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Case Report

Sacral Chondroma: A Case Report and Review of the Literature

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Abstract

Chondromas are benign cartilaginous tumors most commonly arising in the small bones, while sacral involvement remains exceedingly rare. We report the case of a 53-year-old male patient presenting with pelvic pain. Imaging studies revealed a pelvic mass that originates from the sacrum. The patient underwent complete surgical excision abdominal approach, postoperative recovery was uneventful, and no recurrence was observed at 5 years follow-up. This case highlights the diagnostic challenge of distinguishing sacral chondroma from low-grade chondrosarcoma and emphasizes the importance of complete resection and long-term surveillance to prevent local recurrence.

1. Introduction

Chordoma is a rare form of malignant neoplasm that originates from remnants of the notochord, a structure present during early fetal development. The tumor manifests primarily in the bones of the skull base and the spine, with a notable predilection for the sacrum.

Chordomas, a distinct type of primary bone tumor, account for approximately 1–4% of all such cases and are recognized as the most common tumors of both the sacrum and the cervical spine [1]. These tumors characteristically progress at a gradual rate and often remain asymptomatic for prolonged periods, frequently resulting in diagnosis at advanced stages when symptoms manifest, such as pain or neurological deficits due to compression of nearby structures [2]. Although chordoma can occur at any age, it most frequently affects individuals between the ages of 40 and 60.

The diagnosis is primarily based on radiological findings, including standard X-rays, CT scans, and MRI scans. The diagnosis is confirmed through histological analysis of the surgical specimen or biopsy [3].

Optimal management of chordomas is predicated on three fundamental pillars: early diagnosis, wide surgical resection and prolonged post-operative follow-up. It is evident that the utilization of chemotherapy as a therapeutic modality for chordomas has not yielded the expected outcomes. High-dose radiotherapy has been demonstrated to exhibit analgesic properties and delay recurrence; however, it does not currently hold any indication for curative treatment, unless when employed in conjunction with surgery [1].

Chordoma is distinguished by its high malignant potential, which carries with it a significant risk of local and locoregional recurrence. In contrast, metastases typically manifest at a later stage, often several years after the initial presentation [4].

2. Case report

We report the case of 53 years old male patient, who manifested symptoms of moderately severe posterior pelvic pain, radiating to the lower limb, alongside pollakiuria. The patient also exhibited very intense constipation, a weight loss of 10 kilograms over a period of six months was noted. A digital rectal examination was performed, and revealed rectal stenosis due to compression of a posterior mass.

A biological work-up was conducted, which revealed no abnormalities, including normal tumor markers (ACE and CA19-9).

Colonoscopy revealed the presence of an extra-mucosal posterior mass with mass effect on the rectum.

Abdominal-pelvic computed tomography (CT) revealed a pelvic mass developed at the expense of the first and second sacral vertebrae (S1 and S2) with anterior development and a mass effect on the rectum, which was deviated anteriorly and to the right, and an effect on the bladder, which was collapsed and deviated upwards.

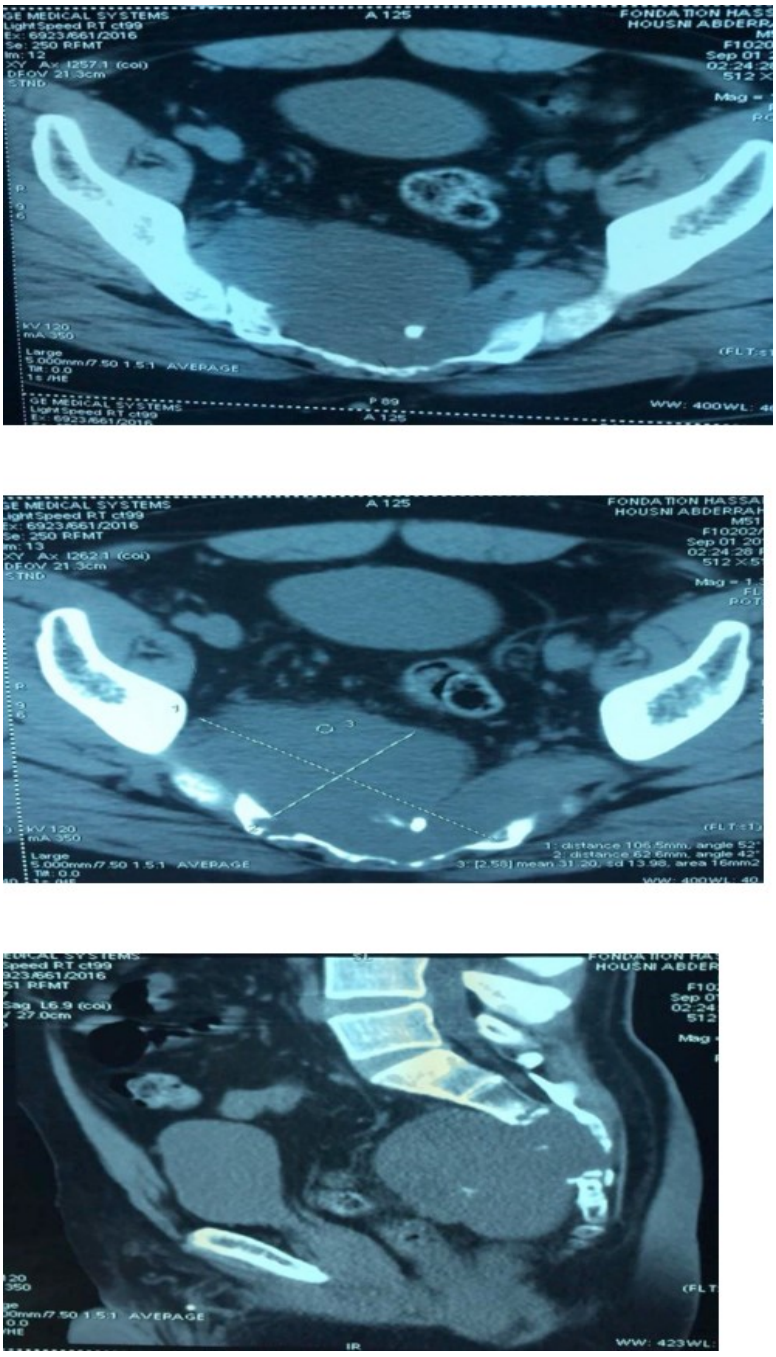

Figure 1: CT in transverse and sagittal sections showing the sacral tumor

MRI of the pelvis was performed to better characterize the lesion. This imaging revealed the presence of a sacral tissue tumoral process developed at the expense of S1-S2. The process has poly-lobed anterior contours and infiltrates posteriorly S3 and S4, with pelvic development and digestive, urinary, and neurological repercussions.

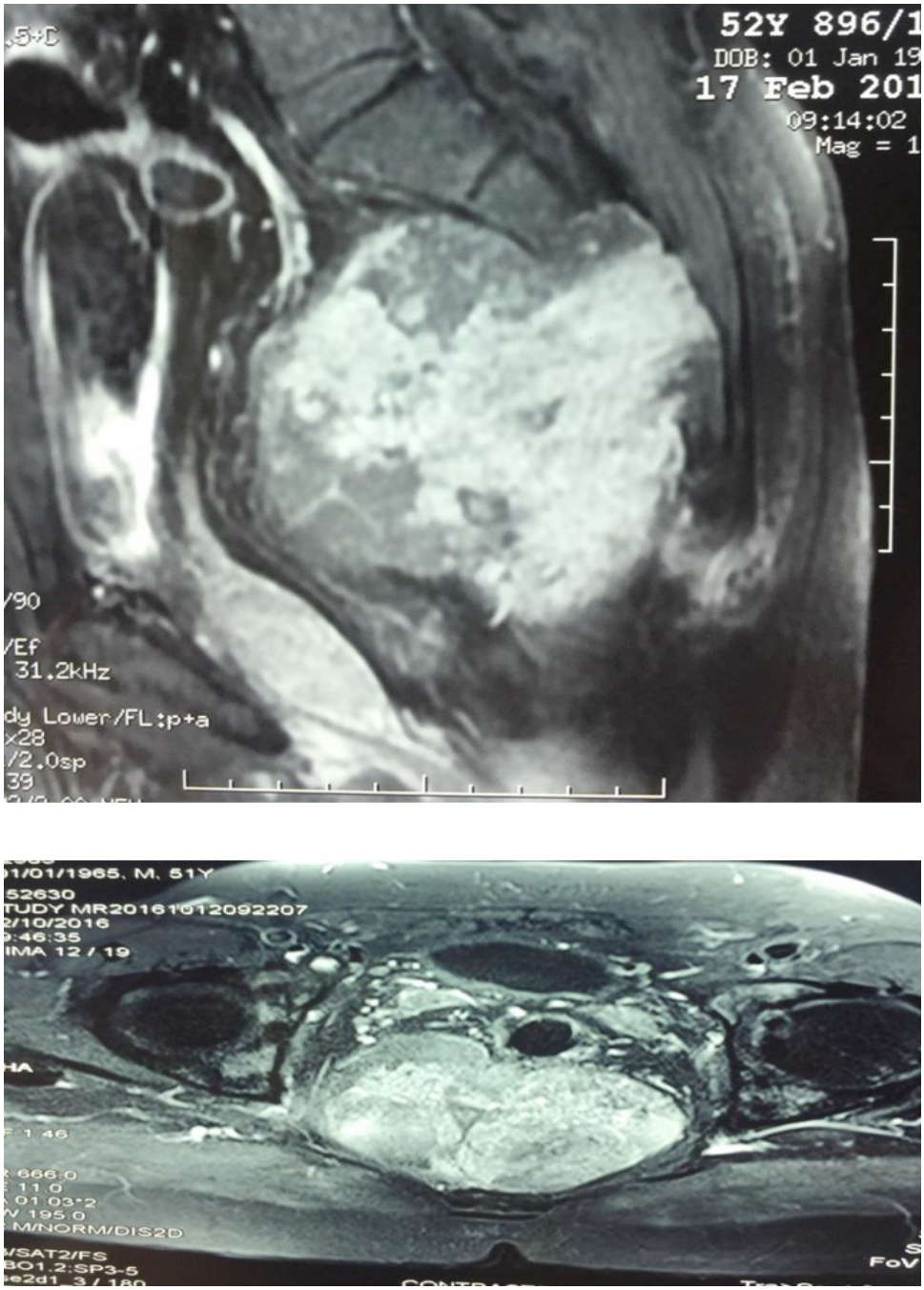

Figure 2: Pelvic MRI, after injection of gadolinium showing the tumor process centered on the sacrum

The patient underwent a surgical intervention involving resection, which entailed creating an incision along the sub umbilical midline. The exploratory procedure yielded no evidence of carcinosis or liver metastases.

Retro-rectal dissection in the avascular zone revealed a whitish, non-encapsulated mass with an cephaloid appearance.

Surgical specimen measured 19*9*4.5cm, immunohistochemical study showed AE1/AE3 positive labelling of tumor cells, CK7 positive, PS100 positive.

The final diagnosis, which was determined based on the morphological appearance and the immunohistochemical profile, was consistent with the diagnosis of sacral chordoma. The postoperative course was uneventful and characterized by a significant regression of neurological symptoms, a complete resolution of pain, and a concurrent resolution of constipation and pollakiuria.

One month after the initial treatment, the patient underwent adjuvant radiotherapy at 50 Gray for one month at the tumor site. This treatment was prescribed to target the intraosseous lesions present in the sacrum.

3. Discussion

Chordoma is a tumor of embryonic origin. It is attributed to the secondary proliferation of persistent cellular islands within the vertebrae or the skull base. These cellular islets are the remnants of an embryonic organ known as the dorsal chord or notochord [4].

Chordoma is a rare tumor. The incidence of this condition varies across studies, ranging from 0.5 to 8 cases per million persons per year [5].

Chordoma constitutes 1-4% of primary malignant bone tumors [4]. The impact of the disease is found to be significantly more pronounced among males than among females (sex ratio $\frac{M}{F} = \frac{3}{1}$) [1].

The clinical manifestations of the tumor are influenced by its dimensions and anatomic location. Such delays are typically attributed to the protracted nature of tumor evolution [1].

The clinic is often dominated by [6].

The predominant symptom is pain. The tumor's potential to evolve and persist without causing pain for an extended period is contingent on its local or local-regional origin [4].

This pain is secondary to the compression and/or invasion of neighboring organs, specifically nerve plexuses and nerve roots in the region, and can thus manifest in several forms.

- Low back pain without radiation.
- Gluteal or sacral pain, aggravated by sitting.

Digestive disorders: Defecation problems, Constipation and sometimes rectal bleeding, Occlusive syndrome.
Urinary disorders: Pollakiuria, Dysuria, Urinary incontinence.
Neurological disorders [7]: The presacral location of the chordoma elucidates the presence of neurological signs resulting from compression or invasion of the various nerve plexuses and roots in the region, manifesting as paresis or paresthesia of the lower limbs, saddle anesthesia, and indications of radicular compression of the last sacral roots or myeloradicular compression.

In the context of chordoma, the utilization of CT has been instrumental in facilitating therapeutic decision-making. This examination is widely regarded as the gold standard for diagnostic confirmation. This technology facilitates precise diagnosis, with the capability to detect malignancies in 100% of cases, even in instances of minute tumors that are not discernible through physical examination. The determination of the nature of the mass can be achieved through the measurement of its density, the assessment of its character, topography, and limits, and the potential extension of its local and distant extent [3].

This examination also enables us to better define the data obtained on the front and profile views of an unprepared abdomen.

In conjunction with CT, pelvic MRI offers high topographical accuracy, particularly for soft tissues in the epidural and intradural space [8]. This modality can be utilized on T1 and T2 sequences in sagittal and axial slices to assess: the lesion's liquid component (intense hyper-signal on T2 sequence) or solid component, its contiguity with the rectum, its limitations.

In order to elicit a diagnosis and delineate tumor extension, with particular emphasis on the upper region, it is imperative to ascertain the optimal surgical access, especially in the sagittal plane.

Furthermore, MRI has been shown to possess superior sensitivity compared to CT in the assessment of nerve root relationships and the presence of any embrochment. Conversely, angiograms and venograms offer a more comprehensive visualization of vascular relationships. The use of magnetic resonance imaging (MRI) with gadolinium injection prior to and following neoadjuvant treatment facilitates the assessment of tumor volume [9].

On MRI, sacrococcygeal chordomas are tumors with hypo- or iso- signal on T1-weighted sequences (compared with muscle masses), and heterogeneous hypersignal on T2-weighted sequences. The chordoma is lobulated in appearance, with extension into the sacral and epidural spaces [9].

Consequently, radiological determination of the tumor's topography in relation to the S1 and S3 vertebrae constitutes a pivotal pre-therapeutic component.

Magnetic resonance imaging (MRI) facilitates the examination of the lesion in all three planes of space, thereby providing the surgeon with invaluable information regarding the tumor's locoregional extension. This includes precise lesion boundaries, potential extensions to the gluteal muscles, rectum, bladder, sciatic nerve, the presence of intracanal extension, and associated lymph node involvement, among other crucial details [9].

The only chance of cure lies in surgical resection "en bloc", with limited excision in healthy areas [9]. When this type of excision is possible, the tumor recurrence rate remains relatively high [4].

Radiotherapy is used to complement surgery, either immediately or in the case of local recurrence, or when surgery is impossible [8].

A retrospective analysis of the clinical experience with chemotherapy reveals its limitations in most cases. However, its utility in the management of secondary localizations remains a subject of further investigation [4].

The prognosis of sacral chordomas is determined primarily by the extent and quality of the surgical excision.

Complications arising from the surgical procedure itself, in addition to secondary complications, may occur. These secondary complications include local recurrence, which is a significant predictor of reduced overall patient survival [4].

In our case, the patient was seen on a regular basis, the prognosis was favorable and there was no local recurrence or tumor metastasis.

4. Conclusion

Adult sacral chordomas represent a rare occurrence.

The prevalence of this condition affects individuals of all ages, with a notable peak in the fifth decade. It exhibits a male predominance, with a ratio of males to females of 3 to 1.

The diagnosis is made very late, due to clinical latency. The presentation of these tumors may manifest as discrete functional symptoms, including atypical pelvic pain, low back pain, signs of compression of the lumbosacral roots, sensory-motor deficits of the lower limbs, and urinary and digestive sphincter disorders.

The positive diagnosis is based on the presence of a fixed, rough, firm presacral mass, subsequently confirmed by computed tomography (CT) and magnetic resonance imaging (MRI).

A definitive diagnosis of the lesion can only be made through an anatomopathological examination following complete surgical removal of the tumor.

Treatment is exclusively surgical, involving total excision wherever feasible to avert complications and the rare instances of recurrence.

Radiotherapy is used to complement surgery, either immediately or in the event of local recurrence, or even when surgery is not possible.

However, surgery to remove a sacral chordoma is a challenging procedure due to the anatomical relationships of the retro rectal space. This anatomical configuration makes the procedure susceptible to intra- and post-operative complications, including hemorrhage, neurological lesions, and injury to the external sphincter.

Sacral chordoma is distinguished by its high malignant potential, resulting in an elevated risk of local and locoregional recurrence. In contrast, metastases manifest at a later stage, often several years after the initial presentation.

Article Information

Ethics Approval and Consent to Participate: Written informed consent of the patient to use clinical information and photographic material for the publication was obtained.

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Disclaimer (Artificial Intelligence): The author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc.), and text-to-image generators have been used during writing or editing of manuscripts.

Competing Interests: Authors have declared that no competing interests exist.

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