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CASE REPORT

Recurrent adamantinoma in the thoracolumbar spine successfully treated by three-level total en bloc spondylectomy by a single posterior approach

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Abstract
Purpose Adamantinoma is a low-grade primary malignant bone tumour with slow growth and local recurrence. Its occurrence in the spine is extremely rare, particularly with multilevel involvement. This paper wants to present the first case involving a patient with recurrent thoracolumbar spinal adamantinoma, who underwent a successful three-level spondylectomy for en bloc resection.
Methods A 24-year-old man with osteolytic masses of T11 and T12 vertebral bodies was performed curettage by a posterior approach in 2008. The pathology report showed the excised neoplasm was a rare adamantinoma. This patient underwent a tumorectomy again because of its local recurrence nearly 3 years later. In 2012, it was unfortunately revealed that the excised tumour had relapsed and had spread to the L1 vertebral body. Due to its repeated recurrence and aggressive lesion, total en bloc spondylectomy (TES) for this malignant tumour was thought to be the best option for preventing repeated recurrence and possible cure. TES for T11–L1 thoracolumbar spine was performed and spinal reconstruction was completed with instrumentation and a titanium mesh cage through a one-stage single posterior approach.
Results After three-level TES, neurological deficits of the patient demonstrated good recovery and no evidence of adamantinoma recurrence or deformity was found at 2-year follow-up.
Conclusions This is the first case involving multilevel thoracolumbar spinal adamantinoma with repeated recurrence to be successfully treated by three-level TES by a single posterior approach.

Keywords Adamantinoma · Total en bloc spondylectomy · Thoracolumbar spine · Tumour · Local recurrence

Introduction

Adamantinoma is a rare low-grade malignant primary bone tumour with slow growth and accounts for <1 % of all primary malignant bone tumours [1–3]. Adamantinomas occur almost exclusively in long bones, with >80 % in the tibia [2–4]. However, adamantinoma involving the spine is extremely rare with six cases reported so far [3, 5, 6]. There are only two cases of primary spinal adamantinoma [5, 6]. To the best of our knowledge, the case presented here is the third case of primary spinal adamantinoma. More importantly, it is the first case of three-level thoracolumbar spinal adamantinoma with repeated recurrence.

Because of the rarity of adamantinoma, there is insufficient clinical data regarding the safest and most effective treatment. Although experience is limited with these tumours, chemotherapy and radiation therapy have both been shown to be ineffective [7]. Therefore, surgery is the treatment of choice for a spinal adamantinoma.

However, spinal tumours remain difficult to treat with curettage or intralesional resection because of the high risk

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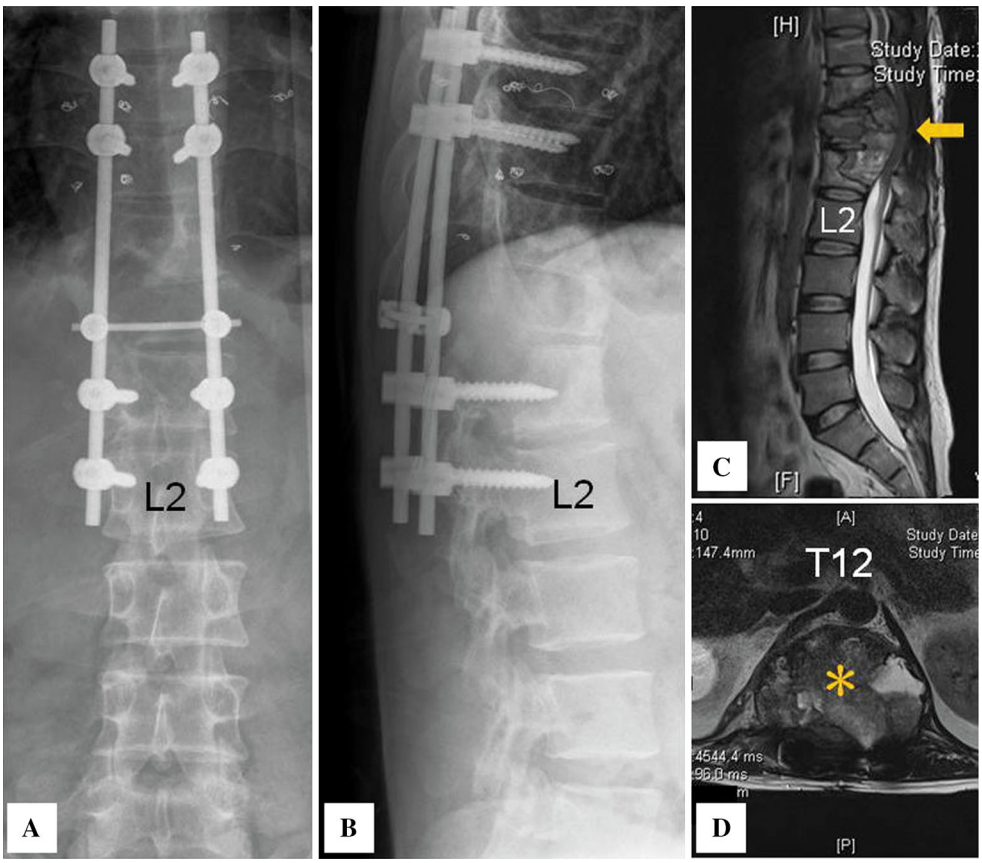

Fig. 1 **a** Anteroposterior and **b** lateral radiographs after the second operation. **c** Sagittal T2-weighted and **d** axial T2-weighted magnetic resonance imaging (MRI) images showing local recurrence of a thoracolumbar spinal adamantinoma involving T11–L1 vertebral bodies, as indicated by the *orange arrow* and *asterisk*

of local recurrence and metastatic progression [8]. By comparison, total en bloc spondylectomy (TES) generally reduces local tumour recurrence and is currently a widely accepted surgical procedure for spinal tumours [9–13]. Single-level TES is routinely performed, although there is little available data for multilevel resections owing to high risk of vital complications [10, 11, 14]. Because of the deranged anatomical structures and the topographic vicinity of the spinal cord and large vessels, revision surgery with TES for recurrent multi-segmental adamantinoma is technically demanding. This usually discourages spine surgeons from performing a revision surgery and ultimately leaves the patient with palliative treatment [11, 14].

Here, we report a case for which three-level TES was successfully performed for a recurrent thoracolumbar spinal adamantinoma by a single posterior approach. Furthermore, we have provided a review of literature.

Case report

A 24-year-old man was admitted to our service for the first time in May 2008. This patient reported progressive back pain with radiating pain in both legs for 1 year. On physical examination, a 4 × 3-cm indolent lump was found at the T12 level of his back, and the right ankle clonus was positive.

Plain X-ray, computed tomography (CT) and magnetic resonance imaging (MRI) scans showed partial osteolytic masses of the T11 and T12 vertebral bodies, which extended into their accessories. Surgery was performed through a posterior approach. A partially destroyed right vertebral body and spinous process were observed during this operation. A frozen section of a partially neoplastic specimen suggested that it had originated from ill-defined epithelial tissue. Therefore, the neoplasm was cut, and curettage was used to enlarge the sclerotic bone. Then, auto-iliac bone was implanted into the intervertebral bodies.

After this operation, the patient's symptoms were in remission, and he achieved good recovery. Three weeks after discharge, the pathology report showed that the excised neoplasm was a rare adamantinoma. Two weeks later, he was administered one course of radiotherapy with a radiation dose of 45 Gy at another hospital.

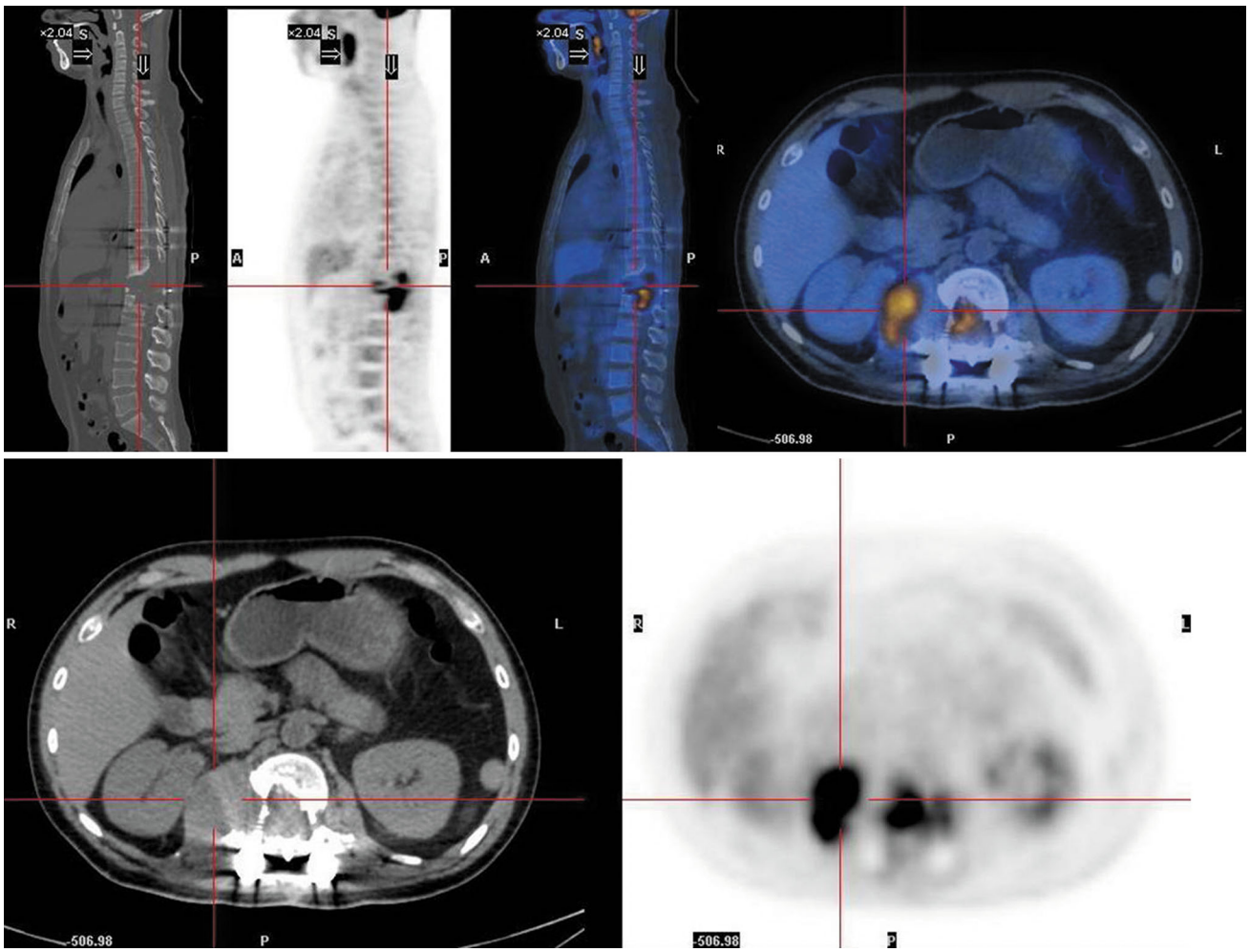

Fig. 2 Positron emission tomography–computed tomography (PET–CT) scan image showing local recurrence of a thoracolumbar spinal adamantinoma involving T11–L1 vertebral bodies, without other metastasis

In February 2011, a follow-up found that the tumour had recurred. This patient was admitted to another hospital and underwent a tumorectomy for the thoracic spine again by a posterior approach. The pathology report again showed that the tumour was adamantinoma. After this surgery, the patient recovered well and was ambulatory.

In February 2012, X-rays and MRI during a physical examination revealed that the excised tumour had relapsed and that it had spread to the L1 vertebral body (Fig. 1). Until June 2012, positron emission tomography (PET)–CT scans showed osteolytic destruction of the T11, T12 and L1 vertebral bodies and the formation of soft tissue masses beside the lesion vertebral bodies, without additional metastatic lesions (Fig. 2). This patient was finally referred to our hospital again for back pain and difficulty in standing and walking after sternutation.

To prevent repeated recurrence of this malignant tumour, TES for three levels of the thoracolumbar spine was performed through a single posterior approach using previously described methods [9, 12]. First, iliac crest bone harvesting was completed prior to the posterior exposure to avoid the risk of any tumour spread. This bone graft was primarily used for posterior fusion at the end of the procedure. Then, a 50-cm-long midline incision from T8 to L4 was made over the involved vertebral segment and the tumour mass. By careful dissection, lateral dissection was performed to expose the lamina of T8–L4. Pedicle screws in T9, T10 and L2 were then changed (Expedium DePuy, T9: 5.5 × 40 mm; T10: 6 × 40 mm and L2: 6 × 45 mm). Furthermore, screws were successfully placed into T8, L3 and L4 (T8: 5.5 × 35 mm; L3 and L4: 6 × 45 mm).

Then, the tumour mass at the L1 level infiltrating into the right paravertebral muscles (5 × 4 × 3 cm) was carefully exposed until it reached the lateral border to avoid any injury to the tumour capsule envelope. The 11th and 12th ribs were transected 4 cm lateral to the costotransverse joint. Inferior articular processes of T10 and superior articular processes of L2 were excised. After the lateral

border of the mass had been identified, gentle blunt dissection was performed to anterior vertebral bodies, and the great vessels were very carefully pulled away.

En bloc laminectomy was performed after T11–L1 pediculotomy using a stainless-steel threadwire saw (T-saw), and bone surfaces of the cut section were immediately sealed with bone wax. Segmental arteries and veins were identified and were bilaterally ligated. If ligation failed, bipolar coagulation was used to control haemorrhage. The bilateral intercostal nerve roots of T11 and T12 were cut, from which the affected vertebrae were planned to be removed by rotation.

A pair of long, homemade, S-shaped protective retractors was inserted with their anterior tips overlapping at the anterior aspect of the affected bodies. One posterior fixation rod was used on the right side to temporarily maintain stability before partially resecting the two discs. After the two discs were thoroughly cut from T10/11 to L1/2, the freed vertebral bodies of T11–L1 were rotated around the spinal cord and carefully removed from the right side (Fig. 3a). The margins of the resection were free of tumour. A total en bloc resection of the three-level vertebrae involved with tumour was achieved (Fig. 4). Another rod was used for fixation on the left side.

Subsequently, a titanium mesh cylinder (MOSS-Miami, DePuy Motech, Warsaw, IN) of an appropriate length was selected and filled with an autogenous bone graft from the iliac bone. The filled titanium mesh cylinder was placed in the middle of the cut space (Fig. 3c). After radiographically verifying the placement with a C-arm, the posterior instrumentation was slightly adjusted to compress the inserted mesh. Finally, after inserting a drain and two chest tubes because of pleural rupture, the wound was closed.

The overall surgery duration was 10 h, and the intraoperative liquid displacement amount was approximately 5,000 ml. However, only 1,800 ml of a homologous blood transfusion was used as a result of the use of intraoperative haemodilution and controlled hypotension anaesthesia. The patient was otherwise healthy. His haemoglobin was 154 g/l before surgery and 108 g/l on the first postoperative day. X-ray examination revealed a good implant position at 1 week after surgery (Fig. 5). He was instructed to wear a thoracolumbar brace for 3 months postoperatively.

Pathological diagnosis was the same as those of the previous two pathological results. Tumour specimens were stained with haematoxylin and eosin (H&E; Fig. 6). Immunohistochemical studies were positive for cytokeratins, keratin, vimentin and Ki-67 and negative for S100 (Fig. 6).

At 3 months postoperatively, X-rays and three-dimensional CT scanning were performed and showed a stable construct (Fig. 7), at which time the patient was ambulatory with a crutch. The patient could walk without a crutch after 6 months postoperatively. No local recurrent tumour was detected on MRI at the last follow-up at 2 years after surgery (Fig. 8d). X-rays and 3-d CT scanning showed the

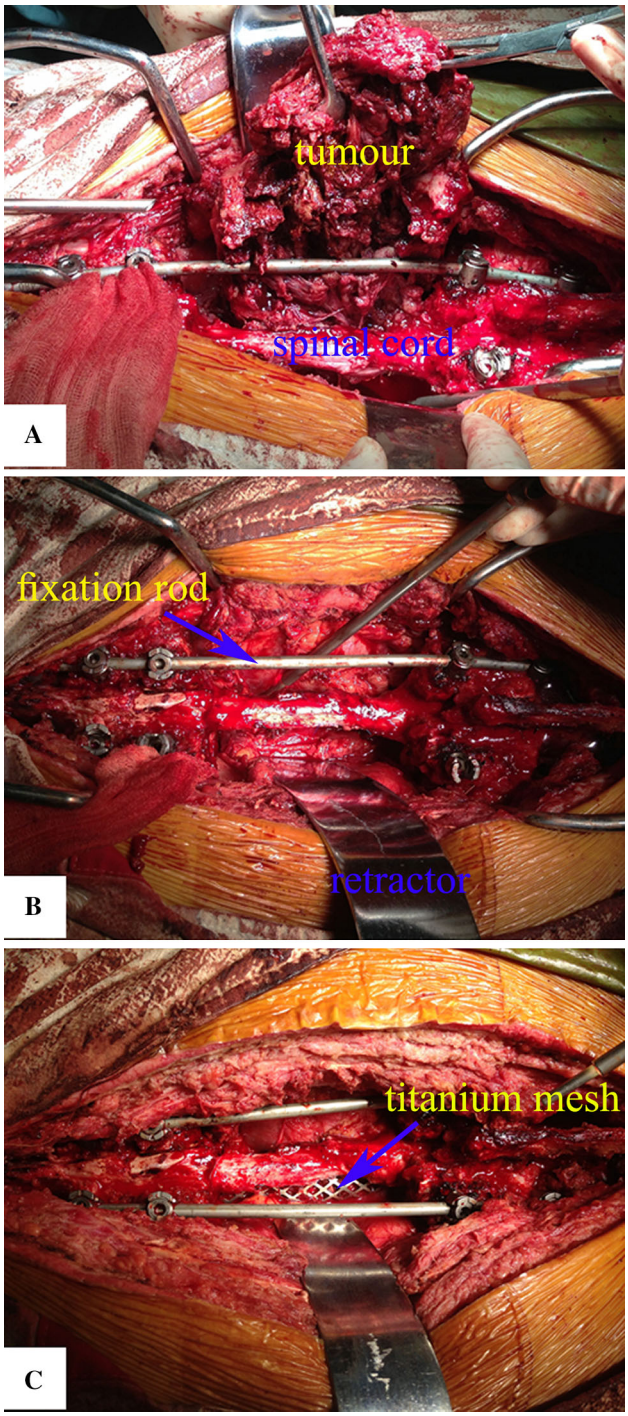

Fig. 3 Intraoperative photographs of en bloc spondylectomy through a single posterior approach. **a** Dissection and careful removal of a three-level thoracolumbar spinal adamantinoma from the right side, **b** after removing the spinal tumour and **c** a titanium mesh cylinder filled with an autogenous bone graft placed in the middle of the cut space

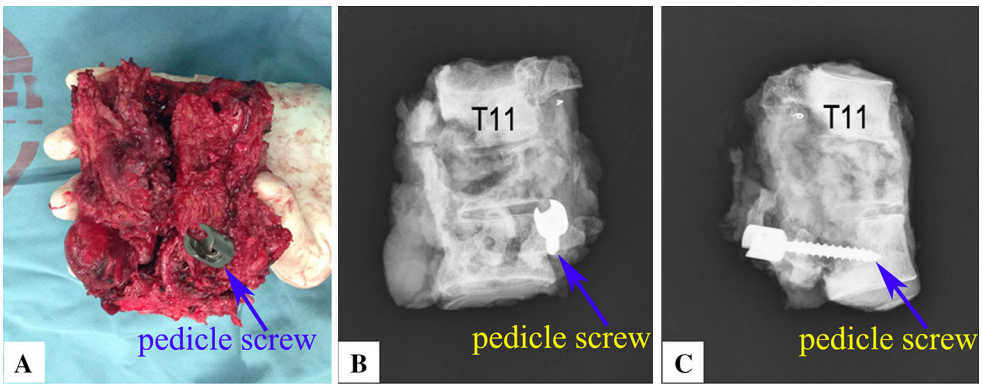

Fig. 4 **a** Specimen of the en bloc resected spinal adamantinoma involving T11, T12 and L1, with the right side pedicle screw of L1. **b** Anteroposterior and **c** lateral radiographs of the excised spinal adamantinoma

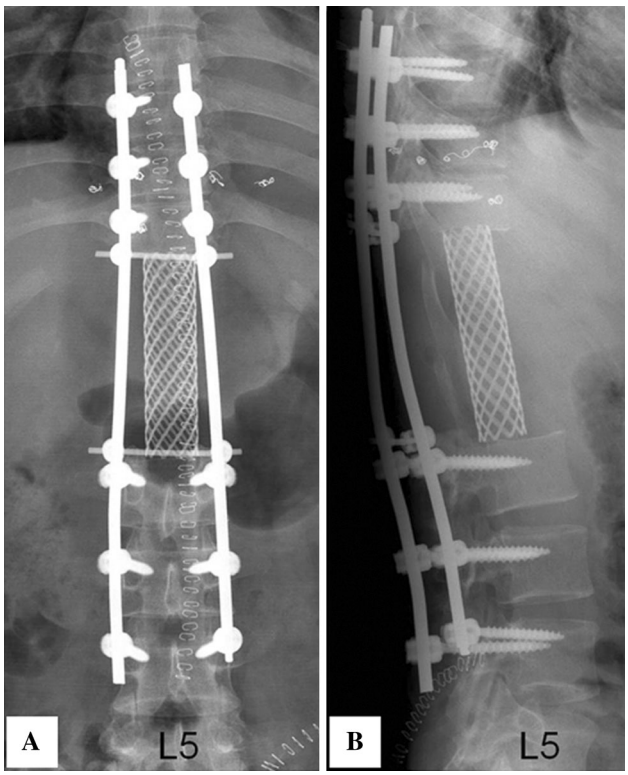

Fig. 5 **a** Anteroposterior and **b** lateral radiographs of the thoracolumbar spine showing a good implant position at 1 week after three-level total en bloc spondylectomy

biologic spinal reconstruction was achieved (Fig. 8a–c). The patient obtained a good functional outcome and had been going to work.

Discussion

Adamantinoma is an extremely rare, low-grade and slow-growing, primary bone tumour with a debated histogenesis. It is believed to account for <1 % of all malignant bone tumours [13, 15]. Epithelial, synovial and endothelial cells have been suggested as potential cells of origin [16]. According to reported data, this type of tumour tends to have a slight male predominance and is most commonly seen in the second and third decades of life [4, 15]. More than 80 % of adamantinomas appear in the tibia, with fibular involvement coexisting in 50 % of these cases, although there have been rare cases of adamantinomas reported in other bones [1, 5, 6, 15, 17]. Adamantinoma involving the spine was only two cases of primary spinal adamantinoma so far, which were the sacral and the cervicothoracic adamantinoma involving C6–T1 [5, 6]. The case reported by us is the third primary spinal adamantinoma and is also the first case involving a three-level thoracolumbar spinal adamantinoma with repeated recurrence.

In addition, histological diagnosis for adamantinoma is sophisticated, and it is difficult to distinguish adamantinoma from other tumours, like Ewing's sarcoma, synovial sarcoma, fibrosarcoma, metastatic carcinoma, fibrous cortical defect or osteofibrous dysplasia [16, 17].

For treating adamantinomas, chemotherapy and radiotherapy have extremely low success rates and are not recommended [7, 17]. Radiotherapy appeared to be ineffective for our patient after the first surgery in 2008. Therefore, the only treatment option for spinal adamantinoma is surgery. However, curettage and intralesional resection are inadequate for treating adamantinoma owing to the very high risk of local recurrence [8, 16]. Moreover, such an inadequate intervention may change the indolent nature of the lesion, causing it to behave in a more aggressive manner or have a risk of metastatic progression [1, 5, 17]. Although follow-up data have indicated a high rate of local recurrence (about 30 %) in the long bones [3], ascertaining accurate recurrence time is difficult because of the extremely rare nature of this tumour, especially in the spine with

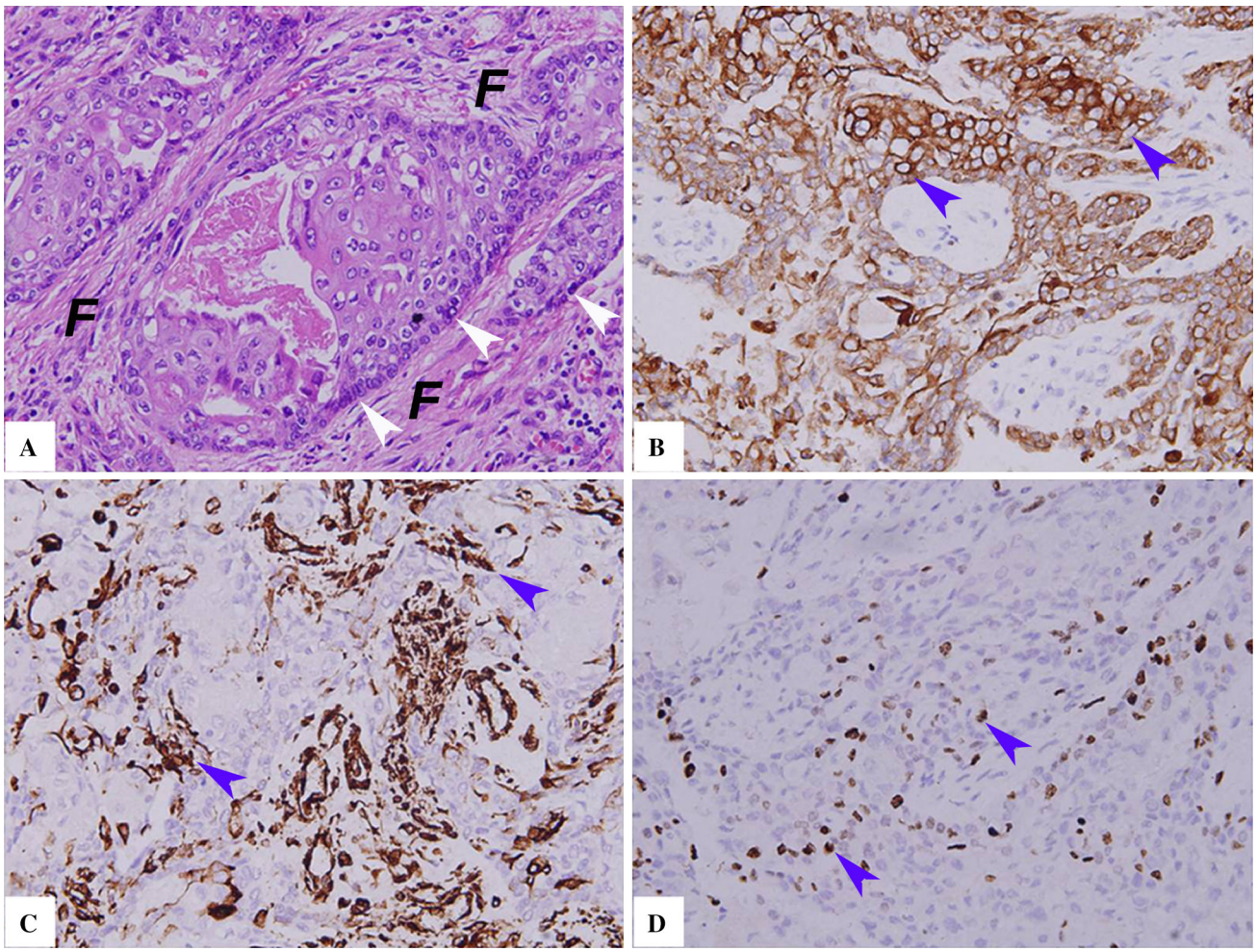

Fig. 6 **a** Photomicrograph showing solid nests of prominent epithelial islands (*white arrows*) embedded in a fibrous stroma [*F* fibrous stroma, haematoxylin and eosin (H&E) ×200]. Immunohistochemistry results for cytokeratin (**b**), vimentin (**c**) and Ki-67 (**d**) showing strong positive signals (*blue arrows*) within the cells of the adamantinoma (×200)

only six cases reported. Dini et al. [5] reported local recurrence in the cervicothoracic adamantinoma 1 year after corpectomy of C6–T1 through an anterior approach and resection of the posterior arch of C6–T1 by a posterior approach. The patient with primary sacral adamantinoma was observed to have a metastatic lesion in the humerus 1 year after resection of tumour by combined anterior and posterior approaches [6]. Similarly, a local repeated recurrence occurred in our case 1 year after the second surgery. In the light of the former twice recurrent time, we could deduce that TES for this case postpones effectively the recurrence of the tumour. Compared with curettage and intralesional resection, TES is currently a widely accepted surgical procedure for spinal tumours because it generally prevents local tumour recurrence and improves the survival rate [7, 9, 11, 15, 16]. Taken together, the result of 2-year follow-up after TES for our case seems to be satisfactory.

The TES procedure consists of en bloc laminectomy and en bloc corpectomy, followed by anterior instrumentation with spacer grafting and posterior spinal instrumentation [9, 12, 13]. This procedure is technically challenging, particularly for multi-segmental thoracolumbar spinal tumours because of the topographic vicinity of the spinal cord and large vessels.

Currently, en bloc resection of a mass is thought to be the best option for long-term local control and a possible cure. Single-level TES is routinely performed, whereas there have been only a few case reports of multi-segmental en bloc excisions [11, 14, 18–20]. Some multi-segmental en bloc resections were performed through multiple-stage combined approaches [11, 14, 20], whereas others were completed by a one-stage single posterior approach [18, 19]. The TES procedure allows both resection and stabilization through a single posterior approach [9, 12, 13, 21].

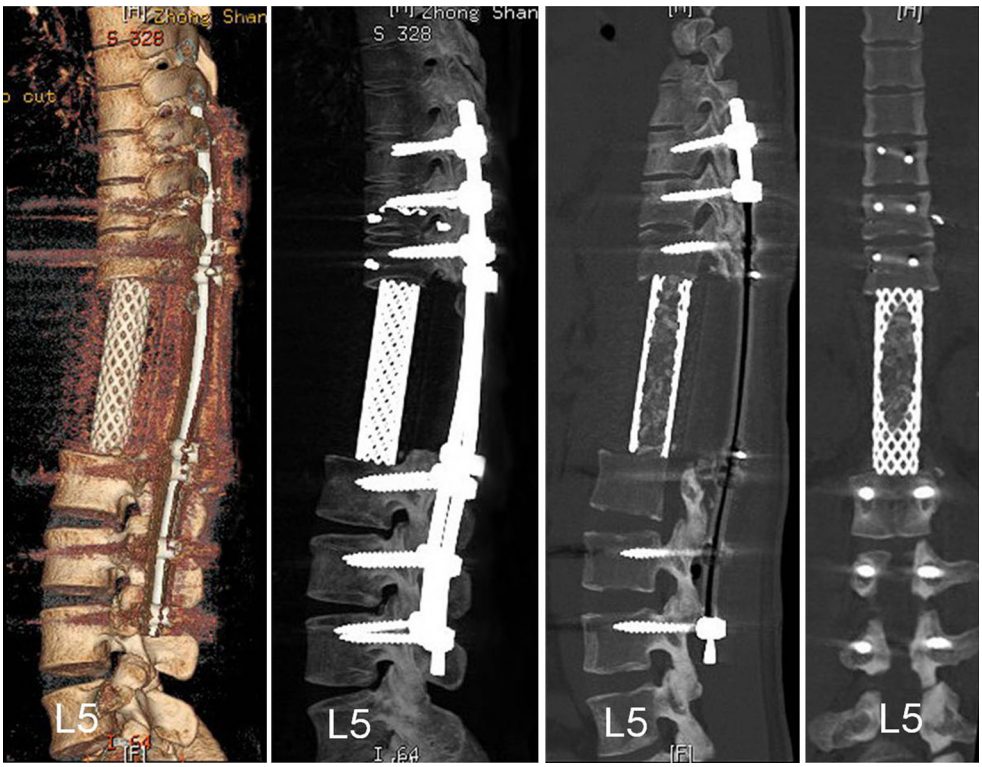

Fig. 7 Three-dimensional computed tomography (CT) scan images showing spinal reconstruction with an anterior titanium mesh filled with an autogenous bone graft for biologic fusion, and a stable construct at 3 months after en bloc spondylectomy

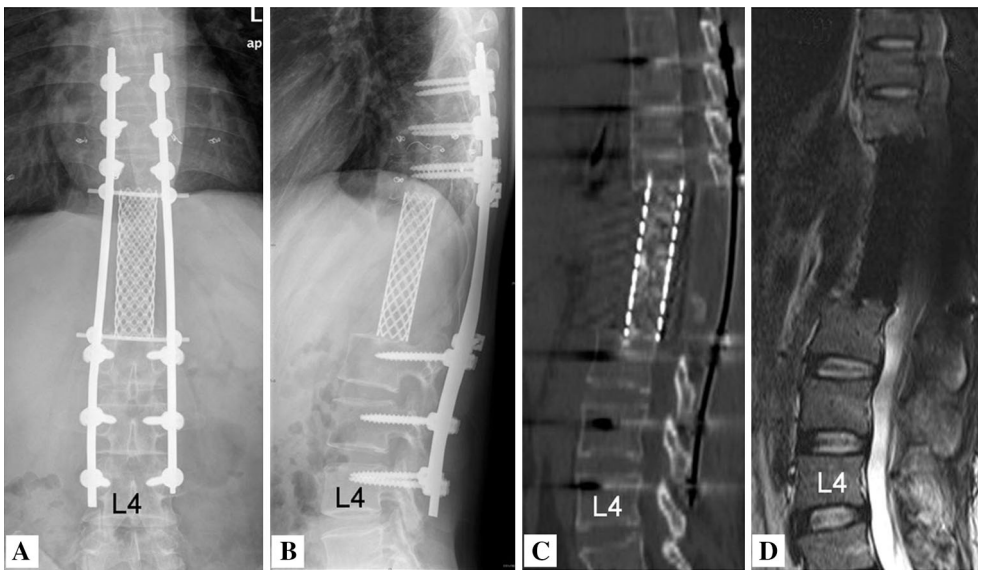

Fig. 8 **a** Anteroposterior, **b** lateral radiographs, **c** three-dimensional CT scan image and **d** MRI image of the thoracolumbar spine showing a stable construct, biological fusion and no local recurrence at 2 years after three-level total en bloc spondylectomy

Therefore, we performed three-level TES for this multi-segmental thoracolumbar spinal adamantinoma with repeated recurrence by a one-stage single posterior approach. Owing to the complicated anatomical structure, the inferior blood supply of the thoracic nervous tissue and serious tissue adhesions after repeated surgeries, revision surgery for recurrent multi-segmental thoracolumbar spinal tumours is technically demanding, which usually discourages spine surgeons and ultimately leaves the patient with palliative treatment [11, 14, 22].

As commonly suggested for TES, instrumentation placement is usually done early during the procedure because of the severely destabilizing effects of removing an entire vertebral body. Screw fixation of three adjacent

levels above and below is recommended [23]. For our case, posterior instrumentation was extended well above (T8) and below (L4) from the resected area of T11–L1, followed by an anterior titanium mesh cylinder filled with an autogenous bone graft. Biomechanical studies have already shown that post-implantation stability after TES is primarily influenced by the number of pedicle screws placed for posterior fixation, and the combination of an anterior titanium mesh and posterior instrumentation provide the best stability and rigidity [10, 23, 24].

Conclusion

Adamantinoma is a low-grade malignant primary bone tumour with slow growth and local recurrence. It is reported to be extremely rare in the spine, particularly with multilevel involvement. ES offers an oncologically en bloc resection option for this type of tumour. This is the first successful case report of a three-level spondylectomy for en bloc resection of a recurrent thoracolumbar spinal adamantinoma by a one-stage single posterior approach.

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Conflict of interest None of the authors has any potential conflict of interest.

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