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CASE REPORT

A rare cause of recalcitrant coccydynia: benign dermoid cyst masquerading as coccygeal pain

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Abstract
Purpose Coccydynia is a common entity in orthopedic practice, and various etiologies have been described for it. However, benign dermoid cyst causing coccydynia has not yet been reported.
Methods A 20-year-old male presented with typical symptoms of coccydynia recalcitrant to conservative treatment for 2 years. Since pain interfered with his daily activities, magnetic resonance imaging was performed which showed a circumscribed precoccygeal cystic lesion.
Results The patient underwent coccygectomy along with cyst excision. Histological examination revealed features of benign dermoid cyst. After surgery, the patient had excellent relief of his symptoms.
Conclusion The case report identifies that the treating surgeon should be aware of benign dermoid cyst as one of the treatable but rare causes of intractable coccydynia, and MRI should be performed in patients with persistent coccygeal pain.

Keywords Coccydynia · Benign dermoid cyst · Magnetic resonance imaging · Coccygectomy

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Introduction

Coccydynia is defined as pain in and around coccyx region. It is mainly post-traumatic and idiopathic [1–3]. Various other rare causes of coccydynia have been described in the form of individual case reports and include bone tumors [4, 5], glomus tumor [6], perineural cysts [7], infection [8] and neural tumors [9, 10]. Usually, the diagnosis is made by typical history and clinical examination, and radiological evaluation is performed only in patients with chronic and persistent symptoms. There are no standard indications described in the literature for performing an MRI scan for patients with coccydynia. Most patients with coccydynia have good relief of symptoms with conservative treatment. Surgical excision is advised in patients with persistent pain.

Case report

A 20-year-old college student presented to us with unbearable pain in the gluteal cleft region near the coccyx for the past 2 years. Pain had started insidiously without any antecedent trauma. It was initially mild but has become unbearable in the last 5 months. The pain typically was aggravated by sitting and not associated with radicular, neurological or constitutional symptoms. There was no history of difficulty in defecation or micturition. The patient's body mass index (BMI) was 22.55 (weight 62 kg and height 166 cm). Local examination revealed normal overlying skin, but there was significant tenderness over the coccyx. Radiograph of the coccyx showed that the morphology of coccyx was normal (Fig. 1). He was treated initially with analgesics and ring pillow and advised to avoid direct pressure over the coccyx. But he did not respond to conservative treatment given for 2 months. So

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Fig. 1 Lateral radiograph of the sacrum with coccyx showing a normal bony anatomy without any significant abnormality

an MRI scan of the coccyx was performed, which showed that the coccyx was normal, but a 1.5 × 1.8 cm well-circumscribed cystic lesion was found situated just anterior arising from the coccyx. The cyst was isointense on T1-weighted image, hypointense on T2-weighted image and hyperintense on STIR (short tau inversion recovery) T1-weighted image (Fig. 2). Since it was difficult to determine the source of pain clinically (either cyst or coccyx), the patient was treated by coccygectomy and cyst excision under general anesthesia. Through a posterior midline approach, subperiosteal dissection was performed and coccyx was removed along with meticulous en bloc resection of the cyst. The anterior cyst wall was situated just posterior to rectum. The cyst was white in appearance and contained cheesy material. The cyst and its contents were sent for histopathological examination which showed an epidermal-lined cyst wall with its keratin contents characteristic of benign dermoid cyst (Fig. 3). The patient's symptoms resolved postoperatively, and he remained asymptomatic at 12-month follow-up.

Discussion

The term coccydynia was coined by Simpson in 1861 for painful coccyx [11]. Coccydynia is a common condition seen in orthopedic practice. Even though it is self-limiting, it can be very debilitating affecting patient's quality of life. The most common cause of coccydynia is trauma [3]. Maigne et al. [3] have suggested that only a traumatic event occurring within 1 month of onset is significant in increasing the risk of instability and subsequent coccydynia. Idiopathic coccydynia is considered a diagnosis of exclusion and has been described in the absence of any obvious pathologic changes involving the coccyx [12]. In idiopathic coccydynia, the pain seems to result from spasticity of the musculature of the pelvic floor [12]. Morphological abnormality of coccyx like spicule formation, increased intercoccygeal angle [1, 2] and retroversion [13] are other possible described causes of idiopathic coccydynia. Obesity is three times more common in patients with coccydynia than in the normal population. Obesity decreases pelvic rotation when patient sits putting

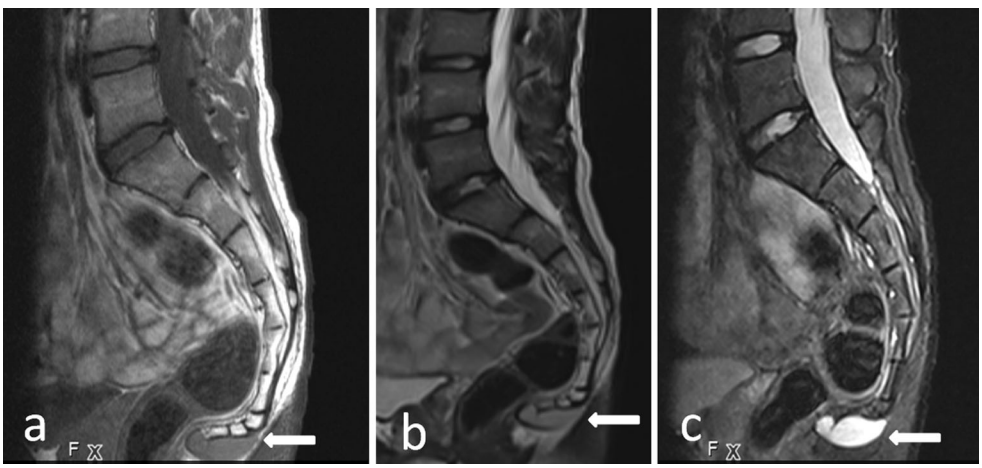

Fig. 2 **a** T1-weighted MRI image showing isointense cystic lesion adjacent to coccyx (*arrow*), **b** MRI T2-weighted image showing hypointense cystic lesion adjacent to coccyx (*arrow*), **c** MRI STIR image showing hyperintense cystic lesion adjacent to coccyx (*arrow*)

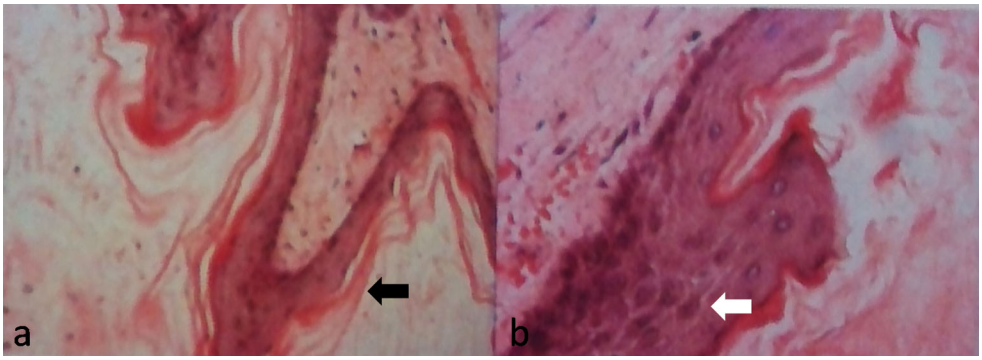

Fig. 3 Histopathology images of benign dermoid cyst showing—**a** epidermal lining of dermoid cyst (*black arrow*), **b** keratin content of cyst (*white arrow*)

coccyx for greater risk of subluxation typically seen in post-traumatic lesion [14].

Though rare, various pathological conditions can present as coccydynia which include tumors of the sacrum/coccyx (hemangioma, carcinoid) [4, 5], glomus tumors of the pericoccygeal tissue [6], perineural cyst [7], infection [8], lumbosacral intradural tumors (schwannoma [9] and ependymoma [10]). Very few cases similar to our case have been reported in the literature. Halefoglu [15] reported pre-coccygeal epidermal inclusion cyst in a 33-year-old pregnant woman who suffered from coccygeal and pelvic pain. Munteanu et al. [16] had reported a case in 18-year-old female with asymptomatic retrorectal mass found incidentally during gynecological examination. Such retrorectal cysts are mostly asymptomatic and discovered incidentally during evaluation for other diseases. Above conditions should be identified and differentiated from idiopathic coccydynia so that appropriate treatment can be initiated. Physical examination should include inspection for the presence of pilonidal sinus or discharge. Palpation may reveal localized tenderness, swelling or mass. Apart from clinical examination, imaging studies could be invaluable in evaluation. Standard AP and lateral radiographs of coccyx may reveal areas of destruction or lysis as in tumors and infection of the coccyx. Fogel et al. [14] proposed acute coccydynia refractory to conservative treatment or symptoms lasting for more than 2 months should be further evaluated with dynamic radiographs and MRI scan. Dynamic radiographs obtained in sitting and standing positions may be more useful than static radiographs to demonstrate sacrococcygeal instability [14]. MRI scan may show inflammatory, infective or tumor pathology [14].

In our case, MRI was performed since the patient had persistent pain and static radiographs did not reveal any abnormality. MRI revealed pericoccygeal cystic lesion. The cyst was excised in toto along with the coccyx, and the patient had good pain relief after surgery. Histopathological examination showed benign dermoid cyst. Pathogenetically, dermoids are ectoderm-lined inclusion cysts and usually contains sebaceous and sweat glands and squamous epithelium. It arises from trapped pouches of ectoderm or from failure of surface ectoderm to separate from neural tube. These are usually benign slowly growing, unilocular, cystic masses and may occur from adolescence to adult life [17]. Although asymptomatic most of the times, dermoid cyst needs excision in symptomatic recalcitrant cases. Local steroid injection and coccyx manipulation are popular treatments for coccydynia. In the absence of MRI, local steroid injection would have been potentially dangerous resulting in infection, in this patient.

Conclusion

Coccydynia is most commonly post-traumatic or idiopathic and usually responds very well to conservative treatment. The purpose of this report was to highlight that MRI scan should be done in cases of coccydynia not responding to prolonged conservative measures, and pericoccygeal dermoid cyst, although very rare, could be one of the possible reasons for pathological coccydynia.

Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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