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SACROCOCCYGEAL TERATOMA POST OPERATIVE FOLLOW UP AND OUTCOME

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ABSTRACT

Aims: To study the post operative complications of sacrococcygeal teratoma and follow up of all the post operative cases every week, 1 month, 6 months and 1 year with clinical examination, digital rectal examination, AFP levels, Ultrasonography.

Materials and methods: It is a prospective & retrospective study of all cases which came during the study period and with minimum follow up for 1 year in 20 cases of sacrococcygeal teratoma.

Results: Commonest age of presentation of SCT is less than 1 month, followed by children between 1year and 5years. Females are more commonly affected than males. Commonest type of Altman presentation was type II, followed by type I and typeIII and type IV. No associated anomalies were noted in the present study. Large size of tumors has poor prognosis and outcome. Cases presenting in the new born period are mostly benign, whereas intrapelvic tumors presenting in older infants and children are mostly malignant. Because of routine antenatal scans most of the cases are detected antenatally, placentomegaly and polyhydramnios was present in almost all the antenatal scans. Almost all cases presented with lump in the low back and lump abdomen in type IV along with anteriorly displaced anus. Commonest presenting complaint was constipation and retention of urine in some cases. Wound infection and wound dehiscence was due continuous soiling of the wound with fecal matter which lead to wound infections and gaping of wounds which can be managed with antibiotics, dressings and secondary suturing. Constipation in post operative patients can be managed with laxatives and dietary advices, which usually subsides with age. It is mandatory to do serum alfa-fetoprotein preoperatively, to note the fall in levels post operatively, serum levels to be done for minimum of 3 years to look for recurrences. Excision of entire tumor with coccyx without rupture of the tumor should be done to prevent recurrences. Chemotherapy is given for all yolk sac tumors that is cisplatin, paclitaxel and mesna 6 cycles. In the follow up period digital per rectal examination, serum alfa-fetoprotein levels and ultrasound abdomen should be done, with minimum 3 years follow up.

Conclusion: Meticulous dissection should be done while removing the tumor to prevent bladder dysfunction post operatively, in many studies bladder dysfunction has been noted.

Keywords: Germ cell tumors, sacrococcygeal teratoma, yolk sac tumor

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INTRODUCTION

Germ cell tumours of the sacrococcygeal region are referred as sacrococcygeal teratomas.[1] Generally present in two distinct types, one is neonates with large predominantly external lesions , which are mostly detected in utero or at birth and are rarely malignant while in older infants and children who present with primarily hidden pelvic tumors have a much higher rate of malignancy. Sacrococcygeal teratomas are the most common extragonadal tumors in neonates, accounting for up to 70% of all the teratomas in childhood. The female to male ratio is around 4:1; it is predominantly seen in females.[1] Reported incidence of 1:40000 live births have been noted. Complete resection soon after birth has excellent prognosis[2].

New born typically present with a mass protruding from sacral region and many cases are detected with prenatal ultrasonography. Caesarean delivery should be considered if the mass is greater than 5 cm, to avoid dystocia and rupture of tumor. In utero shunting can lead to fetal hydrops which is associated with high mortality. Adzik[14] and colleagues performed the first successful resection in a fetus that developed placentomegaly and polyhydromnios, underwent resection of the immature teratoma Makin and colleagues reported 77% survival among 41 antenatally diagnosed SCT but noted survival of 50% in those undergoing fetal interventions, and only 14% survival if intervention was for hydrops. Fetal interventions that can be done are cyst drainage, laser ablation or alcohol sclerosis. Older infants and children typically present with symptoms related to compression of bladder and rectum[2]. If a mass has been noted at birth and left in place, there will be an increased rate of malignancy in future. AFP levels are normally elevated in new born, and the levels should be obtained and then followed to ensure return to normal levels by 9 months of age . We studied post operative complications of sacrococcygeal teratoma Follow up of all the post operative cases every 1 week, 1 month, 6 months and 1 year with clinical examination, digital rectal examination, AFP levels, Ultrasonography.

MATERIALS AND METHODS

All cases of sacrococcygeal teratoma which came to Kakatiya Medical College from December 2017 to December -2022 were included in the study. 20 cases of sacrococcygeal teratoma were included in the study group. It is a prospective & retrospective study of all cases which came during the study period and with minimum follow up for 1 year.

Patient demographics including antenatal details, mode of delivery, and age at presentation, sex, associated anomalies, and tumor histology were recorded. Surgical management with respect to resection of the primary tumor, and early and late postoperative complications were noted. Outcomes relating to recurrence-free survival, and bowel and urinary tract function were analyzed from clinical follow-up.

The diagnosis was based on the clinical suspicion subject to radiological confirmation (ultrasonography with or without contrast-enhanced computed tomography (CT). The management protocol included upfront resection for tumors with age-appropriate AFP levels. Metastatic workup was done for children with raised AFP. Neoadjuvant chemotherapy with two cycles of cisplatin, etoposide, and bleomycin (PEB) (cisplatin at 35 mg/m^2/day on days 1–3, etoposide at 100 mg/m^2/day on days 1–3 and bleomycin at10 units/m^2/day on days 1–3;

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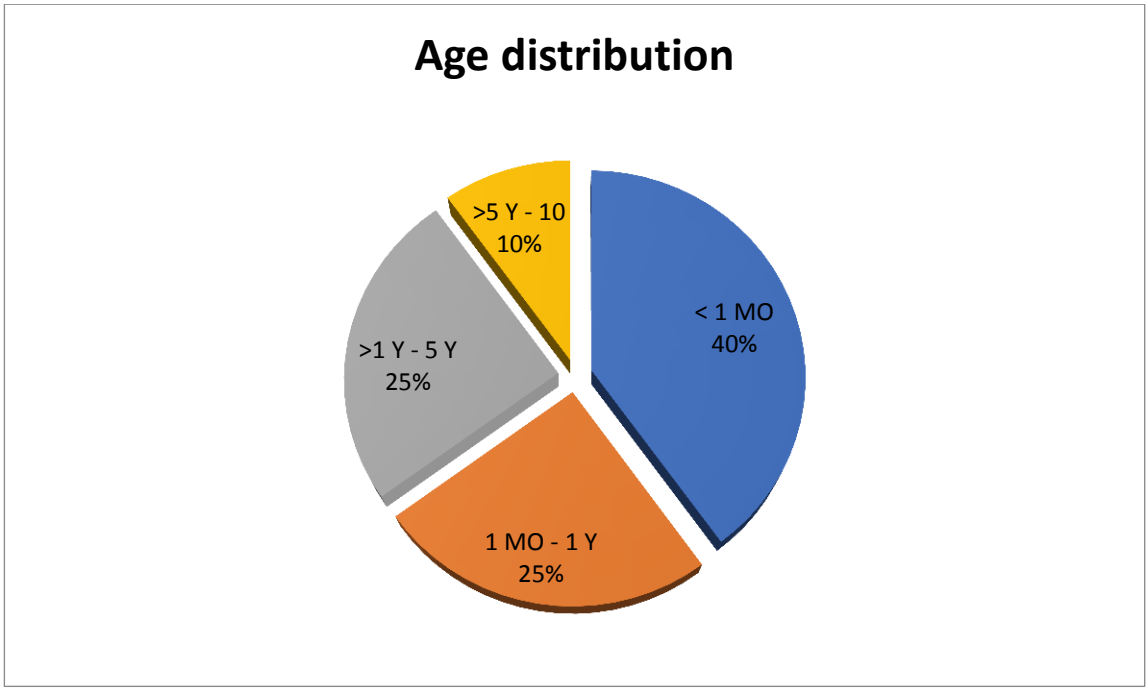
the chemotherapy cycle was repeated after 21 days) was administered to those not fit for upfront resection. Those with un-resectable tumors or persistent chest involvement post two cycles PEB were administered two more cycles.

The follow-up was governed by clinical examination, radiological evaluation, and AFP levels. AFP levels were repeated monthly (in cases with malignancy) or 3 monthly for 6 months followed by yearly for 3-years. Follow-up radiological evaluation with alternate ultrasonography or CT was done at 3 monthly (for 1 year) followed by 6 months' intervals. Cosmetic and functional results were recorded at each follow-up. Functional results were evaluated clinically and radiologically (ultrasonography of kidney, ureter, and bladder with postvoid residue).

RESULTS

Figure-1: Age of Presentation



The commonest age of presentation during the study was of less than 1 month of age followed by more than one month to 5 years. Less than 1month are 8cases(40%), 1 month to 1 year are 5 cases(25%),1year to 5 years are 5 cases(25%) and 5 to10 years are 2 cases(10%).

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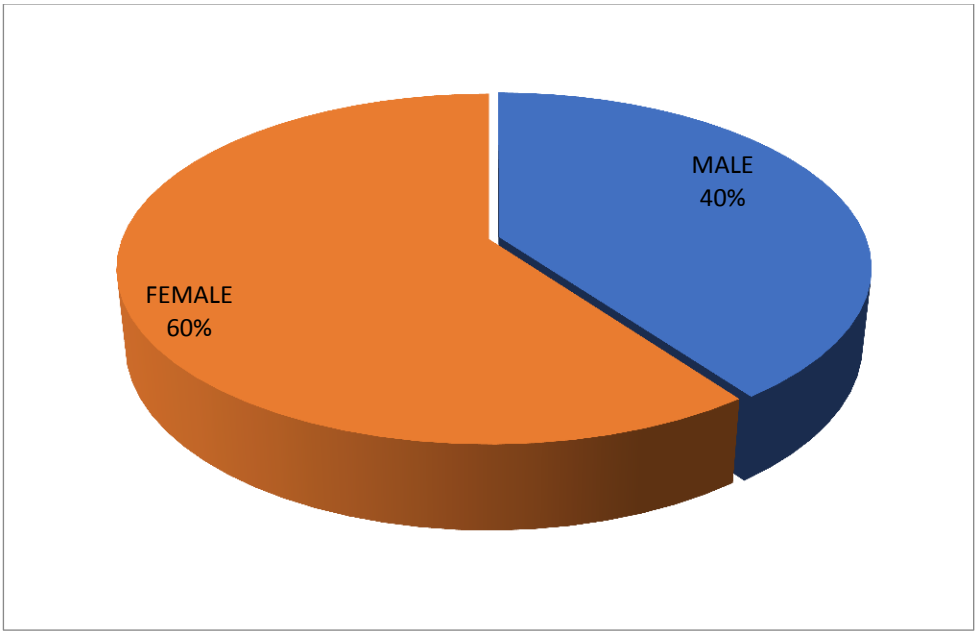
Figure-2: Sex Distribution



The most common sex group affected in the present study was female gender. 60% of the affected patients were females where as 40% were males.

Tabl-1: Clinical Presentation

Symptoms & Signs	Number of Patients
Antenatally Diagnosed	6
Mass Below The Sacrum	20
Constipation	6
Retension of Urine	2
Anus Anteriorly Displaced	20
Rupture of Tumour	1

6 cases were antenatally detected, all the 20 cases presented with mass over the low back, constipation was present in 6 cases, 2 cases presented with acute retention of urine, all the 20 cases anus was anteriorly displaced, one case presented with rupture of the tumor.

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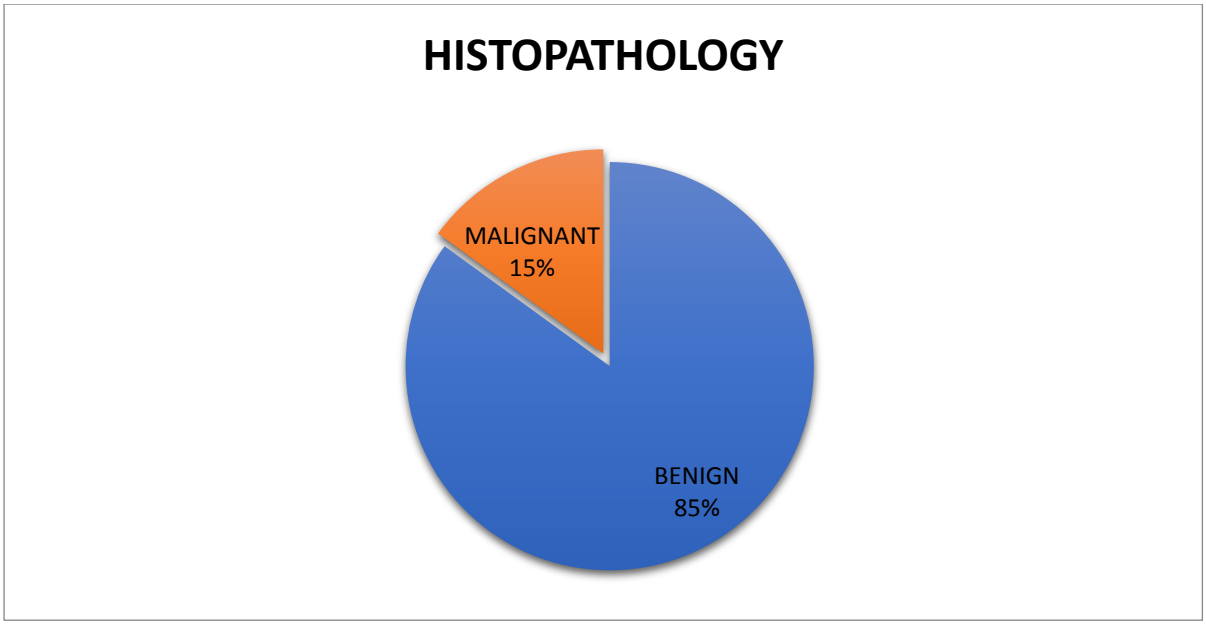
Table-2: Clinical Altmann Stage Of Presentation

TYPE I	TYPE II	TYPE III	TYPE IV
4	12	2	2

4 cases presented with type I (20%), 12 cases presented with type II (60%), 2 cases were type- III (10%), and 2 cases type- IV (10%).

Figure-3: Histopathology



Of the total 20 cases, only 19 cases were operated and biopsy specimen sent for histopathological examination. Of the 19 cases, 16 cases showed benign mature teratomas and only 3 cases were of malignant yolk sac tumors.

Table-3: Post Operative Complications

Post Op Complications	**Number of Patients**
Constipation	4

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Fecal Incontinence	1
Wound Infection	4
Wound Dehiscence	2
Recto-Urethral Fistula	1
Urinary Incontinence	0
Recurrence Of Tumour	0
Mortality	2

Table-4: S**ize of tumor**

S.NO	TUMOR SIZE INTRA OP
1	3x3
2	5X5 cm
3	9X4X3 cm
4	7X5X3 cm
5	15X10 cm
6	12X10 cm
7	5X3 CM
8	3.5X3X3 cm
9	7X6X5 cm
10	4X3cm
11	9X5 cm
12	12X10X8 cm
13	6X5 cm
14	12X10 cm
15	5X4 cm
16	6X5 cm
17	12X10X10 cm
18	6X3cm
19	12X10 cm
20	17X14 cm

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Size of tumor had significant relevance to the tumor out come. Larger the size of the tumor, outcome was poor. One case in which the tumor was 15cm died without operating and the other child with tumor size 17 cm died on 9th post op day.

Figure-4: Mature teratoma with glandular differentiation

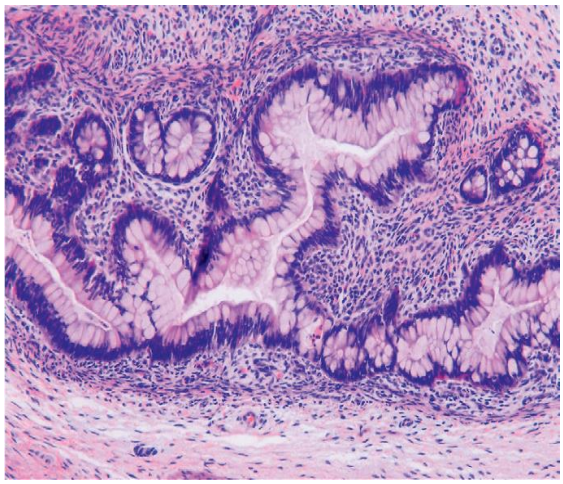

Figure-5: Immature teratomas are graded according to their content of primitive neuroepithelium, and rosette formation .

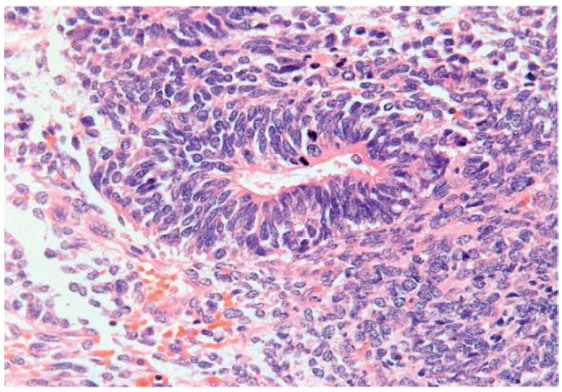

Figure-6:Yolk sac tumor - loose myxoid stroma is filled with a labyrinthine network of cysts lined by clear, flattened epithelial cells. Schiller-Duval body.

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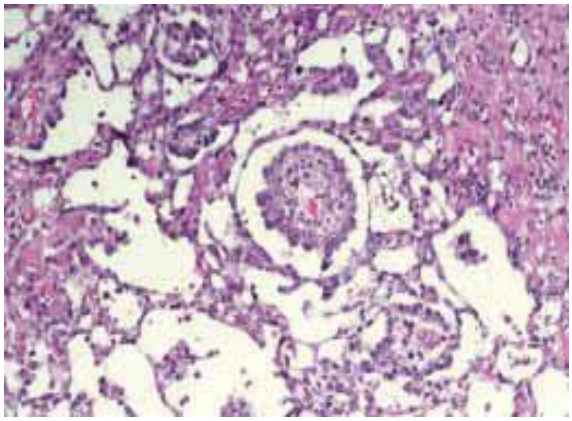
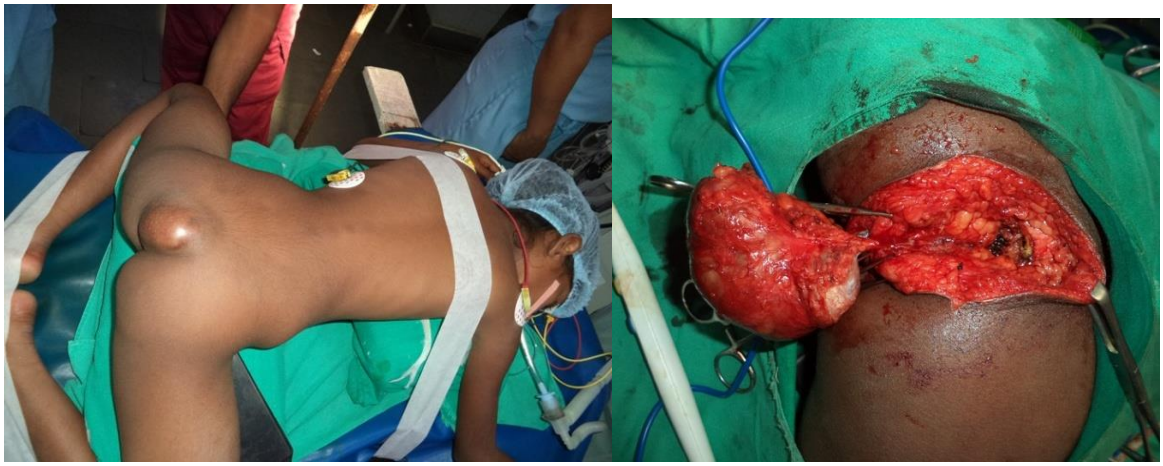


Figure-7:Clinical image of a neonate with a sacrococcygeal teratoma.



DISCUSSION

Commonest age of presentation of SCT was less than 1 month that is about 40% patients; next common age was seen between 1yr to 5yrs. Female patients were commonly affected that is about 60% is females and rests of 40% are males; this type of sex distribution is seen in most of the series. In the present study the commonest type of Altman presentation was type II followed by type I, IV and type III. According to Altman et al[3] types I and II, 56% and 81%, respectively and types III and IV, 44% and 19%, respectively. Because of the antenatal screening[21] with ultrasound which is routinely done, about 6 cases were antenatally diagnosed. In developed countries, many more cases are diagnosed antenatally and therefore antenatal intervention and or planned Caesarian delivery is done for large tumors to avoid dystocia, tumor rupture, hemorrhage, or death.

In all the antenatally diagnosed cases placentomegaly and Polyhydramnios was present.[4] Over half of our patients presented during the neonatal period. This may be due to the fact that most of the tumors were external.[4] Most commonly SCT cases are associated with other anomalies, but in the present study no associated anomalies were noted. Patients with sacrococcygeal teratoma generally present in two clinical patterns; those presenting during the neonatal period with predominantly benign tumors and those presenting after the neonatal period, usually as older infants and children with primarily intrapelvic tumors with a likely malignant, according to chirdan et al[4] Nigerian study. Almost all cases presented with mass

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low back or lump abdomen as in type IV type. 2 cases presented with acute retention of urine and 6 cases presented with constipation.

As one patient with presented with intestinal obstruction , initially diversion that is colostomy and Suprapubic cystostomy was done but later he developed recto-urethral fistula post operatively after tumor excision which is rare complication noted in present study. One case died on 2nd day after delivery due to cardiac failure and tumor rupture. CT scan and magnetic resonance imaging are both reliable and helpful diagnostic modalities which can add to the initial preoperative assessment in determining the anatomic relations of the tumor and the degree of intraspinal tumor extension. Abdomino perineal approach was required in 2 cases, while sacroperineal approach was done in rest of the 17 cases. The surgical approach to SCT has been well established and consists of complete removal of the tumor through a posterior (sacral) or combined abdomenosacral approach, with removal of the coccyx. The importance of removing the coccyx with the tumor was emphasized by Gross[18] and others who reported high recurrence rates of up to 37% when the coccyx had not been excised at the initial procedure. Size of tumor ranged from 3cm to 17 cm, in the present study. Size is very important because very large tumors had poor outcome, as one new born with large tumor died on 2 nd day of life, and the other case died on the 9 th pod. Similar findings were noted by Hambreus et al[5]

In the present study it is identified that impairment was mainly associated with those tumors with a major intrapelvic extension requiring an abdomino-perineal approach for excision. In retrospect, it was impossible to know whether dysfunction was a result of the tumor itself or of the surgery. In the present study 2 patients developed wound dehiscence, secondary suturing was needed in these 2 cases. Wound infection was noted in 4 cases , which were minor complications which resolved with antibiotics and dressings, mainly wound infection was due continuous soiling of the wound with fecal matter which lead to wound infections.

Constipation was noted in 4 cases post operatively, which were managed with laxatives and dietary advices. The management of chronic constipation with soiling should be conservative because it usually improves with age told by Malone et al.[6] No urinary incontinence is noted in present study, but it has been noted in some of studies. Urinary frequency, urinary stress incontinence, and urinary straining in children with sacrococcygeal teratoma may improve over time. (Draper et al)[7] Besides, some of the patients with predominantly intrapelvic tumors may present
with urinary or bowel obstruction as in one of our patients, which mostly suggest malignant diseases at presentation. The high rate of malignancy with late presentation had been noted in many studies. Post-operative morbidity noted in 6 cases, was mainly wound infection and wound dehiscence that could be because of fecal soilage of the wound. Recurrence after excision of benign or immature SCT has been documented and is common after the excision of malignant disease. The initial operation in all these cases was removal of the entire tumor together with the coccyx. It was reported however to be a factor in the postoperative survival due to intraoperative and postoperative complications.

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Serum alfa-fetoprotein in the first month of life and immunohistochemical markers were not found to be of any prognostic significance in patients with SCT. After total resection of the primary SCT together with the coccyx, an elevated serum a-fetoprotein level has been found to be a reliable marker for a recurrence in yolk sac tumor.

In the present series the mean serum alfa -fetoprotein level in the malignant recurrent group was markedly elevated in comparison with that of the benign group. Its real value lies in assessing the extent of the tumor and the presence or absence of distant metastases. The mean time required for AFP level normalization was 9 months. Since none of the parameters which were checked in this series except the tendency of the tumor size were found to be reliable predictors of tumor recurrence, and poor prognosis.

In the present study every 1 month, 6 months and 1 year minimum follow up was done post operatively. The first 3 postoperative years routine meticulous physical examination is essential and indeed the best means of detection of early recurrence.[8]

Serum alpha-fetoprotein levels should regularly be measured. If elevated despite the apparent absence of a clinically detectable tumor, radiographic investigations should promptly be undertaken to locate the presence of a possible recurrent tumor. Bilik et al[9] recommended that every recurrence of SCT should be regarded as being SCT, including gross or microscopic incomplete resection, unresected coccyx, tumor rupture or spillage before or during surgery, immature histology, the presence of microscopic yolk sac tumor foci within the teratoma, female sex, older age, and Altman's classification. However, the relevance of some of these risk factors is still debated. In the present study it has been found no significant association between the occurrence of yolk sac tumor after SCT resection with sex, age, or histology.

Wakhlu et al, 2002[8] had found Zero recurrence in his study. Similar findings are noted in the present study, as there are no recurrences in present study. Similarly, other reports and in some studies revealed that many microscopic Yolk sac tumor foci in SCT were unrecognized at initial routine examinations alone, and they were only identified during central review or re-examination of slides after Yolk sac tumor development. Realistically, meticulous histological examination of huge SCTs, in order to determine the microscopic margin or to search for microscopic Yolk sac tumor foci, may be too labor intensive and unpractical for routine pathological practice.

The mechanism involved in the development of Yolk sac tumor after SCT resection is not fully understood. Some investigators have suggested that Yolk sac tumor develops directly from the teratoma via malignant transformation, while others suggest that microscopic Yolk sac tumor foci present in the preceding teratoma, if not recognized initially, ultimately predominate in the recurrent tumor.(Yoshida et al).[10] The role of postoperative chemotherapy in the management of patients with SCT is still unclear. Some authors have reported that postoperative chemotherapy for immature sacrococcygeal teratomas may decrease malignant relapses[28] while others recommended adjuvant therapy alone for incompletely resected immature teratomas with or without microscopic YST foci .[11] However, many authors do not recommend chemotherapy to avoid treatment-related adverse

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effects, and because salvage therapy would be effective even after the development of Yolk sac tumor **.** Postoperative chemotherapy was not given to any of our patients with mature teratoma. Whereas all the yolk sac tumors were given chemotherapy A stratified, randomized controlled study would help to assess the benefits of adjuvant chemotherapy for SCT. Taken together, we believe that complete tumor resection with coccygectomy and routine histological examination is sufficient for the initial management of patients with SCT. Rectal examination and diagnostic imaging studies may be useful for early detection of recurrence, which was the protocol followed in present study.

One case in present study initially presented with obstruction for which diversion was done that is suprapubic cystostomy and colostomy and after chemotherapy , laprotomy and excision of tumor was done, that child later developed rectourethral fistula post operatively, which is rare complication noted, later soaves pull through was done for that case. However, reconstructing the skeletonized pelvic floor, cosmetic buttock contouring, and preservation of sphincter function are also likely to be important to functional outcome. Fishman et al[16] described a buttocks-contouring closure achieved by bringing the ventral portion of the lateral flaps to a more central posterior location leaving 2 vertical incisions for closure in the midportion of each buttock. It has been reported an increased incidence of malignancy in relation to the surgical type (38% in type IV vs. 8% in type I) and older age at presentation (under 2 months of age, 7% girls and 10% boys have malignant lesions; over 2 months, 48% girls and 67% boys have malignant lesions). Rescorla et al[13] reported that all children presenting after the age of one in their study had malignant tumors. Gabra et al.[12] also reported that 71% of their patients after the neonatal period were malignant.

Duration of study follow up which was only 1 year is the drawback of study. 2 cases needed laprotomy for excision of tumor, and rest of 17 cases were excised by sacral approach. Altman type IV tumors were more likely to require more than one surgical intervention and clearly had the worst outcome regarding their bladder dysfunction compared to other Altman types.(patridge et al).[14]

Ozkan et al[15] reported high rates of hydronephrosis (43%), vesicoureteral reflux (50%), and neurogenic bladder (86%). These findings support a role for direct injury to neural structures supplying the bladder and/or sphincter. Krammer et al[16] has found that on SCT resection in girls may result in diminished sexual function at adult age with worse self-perceived body image. The possibility of sexual complaints should be integrated in the surveillance strategies for these patients. K.O.Soundappan, Walker et al[17] have concluded in their study that overall survival of neonatal SCT is high. While this is a small series, the results are consistent with the literature. Important components of management include timely diagnosis, multidisciplinary planning, long-term follow-up and intervention for functional sequelae. Dorris et al[18] has done study on long term psycho-social outcome in post operative cases of SCT and has found that there is low levels of appearance related distress and has highlighted those in need of psychological intervention.

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COMPARISON WITH OTHER SERIES

study	**No.of patients**	**Male : female**	**Follow up In months**	**Wound dehiscence**	**recurrence**	**Bladder dysfunc-tion**	**Fecal incontin-ence**	**Mortality**
Chirdan et al[4] (2009)	38	7:31	72	8(21%)	3(8%)	3(8%)	2(10%)	3(8%)
Sinha et al[19] (2013)	10	3:7	25	1(10%)	0(0%)	1(10%)	0(0%)	0(0%)
Gabra et al[12] (2006)	33	5:28	96	4(12%)	3(15%)	7(35%)	6(30%)	2(6%)
Present study	20	8:12	12	2(10%)	0(0%)	0(0%)	1(5%)	2(10%)

Similar findings of present study were noted in study done by sinha et al[19], wound complications were noted in 10% of cases, there were no recurrences, but no bladder dysfunction was noted in present study, while the mortality was 10% in present study.

Main difference in comparison to other study groups were the follow up was for long for other study groups while only 12 months follow up was done in present study. This might be a cause that no recurrences and no cases of bladder dysfunction. Wound related complications were almost similar to other studies; Sinha et al[19] also did not found any recurrences and bladder dysfunction cases. Fecal incontinence cases were more in study done by Gabra et al[12]. Mortality was almost same in all study groups.

CONCLUSIONS

Meticulous dissection should be done while removing the tumor to prevent bladder dysfunction post operatively, in many studies bladder dysfunction has been noted. A rare complication was noted in present study that is recto-urethral fistula post-operatively, for which soaves pull through was done and child recovered. Main drawback of the study was follow up was done for one year only that might me the cause that no recurrences are picked in the present study. Concomitant surveillance on urologic and anorectal dysfunction is also essential.

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