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Research Paper Medical Science

Large Adult Sacrococcygeal Teratoma: A Case Report And Review Of Literature.

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ABSTRACT

Sacrococcygeal teratomas (SCT) are neoplasm containing tissue derived from more than one primitive germ layer and its cause remains unknown. They (SCT) is one of the most common tumours in infants, but are rare in adults.The incidence inneonates is 1 in 40000 with a female to male ratio of 10:1[1]. It is rare in adults with less than a hundred cases being documented in the literature[2].Most of the sacrococcygeal tumours are cystic and benign with 12% being malignant. Treatment consistsof early complete surgical resection, as it has the potential for malignant transformation which increase with age[3]. We present a case of a large adult sacrococcygeal teratoma in an female and review of its literature. The clinical features, radiological findings and outcome are described.

Keywords : Sacrococcygeal teratoma, presacral teratoma, germ cell tumours

A 45 years old female presented with a large mass in the right glueteal region. She had difficulty in sitting, no pain, no bowel or bladder complains. She had noticed the mass which had slowly grown over the last twenty years to attain the present size. She had not consulted a doctor as it was painless, but due to its large size she had difficulty in sitting. The patient has had a normal obstetric history. On admission we did her routine investigations which were within normal limits. On examination a mass in the right gluteal region measuring about 12 x 6 cms, oval in shape, skin was stretched with a few dilated veins, it was non-tenderwith firm consistency.

On CT scan alarge mass was seen anterior to the sacrum and coccyxcompressing the urinary bladder and sigmoid colon and anteriorly displacingthe rectum. It was extending inferiorly into the right ischiorectal fossa. It was a complex mass with an intact sacrum and no invasion of surrounding structures.The patient was operated under general anesthesia with endotreacheal intubation in a jack knife position, a vertical incision was made and a large mass present in the gluteal region was excised. The mass was seen extending up between the sacrum and rectum, this retro rectal mass could be excised by creating a plane between the mass and rectum. Coccygectomy was done to prevent recurrence. Haemostasis was secured and two drains were inserted. Histopathology examination confirmed the diagnosis of benign mature teratoma, with the presence of tissue derived from all three germ layers. There was no evidence of immature cell types or any malignant change.Post-operative recovery was uneventful. The patient was called for follow up every two months in the surgical OPD for the first year. At the end of one year no recurrence was seen on CT scan, she has been advised regular follow up once in six months for another two years.

Discussion

A teratoma is a tumour composed of multiple tissues or cell types foreign to the site of origin.Sacrococcygeal teratomas (SCT) are neoplasm containing tissue derived from more than one primitive germ layer3 and its cause remains unknown4,5. It is the most common tumour in newborns and the incidence is 1 in 40000 with a female to male ratio of 10:11. It is rare in adults with less than a hundred cases being documented in the literature2. The incidence of malignancy increases with age3.Sacrococcygeal area is one of the most common sites for teratoma in the new born6,but in adults the tumors at this site are very rare and seen in the gonads (ovaries and testes), the anterior mediastinum, the retroperitoneal space, the presacral and coccygeal areas, pineal and other intracranial sites, the neck, and abdominal viscera7. The tumours may be cystic, solid or mixed8,9 They have been classified morphologically according to their relative extent outside and inside the body by the Altman classification as:-

Type I entirely outside, sometimes attached to the body only by a narrow stalk

Type II mostly outside

Type III mostly inside

Type IV entirely inside; this is also known as a presacral teratoma or retro rectal teratoma.

Little is known about the embryogenesis of SCT, but it has been hypothesized that the anomaly is derived from a primitive knot, also referred to as Henson's node10. SCTs are thought to originate from multipotential cells in Henson's node, which migrates caudally to rest in the coccyx. Various other theories exist to explain the origin of SCT.These include nonsexual reproduction of germ cells within the gonads or in extragonadal sites; 'wandering' germ cells of non-parthenogenetic origin left behind during the migration of embryonic germ cells from yolk sac to gonad; or origin in other totipotential embryonic cells11.

Sacrococcygeal teratomas (SCT) are the most frequent solid congenital tumours in the foetus and the new born. It is rare in adults with only a few published cases in literature12.The sacrococcygeal area is the most frequent site of teratoma in infancy. This tumor rarely presents in adulthood, and is

A female, rather than male, predominance exists2,12with a ratio of about 10:112,13. The clinical presentation varies on the location and extent and symptoms are related to mass effect such as back pain, bowel & bladder dysfunction or they may have neurological symptoms like lower extremity numbness10,11.

Biochemical markers like AFP, carcinoembryonicantigen, and HCG may be elevated in patients with malignant lesions and also help in detection of recurrence after surgery14.

Large mass may cause displacement of the uterus and rectum which can be felt on digital examination. Patients may also present with abscess or perirectal fistulas15. Unlike teratomas in infants, which are externally visible in 90% of cases, sacrococcygeal teratomas in adults are confined mostly to the intrapelvic space16. There is a tendency in the neonates toward malignant transformation of SCTs with increasing age17.However in adult patients benign tumors predominate18.Adult patients with SCT may be asymptomatic with the diagnosis being an incidental finding in a digital rectal examination or as a retro- rectal mass on CT Differential diagnoses are anterior meningeocele, rectal duplication cysts or anal gland cysts. Complete surgical excision is the treatment of choice. Excision of the coccyx may be necessary because the bone may contain a nidus of pluripotent cells with a risk of recurrence19.For benign disease, no further adjuvant therapy may be necessary. Traditional treatment of sacrococcygeal teratomas favors always removing the coccyx with the tumor, since failure to do so results in an approximately 30% to 40% recurrence rate. Many case reports fail to recognize the important distinction between isolated presacral teratomas (defined here as those with no apparent tissue attachment to the coccyx) and true sacrococcygeal teratomas which have attachments to the coccyx.

Symptoms specific to the sacrococcygeal and presacral tumor locations are those of mass effects displacing contiguous structures, rectal pain, backache, paresthesias,

numbness in the so-called saddle area and legs, and loss of bladder and rectal control. Failure to differentiate between the two entities could lead to unnecessary coccygectomies. Complications seen with coccygectomy are delayed wound healing

with increased wound infection rate, increased recovery time, and rarely, bowel

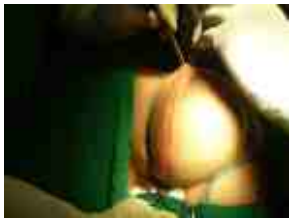
herniation20,21. Access to the tumour could be by transabdominal approach or the transperineal route using the jack-knife position or a combined approach. Complete excision has extremely good prognosis, and is necessary to prevent local recurrence and potential malignant transformation. The sacrococcygeal approach or combined abdominal-sacral approach is recommended22.

Conclusion

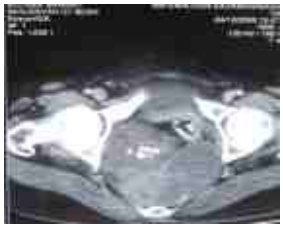
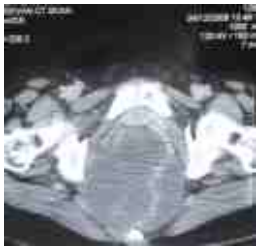
Although rare in adults, sacrococcygeal teratoma should be considered in the differential diagnosis of patients with a pelvic mass presenting with obstructive symptoms. Long-term survival is possible with adequate resection of the tumor. The presence of leptomeningeal involvement and malignant transformation are associated with a less favorable outcome.Surgical removal is generally indicated at the time of detection, as these lesions carry a significant malignant potential. Differentiation between presacral teratoma and sacrococcygeal teratoma is important to prevent unnecessary coccygectomies.

Large sacrococcygeal teratoma in adult


Intra op Post-op




Ct scan

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