CASE REPORT

Rare cause of neck pain: tumours of the posterior elements of the cervical spine

Yoshihiro Katsuura, Garrick Cason, James Osborn

University of Tennessee Health
Science Center College of
Medicine, Chattanooga,
Tennessee, USA

Correspondence to
Dr Yoshihiro Katsuura,
yoshikatsuura@gmail.com

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SUMMARY

Here we present two cases of primary bone tumours of the cervical spine in patients who had persistent neck pain—in one case, lasting 8 years. In each case, there was a delay in diagnosis and referral to a spine specialist was prolonged. Primary bone tumours of the spine are rare, which is in contrast to the wide prevalence of cervical neck pain. Many primary care providers may go an entire career without encountering a symptomatic primary cervical spine tumour. In this paper, we discuss the clinical course and treatment of each patient and review the current literature on primary bone tumours of the spine. Owing to the subtle roentgenographic findings of primary cervical tumours, we highlight the importance of advanced imaging in the clinical work-up of simple axial neck pain lasting >6 weeks to avoid misdiagnosis of serious pathology.

BACKGROUND

The cervical spine is the least common location for a primary bone tumour of any variety.^{1 2} This is in stark contrast with the high rate of neck pain in adults.³ Unfortunately, because the most common presenting symptom for tumours of the cervical spine is simple axial neck pain, they go undiagnosed for months to years.^{4 5} Neurological symptoms are rare with benign tumours of the cervical spine but can result from encroachment into the spinal canal or neuroforamina.^{4 5} Here we present two varieties of cervical spine tumours in adults to highlight an uncommon and easily missed cause of neck pain. In both cases, neck pain was the primary symptom and was misdiagnosed initially as simple cervicgia despite prolonged symptomatology.

CASE PRESENTATION**Case 1**

A 54-year-old woman was referred from her primary care provider (PCP) with an 8-year history of lower posterior cervical pain and left upper extremity numbness and tingling. She described her pain as a dull ache, exacerbated by any motion of the cervical spine. She was otherwise healthy with no significant medical or surgical history. Her pain had been continually growing worse, and she had failed attempts at conservative management by her PCP. On examination, she was noted to have hyperreflexia in her left biceps and a positive Hoffmann's sign on the right, but an otherwise normal neurological examination.

Initial plain film X-rays showed a subtle radiolucency in the C5 spinous process as well as a

Figure 1 (A) Lateral preoperative radiograph demonstrating mixed radiopaque (blue arrow) and lucent lesion (white arrow) at C5; (B) AP radiograph and (C) postoperative lateral plain film.

radiodense lesion in the spinal canal ([figure 1](#)). CT imaging revealed a lesion with high attenuation originating on the anterior aspect of the lamina of C5, protruding against the ligamentum flavum into the spinal canal. The lesion had a smooth hemispheric appearance of cortical bone consistent with a radiographic diagnosis of osteoma ([figure 2](#)). Subsequent MRI demonstrated the lesion was of low signal on T2-weighted imaging. The lesion itself was 8 mm in diameter and compressed the spinal cord at this level, leaving a remaining 8 mm space available for the cord ([figure 3](#)).

Case 2

A 63-year-old woman presented with a 5-month history of neck, low lumbar and right lower extremity pain. She described her axial pain as a constant ache, while her extremity pain was sharp and radiating. On further questioning, she endorsed progressive difficulty with balance, navigating uneven surfaces, and to using a handrail whenever possible. She also reported increasing clumsiness with her upper extremity, manifested by frequently dropping objects. Before being referred, she had received non-steroidal anti-inflammatory pain medication from her PCPs, which failed to provide relief.

On examination, she was found to be hyperreflexic in her upper extremities with 3+ biceps and

Figure 2 CT scan showing sclerotic lesion (blue arrow).

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Figure 3 (A) T2-weighted MRI showing hypointense lesion at C5 (white arrow); (B) T1-weighted MRI; (C) Short tau inversion recovery (STIR) sequence MRI and (D) T1 post-contrast.

brachioradialis reflexes bilaterally, as well as positive Hoffmann's signs bilaterally. Of note, the patient had a history of metallic fragments from a gunshot to the face and thus could not undergo MRI. Plain films of her cervical spine showed significant kyphotic angulation and diffuse degeneration. There was a minor radiolucent area noted on the spinous process of C6 (figure 4). CT myelogram was subsequently performed demonstrating a relatively radiolucent mass with a homogenous pattern of osteoid. The lesion measured ~19 mm in diameter and had the poor mineralisation consistent with a radiographic diagnosis of osteoblastoma. The lesion was located in the posterior elements of C6 and was compressing the spinal cord (figure 5). Given the radiolucent appearance of the lesion, the patient underwent a tumour work-up for metastatic disease, which was negative.

INVESTIGATIONS

Case 1 (patient 1)

1. Plain films of cervical spine: anterior-posterior, lateral, flexion and extension views.
2. CT scan (useful for determining bony architecture).
3. MRI of cervical spine with contrast.
4. Preoperative laboratory testing: complete blood count and differential; comprehensive metabolic panels preoperatively (found to be normal).

Case 2 (patient 2)

1. Plain films of cervical spine: anterior-posterior, lateral, flexion and extension views.
2. CT myelogram of cervical spine (the patient had a contra-indication to MRI imaging).
3. Tumour work-up: CT thorax, abdomen and pelvis; complete blood count and differential; comprehensive metabolic

Figure 4 Gross pathology. The specimen was composed of dense sclerotic bone attached to the lamina consistent with an osteoma.

Figure 5 (A) PA film showing left sided listing. (B) Lateral plain film showing radiolucent lesion in the C6 spinous process (blue arrow).

panel; serum and urine protein electrophoresis (all were negative for systemic disease).

DIFFERENTIAL DIAGNOSIS

Relevant causes of local mechanical cervical pain and neuropathic pain

1. Spinal causes
 - A. Cervicalgia/cervical strain (ie, non-specific muscular strain).
 - B. Cervical spondylosis.
 - C. Cervical discogenic pain.
 - D. Cervical facet degeneration.
 - E. Myofascial pain.
 - F. Infection: epidural abscess, osteomyelitis, diskitis, tuberculosis.
 - G. Systemic illness: rheumatoid arthritis, ankylosing spondylitis, diabetic neuropathy.
 - H. Malignancy: metastatic disease, myeloma, lymphoma.
 - I. Primary tumour: osteoid osteoma, osteoblastoma, osteoma, aneurismal bone cyst.
2. Non-spinal causes
 - A. Irritation of peripheral nerve: brachial plexus/thoracic outlet syndrome.
 - B. Myocardial infarction.
 - C. Referred pain from shoulder: rotator cuff pathology.

TREATMENT

Patient 1

Non-operative care was discussed with the patient; however, considering her myelopathic presentation, surgery was strongly advised. Given the location of the lesion, a posterior approach to the cervical spine was selected. Lateral mass screws were then placed under fluoroscopic guidance from C4 to C6. An en bloc resection of the tumour at C5 via laminectomy was performed using a Kerrison rongeur. The specimen consisted grossly of a hemisphere of dense sclerotic bone consistent with an osteoma (figure 6). The case was completed with decortication of the remaining posterior elements. The wound was thoroughly irrigated and a combination of crushed local autograft, demineralised bone matrix and cancellous allograft was packed into the lateral gutters. The incision was closed in layers and a superficial drain placed. The material was sent to pathology whose report was consistent with osteoma.

Figure 6 (A) Sagittal CT myelogram showing hypoattenuation at lesion of posterior elements consistent with osteoblastoma (blue arrow). (B) Axial CT myelogram showing destructive lesion at posterior elements of spine (white arrow).

Patient 2

Again, in this case there was no role for conservative therapy and surgical treatment was selected. Considering the significant degeneration throughout the cervical spine, a dual approach operation was planned for this patient. The first stage was a multilevel C3–C7 anterior cervical discectomy and fusion (ACDF) to decompress the anterior cord. The second, performed 2 days subsequent, was a C3–C7 posterior spinal instrumentation and fusion (PSIF) with laminectomy for excision of the lytic mass.

For the anterior procedure, the patient was placed supine in a Mayfield headrest in 20 lbs of mandibular traction. A standard anterior Smith-Robinson approach to the cervical spine was performed with discectomies from C3 to C7. A 68 mm anterior plate was then selected and fixed to the cervical spine with screws. The patient recovered uneventfully in the intensive care unit (ICU).

Two days following the index procedure, the patient was taken back to the operating room for a posterior approach. A similar operation was performed as in the previous case, with the exception of fusion from C3 to C7. A burr was used to create a trough at the lamina-lateral mass junction, and then a 2 mm Kerrison rongeur was used to carefully complete the laminectomy. A Murphy Ball probe was then used to tease the tumour off the dura for a complete en bloc resection (figure 7). Pathology reported the specimen to contain a thin cortical shell with a central cavity consistent with an osteoblastoma (figure 7).

OUTCOME AND FOLLOW-UP

Both patients were admitted to the ICU following their surgery and transferred to the floor when adequate pain control had been established. Both patients were discharged on the fourth postoperative day and had uneventful recoveries following their hospitalisations. At 1-year follow-up, both patients had complete resolution of their neck pain with improvement in neurological symptoms.

DISCUSSION

The prevalence of non-specific cervical neck pain (cervicalgia) in the general population is 30–50% and is the fourth leading cause of disability in the USA.^{6–8} The differential diagnosis of cervicalgia is diverse but includes common entities such as degenerative disc disease, cervical strain, facet-mediated pain, whiplash, cervical radiculopathy and spondylotic myelopathy.⁹ Most neck pain is self-limiting and resolves within 6 weeks.

Figure 7 Gross pathology. The specimen was composed of a thin-walled cortical shell and central nidus of reactive bone consistent with osteoblastoma.

Cases of subacute or chronic neck pain (lasting >6 weeks) or cases presenting with ‘red flags’ (including constitutional or neurologic symptoms) require immediate investigation with advanced imaging.¹⁰ Tumours of the spine account for only a fraction of neck pain cases and, generally, are due to metastatic disease.¹¹ Primary bone tumours of the spine are exceedingly rare, even more so in the cervical spine, yet must be considered because of their potential to cause neurological decline.^{1 2} Here we present two cases of primary bony tumours of the cervical spine to contribute to the literature surrounding these entities, heighten awareness of them as a potential source of neck pain, and provide direction on diagnosis and treatment.

Primary spinal tumours may be classified as affecting either the anterior (vertebral body) or posterior elements (pedicles, lamina, spinous process). The differential diagnosis of posterior element tumours is unique to this bony area and includes osteoma, osteoid osteoma, osteoblastoma, osteochondroma and aneurismal bone cysts (figure 8).^{4 12} These lesions tend to be much more common in the thoracic and lumbar spine and rarely affect the cervical spine.^{1 2} Tumours of the anterior elements are generally metastatic and are much more common than posterior element tumours.¹¹ Weinstein *et al*¹ identified only 6 cervical lesions in their series of 82 primary tumours of the spine. Moreover, in a survey of the Leeds Regional Bone Tumour Registry, Dreghorn *et al* reported primary spine tumours accounting for only 2.8% of all axial skeleton tumours. In this series of 55 primary bone tumours, only 5 occurred in the cervical spine.² The most common tumours of the posterior elements are generally osteochondroma and osteoid osteoma.^{4 5 13}

Osteomas are benign primary bone tumours which are rare outside of the skull.¹⁴ They consist of compact lamellar bone

Figure 8 Diagram of tumours affecting the posterior elements of the cervical spine: (A) Normal; (B) osteoma, composed of dense sclerotic cortical bone; (C) osteoid osteoma, <2 cm in diameter with a characteristic radiolucent nidus; (D) osteoblastoma has similar radiographic appearance to osteoid osteoma but >2 cm; (E) osteochondroma, exophytic appearance with cortical continuity; and (F) aneurysmal bone cyst, expansile, aggressive lesion with characteristic fluid levels on MRI.

and grow at a very slow rate, presenting with pain but occasionally can result in myelopathy if large enough.¹⁴ Symptomatic osteomas of the cervical spine are rare with only several other documented cases reported in the literature.^{14 15} Radiographically and histologically, they have the appearance of dense cortical bone and may be easily overlooked.¹⁵

Osteoblastoma itself accounts for ~1% of primary bone tumours and occurs in the posterior elements of the lumbar spine 40% of the time.^{16–18} It is an osteoid-producing primary bone tumour that presents with pain and occasional scoliosis.¹⁹ On imaging, they are characteristically lytic in appearance with a size >2 cm differentiating them from osteoid osteoma.^{17 20 21}

The majority of tumours of the cervical spine identified before the age of 20 are benign, while only 30% of tumours occurring in those over 20 years are benign.^{4 5} The most common presenting symptom of a cervical tumour is pain but may also present as neck stiffness or torticollis.^{4 5 13} This pain results from irritation and compression of nearby structures including the ligamentum flavum.^{4 7 8} Some tumours such as osteoblastoma and osteoid osteoma are also capable of releasing prostaglandins that cause local pain.²² Tumours of the cervical spine may also result in spinal cord compression causing the syndrome known as cervical myelopathy, which has a diverse presentation of signs and symptoms.²³ In addition to neck pain, patients may present with subtle difficulties with balance,

diminished dexterity and proprioception. Interestingly, cervical myelopathy may also cause non-dermatomal extremity pain or even low back pain, known as a false-localising sign, as was present in case 2. Although the mechanism of this phenomena has not been fully established, it is proposed to result from a disorder of the spinothalamic tract.^{24 25}

Spinal cord compression tends to be less frequent with benign disease compared to malignant.⁴ Owing to the ubiquitous nature of these symptoms, the diagnosis may be delayed for years.^{4 5} The majority of primary bone tumours are visible on plain radiographs, but diagnosis can usually only be established with CT or other advanced imaging.⁴ More often than not, histopathologic evaluation can be difficult and inconclusive.⁴

Treatment for benign cervical lesions consists of intralesional biopsy and curettage or en bloc excision but obviously must be tailored to the location, pathology and patient.²⁶ Fusion is generally reserved for patients with removal of facets or instability following laminectomy.²⁷

Here, we present two cases of benign bone tumours of the cervical spine in order to highlight them as rare but potential sources of axial neck pain, which is an otherwise very common symptom in the general population.³ Any patient with non-traumatic neck pain greater than the age of 50 should have plain films obtained on presentation.¹⁰ While many of these pathological processes listed are common and do not require advanced imaging, clinicians should pay careful attention to any patient with moderate neck pain lasting more than 6 weeks or patients with neurological signs or symptoms including hyperreflexia. In these patients, advanced imaging such as MRI or CT should be obtained as they can rule out serious pathology and prevent delayed diagnosis.¹⁰

Learning points

- ▶ Primary tumours of the cervical spine are rare but are potential sources of axial neck pain, which is an otherwise common symptom in many cervical diseases.
- ▶ To rule out serious pathology, advanced imaging should be obtained on any patient with persistent neck pain lasting more than 6 weeks or in any patient with neurologic signs or symptoms.
- ▶ The diagnosis of primary bone tumours of the spine is based on characteristic radiographic appearance and can be made with MRI or CT imaging.

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