Case report

Isolated dorsal column dysfunction due to an intraspinal Osteolipoma – Case report and review of literature

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ABSTRACT

Osteolipoma is a rare variant of the ubiquitous lipoma. Published literature appears in the form of isolated case reports affecting soft tissue structures in the head, neck and rarely affecting the spine. We present a unique instance of an intraspinal osteolipoma in the cervical spine, without evidence of accompanying spinal dysraphism and an atypical clinical presentation of isolated **dorsal column** dysfunction. We describe the clinical presentation, operative procedure and post-operative outcomes with histopathological findings of this rare entity. A 61-year-old male presented with impaired **dorsal column** sensation due to an intraspinal extradural ossifying lesion in the cervical spinal canal. The patient underwent excision of the lesion with complete resolution of symptoms. Ossifying lesion in the spinal canal may be frequent finding on radiological imaging but presence of possible adipose tissue in the lesion should raise suspicion of rare clinical scenario of an osteolipoma.

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1. Introduction

Lipomas are ubiquitous tumours of mature adipose tissue and are rarely a cause of clinical concern. They may sometimes contain other mesenchymal elements, such as fibrous connective tissue (Fibrolipoma), blood vessels (Angiolipoma) and cartilaginous/osseous metaplasia (Chondrolipoma, Osteolipoma).^{1–4} Osteolipoma is a rare variant and published literature, appears in the form of isolated case reports affecting soft tissue structures in the head and neck; rarely affecting the spine. The diagnosis of these lesions can be challenging on imaging, especially when they involve an intraspinal location. Ossified ligamentum flavum and ossified posterior longitudinal ligament are common intraspinal lesions which present with compressive myelopathy and can mimic appearance of an Osteolipoma on imaging.

We present a rare case of cervical intraspinal Osteolipoma causing compressive myelopathy with a predominant dorsal column dysfunction and review the relevant literature.

2. Case report

A 61-year-old gentleman presented to our clinic with complaints of neck pain for duration of 1 year. His symptoms included paraesthesias in both upper and lower limbs along with gait clumsiness which had worsened over the past 4 months.

The case showed a unique presentation with isolated dorsal column dysfunction evidenced by impaired joint position sense, vibratory sense in both lower limbs with sensory ataxia. Physical examination revealed normal tone, pain and temperature sensation were preserved with normal motor power in all four limbs. There was no difficulty in performing fine motor activity in the hands, no evidence of loss of dexterity and the impairment was predominantly seen in both lower limbs. There was no evidence of neurocutaneous markers suggesting spinal dysraphism. Bowel and bladder control were normal.

MRI scan revealed a posterior midline lesion measuring approximately 8 mm × 9 mm at the level of C5 vertebra. The periphery of the lesion was hypointense on T1 and T2 suggestive of ossification accompanied by a central focus of hyperintense signal change in both T1 and T2 weighted images. T1 hyper intensity within the lesion was not evident on the fat suppressed sequences; consistent with a diagnosis of lipomatous tissue (Fig. 1). There was severe canal stenosis (4.0 mm) with the compressed cord showing altered signals with reduced calibre and flattening.

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Fig. 1. MRI images of the case (A) T₂ weighted sagittal section shows the epidural ossified lesion (arrow) causing significant cord compression with altered hyperintense signal changes in the spinal cord. (B) T₂ weighted axial images through the level of the lesion showing hyperintense signals (arrow) in the ossified lesion with significant encroachment of the spinal canal with reduced calibre of the cord and hyperintense signal changes in the spinal (C) cord T₁ weighted sagittal section shows a posterior midline epidural space lesion with hyperintense signal intensity noted in the lesion at C5 level. (D) Fat suppressed axial sequences show attenuation of the signals inside the lesion (E) Axial CT scan through the level of the lesion shows an ossifying lesion encroaching into the spinal canal (F) CT sagittal image shows a posterior midline ossifying lesion (asterix) inside the spinal canal arising at the posterior spinolaminar line at the C5 level.

CT scan documented the ossific lesion occupying the spinal canal at the C5 level along the posterior spinolaminar line in close proximity to the lamina posteriorly (Fig. 1).

The patient was planned for a posterior decompression with a laminectomy. Through a posterior midline approach, the C4–C6 laminae were exposed and en-bloc laminectomy was performed. Using a high speed burr a trough was created at the lamina and lateral mass junction. The laminectomy was completed by Kerrison rongeurs to avoid introducing any instrument into the spinal canal. No neuromonitoring was used during the surgery. A well-delineated, oval, encapsulated mass was identified anterior to the ligamentum flavum with a clear plane of demarcation. The mass was removed en bloc and the spinal cord was completely decompressed. There was no evidence of spinal dysraphism and no intradural extension/extraspinous extension of the lesion was noted.

There were no intraoperative complications and postoperative period was uneventful.

The patient's signs and symptoms of dorsal column dysfunction improved early in the postoperative period. At 6-week follow-up, the patient showed good clinical improvement in joint position sense and gait pattern. By 3-month follow-up the patient was asymptomatic with no functional impairment. Patient was asymptomatic at final 24-month follow-up.

Gross examination of the specimen showed an oval mass measuring 2 cm × 1 cm × 1 cm; with smooth surface and a well-defined capsule. The tumour was firm to hard in consistency with gritty areas on cut sections. Histopathological study revealed lipomatous proliferation of mature adipose tissue with islands of bone and cartilage (Figs. 2 and 3). Osteoblastic rimming was noted at the edge of new bone formation. There was no evidence of

Fig. 2. Histopathology images of the lesion Haematoxylin and eosin stain (A) Low magnification (4×) microscopy image of the lesion showing islands of osteoid (asterix) with adjoining areas of adipose tissue (B) high magnification (40×) image of the lesion showing osteoid.

Fig. 3. Gross image showing a greyish, firm to hard consistency mass arising from the under surface of the lamina held by the forceps.

atypical hyperchromatic stromal cells or cytological atypia within the fatty component. These findings were consistent with the diagnosis of a benign osteolipoma.

3. Discussion

While lipomas are most common soft tissue tumours, Osteolipoma is a very rare variant and accounted for less than 1% of lipomas.¹ Various terms have been used to describe this entity which includes ossifying lipoma, Osteolipoma and osseous lipoma. These tumours have been reported at many sites; especially in the head and neck region including hypothalamus, suprasellar region, tuber cinereum, oral cavity, wrist, knee and thigh.^{2–7}

The differential diagnosis of osteolipoma includes other benign tumours that may contain bone-like Teratoma or Dermoid cysts. In addition to these, tumour calcinosis, calcification in a bursa, ossifying fibromas, myositis ossificans, and Osteosarcomas should be taken into consideration.

Although several theories have been put forth, pathogenesis of osteolipoma still remains unclear. Fritchie et al. reported cytogenetic analyses of three osteolipomas and concluded that the translocations involving 12q13–15 in all cases were consistent with the features of osteolipomas.⁶

Presence of adipose tissue in the intradural or extradural space is a commonly associated with spinal dysraphism.^{8,9} Jaiswal and coworkers reported a case of ossifying lipoma in the lumbar spine associated with a dermal sinus(spinal dysraphism).⁸ They excised the subcutaneous osseous component first which was connected to the intradural lipoma via a defect in the dura. Brones et al. presented the case of a 21-month-old girl with a posterior cervical subcutaneous/spinal lipoma that contained heterotopic bone; however the lesion was associated with a dermal sinus tract which extended to the dura without any intradural extension.⁹ Jin Seo Yang et al. reported a case of C2–C6 posterior muscular component Osteolipoma without presence of any extension into spinal canal.¹⁰

The presence of an intraspinal Osteolipoma variant in the absence of accompanying spinal dysraphism is extremely rare. To the best of our knowledge, there is only one other reported occurrence of Osteolipoma in an intraspinal location.¹¹ Ying-Chao Lin et al. described the first case of an intraspinal Osteolipoma located anterior to the cord in the cervical spine causing severe compressive myelopathy in a young adult. The patient had severe upper motor neuron type of weakness in all 4 limbs. They

successfully excised the tumour after performing C5–T1 laminectomy and demonstrated improvement in patient's motor functions.¹¹

Ossified ligamentum flavum or ossification of the posterior longitudinal ligament are frequent intraspinal lesion which present as compressive myelopathy with motor, sensory and dorsal column dysfunction. Isolated dorsal column dysfunction has been described most frequently with noncompressive myelopathy associated with Vitamin B 12 deficiency or Copper deficiency.^{12,13} Rarely other noncompressive aetiologies such as chemotherapy, diabetes and demyelination have been suggested.^{12,13}

In this case the preoperative x-ray showed a maintained cervical lordosis so we planned a cervical laminectomy without instrumentation. The osteolipoma was in the midline, in the posterior epidural space arising from the lamina, we preferred an en bloc laminectomy to laminoplasty; thus allowing for complete excision of the lesion.

In our patient the Osteolipoma resulted in a compressive myelopathy with isolated dorsal column involvement which appears to be an atypical presentation and the second reported case of intraspinal Osteolipoma without spinal dysraphism.

4. Conclusion

This case represents a unique presentation of an extremely rare benign tumour. CT and MR imaging are excellent aides in the diagnosis of these tumours. Presence of adipose tissue in an ossifying lesion should raise suspicion of Osteolipoma and complete resection is the treatment of choice and gives excellent results.

Ethical concerns

Informed consent was obtained from the participant.

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Conflict of interest

The authors have none to declare.

References

- Heffernan EJ, Lefavre K, Munk PL, Nielsen TO, Masri BA. Ossifying lipoma of the thigh. *Br J Radiol.* 2014 Jan 28.
- Allard RH, Blok P, Kwast WA, Waal I. Oral lipomas with osseous and chondrous metaplasia; report of two cases. *J Oral Pathol Med.* 1982;11(January (1)):18–25.
- Blanshard JD, Veitch D. Ossifying lipoma. *J Laryngol Otol.* 1989;103(April (04)):429–431.
- Sinson G, Gennarelli TA, Wells GB. Suprasellar osteolipoma: case report. *Surg Neurol.* 1998;50(November (5)):457–460.
- Wittig H, Kasper U, Warich-Kirches M, Dietzmann K, Roessner A. Hypothalamic osteolipoma: a case report. *Gen Diagnos Pathol.* 1997;142(June (5–6)):361–364.
- Fritchie KJ, Renner JB, Rao KW, Esther RJ. Osteolipoma: radiological, pathological, and cytogenetic analysis of three cases. *Skeletal Radiol.* 2012;41(February (2)):237–244.
- Demiralp B, Alderete JF, Kose O, Ozcan A, Cicek I, Basbozkurt M. Osteolipoma independent of bone tissue: a case report. *Cases J.* 2009;2(September):8711.
- Jaiswal AK, Garg A, Mahapatra AK. Spinal ossifying lipoma. *J Clin Neurosci.* 2005;12(August (6)):714–717.
- Brones A, Mengshol S, Wilkinson CC. Ossifying lipoma of the cervical spine: case report. *J Neurosurg Pediatr.* 2010;5(March (3)):283–284.
- Yang JS, Kang SH, Cho YJ, Choi HJ. Pure intramuscular osteolipoma. *J Kor Neurosurg Soc.* 2013;54(December (6)):518–520.
- Lin YC, Huang CC, Chen HJ. Intraspinal osteolipoma: case report. *J Neurosurg Spine.* 2001;94(January (1)):126–128.
- Joseph PJ, Reyes MR. Dorsal column myelopathy following intrathecal chemotherapy for acute lymphoblastic leukemia. *J Spinal Cord Med.* 2014;37(January (1)):107–113.
- Prick JJ, Prevo RL, Hoogenraad TU. Transient myelopathy of the cervical posterior columns in a young man with recently diagnosed diabetes mellitus. *Clin Neurol Neurosurg.* 2001;103(December (4)):234–237.