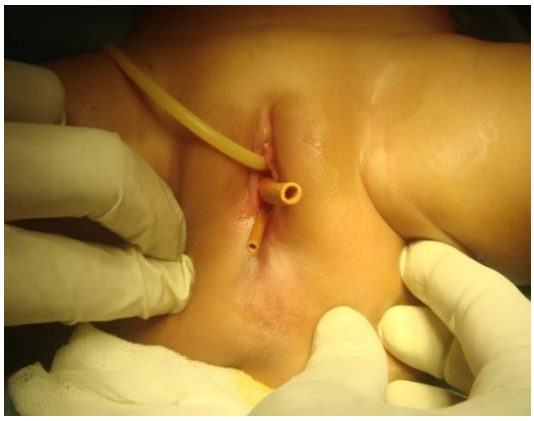

Figure 2: Foley's catheter was inserted in urethra. Larger rubber catheter was in vagina and smaller was in anus through vestibule.

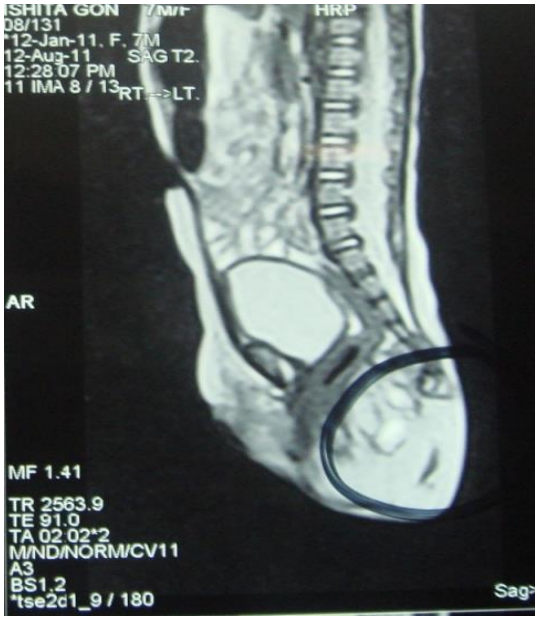

Figure 3: MRI. Indicator shows sacrococcygeal teratoma

PSARP was carried out at 4 months of age under general anesthesia with a curvilinear extension of the classical midline incision for proper exposure of the mass (Figure 4). The complete resection of SCT with the coccyx was done along with PSARP without any difficulty.

The serum AFP was repeated at 6 weeks, 3 months and 1 year following PSARP, which were found to be within normal limits. The histopathology of the mass came out to be a mature teratoma without any immature elements.

The patient was later underwent colostomy closure at 8 months of age. The baby was followed up at regular interval for next one year following operations. She has achieved all developmental milestones without any evidence of recurrence of the teratoma.

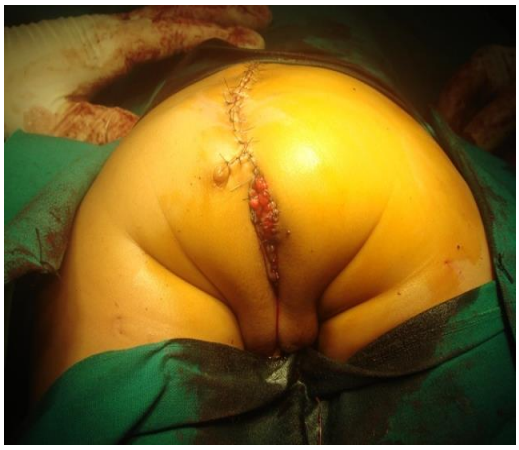

Figure 4: After completion of PSARP and excision of mass and coccyx.

DISCUSSION

Sacrococcygeal teratoma is the most common congenital tumor in the neonate. Incidence is 1 in 35000 to 1 in 40000 live births with a female preponderance (F: M ratio 4:1). Mature teratomas are the most frequent in neonates (68%). Though immature teratomas are cystic in nature, preponderance of solid component indicates malignant nature of the teratoma. Over 50% of SCT's have calcification & ossifications within them (1).

Chirdan et al in a study of Nigerian children described the presentation of SCT's in neonatal period (between 1- 18 days) in 60.5% cases, and in post neonatal period in 39.5% cases. In our case, the baby presented to us with protuberant gluteal mass at the age of 4 month (2).

Sacrococcygeal teratoma's may grow posterior direction to form a gluteal mass as in our case or may dissect anteriorly distorting regional organs (rectum, bladder & vagina etc.) without invading them (3). Another rare variety of anorectal malformations were detected by Salman et al which were associated with widely open anorectum (4). Curraino et al. had detected an association of anorectal malformation with sacrococcygeal bony defect and presacral mass due to persistent neurenteric malfor-

mation (5). In our case, sacrococcygeal bony structure was found to be normal.

Recurrence of SCT's varies between 7.5%-22%, but it can reach up to 37% if coccyx not excised. Tumors with large mean diameters show more recurrences because large tumors may well harbor undetectable small foci of malignant endodermal sinus cells (6). In this case, the baby was treated with PSARP a curvilinear extension of the classical midline incision for proper exposure of the mass. The complete resection of SCT with the coccyx was done along with PSARP.

Regular estimation of AFP is important for clinically undetectable case because raised AFP levels are needed to be regarded as recurrences which are potentially malignant (7). In the present case, the AFP levels came down to normal after operation and remained normal after 1 year follow-up.

Neither ARM nor SCT are rare. But their association is rare. ARM with delayed appearance of SCT with atypical site (in the gluteal region) so far not reported in literature. Here we present a case of ARM (RVF) with SCT noticed in gluteal region at the age of 3 month

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