

SACRO - COCCYGEAL TUMOURS

(A report of two cases and discussion)

by

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Though the occurrence of sacro-coccygeal tumours has been recognised for many centuries, they still remain uncommon tumours and their mode of occurrence still remain obscure. A Surgeon throughout his career will have a chance to see not many of those tumours. The earliest record of a sacro-coccygeal Terratoma seems to be the one made on a cuneiform tablet of Babylonian origin circa 2000 B. C.

In the last 5 years in this Koshi Anchal Hospital we had a chance to see 3 cases. Two of them were operated here and diagnosis was confirmed by histopathological examination; the other one refused operation and only a clinical diagnosis of sacro-coccygeal Terratoma was made. (This case therefore is not included in this report.)

CLINICAL DATA :

Case 1 :—Saraswati, aged 3½ years, female, was admitted in this hospital on 9.6.2027 with the complaint of a tumour on the sacro-coccygeal area which had been present since birth and had been steadily increasing upto the present size. About 3 months back the summit of the tumour had ulcerated at 2 places and was discharging pus through those openings.

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On examination she was a rather undernourished, anaemic female child. Her growth was normal. There were no other congenital anomalies.

There was a globular tumour 5"x4½" arising from the sacro-coccygeal region. The skin over it was ulcerated at two places, discharging pus. The feel was varied, solid at places and cystic at others. The base of the tumour was broad and was adherent to underlying structures in the middle. A clinical diagnosis of sacro-coccygeal teratoma was made and after a short course of parenteral and oral haematinics the patient was operated on 16.6.2027.

The operation was carried out in general anaesthesia with the patient in prone position. An elliptical incision was made in vertical axis, leaving as much skin just sufficient to cover the defect during closure. The tumour was well encapsulated, but has to be separated from gluteal muscles of both the



Fig (a)



Fig (b)

sides, and lower 3 pieces of sacrum by sharp dissection with knife, but the tumour was intimately blended with the coccyx. The coccyx was excised along with the tumour safeguarding the rectum. There was no pelvic extension. The wound was closed with a drain, which was taken out after 48 hours and a padded dressing was applied. She made an uneventful recovery and was discharged from the hospital 10 days after the operation, having no sphincteric or neurological disturbances. The patient is healthy since then.

The specimen showed solid and cystic areas with pieces of cartilage here and there. The cysts contained gelatinous fluid.

Histological examination confirmed Teratoma with no evidence of malignancy.

Case 2 :

Dev Kumari, aged 5 years, female, was admitted in this hospital on 22.9.2028 with a lump and a leg hanging from sacral region present since birth which has been growing steadily along with the growth of the child. Fig. (a) and (b).

This child was one of the twins, her twin was also a female, alive and healthy uptill now and has no congenital anomalies. She has an elder brother 2 years senior, who is

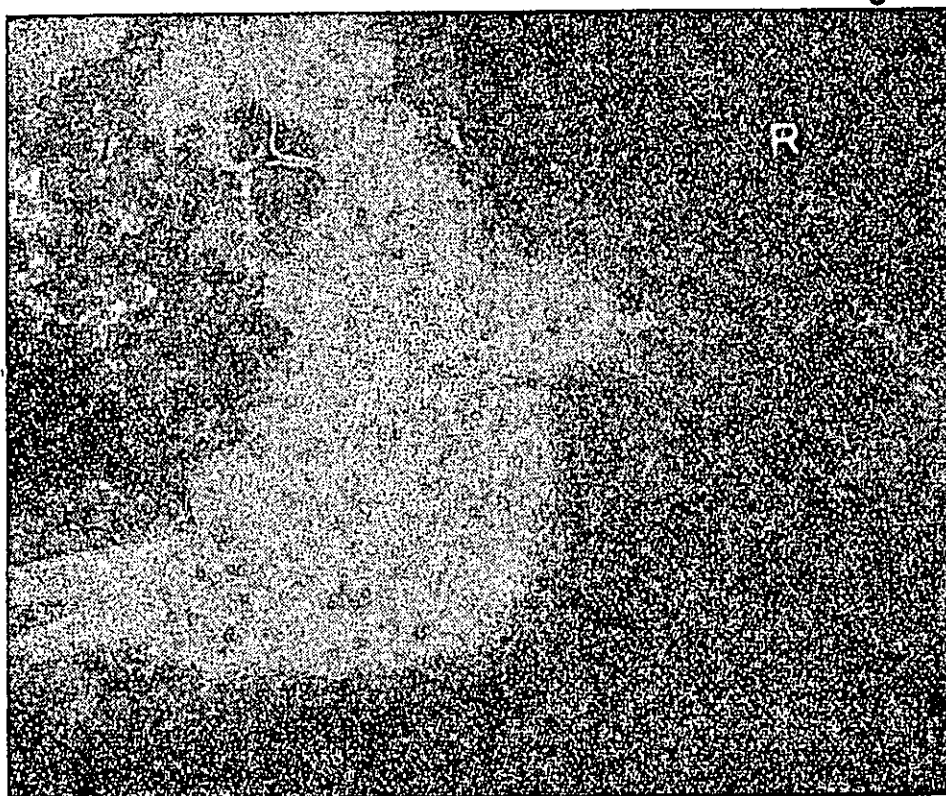


Fig. (c)

healthy. On examination the patient was a very uncooperative child with a fair growth having no congenital anomalies except the tumour. The tumour was situated in the sacro-coccygeal region, 15 cm. x 10 cm. with a leg hanging from its right side reaching upto upper 1/3rd. of the patient's thigh. (obviously it is the left leg of undeveloped triplet). The other leg was missing but was replaced by a small swelling with a scar on its top at the opposite side. The parents had tied hair at the base of this ? growing leg in infancy and the leg was cast off by process of avascular necrosis. A fairly developed penis was protruding from the summit of the growth with well developed prepuce and glans

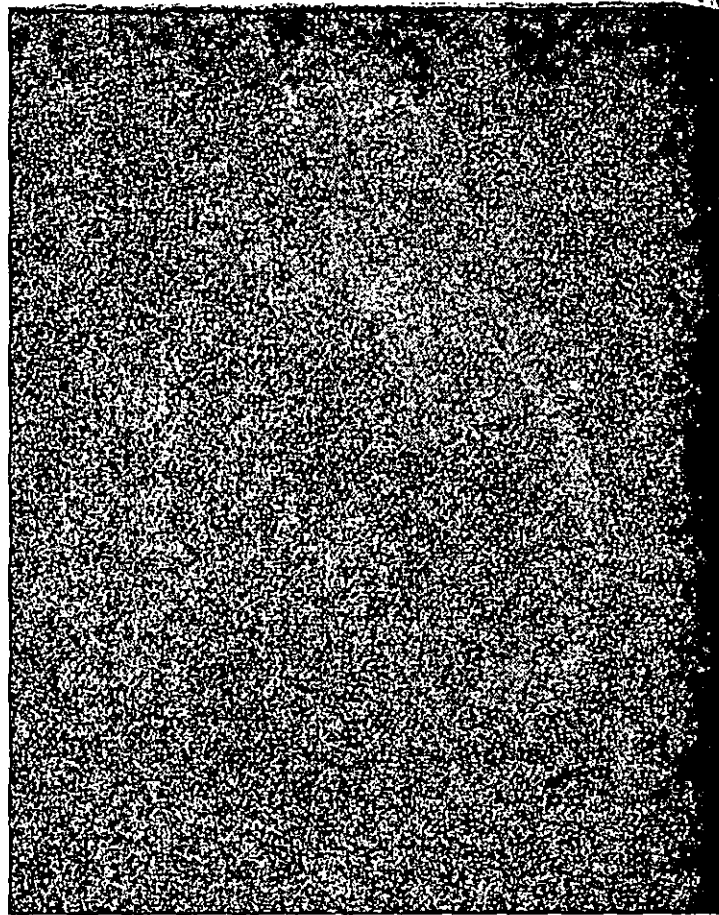


Fig. (d)

which was exuding drops of pus-like fluid through the urethral opening.

X-ray (fig. c and d) shows underdeveloped pelvic bones attached to upper sacrum with a long bone in the limb with a prominent lower epiphysis (? underdeveloped Femur), then a short bone (? underdeveloped Tibia), then small rounded bone (? underdeveloped Tarsus) then 2 metatarsals, one of them having another metatarsal attached to it, the phalanges of 2 toes.

Chest X-ray and haemogram of the patient were normal.

The patient was operated on 26.9.2028 in general anaesthesia with endotracheal intubation in prone position.

An elliptical incision was made in a transverse axis through skin and subcutaneous tissues. The tumour was freed easily on the right side but the pelvic bones of the tumour had formed a cartilaginous joint with the left Iliac crest and sacrum of the patient which had to be excised by chiselling off using bone cutting forceps. In the centre

underneath the tumour there was a sac with boggy feel which lodged in a cavity made partly by deficiency of sacrum in the middle. This was enucleated from the defect and pelvic cavity without much difficulty. There was a fairly developed vessel and a nerve coming from the spinal canal supplying the tumour. The nerve was divided, the vessels ligated and divided and the tumour was removed. The coccyx was not involved at all. The gap in the sacrum was closed by flaps of fascia and muscle mobilized from glutei and sutured together.

The removed specimen is shown (Fig. e and f). A probe passed through the urethra reached the sac (tip of probe held by forceps in Fig. e). The sac when opened contained about 2 ozs. of thick greasy material, not much unlike meconium.

The histopathological report reads as follows :

Report :

Specimen : Mass of tissue complete with skin measuring 10 X 7. cm (excluding leg) appears like a half pelvic part with an attached leg and a rudimentary penis. No scrotum

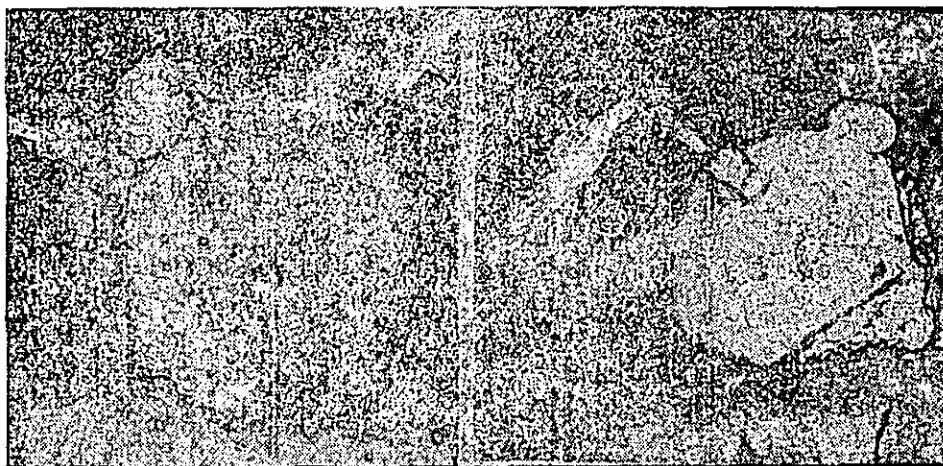


Fig. (e)

Fig. (f)

or scrotal fold seen. The external surface is covered by skin and the inner tissue mass is formed by bone and fibrofatty tissue. The penis protrudes 0.5 cm. The exposed glans penis is partly covered by dark coloured prepuceal skin. The leg is attached to one side of the tissue mass (left leg). Right leg is missing. The length of the leg is 12 cm and is slightly curved with convexity outward. No knee joint found. Smoothly curved leg ends in a foot which measures 10 cm from heel to toes. There are only two rudimentary toes. The shaft of the penis when dissected is 9 cm. long and ends in a blind pouch measures 5x4 cm. The pouch is lined by velvety mucosal folds. Section shows intestinal pattern of mucosa.

The patient had postoperative wound infection which settled with antibiotics and dressing and was discharged home on 3.11.2028, having no neurological complications.

DISCUSSION :

Classification of sacro-coccygeal tumours is difficult, because, though they have many things in common yet their histologic pattern is varied. For convenience they can be classified into three groups.

GROUP — A

Tumours of the Notochord : Chordoma :— Notochord is the primitive entodermal axis around which the mesoderm of vertebral column develops. It disappears in early foetal life almost completely, but traces may persist at the base of skull, Nucleus pulposus of intervertebral disc and in the region of coccyx.

The tumour arising from this is called chordoma and is usually locally malignant, infiltrating and destroying the surrounding structures, bone, nerve, pelvis, bladder, rectum etc. Occasionally there is metastasis. The tumour and infiltrating mass is gelatinous.

The treatment consists of radical resection, but the prognosis is poor, due to local recurrence and for the fact that they are radio-resistant.

GROUP — B

Tumours arising from anomalous development in the early Embryo :—These tumours are commoner in females. Both of our cases were also females. This group for convenience can be subclassified into two groups.

- (a) Parasites composed of definite organs or their rudiments e. g. limbs, bones, viscera, forming an incomplete twin. These tumours have no more chance to get malignant than any other portion of the body of the host. Probably these arise from a single Blastomere. One develops completely, the autosite, and another incompletely, the parasite.

Our case No. 2 falls in this group but is interesting for the fact that the parasite represents a triplet, if developed completely, the score of the mother would be three at a time, 2 daughters and a son, because the parasite represents the lower part of the body of a male. The treatment is excision as we did, the incision used is a matter of choice, necessity and convenience.

- (b) Sacro-coccygeal Teratoma—Our case No. 1 falls in this group. The origin of this tumour is still not very clear, but it is very likely that they arise from an abortive attempt at forming twins, the tissues of the twin getting incorporated into the body of the sister or brother. This seems plausible, remembering that most conjoined twins are attached to each other in the sacro-coccygeal area. These tumours are of multiple cell types having preponderance of tissues developed from two or more primitive cell types, ectodermal, mesodermal and or entodermal. So they may have areolar and

fibrous tissue, cartilage, bone, mucoid or nervous tissues. Mostly they are situated on the dorsal aspect of coccyx and sacrum, but may be present anteriorly or in the pelvic aspect or might be a dorsal one with pelvic extension. They may be of any size, varying from a marble to large one causing unsightly swelling or pressure effect in bladder, rectum or even dislocating the child's hips. Usually they develop in infancy or are present since birth but occasionally they are noticed in adult life. Mostly these tumours are benign but if left alone may turn malignant causing distant metastasis. There may be few which might be malignant to start with. The treatment consists of excision using either an inverted V incision or elliptical incision (as we did in case 1) for a dorsal type tumour. The coccyx should be removed in all cases with the tumour to prevent recurrence. If there is a pelvic protrusion it can either be enucleated through the same incision or a laparotomy can be performed on the same sitting or dorsal portion removed in one sitting and pelvic portion removed after few days by laparotomy.

GROUP — C

Tumour arising from persistent rudimentary structures :—These tumours arise from remnants of neurenteric canal, the postanal gut and proctodeal membrane. They are usually ventral to sacrum and coccyx but may get displaced dorsally later on. They are usually tumours containing thick mucus, congenital anal post cysts and pilonidal cyst or sinuses also arise in relation to the skin dimple at the tip of coccyx; they are formed either by traction due to retrogression of the tail bud causing traction dermoid or from the remnant of coccygeal vestige of spinal column. Treatment is excision.

SUMMARY :

2 cases of sacro-coccygeal tumours have been discussed. Both were females. In both the tumours seem to have originated from abortive attempt at forming twins, forming a Teratoma in case No. 1 and a parasite in case No. 2. It is interesting to note that in case No. 2 the tumour was an abortive attempt at forming a triplet.

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