Case Report
Orthopedics and Traumatology

Medical Journal of Islamic World Academy of Sciences
doi: 10.5505/ias.2024.15986 **2024;31(1): 43-46**

A Rare Cause of Coccydynia: Sacrococcygeal Chondrosarcoma

Recep ÖZTÜRK[1], İsmail Burak ATALAY[1], Fisun ARDIÇ YÜKRÜK[2]

[1] Department of Orthopedics and Traumatology, Ankara Oncology Training and Research Hospital, Ankara, Türkiye
[2] Department of Pathology, Ankara Oncology Training and Research Hospital, Ankara, Türkiye

Correspondence
Recep Öztürk
Mehmet Akif Ersoy Mahallesi 13, Cadde No. 56, Yenimahalle/, Ankara, Türkiye. Tel: 0 312 336 09 09, Fax: 0 312 334 03 5 06200.
e-mail: ozturk_recep@windowslive.com

ABSTRACT

Spinal chondrosarcomas are rare, with only some case series reported in the literature. Less than 10% of chondrosarcomas are localized in vertebrae, with the thoracic vertebrae being the most common site.

Sacral localization is rare; however, the case reported in our study involved a unique instance of chondrosarcoma affecting the distal sacrum and first coccygeal vertebrae. The patient presented with coccydynia. In this case, the grade 2 tumoral lesion was resected with wide margins, without chemotherapy and radiotherapy, and no relapse was reported at the 6-month follow-up.

Keywords: Chondrosarcoma, coccyx, complication, sacral tumor, sacrectomy

INTRODUCTION

Chondrosarcoma is a malignant cartilage forming bone neoplasm, accounting for almost 10% of all bone tumors (1). It is the third most common primary malignant bone tumor and is mostly commonly observed in individuals aged 30–60 years. It is classified as primary and secondary, with primary chondrosarcomas originating directly from bone and soft tissue and secondary chondrosarcomas arising from pre-existing benign cartilage (enchondroma or osteochondroma) (2,3).

Less than 10% of chondrosarcomas are localized in vertebrae, with the thoracic vertebrae being the most common site (4). Sacrococcygeal localization is rare, and here we present a case of sacrococcygeal chondrosarcoma.

CASE PRESENTATION

A 39-year-old male patient with coccydynia was admitted to our clinic. He had a 1-year history of a fall on his buttocks and pain over the coccyx since that time.

At the beginning, the pain was thought to be secondary to the trauma, and the patient waited for the pain to be relieved by itself. As the pain worsened, the patient intermittently sought treatment, receiving intramuscular injections, a donut cushion, and non-steroidal anti-inflammatory drugs. He underwent an x-ray examination only once in the past and was told that he had no pathological condition.

The skin over the coccyx was normal, with no hyperemia, edema, drainage, or any other finding on the physical examination. But the patient felt localized pain upon palpation.

The anteroposterior and lateral radiographs revealed fading of the distal sacrum and proximal coccyx. Magnetic resonance imaging (MRI) was performed with a prediagnosis of a tumoral lesion (Fig. 1a and 1b).

The MRI revealed a 7.5 × 5 cm^2 lobulated lesion with prominent hyperintense cystic areas. After intravenous contrast injection, expansion of the distal sacrum and coccyx was detected, with the tumor extending through the pelvic base and perirectal adipose tissue but sparing the rectal wall (Fig. 2a–2e).

No tumoral lesion was detected on thoracic CT and abdominal ultrasonography. Pathologic tracer activity was noted only in the sacrococcygeal area during whole-body bone scintigraphy.

An incisional biopsy performed in our clinic confirmed chondrosarcoma. The patient underwent wide resection, including the sacral third vertebra, while sparing all the branches (Figs. 3a, 3b, and 4a–4c). A plain radiograph was taken on the first postoperative day (Fig. 5a and 5b). The histopathologic examination diagnosed grade 2 chondrosarcoma, with clean surgical margins. The bladder and intestinal functions were perfect on the postoperative examination, and no perioperative complications were noted.

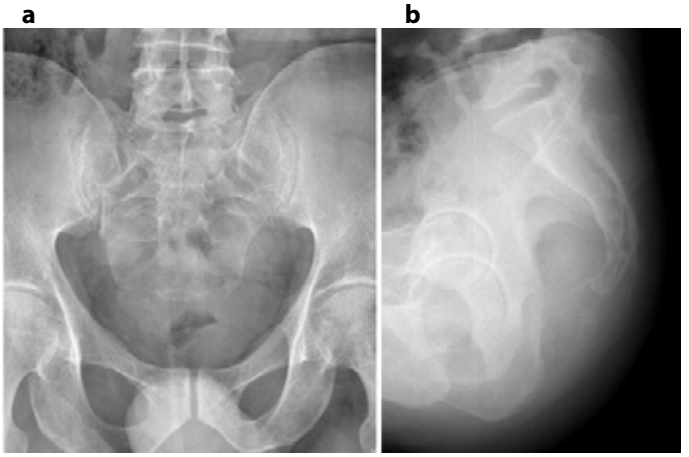

Figure 1 (a and b) Sacrococcygeal lytic lesion on two-plane plain radiography in the patient with coccydynia.

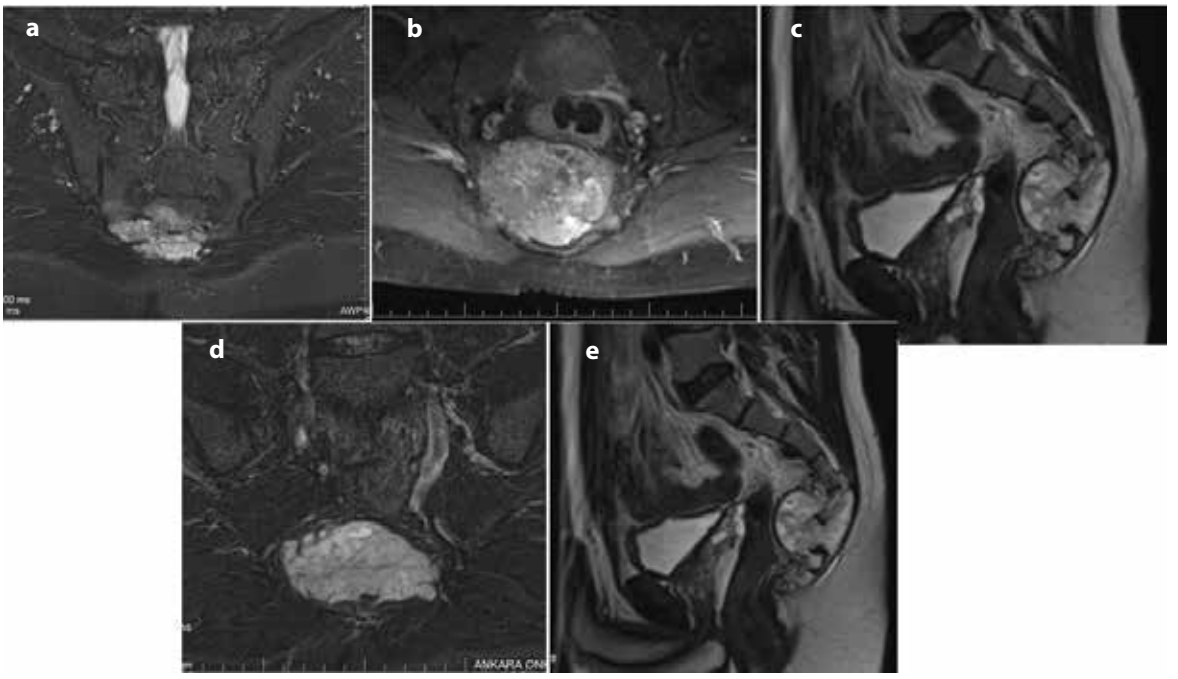

Figure 2 (a - e) MRI showing a 7.5 × 5 cm^2 lobulated lesion with prominent hyperintense cystic areas. After intravenous contrast injection, expansion of the distal sacrum and coccyx was observed.

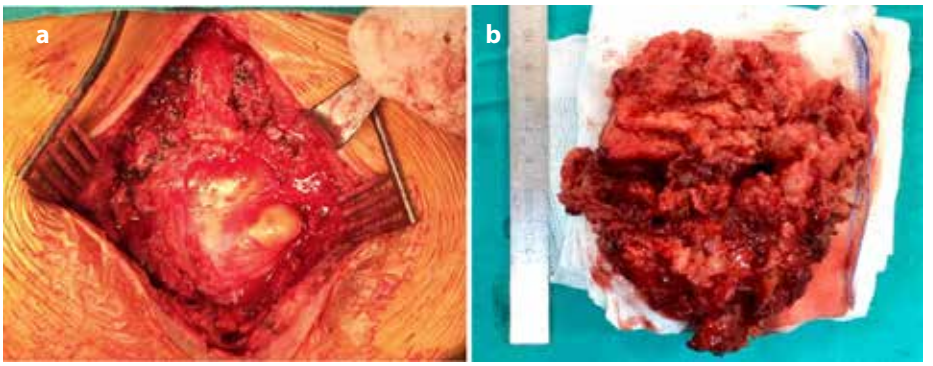

Figure 3 **(a)** Intraoperative view of the sacrococcygeal tumor. **(b)** Macroscopic image after tumor resection.

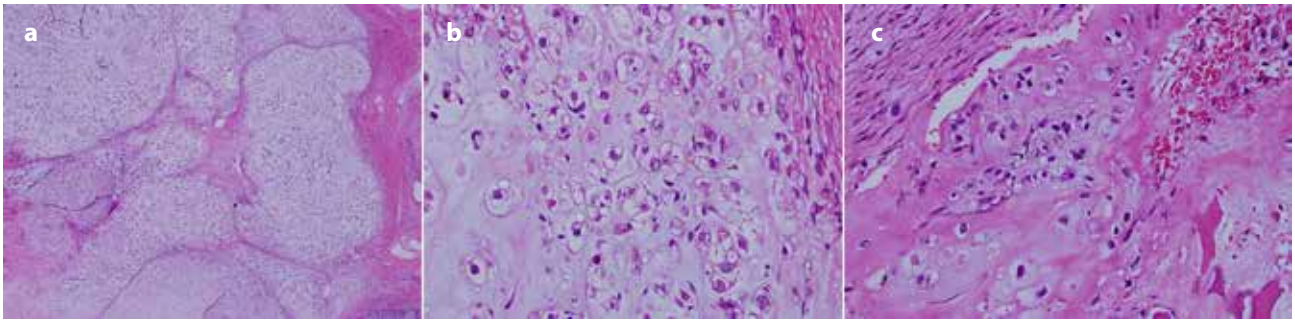

Figure 4 **(a)** Irregular lobules of malignant cartilage with fibrosis and partially myxoid matrix. **(b)** Atypical chondrocytes with varied nuclear shapes and sizes and hypercellularity, consistent with grade 3 chondrosarcoma. **(c)** Bone permeation and irregularly shaped dead bone spicules.

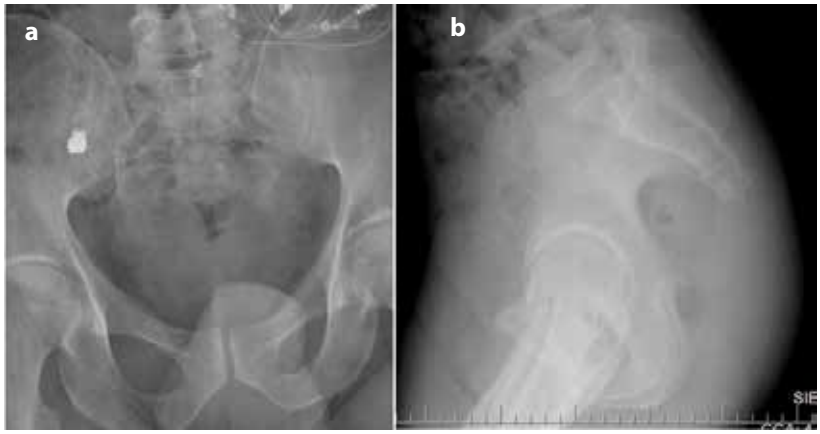

Figure 5 (a and b) Postoperative plain radiographs taken on day 1 following tumor resection.

The patient was readmitted with smelly drainage on the 12th day postoperatively, with wound edge separation. Debridement was performed for wound infection, culture was obtained, and antibiotic therapy was prescribed. The sutures were removed on the 15th day postoperatively, revealing a clean wound.

The evaluation in the first year postoperatively showed that despite no wound-related complications, no relapse was reported during the examinations (Fig. 6). The patient was satisfied with the quality of life, and the bladder–intestine functions remained normal.

DISCUSSION

Spinal chondrosarcomas are rare, with some case series reported in the literature (4,5). Although vertebral chondrosarcomas most frequently occur in thoracic vertebrae, cervical and lumbar chondrosarcomas also exist (5). Chondrosarcomas located around the sacrum are much rare. Nevertheless, the case we discussed in this study involved the distal sacrum and first coccygeal vertebrae, and the patient presented with coccydynia.

Chondrosarcomas affect men more than women. The most affected age interval is 30–60 years (3,4). In this study, the patient was a 39-year-old man. The tumors often appeared as vertebral or paraspinal calcified lesions (1).

The most common symptom of vertebral chondrosarcomas is a painless bump, followed by pain and neurologic findings (6). Coccydynia related to probable coccygeal involvement besides sacral pain was reported in the present case. The patient experienced aggravated pain only when sitting. He did not have much pain when standing.

Histologically, chondrosarcomas invade the bone, leading to the formation of irregularly shaped lobules with some fibrous bands. The atypical chondrocytes are embedded within the cartilaginous matrix, which may

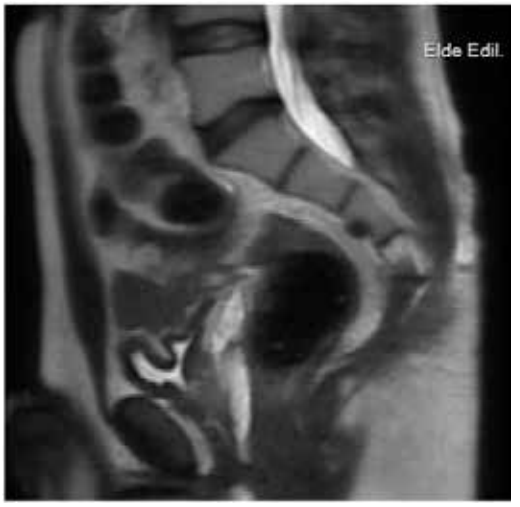

Figure 6 MRI at the 6-month follow-up showing no relapse.

sometimes be myxoid. The morphological features vary with tumor grade, and more aggressive tumors have higher degrees of cellularity and nuclear pleomorphism. Differential diagnosis should be made with chordoma, as chondromas are the most common primary malignant tumors in this region. The hallmark cells reflecting notochordal origin are the physaliphorous cells, which are absent in chondrosarcoma. If present, a myxoid matrix and lobular growth pattern may cause confusion with the chordoma. Immunohistochemistry is useful in difficult cases. Chordomas test immunopositive, whereas chondrosarcomas test immunonegative for EMA and CK (2,4,5-7).

The effective treatment is en bloc resection of the tumor (7). This treatment requires a multidisciplinary team including a neurosurgeon, an orthopedist, and a plastic and reconstructive surgeon (4,8). The surgeons in other departments were not needed because of the absence of spinal canal involvement and the need for skin reconstruction in our case.

Chondrosarcomas generally resist chemotherapy and radiotherapy (3). Radiotherapy may be considered for some inoperable patients for pain palliation or local control, and chemotherapy may be effective in certain high-grade chondrosarcomas (9,10).

In this case, the grade 2 tumoral lesion was resected with wide margins, and neither chemotherapy nor radiotherapy was administered. No relapse was reported at the 6-month follow-up.

The recurrence rate was 70% after resection with inadequate surgical margins and 10% after adequate surgical margins (11). The 5-year survival rates were 80%–90% for grade 1 chondrosarcomas, 50%–80% for grade 2 chondrosarcoma, and 40% for grade 3 chondrosarcomas (12). Larger, multicenter studies are needed to gain a better understanding of sacral chondrosarcomas.

CONCLUSIONS

Sacrococcygeal chondrosarcomas, though rare, should be considered in patients with coccydynia. Clinical and radiological evaluations of coccydynia should include the sacrum besides the coccyx. Wide resection remains the most effective treatment, as chemotherapy and radiotherapy are generally ineffective.

REFERENCES

1. Sundaresan N, Rosen G, Boriani S. Primary Malignant Tumors of the Spine. Orthop Clin North Am. 2009;40(1):21–36. https://doi.org/10.1016/j.ocl.2008.10.004
2. Stuckey RM, Marco RA. Chondrosarcoma of the mobile spine and sacrum.Sarcoma. 2011 : 274-281 https://doi.org/10.1155/2011/274281
3. Öztürk R, Ulucaköy C, Atalay İB, Yapar A, Karakoç Y. Management and retrospective analysis of pelvic ramus tumors and tumor-like lesions: Evaluation with 31 cases. Jt Dis Relat Surg. 2020;31:184–192. https://doi.org/10.5606/ehc.2020.72762
4. Strike SA, McCarthy EF. Chondrosarcoma of the spine: a series of 16 cases and a review of the literature. The Iowa orthopaedic journal. 2011;31:154–159
5. Katonis P, Alpantaki K, Michail K, Lianoudakis S, Christoforakis Z, Tzanakakis G et al. (2011) Spinal Chondrosarcoma: a review. Sarcoma;1–10 https://doi.org/10.1155/2011/378957
6. Merchant S, Mohiyuddin SM, Rudrappa S, Deo RP, Sagayaraj A, Menon LR.Cervical Chondrosarcoma- Rare Malignancy: A Case Report.Indian J Surg Oncol. 2014;5(4):293-6. https://doi.org/10.1007/s13193-014-0350-3
7. Boriani S, Biagini R, De Iure F, Bandiera S, Di Fiore M, Bandello L, et al. Resection surgery in the treatment of vertebral tumors. Chir Organi Mov 1998;83:53–64.
8. Ozturk R, Aydın M, Arıkan ŞM et al (2018) Extremity soft tissue sarcomas of uncertain differentiation: presentation, treatment, and outcomes in a clinical series of 59 patients. Bakırköy Tıp Dergisi 14:31–9. https://doi.org/10.5350/BTDMJB.20161217033551
9. McNaney D, Lindberg RD, Ayala AG, Barkley HT Jr, Hussey DH. Fifteen year radiotherapy experience with chondrosarcoma of bone. İnt J Radiat Oncol Biol Phys. 1982;8(2):187-90 https://doi.org/10.1016/0360-3016(82)90512-0
10. Gelderblom H, Hogendoorn PC, Dijkstra SD, et al. The clinical approach towards chondrosarcoma. Oncologist. 2008;13:320-9. https://doi.org/10.1634/theoncologist.2007-0237
11. Gibbs Jr CP, Weber K, Scarborough MT, Malignant Bone Tumors. İn: Beaty JH, ed. İnstructional Course Lectures. Rosemont, IL: AAOS;2002. p. 413-28
12. Yıldız Y. Malign Kemik Tümörleri. Türkiye klinikleri J Surg Med Sci. 2(52), 29-36, 2006.