



Case Report

Bursitis of the coccyx in an adult with rheumatoid arthritis mimicking a sacrococcygeal meningocele

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ABSTRACT

Background: Bursitis is a chronic inflammatory condition characterized by the deposition of cholesterol, macrophage infiltration, and bursal wall calcification. Bursitis is, however, rarely found in the sacrococcygeal region where it may present as a space-occupying mass.

Case Description: A 64-year-old male with rheumatoid arthritis presented with 3 years' duration of difficulty sitting and walking due to a soft-tissue mass involving the coccyx region. Once the patient's MR demonstrated a cystic lesion with erosion of the coccyx, the patient underwent gross total resection of the lesion that proved to be pathologically consistent with bursitis. Postoperatively, the patient's complaints fully resolved.

Conclusion: Bursitis may present as a soft-tissue tumor-like lesion in the coccyx that favorably responds to gross total surgical excision.

Keywords: Bursitis, Rheumatoid arthritis, Sacrococcygeal

INTRODUCTION

Bursae are fluid-filled sacs located around joints, and between bone and surrounding soft tissues.^[3,9] Degeneration of the muscle, tendons, and skin overlying bony processes may lead to repeated friction and thickening of the bursa's connective tissue wall.^[6] The resulted increased secretion of synovial fluid then contributes to the formation of a bursal cyst, with chronic inflammation leading to bursitis that is pathologically characterized by the deposition of cholesterol, macrophage infiltration, and bursal wall calcification.^[6]

Here, we present a 64-year-old male with a history of rheumatoid arthritis (RA) whose low back pain attributed to a large bursal cyst/erosion of the coccyx responded favorably to surgical excision.

CASE REPORT

A 64-year-old male with a history of RA presented with pain attributed to an enlarging coccygeal soft-tissue mass for the past 3 years.

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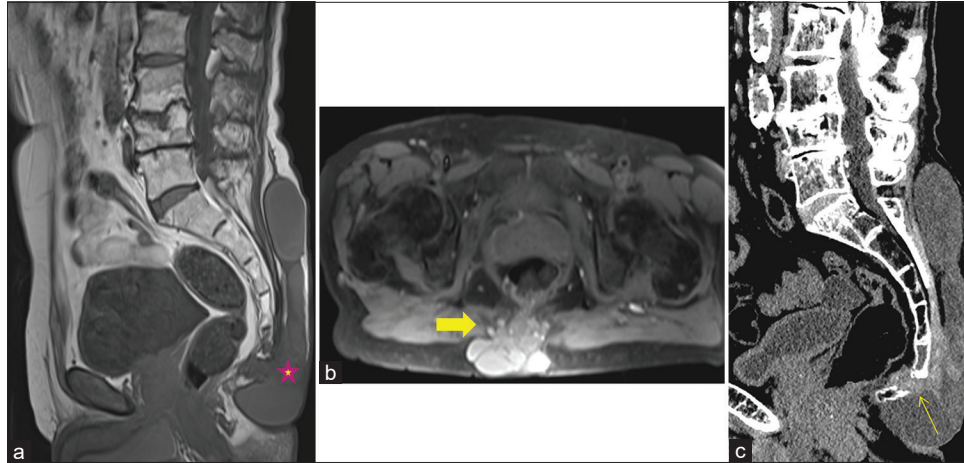


Figure 1: Preoperative: (a) sagittal T1-weighted image of the lower lumbar spine, sacrum, and coccyx reveals an extensive, well-circumscribed subcutaneous lesion (star) at the dorsal aspect of the pelvis, which returns homogenous low signal and infiltrates the middle osseous part of the coccyx. (b) Axial T2-weighted image with fat saturation at the level of the coccyx shows the communication of the subcutaneous multiseptated lesion (thick arrow), which returns high signal indicative of fluid and/or proteinaceous material with the destroyed infiltrated central part of the coccyx. (c) Sagittal CT reconstruction of the same ROI shows the different density between the fluid-filled main subcutaneous lesion and the higher, soft-tissue like density of the lesion that causes osteolysis of the coccyx (arrow).

The lesion was soft and mobile, and there were no signs of infection. The magnetic resonance imaging demonstrated a cystic lesion, extending from the coccyx up to the L5 vertebral body. The lesion was hyperintense on T2-weighted images, similar to cerebrospinal fluid but there was no evidence of a connection between the lesion and the subarachnoid space [Figure 1a and b]. The computed tomography (CT) scan further documented focal erosion of the coccyx at the lesion site [Figure 1c].

Aspiration

Initially, the patient underwent aspiration of the fluid component of the cyst; it was yellow, thick, and contained amorphous floaters. The culture revealed no microorganisms.

Surgery

The patient underwent gross total resection of the cyst with removal of the coccyx. At surgery, the cyst was whitish-gray in color, multiloculated, adherent to the periosteum of the sacral bone and coccyx, and continuous with the fascia of the paravertebral muscles [Figure 2]. The postoperative CT confirmed adequate lesion removal [Figure 3].

Pathology

The pathological examination showed chronic inflammation and fibrosis consistent with a diagnosis of bursitis. In addition, there was amorphous necrotic material similar to fibrinoid necrosis surrounded by a histiocytic reaction with granulomatous features [Figure 4a and b]. This morphological appearance was



Figure 2: Intraoperative image showing a multiloculated cyst with secretion of yellowish thick fluid (star) through its wall.

similar to rheumatoid nodules, and consistent with the inflammation seen with RA.

Postoperatively, the patient recovered uneventfully, and the lesion did not recur over 8 months duration.

DISCUSSION

Bursae are lubricated fluid-filled thin sacs located between bone and surrounding soft tissue, tendons, and muscles around joints. With friction and microtrauma, the inflammatory pathway is activated, and the bursae wall becomes thickened thus resulting in bursitis.^[3] Bursitis is thus characterized by a chronic inflammatory reaction resulting

Table 1: Cases of sacrococcygeal bursitis.

Author	Year	Cases	Age	Symptoms	Histopathology	Treatment
Kai <i>et al.</i> ^[4]	2001	1	69	Pain	Yes	Surgical, recurrence
Goishi <i>et al.</i> ^[1]	2005	1	72	NR*	Yes	Surgical
Samura <i>et al.</i> ^[6]	2008	1	10	Pain	Yes	Surgical
Seike <i>et al.</i> ^[7]	2008	2	58	NR	NR	Surgical, recurrence
			14	Pain	NR	Surgical, recurrence
Maigne <i>et al.</i> ^[5]	2011	6	<16	NR	NR	NR
Hayashi <i>et al.</i> ^[2]	2012	1	71	NR	Yes	Surgical, sclerotherapy
Shih <i>et al.</i> ^[8]	2016	1	31	Pain	Yes	Surgical
Present case	2020	1	64	Pain	Yes	Surgical

*NR: Nonreported

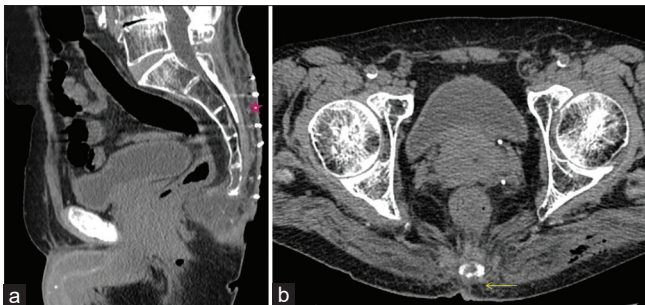


Figure 3: Postoperative: (a) postoperative sagittal CT reconstruction shows complete removal of the subcutaneous tissue (star) and surgical excision of the distal part of the coccyx. (b) The axial CT image shows minor changes at the level of the distal part of the coccyx without evidence of subcutaneous recurrent mass or collection (arrow).

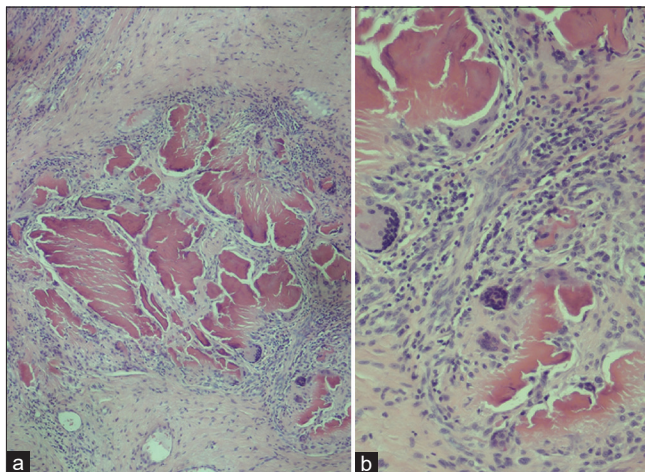


Figure 4: Necrobiotic areas containing amorphous eosinophilic material that is surrounded by inflammatory reaction with prominent macrophages, including multinucleated giant cells: (a) $\times 40$ and (b) $\times 100$.

in deposition of cholesterol, macrophage infiltration, and bursal wall calcification.^[6] The most common causes include trauma, inflammation, infection, and crystal deposition.^[3,6,9]

There are very few cases of isolated sacrococcygeal bursitis reported in the literature.^[1,4,6-8] Maigne *et al.* reported six cases of sacrococcygeal bursitis without providing any further details.^[5] Hayashi *et al.* described a 71-year-old patient with RA and multiple rheumatoid bursal cysts who had multiple bursal cysts on his elbow, knee, hand, back, and buttock; the patient was treated with a combination of surgery and sclerotherapy.^[2] In our case, the patient additionally had underlying RA [Table 1].

CONCLUSION

In patients with a history of RA, soft-tissue tumor-like lesions over the coccygeal region may represent RA-related bursitis. For these individuals, gross total lesion excision will likely result in symptom resolution.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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