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Spinal intraosseous schwannoma without spinal canal and neuroforamina involvement: A case report

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Abstract

BACKGROUND

Spinal intraosseous schwannomas (SIS) are rare, and as yet have not been fully described in the literature. The first case of SIS was reported in 1971, and 24 cases of SIS have been reported so far. However, including the present case, there are only seven cases without spinal canal and neuroforamina involvement.

CASE SUMMARY

A 56-year-old man presented with a history of neck pain for 2 years. An obvious osteolytic destruction of the seventh cervical (C7) vertebra was observed on imaging examination. Magnetic resonance imaging of the cervical spine showed space-occupying lesions in the C7 vertebra, and destruction of the anterior cortex of the vertebra. The lesions had an exophytic component that extended from the C7 vertebra into the soft tissue on the front side. The foramen transversarium on both sides were intact. The patient underwent surgical biopsy and focal excision of the C7 lesion. The diagnosis of "schwannoma" was verified by postoperative pathological examinations. In a review of the literature, this is the seventh case of SIS without spinal canal and neuroforamina involvement, and the third reported case of type VIII SIS. We discussed our case with respect to reported classification characteristics of SIS.

CONCLUSION

SIS is a very rare tumor. We report a rare case that may be important for further classification of osteo-schwannoma. The establishment of a complete disease classification is of high importance for the treatment and prognosis of this disease. Thus, more basic studies and retrospective analysis of related cases are necessary.

Key words: Spinal intraosseous schwannoma; Neurilemmoma; Spinal tumor; Classification; Case report; Cervical

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Core tip: Intraosseous schwannomas are extremely rare, accounting for < 0.2% of primary bone tumors. Spinal intraosseous schwannomas (SIS) are even rarer. We present a rare case of SIS of the seventh cervical (C7) vertebra in a 56-year-old man presenting with a history of neck pain for 2 years. An obvious osteolytic destruction of the C7 vertebra was observed on imaging examination. The patient underwent surgical biopsy and focal excision of the C7 lesion. The diagnosis of “schwannoma” was verified by postoperative pathological examinations. In a review of the literature, this is the seventh case of SIS without spinal canal and neuroforamina involvement, and the third case of type VIII SIS. It is important for further classification of osteo-schwannoma.

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INTRODUCTION

Intraosseous schwannomas are extremely rare, accounting for < 0.2% of primary bone tumors. The most common intraosseous lesions include the mandible and sacrum, while spinal lesions are less common. Spinal intraosseous schwannomas (SIS) are even rarer. Only 24 cases of SIS have been reported, of which 18 cases had lesions extending into the spinal canal or neuroforamina. We here present the seventh case of SIS without spinal canal and neuroforamina involvement, and the third case of type VIII SIS, with a review of the literature and discussion of this rare tumor.

CASE PRESENTATION

Chief complaints

A 56-year-old man presented with chronic and persistent neck pain for three months.

History of past illness

He had no previous trauma and medical problems.

Physical examination

There was local pain elicited by percussion of cervical spine. The range of cervical spinal motion was decreased, especially with regard to the flexion. Neurological examination revealed no abnormalities.

Laboratory examinations

Routine blood examination and the C reactive protein concentration were normal.

Imaging examinations

Conventional radiograph of the cervical spine showed loss of height of the seventh cervical (C7) vertebral body (Figure 1A). Magnetic resonance imaging (MRI) of the cervical spine showed space-occupying lesions in the C7 vertebra, and destruction of the anterior cortex of the vertebra. The lesions had an exophytic component that extended from the C7 vertebra into the soft tissue on the front side. The foramen transversarium on both sides were intact. The lesion area was well-circumscribed, showing a low signal on T1-weighted MRI and a high signal on T2-weighted MRI (Figure 1B and 1C). There was no extension of the lesion into the spinal canal, and no involvement of the spinal cord nerve. Computed tomography (CT) showed a C7 vertebral body lesion with lytic features indicative of a malignant etiology, and a bony defect of the C7 body, although the bony cortex of the vertebra was well preserved (Figure 1D-F).

Histological examination

Histological examination of the lesion showed Antoni A and B areas with hypercellular, palisaded cells alternating with hypocellular stroma (Figure 2A). There were also regions with nuclear palisading suggestive of Verocay body formation (Figure 2B). Immunohistochemical staining with S100 protein and glial fibrillary

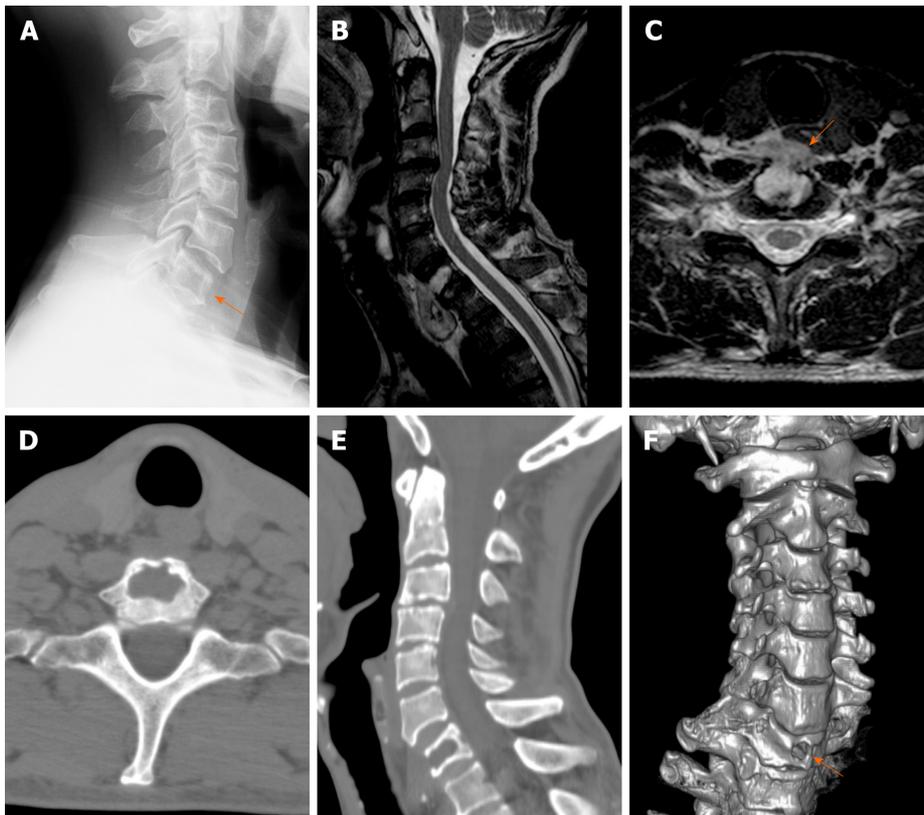


Figure 1 Imaging findings before treatment. A: Cervical X-ray showed loss of height of the seventh cervical (C7) vertebral body; B: Sagittal T2-weighted magnetic resonance imaging of the cervical spine showing a hyperintense prevertebral mass; C: Axial magnetic resonance imaging showed a well-demarcated lesion seemingly originating in corpus C7 without spinal canal and neuroforamina involvement; D: Axial computed tomography (CT) shows lytic changes in the anterior cortical margin of C7 body. Posterior cortical margin is intact; E: CT scanning exhibited a bony defect of the C7 body; F: Three-dimensional CT reconstruction shows a circular defect in the anterior edge of the C7 vertebra.

acidic protein were diffusely positive (Figure 2C and 2D). The features were of a schwannoma (World Health Organization grade I). No nerve root remnants were identified in the specimen.

FINAL DIAGNOSIS

The final diagnosis of the presented case is spontaneous cerebral abscess due to spinal intraosseous schwannoma.

TREATMENT

Based on the findings of typical vertebral lesions, suspected malignancy, with no evidence of a primary malignant process, surgical treatment was considered. The patient was operated upon via an anterior approach. A well-capsulated mass (3 cm × 2 cm) was found, which seemed to extrude from the anterior of the C7 vertebral body. The exophytic component was excised and the tumor was totally removed from the vertebral body using curettes, followed by C7 fusion using an iliac bone autograft.

OUTCOME AND FOLLOW-UP

One week after surgery, postoperative computed tomography and X-ray did not reveal any evidence of tumor recurrence (Figure 3A-3D). The patient recovered well in the postoperative period. The neck pain was disappeared and at 1-year follow-up, the patient was doing well without any deficits.

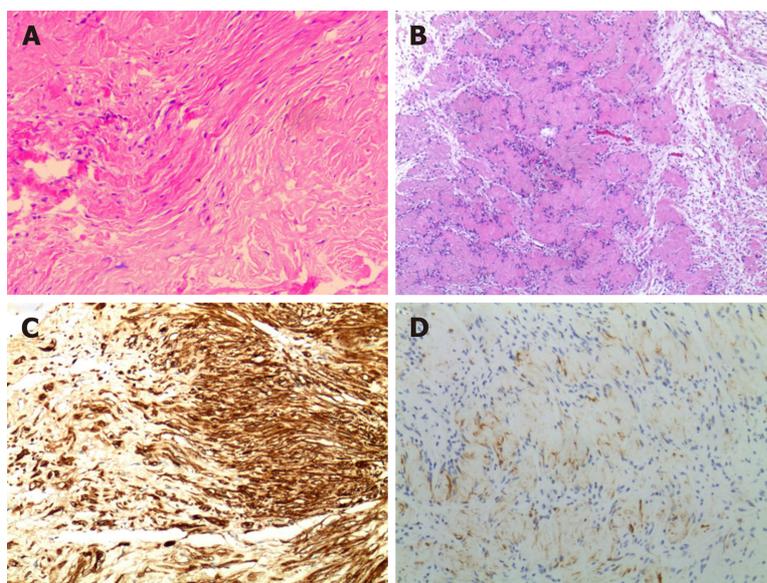


Figure 2 Histological examination of the lesion (hematoxylin and eosin; $\times 200$). A: Histology revealed hypercellular (Antoni A) and hypocellular (Antoni B) areas, indicating a typical schwannoma; B: Tumor cells constitute the Verocay; C: Immunohistochemical study for S100 protein is diffusely positive; D: Immunohistochemistry showed positive glial fibrillary acidic protein.

DISCUSSION

Intraosseous schwannomas are rare benign tumors, accounting for $< 0.2\%$ of primary bone tumors. The most common intraosseous lesions include the mandible and sacrum, while spinal lesions are less common. The first case of SIS of a lumbar vertebral body was reported by Dickson *et al*^[1] in 1971, where the lesion was located at the level of L3, but with extension to the spinal canal resulting in compression of the thecal sac. Only 24 cases of SIS (Table 1) have been reported, of which 18 cases had lesions extending into the spinal canal or neuroforamina. These tumors originally arose from the abutting nerve root, extending into the spinal canal and invading the vertebrae, suggesting that the SIS lesions likely involved intraosseous invasion of the extraosseous nerve sheath tumor originating from the spinal nerve root. This hypothesis is supported by most reports on SIS. However, the classification of SIS has not been fully described in the literature.

Wang *et al*^[2] described the largest series of cases of SIS to date, with a retrospective analysis of the clinical features, surgical strategies, and outcomes of 20 cases of SIS. In that study, all of the lesions extended from layers A to D of the Weinstein-Boriani-Biagini classification, and all lesions exhibited extraosseous components extending into the spinal canal. In 2001, Sridhar *et al*^[3] classified benign nerve sheath tumors, in which V-type tumors caused vertebral erosion. Park *et al*^[4] modified this classification by adding type VI and type VII lesions; type VI is a complete intraspinal tumor, while type VII is an intraspinal tumor with vertebral erosion and expansion into the nerve foramen. However, the present case does not fit into any of these subtypes.

Mohanty *et al*^[5] proposed a new subtype (type VIII) involving intraosseous tumors with an exophytic component, but without any intraspinal or neural foraminal extension. The present case is the third case of this VIII type SIS (Figure 4). The first case was published by Schreuder *et al*^[6] in 2001, who described a lesion that extruded from the C6 vertebral body, and passed in front of the C7 vertebral body, although no specific category was described. We believe that the establishment of a VIII type is very important for SIS, and that the VIII and V types are associated with very different symptoms, surgical methods, and prognosis. Type VIII, which has an exophytic component, may present with symptoms related to compression of the surrounding structures such as dysphagia or pain, while no radicular pain or myelopathic features are present. Because the tumor does not involve the spinal canal or neuroforamina, it is not necessary to separate the tumor from the nerve root, making the operation safer. Postoperative nerve root numbness was reported in some patients.

A small number of patients with type VII relapsed during follow-up time, while no patient with type VIII relapsed. This may relate to the small number of patients with VIII, as well as the inability to fully dissect the nerve and tumor during type VII

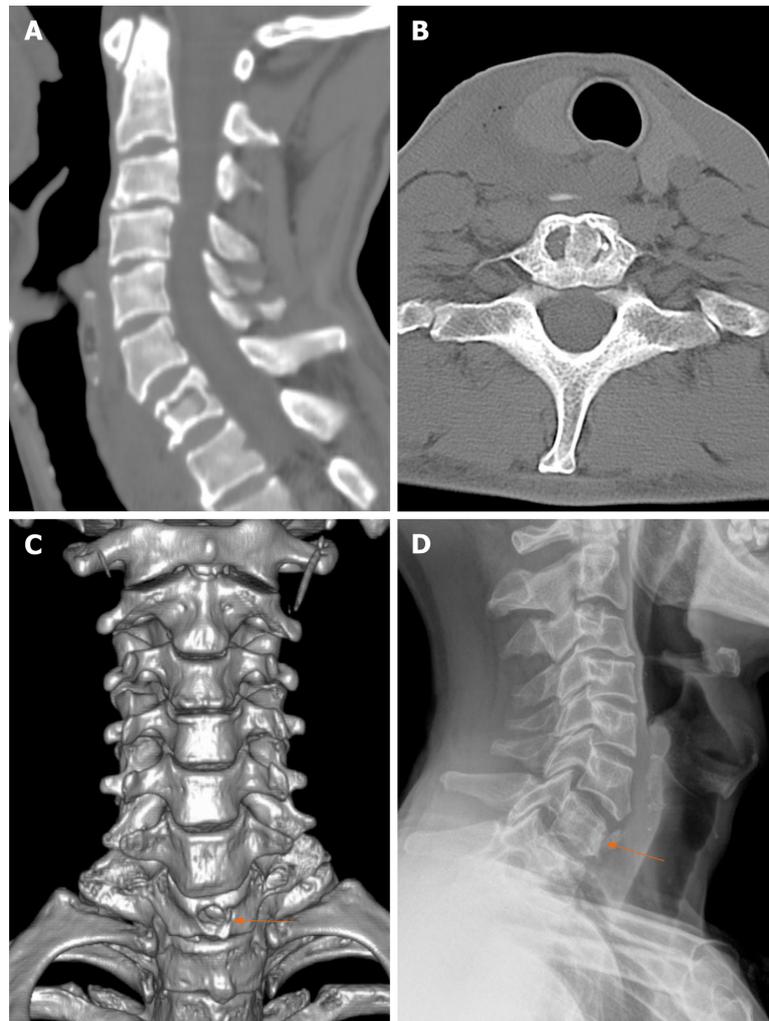


Figure 3 Imaging findings after treatment. A-C: The axial computed tomography (CT) images (A), sagittal CT images (B) and the Three-dimensional CT reconstruction (C) of the cervical vertebra after the operation showed a good position for bone grafting; D: The last follow-up cervical X-ray showed no radiological recurrence.

surgery. Most authors believe that vertebral resection and external fixation are appropriate surgical options, and the other two reported type VIII cases^[5,6] utilized vertebral resection and external fixation. In the present case, the lesion was circumscribed within the cortical bone, the pedicle and facet joints were intact, and the stability of the cervical spine was not significantly decreased. Thus, after the lesion was completely scrapped, we used iliac bone grafting at the anterior edge of the vertebral body, without spinal external fixation. According to the follow-up and re-examination at 1 year after surgery, the prognosis of the patient was favorable, and no recurrence was observed (Figure 3D).

CONCLUSION

SIS is a very rare tumor. We report a rare case that may be important for further classification of osteo-schwannoma. The establishment of a complete disease classification is of great importance for the treatment and prognosis of this disease. Thus, more basic studies and retrospective analysis of related cases are necessary.

Table 1 Summary of all published cases of spinal intraosseous schwannomas

No.	Ref.	Gender/age	Symptoms	Level	Classification	Stabilization	Final follow-up
1	Dickson et al ^[1] , 1971	F/51	Radicular pain	L3	VII	Iliac crest bone graft	No recurrence
2	Polkey et al ^[7] , 1975	F/34	Pain and weakness	C6-C7	VII	Iliac crest bone graft	No recurrence
3	Naidu et al ^[8] , 1988	M/50	Weakness	C3-C4	VII	-	No recurrence
4	Nooraie et al ^[9] , 1997	M/46	Asymptomatic	T12	VII	Fusion + fixation	No recurrence
5	Chang et al ^[10] , 1998	M/58	Pain and numbness	L4-L5	VII	Fusion + fixation	No recurrence
6	Schreuder et al ^[6] , 2001	F/38	Dysphagia	C6	VIII	Fusion + fixation	No recurrence
7	Ramasamy et al ^[11] , 2001	M/37	Local pain, weakness	T12	VII	Fusion + fixation	No recurrence
8	Gupta et al ^[12] , 2005	F/30	Local pain, weakness	L2	VII	Fusion + fixation	No recurrence
9	Nannapaneni et al ^[13] , 2005	M/42	Asymptomatic	C5	VIII	Fusion + fixation	No recurrence
10	Singrakhia et al ^[14] , 2005	M/43	Numbness, weakness	C3-C4	VII	Fusion + fixation	Recurrence
11	Singrakhia et al ^[14] , 2006	M/45	Radicular pain, weakness	C3	VII	Fusion + fixation	Recurrence
12	Choudry et al ^[15] , 2007	M/18	Local pain, weakness	T12	VII	Fusion + fixation	No recurrence
13	Cetinkal et al ^[16] , 2009	F/55	Local pain, numbness	T12	VII	-	No recurrence
14	Park et al ^[4] , 2009	F/46	Local pain	L4	VII	Fusion + fixation	No recurrence
15	Kojima et al ^[17] , 2010	M/60	Weakness, numbness	T9	VII	Fixation	No recurrence
16	Mizutani et al ^[18] , 2010	F/44	Paresthesia	C4	VI	-	No recurrence
17	Peng et al ^[19] , 2011	M/44	Weakness	C3	VII	Fusion + fixation	Recurrence
18	Youn et al ^[20] , 2012	M/65	Local, numbness	L2	VII	Fusion + fixation	No recurrence
19	Mohanty et al ^[5] , 2013	M/10	Local pain, dysphagia	C4	VIII	Fusion + fixation	No recurrence
20	Song et al ^[21] , 2014	M/44	Numbness	L5	VII	Fusion + fixation	No recurrence
21	Zhang et al ^[22] , 2015	M/71	Radicular pain, weakness	L4	VII	Fusion + fixation	No recurrence
22	Zhang et al ^[22] , 2015	F/54	Radicular pain, weakness	T9	VII	Fusion + fixation	No recurrence
23	Jia et al ^[23] , 2018	F/64	Radicular pain, weakness	T7-T8	VI	Fusion + fixation	No recurrence
24	Zaidman et al ^[24] , 2019	F/56	Asymptomatic	T1	VI	Fusion + fixation	No recurrence
25	Present study	M/56	Local pain	C7	VIII	Iliac crest bone graft	No recurrence

C: Cervical; T: Thoracic; L: Lumbar.

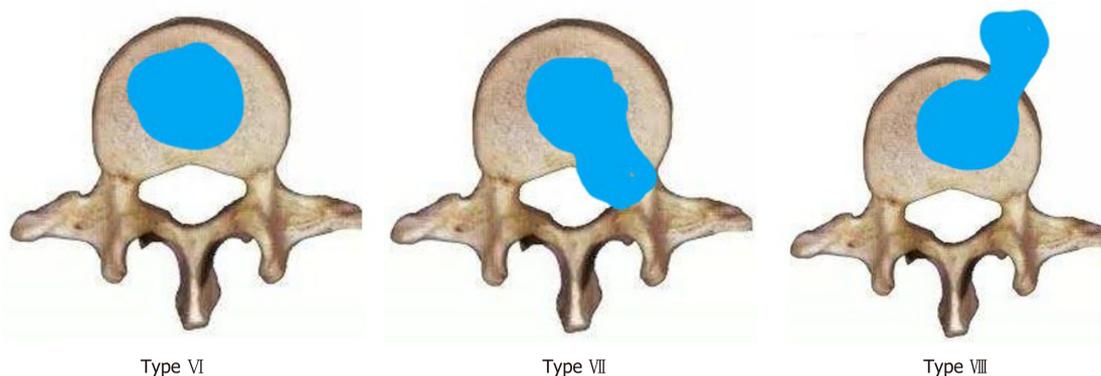


Figure 4 Modified classification of spinal intraosseous schwannomas. Type VI: Tumors in entirely intravertebral location without intraspinal portion; Type VII: Intraspinal tumor with erosion into vertebral bodies (invasive tumor) and extension into nerve root foramen; Type VIII: Tumors including intraosseous tumors with exophytic component without intraspinal or neural foraminal extension.

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