Original Article

An observational study on Sacrococcygeal teratoma: a pediatric tumor at Liaquat University Hospital, Jamshoro, Pakistan

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Objective: To high light and add to local literature regarding sacrococcygeal
Teratoma (SCT).
Methodology: This descriptive study was conducted in the department of Pediatric surgery of Liaquat University Hospital, Sindh, Pakistan from January 2010 to December 2015. A total of 10 patients with SCT were included in the study. Surgery was performed all cases. Data were analyzed using SPSS version 18.
Results: Out of 10 patients, 6 were males, 4 were females with age ranging between 20 hours to 08 months. Nine were full term while only one was preterm baby. Four were delivered by normal vaginal delivery and six by cesarean section. Only three were diagnosed by ultrasound in antenatal period while seven in postnatal period.
Conclusions: SCT requires surgical excision by team of experts including pediatric surgeon, neonatologist, neurosurgeon and anesthesiologist. Better surgical outcomes are possible in our setup provided surgery in proper time and be properly done. (Rawal Med J 201;43:111-114).
Keywords: Congenital tumors, sacrococcygeal teratoma, neonatal tumors.

INTRODUCTION

Sacrococcygeal teratoma (SCT) is the most frequent congenital tumor of newborns, seen in 1/35 000 to 1/40 000 live births with a ratio of 4:1 female to male.[1] These tumors arise from remnants of primitive streak. It may be benign (mature) which is most common or malignant (immature). SCT may contain any tissue for e.g. brain, skin, respiratory, gut tissue, cartilage, smooth and striated muscle frequently while bone, pancreatic tissue, choroid plexus and adrenal tissues may also be seen in some cases.[2,3]

Usually SCT is of 8 cm, ranging from 1-30 cm. The tumors are most commonly seen in sacrococcygeal region, in rare cases may be seen in mediastinum, testes, retroperitoneum, brain, head and neck, vagina or stomach. They may continue to grow posteriorly to form an external protrusion, or grow anteriorly, distorting regional organs (rectum, vagina, and bladder) without invading them.[4,5]

The extent of tumor may be classified as follows: Type I: Predominantly external with minimal presacral component (47%); Type II: Present externally but with significant intrapelvic extension (37%); Type III: Apparent externally but predominantly a pelvic mass extending into the abdomen (09%) and Type IV: Presacral with no external presentation (10%).[6] They are graded histologically as under: Grade 0: contains only matures tissue, Grade 1: contains rare foci of immature tissues, Grade 2: contains moderate quantities of immature tissues and Grade 3: contains large quantities of immature tissue.[7]

SCT are successfully diagnosed on antenatal ultrasonography between 22nd and the 34th week of gestation. Investigations like x rays of spine, abdomen, chest, and pelvis to detect evidence of tumor or metastasis, ultrasound for extent, nature (cystic/solid) of the mass, calcification and intrapelvic extension are required. MRI can delineate the bony and muscular pelvic structures and to identify small areas of intrapelvic extension. SCT should be excised soon after birth to avoid tumor ulceration, bleeding and to reduce the risk of malignant change, which increases with increasing age. In neonates, the frequency of malignant transformation is 7%, which increase above 2 years up to 50%.[8,9]

Total resection of primary sacrococcygeal teratoma together with the coccyx is the treatment of choice

as microscopic neoplasmic foci seen in/near coccyx. Post-operative complications like weakness of lower limbs, paralysis, constipation, fecal and urinary incontinence may occur. Post-operative follow up is carried out by patient examination and assay of serum fetoprotein serum alpha-fetoprotein, bimonthly for two years for early detection of malignant tumor recurrence/ prognosis.[10,11]

METHODOLOGY

This descriptive study was conducted in the department of Pediatric surgery, Liaquat University Hospital, Jamshoro, Sindh, Pakistan from January 2010 to December 2015. Patients presenting with the SCT were included in the study. Informed consent was obtained from parents of patient. Detailed history was taken and complete physical examination was performed. Surgery was performed in all cases after proper investigation, arrangement of blood/donor. Surgical procedures included excision of tumor with coccygectomy in all cases while laparotomy was performed in two cases. Regular follow up was advised for at least two years on discharge from hospital. Data was recorded on proforma and analyzed using SPSS version 18.

RESULTS

During study period, 10 patients were admitted and operated. Out of these, 6 were males, 4 were females with age ranging between 20 hours to 8 months. Nine were full term and one was preterm baby. Four were delivered by normal vaginal delivery and six by cesarean section. Only 3 were diagnosed by ultrasound in antenatal period while 7 in postnatal period (Table 1).

Fig. Huge SCT in two months old girl.

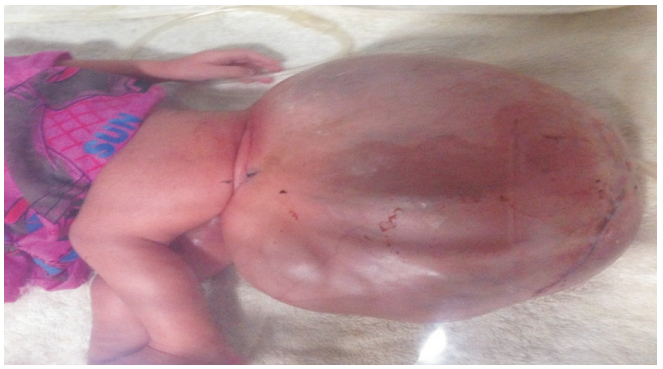

Table 1. Demographics of patients.

	Gender	Age	Mode of delivery	Diagnosis during antenatal period	Gestational Age
1	Male	01 day	Normal vaginal	No	Full term
2	Female	20 hours	Cesarean section	Yes	Full term
3	Male	05 months	Cesarean section	Yes	Full term
4	Female	08 months	Normal vaginal	No	Full term
5	Male	03 days	Cesarean section	No	Full term
6	Male	07 days	Normal vaginal	No	Pre term
7	Female	01 month	Cesarean section	Yes	Full term
8	Male	20 days	Normal vaginal	No	Full term
9	Female	04 months	Cesarean section	No	Full term
10	Male	05 days	Cesarean section	No	Full term

Table 2. Features of tumor.

Variable	Type 1	Type 2	Type 3	Type 4
	07	02	01	00
Size of tumor	Small 2-5cm	Medium 5-10cm	Large >10cm	
	00	02	08	
Type on histology	Mature	Immature		
	08	02		
Surgical procedure	Total excision	Coccygectomy	Laporotomy	
	10	10	02	
Regular Follow up	Yes	No		
	09	01		
Post-operative complications	Death	Wound infection	Recurrence	
	01	01	01	

Most tumors were Type I, 2-5 cm in size with one huge in size (Fig.), with one death and one recurrence (Table 2).

DISCUSSION

SCT is usually found in the newborn babies mostly diagnosed antenatal, at birth, within first few days of life, less than 10% diagnosed after the age of two year. Females are affected ten times more.[12] In this study, ratio of male patients was more than females i.e. 3:2. The probable reason behind this is difference in geographical locale because less number of cases or negligence/poor compliance for female child, due to illiterate and poor societies of Indo Pak.[9,12]

Modern technology using three dimensional ultrasound, SCT can be diagnosed in prenatal period even in the first trimester with more ease in second trimester, depending on observer.[13] In our study, only 3 were diagnosed before birth. Among 7 undiagnosed cases, mothers of 4 patients never had antenatal scan while in three cases it was not diagnosed, so medical negligence is not only

blamed. A study from India reported only two cases diagnosed before birth out of 10 cases.[14]

The prognosis of SCT is excellent depending on the time of diagnosis, malignant changes and possibility of surgical excision, which is the mainstay of management. Prenatal diagnosis in first trimester is associated with high fetal morbidity and mortality while presentation after 30 weeks is considered as relatively good prognostic indicator for fetal survival. Early diagnosis helps in planning delivery by cesarean section in a setup with good neonatal care and surgical intervention for new born is possible.[15-17]

After complete excision, mature teratoma tends to reoccur in 11-22% cases. In malignant cases, incomplete resection of tumor, tumor spillage during surgery and no coccygectomy may be the cause of recurrence. Regular follow up include examination of patient and serum alpha fetoprotein levels.[18,19]

In our study, nine out of 10 patients had regular follow up only 01 patient not remained in contact. Excellent recovery was seen in all patient except one patient died postoperatively because of infection and recurrence is seen in only one patient probably because of huge size and rupture during surgery. Similar survival rates are seen by other authors.[14,20]

CONCLUSION

SCT is tumors of neonates and infants. A team of experts is needed who are competent in surgical excision of tumor mass extending in nearby viscera, good anesthetic approach as hyperkalemia, cardiac arrest and bleeding in highly vascular tumor. All patients with such lesions should be referred to tertiary care centers with pediatric surgical facilities for optimal care and management.

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Conception and design: Pushpa
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Drafting of the article: Pushpa
Critical revision of the article for important intellectual content: Samreen Memon, Nand Lal
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