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CASE REPORT

Solitary bone plasmacytoma of the upper cervical spine: A case report

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Author contributions: Li RJ, Li XF and Jiang WM participated in the diagnosis and performed the surgery; Li RJ wrote the manuscript; Li XF revised the manuscript; All authors read and approved the manuscript.

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Abstract

BACKGROUND

Solitary bone plasmacytoma (SBP) of the upper cervical spine is a rare diagnosis. The exact role of surgery for SBP remains unclear.

CASE SUMMARY

We present the first case of SBP of the C2. A 69-year-old Chinese woman presented with severe neck pain and limitation of rotative activity for 2 mo. She underwent anterior one-stage debridement combined with cement augmentation in the C2 to reconstruct stability of the spine. The patient did not receive postoperative radiotherapy. She now remains disease free with no neck pain or neurological deficit after follow-up of 3 years.

CONCLUSION

Anterior one-stage debridement combined with cement augmentation of the upper cervical spine may be an alternative treatment for SBP.

Key Words: Solitary bone plasmacytoma; Multiple myeloma; Cervical spine; Surgical treatment; Mini-invasive surgery; Case report

Core Tip: We present a case of solitary bone plasmacytoma on the C2 with follow-up of 3 years. The patient underwent anterior one-stage debridement combined with cement augmentation. The results of this case provide a choice for surgical treatment of solitary bone plasmacytoma. They also show that radical surgery without radiotherapy is a rational choice for treating solitary bone plasmacytoma with spinal instability.

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INTRODUCTION

Multiple myeloma (MM) presents as a malignant tumor with abnormal proliferation of plasma cells with major clinical manifestations of anemia, bone pain, hypercalcemia and renal insufficiency. When myeloma cells infiltrate the bone marrow, the balance of bone resorption and destruction is destroyed. It commonly affects the spine, presenting as either a solitary plasmacytoma or part of a systemic disease. The level of the cervical spine is the location of 10%-17% of neoplastic disease of the vertebral column, including MM[1]. Solitary bone plasmacytoma (SBP) is defined as a localized tumor in the bone comprised by a single clone of plasma cells. The diagnosis of SBP is currently based on histopathological examination through tissue biopsy and radiological confirmation[2]. SBP is a rare diagnosis for which the primary treatment is local radiotherapy. The exact role of surgery for SBP remains unclear[3-5]. Although MM is a common disease when compared to other plasma cell neoplasms, SBP in the vertebral body is a rare condition with little published literature[3]. Meanwhile, the upper cervical spine (C1/2) has a unique function and biomechanical and anatomical characteristics. Lesions in this region mainly present with pain from instability rather than neurological deficit from cord compression.

Here, we report a 69-year-old woman with SBP of the upper cervical spine to improve our understanding of the clinical manifestations and to report successful radical surgery of SBP.

CASE PRESENTATION

Chief complaints

A 69-year-old Chinese woman presented with severe neck pain and limitation of rotative activity for 2 mo.

History of present illness

The patient noticed intermittent neck discomfort 10 years ago, which improved after rest and became aggravated after activity. Two months ago, the neck pain aggravated suddenly.

History of past illness

The patient complained about neck discomfort for > 10 years but did not go to the hospital or take any medication. There was no significant change in body weight for 10 years.

Personal and family history

She denied any personal or family history of other diseases.

Physical examination

Physical examination revealed localized tenderness on the cervical spine. The strength of both upper limbs was normal, while the sensation of both hands decreased. No sensory loss of the lower extremities was noted, and muscular tension was normal. Pathological signs were negative, and no hyperactivity of tendon reflex was found. The spinal instability neoplastic score was 8.

Laboratory examinations

Hemoglobin, calcium and creatinine were normal. C2 biopsy showed multilobe mature plasma cells with no histopathology. No tumor cells were found in the bone marrow 1 year postoperatively (Figure 1).

Imaging examinations

X-rays showed mild cervical kyphosis and cystic lesions in (CK1) C2. Computed tomography (CT) reconstruction revealed bone destruction in the pedicle of C2 with

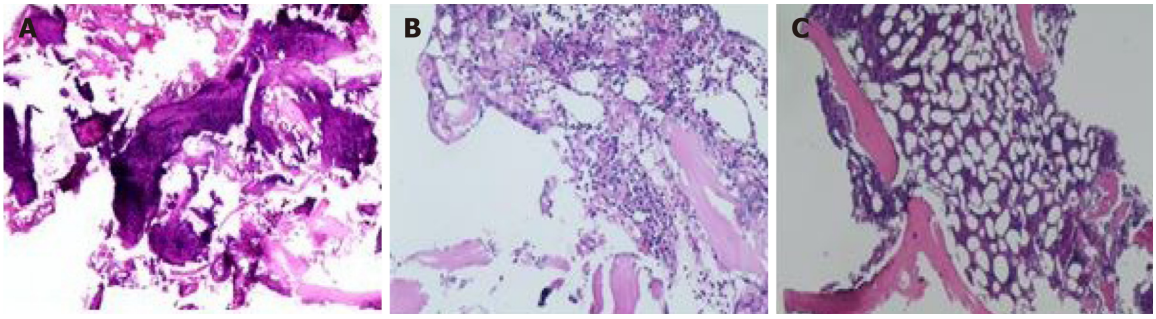

Figure 1 Laboratory examinations. A and B: C2 biopsy showed multilobe mature plasma cells with no histological pathologies; C: No tumor cells were found in the bone marrow 1 year postoperatively.

absence of the posterior wall. Magnetic resonance imaging showed a high signal mass on the right side of the C2 with clear margin. No lymph node metastasis was found in the magnetic resonance imaging. Cervical disc herniation could be found in C4-6, while no garrulous blade or swollen discs were observed toward the spinal cord in C2 (Figure 2 and 3). Single photon emission CT/CT showed high bright signal intensity in C2 and the teeth (Figure 4).

FINAL DIAGNOSIS

According to clinical symptoms, imaging findings and postoperative biopsy, SBP was diagnosed.

TREATMENT

CT revealed severe bone destruction in the (CK1) C2, causing instability of the cervical spine. The patient was asked to wear a neck brace and forbidden to get out of bed. She underwent anterior one-stage debridement combined with cement augmentation in C2 to reconstruct stability of the spine. Methylprednisolone was given intravenously in the first 3 d after surgery, and dexamethasone was used in the last 4 d. The patient was reviewed in the hematology department every 6 mo. This patient did not receive postoperative radiotherapy.

OUTCOME AND FOLLOW-UP

The cervical activity of the patient was restored to normal, and neck pain was significantly relieved. Postoperative X-rays and CT showed that the bone cement filled well, and no leakage occurred during the 3 years of follow up (Figure 5). X-rays showed good stability, and the patient remained disease free with no neurological deficit at 3 years postoperatively. The serum markers of MM were normal at the 3-year follow-up.

DISCUSSION

In patients with plasma cell dysplasia, SBP is a rare condition with a cumulative incidence of 5%[6]. As long as there is initial bone involvement, it can still be classified as SBP, even if soft tissue extension occurs[7]. SBP can be diagnosed by biopsy demonstrating monoclonal plasma cell infiltration of a single lesion. The median age at diagnosis of SBP is 10 years younger than that of MM[3]. To date, the treatment choices of tumors in the upper cervical spine are controversial, especially in terms of the surgical approaches, extent and reconstruction. The most common symptom may be pain, but other neurological symptoms may be caused by nerve compression[8-10].

Molloy *et al*[4] reported that treatment of SBP involving the spine is medical, including radiotherapy, chemotherapy or both modalities. However, instability is the

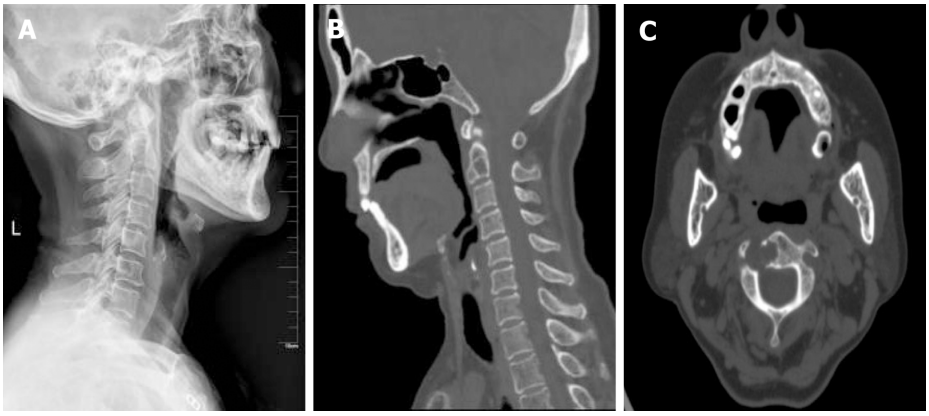

Figure 2 Sagittal X-ray. A: Mild cervical kyphosis and cystic lesions in the C2; B and C: Sagittal and plain computed tomography scan revealed bone destruction in the pedicle of C2. The posterior wall of the vertebrae was destroyed.

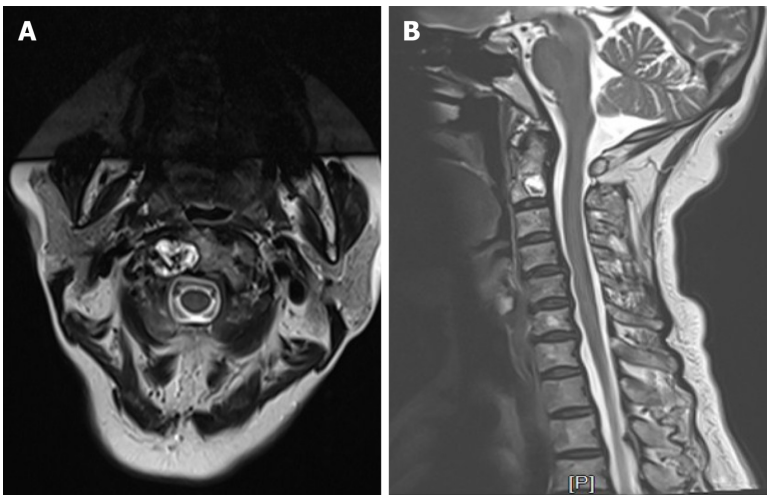

Figure 3 Magnetic resonance imaging. A: A high signal mass on the right side of C2 with an irregular boundary; B: Cervical disc herniation could be found in C4-6, while no garrulous blade or swollen discs were observed toward the spinal cord in C2.

main reason for pain, which can only be relieved completely by surgery. Yang *et al*[11] recommended aggressive resection of upper cervical spine tumor to reduce mortality. Huang *et al*[12] reported four cases of cervical SBP that showed unsatisfactory outcomes after surgical treatment due to spinal instability and neurological deficits in the cervical spine. They demonstrated that the vertebral body was reconstructed by a titanium mesh cage filled with iliac crest combined with an anterior titanium plate for lesions in the (CK1) C2. Von der Hoeh *et al*[3] reported that surgery combined with radiotherapy are necessary because of spinal instability in SBP. They demonstrated that total spondylectomy and postoperative radiotherapy minimized the risk of local recurrence and reduced the risk of conversion to MM. Molloy *et al*[4] argued that spinal stability was not an issue in SBP because the vertebral body had compensated for the osteolytic defects by thickening its cortices. Some researchers[13-15] believe that the posterior approach is preferred for this region, and lesions of C1 or C2 are needed to be fused into the occiput for the spinal stability (CK2)[3].

To date, the recommended treatment of SBP is local radiotherapy without surgery. However, CT reconstruction revealed bone destruction in the C2 (CK4) with absence of the posterior wall and involvement of the pedicle (CK5), and adjuvant therapy cannot maintain the spinal stability. Once neck trauma occurs, it can easily lead to cervical cord compression. Generally speaking, adjuvant therapy such as radiotherapy should be carried out after surgery rather than before. Von der Hoeh *et al*[3] reported a similar case in 2014 with the diagnosis of SBP with a 1 year history of low back pain. He chose to perform a total spondylectomy in the lumbar spine. Postoperative local radiotherapy was performed with the rationale of improving the prognosis regarding local recurrence and decreasing the risk of progression to MM. After 4 years of follow-up, laboratory and imaging diagnostics showed no pathologic recurrence. In the

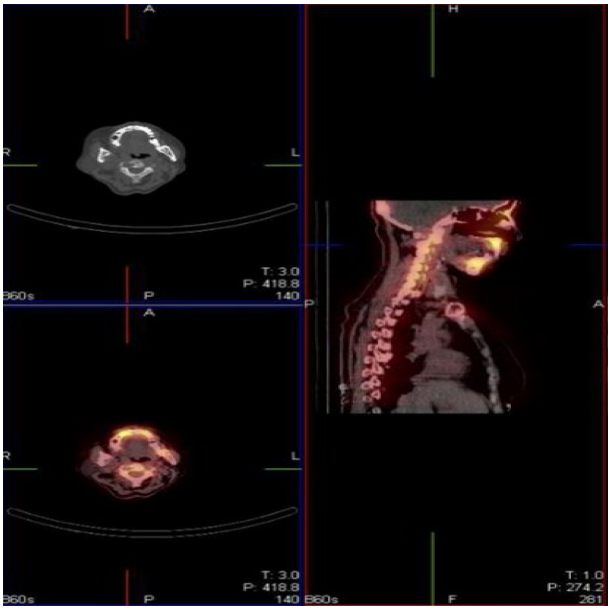

Figure 4 Single photon emission computed tomography/computed tomography showed high bright signal in the C2 and teeth.

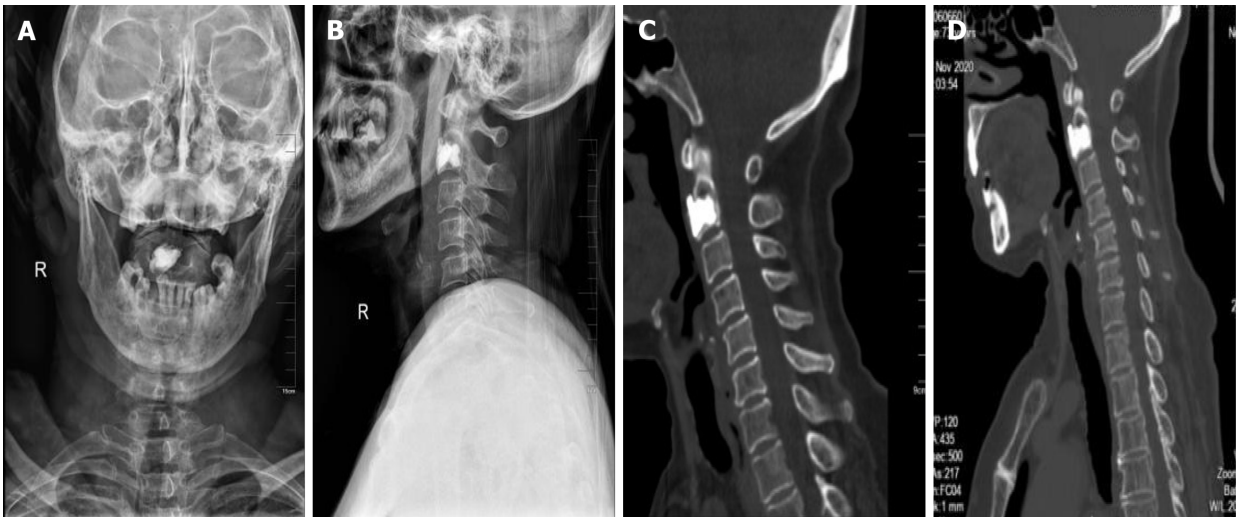

Figure 5 Postoperative X-rays and computed tomography showed that the bone cement filled well, and no leakage occurred during 3 years of follow-up. A and B: The anteroposterior and lateral X-ray was taken at 2 d after surgery; C and D: The two sagittal computed tomography scans were taken at 6 mo and 3 yr after surgery. They showed that no leakage of the bone cement occurred, and the shape of the C2 was maintained.

present case, the spinal instability neoplastic score was 8, and surgical intervention is recommended when the score ranges from 7 to 18[16]. The patient underwent anterior one-stage debridement combined with cement augmentation, with the advantages of less trauma and blood loss. A definitive surgical excision alone was considered because of the small size of the tumor and a clear margin. In addition, no lymph node metastases were found in the magnetic resonance imaging.

Meanwhile, the patient's own decision was necessary for the choice of treatment. After the surgery, the shape of the vertebral body and immediate spinal stability were restored by filling with bone cement. Temperature elevation caused by bone cement played a role in the destruction of tumor cells[17]. It can be seen that no leakage of bone cement occurred, and the shape of the C2 was maintained. Normal laboratory examinations indicated no pathological recurrence or progression to MM. This case provides evidence that surgical treatment of SBP can be successful and shows that radical surgery without radiotherapy is a rational choice for treating SBP with spinal instability. However, follow-up of 3 years is limited and further follow-up is needed.

CONCLUSION

Anterior one-stage debridement combined with cement augmentation without radiotherapy of the upper cervical spine may be an alternative choice for treatment of SBP.

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