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Case Report

A rare primary sacral glomus tumor presenting as intradural-extramedullary tumor: A Case report and review of literature

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ABSTRACT

Background: Glomus tumors are very infrequent in the spine where they can grow intraosseously at any level. We were able to identify only eight such cases in the literature, with only one occurring in the sacrum. Here, a 48-yearold male with sacral S1/S2 radiculopathy was found to have a rare glomangioma/glomus tumor of the sacral region.

Case Description: A 48-year-old male presented with left-sided S2 radiculopathy characterized by left lower extremity weakness/paresis. The magnetic resonance showed an intradural extramedullary mass measuring $1.8 \times 1.9 \times 4.3$ cm at S1-S2 extending through the left foramen, inhomogeneously enhanced with contrast. He underwent an S1-S2 durotomy with gross total excision of the mass. Pathologically, it proved to be a glomus tumor. Two months postoperatively, he ambulated without the support and demonstrated no tumor recurrence at 1 postoperative year.

Conclusion: Glomus tumors involving the sacral region are rare and can be successfully excised resulting in good clinical outcomes.

Keywords: Glomoid cells, Glomus tumor, Intradural extramedullary, Intraosseous, Sacrum

INTRODUCTION

Glomus tumors are benign mesenchymal tumors that comprise <2% of soft tissue tumors. [3] Many are clinically misdiagnosed as hemangiomas or venous malformations. These lesions are usually located near the distal phalanges, where they sometimes erode the bone. We identified 8 reports of these tumors involving the spine, with five occurring in the thoracic, one at the thoracolumbar region, [5] one in the sacrum, and one in the coccyx. [2] Here, we present a 48-yearold male who presented with a left-sided S1/S2 radiculopathy attributed to a glomus sacral tumor that was successfully surgically excised.

CASE REPORT

A 48-year-old male presented with chronic backache, left-sided radicular pain in the S1/S2 dermatome, and 4/5 left leg weakness of 3 - 4 years duration. The lumbosacral

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magnetic resonance imaging (MRI) revealed a lobulated heterogeneous lesion measuring $1.8 \times 1.9 \times 4.3$ cm that was ISO intense on T1-weighted images, ISO/hyperintense on T2-weighted (T2W)/short tau inversion recovery images, and enhanced homogeneously with contrast [Figures 1 and 2]. The lesion occupied the left anterolateral spinal canal, was posterior/posterolateral to the S1/S2 vertebra, and extended through the left S1/S2 intervertebral foramen [Figure 3]. Previously, it was diagnosed as a neurofibroma measuring $0.7 \times 1.7 \times 2.8$ cm.

Surgery

At our institution, utilizing an operating microscope and intraoperative neuromonitoring, the patient underwent reexploration of the tumor. The sacral dura was exposed and opened in the midline at the S1/S2 level. Nerve roots were mobilized, and an anterior durotomy exposed the extradural



Figure 1: T1-weighted image showing hypo to isointense lesion from S1 to S2.



Figure 2: T2-weighted image hyperintense lesion from S1 to S2.

and highly vascular lesion. A clear dissection plane was identified between the tumor and the nerve roots, allowing for gross total excision in a piecemeal fashion.

Pathology

The histopathology was consistent with a glomus tumor. Cells were arranged in sheets with cells showing mild pleomorphism in a perivascular pattern. They were round to oval with vesicular chromatin, inconspicuous nucleoli, and a moderate amount of clear cytoplasm. Approximately 2-3 mitosis were observed per 10 high power fields. No necrosis was noted. The tumor cells expressed vimentin and smooth muscle actin (SMA), lacked expression of S100, Glial fibrillary acidic protein (GFAP), synaptophysin, CD34, Human Melanoma Black 45 (HMB45), and PanCK. Ki 67 proliferation index was 2-4% in the highest proliferative areas.

DISCUSSION

The glomus body is a specialized form of arterial-venous anastomosis that consists of nerves, smooth muscles, and blood vessels, usually located at the dermal subdermal junction of fingers. Multiple tumors are usually between 0.1 and 0.3 cm in diameter but occasionally have been reported to reach up to 5 cm; in this case, the maximum dimension was 4.3 cm.^[7] On computed tomography studies, these lesions may appear as ovoid, well-circumscribed lesions with accompanying soft tissue changes and possible bone erosion. Kuo et al.[4] reported an epidural glomus tumor in a 26-year-old male with compressive myelopathy at the T11 level that showed reactive sclerotic changes over the vertebral body/lamina. On MRI, spinal glomus lesions are ovoid and well-defined lesions showing inconsistent findings on T1 but clear hyperintensity on T2W MRI images; further, they markedly enhance with contrast. [6]

Treatment

Treatments for spinal glomus lesions include sclerotherapy, laser, or surgical excision (i.e., based on lesional location and clinical presentation). Preoperative embolization can also help

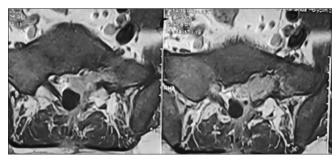


Figure 3: Postcontrast image showing non-homogeneous postcontrast enhancement extending into the left intervertebral foramen.

reduce intraoperative blood loss, especially in larger lesions.^[1] These lesions are best managed with gross total tumor excision.

CONCLUSION

Glomus tumors of the spine are rare and are best managed with gross total tumor excision.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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