Case report
Title of paper:
Complex spinal fixation of a cervical vertebra brown tumour: report of an unusual case

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Abstract:
Brown tumours affecting the cervical spine are a rare but recognised complication of renal failure-related secondary hyperparathyroidism. We present a case of a 26 year-old female with radiculopathy who was managed successfully with 360° cervical spine fixation and parathyroidectomy.

Keywords:
Spine; cervical spine; spinal tumour; vertebra

Clinical details

A 26 year old female presented to her local hospital with a subacute history of bilateral but predominantly left-sided C6 radiculopathy. Her medical background was significant for end stage renal failure (ESRF) due to congenital dysplastic kidneys. This was managed originally with a cadaveric transplant 10 years ago which failed 2 years later due to acute cellular rejection. Since then she had been maintained on thrice weekly haemodialysis, but due to longstanding ESRF had developed renal bone mineral disease with secondary hyperparathyroidism. She was poorly compliant with all her medications including calcium supplementation and phosphate binders. At the time of presentation with radiculopathy, neurological examination revealed no motor deficits in the upper or lower limbs. Her blood tests on admission showed corrected calcium 2.41mmol/L, phosphate 0.73mmol/L, ALP 2195iu/L, parathyroid hormone (PTH) 277pmol/L (the latter two results having been chronically significantly raised for several years).

She was transferred to a specialist neurosurgical centre after MR imaging of the cervical spine showed an expansile lesion obliterating most of the body and posterior elements of C_5 vertebra, with compression of the exiting C6 nerve roots and impending compression of the spinal cord. CT of the cervical spine showed near complete erosion of the bony elements of C_5 vertebra (Figure 1).

The patient was managed with 360° decompression and surgical fixation in a two-stage procedure, beginning with a C_5 corpectomy, cage insertion with autologous bone from iliac crest, cement augmentation of C_4 and C6 vertebral bodies followed by plate and screw insertion. One week later she underwent posterior decompression with C_4-6 lateral mass screw fixation (Figure 2). She was kept in a Miami J collar post-operatively when sitting up to >30°, due to poor bone quality. Histopathological analysis of the lesion revealed an osteoclast rich lesion of bone with mononuclear stromal cells showing no atypia, and focal brown staining due to haemosiderin deposition as a consequence of old haemorrhage. In the context of the history of ESRF the histological diagnosis was of a brown tumour.

The 360° decompression and complex spinal fixation was successful in ameliorating the patient's symptoms and she returned to her local hospital for a parathyroidectomy, at which four hyperplastic parathyroid glands were identified in the neck. Following this, PTH levels fell to 3.2pmol/L and ALP to 569iu/L. At the time of her latest clinical review 6 months after fixation, her neurological symptoms had disappeared completely.

Discussion

Brown tumours are uncommon bone lesions. They occur in hyperparathyroidism due to increased osteoclastic activity and aberrant bone turnover, with coexistent hypocalcaemia leading to poor mineralisation of newly formed bone. It is important to note that despite the nomenclature, this is not a neoplastic process but rather a reparative cellular one. Macroscopically, brown discoloration of the lesion is observed due to repeated haemorrhage and haemosiderin deposition. Microscopically, the lesion possesses numerous giant cells with

interstitial hemorrhage, hemosiderin, microfractures, and ingrowth of vascularized fibrous tissue with fibroblasts. Histologically, they are analogous to giant cell tumour of bone (and both are technically osteoclastomas), but in the setting of a raised serum PTH (whether primary, secondary or tertiary), the term brown tumour is applied.

In pathophysiological terms, brown tumours are localised forms of osteitis fibrosa cystica (previously known as von Recklinghausen's disease of bone) a classic manifestation of renal osteodystrophy. The clinical presentation of brown tumours is usually with bone pain, although when located in vertebra they can – as in this case – present acutely due to neurological compromise or vertebral fracture. Radiographic signs of well-defined lytic lesions with thinned cortex and little reactive bone, are commonly seen. The term chronic kidney disease-mineral bone disorder (CKD-MBD) has now replaced renal osteodystrophy, in order to reflect other facets of this variegated pathology, such as adynamic bone disease, osteomalacia, and vascular calcification which leads to excess mortality through cerebro- and cardiovascular disease.

Brown tumours occur in up to 13% of patients with secondary hyperparathyroidism (Fargen et al 2012), with a recent increasing trend in incidence as ESRF survival has improved alongside superior renal replacement techniques. Incidence in primary hyperparathyroidism is estimated at 4.5% (Arsalanizadeh & Westacott, 2013), and in this setting is usually due to a single functioning parathyroid adenoma. Brown tumours more commonly affect the pelvis, sternum, jaw, long bones and ribs. 38 cases have been reported in world literature of brown tumours affecting spinal elements, with no predilection for vertebral bodies or posterior elements; a majority (57%) affect the thoracic spine.

11 cases of cervical spine brown tumours have been reported previously (Ozyurek & Atik, 2015); 8 were in female patients, with a mean age of 36.1 years, and 6 of these were in the context of secondary hyperparathyroidism. Three of these patients underwent some form of cervical spine fixation, but this is the first case reported which describes a comprehensive 360° fixation with good effect. Despite the poor bone quality and due to lack of alternative treatment option for instability and impending cord compression, the 360° spinal fixation with cement augmentation provided the best solution in this complex spinal and clinical case.

The ultimate treatment of brown tumours is underpinned by successful management of secondary hyperparathyroidism. In this case, patient adherence to medical treatments was suboptimal and therefore a parathyroidectomy was undertaken, resulting in the restoration of biochemical normality, which would promote bone remineralisation and prevent further tumour development (Arsalanizadeh & Westacott, 2013). The involvement of neurosurgeons in the management of brown tumours is reserved for cases where neurological deterioration has occurred, or is impending. In such cases, timely fixation should be as comprehensive as possible.

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Informed Consent
Informed consent was obtained from the participant included in the study.

Conflicts of Interest
The authors disclose no conflicts of interest in submitting this manuscript.

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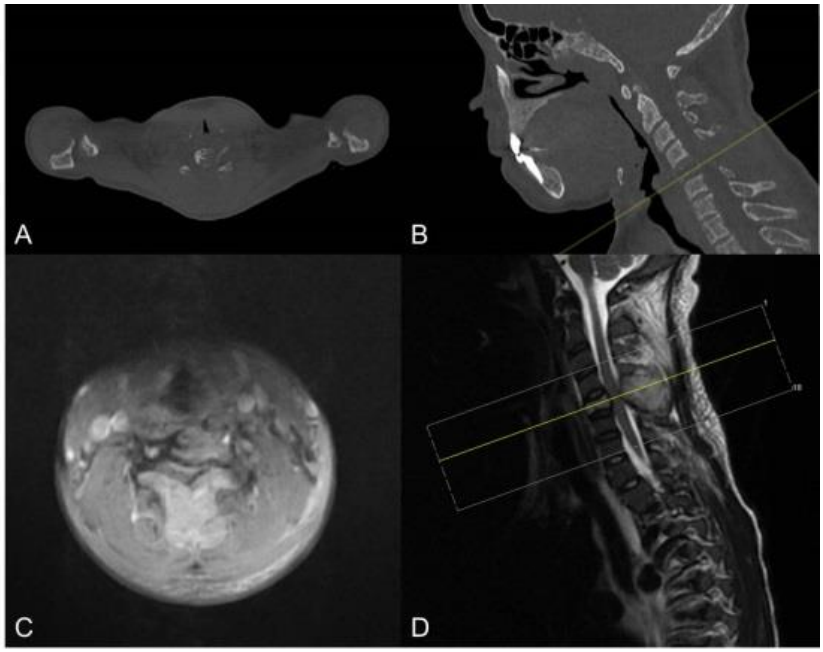

Figure 1. Pre-operative imaging. A, C5 axial and B, mid-sagittal slice of bone-windowed CT scan taken at presentation. C, C5 axial and B, mid-sagittal slice of T2-weighted MR scan taken at presentation.

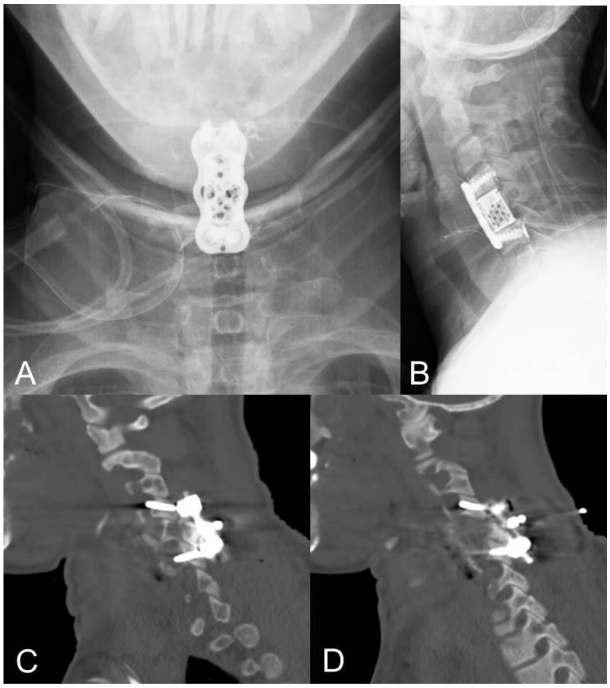

Figure 2. Post-operative imaging. A, anteroposterior and B, lateral plain radiographs following stage 1 procedure (C_5 corpectomy, cage insertion with autologous bone from iliac crest, cement augmentation of C_4 and C6 vertebral bodies and plate and screw insertion). C, right parasagittal and D, left parasagittal slices of CT scan following stage 2 procedure (posterior decompression with C_4-6 lateral mass screws).