Osteoid Osteoma of the Spine is an Important Cause of Back Pain: Two Cases and Review

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CASE REPORT

Abstract:
Two cases are reported and discussed of osteoid osteoma in the spines of two children, presenting as nocturnal low back pain irrelevant to any position or activity. Plain radiographs of the lumbo-sacral spine, Technetium-99 bone scan and computerized tomography (CT) were used to locate and verify the lesions. Both were then excised by open surgery and confirmed by a post-operative CT scan. A review of all the known and updated modalities of diagnosis and treatment is presented and it is emphasized that this uncommon lesion should be considered in the differential diagnosis of back pain in children.

Key words: Osteoid osteoma, Spine, Back pain

Introduction:
Primary bone tumors of the spine are relatively infrequent lesions compared with metastatic diseases, multiple myeloma and lymphoma which are more frequent and are usually multifocal lesions posing few problems of diagnosis(1). Osteoid osteoma is one of the most common benign tumors of the spine and should be included in the differential diagnosis of any young patient with pain in the back, and it is also the most common cause of painful scoliosis in adolescents(1-3). It is a benign bone lesion with a nidus of less than two centimeters surrounded by a zone of reactive bone and it accounts for approximately 10% of all benign tumors, 13% percent of which occur in the spine.

Case one:
A seven-year-old girl complained of recurrent attacks of pain in the lower back, right buttock and posterior aspect of the right thigh for about four months prior to admission. The pain was almost constant at night but could be relieved by a mild analgesic (acetaminophen). Clinical examination showed no local tenderness over the lower back and buttck; the straight-leg raising test and gait were normal; there were no neurological deficits or scoliosis. Blood investigations were normal.

Plain radiographs of the lumbo-sacral spine showed widening in the right hemilamina of L5 vertebra in the antero-posterior view, but no scoliosis (Figure 1-A). Bone isotope (Technetium-99) scan showed a small focal active lesion confined to the right side of L5 vertebra (Figure 1-B). The computerized tomographic (CT) scan identified a bone defect with a central nidus measuring about 7mm in diameter involving the right hemilamina of L5, suggesting osteoid osteoma (Figure 1-C).

The vertebra was explored through a midline incision in the prone position and the lesion was identified by an obvious local swelling overlying the nidus, which was unroofed and the central material was curetted and sent for histopathology. The lesion was found to be connected to the facet joint.

A post-operative CT scan showed complete resection of the lesion (Figure 1-D). The histopathological report confirmed the diagnosis of osteoid osteoma. The symptoms had disappeared completely in the immediate post-operative period and the child had a smooth recovery with no recurrence of symptoms for three years.

Figure 1-A:
Plain radiograph showing widening of right hemilamina of L5 vertebra.
Case two:

An eight-year-old boy complained of long-standing non-radiating low back pain that was worse at night but also could be relieved by a mild analgesic (acetaminophen). During the attacks he could hardly bend his back and walked with an obvious stiffness with an inclination to the right side. On clinical examination his active spinal movements were restricted; passive straight-leg raising on the right was also limited; no neurological deficits were found. Blood investigations were not significant.

Plain radiographs of the lumbo-sacral spine showed a translucent swelling with a central calcification in the right hemilamina of S1 vertebra, with no scoliosis (Figure 2-A). Technitium-99 scintigraphy of this possibly doubtful lesion showed a well-circumscribed intense rounded focal uptake in the right side of S1 vertebra (Figure 2-B).
A CT scan matched the lesion seen in the plain X-rays and showed focal bony destruction in the right hemilamina of S1 with spotty calcification in a central mass of soft tissue suggestive of osteoid osteoma (Figure 2-C).

The spine was explored by a midline incision in the prone position and the lesion was easily identified by obvious local swelling. It was unroofed first to visualize the nidus and, as the remaining shell was thin, the whole lesion was excised with a wide zone of healthy bone. A post-operative CT scan confirmed full excision (Figure 2-D), while the histopathological result confirmed the diagnosis. The child had an uneventful recovery and the symptoms disappeared completely in the early post-operative period with no recurrence recorded for two years.

Discussion:

Osteoid osteoma is a benign osteoblastic neoplasm most often seen in young males, frequently in the first three decades of life and may be found in the cortical or cancellous bone with a further five per cent reported as subperiosteal, and multicentric foci were also reported. When the lesion is periarticular or eroding the articular surface, as in the first case, symptoms will be related to the hypertrophic degenerative arthritis resulting from the concomitant pathologic changes in the joint surfaces and synovial tissues. This explains the picture of sciatica in our patient, in addition to the bony swelling caused by the tumour itself.

Preliminary diagnosis carries some difficulties, mostly due to unawareness of the condition, leading to delay in the management. The diagnosis of persistent low back pain in children and adolescents may require various procedures to reach a final result, but the clinical features and awareness of the presence of this lesion will give a better hint and direction to the goal. The first imaging procedure is usually the plain radiography, which shows different pictures depending on the location of the lesion (whether medullary or cortical) and the degree of reactive sclerosis surrounding the nidus. Typically it appears as a radiolucent round or oval nidus with a dense sclerotic reactive zone, particularly in the diaphyseal cortical bone, but this zone may be absent or the nidus may be opacified by calcification or ossification, while lesions in the pelvis and spine are usually difficult to identify by plain radiographs. The most important plain radiographic differential diagnosis includes osteoblastoma, Brodies abscess, stress fracture and enostosis.

Although radioisotope imaging is not pathognomonic for osteoid osteoma, it can be very useful in diagnosis and anatomic location, intra-operative and post-operative identification. The sign of “double density image” is usually created, which is fairly typical of osteoid osteoma. Occasional negative bone scans have been reported.

Computerized tomography is considered by many authors to be the imaging method of choice in demonstrating the nidus, particularly when extensive sclerosis is obstructing it, or when the tumour is localized in the pelvis or spine, as in our cases. We also believe that CT is essential for optimal planning before surgery is performed and for postoperative confirmation of complete resection. Magnetic imaging resonance although sometimes used, is considered less specific in the diagnosis.

Treatment can be surgical or conservative. To achieve a surgical cure the entire nidus must be removed completely; this can be achieved either by open methods or minimally invasive techniques. En block resection can be performed much more easily in the diaphysis of the long bones in which the intraoperative localization of the nidus, if needed, can be achieved ei-
ther by radioisotopes or fluorescence techniques\(^5\textendash}^9\). **Open intralesional resection** of the nidus by unroofing the lesion is another method that is more helpful in regions where wide resection may endanger the bone strength itself or the surrounding structures, as in the pelvis, proximal femur, long bones diaphysis, and the spine\(^6\textendash}^7\textendash}^9\). Campanacci et al\(^9\) claimed 100% primary cure using both these methods. Others reported recurrence even after en bloc resection\(^1\textendash}^6\). Removal of the nidus by **minimally invasive** techniques is becoming more widespread although, due to incomplete removal, this may be less successful at curing the pain and preventing recurrence\(^7\textendash}^9\). CT guidance is used, while the extraction is done percutaneously either by intraleisonal excision\(^5\textendash}^7\textendash}^11\textendash}^17\), by thermocoagulation (radiofrequency thermal ablation)\(^18\textendash}^19\), by laser photocoagulation\(^20\) or by ethanol injection\(^9\). Donahue et al\(^22\) and Katz et al\(^14\) claimed a 100% cure rate with the percutaneous CT-guided excision, while Sans et al\(^15\) claimed about 84% cure rate. Barei et al\(^17\) using the percutaneous CT-guided thermal ablation had a success rate in 10 out of 11 in his series, but they advised its use only on extra spinal lesions that are not immediately adjacent to neurovascular structures, while Cove et al\(^18\) used it successfully in two cases with lumbar spinal lesions. Campanacci et al\(^9\) found 83% primary cure in a collection of 247 cases reviewed in the literature, in which the percutaneous technique of nidus removal had been performed either by excision or coagulation.

Medical management of osteoid ostemata is well recognized and is usually needed whenever the lesion is inaccessible even with the most carefully planned surgical exposures, or is adjacent to a vital neurovascular structures, or when the patient refuses any operative interference. Salicylates or other non-steroidal anti-inflammatory drugs can relieve the pain and achieve a cure if used for a long period approaching an average of thirty-three months\(^5\textendash}^7\textendash}^21\textendash}^22\). Intolerance to such drugs will over-rule this management.

In conclusion, we believe that any child or adolescent complaining of back pain, particularly long-standing, should be investigated thoroughly, keeping in mind the possibility of osteoid osteoma and that a plain radiograph may not be significant in detecting these lesions. It is obvious that multiple means of excision are within the reach of surgeons to be used accordingly in different sites of the body.

**References:**