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Case Report

Primary Retroperitoneal Mature Cystic Teratoma (Dermoid Cyst) in Pelvic Musculature

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Abstract

Dermoid cysts, or mature cystic teratomas containing ectoderm-derived skin adnexa, rarely occur intramuscularly, especially within the male pelvic musculature. We present a male patient with a dermoid cyst within the obturator internus spreading into the adductor compartment and the pelvic cavity. In contrast to dermoid cysts in previous reports, the cyst appeared centered in muscle rather than simply perforating muscle via an extension. The case is also notable for the rapid recurrence of the cyst after incomplete resection and intraoperative rupture.

Introduction

Mature cystic teratomas (MCTs) primarily demonstrating skin structures of ectodermal origin may be referred to as dermoid cysts. Dermoid MCTs have a characteristic keratinized squamous epithelial lining, frequently punctated by hair follicles, with hairs directed into the cystic lumen or the lining itself [1,2]. Intramural adnexal structures, including eccrine and sebaceous glands, may drain their contents into the lumen, accumulating sebum, keratin, and hair [3]. Giant cell reactions and focal calcifications may also be present [2,4]. At smaller sizes, MCTs may be asymptomatic, incidental findings [5,6]. Mass effects account for most symptoms as the teratomas gradually grow, with the exact manifestation dependent on their location and extent [7]. Patients with MCTs in the abdomen may report progressive abdominal pain and distention, urinary issues, neurological symptoms, or infections [6-8]. Rupture may lead to further complications, such as chemical peritonitis [6]. Dermoid cysts and other MCTs are typically benign, though the development of malignancies, including squamous cell carcinoma, has been reported [6,9]. Resection is the standard treatment and is also necessary for the histopathological confirmation of the diagnosis [6,10]. We herein present, to our knowledge, the third documented manifestation in English literature of an extragonadal intramuscular dermoid MCT within the pelvic musculature in a male patient, not associated with the presacral space. Extragonadal MCTs in the adult male are a rarity; those within the male pelvis are even more so. A literature search for intramuscular dermoid cysts in the pelvis returned three cases, all in male patients [2,8,11,14]. In contrast to previous instances, which had extensions of the tumor through the levator ani, the present case appeared centered in the obturator internus. This report may prove valuable in expanding the differential diagnosis for similar lesions in the pelvic region. Moreover, the recurrence of the cyst supports the importance of high vigilance during surgical removal to achieve a complete resection of such tumors.

Case presentation

A 30-year-old male presented to the ER with pain in his left hip and abdomen. A CT scan revealed a large pelvic mass of unclear etiology. He was referred to orthopedic oncology, where his initial diagnosis was suspected to be an unusual synovial chondromatosis. Multisequence multiplanar MRI revealed an indeterminate, intramuscular cystic mass measuring, along its greatest dimension, 7.0 cm in the transverse plane. Spanning through the obturator foramen, the mass involved the body

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of the left obturator internus, the obturator externus, and the adductor compartment. Viewed with T1 weighting, faint peripheral enhancement of the cyst was observed. It appeared to contain multiple solid bodies, which were mildly hyperintense with T1 weighting and suppressed on fat-saturated sequences. Prohance contrast images did not enhance the bodies (Figure 1). Open resection of the mass was pursued through a retropubic approach via a Pfannenstiel incision to reach the involved pelvic musculature. Adherence to the iliac vesicles, bladder wall, prostate, rectum, internal iliac, obturator vessels, and local nerves made for a challenging surgery, and intraoperative rupture occurred. Cystic contents included abundant hair in a yellow, noncellular fluid. Frozen sections obtained during surgery suggested a potential mature teratoma, and the remainder of the specimen was submitted entirely for permanent diagnosis. Viewed under microscopic examination with hematoxylin and eosin staining, the cyst wall contained keratinized squamous epithelium with several sebaceous glands and hair follicles (Figure 2). As mesodermal- and endodermal-derived structures were absent, the diagnosis of a dermoid MCT was favored. The patient reported pain relief immediately after surgery. However, within two months, he experienced a recurrence of abdominal pain, pain radiating down his left leg, as well as loss of appetite, nausea, bloating, changes in stooling frequency, and difficulty urinating. MRI showed an increasing fluid component of a recurred cyst extending from the obturator fossa into the pelvis. Potential residual teratomatous lesions were postulated. A fluid collection in the surgical cavity with a hypoechoic nodule at the left epididymal tail, hyperemia of the left epididymis, and small bilateral hydroceles were also noted. Given the rapidity of its growth and the return of the patient's pain, resection was once again sought. The robot-assisted laparoscopic excision achieved a visually complete removal of the cyst and cyst wall in three distinct segments. Histopathological examination of the mass demonstrated the same features as the initial excision, including a cyst with a lining of stratified squamous epithelium, in addition to fibrous and granulation tissue. Within six weeks, abdominal pain complicated the patient's course. Follow-up CT findings of a gas and fluid collection in the surgical bed were concerning for an abscess, which was subsequently drained.

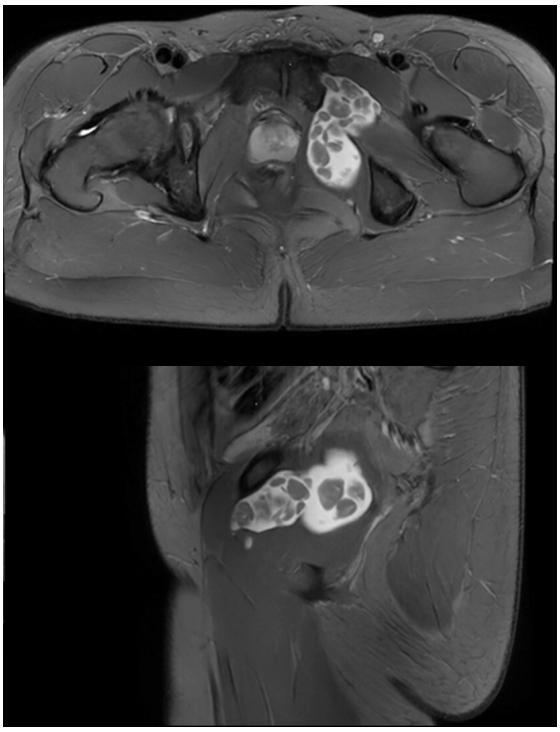

Figure 1: Coronal (Top) and sagittal (Bottom) T2-weighted pelvis MRI images show a well-circumscribed multicystic mass. The mass is hyperintense on T2 with multiple small bodies which are less intense than the surrounding fluid. The mass involves the left obturator internus as well as the obturator externus and the adductor compartment.

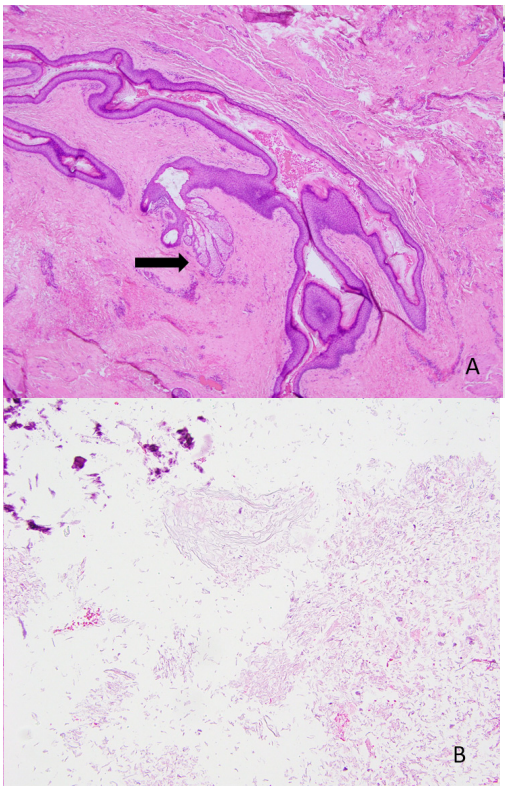

Figure 2: A: Hematoxylin and Eosin, 4x10: The tumor shows a cystic structure lined by stratified squamous epithelium. Mature skin appendages such as hair follicles and sebaceous glands (arrow) are connected to the cyst. **B:** Hematoxylin and Eosin, 10x10: The cyst contains keratin debris, hair shafts, and calcification.

Discussion

Teratomas are germ cell tumors that can contain derivatives of all three embryonic germ layers. The etiology is uncertain, and the exact mechanism may vary by location [15]. Many theorize that they stem from aberrant primordial germ cells that have strayed from their normal migratory path [16]. They may be classified as either mature, if they contain less than 10% undifferentiated tissue, or immature [6]. Additionally, the contents may be used to classify teratomas as either solid, cystic, or mixed. Immature and solid teratomas tend to present relatively greater potential for malignancy [6]. Mature cystic teratomas (MCTs) contain differentiated tissues derived from at least two of the three embryological germ layers. Dermoid cysts are MCTs that contain primarily ectoderm-derived skin structures. While ovarian dermoid cysts are the most common MCT in adults, extragonadal manifestations also exist [17]. MCTs in the retroperitoneal space are rare, accounting for less than 3% to 5% of all teratomas [18,20]. This space is confined by the posterior parietal peritoneum and the posterior body wall, with the

diaphragm forming the superior boundary and the pelvic diaphragm at the inferior extent [21]. Here, MCTs are diagnosed most often within the first year of life, though a second peak of incidence occurs in young adulthood, with only 10% to 20% of cases reported in patients older than 30 [6,22]. They occur in females roughly two to three times as often as in males [19, 22]. Thus, few reports of dermoid cysts in the adult male pelvis exist [19,22]. The present case is notable for three main aspects: (i) its rarity as an intramuscular dermoid MCT in the male pelvis, (ii) its potential to expand the differential diagnosis for masses of the pelvic musculature, and (iii) its rapid recurrence in the context of incomplete resection and intraoperative rupture. The site and muscular involvement of the current case are unique. It has no presence in the presacral space but rather is situated anterolateral to the rectum. This distinguishes it from type IV sacrococcygeal teratomas, which occupy the presacral space posterior to the rectum and anterior to the sacrum [7]. A PubMed search of English literature for extragonadal dermoid MCTs in the pelvis anterior to the rectum returned four cases [2,11,13,14]. The intramuscular nature of the mass further distinguishes it. Three of the four previous reports of intramuscular dermoid cysts outside of the orbit involve pelvic muscles. Tanaka et al. describe a case in the erector spinae in a 67-year-old male [5]. Lukanovic and Patrelli describe a 24-year-old female with a paravesical mass that contacted the obturator internus, extended to the ischiorectal fossa, and integrated with the levator ani [11]. Choudur et al. and Van Gelderen et al. likewise describe pelvic dermoid cysts with extensions through the levator ani in two male patients [12,13]. None of these instances replicate the anatomy of the present case, which centered within the obturator internus and involved the obturator foramen, obturator externus, and adductor compartment, with adhesions to the bladder, prostate, and rectum. While little precedent exists for a dermoid MCT occurring with this anatomical involvement, radiological and pathological evidence support the diagnosis. Magnetic resonance imaging of a dermoid MCT often reveals a hyperintense mass on T2-weighted imaging [5,23,24]. The mass characteristically contains several smaller bodies less intense than the surrounding cystic fluid. Some refer to this appearance as a "sack of marbles" [5,25]. T1-weighted imaging may show somewhat hyperintense nodules compared to the surrounding fluid [5,23,24]. Fat saturation suppresses the intensity of the nodules in T1-weighted images, supporting a fatty composition [5]. This description aligns well with the MRI results for this case. The pathological report likewise indicates a dermoid MCT. No undifferentiated cells were observed, and ectodermal skin structures primarily were seen, as noted in a previously documented dermoid MCT in the male pelvis [2]. The differential diagnosis for a cystic mass in this region includes hydatid cyst [26], epidermoid cyst [27], mycetoma [4], liposarcoma [28], hematoma, sarcoma, and carcinoma. Notably, the literature is inconsistent in the discrimination, or lack thereof, between the often subcutaneous dermoid cysts and dermoid MCTs. Some authors make a distinction based on origin and the potential to find structures from multiple germ layers. In this classification, dermoid MCTs, which develop from primordial germ cells, are lesions of mostly ectoderm-derived structures, with minimal presence of other germ layers [29]. Dermoid cysts, as commonly occur on the face of neonates, arise specifically from entrapped ectoderm, with more limited potential for differentiation [30,31]. Alternatively, some view all dermoid cysts as a subcategory of mature cystic teratomas [11].

Due to the extreme rarity of intramuscular MCTs, the effect of muscular involvement on the recurrence risk is not well defined. Incomplete resection increases the recurrence rate among sacrococcygeal teratomas and ovarian dermoid cysts [7,10,32]. Recurrence may be especially high in sacrococcygeal teratomas due to the relatively high frequency of incomplete capsules and the potential for the coccyx to harbor residual totipotent cells [7]. Similarly, the interaction of muscle with the dermoid cyst may have played a role in the recurrence observed in this case. Rupture may also be associated with an increased risk for benign recurrence, as noted in ovarian dermoid cysts [33]. As the initial resection was incomplete and intraoperative rupture occurred, the rapid recurrence cannot be attributed with certainty to the intramuscular character of the teratoma [33]. The potential interaction between intramuscular location and recurrence risk requires further investigation.

Conclusion

Dermoid MCTs in the male pelvic retroperitoneum are extremely rare, and those in the pelvic musculature are even more so. This report documents what is, to our knowledge, the first occurrence of an intramuscular dermoid MCT within the obturator internus. We aim for this rare case to heighten awareness about this tumor's rarity, broaden the spectrum of differential diagnoses concerning retroperitoneal intramuscular tumors, and underscore the significance of complete resection during surgery.

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