De Novo Plasmacytoma at the Site of Previous Anterior Cervical Fusion

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BACKGROUND AND IMPORTANCE: Solitary plasmacytoma of bone (SPB) is a rare tumor that forms from monoclonal plasma cells within bone. These tumors are typically found in patients in their fifth to sixth decade of life and often progress to multiple myeloma. Previous reports have noted the formation of these tumors at the site of previous procedures or trauma, yet none have reported the development of SPB at the same site of a previous spinal surgery. **CLINICAL PRESENTATION:** The authors report on a 47-year-old woman who developed a de novo plasmacytoma 6 years after anterior cervical discectomy and fusion at the surgical site.

CONCLUSION: Although SPB is a rare tumor primarily seen in bone that has been unaffected by surgery, it should be included in the differential after finding a tumor at the site of a previous spine surgery. Furthermore, biopsy should be considered once a lesion is identified in the area of a previous spine surgery, as SPB may progress to multiple myeloma, resulting in worse outcomes.

KEY WORDS: Multiple myeloma, Solitary plasmacytoma

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Solitary plasmacytoma of bone (SPB) is a rare neoplasm arising from plasma cells within bone marrow. These tumors can be found in any bone but most commonly occur in the axial skeleton.¹ Incidence of SPB is 0.15 per 100 000 person-years and predominantly affects older patients, with median age at diagnosis between 55 and 66 years.²⁻⁵ SPB often progresses to multiple myeloma (MM), especially in older individuals.⁶ The 10- and 15-year rate of progression from SPB to MM is 65%–84% and 65%–100%, respectively.^{2,6}

Although vertebrae are the most common site for SPB, this is generally in the setting of a surgically naïve vertebral level.^{7,8} Here, we describe a case of de novo SPB that developed in a 47-year-old patient 5 years postcervical fusion to report a unique case and improve our understanding of a rare tumor.

CLINICAL PRESENTATION

This patient is a 47-year-old woman who presented in 2017 after feeling her neck "pop" while at work. Immediately after, she

ABBREVIATIONS: MM, multiple myeloma; SPB, solitary plasmacytoma of bone.

developed numbress down the entire right side of her body, which progressed to bilateral numbness. The patient underwent treatment with cyclobenzaprine, ibuprofen, and prednisone. Four days later, she received a computed tomography (CT) scan of her cervical spine where degenerative disk changes were discovered at cervical 5-6. She returned to work with a 10-pound lifting restriction but continued to have severe pain in the back of her neck and had difficulty flexing her neck. The patient returned to the emergency department with reported progressive numbness of her face in addition to her body. MRI showed "a large central and rightsided cervical 5-6 disk herniation with severe canal stenosis, cord compression, and a small single area of myelomalacia" (Figure 1). Neurological examination was unremarkable with normal strength and no signs of myelopathy. A 5-6 anterior cervical discectomy with fusion was performed by a local orthopedic spine surgeon. A fibular allograft with small portions of autograft was used to repair the surgical defect without the addition of bone morphogenetic protein. Postoperatively, the patient's numbress resolved completely. Postoperative images showed stable hardware (Figure 2).

The patient was lost to follow-up until presenting in early 2023 with neck pain, decreased sensation in her extremities, gait disturbances, and weakness that started after lifting a crate. CT (Figure 3A and 3B) and MRI (Figure 3C and 3D) revealed

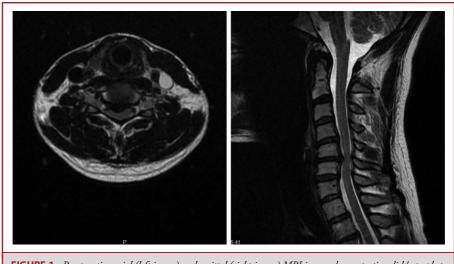


FIGURE 1. Preoperative axial (left image) and sagittal (right image) MRI images demonstrating disk/osteophyte complex causing severe stenosis at the cervical 5/6 level.

complete erosion of the cervical 5 and 6 vertebral bodies, along with an enhancing soft tissue mass causing severe spinal cord compression. Given the findings, the patient consented to a 2level corpectomy with the removal of prior instrumentation. Intraoperative frozen sections were consistent with a plasma cell



tumor. There were no complications during this surgery. The next day, the patient underwent posterior instrumented arthrodesis from cervical 4-thoracic 2. Postoperative MRI (Figure 4A and 4B), CT (Figure 4C and 4D), and x-ray (Figure 4E and 4F) showed stable instrumentation with no evidence of residual soft tissue tumor.

Pathology from the surgical specimen was consistent with plasmacytoma, showing 40%–50% plasma cell infiltration. The patient subsequently underwent postoperative radiation to the cervical spine, 30 Gy in 10 fractions over 2 weeks. After completion of radiation therapy, the patient was started on daratumumab (DARZALEX[®], Janssen Biotech), bortezomib (Velcade[®], Millennium/Takeda and Janssen Pharmaceutical Companies), lenalidomide (Revlimid[®], Bristol Myers Squibb), and dexamethasone (Decadron[®], Hikma Pharma). The patient is currently still on chemotherapy with plans for future bone marrow transplant.

DISCUSSION

Plasmacytomas are solitary tumors originating from monoclonal plasma cells within bone marrow and can be found throughout the body. SPB, a type of plasmacytoma that occurs on bone, is most commonly found within the vertebral column.² Patients with SPB present with pain near the site of the tumor due to local destruction of bone, as well as neuralgias and radiculopathy from the tumor compressing nearby nerves and nerve roots.^{1,3} Diagnosis is based on tissue biopsy, histology, and immunohistochemistry, requiring there to be less than 10% monoclonal plasma cell infiltration, no evidence of systemic disease, and an absence of MM hallmarks, such as monoclonal proteins on serum electrophoresis.⁹



FIGURE 3. Computed tomography A, and B, and MRIC, and D, images revealing a compressive mass with erosion of the vertebral bodies and allograft at the site of anterior C5–C6 instrumentation.

The most unusual aspect of this patient's history is that she eventually developed a plasmacytoma in the same vertebral level where she had previously had surgery for a cervical disk herniation. Plasmacytoma developing at the site of a previous surgery is very rare and has only been documented a few times in the literature. Larrea et al reported a plasmacytoma at the site of a previous bilateral humeral fracture.¹⁰ This presented as a painful, "prominent swelling" near the deltoid area bilaterally, which rapidly progressed to pain in both shoulders and adjacent thorax. Similarly, Muchtar et al and Moreno et al¹² also present patients who developed plasmacytomas at the sites of previous procedures; however, these were the extramedullary subtype.^{11,12} In each of these cases, patients were diagnosed with MM shortly after the discovery of their plasmacytomas, suggesting that the presence of this tumor at the site of a previous surgery may be a warning sign of future progression to MM.¹⁰⁻¹² In another report, a young

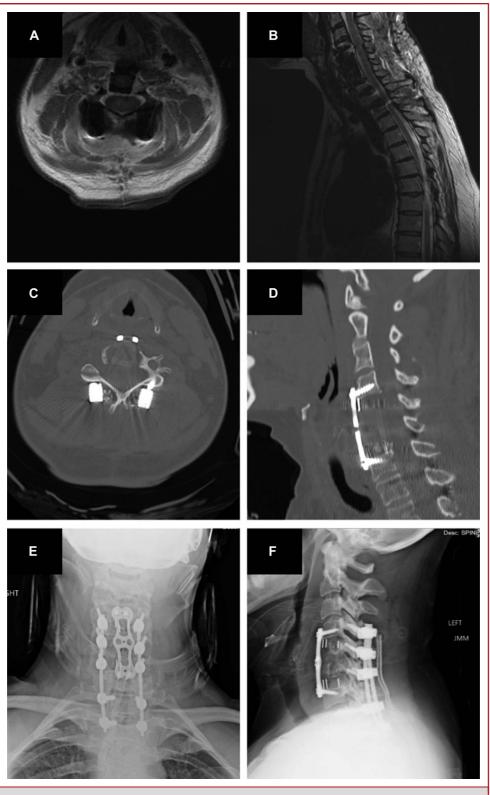


FIGURE 4. Postoperative cervical MRI **A**, and **B**, computed tomography **C**, and **D**, and upright x-ray imaging **E**, and **F**, demonstrating tumor gross total resections and anterior cervical 5/6 corpectomy and posterior cervical 4 thoracic 2 instrumented fusion.

patient who suffered low impact trauma to their lower back and pelvic region was subsequently discovered to have a SPB at the same location as their injury.¹ This demonstrates an additional instance in which plasmacytoma developed at the site of a previous injury of tissue. Although exact mechanisms of tumorigenesis in previous surgical sites are unknown, it has been speculated that inflammatory processes may promote the migration of atypical, malignant cells to the site of trauma.

Although plasmacytomas are believed to be an early form of MM, they can coincide with the disease, with 3.5%–18% of newly diagnosed MM patients being found to also have plasmacytoma.¹³ In our patient's case, it was initially believed that her mass was a SPB. However, bone marrow biopsy showed 40%–50% plasma cell infiltration, indicating that she may now have MM. SPB can progress to MM, with a 64% 5-year rate of progression and median time to progression being 19 months.^{14,15}

Limitations

Owing to our patient's lengthy loss to follow-up, it is difficult to say whether her plasmacytoma occurred first and then progressed to MM or if she developed MM which then manifested as a solitary lesion.

Soft tissue tumors arising at the site of a previous procedure of the spine are rare but have been reported in the literature. However, none of these were found to be SPB but rather desmoid tumors.^{16,17} Desmoid tumors are primarily managed with active surveillance with eventual medical and surgical treatment if evidence of progression.¹⁸ This differs from SPBs in which radiotherapy is standard of care.² In cases where there is compression of the spinal cord or instability of the surrounding vertebrae, as in our patient, surgery is pursued in combination with radiation.²

CONCLUSION

SPB is a rare tumor that is most often found in the vertebral column. Yet, if a patient is discovered to have a tumor at the site of a previous spinal surgery, SPB should be considered in the differential. Even in the case where a patient is not a candidate for surgery to remove the tumor, a biopsy should be performed to characterize the lesion and to assess for increased plasma cell infiltration, indicating a more severe diagnosis of MM.

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