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CASE REPORT

Non-traumatic Ischiorectal Epidermoid Cyst

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ABSTRACT

Epidermoid cysts in the pelvis are rare and usually associated with local trauma or surgery. They may mimic more threatening lesions and develop complications when they become large. We report a case of epidermoid cyst arising from the ischiorectal fossa in a woman with no history of local trauma or surgery.

Key Words: *Epidermal cyst; Magnetic resonance imaging; Pelvis*

中文摘要

非創傷性坐骨直腸表皮樣囊腫

李家彥、李家灝、朱志揚、李曉蕾、邱麗珊

在盆腔中的表皮樣囊腫很罕見，並通常與局部創傷或手術相關。當它們夠大時可模擬更惡性的病變並引致併發症。本文報告一例坐骨直腸窩表皮樣囊腫，發生在一名沒有局部創傷或手術的女性。

INTRODUCTION

Perirectal cystic lesions, including those arising from the ischiorectal fossa, encompass a histologically diverse group owing to multiple embryologic remnants in this region. In contrast to rectal tumours, perirectal cystic lesions are usually benign, congenital, and asymptomatic.[1] When the lesion is large, the local mass effect may mimic a rectal tumour or complications such as infection, fistula, or malignant degeneration.[2] Epidermoid cyst in the ischiorectal fossa is rare,[2] and it is even rarer in patients with no history of local trauma. We report a case of epidermoid cyst arising from the ischiorectal fossa in a woman with no history of local trauma or surgery.

CASE REPORT

In January 2015, a 24-year-old woman presented to the Pamela Youde Nethersole Eastern Hospital with epigastric pain and vomiting. She had a history of familial hypercalciuria and congenital renal anomaly. Computed tomography scans of the abdomen and pelvis were unremarkable, except for an incidental finding of a well-defined hypodense lesion with rim enhancement in the left ischiorectal fossa. The patient had no history of local trauma or surgery. The initial diagnosis was gastroenteritis with a developmental cyst. She was discharged home after resolution of the symptoms.

Three months later, the patient still had pressure

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symptoms during defaecation. Magnetic resonance imaging revealed a 6-cm cystic lesion at the left ischiorectal fossa that was intermediately intense on T1-weighted images and hyperintense on T2-weighted images, with mild gadolinium enhancement of the rim and restricted diffusion (Figure 1). The mass displaced the rectum and left levator ani muscle. No fistulous connection to the skin or extension into the sacral foramina was noted. The imaging diagnosis was an epidermoid cyst.

Four months later, the patient underwent excision of the cyst, which was a tan-coloured nodule containing yellowish creamy material. Microscopically, the cyst was lined with stratified squamous epithelium and contained laminated keratinous material (Figure 2), with no evidence of malignancy. The pathological diagnosis was epidermal cyst. At the 3-month follow-up visit, the patient reported resolution of all symptoms. No further follow-up was arranged.

DISCUSSION

Developmental cysts are the most common congenital lesions within the perirectal region.[1,3] Tailgut cysts account for half of all developmental cysts, followed by epidermoid or dermoid cysts, rectal duplication cysts, and neurenteric cysts. Tailgut cysts and rectal duplication cysts are classified as enteric cysts.[1] Other congenital lesions in this region include sacro-coccygeal teratoma, lymphangioma, and anterior sacral meningocoele.[3]

Epidermoid cysts arise from epidermal cell proliferation within a confined dermal space.[4,5] The most common pathological mechanisms include misplacement of ectodermal elements during embryogenesis, occlusion of the pilosebaceous unit, or traumatic or surgical implantation of epithelial tissue.[4,6] Most such cases are related to pelvic trauma, trauma from giving birth, surgical procedures such as vaginal hysterectomy and episiotomy, or female genital mutilation. Epidermoid cysts in the pelvis without a history of local trauma or surgery are rare; only two such cases, presenting as a buttock mass and obstructed labour, have been reported.[5]

About 50% of perirectal developmental cysts are asymptomatic. Clinical presentation depends on the size and location of the lesions. Symptoms include rectal compression, constipation, painful defaecation, rectal fullness, buttock or lower abdominal pain, lower urinary tract compression, urinary frequency, dysuria, and, rarely, obstructed labour and sciatic pain.[1] Patients may also present with local complications such as infection with perianal fistula or rectal bleeding, as well as the Currarino triad of anorectal malformation, sacrococcygeal osseous defect, and a presacral mass (anterior sacral meningocoele, tailgut cyst, dermoid cyst, or teratoma).[3,4]

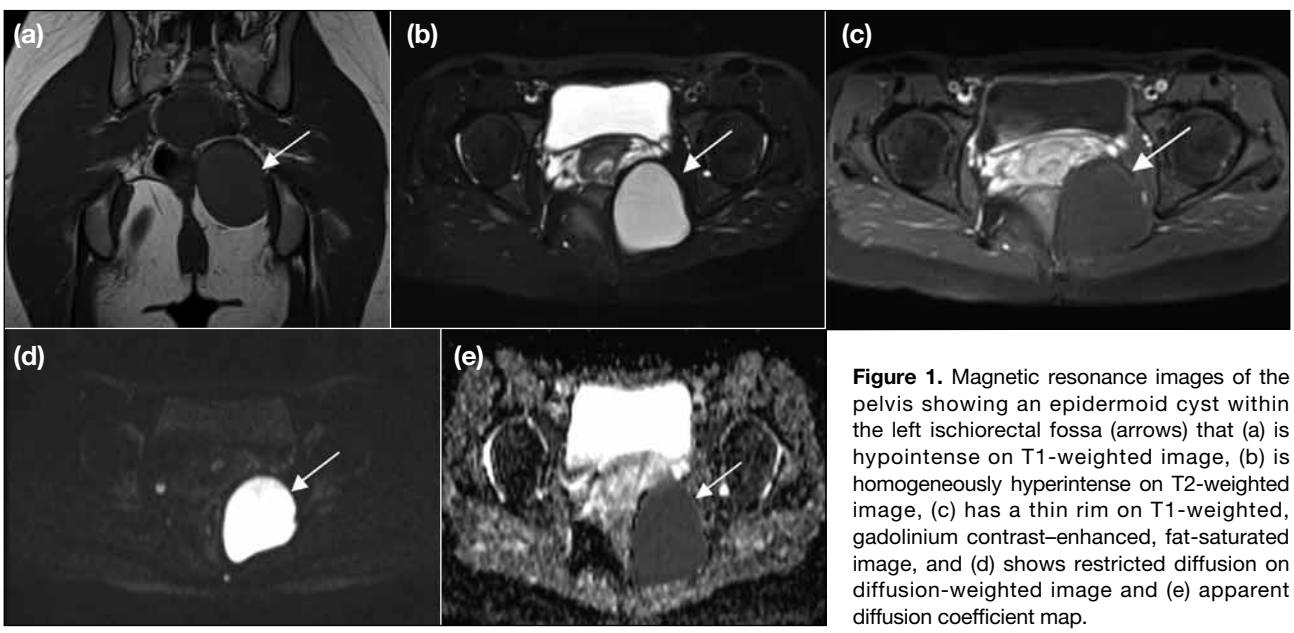

Figure 1. Magnetic resonance images of the pelvis showing an epidermoid cyst within the left ischiorectal fossa (arrows) that (a) is hypointense on T1-weighted image, (b) is homogeneously hyperintense on T2-weighted image, (c) has a thin rim on T1-weighted, gadolinium contrast–enhanced, fat-saturated image, and (d) shows restricted diffusion on diffusion-weighted image and (e) apparent diffusion coefficient map.

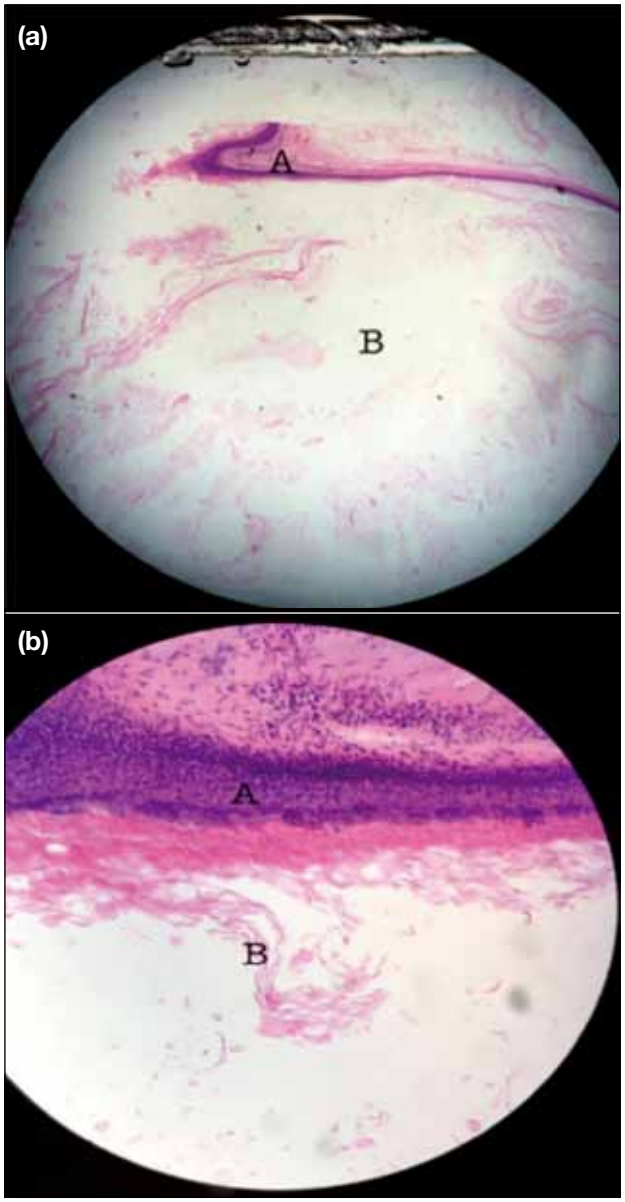

Figure 2. (a) Low-power field and (b) high-power field photomicrographs (H&E, 20x and 60x, respectively) showing (A) the epidermal cyst (infundibular type) wall formed by stratified squamous epithelium and (B) laminated keratinous cyst content.

Epidermoid cysts are lined with stratified squamous epithelium and are usually unilocular with internal clear fluid.[5,6] Unruptured epidermoid cysts are typically well defined, round or ovoid, and homogeneously hyperintense on T2-weighted and hypointense on T1-weighted images. Mild internal signal hyperintensity on T1-weighted images might correspond to mucoid or proteinaceous materials.[1,6] Restricted diffusion, along with a thin rim of gadolinium enhancement, are usually observed. Features that are suggestive of rupture include internal septation, thick and irregular rim of cyst wall enhancement, and fuzzy enhancement in the surrounding subcutaneous tissue.[5]

In dermato-pathological terms, epidermoid cysts are known as follicular or epidermal cysts, of infundibular type. The most typical histological features include a cystic cavity filled with laminated keratin and lined with stratified squamous epithelium identical to the epidermis. A punctum opening connecting the cystic lesion in the dermal layer to the epidermis on the surface may also be seen in cases located in the skin. Granulomatous inflammatory changes may be evident in cases of rupture.[7]

Imaging can help differentiate epidermoid cysts from other cystic lesions in the ischiorectal fossa. Dermoid cysts contain fat elements (owing to sebaceous glands in the skin appendages) and are hyperintense on T1-weighted images and hypointense on fat-suppressed images at fat-containing regions. Dermoid cysts less commonly demonstrate restricted diffusion and can be multiple in number, compared with epidermoid cysts.[3,8] Tailgut cysts are typically multiloculated in appearance or have a small peripheral cyst in additional to a large cyst.[9] They are usually hypointense on T1-weighted images and hyperintense on T2-weighted images. They have a 7% risk of malignant degeneration, with suggestive imaging features such as solid-enhancing components or asymmetric irregular wall-thickening.[9] Rectal duplication cysts are commonly contiguous with the rectal wall, show luminal communication, and contain bowel content.[10] A well-circumscribed two-layer smooth-muscle wall may be observed.[3] Calcification is uncommon in dermoid and tailgut cysts.[1,7] Lymphangioma extends into different compartments without significant mass effect.[3]

Perirectal lesions are commonly benign and congenital, in contrast to rectal lesions, which are commonly neoplastic and malignant.[1,3] Perirectal lesions can be neurogenic in origin (such as schwannoma and neurofibroma)[11] or osseous in origin (such as chordoma and giant-cell tumour).[12] Abscess and endometriosis may also arise in the perirectal region.[3]

Total excision (cyst removal with intact capsule as a whole) is the mainstay of treatment.[13] When total removal is not feasible, such as in intracranial

epidermoid cysts, the extent of excision is a determinant of the time to recurrence.[14]

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