

SACROCOCYGEAL TERATOMA IN AN ADOLESCENT

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Sacrococcygeal teratoma (SCT) is a neoplasm derived from more than one primitive germ cell layer. SCTs have rarely been reported in adults. We present a case of a 17-year-old youth who complained of a painful mass over his left buttock. The mass had been growing gradually since his childhood. Magnetic resonance imaging showed a well-defined soft-tissue mass which contained mainly fat tissue and large intestine. The patient had complete excision of the tumor and coccyx. The gross impression and histology revealed a mature teratoma.

Key-word: Teratoma.

Sacrococcygeal teratoma (SCT) is a neoplasm arising in the sacrococcyx and contains tissue derived from more than one primitive germ cell layer. Most SCTs are found in neonates, but they occur rarely in adults (1-4). We present a case of SCT in an adolescent.

Case report

A 17-year-old young man suffered from a painful mass over his left buttock. The mass had been growing gradually since his childhood. The mass was elastic, with mild tenderness and without local warmth or erythema, and pushed the anus from the midline. The skin and anal opening were intact. No bowel habit discomfort was noted. Laboratory findings, including beta human chorionic gonadotrophin and serum alpha-fetoprotein, were negative. Magnetic resonance imaging (MRI) showed a well-defined, lobulated, cystic and fat-containing mass lesion in the left gluteal region (Fig. 1A-C), with mild peripheral enhancement on postcontrast images (Fig. 2A, B). There was no communication to the anus or thecal sac. Computed tomography (CT) scan showed no calcification within the mass. The preoperative diagnosis was sacrococcygeal teratoma. The tumor and coccyx were completely excised (Fig. 2C, D). Histopathology showed that the mass included a large cyst with a brownish fibrous wall. Two protruding nodules were found on the anterior surface. One nodule was attached to a small fragment of the coccygeal tip. The mass also contained fat tissue.



Fig. 1. — A. Sagittal T2-weighted image shows colonic haustrations with fluid retention in the mass lesion (arrow). B. In-phase MR image shows a hyperintense area (arrow) in the mass lesion. (C) Out-of-phase image shows signal loss at the interface (white arrow) of the fatty component (black arrow) and the water-based cystic mass.

Microscopically, the tumor was diagnosed as a mature teratoma with multiple cystic spaces, mainly composed of intestinal tract tissue (Fig. 3). The mucosa of the intestinal tract showed marked degeneration. The other components included scant residual simple mucinous epithelium, respiratory tract wall of ciliated pseudostratified columnar epithelium, a muscular layer with

scant cartilage, pancreatic tissue, meningeal tissue with meningo-epithelial cell proliferation, and psammoma bodies. Fat necrosis with infiltrating macrophages and focal hemorrhage were present. No focal malignancy was noted.

Discussion

Sacrococcygeal teratoma (SCT) is a neoplasm arising in the sacrococcygeal region and contains tissue derived from more than one primitive germ cell layer. The prevalence of benign SCT is approximately one in 35 000 to 40 000 births (5), mainly in females (10:1), and 90% are diagnosed in the newborn period and are benign (6). Most adult SCTs are intrapelvic, whereas external masses

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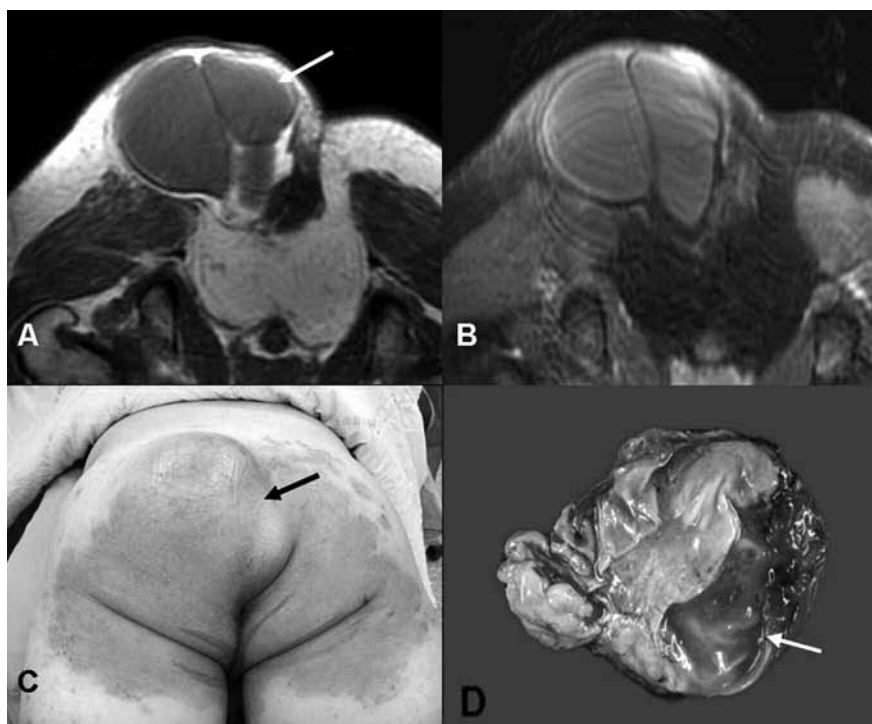


Fig. 2. — A. Axial T1-weighted MR image and B. postcontrast fat-saturation T1-weighted image show cystic mass lesion (white arrow) with fat component. Mild peripheral enhancement of the tumor is noted. C. A protruding huge mass (arrow), with redness of overlying skin, over the patient's left buttock. D. Surgical gross appearance of the buttock mass, with cystic component (arrow).

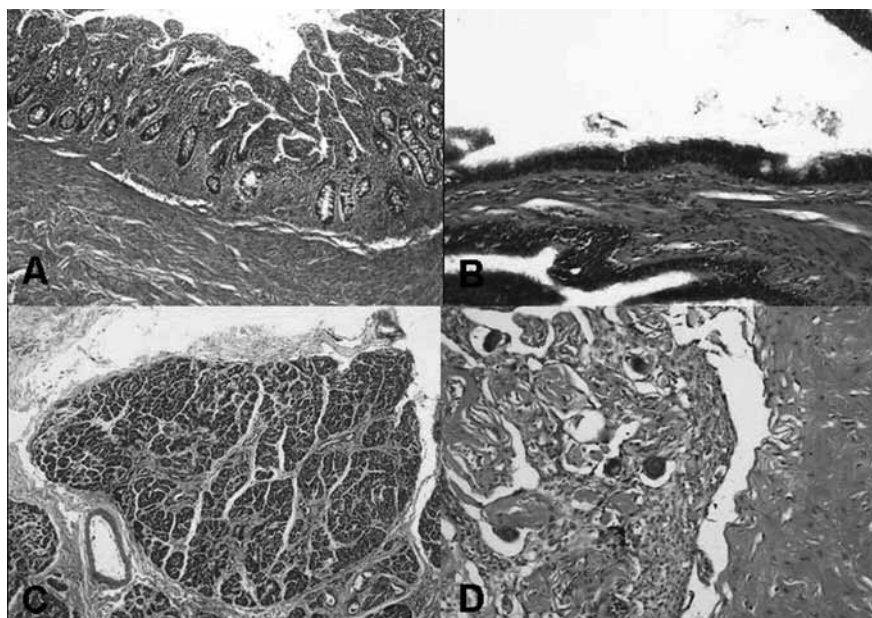


Fig. 3. — Photomicrographs show (A) colon tissue with mucosa and muscle layers, (B) respiratory tract structure with ciliated pseudostratified columnar epithelium, (C) pancreas tissue, and (D) meningeal tissue with meningotheelial cell proliferation.

are common in infants. Most adult SCTs are cystic, and only 1 to 2% are malignant (2, 3, 5).

SCTs are classified into three histopathologic categories (5): mature,

immature, and malignant. Mature teratoma (also known as benign teratoma) contains epithelium-lined structures, mature cartilage, and striated or smooth muscle. Immature

teratoma has areas of primitive mesoderm, endoderm, or ectoderm mixed with more mature elements in higher cellular stroma, with mitotic figures. Malignant teratoma has malignant tissue of germ cell origin, such as germinoma and choriocarcinoma. Malignant teratoma has a predominant solid component; hemorrhage and necrosis are common (7). Tumors containing malignant, nongerminoma elements, including adenocarcinoma and squamous cell carcinoma, are referred to as teratoma with malignant transformation.

The role of imaging is to determine the tumor location and extent. There are four types of SCT (5, 7): type I, predominantly external masses with a small presacral component; type II, external masses with a significant intrapelvic component; type III, external masses with a pelvic and abdominal component; and type IV, internal masses with an intrapelvic and abdominal location. Approximately 50% of SCTs are type I lesions, and 38% of type IV tumors are malignant (7). Our case can be classified as type I SCT.

CT scan is the most sensitive method of demonstrating calcification or ossification (which may be visible in over 50% of benign tumors) and integrity of adjacent bone. Calcifications are seldom observed in malignant tumors (7). At CT or MRI, SCTs appear as heterogeneous, well-defined masses with mixed cystic and solid components. Fat-fluid or fluid-debris levels may appear in SCTs with complex cyst contents; entirely cystic SCTs are uncommon (6). MRI better depicts the cystic elements, the contents of which might be inferred from signal-intensity patterns on different pulse sequences. Fat components, which occur in 50% of SCTs, can be identified by either CT or MRI. CT and MRI are complementary in the evaluation of sacrococcygeal column anomalies, such as spinal dysraphism, sacral agenesis, pressure erosion, and remodeling, which can be associated with SCT (3).

Differential diagnosis of SCTs should include meningocele, rectal duplication cyst, and anal gland cyst. Anterior sacral meningocele is defined as a meningeal cyst that occurs in the presacral space secondary to agenesis of a portion of the anterior sacrum. Meningoceles are usually seen with sacral defects, hernial sac, nerve roots, dysraphism and the meningocele neck, which communicates with the thecal

sac (7). This could be excluded by our MRI findings. Rectal duplication cysts most frequently are spherical cystic lesions in the presacral region. They may communicate internally with the anorectal lumen or externally with the skin surface (7). Rectal duplication cysts seldom contain fat or calcification, and are not usually solid or multiloculated (2). Typical anal gland cysts are mucus-secreting cysts, communicating with the dentate line or anal duct and occurring anywhere around the anorectum. They are commonly a uniloculated or multilocular cystic lesion near the anal sphincter in the retrorectal space, and may involve the coccyx (6). Our case, unlike an anal gland cyst, contained both fat and cystic components.

Complications are most frequently seen in type IV SCTs. Fetal hydrops,

hemorrhage, or rupture of the SCTs are the main complications, associated with a high mortality rate in neonates (6).

Complete excision of the tumor and coccyx is the treatment of choice. Failure to remove the coccyx has been associated with a high risk of recurrence. Mature SCT is potentially curable and the prognosis is excellent.

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