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# Osteoclastomas ('brown tumours') and spinal cord compression: a review

Bahareh Arsalanizadeh[1] and Rachel Westacott[2]

[1]Peterborough and Stamford Hospitals, NHS Foundation Trust, Peterboroug, UK and [2]John Walls Renal Unit, University Hospitals of Leicester, Leicester General Hospital, Leicester, UK

*Correspondence and offprint requests to*: Bahareh Arsalanizadeh; E-mail: bahar.arsalani@gmail.com

**Abstract**
Brown tumours are an uncommon manifestation of primary and secondary hyperparathyroidism. There are numerous case reports of brown tumours arising in various parts of the skeleton. They can therefore present a wide range of clinical manifestations. A recent case highlighted the need for heightened awareness of the diagnosis and prompted a literature review.

**Keywords:** brown tumour; secondary hyperparathyroidism; spinal cord compression

## Introduction

Many patients on long-term dialysis develop secondary hyperparathyroidism. Brown tumours, an unusual but recognized complication of both primary and secondary hyperparathyroidism, have been reported to occur in 4.5% of patients with primary and 1.5–1.7% of those with secondary disease [1].

## The case

A frail 57-year-old male haemodialysis patient with known severe peripheral vascular disease presented to the vascular surgeons with a 4-week history of worsening leg weakness. A magnetic resonance imaging (MRI) scan of the spine (Figure 1) showed multiple lesions, the largest of which was at T12, causing cord compression. He was subsequently transferred to Oncology and was initiated on steroids and radiotherapy. Subsequently, a bone biopsy was performed. This showed portions of bone containing a cellular spindle cell proliferation with abundant brown, granular and globular material and scattered groups of multinucleate giant cells of the osteoclast type, which, in the context of high parathyroid hormone levels, was diagnosed as a brown tumour of hyperparathyroidism. Despite significant cord compression, our patient did not undergo surgical decompression due to significant comorbidities. He was initially commenced on increased medical therapy for his secondary hyperparathyroidism including cinacalcet. However, after 6 weeks the patient had not responded to treatment, and he therefore underwent a four-gland parathyroidectomy. He regained some but not all muscle power following prolonged physiotherapy.

## Discussion

Brown tumours (or osteoclastomas) are an unusual but recognized complication of both primary and secondary hyperparathyroidism and have been reported to occur in 4.5% of patients with primary and 1.5–1.7% of those with secondary disease [1]. They are composed of multinucleated osteoclasts, stromal cells and matrix [2, 3] and are benign in nature, although they can cause significant morbidity due to secondary effects such as spinal cord compression. The name derives from their vascularity, haemorrhage and consequent haemosiderin deposition, which give these tumours their characteristic colour [2, 3].

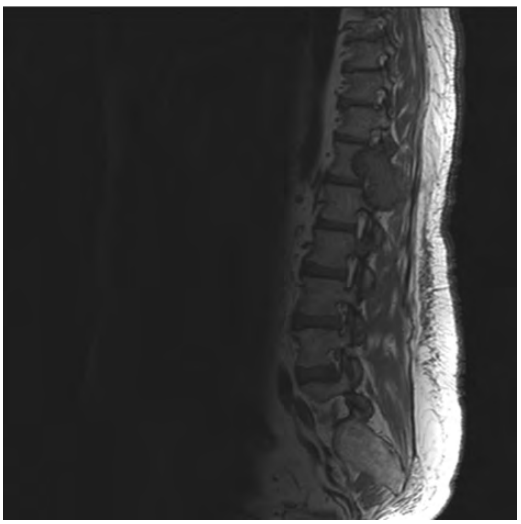

**Fig. 1.** MRI of spine showing the tumour at T12.

**Table 1.** Summary of the spinal brown tumour cases reported in the literature

| | Year published | Sex/age (years) | Hyperparathyroidism | Spine involved | Symptoms | Treatment |
|---|---|---|---|---|---|---|
| Shaw and Davies [11] | 1968 | F/58 | Primary | T10 pedicle | Paraparesis and urinary retention | Surgical resection and parathyroidectomy |
| Shuangshoti *et al*. [12] | 1972 | M/32 | Primary | L4 posterior elements | Progressive paraparesis and reticular pain | Surgical resection and parathyroidectomy |
| Sundaram and Scholz [13] | 1977 | F/63 | Primary | T10 body and pedicle | Paraplegia and urinary retention | Surgical resection and parathyroidectomy |
| Siu *et al*. [14] | 1977 | F/64 | Primary | T10 | Paraplegia, sensory loss, urinary retention | Surgical resection, Parathyroidectomy |
| Ericsson *et al*. [9] | 1978 | F/47 | Secondary | Cervico-thoracic | Paresis | Surgical resection and parathyroidectomy |
| Ganesh *et al*. [15] | 1981 | M/40 | Primary | T2 body and pedicle | Paraparesis, radicular pain | Parathyroidectomy |
| Bohlman *et al*. [7] | 1986 | F/69 | Secondary | Thoracic | Incipient Paraplagia | Steroid therapy |
| Yokota *et al*. [16] | 1989 | F/58 | Primary | T5 pedicle | Paraparesis and numbness | Surgical resection and Parathyroidectomy |
| Pumar *et al*. [17] | 1990 | F/24 | Secondary | Thoracic | Incipient Paraplagia | Surgical resection |
| Kashkari *et al*. [18] | 1990 | F/51 | Primary | T6 and T7 bodies | Paraparesis | Surgical resection and parathyroidectomy |
| Barlow and Archer [19] | 1993 | F/31 | Secondary | Cervical | Neck pain and cervicobrachial neuralgia | Parathyroidectomy and Minerva jacket |
| Sarda *et al*. [20] | 1993 | F/23 | Primary | T3-4 | Paraparesis and radicular pain | Surgical resection and parathyroidectomy |
| Motateanu *et al*. [21] | 1994 | M/57 | Primary | L4-5 facet | Lower limb radicular symptoms | Surgical resection |
| Mourelatus *et al*. [22] | 1998 | M/48 | Secondary | T2 body and posterior elements | Paraparesis and incontinence | Not reported |
| Fineman *et al*. [23] | 1999 | F/37 | Secondary | Thoracic | Incipient oaraplagia | Surgical resection and parathyroidectomy |
| Azria *et al*. [24] | 2000 | F/40 | Secondary | Thoracic | Back pain | parathyroidectomy |
| Masutani *et al*. [25] | 2001 | F/39 | Secondary | Thoracic | Paraplegia | Surgical resection and parathyroidectomy |
| Paderni *et al*. [26] | 2003 | F/45 | Secondary | L2-L3,L5,S1 | Paraparesis | Surgical resection and parathyroidectomy |
| Mustonen *et al*. [27] | 2004 | M/28 | Primary | L2 | Lower limb radicular pain and numbness | Parathyroidectomy |
| Vandenbussche *et al*. [5] | 2004 | F/34 | secondary | Thoracic | Spinal cord compression | Decompression and parathyroidectomy |
| Tarass *et al*. [6] | 2006 | M/42 | secondary | Sacral | Cauda equina syndrome | Surgical decompression and parathyroidectomy |
| Haddad *et al*. [28] | 2007 | F/62 | Primary | T2-4 | Paraparesis | Surgical resection and parathyroidectomy |
| Kaya *et al*. [29] | 2007 | M/72 | Secondary | T1 body and transverese process | Unilateral arm pain and paresis | Radical excision |
| Khalil *et al*. [30] | 2007 | M/69 | Primary | L2 body and pedicle | Lower limb radicular pain | Surgical resection |
| Wiebe *et al*. [3] | 2008 | F/33 | Secondary | Thoracic | Paraparesis | Surgical decompression and parathyroidectomy |
| Hoshi *et al*. [31] | 2008 | F/23 | Primary | Sacrum | Lower limb radicular pain | Parathyroidectomy |
| Ren *et al*. [32] | 2008 | M/47 | Secondary | T4 body and pedicle | Paraparesis and numbness | Surgical resection |
| Mak *et al*. [4] | 2009 | F/65 | Yes | Thoracic | Back pain and paraplegia | Surgical decompression |
| Pavlovic *et al*. [33] | 2009 | M/40 | Secondary | T9 body and pedicle | Paraplegia | Biopsy and surgical resection |
| Noman Zaheer *et al*. [34] | 2009 | M/30 | Secondary | Thoracic | Back pain and minimal neurological problem | Surgical resection |
| Kampschreur *et al*. [35] | 2010 | M/43 | Secondary | Thoracic | Upper abdominal pain radiating to back | Surgical resection and subtotal parathyroidectomy |
| Gheith *et al*. [36] | 2010 | M/19 | Secondary | Lumbar | Back pain and paraparesis | Surgical decompression and parathyroidectomy |
| | | F/25 | Secondary | Cervical | Neck pain and paraparesis | Surgical decompression and parathyroidectomy |
| Mateo *et al*. [37] | 2011 | F/34 | Secondary | C2 | Neck pain | Biopsy and parathyroidectomy |
| Fargen *et al*. [8] | 2011 | F/33 | Secondary | L1 | Paraparesis | Laminectomy and bracing |
| Araujo *et al*. [38] | 2012 | M/47 | Secondary | Lumboscral | Back pain and difficulty in gait | Posterior laminectomy and tumour excision |

Brown tumours are histologically similar to other giant cell tumours and diagnosis therefore depends on the histological findings along with a raised PTH level [1, 4, 5].

In terms of the underlying pathology, brown tumours are a localized form of osteitis fibrosa, the classical histological form of high-turnover renal osteodystrophy. They most commonly arise from the tuberous parts of the jaw, and in long bones and ribs [1]. Spinal cord lesions are less common [5–7] and historically reported more frequently in patients with primary

hyperparathyroidism. However, a recent literature review noted increased reporting of vertebral brown tumours in patients with ESRD over the last few decades for which this may simply reflect increased reporting or be a marker of increased survival in dialysis populations resulting in a true increase in the incidence of cases [8].

The first case of brown tumours involving the spine in a haemodialysis patient was reported in 1978 by Ericsson *et al.* [9]. In total and excluding the case presented here, 36 other cases of spinal cord compression secondary to brown tumours have been reported in the literature (Table 1). Of these, 64% (23 of 36) of cases have been reported in patients with secondary hyperparathyroidism due to chronic kidney disease (CKD). It is more common in females (61.1%). The mean age of the patients with secondary hyperparathyroidism was 43.67 ± 14.9 years. Most of the cases (58.3%) reported involvement of thoracic spine. These are similar findings to those quoted in a recently published review [8]. All cases presented with signs and symptoms of cord compression, 77.8% (28 of 36) had surgical resection of tumour and 69.4% had parathyroidectomy. One case did not report the modality of treatment.

Vertebral brown tumours can either present acutely due to cord compression with progressive neurological deficit or with symptoms caused by vertebral fracture [1, 4]. Radiological findings depend on the modality used. On plain X-rays, brown tumours usually present as an osteolytic lesion [1, 10]. In the long bones, these are usually well demarcated but in the spine the margins can be difficult to see. A computed tomography scan can confirm an osteolytic lesion with no cortical disruption or periosteal reaction [1, 5] but an MRI scan will provide a far more detailed image and often demonstrates the fluid cysts which are a highly suggestive marker of a brown tumour [5]. Brown tumours may mimic metastases on bone scan due to the presence of 'hot spots', a result of intense osteoclastic activity [8].

Treatment of brown tumours centres around treatment of the secondary hyperparathyroidism. Medical treatment includes aggressive dialysis, treatment with phosphate binders, vitamin D supplements and cinacalcet, although previous case reports have questioned the effectiveness of the latter [3, 8]. Parathyroidectomy is also commonly performed but requires a patient to be fit enough for a general anaesthetic. Normalizing bone biochemistry and parathyroid hormone levels generally result in bone remineralization and resolution of the tumours. However, remineralization may not happen in a spinal lesion; this is presumed to be due to the reduced amount of mechanical stress in comparison to long bones [6].

In conclusion, the possibility of a brown tumour should form part of the differential diagnosis in patients with advanced kidney disease presenting with peripheral neurological symptoms and a mass lesion. This case demonstrates the need for a high index of suspicion and highlights the need for nephrologists to be involved in the ongoing care of dialysis patients admitted to other specialties.

*Conflict of interest statement.* None declared.

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