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CASE REPORT

Diagnostic Dilemma in Ruptured Cystic Sacrococcygeal Teratoma: A Case Report

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ABSTRACT

Sacrococcygeal teratoma (SCT) is the most common tumor of the newborn. Diagnosis is clinical and frequently straightforward. However, varied presentations can occasionally result in a diagnostic dilemma. We present a case of ruptured cystic Altman Type 1 SCT along with pertinent review of literature. The purely cystic variant of SCT is rare in itself and presenting as a ruptured lesion added to the dilemma in diagnosis and management.

Key words: Cystic sacrococcygeal teratoma; Ruptured myelomeningocele; Neonate

INTRODUCTION

A sacrococcygeal teratoma (SCT) is the most common tumor in the neonate. These are most often mixed solid/cystic, although purely cystic types can occur in ~15% of cases. Predominantly, cystic lesions are rare and almost always benign [1-3]. A case of pure cystic variety of SCT (Altman Type 1) is reported that presented as ruptured lesion.

CASE REPORT

A full-term vaginally-delivered female neonate (birth weight = 2.8 kg) presented in the emergency with a ruptured cystic lesion over the lower back with watery discharge, on the 2nd day of life. On examination, she had a skin-covered sac at the sacrococcygeal region extending from the coccyx to anal verge and extending over both buttocks (Figure 1a). The length of sac was around 8 cm beyond the coccyx and the terminal skin of sac was necrotic. Rectal examination showed a lax sphincter; there were no other obvious neurological deficits. The child was admitted with a provisional diagnosis of ruptured myelomeningocele (MMC) in the department of neurosurgery. The magnetic resonance imaging, however, suggested no spinal cord communication with the lesion and no spinal dysraphism (Figure 1b). The neonate was then transferred to pediatric surgery department for further management. The alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin were within normal limits for her age. The baby was taken up for surgery and the lesion was excised completely along with the coccyx (Figure 2). Biopsy was suggestive of mature cystic SCT. The infant is under follow-up and is continent for bowel and bladder.

DISCUSSION

Purely cystic variants of SCT are rare [3]. Rarely, if these cystic lesions get ruptured they can add dilemma in diagnosis and management.

Large fetal SCTS can cause hemorrhage, preterm labor, dystocia, and obstructed labor. However, cystic variants also run the risk of rupture during delivery. Solid SCTs are associated with increased AFP levels and risk of high-output cardiac failure and hydrops [4]. To minimize risk of rupture, cesarean delivery is recommended for fetuses with SCTs of >5 cm in diameter.

There have been case reports where a cystic SCT has been misdiagnosed as other lesions. The differential diagnosis for sacrococcygeal masses includes meningocele, MMC, myelocystocele, teratoma, lipoma, hamartoma, lymphangioma, hemangioma, chordoma, and ependymoma [5]. Our case was also misdiagnosed in the emergency room as MMC. The clinical points that were in favor of SCT that the lesion extended from the coccygeal region to the anal margin and involving both

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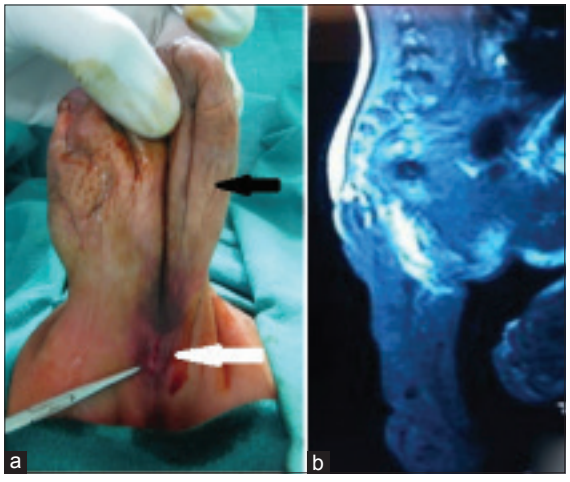

Figure 1: (a) Clinical presentation. Black arrow; protruding sac (cystic lesion). White arrow; anal opening. Note the close approximation of anal opening with the sac. (b) Magnetic resonance imaging of the lesion; no spinal cord communication with the lesion and no spinal dysraphism

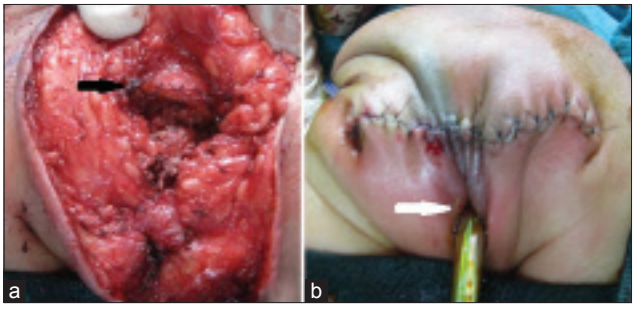

Figure 2: (a) Intraoperative image; completed dissection. Black arrow; removal of coccyx along with the lesion. (b) Immediate postoperative image. White arrow; Hegar dilator in anal opening

buttock areas (slightly caudal to where one expects a sacral MMC) and no neurological deficits. However, there have been case reports of SCTs extending into the spine and causing neurological deficits. The key imaging distinction between these cystic masses is the dysraphic spine and the continuity of the protruding sac, with the spinal cord central canal. SCT is not a neural tube defect, and there is typically no spinal dysraphism, but all of the four Altman types might have an intraspinal component [6].

Spinal involvement may be associated with neurologic deficit. Furthermore, extensive dissection during surgery in the pelvic and perineal region can lead to disruption of nerves and muscles that can compromise urinary and fecal continence in post-operative period.

The recommended treatment of a cystic SCT is same as for any SCT, i.e., complete excision (including coccygectomy), as in this case, and it is adequate if the tumor is benign. Chemotherapy and radiotherapy are, however, indicated for malignant cases.

To conclude, the cystic variety of SCT is rare but should always be considered as a differential in any cystic lesion over the sacrococcygeal region even if it is ruptured. The further caudal location of SCT is a helpful indicator. Cesarean section should be considered for cystic SCT. The management of cystic variants remains the same and prognosis is good, especially since these lesions are rarely malignant.

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