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CASE REPORT

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Osteoid osteoma of the lateral mass of C5. Should excision be combined with fusion?

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Abstract A 10-year-old girl presented with a 1-year history of pain and stiffness in her neck associated with left shoulder and arm pain. This was found to be caused by an osteoid osteoma of the lateral mass of C5. Surgical excision of the tumour was performed through a posterior approach. Following surgery, the patient's pre-operative pain resolved. However, 3 months later she developed a recurrence of neck pain secondary to cervical instability. Further investigation revealed a grade II spondylolisthesis at the C5/6 level. A combined anterior and posterior fusion was performed and the patient's instability pain rapidly resolved. At her 18-months' follow-up a solid fusion was confirmed radiologically and the patient remained asymptomatic with no evidence of tumour recurrence. The purpose of this report is to highlight the difficulty in diagnosing this condition as well as to emphasise the surgical technique required. It would appear that excision of the lateral mass will result in instability. To prevent this, fusion of the spine should always be considered at the time of surgical excision of the tumour.

Key words Cervical spine · Osteoid osteoma · Spinal stabilisation

Introduction

Prior to 1954, vertebral osteoid osteoma was reported in the Italian literature. MacLellan and Wilson reviewed the literature in 1967 and found 36 cases of osteoid osteoma of the spine [7]. In 1975, Keim and Regina reviewed 36 cases of osteoid osteoma, 9 of which were in the spine [5]. All were located in the posterior elements. In 1992, Levine et al. reviewed 40 cases of benign tumours of the cervical spine, 18 of which were osteoid osteoma [6]. All were situated in the posterior elements.

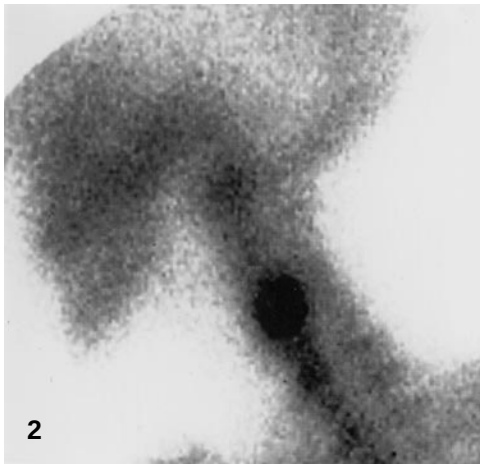
We report a case of an osteoid osteoma of the left lateral mass of the fifth cervical vertebra in a 10-year-old girl. This case highlights the importance of fusing the spine at the time of tumour excision.

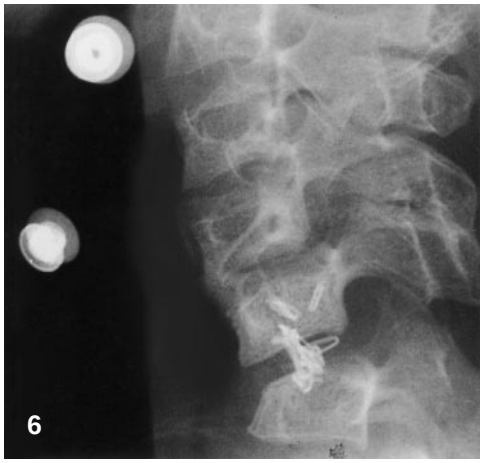
Case report

A 10-year-old girl presented with a 1-year history of neck pain and left C6 radicular pain. There was no history of trauma. On examination, an area of tenderness over the lower cervical spine was elicited on deep palpation posteriorly. Movement of the spine was limited at the extremes in all directions. However, rotation was greater on the right side than the left. Sensory and motor examination of the limbs revealed no abnormalities.

A plain radiograph of the cervical spine was reported as normal on the first presentation. A subsequent radiograph of the cervical spine performed a few months later revealed sclerosis of the left lateral mass of C5 (Fig. 1). A corresponding area of increased uptake of radionuclide in C5 was also noted on bone scan (Fig. 2). A computerised tomographic scan of the relevant cervical region revealed a lucent zone with a central nidus surrounded by sclerosis in the left articular mass of C5 extending into the lamina (Fig. 3). An MRI scan showed a predominantly low signal on T1- and a high signal on T2-weighted images in relation to the body of C5 (Fig. 4).

The findings of systems review and laboratory investigations were normal. The patient underwent excision of the lesion via a





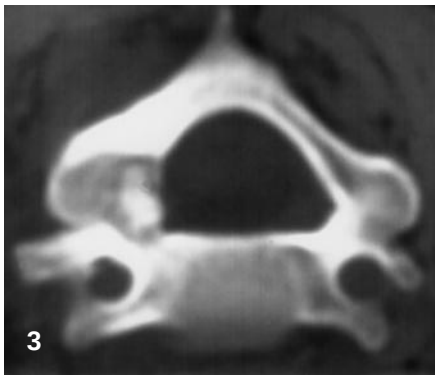


Fig. 1 Pre-operative lateral cervical radiograph showing sclerosis in the lateral mass of C5

Fig. 2 Technetium bone scan revealing increased uptake in the lateral mass of C5

Fig. 3 Computed tomography of the cervical spine showing a lucent area with a central nidus surrounded by a zone of sclerosis involving the lateral mass of the C5 vertebra

Fig. 4 T-2 weighted MR image of the cervical spine, showing the lesion in C5 with increased signal intensity

Fig. 5 Delicate osteoid trabeculae separated by fibrovascular tissue and lined by osteoblasts, typical of osteoid osteoma (H&E; ×80)

Fig. 6 Lateral radiograph of the cervical spine taken 3 months after surgery showing a kyphotic deformity at the C5/6 level

Fig. 7 Radiograph of the cervical spine taken 18 months after combined anterior and posterior fusion, showing a solid fusion

posterior approach. The posterior elements of C5 were exposed. A left C5 hemilaminectomy combined with excision of the lateral mass was performed. The left inferior facet at C5 was removed with part of the superior lamina of C6. The vertebral artery was exposed. The vertebral artery was accidentally injured during the procedure. The vessel was clipped to control the bleeding. There were no changes on cord monitoring intra-operatively.

The patient made an uneventful post-operative recovery without any neural or vascular complications.

Fusion at the time of excision of the tumour was not contemplated at the index operation in view of the age of the patient. It was felt that if fusion was performed posteriorly there would be further growth anteriorly leading to increased cervical lordosis.

The patient had complete resolution of her neck pain following excision of the tumour. Histopathological examination of the lesion confirmed it to be an osteoid osteoma (Fig. 5).

Three months later the patient presented with symptoms consistent with cervical instability. Radiological examination of the cervical spine revealed a grade II spondylolisthesis of C5 on C6 with a kyphotic angle measuring 30° (Fig. 6). These symptoms were not felt to be caused by recurrence of the tumour, because of the difference in the character of the pain compared with her previous history and also no recurrence was evident on the radiological findings.

A combined anterior and posterior fusion was undertaken using a tricortical iliac crest strut graft anteriorly and sublaminar wire stabilisation posteriorly. This procedure was carried out with intraoperative somatosensory monitoring of the spinal cord. No complications arose from the second procedure. At a follow-up examination 18 months after the operation the patient remained asymptomatic (Fig. 7).

Discussion

Osteoid osteomata constitute 2.5% of all bone tumours and 10% of all benign lesions of bone. Approximately 10% of osteoid osteomata occur in the spine, most commonly in the lumbar spine (56%). Of these, 20% occur in the articular process. Less commonly they occur in the transverse process, spinous process and vertebral body. There is a male predominance, with the male-to-female ratio being 2 : 1 [1]. Following the lumbar spine, the next most common site of occurrence of osteoid osteomata is the cervical spine. Here too, the majority are situated in the posterior elements. However, rarely they may involve the vertebral body [9].

Osteoid osteomata of the spine are difficult to diagnose. The average delay in diagnosis is approximately 18–24 months [6]. Because of their small size osteoid osteomata of the cervical spine are generally not visible on plain radiographs. MRI scans can be misleading [4]. However, there has never been a false-negative bone scan of an osteoid osteoma reported in the literature [8].

The plain radiograph of our case was reported as normal and the diagnosis was only made almost 1 year from the onset of symptoms. The diagnosis was established with the aid of a radionuclide scan, CT scan and MRI. Excisional biopsy of the lesion confirmed the diagnosis.

In patients with osteoid osteoma, it is important to excise the complete nidus. This results in immediate relief of pain, allows histological confirmation of the diagnosis and affords an early return to full spinal mobility to avoid structural deformity in the skeletally immature spine [2].

Benign tumours of the cervical spine present a number of problems peculiar to this region. An osteoid osteoma involving the posterior elements of the cervical spine may be treated adequately with excision biopsy by curettage. An osteoid osteoma of the vertebral body, on the other hand, is best treated by anterior excision and fusion [6]. Excision or curettage of an osteoid osteoma involving the lateral mass may be followed by segmental instability, as the current case demonstrates. A combined anterior and posterior fusion therefore needs to be considered at the time of the initial operation. This ultimately depends on the amount of bone and soft tissue resected to allow access to the tumour. The preservation of the sagittal contour of the cervical spine depends on the integrity of the posterior and anterior elements. When there is extensive loss of the posterior elements a progressive kyphos may develop, particularly if the facet joint or its capsule are disrupted. As this develops, the posterior column is placed under tension and the anterior column is placed under compression. The incidence of late deformity may be reduced by an appropriate posterior fusion [3]. This may be insufficient if the lateral mass is removed to afford complete excision of the tumour as demonstrated by the above case. In this situation a posterior fusion may need to be supplemented with an anterior fusion to avoid the development of cervical hyperlordosis in the immature spine. The vertebral artery is susceptible to injury during lateral mass excision; great care and experience is required to avoid this complication.

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