

## Case Report

# Plasmacytoma-induced Pathological Axis Fracture: A Case Report and Literature Review



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**Citation** Abouie-Mehrizi M, Haghir AM, Ghanavision M, Mojazi-Amiri M, Maleki-pour B, Mohazzab-Torabi S. Plasmacytoma-induced Pathological Axis Fracture: A Case Report and Literature Review. *Iran J Neurosurg*. 2025; 11:???. <http://dx.doi.org/10.32598/irjns.11.7>

<http://dx.doi.org/10.32598/irjns.11.7>

### Article info:

**Received:** 06 Jan 2024

**Accepted:** 01 Feb 2025

**Available Online:** 27 May 2025

### Keywords:

Spinal tumor, Plasmacytoma, Spinal fusion

## ABSTRACT

**Background and Importance:** For the cervical spine, managing the pathological body of the axis tumor is controversial. In older patients, major procedures may be hazardous, and non-operative treatment is associated with cervical instability.

**Case Presentation:** A 70-year-old woman presented with chronic neck pain, mild quadriparesis, and a "sensation" of skull instability. Computed tomography (CT) showed a lytic lesion involving almost the entire body of the C2 vertebra. Magnetic resonance imaging (MRI) revealed a hypotensive infiltrative lesion involving the axis. Posterior cervical fusion was suggested to improve clinical symptoms and prevent future vertebral collapse with catastrophic neurological consequences. Under general anesthesia and neurophysiological monitoring, one-stage surgery was performed with C1 laminar hook and C3-C4 lateral mass screw insertion. A cytological sample from the lytic lesion was transpedicular through C2. Her neurological function recovered well, and he could walk independently 15 days postoperatively. Histopathology with further immunohistochemistry (IHC) of the C2 vertebral body also showed plasmacytoma.

**Conclusion:** No specific approach is observed to treat the axis's rare, solitary pathological lesions. Posterior cervical fixation has proven safe and effective in palliative treatment, improving neurological function and cervical stability at the beginning of early chemotherapy.

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## Highlights

- Posterior cervical fixation was successfully performed to treat solitary plasmacytoma-induced pathological axis fracture in a 70-year-old female.
- The patient experienced a significant reduction in postoperative cervical pain and improved craniocervical stability.
- Cervical fixation combined with radiation therapy has proven effective in managing tumor-induced pain and preventing further instability.

## Plain Language Summary

This case report discusses a rare medical condition in which a type of cancer called plasmacytoma causes a fracture in the neck bone (axis). The patient was a 70-year-old woman who had severe pain and difficulty moving her neck due to this fracture. Plasmacytoma is a rare form of cancer that affects plasma cells in the bone and can cause damage, such as bone fractures, when it spreads. In this case, the tumor caused instability in the neck, leading to fracture. To treat the fracture and instability, doctors perform surgery to fix the neck bones and prevent further damage. They also used a special needle to collect a small sample of bone tissue for testing, confirming the plasmacytoma diagnosis. After the surgery, the patient experienced a significant reduction in pain and could move her neck again without a collar just seven days later. The findings from this case are crucial because they show that even in rare cases, such as this one, surgical treatment can greatly improve patients' quality of life. The study also highlights the importance of correctly diagnosing the condition and providing a combination of treatments, such as surgery and radiation therapy, to address both the cancer and pain caused by the fracture. This is especially relevant for the public because it shows that even difficult and rare medical conditions can be managed successfully with the right treatment, improving patient mobility and comfort.

### 1. Background and Importance

**S**olitary bone plasmacytoma (SBP) is a rare hematological malignancy defined as the localized proliferation of neoplastic monoclonal plasma cells in the absence of multiple myeloma (MM) with <10% infiltration of plasma cells into the bone marrow [1, 2]. SBP is a malignant condition that often develops in the posterior arch of the lumbar spine, followed by the thoracic and less common cervical regions. Metastases to the cervical junction (C1 and C2) are unconventional and account for <1% of all spinal metastases [3, 4]. Their malignant nature tends to be locally aggressive and can result in pathological fractures and catastrophic chronic or acute neurological complications [5]. SBP should be misdiagnosed in pathognomonic conditions based on its radiological appearance, consisting of lytic bone lesions identical to other malignancies, such as MM, aneurysmal bone cyst, and metastasis [6-8]. The treatment of SBP is controversial and challenging and requires consideration of the patient's comorbidities, quality of life, and overall survival. Multiple surgical approaches have been used with variable improvements.

We report the case of an unusual patient with SBP at C2 who presented with severe axial mechanical neck pain and craniocervical instability that was successfully treated with posterior decompression/stabilization followed by postoperative radiotherapy.

### 2. Case Presentation

A 70-year-old woman presented with 6 months of progressive, severe axial mechanical cervical pain and "sensation" of skull instability. Primarily, she had complete concessions with intact upper and lower extremities without sensory or upper motor neuron symptoms. Computed tomography (CT) showed a lytic expansile lesion involving the body of C2 and left lamina but thinned anterior and posterior cortical borders with some disruption (Figure 1). Furthermore, magnetic resonance imaging (MRI) showed hypo-signal lesions in the T1 sequence and hyper-signal in T2 with heterogenous enhancement in T1 + generalized anxiety disorder (GAD) (Figure 2). A whole-body bone scan only showed solitary axis involvement, and no abnormalities or indications of any metastatic conditions were detected. Laboratory tests, plasma protein electrophoresis, paranoid personality disorder (PPD) test, and brucellosis markers were all normal.



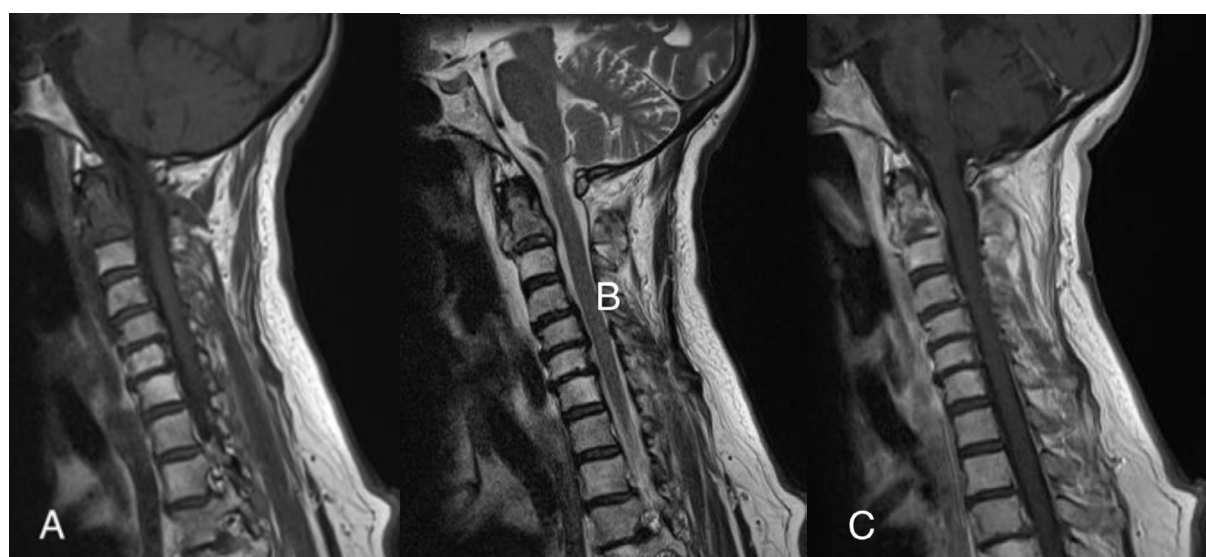
**Figure 1.** Sagittal (A), coronal (B), and axial (C) CT scan of cervical spine

CT scan: Computed tomography scan.

Given the symptoms, patient age, and structural instability of the craniospinal junction with the lytic C2 lesion, we decided to perform posterior cervical fusion with three main objectives: To improve the clinical symptoms, stabilize the cervical spine, and prevent devastating neurological consequences. Surgery was performed under general anesthesia and neurophysiological monitoring. The patient was premedicated with intravenous antibiotics (2 g of sodium cefazolin). The patient was placed in the prone position with an adequate cushion, and his head was fixed using a Mayfield clamp under general anesthesia. After division of the skin and posterior neck muscles, the Laminar hook on the C1 and C3-C4 lateral mass was inserted. An additional posterior transpedicular biopsy of the bone marrow was performed using the Jamshidi

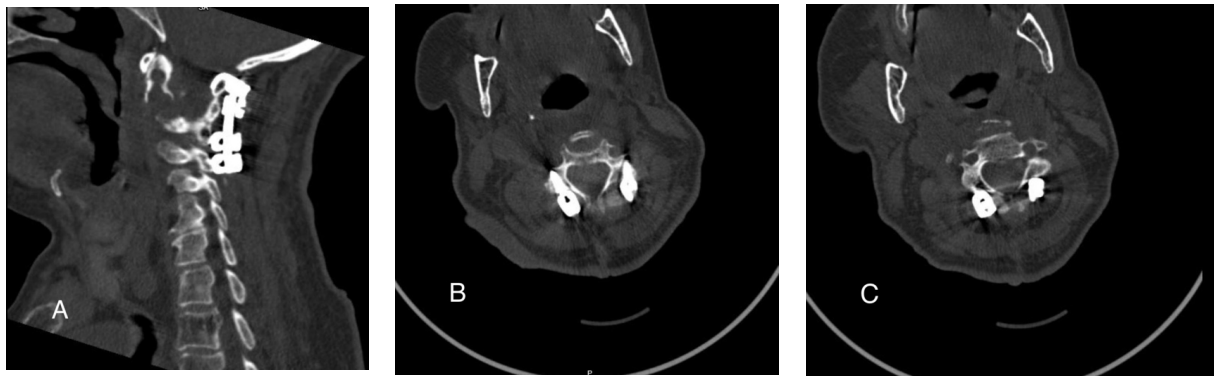
needle. Within 1 d postoperatively, the patient's pain resolved without new neurological deficits, and a CT scan confirmed cervical stability (Figure 3). The patient was ambulated on the third day and discharged on the fifth postoperative day using a Miami collar.

Histopathological and immunohistochemical studies of the lesion were consistent with those of the plasmacytoma. Anti-CD56, anti-CD166, anti-CXCR4, anti-Ki-67, and anti-c-MYC antibodies were used for immunohistochemical (IHC) analysis of plasmacytoma biopsies. Where staining with antibodies was observed, the following expression values were considered positive:  $\geq 10\%$  for CD56,  $\geq 50\%$  for CD166,  $\geq 20\%$  for CXCR4, and  $\geq 40\%$  for c-MYC. The level of proliferative activity was expressed as a percentage and assessed based on the expression of the Ki-67 marker



**Figure 2.** T1 sagittal (A), T2 sagittal (B), T1 + GAD sagittal (C) MRI of cervical spine

GAD: Generalized anxiety disorder; MRI: Magnetic resonance imaging.



**Figure 3.** Post-operative sagittal (A) and axial (B & C) CT scan of cervical spine

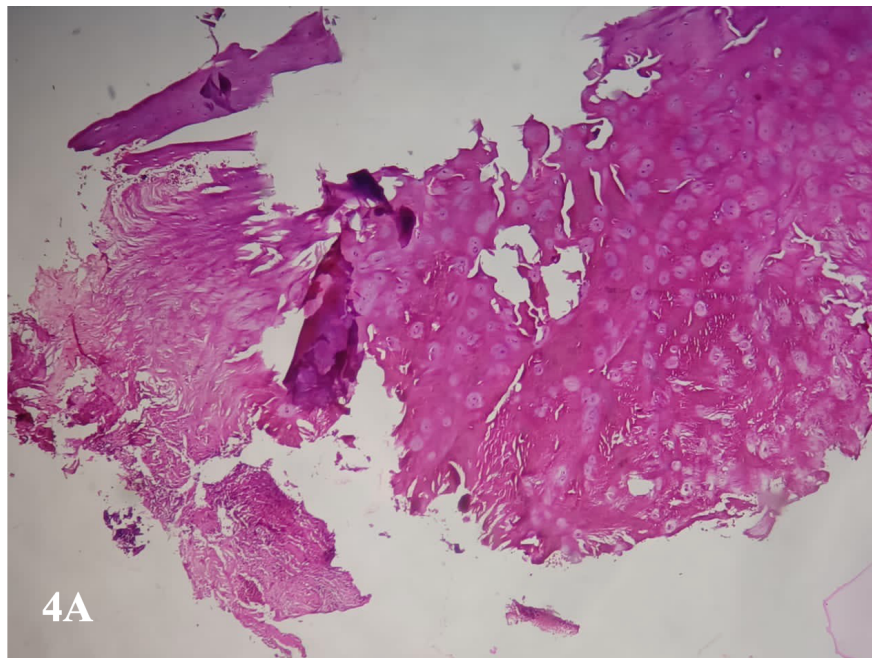
CT scan: Computed tomography scan.

in tumour cells. The diagnosis of SPB was confirmed by immunohistochemistry (IHC). Subsequently, the patient underwent radiotherapy (RT) (25 sessions [45 Gy]) with complete remission (Figure 4).

In postoperative course, conventional radiotherapy (CRT) (25 sessions: 40 Gy) was started in the fifth postoperative week. Six months later, a whole-body bone scan revealed adequate local control, no new bone lesions, and good therapeutic response. After one year of follow-up, the patient was pain-free and had a normal life. Subsequent cervical CT and whole-body scans showed stability and no evidence of recurrence.

### 3. Discussion

To our knowledge, cases of posterior cervical fixation for solitary plasmacytomas of the axis are rare in literature. Cervical pain in patients with metastatic lesions involving the spine can be caused by the tumor itself or by junctional instability (e.g. pathological fractures) [9]. We describe a 70-year-old woman who complained of severe chronic pain, exacerbated by range-of-motion activities, for the past 6 months.



**Figure 4.** Histopathological view of SBP

4A) Invasion of mature, and plasmablastic and anaplastic plasma cells in bone tissue (H & E, 100x)



SBP accounts for only 3% of all plasma cell neoplasms and is extremely rare (0.5%) in the craniocervical region [10]. It predominantly affects men (65%), and the mean age of patients diagnosed with SBP is 55 years [2]. Without effective preventive treatment, there is a high tendency for progression from SBP to MM.

The **International Myeloma Working Group (IMWG)** describes the criteria for SBP as follows: A solitary bone or soft tissue lesion, confirmed by bone biopsy showing clonal plasma cells; normal bone marrow, with no evidence of plasma cells or, at most, less than 10% involvement; no evidence of bone lesions other than the primary solitary lesion on MRI and/or CT of the spine and pelvis; and the absence of any target organ damage (e.g. hypercalcemia, renal failure, or anemia) [11].

Histological findings, including identifying monoclonal plasma cell infiltrates, are essential to establish a primary diagnosis. Our patient exhibited a lytic lesion on both MRI and CT, indicative of a malignant tumor. The CT showed expansive bone destruction, preserving the anterior and posterior borders. Patients presenting with tumor-induced pain may be good candidates for palliative therapies, such as radiation or bisphosphonate treatment, depending on the primary tumor type [12]. Our patient's pain was mechanical and caused by craniocervical instability resulting from a pathologic fracture. After a comprehensive whole-body metastatic assessment, the primary tumor remained undiagnosed. Consequently, we opted for an invasive treatment approach as the patient was in good overall condition but symptomatic.

Moreover, the fragile outer borders of the C2 body pose a significant fracture risk, with potentially catastrophic consequences. Single-session posterior cervical fixation is rarely recommended for treating metastatic cervical instability. To our knowledge, the transpedicular approach to access the C2 body is not a safe option due to the vertebral arteries' proximity and the axis pedicles' small size [3]. The transoral approach provides the most straightforward access, offering the advantage of a short distance to the C2 vertebral body, thereby minimizing the risk of injury to crucial vascular structures [13, 14]. However, we decided against a second surgery with a higher risk of infection and bleeding due to the undiagnosed nature of the lesion. Therefore, posterior transpedicular biopsy via a Jamshidi needle was the optimal option for confirming the diagnosis of SBP. The posterior approach also allowed stabilization of the craniocervical junction. After careful consideration, the posterior approach was selected, and C1 laminar hooks with C3-C4 lateral mass fixation were performed along with C2 transpedicular biopsy.

Several authors have recommended injecting selective bone cement to prevent vertebral collapse [2, 3, 5, 15]. However, we considered our patient a high-risk case for cement injection due to the thinness of the vertebral borders, which could easily be disrupted, potentially leading to catastrophic neurological consequences. Cervical fixation significantly improves the patient's quality of life. Our patient experienced a reduction in cervical pain 7 days after surgery and was able to ambulate without a soft neck collar.

Radiation therapy is the first-line treatment for SBP in several patients. However, in cases of spinal compression, neurological symptoms, or severe instability, radiation therapy should be combined with spinal stabilization. Additionally, radiation therapy during surgery helps eliminate most tumor cells and delays local recurrence, with better outcomes in managing tumor-related pain.

One of the main limitations in discussing plasmacytoma and C2 pathological fractures is the scarcity of comprehensive studies and research specifically focusing on this topic. Most available literature consists of case reports and small case series, limiting the findings' generalizability. Due to the rarity of plasmacytoma and C2 pathological fractures, studies often have small sample sizes, making it challenging to draw definitive conclusions. Large-scale studies involving diverse populations are required to obtain a more accurate understanding of this condition. Most of the available literature is retrospective and relies on medical records and historical data. Prospective studies that follow patients over time provide more robust evidence and allow for a better characterization of disease progression and treatment outcomes. Plasmacytoma and C2 pathological fractures can manifest in various ways, leading to differences in clinical presentation, radiographic findings, and treatment approaches. The lack of standardized diagnostic criteria and treatment protocols contributes to the heterogeneity of reported cases, making it challenging to compare and synthesize available evidence. Many studies have limited follow-up periods, often ranging from months to years.

## 4. Conclusion

Finally, SBP involving the C2 cervical vertebral body is very rare; however, posterior cervical fixation combined with neoadjuvant radiotherapy may constitute a safe and effective treatment to stabilize SBP, compromising the stability of C2, improving mechanical pain, and improving quality of life. Lytic lesions affecting the vertebral body, especially in the upper cervical region, must be considered when making a differential diagnosis and precise surgical approach.

## Ethical Considerations

### Compliance with ethical guidelines

Written informed consent was obtained from the patient to publish this case report and any accompanying images. The patient were fully informed about the nature of the report and the potential for her medical information to be shared in scientific literature, with all efforts made to maintain confidentiality and privacy. The patient signed the consent before submitting or publishing case details.

### Funding

This research did not receive any grant from funding agencies in the public, commercial, or non-profit sectors.

### Authors' contributions

Conception and design: Saman Mohazzab-Torabi, Mohammad Ali Abouie Mehrizi; Data collection: Saman Mohazzab-Torabi, Mostafa Ghanavizian, Milad Mojazi-Amiri; Data analysis and interpretation: Saman Mohazzab-Torabi, Milad Mojazi-Amiri; Drafting the article: Saman Mohazzab-Torabi, Amir Hossein Haghir; Critically revising the article: Amir Hossein Haghir, Mohammad Ali Abouie Mehrizi, Bahador Maleki-poor; Reviewing the submitted version of the manuscript: Saman Mohazzab-Torabi, Amir Hossein Haghir; Final approval: All Authors.

### Conflict of interest

The authors declared no conflict of interest.

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