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Osteoid Osteoma of the Sacrum - Case Report and Review of Literature

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Authors' contributions

This work was carried out in collaboration among all authors. Authors VVN and AR designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors VVN, AR and AA managed the analyses of the study. Authors AR and AA managed the literature searches. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Sacrum is an extremely rare site for osteoid osteoma. We present a 25 year-old male, a computer operator who presented with chronic back pain of 4 years duration. He was initially investigated with routine x-rays and lab investigations and was treated for chronic back pain at another facility. Extensive work up at our clinic revealed a diagnosis of Osteoid Osteoma of Right 2nd Sacral vertebral arch Interlesional resection of the tumor was done and this was verified on histopathology. The patient had complete relief of pain following surgery. Repeat CT scan done at one year follow up. It showed complete removal of the Nidus. To the best of our knowledge there have not been many case reports in English Literature from the Indian subcontinent. There are many studies in which tumor of lumbar region is one of the causes of radiating pain in lower extremities but to the best of our knowledge this finding is unique and can also be an important cause of radicular pain.

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1. INTRODUCTION

Jaffe was the first to designate a benigh condition of the bone characterized by the formation of a nidus of vascularized osteoid tissue with sclerosis of surrounding bone (Jaffe-1) and is usually seen in lower extremity in young adults. Further elaboration of this entity was done by Bregsternd [1]. Despite the small size it can cause considerable pain which is aggravated at night time and classically responds to Non steroidal anti inflammatory drugs (NSAIDS)/salicylates). On quite a few occasions symptomatology predates x-ray appearance [2].

The incidence of osteoid osteoma in vertebrae is about 0-20% with average of about 10% [1]. Our patient had a history of chronic back pain more at night associated with radiating pain to his right lower limb mimicking Disc prolapse for which he was treated elsewhere with medication and physiotherapy for two years and had no relief. MRI of the lumbosacral spine done at our institution was suggestive of infection and a CT scan osteoid osteoma which was again confirmed on final histopathology. Our case highlights the importance of the fact that one should go for better imaging modalities when patient is not responding to conventional treatment.

2. CASE REPORT

A 25 year-old male a computer operator was admitted with chronic back pain and radiating pain to his right lower limb and going till the foot (where posterior or anterior, specify) and had been waxing and waning for 4 years. Pain was worse at night and better with medications. He had been treated elsewhere or the above mentioned symptoms with working diagnosis of lumbosacral strain/possible prolapse. Enlarged x-rays revealed possible destruction in sacrum (Figs. 1 and 2). MRI done few months ago was suggestive of infection (only available-no images)-tuberculosis although is sputum analysis and blood work was negative. His physical examination of the revealed he had point tenderness over the right side of the Sacrum and over the right Sacroiliac joint. Straight leg raising test was negative. Hip examination was normal. Spinal movements

were normal. No neurological deficit was noticed. Patient was offered a CT- guided biopsy of the lesion to prove/disprove infection and he declined it. CT scan (Figs. 3 and 4) was done and revealed, the typical feature of Osteoid Osteoma of the right 2nd sacral vertebral lamina suggested by a central nidus measuring <0.5 mm, surrounded by sclerosed bone measuring 0.5 mm. Patient was offered intralesional excision and he agreed for the procedure. Since we do not have facility of CT guided excision we decided to proceed with open excision.

Under general anesthesia, patient positioned in knee chest position. Mid line skin incision was made over the sacrum measuring about 8 cms. The paraspinal muscles were striped. Second sacral arch was identified under image guidance. A flake of bone got dislodged from the site of the lesion while stripping the paraspinal muscles over the S2 sacral lamina on the right side and on deroofing the lesion, a brownish red tissue was seen surrounded by sclerosed bone. The lesion over the S2vertebral lamina on the right side was removed by Interlesional excision. The under lying nerve root was also exposed and was found to be free. The specimen was sent for histopathology which confirmed the diagnosis of osteoid osteoma. Wound lavage was given and the skin was closed in layers. Patient had complete relief of pain following surgery. The radiating pain had also completely subsided. He had no night pain thereafter and had complete resolution of symptoms. Skin staples were removed on the 12 post-operative day. Repeat CT- scan of the Sacrum done at 6 months (Fig. 5) showed complete removal of the lesion.

3. DISCUSSION

Osteoid Osteoma is a benign osteoblastic tumor that has been coined by Jaffe and elaborated by Bergstrand [4,1]. The usual size of Osteoid Osteoma about 1.5-2 cm and characterized by an osteoid-rich nidus in a highly loose, vascular connective tissue. The nidus is well-demarcated and may contain a variable amount of calcification and is rich in nerve fibers and high concentration of prostaglandins [2]. Surrounding the nidus is a zone of sclerotic but otherwise normal bone.



Fig. 1. X-ray of pelvis with sacrum with no definite pathology (mark the lesion with arrow) [3]



Fig. 2. Enlarged view of x-ray showing possible destruction (mark the lesion with arrow)



Fig. 3. Arrow shows nidus and sclerosis



Fig. 4. Arrow shows sclerosis of the sacrum



Fig. 5. Post-op CT-scan showing laminectomy defect and complete absence of the nidus and the surrounding sclerotic bone[3]

Osteoid osteoma is seen in spinal locations in 7-20 percent of cases and is predominantly seen in cervical and lumbar regions lumbar [2-11]. The usual clinical manifestation is pain in affected part although painless variants have been reported [11]. The age of presentation is usually 10-30 years [7]. Our patient had a similar presentation.

The tumor characteristically spares the vertebral body and has a predilection for the posterior elements, most commonly affecting the cancellous lamina, spinous process, and pedicle [5,6]. The same authors also reported that this predilection is seen in 75% of cases, with 33% involving the lamina, 20% involving the articular facets, and 15% involving the pedicles. Lumbar

spine location predominates in 60% of the cases and sacrum is involved in only 2 percent of cases.

Osteoid Osteoma of Sacrum can be a great mimicker and symptomatology can be confused with infection/ disc prolapse. This finding was demonstrated in our case as patient suffered for 4 years. Even a sensitive investigation like MRI did not lead to a diagnosis which was definite. Combined with normal blood work, it was a diagnostic dilemma .No bone scan was done in our case because of cost constraints. The only classical feature was response to salicylates. The characteristic radiology was picked up on CT scan and revealed a radiolucent nidus surrounded by sclerosis. Ct scan has been shown superior to MRI because it is unaffected by calcification [12]. We also had similar observations.

Since 1990 trend has been towards CT guided excision of the tumor /radiofrequency ablation of the lesion and these have resulted in good outcomes [13-18]. It is of immense benefit in inaccessible locations. However incomplete resections and recurrence rate remains high with the minimally invasive techniques [15]. We did not have the option of minimally invasive techniques and hence went in for open excision. Patient was doing well at 1 year follow up.

4. CONCLUSION

To summarize young adults with unresponsive spinal pain should undergo fine cut Ct scan to rule out rare etiologies like osteoid osteoma. Open excision still remains a viable option if more technology savvy options are not available.

CONSENT

Patients consent was obtained for reproducing his images.

ETHICAL APPROVAL

As per international standard ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

- Jaffe HL. Osteoid osteoma. A benign osteoblastic tumor composed of osteiod and atypical bone. Arch. Surg. 1935;31(1): 709-7335.
- Capanna R, Ayala A, Bertoni F, Picci P, Calderoni P, Gherlinzoni F, Bettelli G, Campanacci M. Sacral osteoid osteoma and osteoblastoma: A report of 13 cases. Arch Orthop Trauma Surg. 1986;105(3): 205-210.
- Rinald AG, Vijaynarasimhan V, Kumar SK. A rare case of osteoid osteoma in second sacral arch. Research Journal of Pharmaceutical Biological and Chemical Sciences. 2015;6(2):2006-2010.
- 4. Paus BC, Kim TK. Oteoid osteoma of the spine. Acta Orhtop. Scandanavia. 1963;33(2):24-29.
- Makley JT, Dunn MJ. Prostaglandin synthesis by osteoid osteoma. Lancet. 1982:2:42.
- Unni KK. Dahlin's bone tumors. General aspects and data on 11,087 cases. 5th Ed. Philadelphia: Lippincott-Raven Publishers; 1996.
- 7. Bruce M, Cameron M, Friend LF. Osteoid osteoma of the sacrum. Report of a case. J Bone Joint Surg Am. 1954;36:876-877.
- 8. Biagini R, Orsini U, Demitri S, Bibiloni J, Ruggieri P, Mercuri M, Capanna R, Majorana B, Bertoni F, Bacchini P. Osteoid osteoma and osteoblastoma of the sacrum. Orthopedics. 2001;24:1061–1064.
- 9. Schmitz A, Diedrich O, Schmitt O. Sacral osteoid osteoma- A rare cause of back pain in childhood and adolescence. Klin Padiatr. 2000;212(3):110-112.
- Nasr FW, Sarru EA, Haddad FS. Sacral osteoid osteoma in a 10 year old girl. J Med Liban. 1992;40(4):216-8.
- Villas C, López R, Zubieta JL. Osteoid osteoma in the lumbar and sacral regions: Two cases of difficult diagnosis. J Spinal Disord. 1990;3(4):418-22.
- 12. Gamba JL, Martienez S, Apple J, Harrelson JM, et al. Computed tomography of skeletal osteoid osteoma. AJR. 1984;142:769-72.
- Voto SJ, Cook AJ, Weiner DS, Ewing JW, Arrington LE. Treatment of osteoid osteoma by computed tomography guided excision in the pediatric patient. J Pediatr Orthop. 1990;10:510–513.
- Rosenthal DI, Hornicek FJ, Torriani M, Gebhardt MC, Mankin HJ. Osteoid

- osteoma: Percutaneous treatment with radiofrequency energy. Radiology. 2003;229:171–175.
- Cribb GL, Goude WH, Cool P, Tins B, Cassar-Pullicino VN, Mangham DC. Percutaneous radiofrequency thermocoagulation of osteoid osteomas: Factors affecting therapeutic outcome. Skeletal Radiol. 2005;34:702–706.
- Chaudhary RK, Acharya S, Chahal RS, Kalra KL. Fluoroscopy guided percutaneous transpedicular biopsy of vertebral body lesion. J Nepal Health Res Counc. 2019;17(2):163-167. DOI: 10.33314/jnhrc.v0i0.1863
- Fukuda S, Susa M, Watanabe I, Nishimoto K, Horiuchi K, Toyama Y, Morioka H. Computed tomography-guided resection of osteoid osteoma of the sacrum: A case report. J Med Case Rep. 2014;18(8):206.
 DOI: 10.1186/1752-1947-8-206
- Garge Shaileshkumar, Keshava Shyamkumar N, Moses Vinu, Chiramel George K, Ahmed Munawwar, Mammen Suraj, Madhuri Vrisha. Radiofrequency ablation of osteoid osteoma in common and technically challenging locations in pediatric population. Interventional Radiology. 2017;27(1):88-91.

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