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Precoccygeal epidermal inclusion cyst presenting as coccygodynia

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ABSTRACT

Although there are numerous aetiologies for coccygodynia described in the medical literature, precoccygeal epidermal inclusion cyst presenting as a coccygodynia has not been reported. We report a 30-year-old woman with intractable coccygodynia. Magnetic resonance imaging showed a circumscribed precoccygeal cystic lesion. The removed cyst was pearly-white in appearance and contained cheesy material. Histological evaluation established the diagnosis of epidermal inclusion cyst with mild nonspecific inflammation. The patient became asymptomatic and remained so at two years follow-up. This report suggests that precoccygeal epidermal inclusion cyst should be considered as one of the differential diagnosis of coccygodynia. Our experience suggests that patients with intractable coccygodynia should have a magnetic resonance imaging to rule out treatable causes of coccygodynia.

Keywords: coccygodynia, coccyx, epidermal inclusion cyst, precoccygeal epidermal inclusion cyst

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INTRODUCTION

Intractable coccygodynia is frequently-labelled as being idiopathic or post-traumatic,[1-3] but various other aetiologies[4-14] of coccygodynia have been described, usually in the form of sporadic individual case reports. We report a previously-undescribed case of precoccygeal epidermal inclusion cyst as cause of coccygodynia, which stresses the importance of advanced investigations for confirmation of diagnosis. A thorough literature review was done to evaluate the various aetiologies of coccygodynia.

CASE REPORT

A 30-year-old nulliparous female patient presented with severe pain in the coccygeal region of five years' duration. The pain was insidious from onset without any antecedent trauma, localised, initially intermittent but continuous for the last two months. Sitting and reclining backwards aggravated the pain. There was no radiation, neurological symptoms, constitutional symptoms, difficulty in defaecation or micturition.

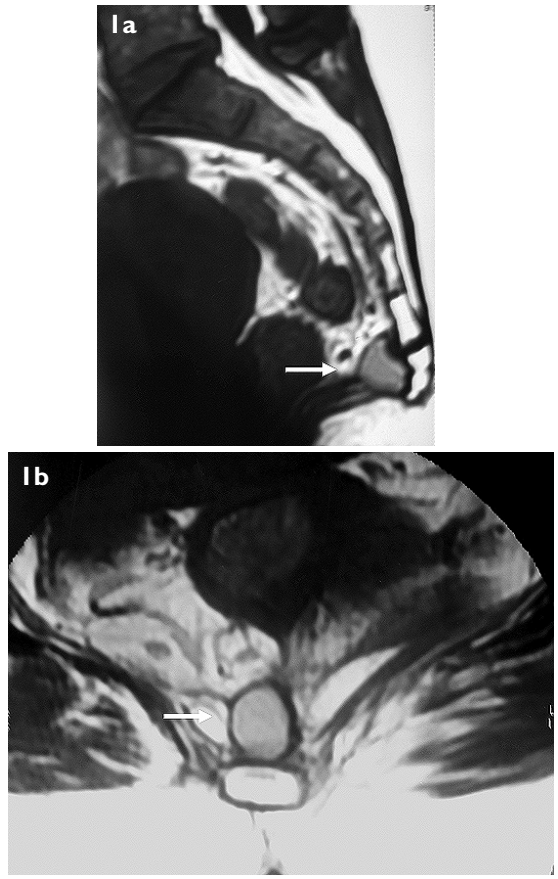

Fig. 1 (a) Sagittal and (b) axial T1-W MR images taken at the level of the coccyx show a 1.5 cm isointense precoccygeal cystic lesion with well-circumscribed margins.

She had undergone conservative treatment in the form of rest, doughnut ring, local heat, and avoidance of direct pressure over the area, with only partial relief. Physical examination revealed normal overlying skin and marked tenderness over the coccyx on deep pressure. Rectal examination revealed an extra luminal retrorectal tender swelling. Radiographs showed type 1 morphology (Postacchini and Massobrio[2]) of coccyx without any evidence of fracture or erosion. There was no instability in the form of subluxation or hypermobility in lateral dynamic views. Haematological investigations including erythrocyte sedimentation rate and C-reactive protein were noncontributory. Magnetic resonance (MR) imaging showed a 1.5 cm precoccygeal cystic lesion with a well-circumscribed margin. The contents of the cyst appeared isointense on T1- and hypointense on T2-weighted images (Figs. 1 & 2).

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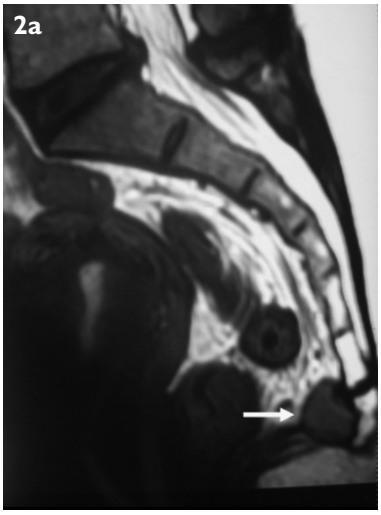

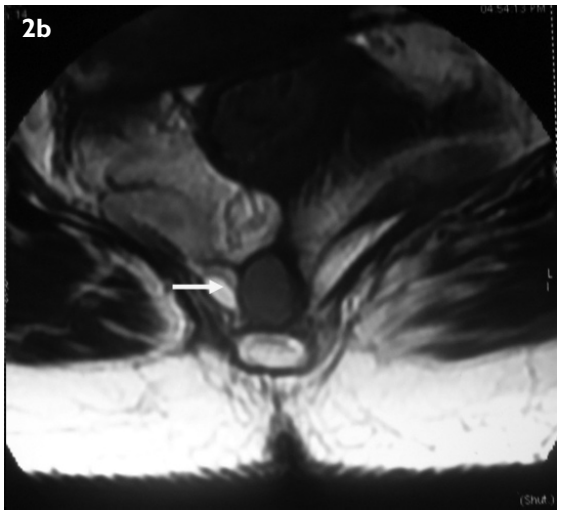

Fig. 2 (a) Sagittal and (b) axial T2-W MR images taken at the level of the coccyx show a 1.5 cm hypointense precoccygeal cystic lesion with well-circumscribed margins.

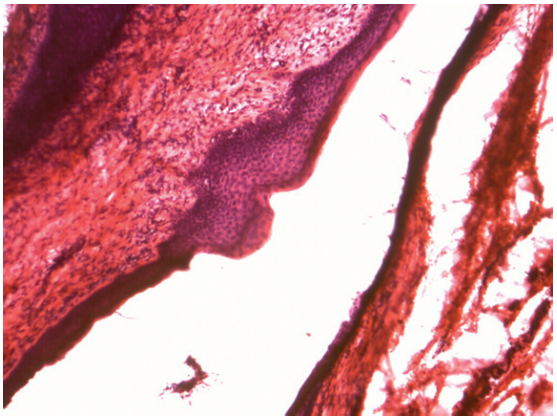

Fig. 3 Photomicrograph of the resected specimen shows keratinaecious debris circumscribed by stratified squamous epithelium with mild nonspecific chronic inflammation in the cyst wall and without any evidence of malignancy. Histological diagnosis is epidermal inclusion cyst (Haematoxylin & eosin, × 100).

Resection of the coccyx and cyst was done with the patient in prone position, using a posterior longitudinal midline incision, careful subperiosteal dissection, coccygectomy and meticulous extirpation of the cyst. The cyst was pearly white in appearance and contained cheesy material. The histological sections of the biopsy demonstrated keratinaecious debris circumscribed by stratified squamous epithelium (Fig. 3). There was no evidence of malignancy. There was a mild, nonspecific chronic inflammation in the cyst wall. There were no granulomata. The histological diagnosis was epidermal inclusion cyst. The patient was completely relieved of symptoms and continued to be asymptomatic at two years follow-up.

DISCUSSION

Coccygodynia is a common entity seen in orthopaedic and spine clinics. The term, coccygodynia, was coined by Simpson in 1861 for painful coccyx.[15] Though it is often being referred to as a disease, it actually describes a symptom. Intractable coccygodynia can be a very debilitating disorder. Although mostly labelled as idiopathic or post-traumatic in origin, various unusual pathological conditions have been described as a cause for coccygodynia.[4-14] Initially thought to be neurotic disorder, it is now known that there is not much difference in psychological profiles of symptomatic and asymptomatic people.[2] Other suggested aetiologies[2] of idiopathic coccygodynia are spasm of the muscles of the pelvic floor,[16] anomalies of the soft tissues in the mid-sacral region, chronic inflammation of an adventitious coccygeal bursa, and arachnoiditis of the lower sacral nerve roots. Morphological abnormality of the coccyx, including increased intercoccygeal angle,[1,2] spicule,[1,2] retroversion,[17] and scoliosis,[18] may be possible causes of idiopathic coccygodynia. Initially, it was thought that even lumbosacral disc prolapse can present as coccygodynia.[19] However, the fact that a substantial number of patients showing successful outcomes with treatment localised to the coccyx refutes the theory of discogenic origin of pain.[2]

Since coccygeal pain often develops after a local injury, it has been most frequently attributed to causes involving trauma. Antecedent trauma due to falls or difficult vaginal delivery can directly injure sacrococcygeal synchondrosis. Furthermore, obesity decreases pelvic rotation when the patient sits, henceforth puts the coccyx at a greater risk for luxation, which is a typical post-traumatic lesion.[3] Instability of the coccyx can be judged radiologically as hypermobility or intermittent subluxation on dynamic sitting views via the method suggested by Maigne and Tamalet.[18] Radiologically-demonstrable instability of the coccyx forms the group which has the maximum chance of recovery after surgery.[1,20]

Some rare, though well-described, pathological conditions which can present as coccygodynia include recent fracture (post-traumatic or intrapartum[21]), dislocation,[22] tumours of the sacrum and coccyx (haemangioma,[4] carcinoid,[5]), glomus tumours of the

pericoccygeal tissue,[6] lumbosacral intradural tumours (schwannoma,[7] ependydoma,[8] arachnoid cysts[9]), perineural cyst,[10] intraosseous lipoma,[11] infectious diseases (tuberculosis[12]), anal duct/gland cyst,[13] and avascular necrosis of the coccyx.[14] Glomus tumour as a cause of coccygodynia has been questioned as they mostly represent normal coccygeal bodies (non-pathological organelle glomus coccygeum).[23,24] Immunohistochemical demonstration of the smooth muscle actin and neuron-specific enolase in glomus cells may be beneficial for accurate identification of this organelle.[24]

In our case, MR imaging revealed a precoccygeal cystic lesion, and the histopathological diagnosis was epidermal inclusion cyst. An epidermal inclusion cyst can be described as a dermal cystic enclosure of keratinising squamous epithelium that is filled with keratin. Pathogenetically, an epidermal cyst may be secondary to congenital development or iatrogenic implantation. Our case appears to be of the congenital variety, in the absence of any antecedent trauma or local injections. Epidermal cyst in this region is an extremely rare condition. Clinically, these cysts manifest as painless, slow-growing, well-circumscribed swellings and may occur at any time from adolescence to adult life,[25] but they may become inflamed or secondarily infected, resulting in pain and tenderness. The epidermal cysts are generally characterised on MR imaging by variability of signal intensity between different cases, and at times between the different parts of the same cysts.[26] Other features include the absence of the oedema in surrounding tissues, fairly well-defined limits and peripheral enhancement on gadolinium injection. The disparity in signal intensity is most likely related to the chemical state of cholesterol, or the relative composition of cholesterol and keratin makes the preoperative diagnosis difficult. Malignancy must be ruled out. Rarely, malignancies, including basal cell carcinoma, Bowen's disease, squamous cell carcinoma, and even mycosis fungoides, have developed in epidermal cysts.[27]

A variety of treatment modalities have been described for coccygodynia. As with any clinical condition, a correct diagnosis is most vital in treatment. This report suggests that precoccygeal epidermal inclusion cyst should be considered as one of the differential diagnosis of coccygodynia, besides other possible aetiologies of coccygodynia mentioned in the literature. A high level of suspicion is warranted before embarking on surgical treatment of intractable coccygodynia. It is suggested that the clinician rules out all possible aetiologies for successful treatment of intractable coccygodynia through further investigations, preferably using MR imaging.

REFERENCES

1. Kim NH, Suk KS. Clinical and radiological differences between traumatic and idiopathic coccygodynia. Yonsei Med J 1999; 40: 215-20.
2. Postacchini F, Massobrio M. Idiopathic coccygodynia. Analysis of fifty-one operative cases and a radiographic study of the normal coccyx. J Bone Joint Surg Am 1983; 65:1116-24.
3. Maigne JY, Doursounian L, Chatellier G. Causes and mechanisms of common coccydynia: role of body mass index and coccygeal trauma. Spine 2000; 25:3072-9.
4. Lath R, Rajshekhar V, Chacko G. Sacral haemangioma as a cause of coccydynia. Neuroradiology 1998; 40:524-6.
5. Krasin E, Nirkin A, Issakov J, et al, Carcinoid tumor of the coccyx: case report and review of the literature. Spine 2001; 26:2165-7.
6. Ho KL, Palc MSY. Glomus tumor of the coccygeal region. J Bone Joint Surg Am 1980; 62:141-2.
7. Kinnett JG, Root L. An obscure cause of coccygodynia. J Bone Joint Surg Am 1979; 61:299.
8. Houlding NR, Mathesan AT. Intrathecal spinal tumour as a cause of coccygodynia. J Bone Joint Surg Br 1961; 43:344-5.
9. Kepski A, Rudnicki S Arachnoid cyst of the cauda equina: a contribution to the etiology of coccygodynia. Neurol Neurochir Pol 1978; 12:109-12.
10. Ziegler DK, Batnitzky S. Coccygodynia caused by perineural cyst. Neurology 1984; 34:829-30.
11. Hanelin LG, Sclamberg EL, Bardaley JL. Intraosseous lipoma of the coccyx. Radiology 1975; 114:343-4.
12. Kumar A, Varshney MK, Trikha V, Khan SA. Isolated tuberculosis of the coccyx. J Bone Joint Surg Br 2006; 88:1388-9.
13. Kulaylat MN, Doerr RJ, Neuwirth M, Satchidanand SK. Anal duct/gland cyst: report of a case and review of the literature. Dis Colon Rectum 1998; 41:103-10.
14. Lourie J, Young S. Avascular necrosis of the coccyx: a cause of coccydynia? Case report and histological findings in 16 patients. Br J Clin Pract 1985; 39:247-8.
15. Simpson JY. Removal of the coccyx and coccygodynia [letter]. Med Times Gazette 1861; 1:317.
16. Thieli GH. Coccygodynia and pain in the superior gluteal region. JAMA 1937: 109;1271-5.
17. Dennell LV, Nathan S. Coccygeal retroversion. Spine 2004; 29: E256-7.
18. Maigne JY, Tamalet B. Standardized radiologic protocol for the study of common coccygodynia and characteristics of the lesions observed in the sitting position. Clinical elements differentiating luxation, hypermobility, and normal mobility. Spine 1996; 21:2588-93.
19. Richards HJ. Causes of coccydynia. J Bone Joint Surg Br 1954; 36:142.
20. Bayne 0, Bateman JE, Cameron HU. The influence of etiology on the results of coccygectomy. Clin Orthop Relat Res 1984; 190:266-72.
21. Jones ME, Shoaib A, Bircher MD. A case of coccygodynia due to coccygeal fracture secondary to parturition. Injury 1997; 28:549-50.
22. Bergkamp AB, Verhaar JA. Dislocation of the coccyx: a case report. J Bone Joint Surg Br 1995; 77:831-2.
23. RS Bell, Goodman SB, Fornasier VL. Coccygeal glomus tumors: a case of mistaken identity? J Bone Joint Surg Am 1982; 64:595-7.
24. Gatalica Z, Wang L, Lucio ET, et al. Glomus coccygeum in surgical pathology specimens: small troublemaker. Arch Pathol Lab Med 1999; 123:905-8.
25. Epstein W L. Epithelial cysts in buried human skin. Arch Dermatol 1957; 76:437-45.
26. Vion-Dury J, Vincentelli F, Jiddane M, et al. MR imaging of epidermoid cysts. Neuroradiology 1987; 29:333-8.
27. López-Ríos F, Rodríguez-Peralto JL, Castaño E, Benito A. Squamous cell carcinoma arising in a cutaneous epidermal cyst: case report and literature review. Am J Dermatopathol 1999; 21: 174-7.