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Tumor Regression Following Radiotherapy for Primary Cutaneous Plasmacytoma in a Young Patient: A Case Report

Masaki Tobikawa [1], Tatsuji Mizukami [1], Kentaro Yamagishi [1], Teruhiko Makino [2], Jun-Ichi Saitoh [1]

1. Division of Radiation Oncology, Department of Radiology, Faculty of Medicine, Academic Assembly, University of Toyama, Toyama, JPN 2. Department of Dermatology, University of Toyama, Toyama, JPN

Corresponding author: Masaki Tobikawa, user.milkcoffee@gmail.com

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Abstract

Primary cutaneous plasmacytoma is a type of extramedullary plasmacytoma and is an extremely rare disease. No standard treatment regimen has been established for this condition; various treatments, including surgical excision, radiation therapy, and chemotherapy, have been employed previously. We report a case of a 15-year-old male who developed a nodule on the left nasal wing and was diagnosed with cutaneous plasmacytoma. He received electron beam irradiation at a dose of 60 Gy in 30 fractions, resulting in gradual regression of the lesion.

Categories: Hematology, Radiation Oncology, Dermatology
Keywords: electron beam, primary cutaneous plasmacytoma, radiation therapy, skin, young person

Introduction

Plasma cell neoplasms are diseases characterized by the monoclonal proliferation of plasma cells, which are the fully differentiated form of B cells. Depending on their characteristics, plasma cell neoplasms are classified into monoclonal immunoglobulinemia, multiple myeloma, solitary plasma cell neoplasms, and plasma cell leukemia. While most plasma cell tumors (PCPs) present as multiple myeloma, approximately 5% present as isolated PCPs in bone or extramedullary tissues [1].

PCPs that arise extramedullary (extramedullary PCPs) frequently occur in the upper respiratory tract or upper gastrointestinal tract [2]. Among extramedullary PCPs, those arising in the skin are classified as secondary tumors, which develop from PCPs originating elsewhere, and primary tumors, which arise exclusively in the skin; the latter are referred to as primary cutaneous PCPs [3].

Primary cutaneous plasmacytoma is a rare disease, with only about 80 cases reported to date; various treatments are employed, including surgical resection [4], radiation therapy [5], chemotherapy [6], and ultraviolet therapy [7]. However, no standard treatment has been established. Furthermore, regarding radiation therapy, various methods are employed, including irradiation with cobalt-60 [8], brachytherapy [9], and electron beam irradiation [10]. Treatment doses also vary widely, ranging from 20 Gy to 50 Gy, and there is no established standard for the irradiation method or dose.

We present a case of solitary primary cutaneous plasmacytoma occurring on the left nasal wing that was treated with electron beam irradiation at 60 Gy/30 fractions, resulting in gradual shrinkage.

Case Presentation

The patient was a 15-year-old male who presented with a nodular lesion on the left nasal wing. As the lesion showed a tendency to enlarge, he consulted a local physician six months after first noticing the symptoms and was referred to our hospital that same month for further evaluation and treatment. The patient had no significant past medical history.

On initial examination, a 9 mm red nodular lesion was noted on the left nasal wing (Figure *1*). A biopsy was performed due to suspicion of a skin tumor. Hematoxylin and eosin staining revealed diffusely distributed atypical plasma cells within the dermis. The immunohistochemical panel included CD10, CD20, and CD138 immunohistochemical stains, as well as kappa and lambda light chain in situ hybridization.

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FIGURE 1: A reddish, nodular lesion measuring 9 mm was observed on the left nostril.

Immunohistochemical staining revealed that the atypical cells were CD138-positive and were considered atypical plasma cells. However, they were negative for CD10 and CD20, and no B-cell infiltration was observed. In situ hybridization revealed that, while the λ chain showed widespread positivity, the κ chain showed only minimal positivity; therefore, the proliferation was considered to be monoclonal. Based on these findings, a diagnosis of plasmacytoma was made (Figure *2*).

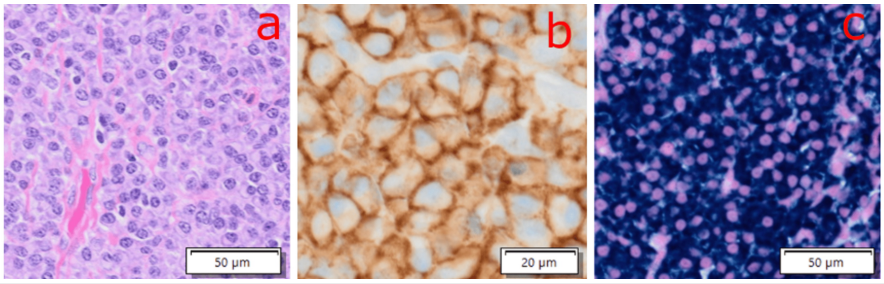


FIGURE 2: (a) Dense plasma cell infiltrate, hematoxylin and eosin (H&E) staining; (b) CD138 immunostaining labeling plasma cells, immunohistochemistry (IHC); (c) most plasma cells were lambda-positive, in situ hybridization (ISH).

A systemic examination was subsequently performed. Blood tests revealed no obvious abnormalities, including calcium (Ca) levels, hemoglobin (Hb) levels, and renal function. CT and MRI scans were performed to evaluate systemic and localized findings, but no obvious abnormalities were identified other than the lesion on the nasal wing. Furthermore, regarding the nasal wing lesion, no obvious bone or nasal mucosal infiltration was observed. No obvious multiple lesions were identified on visual examination either. Immunoelectrophoresis and urinalysis revealed no evidence of M protein. Bone marrow examination showed a plasma cell percentage of 1.8%, with no obvious abnormalities noted. Based on these findings, a diagnosis of primary cutaneous plasmacytoma was made.

Given the location of the lesion, surgical resection was deemed difficult, and a course of radiation therapy was planned. Treatment was performed using a 6 MeV electron beam. The GTV was defined based on imaging and physical examination, and the CTV was defined as a 2 cm margin around the tumor. The treatment plan called for delivering a total dose of 50 Gy in 25 fractions to a 5 cm diameter region centered on the tumor to ensure adequate coverage of the entire volume. Although there was a tendency toward shrinkage at 46 Gy, the response was poor. A dose of 40-50 Gy is recommended for extramedullary plasmacytoma, and there was a possibility that the tumor might shrink even if treatment were completed as planned at 50 Gy. However, in this case, local recurrence would make reirradiation or surgical resection difficult and might necessitate systemic chemotherapy; furthermore, given the patient's age, the physical and social burden would be significant. Thus, although there is a risk of adverse events associated with increasing the dose, and considering the potential for improved local control with additional radiation, we decided-after thorough consultation with the patient and family-to increase the dose from 50 Gy to 60 Gy (Figure *3*).

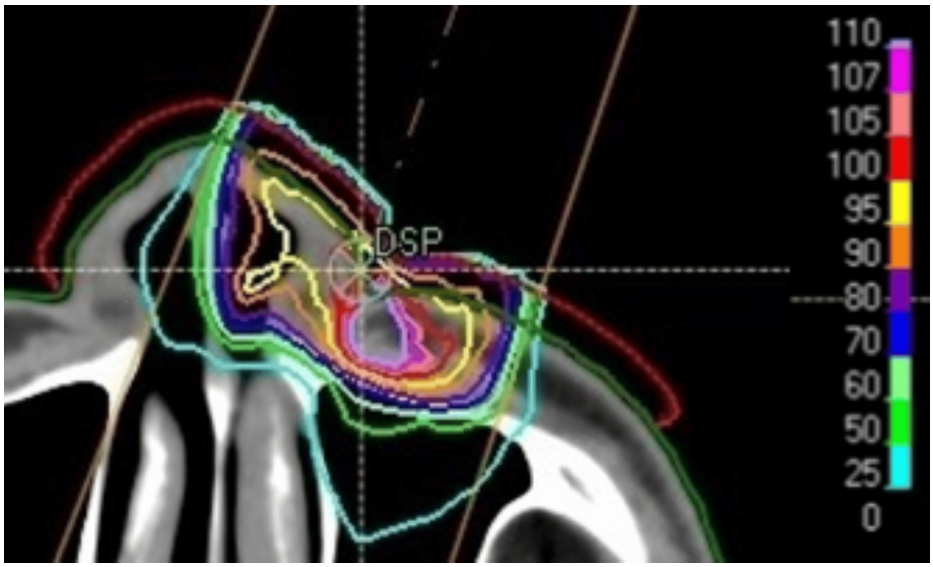


FIGURE 3: A total dose of 60 Gy was administered in 30 fractions using electron beams, centered on the tumor, with a diameter of 5 cm.

At the 60 Gy dose, the lesion had crusted over and shrunk to 7 mm in size; the only adverse event observed was mild dermatitis. Seven months after completion of treatment, the lesion had flattened and further shrinkage was noted (Figure *4*).



FIGURE 4: Seven months after the end of treatment, the lesion had flattened, and further shrinkage was observed.

Discussion

Solitary plasmacytoma is classified into two types: those that arise in the bone and those that arise outside the bone. The diagnostic criteria for solitary plasmacytoma are as follows: the presence of bone or soft tissue consisting of clonal plasma cells, as confirmed by biopsy; the absence of clonal plasma cells in the bone marrow (<10%); the absence of abnormalities on bone X-rays, vertebral, and pelvic MRI (or CT) other than the solitary plasmacytoma lesion; no organ dysfunction (Ca: serum Ca > 11 mg/dL or an increase exceeding 1 mg/dL above the upper limit of normal; renal failure: CrCl < 40 mL/min or serum Cr > 2.0 mg/dL; anemia: Hb < 10 g/dL or a decrease of more than 2 g/dL below the lower limit of normal; bone lesions: one or more lesions detected on bone X-ray, CT, or PET/CT) [11].

Solitary plasmacytoma accounts for about 5% of all plasmacytomas [12], with more than 80% occurring in the upper respiratory tract or upper gastrointestinal tract [11]; primary cutaneous plasmacytoma, which primarily affects the skin as in this case, is an extremely rare condition.

Primary cutaneous plasmacytoma is more common in men and predominantly occurs in the elderly or middle-aged; it rarely occurs in young adults, as seen in this case [12].

Although the exact etiology of primary cutaneous plasmacytoma remains unknown, it has been suggested that the condition may develop in association with some form of stimulus, such as trauma, infection, or malignant tumors [13]. Li et al. reported a case of primary cutaneous plasmacytoma that developed at the site of a postoperative scar following cardiac pacemaker implantation [14].

In a review of reports involving patients with primary cutaneous plasmacytoma who underwent surgery, radiation therapy, or chemotherapy and were followed for more than five years, it was reported that approximately half experienced recurrence; therefore, the prognosis cannot be considered favorable [15]. However, solitary primary cutaneous plasmacytomas, such as the present case, are said to have a better prognosis compared to multiple cases [16].

In our case, we treated a primary cutaneous plasmacytoma that presented as an isolated lesion on the skin by administering a total dose of 60 Gy in 30 fractions at 6 MeV using a 5 mm bolus centered on the tumor within a 5 cm diameter. This resulted in gradual tumor shrinkage.

Although reports of radiotherapy for primary cutaneous plasmacytoma are sporadic, there were only nine cases, including the present one, in which radiotherapy alone was used to treat an isolated primary cutaneous plasmacytoma (Table *1*).

Case	Age	Sex	Size	Modality	Dose/fraction	Treatment progress
Klein et al. (1977) [9]	52	M	1.5 cm	Mold containing radon seed	48.18 Gy/7 fr	No recurrence observed during 18 months of follow-up.
van der Putte et al. (1984) [17]	54	M	6 cm	X-ray irradiation	40 Gy/unknown	Complete remission (CR) after treatment. The duration of follow-up is unknown.
Wong et al. (1994) [18]	80	M	4×6 cm	Electrons	50 Gy/25 fr	No recurrence observed during 72 months of follow-up.
Gambichler et al. (2008) [19]	80	M	Unknown	Unknown	30 Gy/15 fr	Complete remission (CR) after treatment. The duration of follow-up is unknown.
Li et al. (2013) [14]	89	M	5×6 cm	Unknown	20 Gy/5 fr	The disease recurred two months later, and the patient died five months later.
Tsang et al. (2016) [5]	48	F	Unknown	Cobalt-60	25 Gy/10 fr	No recurrence observed during 95.9 months of follow-up.
Tsang et al. (2016) [5]	69	M	4.5×1.5 cm	Ortho-225 kVp voltage	35 Gy/10 fr	No recurrence observed during 11 months of follow-up.
Him et al. (2017) [20]	65	M	1.5×1 cm	X-ray irradiation	50 Gy/25 fr	Shrinkage occurred after treatment. The duration of follow-up is unknown.
Tobikawa et al. (2026) (current study)	15	M	9 mm	Electrons	60 Gy/30 fr	No recurrence observed during 7 months of follow-up.

TABLE 1: A case of isolated primary cutaneous plasmacytoma treated with radiation therapy alone.

The total dose ranged from 20 to 60 Gy (median: 40 Gy). Among the nine cases with reported follow-up periods, six cases had follow-up durations ranging from 5 to 95.9 months (median: 14.5 months). Recurrence was confirmed in one of the six cases.

It is reported that the treatment outcomes for solitary primary cutaneous plasmacytoma are better than those for multiple primary cutaneous plasmacytoma [20]; in a study of 35 cases of solitary primary cutaneous plasmacytoma, recurrence occurred in nine of 35 cases, and it was reported that most of the cases that recurred did so within three years [5]. Even when examining the nine cases treated with radiation therapy alone, the single case that relapsed did so within two years, indicating that the course of the disease was similar to that observed with other treatment methods.

Various treatment methods, including surgery, radiation therapy, and chemotherapy, have been employed for primary cutaneous plasmacytoma.

Tsang et al. compared recurrence-free survival and overall survival in 52 cases with follow-up data between those treated with radiation therapy or chemotherapy and those treated with surgery; they reported no significant difference in treatment outcomes [5].

When selecting a treatment for primary cutaneous plasmacytoma - such as surgery, radiation therapy, or chemotherapy - various factors should be considered, including the number and size of lesions, the site of onset, the patient's age, and general health status. Although this case involved a solitary lesion, surgical treatment was considered difficult due to its location on the face; therefore, treatment using electron beams was performed.

Conclusions

Although various treatment modalities have been used for primary cutaneous plasmacytoma, in this case, we administered radiation therapy using electron beams. While there is no established consensus regarding the radiation dose for primary cutaneous plasmacytoma, based on previous case reports, we initially considered a treatment regimen of 50 Gy in 25 fractions. However, since there was little change in tumor size during the course of treatment, we ultimately added 10 Gy in five fractions, bringing the total dose to 60 Gy in 30 fractions, and tumor shrinkage was observed in this case.

For cases of primary cutaneous plasmacytoma where surgery is difficult, radiation therapy may be a viable treatment option, and further prospective evaluation is warranted.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Masaki Tobikawa, Jun-Ichi Saitoh, Kentaro Yamagishi, Tatsuji Mizukami, Teruhiko Makino

Acquisition, analysis, or interpretation of data: Masaki Tobikawa, Jun-Ichi Saitoh

Drafting of the manuscript: Masaki Tobikawa, Jun-Ichi Saitoh

Critical review of the manuscript for important intellectual content: Masaki Tobikawa, Jun-Ichi Saitoh, Kentaro Yamagishi, Tatsuji Mizukami, Teruhiko Makino

Supervision: Jun-Ichi Saitoh

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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