Paediatr Croat. 2025;69:186-91
ORIGINAL SCIENTIFIC PAPER / *IZVORNI ZNANSTVENI RAD*
journal.paedcro.com
http://dx.doi.org/10.13112/pc.1110

Osteoid osteoma of the spine and scoliosis: about 5 cases

Zied Jlalia[1], Zied Mansi[2], Guermazi Bassem[1], Arfa Wajdi[1], Sami Bahroun[1], Atef Ltifi[2], Hedhili Guedhami[2], Abderrahmen Souid[3], Slah Soui[4], Zaidi Bacem[5], Samir Daghfous[1]

Introduction: Scoliosis, a sideways curvature of the spine, is generally asymptomatic in children but may be associated with pain in adults. This study explores five cases of painful scoliosis caused by osteoid osteoma (OO), a benign lesion often misdiagnosed due to its rarity and subtle presentation. The aim was to examine the diagnostic challenges and treatment outcomes in cases of spinal OO presenting with scoliosis.

Methods: A retrospective study was conducted on five patients treated between 1986 and 2016, who presented with painful scoliosis attributed to OO. Radiographic imaging, including computed tomography (CT) in four cases and magnetic resonance imaging (MRI) in one case, was used for diagnosis. All patients underwent surgical resection of the OO by posterior approach with postoperative bracing. The average follow-up period was 5 years and 5 months.

Results: The average age of the patients was 12.2 years. The time to diagnosis averaged 13 months. Scoliosis was present in all cases, with two cases showing dorso-lumbar and three showing lumbar curvature. CT scans identified the OO in all patients. The lesion sites included the pedicle (2 cases: L3, L4), articular facets (1 case: L4), posterior hemi-arch (1 case: D12), and lamina (1 case: D10). Surgical resection resulted in spontaneous correction of scoliosis in three patients within an average of seven months, while one patient required additional corrective surgery due to severe scoliosis (57 °) diagnosed after 24 months. Spondylodesis with autologous bone graft was performed in two cases to ensure stability, and one patient required revision surgery due to resection at the wrong vertebral level. Postoperative bracing was used for three months in four patients. Histological analysis confirmed the diagnosis of OO in all cases. Outcomes were favorable in four patients, with resolution of pain and improved function. One case of graft nonunion occurred after revision surgery; however, the patient remained satisfied at 10 years of follow-up. The results of our series are summarized in Table 1.

[1] Orthopedic pediatric department. Kassab institute of orthopedic surgery, Workers' Street, Ksar Saïd, Manouba 2010 Tunis, Tunisia
[2] Department of Orthopedic Surgery, IBN EL JAZZAR University Hospital of Kairouan, Ibn Al Jazzar Street 3100 Kairouan, Tunisia
[3] Department of Orthopedic Surgery, Hospital Houcine Bouzayaiene of Gafsa, Douali 2100 Gafsa, Tunisia
[4] Department of anesthesia, IBN EL JAZZAR University Hospital of Kairouan, Ibn Al Jazzar Street 3100 Kairouan, Tunisia
[5] Department of General Surgery, IBN EL JAZZAR University Hospital of Kairouan, Ibn Al Jazzar Street 3100 Kairouan. Tunisia

Conclusion: Spinal OO should be considered in cases of painful scoliosis, especially in children. Early diagnosis through CT imaging is crucial for effective management. Surgical resection provides good outcomes, and timely intervention prevents the development of structural scoliosis.

Key words: SPINE; BONE NEOPLASMS; OSTEOMA, OSTEOID; MAGNETIC RESONANCE IMAGING

INTRODUCTION

Scoliosis is a sideways curvature of the spine that most often occurs before puberty. The cause of most scoliosis is unknown. While back pain is the first sign of scoliosis in adults, scoliosis is rarely painful in children. This study aims to describe five cases of scoliosis caused by osteoid osteoma (OO) that posed diagnostic and therapeutic challenges.

MATERIAL AND METHODS

A retrospective study was performed and included five patients presenting with painful scoliosis related to OO. All patients were treated between 1986 and 2016. Radiographs were performed in all cases, CT in four cases, and MRI in one case. MRI was performed in this single case due to diagnostic uncertainty at the time, although CT is generally considered more reliable. All patients underwent open resection of the OO en bloc by posterior approach, with bracing for three months. Postoperative radiographs were performed for all cases. The mean follow-up was 5 years and 5 months.

RESULTS

The average age was 12.2 years (range: 9–16 years). The average time to diagnosis was 13 months (range: 1 month–3 years). The main presenting complaint was painful spinal deformity in all cases. Radiographs showed scoliosis in all patients: dorso-lumbar in two cases and lumbar in three cases. CT scanning demonstrated the lesion in all cases. The OO was located in the pedicle (2 cases), articular facet (1 case), posterior hemiarch (1 case), and lamina (1 case). En bloc resection of the OO was performed in all cases. Spondylodesis with autologous bone graft was used in two patients to ensure vertebral stability. Revision surgery was required in one case due to an initial resection at the wrong vertebral level. Postoperative bracing was applied for three months in four patients. Histology confirmed OO in all cases. Scoliosis was corrected spontaneously in three patients within an average of seven months after resection. One patient with severe scoliosis (57°) diagnosed after 24 months required surgical correction with internal instrumentation (Figure 1). In one case, scoliosis persisted after two years of follow-up. Overall outcomes were favorable in four patients, with pain relief and functional improvement. A single case of graft nonunion occurred; however, the patient remained satisfied with the outcome at 10 years of follow-up. The results of our series are summarized in Table 1.

DISCUSSION

OO is a benign lesion first described in 1935 by Jaffe. OO may occur at any age, but it is most common in the femur and tibia; fewer than half of cases involve other locations (1). Spinal OOs are most often located in the lumbar spine (60 %), followed by cervical (27 %), thoracic (12 %), and sacral (2 %) regions (2). According to Janin et al., the most frequent sites are the lamina, pars interarticularis, pedicle, and transverse process (3).

OO should be considered as a potential cause of neck or back pain, or painful scoliosis, in young patients (1). Nocturnal pain is a typical feature and is often severe (4, 5). Pain relief with salicylates occurs in approximately 30 % of cases. In atypical scoliosis, such as left convex thoracic scoliosis or right convex lumbar scoliosis (Figure 2), OO should be suspected, as the lesion is usually located at the apex of the concavity (6).

Some authors suggest that scoliosis results from muscle spasm, which may extend across several vertebral levels (7). Saifuddin et al. proposed that the asymmetric location of OO is a key factor in scoliosis development (8). Delayed diagnosis, sometimes extending over months or years, is common; in our series, the mean diagnostic delay was 13 months, consistent with reports in the literature (9). Misdiagnosis often results from the

Table 1. Summary of the results of our series

Patients (N)	Age (Y)	Gender	Pain duration (month)	Reason for consultation	Pre operative symptoms	Site	Part	Imaging	Operation	Follow up (months)
1	14	M	12	Night time pain Spinal deformity	Scoliosis	D 12	Posterior hemi-arch	Radiographs CT	Resection+ Internal fixation	44 Asymptomatic Spine aligned
2	9	M	36	Low back pain	Scoliosis	L 4	Articular facet	Radiographs	Laminectomy + Bracing	76 Asymptomatic
3	16	M	12	Low back pain	Scoliosis	L 4	Pedicule	Radiographs CT	Resection + Spondylodesis + Bracing	120 Recurrence of pain Pseudarthrosis
4	12	M	4	Low back pain Spinal deformity	Scoliosis	D 10	Lamina	Radiographs CT	Resection + Spondylodesis + Bracing	64 Asymptomatic Spine aligned
5	10	W	1	Low back pain	Scoliosis Cruralgia	L 3	Pedicule	Radiographs CT MRI	Resection + Bracing	24 Asymptomatic Persistance of scoliosis

Figure 1. Standard radiograph of the spine, showing a dorsal scoliosis without any other abnormality

complexity of spinal anatomy and the subtle radiographic findings.

Radiographs may appear abnormal in over 70 % of cases, but the nidus is often difficult to detect, particularly in the cervical and thoracic regions. Most OOs are osteosclerotic with or without a visible nidus (Figure 3). In the spine, due to its anatomical complexity and smaller cancellous spaces, sclerosis is often less pronounced (10). Differential diagnoses include osteoblastoma, osteoblastic metastasis, enostosis (bone island) (Figure 4), infection, lymphoma, and facet joint abnormalities.

Figure 2. Standard radiograph of the spine, showing a lumbar right scoliosis with a calcification of the left pedicle of L12

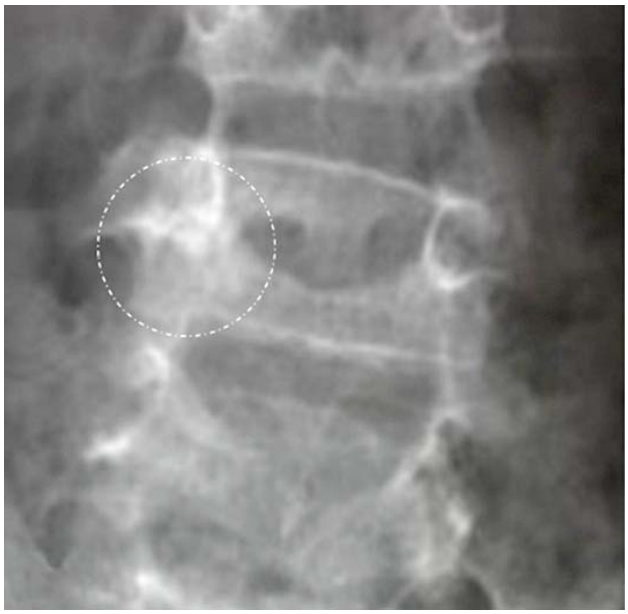

Figure 3. Standard radiograph of the spine, showing an osteosclerotic lesion of the pedicle of the vertebra with or without recognition of a nidus

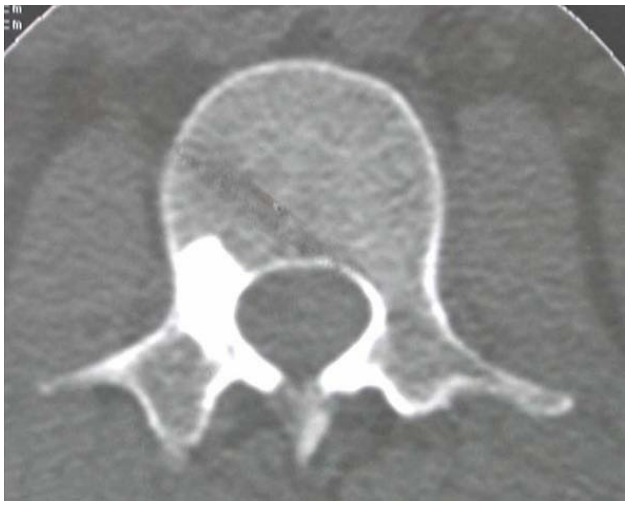

Figure 4. Radiographic differential diagnosis for a dense pedicle includes osteoblastoma, osteoblastic metastasis, and enostosis (Bone Island)

CT is widely regarded as the gold standard for identifying OO (11, 12) and is also essential for surgical planning and verifying complete resection (13). MRI is less specific, with variable appearances that may mimic malignancy or inflammation (14, 15). However, MRI remains useful when neurological symptoms are present or when soft tissue involvement is suspected. Scintigraphy can also be useful if radiographs are inconclusive, particularly in cases of back pain without deformity (16).

In our series, all patients underwent surgical resection. Although spontaneous resolution of OO has been reported (17, 18), surgery is generally recommended to prevent painful scoliosis from becoming structural (1, 7, 19). The surgical approach depends on the severity of scoliosis, the location of the lesion, and spinal stability. Posterior fusion is not always necessary and should be reserved for cases where stability is compromised (20). In our series, spondylodesis was performed in two patients for stabilization. Pain was relieved within hours to days after surgery in four cases, consistent with previous reports (1). Persistent symptoms typically occur after incomplete resection, as seen in one of our cases with resection at the wrong vertebral level.

CONCLUSION

Spinal OO is rare but should be suspected in children presenting with painful scoliosis. Radiographs are often insufficient for diagnosis, and CT should be performed promptly to avoid delays. MRI may be helpful in cases with neurological in-

volvement, but CT remains the most reliable tool. Early diagnosis and complete surgical resection are crucial to achieving pain relief and preventing scoliosis from becoming structural.

REFERENCES

1. Mehta MH. Pain provoked scoliosis. Observations on the evolution of the deformity. Clin Orthop Relat Res 1978: 58–65.
2. Sapkas G, Efstathopoulos N, Papadakis M. Undiagnosed osteoidosteoma of the spine presenting as painful scoliosis from adolescence to adulthood: a case report. Scoliosis Journal 2009;4.
3. Janin Y, Epstein JA, Carras R, Khan A. Osteoid osteomas and osteoblastomas of the spine. Neurosurgery 1981;8: 31–8. doi:10.1227/00006123-198101000-00007.
4. Zileli M, Cagli S, Basdemir G, Ersahin Y. Osteoid osteomas and osteoblastomas of the spine. Neurosurg Focus 2003; 15:E5. doi:10.3171/foc.2003.15.5.5.
5. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg 2011;19:678–89. doi:10.5435/00124635-201111000-00004.
6. Kransdorf MJ, Stull MA, Gilkey FW, Moser RP Jr. Osteoid osteoma. Radiographics 1991;11:671–96. doi:10.1148/radiographics.11.4.1887121.
7. Bléry M, Leroux B. Ostéome ostéoïde vertébral. A propos de six cas. Ann Radiol 1978;21:59–63.
8. Saifuddin A, White J, Sherazi Z, Shaikh MI, Natali C, Rransford AO. Osteoidosteoma and osteoblastoma of the spine, factors associated with the presence of scoliosis. Spine 1998;23:47–53.
9. Kirwan EO, Hutton P. Osteoid osteoma and benign osteoblastoma of the spine: clinical presentation and treatment. J Bone Joint Surg [Br] 1984;66:21–6.
10. McLeod RA, Dahlin DC, Beabout JW. The spectrum of osteoblastoma. AJR Am J Roentgenol 1976;126:321–5. doi: 10.2214/ajr.126.2.321.
11. Schmitz A, Diedrich O, Schmitt O. Sacral osteoid osteoma: A rare cause of back pain in childhood and adolescence. Klin Padiatr 2000;212:110–2.
12. Bloem JL, Kroon HM. Osseous lesions. Radiol Clin North Am 1993;31:261–78. doi:10.1016/s0033-8389(22)02857-3.
13. Katz K, Kornreich L, David R, Horev G, Soudry M. Osteoid osteoma: resection with CT guidance. Isr Med Assoc J 2000;2:151–3.
14. Jalel OA, Wulff C, Varmarken JE. Osteoid osteoma of the cervical spine. Ugeskr Laeger 2000;162:2446–7.
15. Farrokh D. Erroneous diagnosis of subperiosteal osteoid osteoma: value of imaging techniques? J Radiol 1999;80: 1572–4.
16. Wells RG, Miller JH, Sty JR. Scintigraphic patterns in osteoid osteoma and spondylolysis. Clin Nucl Med 1987;12: 39–44. doi:10.1097/00003072-198701000-00010.
17. Ilyas I, Younge DA. Medical management of osteoid osteoma. Can J Surg 2002;45:435–7.
18. Jayakumar P, Harish S, Nnadi C, Noordeen H, Saifuddin A. Symptomatic resolution of spinal osteoid osteoma with conservative management: imaging correlation. Skeletal Radiol 2007;36 Suppl 1:S72-6. doi:10.1007/s00256-006-0198-x.
19. Ozaki T, Liljenqvist U, Hillmann A, Halm H, Lindner N, Gosheger G, et al. Osteoid osteoma and osteoblastoma of the spine: experiences with 22 patients. Clin Orthop Relat Res 2002;397:394–402. doi:10.1097/00003086-200204000-00046.
20. Swee RG, McLeod RA, Beabout JW. Osteoid osteoma. Detection, diagnosis, and localization: Detection, Diagnosis, and Localization. Radiology 1979;130:117–23. doi: 10.1148/130.1.117.

Correspondence to:

Dr. **Zied Mansi**
Department of Orthopaedic Surgery
IBN EL JAZZAR KAIROUAN University Hospital
3100 Kairouan, Tunisia
E-mail: doc.zm@hotmail.fr

SAŽETAK

Osteoidni osteom kralježnice i skolioza: prikaz 5 slučajeva

Zied Jlalia, Zied Mansi, Guermazi Bassem, Arfa Wajdi, Sami Bahroun, Atef Ltifi, Hedhili Guedhami, Abderrahmen Souid, Slah Soui, Zaidi Bacem, Samir Daghfous

Uvod: Skolioza, bočna zakrivljenost kralježnice, uobičajeno je asimptomatska kod djece, ali može biti povezana s boli kod odraslih. Ova studija istražuje pet slučajeva bolne skolioze uzrokovane osteoidnim osteomom (OO), benignom lezijom koja se često pogrešno dijagnosticira zbog svoje rijetkosti i suptilne prezentacije. Cilj je bio ispitati dijagnostičke izazove i ishode liječenja u slučajevima spinalnog OO koji se prezentira sa skoliozom.

Metode: Retrospektivna studija provedena je na pet pacijenata liječenih između 1986. i 2016. godine, koji su se prezentirali s bolnom skoliozom pripisanom OO. Za dijagnozu su korištena radiografska snimanja, uključujući kompjuteriziranu tomografiju (CT) u četiri slučaja i magnetsku rezonancu (MRI) u jednom slučaju. Svi pacijenti podvrgnuti su kirurškoj resekciji OO stražnjim pristupom s postoperativnom ortozom. Prosječno razdoblje praćenja bilo je 5 godina i 5 mjeseci.

Rezultati: Prosječna dob pacijenata bila je 12,2 godine. Vrijeme do dijagnoze u prosjeku je bilo 13 mjeseci. Skolioza je bila prisutna u svim slučajevima, s dva slučaja koja su pokazivala dorzo-lumbalnu, a tri lumbalnu zakrivljenost. CT snimke su identificirale OO kod svih pacijenata. Mjesta lezije uključivala su pedikul (2 slučaja: L3, L4), zglobne fasete (1 slučaj: L4), stražnji hemi-luk (1 slučaj: D12) i laminu (1 slučaj: D10). Kirurška resekcija rezultirala je spontanom korekcijom skolioze kod tri pacijenta u prosjeku od sedam mjeseci, dok je jednom pacijentu bila potrebna dodatna korektivna operacija zbog teške skolioze (57°) dijagnosticirane nakon 24 mjeseca. Spondilodeza s autolognim koštanim transplantatom provedena je u dva slučaja kako bi se osigurala stabilnost, a jednom pacijentu je bila potrebna revizijska operacija zbog resekcije na pogrešnoj razini kralješka. Postoperativna ortoza korištena je tri mjeseca kod četiri pacijenta. Histološka analiza potvrdila je dijagnozu OO u svim slučajevima. Ishodi su bili povoljni kod četiri pacijenta, s povlačenjem boli i poboljšanom funkcijom. Jedan slučaj nezarastanja grafta dogodio se nakon revizijske operacije; međutim, pacijent je ostao zadovoljan nakon 10 godina praćenja. Rezultati naše serije sažeti su u Tablici 1.

Zaključak: Spinalna OO treba se uzeti u obzir u slučajevima bolne skolioze, posebno kod djece. Rana dijagnoza putem CT snimanja ključna je za učinkovito liječenje. Kirurška resekcija pruža dobre ishode, a pravovremena intervencija sprječava razvoj strukturne skolioze.

Ključne riječi: KRALJEŽNICA; KOŠTANE NEOPLAZME; OSTEOM, OSTEOIDNI; MAGNETSKA REZONANCIJA