Sacrococcygeal teratoma

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- A familial case of VACTERL association with co-occurring sacrococcygeal teratoma: a case report

Extragonadal germ cell tumors were thought initially to be isolated metastases from an undetected primary tumor in a gonad, but it is now known that many germ cell tumors are congenital and originate outside the gonads. The most notable of these is sacrococcygeal teratoma, the single most common tumor diagnosed in babies at birth.

It is found predominantly in females and discovered in the neonatal period.

These tumors are believed to arise from embryologically multipotent cells of Hensen's node, which lies within the coccyx [1)].

Classification

The tumors are classified, according to Altman Classification of the Surgical Section of the American Academy of Pediatrics, into four types:

Type I, those tumors that are predominantly external projecting from the sacrococcygeal region and presenting with distortion of the buttocks

Type II, those tumors that are predominantly external, but have a large intrapelvic component

Type III, those that are predominantly intrapelvic with a small external, buttock mass

Type IV, those that are entirely internal with no external or buttock component.1 Type IV sacrococcygeal teratomas may occur as a familial form inherited as an autosomal dominant condition. In this entity, a presacral mass (teratoma, anterior meningocele, dermoid cyst, lipoma, neurofibroma, enteric cysts, or hamartoma) is associated with anal stenosis and typical Scimitar defect of the sacrum. This is often referred to as Currarino triad. Approximately 45% of teratomas is Type I, 35% is Type II, 10% Type III, and 10% Type IV.

Intradural sacrococcygeal teratoma (SCT) is a rare entity that has been reported in only a few cases previously. Shahjouei t al. present the case of a 2-week-old, otherwise healthy neonate with a mass in the buttock. The imaging findings and the high level of serum alpha-fetoprotein were highly

suggestive of SCT. On operation the authors found intradural extension of the teratoma. The lesion was managed successfully without any remaining sequelae [2].

Complications

Yolk sac tumors (YSTs) developed after sacrococcygeal teratoma (SCT) resection not only in patients with previously reported risk factors. Therefore its recommended that patients undergo serum AFP monitoring every 3months for ≥3years after SCT resection [3].

Case reports

Serratrice et al. described an immature ruptured giant sacrococcygeal teratoma (GSCT) complicated by hemorrhagic shock at a 32-week gestation boy requiring an emergency delivery, followed immediately by urgent surgical removal. A brain lesion resembling a giant hypothalamic hamartoma (GHH) was also present on the antenatal MRI. In order to exclude metastatic immature teratoma or glioma, a biopsy was performed by a retro-sigmoidal approach, which confirmed the nature of the hamartoma. Serratrice et al. described for the first time the association of a ruptured immature GSCT associated with a GHH [4].

1)
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2)
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3)
Yoshida M, Matsuoka K, Nakazawa A, Yoshida M, Inoue T, Kishimoto H, Nakayama M, Takaba E, Hamazaki M, Yokoyama S, Horie H, Tanaka M, Gomi K, Ohama Y, Kigasawa H, Kitano Y, Uchida H, Kanamori Y, Iwanaka T, Tanaka Y. Sacrococcygeal yolk sac tumor developing after teratoma: a clinicopathological study of pediatric sacrococcygeal germ cell tumors and a proposal of the pathogenesis of sacrococcygeal yolk sac tumors. J Pediatr Surg. 2013 Apr;48(4):776-81. doi: 10.1016/j.jpedsurg.2012.08.028. PubMed PMID: 23583133.
4)
Serratrice N, Faure A, de Paula AM, Girard N, André N, Scavarda D. Description of a giant hypothalamic hamartoma associated with an immature ruptured giant sacrococcygeal teratoma: a case report. Childs Nerv Syst. 2020 Sep 25. doi: 10.1007/s00381-020-04894-y. Epub ahead of print. PMID: 32978641.