



# Intraosseous schwannoma of the mobile spine: a report of twenty cases

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## Abstract

**Purpose** To clarify the clinical features, surgical strategies, and outcomes of intraosseous schwannoma (IOS) of the mobile spine.

**Methods** We retrospectively reviewed patients with primary benign spinal schwannoma who underwent surgery in our orthopedic department.

**Results** A total of 101 patients with primary benign schwannoma located in the mobile spine underwent surgery in our orthopedic department from 2005 to 2015. Twenty-five patients presented with aggressive features. Twenty patients were regularly followed up, twelve with lesions in the cervical spine, six with lesions in the thoracic region, and two with lesions in the lumbar spine. Preoperative CT-guided biopsy was performed in fourteen cases; the accuracy of diagnosis was 100%, and IOS is not histologically different from conventional schwannoma. The computed tomography (CT) scan revealed expansile and osteolytic bone destruction in all these cases, with six patients having pathological fracture. On T2-weighted magnetic resonance imaging, the lobulated schwannomas showed heterogeneous signal intensity and significant heterogeneous enhancement on post-contrast images. Gross total resection was performed in seventeen patients and subtotal resection in three. Tumor-involved nerve roots resection were documented to decrease local recurrence in fourteen cases. The visual analog scale score decreased from  $5.66 \pm 1.79$  preoperatively to  $1.16 \pm 1.77$  at the final follow-up. No local recurrence was noticed at the final follow-up.

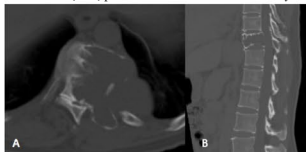
**Conclusion** CT-guided biopsy is effective for the preoperative diagnosis of spinal IOS. Total resection is the optimal treatment for IOS, whereas subtotal resection could be an alternative choice for high-risk cases.

**Graphical abstract** These slides can be retrieved under electronic supplementary material.

Spine Journal

### Key points

Spinal schwannomas are usually present as extramedullary, intradural tumors. And intraosseous schwannomas (IOSs) present features of invasive and osteolytic destruction.

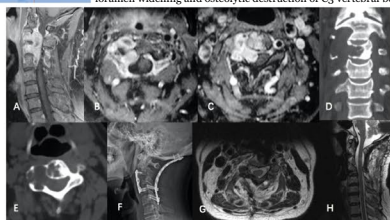


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Cervical intraosseous schwannoma with C2-3 intervertebral foramen widening and osteolytic destruction of C3 vertebral body



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### Take Home Messages

1. Intraosseous schwannoma (IOS) is not as rare as previously thought.
2. CT-guided biopsy is effective for preoperative diagnosis of spinal IOS.
3. Total resection is the optimal treatment for IOS, whereas subtotal resection could be an alternative choice for high risk cases.

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**Keywords** Intraosseous schwannoma · Mobile spine · Surgery · Prognosis · Biopsy

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Extended author information available on the last page of the article

## Introduction

Spinal schwannomas are benign tumors that usually present as extramedullary intradural tumors [1] and seldom erode the bone [2]. Sometimes, they present features of invasive and osteolytic destruction of the spine, making it difficult to distinguish them from malignant spinal tumors, and such tumors are defined as intraosseous (invasive) schwannomas (IOSs). There is little consensus on the definition of IOS, which is Type V in both Sridhar's classification [3] and PUTH classification [4].

IOSs are rare, accounting for <0.2% of primary bone tumors [5]. Accordingly, it is difficult to obtain a large sample size to assess a surgical procedure for treating these tumors. Relevant literature available are mostly case reports. En bloc resection is the ideal treatment for IOS, but sometimes difficult to accomplish.

In this study, we retrospectively analyzed twenty cases of IOS treated in our orthopedic department and clarified its

clinical features, surgical strategies, and outcomes, which have been the focus of controversy among authors. To the best of our knowledge, this has been the largest case series to date. The ethics committee of our university hospital approved this study.

## Materials and methods

### Inclusion criteria

Patients were selected according to the following criteria: (1) The treatment was conducted between 2005 and 2015; (2) the diagnosis of "schwannoma" was verified by post-operative pathological examinations; (3) obvious osteolytic destruction was observed on computed tomography (CT); and (4) there was a minimum of 2-year follow-up.

**Table 1** Patients' demographics and operative details

| No. | Gender/<br>age | Symptom                             | Level  | WBB clas-<br>sifications:<br>La/S | Approach                        | Resection | Instrumentation           | F/U (yrs) | Final follow-up |
|-----|----------------|-------------------------------------|--------|-----------------------------------|---------------------------------|-----------|---------------------------|-----------|-----------------|
| 1   | M/48           | Local pain, numbness, weakness      | C2–C3  | 6–9/A–D                           | Anterior + pos-<br>terior       | Total     | P: Occiput–C5<br>A: C2–C4 | 12        | No recurrence   |
| 2   | M/38           | Local pain, numbness, weakness      | C1     | 2–5/A–D                           | Posterior                       | Total     | Occiput–C3                | 12        | No recurrence   |
| 3   | M/50           | Asymptomatic                        | L5     | 4–8/A–D                           | Posterior                       | Subtotal  | L3–L5                     | 11        | Residual        |
| 4   | F/50           | Numbness, weakness                  | C2–C3  | 7–9/A–D                           | Posterior                       | Total     | C2–C4                     | 9         | No recurrence   |
| 5   | M/46           | Local pain                          | C1     | 2–6/A–D                           | Posterior + pos-<br>terolateral | Subtotal  | Occiput–C3                | 8         | Residual        |
| 6   | F/49           | Weakness, numbness, local pain, BBD | T9–T11 | 2–8/A–D                           | Posterior                       | Total     | T8–L1                     | 8         | No recurrence   |
| 7   | F/44           | Numbness, BBD                       | T2–T4  | 7–10/A–D                          | Posterior                       | Total     | T1–T3                     | 7         | No recurrence   |
| 8   | F/33           | Local pain, weakness                | T7     | 1–6/A–D                           | Posterior                       | Total     | T5–T9                     | 7         | No recurrence   |
| 9   | M/42           | Asymptomatic                        | C5–C6  | 3–6/A–D                           | Anterior                        | Total     | C5–C7                     | 4         | No recurrence   |
| 10  | F/45           | Radicular pain, numbness            | C6     | 4–8/A–D                           | Anterior                        | Total     | C5–C7                     | 4         | No recurrence   |
| 11  | F/29           | Local pain, numbness                | C3–C5  | 3–9/A–D                           | Anterior                        | Subtotal  | C1–C5                     | 4         | Residual        |
| 12  | F/55           | Local pain                          | T12    | 5–9/A–D                           | Posterior                       | Total     | T10–L2                    | 5         | No recurrence   |
| 13  | F/36           | Local pain, numbness                | L5     | 1–7/A–D                           | Posterior                       | Total     | L4–S1                     | 4         | No recurrence   |
| 14  | M/29           | Radicular pain, numbness, weakness  | C4     | 4–8/A–D                           | Posterior                       | Total     | C2–C6                     | 4         | No recurrence   |
| 15  | F/34           | Radicular pain, numbness            | C6–C7  | 6–9/A–D                           | Anterior + pos-<br>terior       | Total     | P: C4–T2 A:<br>C5–T1      | 4         | No recurrence   |
| 16  | M/32           | Numbness, weakness                  | C7     | 6–9/A–D                           | Anterior                        | Total     | C6–T1                     | 3         | No recurrence   |
| 17  | M/49           | Numbness, weakness                  | C5     | 4–9/A–D                           | Anterior + pos-<br>terior       | Total     | C4–C6                     | 3         | No recurrence   |
| 18  | M/64           | Asymptomatic                        | T8     | 8–10/A–D                          | Posterior                       | Total     | T7–T10                    | 2         | No recurrence   |
| 19  | F/26           | Asymptomatic                        | C5     | 6–9/A–D                           | Anterior                        | Total     | C4–C6                     | 2         | No recurrence   |
| 20  | F/68           | Local pain, numbness, weakness      | T5–T6  | 5–9/A–D                           | Posterior                       | Total     | T3–T7                     | 2         | No recurrence   |

C, cervical; T, thoracic; L, lumbar; F/U, follow-up; yrs, years; BBD, bowel and bladder dysfunction; WBB, Weinstein–Boriani–Biagini; La, layers; S, sectors; P, posterior; A, anterior

## Exclusion criteria

The exclusion criteria were: (1) prior spinal tumor resection; (2) malignant cases or neurofibromatosis cases; (3) conventional spinal schwannomas (e.g., cortex scalloping); (4) recurrent tumors; and (5) tumors in the sacrum.

## Methods

### General information

Medical records, pathological reports, and radiographic studies were reviewed retrospectively. The study parameters included patients' age, gender, tumor location, preoperative and postoperative symptoms, estimated blood loss, and surgical complications.

### Imaging and biopsy

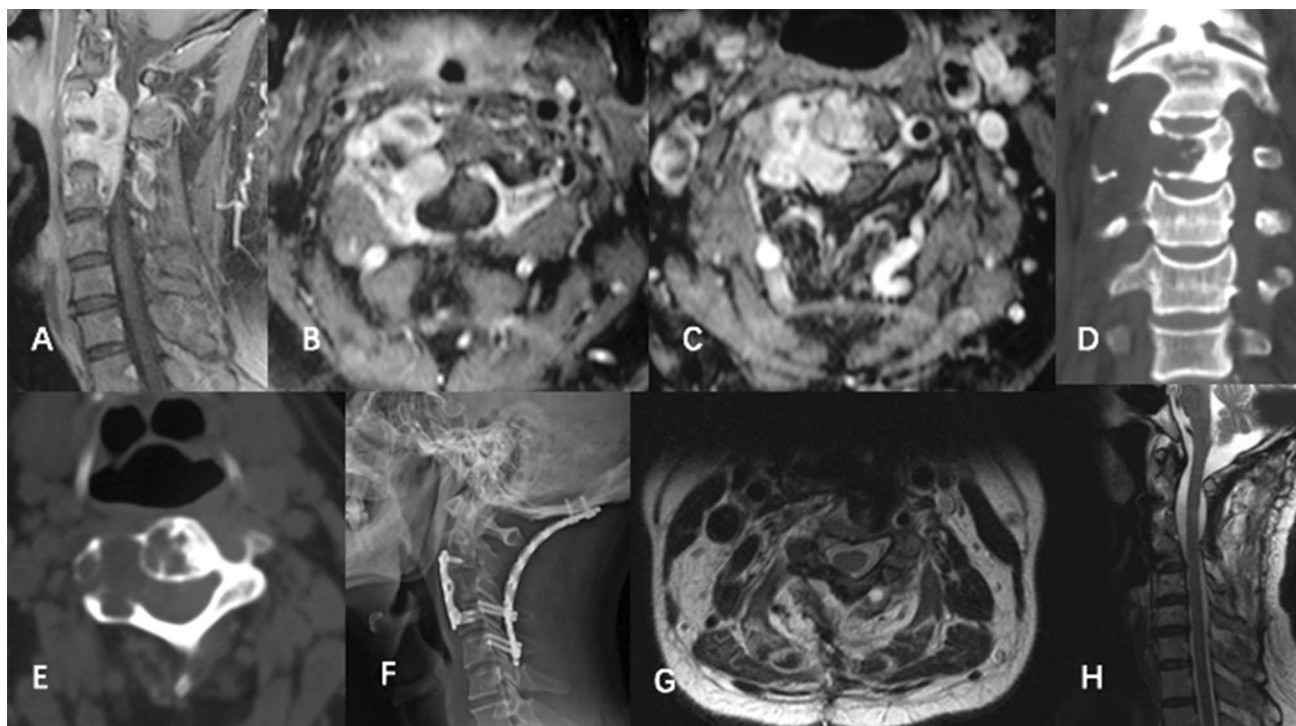
We routinely took anteroposterior radiographs, reconstructive CT and MRI before surgery, at 3, 6, 12, and 24 months after surgery, and annually thereafter. Percutaneous

CT-guided trocar biopsy under local anesthesia was performed for suspected malignant tumors.

### Surgical strategy

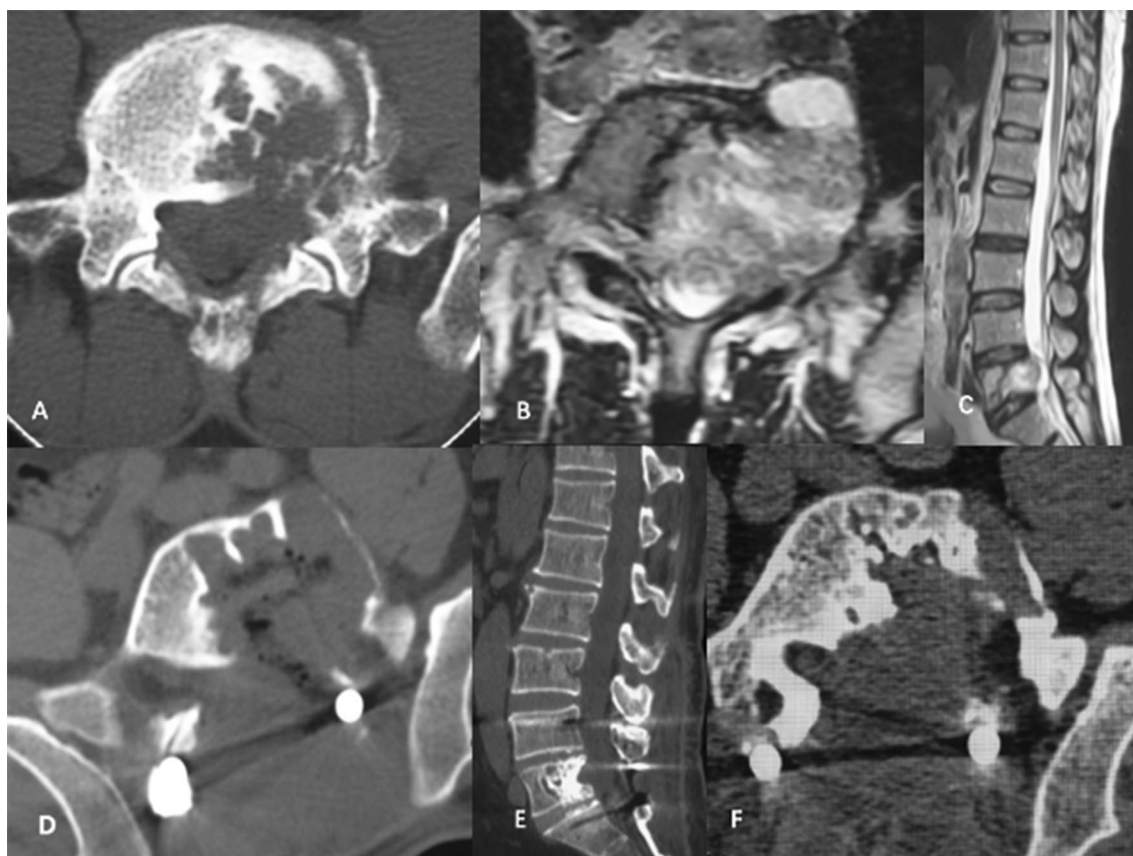
Although total resection could provide cure, it is difficult to accomplish it, given the potential for neurological deficit. The individualized surgical strategy was balanced between cure and surgical complications, and usually intralesional resection was preferred. Extensive curettage was usually performed for the bony lesions, followed by the use of a burr to remove the residual lesions in the niches; for the paraspinal and epidural lesions, marginal resection was achieved and the tumor was removed en bloc with the capsule. However, in cases with possible vascular injury, we preferred intralesional curettage, and the residual tumor capsule was coagulated with bipolar forceps. In cases with suspected benign lesion without a preoperative biopsy, our surgical strategy (intralesional resection) was the same as in those cases where biopsy was performed, with pathological diagnosis confirmed intraoperatively by frozen pathology.

The approach adopted was usually determined by the location of the tumor. Posterior approach was usually



**Fig. 1** (Case 1). A 48-year-old man complained of neck pain that had persisted for 2 years. He experienced right arm weakness for 1 year and weakness in both lower extremities for 3 months. His VAS score was 6, and his JOA score was 8. **a** T1-weighted sagittal magnetic resonance imaging (MRI) scan. **b** Preoperative axial MRI scan at the C2 level. **c** Preoperative axial MRI scan at the C3 level. **b, c** Demonstrate compression of the spinal cord. **d** Coronal reconstructive computed

tomography (CT) scan showing C2–3 intervertebral foramen widening and osteolytic destruction of C3 vertebral body. **e** Preoperative axial CT scan at the C3 level. He had total tumor removal and reconstruction with by a two-stage posterior and anterior approach. **f** Postoperative lateral radiograph. **g** Axial MRI scan at the 12-year follow-up. **h**, T2-weighted sagittal MRI scan at 12-year follow-up showing no recurrence



**Fig. 2** (Case 3). A lesion was accidentally revealed in an asymptomatic 50-year-old man. **a** Axial CT scan showing osteolytic bony destruction of L5. **b**, **c** Axial and sagittal T2-weighted MRI. The

patient had subtotal resection and pedicle screw fixation. **d** Postoperative CT scan. **e**, **f**, CT scan showing no recurrence at the 11-year follow-up

preferred for thoracic and lumbar lesions. While in the cervical lesion, surgical approach was decided by tumor location and usually combined approaches were required [4]. Instrumentation with fusion was carried out for spinal instability after extensive tumor resection.

### Functional outcome measures

Pain was evaluated by visual analog scale (VAS), and neurological deficit was evaluated using the American Spinal Injury Association (ASIA) impairment scale.

### Search strategy and selection of studies

We reviewed the spinal IOS cases published between January 1960 and December 2017 in the PubMed, EBSCO, Ovid, and Springer databases. In addition, the reference section of each paper was reviewed to identify additional cases. The inclusion and exclusion criteria were the same as those listed above. Cases with insufficient clinical information or unclear images were excluded.

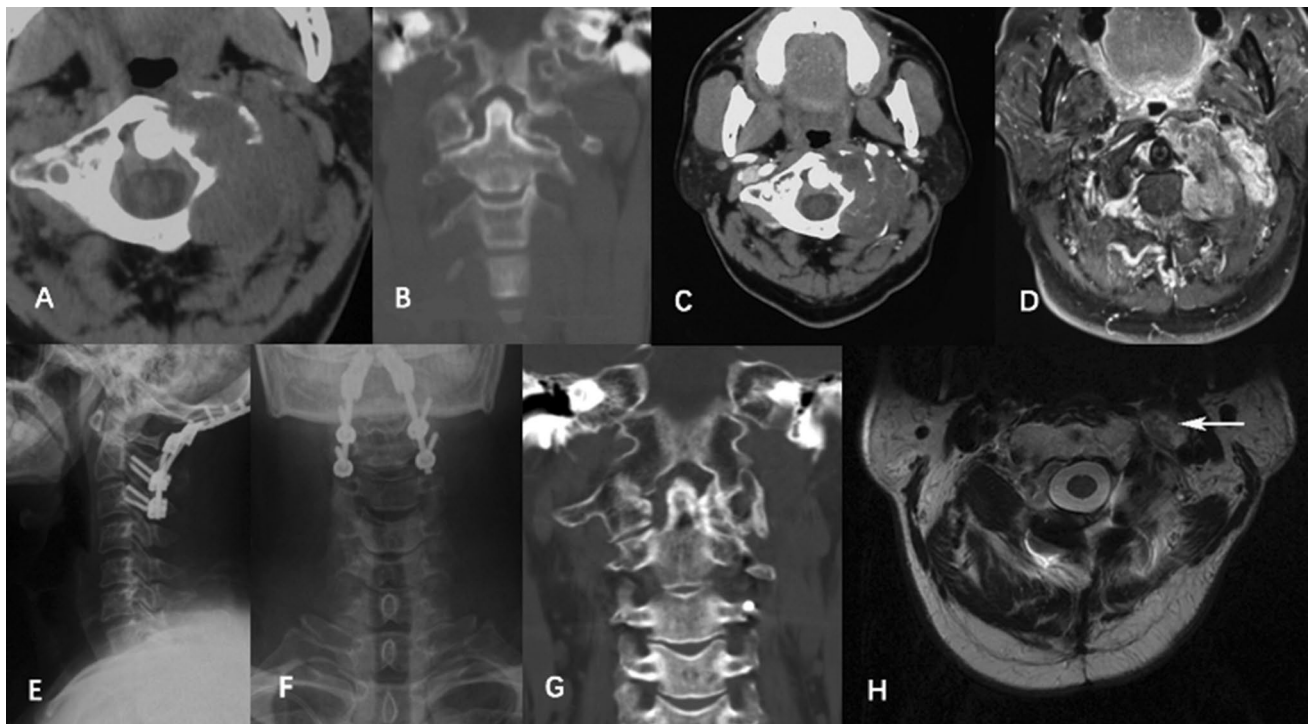
## Results

### Clinical presentations

Between January 2005 and November 2015, a total of 101 consecutive patients with primary benign spinal schwannoma involving the mobile spine underwent surgery in our orthopedic department. Definitive diagnosis was made by postoperative histological examination in all patients. Invasive and osteolytic bony destruction was noted in twenty-five patients, and five patients were lost to follow-up. Therefore, a total of twenty patients were included in this study.

The age of these patients at diagnosis ranged from 26 to 68 years (mean,  $43.3 \pm 11.1$  years); nine patients were male and eleven were female. The median duration of preoperative symptoms was 29 months (ranged from 20 days to 120 months). The most common symptoms were numbness ( $n = 13$ ), myelopathy ( $n = 10$ ), and local pain ( $n = 9$ ); three patients presented with radicular pain, four showed no significant symptoms before surgery, and one had an asymptomatic mass in the neck for 6 years (Table 1).





**Fig. 3** (Case 5). **a** A 46-year-old man presented with progressive neck pain with no neurological symptoms for 4 months. His VAS was 8. The patient underwent a two-stage operation. A posterior approach was carried out first with tumor removal, fusion, and occipitocervical fixation. However, only subtotal resection could be achieved during the lateral approach. **a** Axial computed tomography (CT) scan showing that the lesion extended from the left anterior arch, and the left lateral mass to the posterior arch. **b** Slightly collapsed left C1 lateral

mass on the coronal reconstructive CT. **c** Contrast CT scan showing that the left vertebral artery was entrapped by the tumor. **d** Gadolinium magnetic resonance imaging (MRI) scan demonstrating heterogeneous enhancement. **e, f** Postoperative lateral radiograph. **g** Coronal reconstructive CT scan showing healing of the lesion at the 8-year follow-up. **h** Axial MRI scan demonstrating residual tumor with little progress (arrow) at the 8-year follow-up

## Tumor location

The cervical spine was most commonly involved ( $n=12$ ), followed by the thoracic spine ( $n=6$ ) and lumbar spine ( $n=2$ ). In the axial plane, all tumors extended from layer A to layer D, as defined by the Weinstein–Boriani–Biagini classification (WBB, Table 1), while longitudinally, the tumors destroyed one bony segment in twelve cases, two segments in five cases, and three segments in three cases (Figs. 1, 2, and 3). However, sometimes the tumors have large extraspinal components, and the lesions were found to extend to two levels in six cases, three levels in two cases, and four levels in three cases.

## Imaging and biopsy

CT scan revealed osteolytic bone destruction in all cases, and pathological fracture was observed in six patients (Figs. 4, 5, and 6). On T1-weighted MRI, the average maximal diameter

of tumors was  $5.67 \pm 1.77$  cm. On T2-weighted MRI, the lobulated mass showed heterogeneous signal intensity with significant heterogeneous enhancement on contrast images. A total of fourteen patients underwent preoperative CT-guided trocar biopsy for suspected malignant tumors. All of them were diagnosed with schwannoma. The histological features of spinal IOS were the same as those of conventional schwannoma, except for blurred bony margins. In the beginning, we prescribed a biopsy for each IOS lesion, but after we gained some experience with IOS cases, we did not perform a biopsy of the large lesions in patients with minor symptoms and slow progression since these indicate benign tumors.

## Surgical outcomes and complications

One-stage single posterior approach operation was performed in eleven patients, one-stage single anterior approach in five, and combined approaches in four patients (Table 1). The average operation time was  $238 \pm 93$  min (range



**Fig. 4** (Case 6). **a** A 49-year-old woman complained of increasing back pain and weakness in the lower extremities during the last 6 months, her VAS score was 9, and the ASIA impairment scale was C. Total vertebrectomy was carried out with reconstruction for the tumor, destroying the major part of the T10 vertebral body. **a** Preoperative axial computed tomography (CT) scan showing that the lesion involv-

ing the vertebral body, vertebral foramen, and left vertebral appendix of T10. **b** Preoperative sagittal CT scan reveals osteolytic changes. **c** She had back pain at the 8-year follow-up; lateral radiography showing the rupturing of the connecting rods. **d** Sagittal CT scan showing the collapse of titanium mesh and solid fusion of T10–T11 levels. **e** CT scan showing no sign of recurrence at the 8-year follow-up

109–405 min), and the average estimated blood loss (EBL) was 1275 mL (range 100–4500 mL). Gross total resection of the tumor was achieved in seventeen cases and subtotal resection in three cases. After tumor removal, all cases had fusion with instrumentation. Case 9 had a vertebral artery injury, which was successfully controlled by ligation. A revision surgery was conducted in Case 6 (Fig. 4) because of instrumentation failure at the 8-year follow-up. Other complications included cerebrospinal fluid leakage in four cases, pleural effusion in two, and pleural rupture in one, all of which were successfully treated conservatively.

Resection of tumor-involved nerve roots was performed to decrease local recurrence in fourteen cases, and postoperatively, one patient had further radicular pain and another

had worsened numbness, but all of them recovered at the third-month follow-up.

### Follow-up

Patients had an average follow-up of  $69.5 \pm 35.2$  months (range 25–139 months). All patients improved during the final follow-up. For the twelve patients with pain, the VAS score decreased from  $5.66 \pm 1.79$  (range 3–9) preoperatively to  $1.16 \pm 1.77$  (range 0–6). For the ten patients who had myelopathy, the ASIA impairment scale increased by an average of 1.1 grades (range 1–2).

After gross total resection, no local recurrence was observed at the final follow-up (Figs. 7, 8, and 9). For those subtotal resection cases, residual tumors were observed with



**Fig. 5** (Case 10). A 45-year-old woman presented with a 2-year history of progressive pain and numbness around the shoulder and upper extremities, and her VAS was 4. **a** Preoperative axial CT scan showing the osteolytic bone destruction of C6. **b** Sagittal CT scan showing the pathological fracture of C6. **c** The tumor was heterogeneous

signal intensity on T2-weighted MRI, and the left lobar thyroid was compressed by the tumor. The tumor was totally removed via anterior approach with fusion and stabilization. **d** The CT scan at 4-year follow-up showing no tumor recurrence

only little progression after 11, 8, and 4 years of follow-up, respectively.

### Published cases of spinal IOS

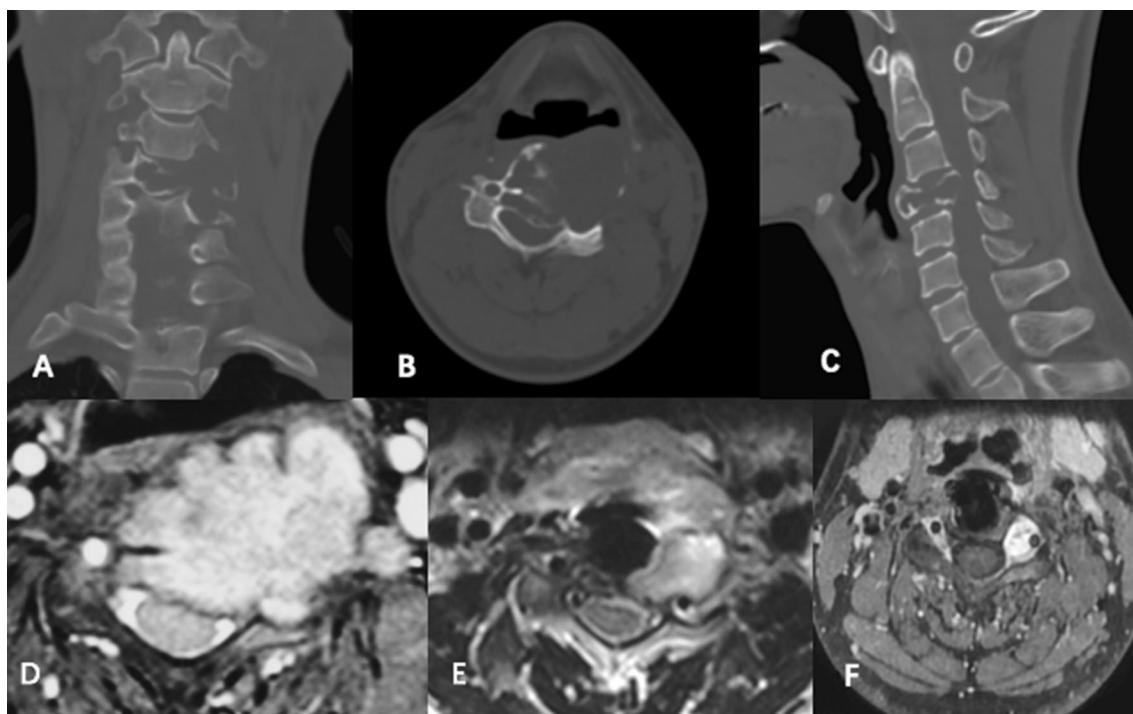
We retrieved twenty-four reported cases of spinal IOS from the currently available English literature since 1960 (Table 2). All studies were case reports. The mean age of the patients in the previous cases was  $43.0 \pm 14.9$  years. There were sixteen male and eight female patients. The tumors were equally distributed in the cervical spine ( $n=9$ ), thoracic spine ( $n=7$ ), and lumbar spine ( $n=8$ ). In these surgical series, the authors used different approaches to achieve total resection ( $n=21$ ), but most of them did not specify the type of tumor resection, and total resection included both en bloc and gross total resection. These studies had an average

follow-up of  $19.9 \pm 14.4$  months (range 1–60 months) and most authors reported no recurrence at the final follow-up, except for one at 2 years' follow-up [6].

### Discussion

#### Definition

In the present study, IOS is defined as a tumor causing invasive and osteolytic destruction of the spine, including small but deep nidus, regardless of the size of the extraosseous portion (PUTH classification Section V). All of our cases were primary spinal tumors with invasive and osteolytic bone destruction. The tumor was originally arising from the abutting nerve root and invading the vertebrae,



**Fig. 6** (Case 11). **a** A 29-year-old man complained of neck pain and numbness of the upper extremities after a football match, and his VAS was 3. We could only get subtotal resection with fusion by anterior approach because of its close relation to the vertebral artery. **a** Preoperative coronal CT scan showing a large lytic lesion involving C3–C5. **b** Axial CT scan at the C4 level. **c** Sagittal CT scan showing

the pathological fracture of C4. **d** Preoperative T2-weighted imaging showing the tumor involving the C4 vertebral body, and the left vertebral artery was entrapped by the tumor. **e** Postoperative MRI. **f** Axial MRI scan demonstrating residual tumor with no significant progress at the 4-year follow-up

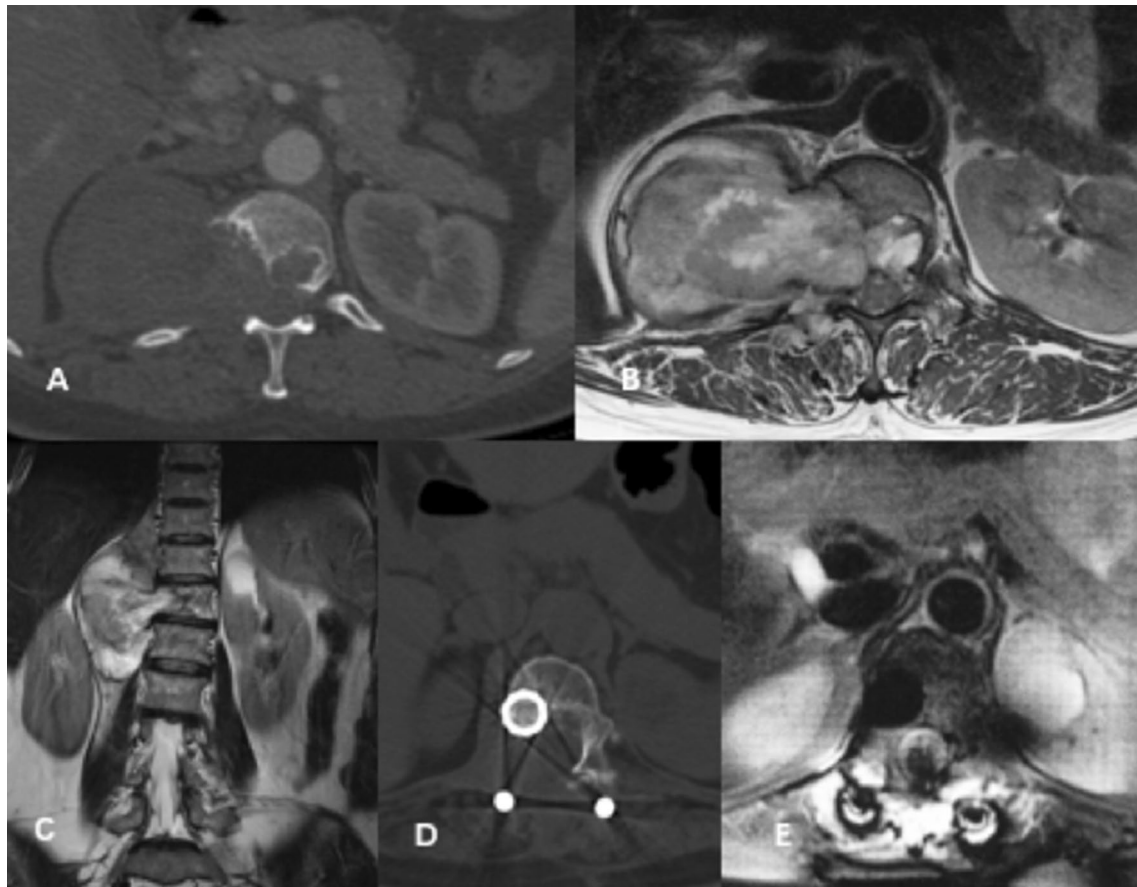
which suggests that the lesion of spinal IOS is probably intraosseous invasion of extraosseous nerve sheath tumor originating from the spinal nerve root. This hypothesis is supported by most reports on spinal IOS [1, 7, 8].

There is little consensus on the definition of IOS. Cohen et al. [9] first described eroding bony changes caused by neurilemmoma in 1964. Dickson et al. [10] first used the term IOS in 1971. In 2001, Inaoka et al. [7] defined IOS as “paravertebral neurinoma with aggressive intravertebral extension.” However, some surgeons preferred to define IOS as infiltrative and invasive changes of the vertebral body or posterior column [1, 10–14]. Sridhar et al. [3] defined “giant invasive spinal schwannoma” as a lesion that erodes the vertebral bodies and extends posteriorly and laterally into the myofascial planes, and is classified as Type V. However, their studies included neurofibromatosis and recurrent cases, leading to confusion about the definition.

## Clinical features

Schwannomas are the most frequent extramedullary, intradural spinal tumors, accounting for approximately 24% of all nerve sheath tumors in adults [15]. However, IOSs are relatively rare. Sridhar et al. [3] reported ten cases of giant invasive spinal schwannomas, which formed 10.9% of the cases of spinal neuromas treated in their center. Yu et al. [16] reported that the overall rate of giant invasive spinal schwannomas in their study was 4.9%. However, the overall rate of spine IOS in our series was 24.7%, significantly higher than that in these reports. There was a male predominance in previous literature, while there was no such predominance in our cohort. In previous reports, spinal IOS was equally distributed. As for the non-intraosseous spinal schwannoma series, the most common location was the lumbar region [15]. However, in our series, the cervical spine was the most commonly affected area. One





**Fig. 7** (Case 12). A 55-year-old woman presented with back pain, and her VAS score was 3. **a** Preoperative axial CT scan showing the osteolytic bone destruction of T12. **b**, **c**, Preoperative axial and coronal MRI scan showing a huge paraspinal mass spanning more than four

vertebral bodies. The tumor was totally removed with reconstruction via posterior approach. **d**, **e**, Postoperative CT scan and MRI showing no recurrence at the 5-year follow-up

possible reason for the differences is that our institute is a tertiary referral center for cervical spine diseases.

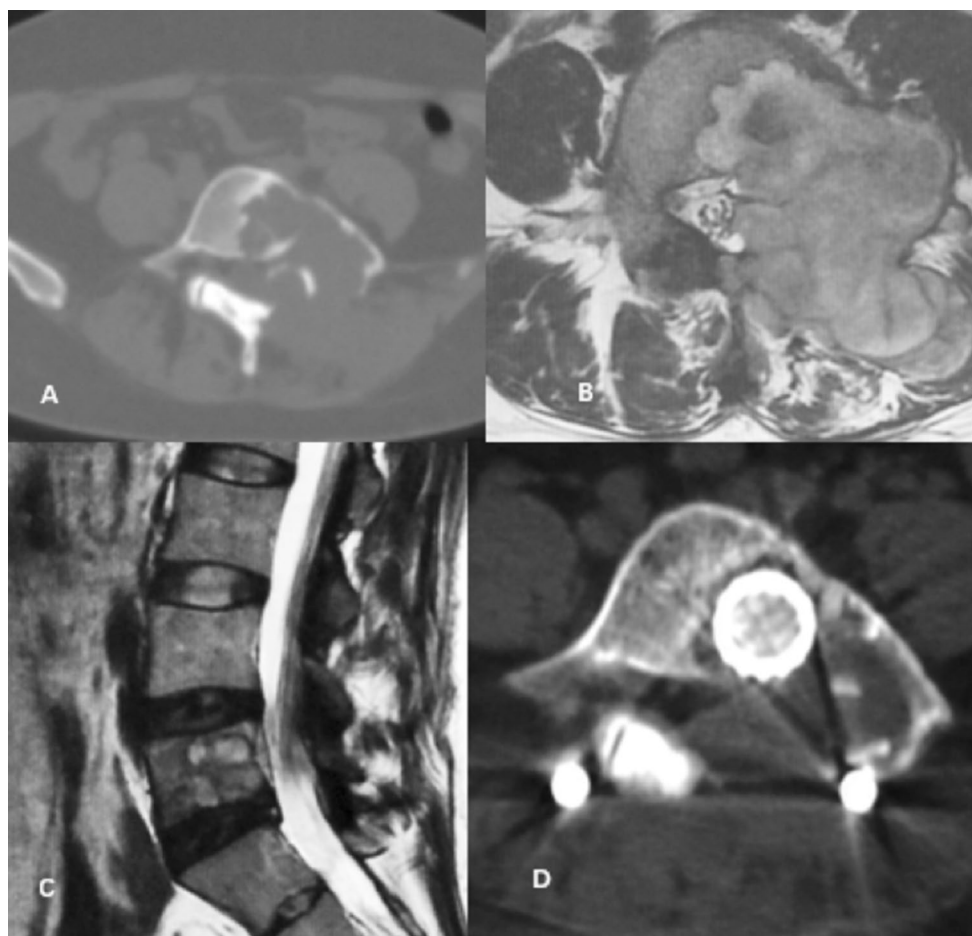
Conventional schwannomas can remain asymptomatic for a long time due to slow tumor growth. The most common symptom is local pain followed by neurological symptoms [17]. IOS could also present with insidious features. Motor weakness may not be obvious until the huge lesion leads to severe neurological compression.

It is difficult to distinguish spinal IOS from other spinal tumors only by imaging. Conventional schwannomas usually have sclerotic and clear margins (compressive change). And the features of IOS on CT often consist of expansile osteolytic lesions without clear margins with/without pathological fractures [18], which could be misdiagnosed as malignant spinal tumors. To differentiate malignant and benign spinal dumbbell tumors, Matsumoto et al. [19] proposed

a dumbbell scoring system based on preoperative images only. A score of more than three points leads to suspicion for malignancy. However, they also pointed out that invasive (intraosseous) schwannoma might have high scores and pathological examination would be essential for preoperative diagnosis.

### Surgical strategies

En bloc resection is recommended usually for conventional spinal schwannomas to prevent recurrence. Fehlings et al. [25] reported a retrospective multicenter study of 169 cases where the recurrence rate in intralesional resection cases was four times higher than in en bloc resection cases. In a retrospective study, Li et al. [20] also stated that total resection could significantly improve the local relapse-free



**Fig. 8** (Case 13). A 36-year-old woman complained of intermittent low back pain and numbness of the left lower extremities. **a** Preoperative axial CT scan showing revealed a lytic lesion in the left vertebral body and lamina of L5. **b, c**, Preoperative axial and sagittal

T2-weighted MRI showing heterogeneous signal intensity of the tumor. The tumor was totally removed and reconstructed via posterior approach. **d** CT scan showing no recurrence at the 4-year follow-up

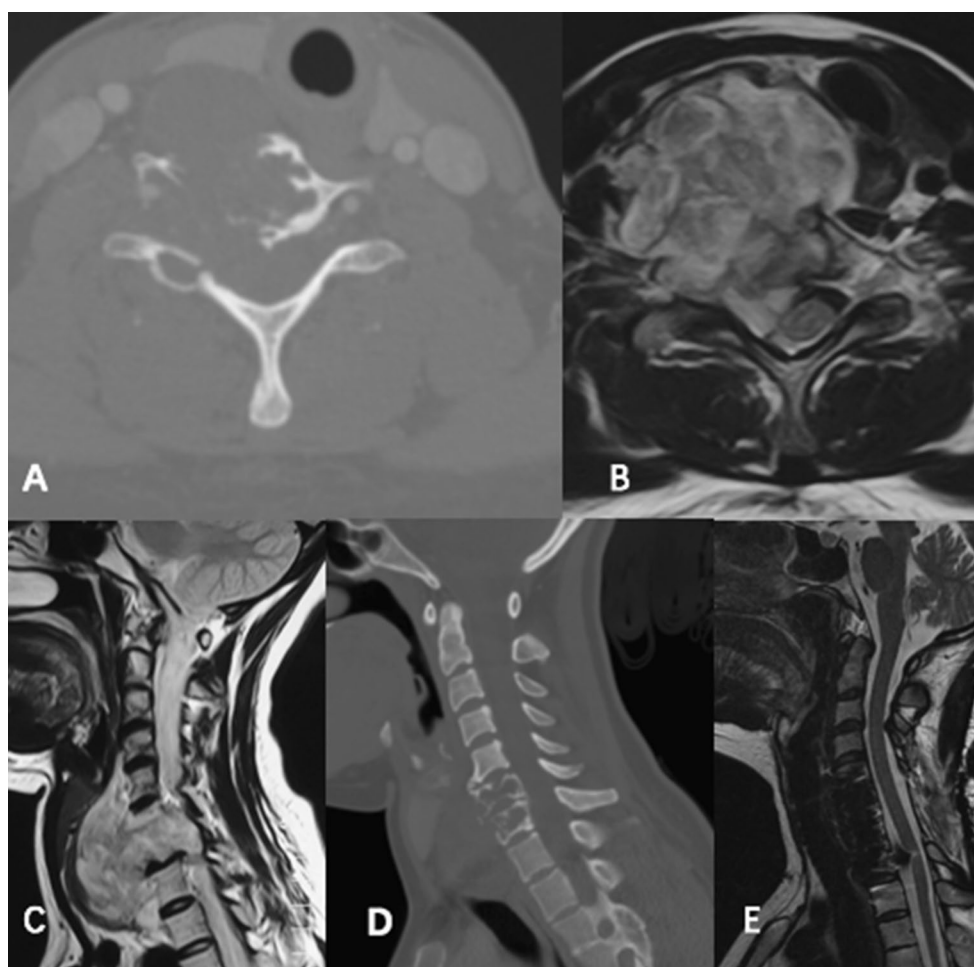
survival; the study included 169 patients (including 12 with malignancies) with cervical spinal nerve sheath tumors, with mean follow-up period of 72.1 months (median 62.0, range 1–158).

However, en bloc resection sometimes may be too dangerous, as when paraspinal lesions extend to adjacent critical structures, such as the vertebral artery or the abdominal aorta. Osteolytic bony lesions could be removed by either en bloc resection or extensive curettage. With IOS being benign in nature, we recommend extensive curettage followed by the use of a burr to remove the residual lesion in the niches.

Complete tumor resection for IOS often requires nerve root removal because of its involvement with the tumor [21]. According to Kim et al. [22], 23% of the total resection of

schwannomas with functionally important nerve roots resulted in the development of neurological symptoms. In the present study, depending on the feasibility, we tried to peel off the tumor with the capsule from the involved nerve root, and if it was not feasible, the involved nerve root was resected; the latter course was followed in fourteen cases, of which, two patients developed numbness and radicular pain, which resolved in 3 months.

To achieve complete resection, an appropriate approach is essential. Sridhar [3] used posterolateral approach and reported that they could safely separate the tumor from the cord. George [23] concluded that complete removal and best neurological recovery were achieved using posterolateral or anterolateral approaches. The posterior midline approach is usually ideal for instrumentation, and the lateral approach



**Fig. 9** (Case 15). A 34-year-old woman had progressive bilateral pain and numbness around the shoulder and extremities in the last 3 months, and her VAS score was 6. **a** Preoperative axial CT scan showing the osteolytic bone destruction. **b** Preoperative axial MRI scan showing the lesion has a huge exophytic component. **c** Sagittal

tal MRI scan showing heterogeneous signal intensity of the tumor. **d** Sagittal CT scan showing pathological fracture of C6 and C7. The tumor was totally removed and reconstructed by two-stage posterior and anterior approach. **e** MRI showing no sign of recurrence at the 4-year follow-up

facilitates the dissection of tumors [24] because most IOS cases involve unilateral bony elements of the vertebrae. In our cases, the choice of surgical approach depended on personalized factors; for the cervical spinal IOS, the surgical approach was strictly based on the PUTH classification [4], and posterior approach was usually recommended for thoracic or lumbar IOS.

### Prognosis

Though the treatment of spinal IOS is more difficult than the conventional spinal schwannoma, there is no significant difference in the prognosis [20]. According to Fehlings et al. [25], the overall rate of local recurrence after en bloc resection of conventional spinal schwannoma was approximately 5% after 1.7 years postoperatively.

Klekamp and Samii noted a recurrence rate of 10.7% at 5 years and 28.2% after 10 to 15 years in patients with spinal nerve sheath tumors [26]. In our series, recurrence was not observed in the average 5-year follow-up period; perhaps, the prolonged follow-up and larger sample size will help improve the accuracy of this conclusion. In previously reported cases of spinal IOS, only Peng et al. [6] reported a case of the recurrence and malignant transformation of spinal IOS after subtotal resection. Some authors reported that younger patients or those having lesions with higher Ki-67 index ( $> 2\%$ ) were more likely to have a high risk of recurrence [6, 16]. Therefore, regular follow-up is very important for patients with high risk of recurrence.

In conclusion, CT-guided biopsy is effective for preoperative diagnosis of spinal IOS. Total resection is the

**Table 2** Previously published reports of IOS in the mobile spine

| No. | Author/year     | Gender/<br>age | Symptom                            | Level | Approach                  | Stabilization          | Resection | F/U<br>(mos) | Final follow-<br>up |
|-----|-----------------|----------------|------------------------------------|-------|---------------------------|------------------------|-----------|--------------|---------------------|
| 1   | Dickson/1971    | F/51           | Pain in bilateral lower limbs      | L3    | Abdominal approach        | Iliac crest bone graft | Total     | 24           | No recurrence       |
| 2   | Polkey/1975     | F/34           | Pain and weakness                  | C6–C7 | Posterior                 | Iliac crest bone graft | Total     | 4            | No recurrence       |
| 3   | Naidu/1988      | M/50           | Weakness and burning sensation     | C3–C4 | Posterior                 | No                     | Total     | 6            | No recurrence       |
| 4   | Nooraie/1997    | M/46           | Asymptomatic                       | T12   | Posterior                 | Fusion + fixation      | Total     | 18           | No recurrence       |
| 5   | Chang/1998      | M/58           | Pain and numbness                  | L4–L5 | Anterolateral + posterior | Fusion + fixation      | Total     | 1            | –                   |
| 6   | Schreuder/2001  | F/38           | Local pain, paresthesia, dysphagia | C6    | Anterior                  | Fusion + fixation      | Total     | 48           | No recurrence       |
| 7   | Ramasamy/2001   | M/37           | Local pain, weakness, numbness     | T12   | Anterior + posterior      | Fixation               | Total     | 18           | No recurrence       |
| 8   | Inaoka/2001     | M/9            | Distention of the back             | T10   | –                         | –                      | Total     | 12           | No recurrence       |
| 9   | Inaoka/2001     | M/39           | Local pain                         | L5    | –                         | –                      | Total     | 36           | No recurrence       |
| 10  | Nannapaneni     | M/42           | Asymptomatic                       | C5    | Anterior                  | Fusion + fixation      | Total     | 18           | No recurrence       |
| 11  | Singrakhia/2005 | M/43           | Numbness, weakness                 | C3–C4 | Anterior                  | Fusion + fixation      | Subtotal  | 12           | Residual            |
| 12  | Singrakhia/2006 | M/45           | Radicular pain, weakness           | C3    | Anterior                  | –                      | Subtotal  | 12           | Residual            |
| 13  | Gupta/2005      | F/30           | Local pain, weakness               | L2    | –                         | Iliac crest bone graft | Total     | 6            | No recurrence       |
| 14  | Choudry/2007    | M/18           | Local pain, thinning of the leg    | T12   | –                         | Fusion + fixation      | Total     | 60           | No recurrence       |
| 15  | Park/2009       | F/46           | Local pain                         | L4    | –                         | Fusion + fixation      | Total     | 21           | No recurrence       |
| 16  | Cetinkal/2009   | F/55           | Local pain, numbness               | T12   | Posterior                 | No                     | Total     | 12           | No recurrence       |
| 17  | Mizutani/2010   | F/44           | Paresthesia                        | C4    | Anterior                  | No                     | Total     | 17           | No recurrence       |
| 18  | Kojima/2011     | M/60           | Local pain, weakness, numbness     | T9    | Posterior                 | Pedicle screw fixation | Total     | 24           | No recurrence       |
| 19  | Peng/2011       | M.44           | Weakness                           | C3    | Posterior + anterolateral | Fusion + fixation      | Subtotal  | 24           | Recurrence          |
| 20  | Youn/2012       | M/65           | Local pain, numbness               | L2    | Posterior                 | Fusion + fixation      | Total     | 12           | No recurrence       |
| 21  | Zhang/2012      | M/71           | Radicular pain, weakness           | L4    | Posterior                 | Fusion + fixation      | Total     | 24           | No recurrence       |
| 22  | Zhang/2012      | F/54           | Gait disturbance, paresthesia      | T9    | Posterior                 | Fusion + fixation      | Total     | 48           | No recurrence       |
| 23  | Mohanty/2013    | M/10           | Mass, dysphagia                    | C4    | Anterior                  | Fusion + fixation      | Total     | 10           | No recurrence       |
| 24  | Song/2014       | M/44           | Radicular pain                     | L5    | Anterior                  | Fusion + fixation      | Total     | 12           | No recurrence       |

C, cervical; T, thoracic; L, lumbar; F/U, follow-up; mos, months

optimal treatment for IOS, whereas subtotal resection could be an alternative choice for high-risk cases.

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### Compliance with ethical standards

**Conflict of interest** There is no other conflict of interest.

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