

Palatal Solitary Plasmacytoma: A Case Report on Post-radiotherapy Challenges

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Abstract

Background/Aim: Plasmacytomas are neoplastic proliferations of plasma cells, usually presenting as solitary lesions distinct from multiple myeloma (MM). Solitary plasmacytomas in the oral cavity and head and neck are very rare, with limited data on clinical course and treatment response.

Case Report: A 78-year-old woman presented with a solitary plasmacytoma on the palate. Imaging ruled out bone or systemic involvement. Pathology showed positivity for CD138, lambda light chain, CD79a, MUM1, Ki-67 (10-15%), and amyloid deposits. Although the patient received definitive radiation therapy (50 Gy/25 fractions), residual disease remained. At the patient's request, six cycles of systemic chemotherapy with daratumumab, lenalidomide, and dexamethasone (DRd) were administered, leading to further shrinkage without complete response.

Conclusion: This rare case of solitary plasmacytoma showed residual disease post definitive radiotherapy. In those residual tumors, close long-term monitoring is crucial.

Keywords: Plasmacytoma, CD138, radiotherapy resistance, multiple myeloma.

Introduction

Plasma cell neoplasms arise from the abnormal proliferation of plasma cells, which are terminally differentiated B lymphocytes, and typically manifest as tumors within the bone or soft tissue. Multiple myeloma

is a malignant plasma cell disorder that can lead to significant organ dysfunction. It commonly affects multiple skeletal sites, including the skull, vertebrae, ribs, and iliac bones. However, plasma cell proliferation occurs as a solitary lesion without evidence of systemic involvement. These lesions are found in the bone – referred to as



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solitary bone plasmacytoma (SBP) – or in soft tissue – known as extramedullary plasmacytoma (1). Occurrence of solitary plasmacytoma specifically in the oral and maxillofacial regions is exceedingly rare (2). Here, we report a rare case of a solitary bone plasmacytoma arising in the palate.

Case Report

A 78-year-old female was referred to our hospital for further evaluation of a suspected plasma cell tumor located in the palate. Intraoral examination revealed a diffuse, elastic, soft mass with a smooth surface extending from the region of tooth #16 Fédération Dentaire Internationale (FDI) notation across the hard palate (Figure 1A). On palpation, the mass was non-tender, with no evidence of fluctuation. Computed tomography revealed no osteolytic lesions suggestive of multiple myeloma, and the lesion was confined to the maxilla (Figure 1B). Additionally, no abnormalities were noted in the lung fields or abdominal organs, and there was no significant cervical lymphadenopathy. Contrast-enhanced magnetic resonance imaging demonstrated infiltration of the right palate and right maxilla, with extension into the right maxillary sinus (Figure 1B). The lesion showed high signal intensity on T2-weighted images and exhibited strong contrast enhancement. Although the mass was in contact with the nasal cavity, no definitive evidence of invasion was identified. Bone marrow aspiration revealed a nucleated cell proportion of approximately 25-30%, with a myeloid-to-erythroid ratio of approximately 2-3. Serum immunoglobulin levels were as follows: IgG, 1920 mg/dl; IgA, 232 mg/dl; and IgM, 160 mg/dl. Immunoelectrophoresis showed no detectable M-protein, and urine analysis for Bence Jones protein was negative.

Histopathological examination. The biopsy specimen was routinely fixed in 10% neutral buffered formalin and embedded in paraffin. Hematoxylin and eosin staining revealed well-demarcated, multilobular nodules composed of lymphoid mononuclear cells with plasmacytoid morphology (Figure 2). The tumor cells

displayed low-grade cytological atypia, and no mitotic figures were observed (Figure 2, Area A). Additionally, nodular regions with eosinophilic deposits were identified in the peritumoral area (Figure 2, Area B).

Immunohistochemical analysis demonstrated that the plasmacytoid tumor cells were positive for CD138, lambda (λ) light chain, and Ki-67 (10-15%) (Figure 3A). CD79a and MUM1 positivity and negativity for cytokeratin, smooth muscle actin, S-100, and GCDFP-15 were also noted (data not shown). Since there was no evidence of tumors in the other organs, the lesion was diagnosed as a solitary plasmacytoma.

The eosinophilic deposits stained positively with Congo red under polarized light microscopy (Figure 3B). The deposits were positive for lambda (λ) light chain, and negative for kappa (κ) light chain and transthyretin (data not shown), confirming the presence of amyloid deposition.

Diagnosis and treatment. Based on the clinical, radiological, and pathological findings, the patient was diagnosed with a solitary plasmacytoma of the palate. As definitive treatment, she underwent radiation therapy with a total dose of 50 Gy delivered in 25 fractions. Post-radiation imaging demonstrated reduction in tumor size, although a residual lesion remained. Hence, she was referred to the Department of Oral and Maxillofacial Surgery for further surgical evaluation. However, based on the patient's preference, the treatment strategy was transitioned to systemic chemotherapy under the supervision of the Department of Hematology. The patient subsequently received six cycles of DRd therapy, comprising daratumumab, lenalidomide, and dexamethasone. Although a reduction in tumor size was observed, complete remission was not achieved at four months after the initiation of chemotherapy (Figure 1A). The patient is currently continuing chemotherapy.

Discussion

We report a rare case of a plasmacytoma arising in the palate, which, despite definitive radiotherapy followed by

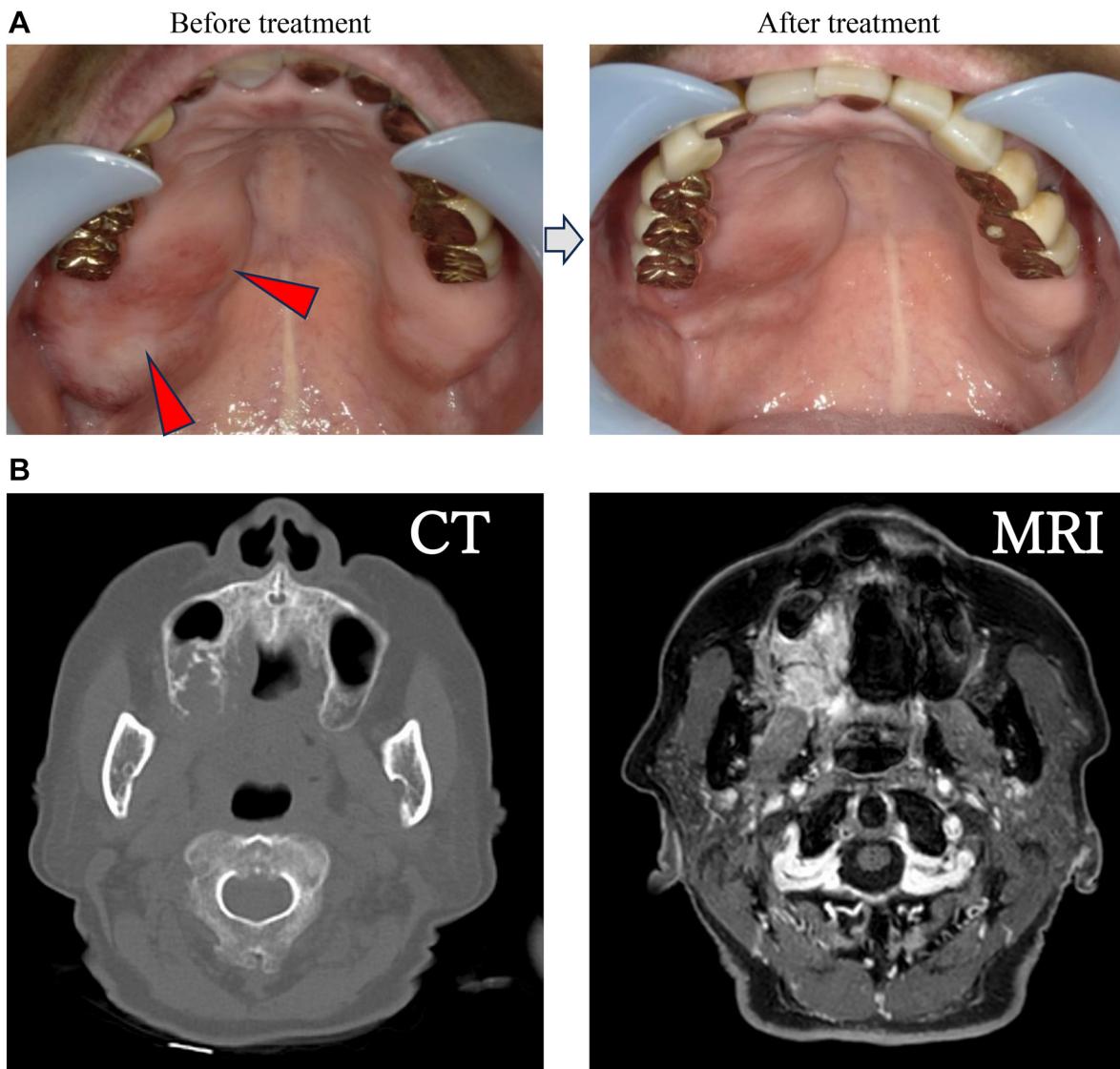


Figure 1. *Clinical observations. A)* Intraoral photographs of the lesion before treatment, and after radiation therapy (total dose of 50 Gy delivered in 25 fractions) and five cycles of chemotherapy. The red arrowheads indicate the lesion at the time of initial diagnosis. *B)* Computed tomography (CT) and T1-weighted contrast-enhanced magnetic resonance imaging (MRI) scans demonstrating the lesion prior to treatment.

chemotherapy, did not achieve complete remission. Plasmacytoma was first described by Schridde in 1905 as a rare, solitary proliferation of neoplastic monoclonal plasma cells. According to the International Myeloma Working Group (IMWG), plasmacytomas are categorized into three types: SBP, extramedullary plasmacytoma, and multiple myeloma. Among these, SBP is the most common,

accounting for 3-5% of all malignant plasma cell neoplasms (3). SBP occurs most frequently in the vertebrae, followed by the pelvis and ribs, while cases involving the jaw are relatively rare. Agostini reported on 50 cases of solitary plasmacytoma of the jawbone, noting a higher incidence in the mandible, particularly in the mandibular ramus and angle regions (4). However,

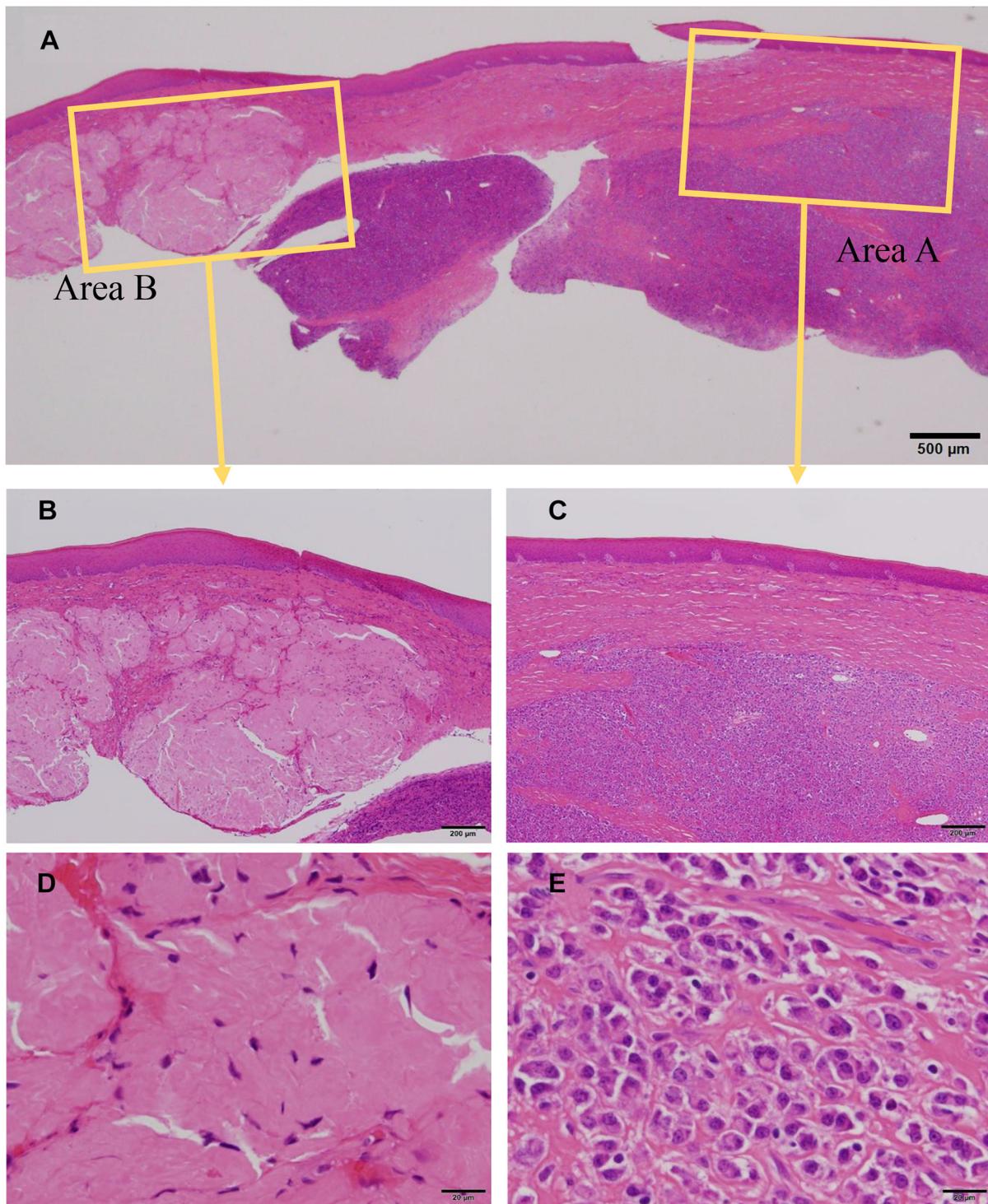
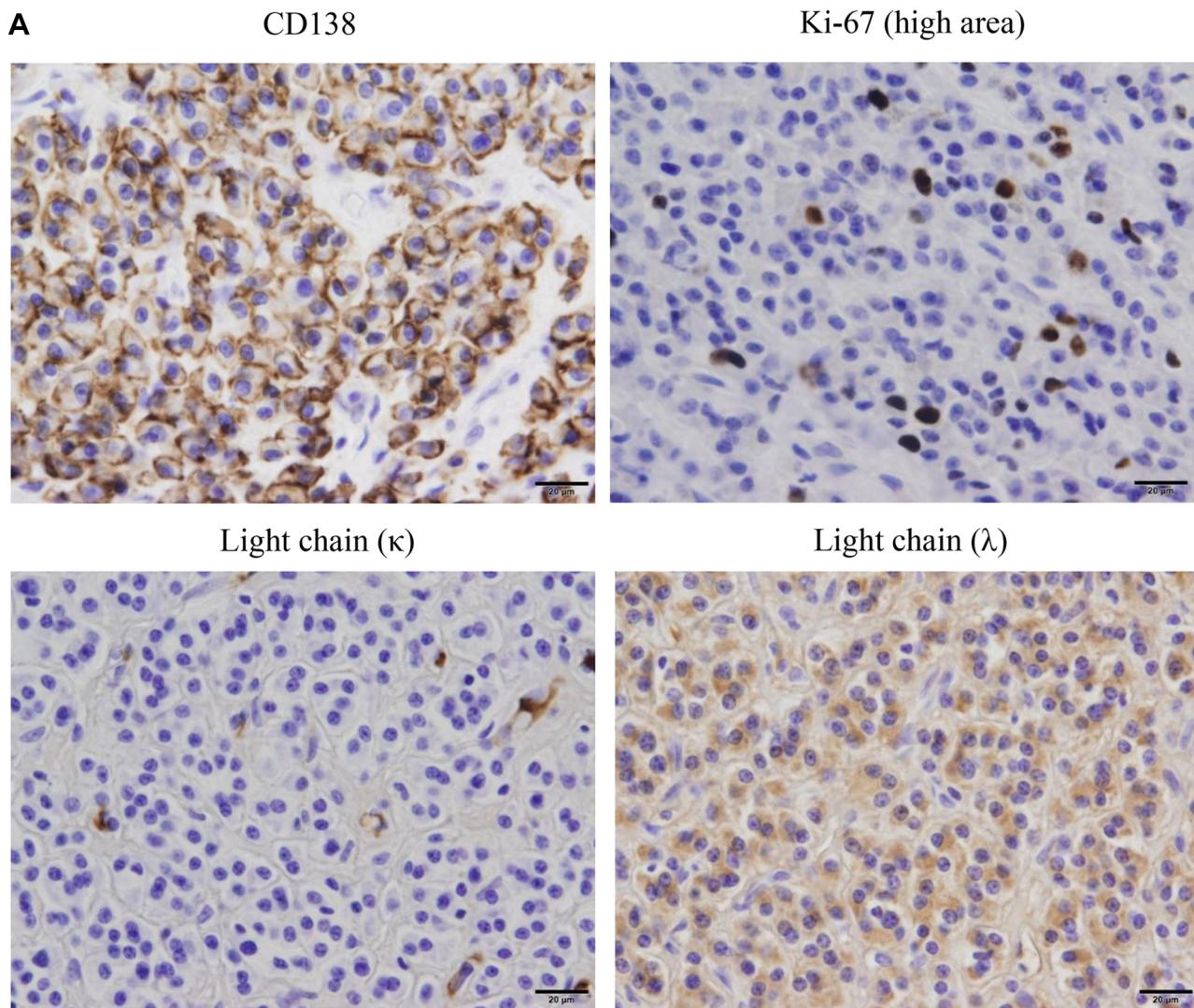


Figure 2. Pathological examination. Hematoxylin and eosin staining of the main lesion (A). High-magnification images of the tumor area (Area A) in Figure 2A are presented in (C) and (E) (scale bars: 200 μ m and 20 μ m, respectively). High-magnification images of the adjacent eosinophilic area (Area B) in Figure 2A are shown in (B) and (D) (scale bars: 200 μ m and 20 μ m, respectively).

Figure 3. *Continued*

occurrence of plasmacytoma in the palate, as seen in our case, is extremely uncommon.

According to the diagnostic criteria defined by the IMWG, the lesion must fulfill all of the following five conditions for a diagnosis of SBP: 1) no M-protein in serum or urine, 2) a single site of bone destruction due to clonal plasma cells, 3) absence of abnormal plasma cells on bone marrow examination, 4) normal skeletal evaluation except at the lesion, and 5) no organ damage (5). Our case met all of these criteria. Additionally, in this

case, the diagnosis was made based on positivity for CD138, CD79a, lambda (λ) light chain, and Ki-67.

Although radiotherapy has been reported to provide good local control (6), cases such as ours demonstrate that residual disease might persist. Furthermore, SBP carries a high risk of progression to multiple myeloma – 65%-84% within 10 years, and nearly 100% within 15 years (7, 8). In addition, patients with solitary bone plasmacytoma (SBP) have been shown to have inferior 3-year disease-free survival (DFS) and overall survival (OS) compared to

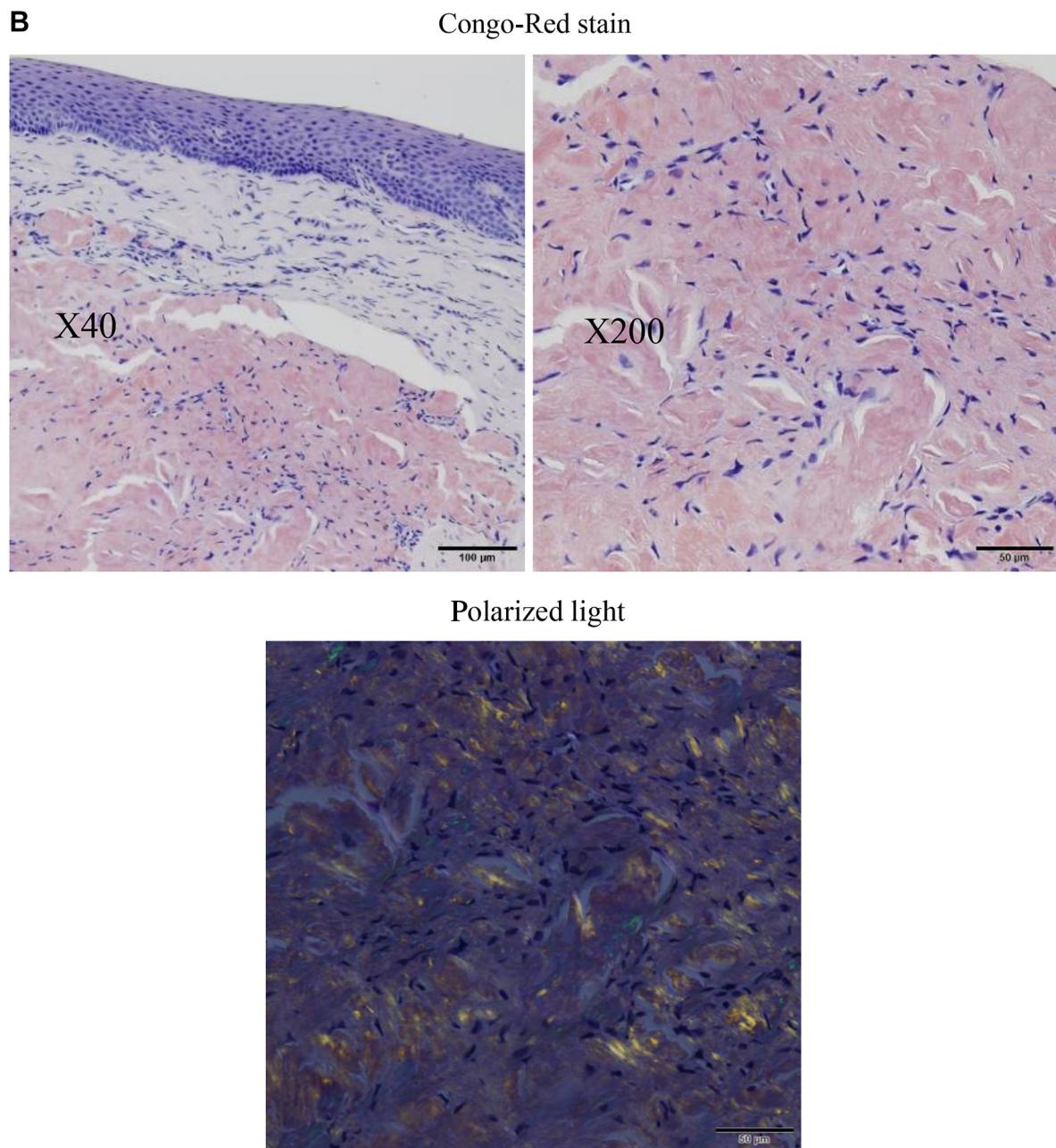


Figure 3. Results of immunohistochemistry and evaluation for amyloid deposition. (A) Immunohistochemistry revealed positivity for CD138, light chains, and Ki-67 (scale bars: 20 μ m). (B) Congo red stained sections assessed using polarized light microscopy (scale bars: 100 μ m, 50 μ m, 50 μ m).

patients with solitary extramedullary plasmacytoma. The higher likelihood of disease progression in SBP patients, as compared to those with solitary extramedullary plasmacytoma, has also been confirmed in a cohort study

from the Mayo Clinic (9). So far, no progression to multiple myeloma has been observed in the present case at one year after the initial diagnosis; however, close follow-up remains essential.

Conclusion

We experienced a case of solitary plasmacytoma arising in the palate that showed tumor progression even after definitive radiotherapy. In those residual tumors, the potential risk of plasmacytoma progressing to multiple myeloma should be kept in mind.

Conflicts of Interest

None of the Authors have any conflicts of interest to declare in relation to this study.

Authors' Contributions

Mayuko Yamashita: Collection of clinical data, histological and immunohistochemical analyses, drafting of the manuscript; Yoshihiko Kondo: histological and immunohistochemical analyses support; Shinya Endo, Nao Nishimura, Yawara Kawano: Responsible for diagnosis and treatment; Akiyuki Hirosue, Ryoji Yoshida: Coordination of the treatment strategies; Hideki Nakayama: Writing – review and editing; Yoshihiro Komohara: Supervision of the entire study.

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Artificial Intelligence (AI) Disclosure

No artificial intelligence (AI) tools, including large language models or machine learning software, were used in the preparation, analysis, or presentation of this manuscript.

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