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CASE REPORT

A case of multiple myeloma with atypical localization of orbital metastases

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Abstract

Multiple myeloma (MM) is a malignant tumor characterized by monoclonal proliferation of plasma cells and is usually seen in older age. In this report, we present a case of atypical localization of a plasmacytoma. Seventy-2-years-old male patient who had been followed up for 3 years for MM presented to our clinic with low vision for 6 months and a mass in the left eye that showed sudden growth in 1 week. An irregular and vascularized mass was observed in the caruncle region of the left eye. Visual acuity was 7/10 in the right eye and hand movements in the left eye. Cataract was present in the left eye and no pathology was detected on ultrasonography. On examination of the right eye, the cornea was clear, the iris was normal, the posterior chamber intraocular lens was present and the fundoscopy was normal. Magnetic resonance imaging revealed an intense contrast enhancement of the mass. A biopsy was performed for pathological examination, and a plasmocytoma was diagnosed. The patient was referred to the hematology clinic for further treatment.

Keywords: Atypical localization; caruncle tumors; metastasis; multiple myeloma; plasmacytoma.

Multiple myeloma (MM) is a malignant tumor characterized by monoclonal proliferation of bone marrow-derived plasma cells, usually occurring in advanced age.[1] MM involvement outside the bone marrow is called plasmacytoma.[2] Plasmacytomas composed of plasma cells are clinically and pathologically divided into three groups: MM, solitary bone plasmacytoma, and extramedullary plasmacytoma occurring in the soft tissue. Orbital plasmacytomas are very rare tumors constituting <1% of orbital tumors.[2] Although orbital involvement is more common than intraocular involvement, cases of plasmacytomas in conjunctiva, iris and ciliary body have been reported.[3] Proptosis, diplopia, pain and decreased vision are common symptoms in cases with orbital involvement.[4] Secondary angle closure and anterior synechiae have also been reported in cases with intraocular involvement.[3] In this case report, we present a patient with orbital involvement for 1 week who was followed up for 3 years due to MM.

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Case Report

A 72-year-old male patient was admitted to our clinic because of a mass in the left eye that grew suddenly in 1 week and had decreased vision for 6 months. MM was diagnosed 3 years ago and complete remission was achieved with 5 cycles of Bortezomib, Cyclophosphamide and Dexamethasone combination therapy. After remission, autologous stem cell transplantation was performed in July 2022. Following nine cycles of lenalidomide maintenance treatment in the post-transplant period, two cycles of bortezomib, thalidomide, and dexamethasone (BTD) treatment were administered to the patient due to disease recurrence. Immunoglobulin G levels decreased after the 1st course of BTD treatment. However, biochemical recurrence of MM and a new mass lesion in the left eye were detected during follow-up after the 2nd course of BTD. The patient had a solid, vascularized mass in the left eye caruncle region, and the left eye deviated outward (Fig. 1). Eye movements were free in all directions. On ophthalmological examination, best-corrected visual acuity was 7/10 in the right eye and hand movements in the left eye. On biomicroscopic examination, the cornea was clear, the iris was normal, posterior chamber intraocular lens was present in the right eye, and cataract was present in the left eye. The intraocular pressure was 13 mmHg in the right eye and 11 mmHg in the left eye. Fundus examination of the right eye revealed no abnormalities. The fundus of the left eye could not be evaluated because of the cataract, but no vitreoretinal pathology was found on ultrasonography. Based on the patient's history and clinical findings, the lesion was thought to be MM metastasis, and radiological imaging was performed. Contrast-enhanced magnetic resonance imaging revealed a 14 × 10 mm mass with intense contrast enhancement in the medial left globe, compatible with metastasis (Fig. 2). In addition, there was another intracranial lesion measuring 12 × 6.6 mm in the posterior left globe with intense contrast enhancement (Fig. 2). A biopsy was obtained from the mass in the medial left globe for pathological examination and differential diagnosis. In the biopsy sample, atypical cells with monotonous plasmacytoid appearance and hyperchromic nuclei forming diffuse infiltration under the conjunctival epithelium were observed (Fig. 3). Immunohistochemical tests showed positive staining for CD138, CD38, and kappa, but no staining was observed for lambda and life cycle assessments (Fig. 3). Hematological tests revealed an increase in monoclonal protein levels. At this stage, a combination of dexamethasone, cyclophosphamide, etoposide, and cisplatin (DCEP) combination chemotherapy was administered. Response evaluation after two cycles of DCEP treatment showed a decrease in monoclonal protein levels and a decrease to normal limits. After treatment, the plasmocytoma in the left eye caruncle regressed both in clinical examination and radiological imaging (Fig. 4).

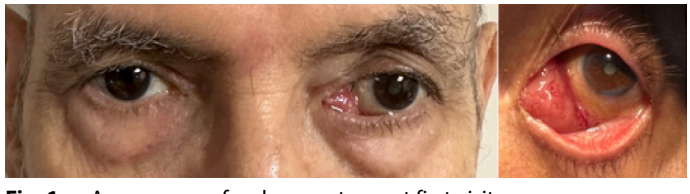

Fig. 1. Appearance of a plasmacytoma at first visit.

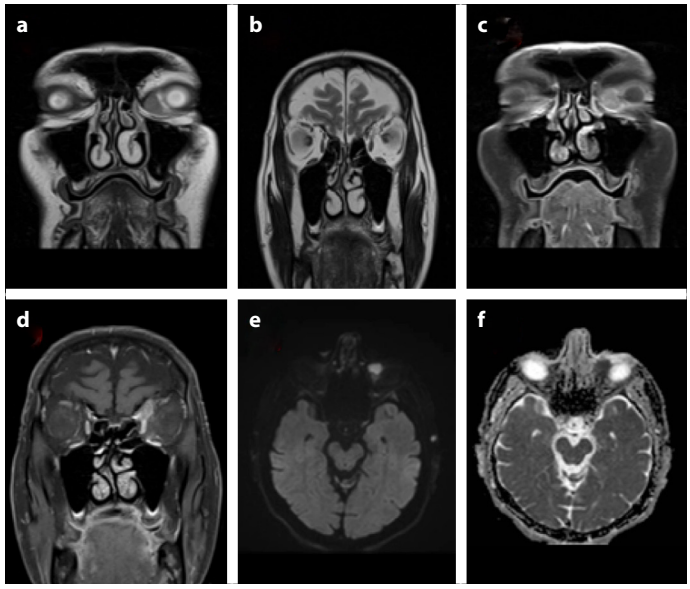

Fig. 2. Magnetic resonance images of the tumor. **(a and b)** There are 2 hypointense solid lesions in the medial left orbit on T2-weighted images. **(c and d)** Intense contrast enhancement of the lesions was observed on contrast-enhanced fat-suppressed T1-weighted images. **(e)** Hyperintense in diffusion weighted imaging. **(f)** Apparent diffusion coefficient was also hypointense and diffusion restriction was observed.

Discussion

Extramedullary plasmacytomas are extremely rare tumors in MM.[5] The head and neck region, paranasal sinuses, upper respiratory tract and salivary glands are the sites of approximately 90% of extramedullary plasmacytomas.[6] Orbital involvement is a very rare clinical entity.[1] In most cases, clinical findings include slowly progressive painful proptosis, diplopia, vision loss, and limitation of eye movements. Since the prognosis is worse in patients with systemic involvement, early diagnosis is important in terms of prognosis.[1] As the first extramedullary

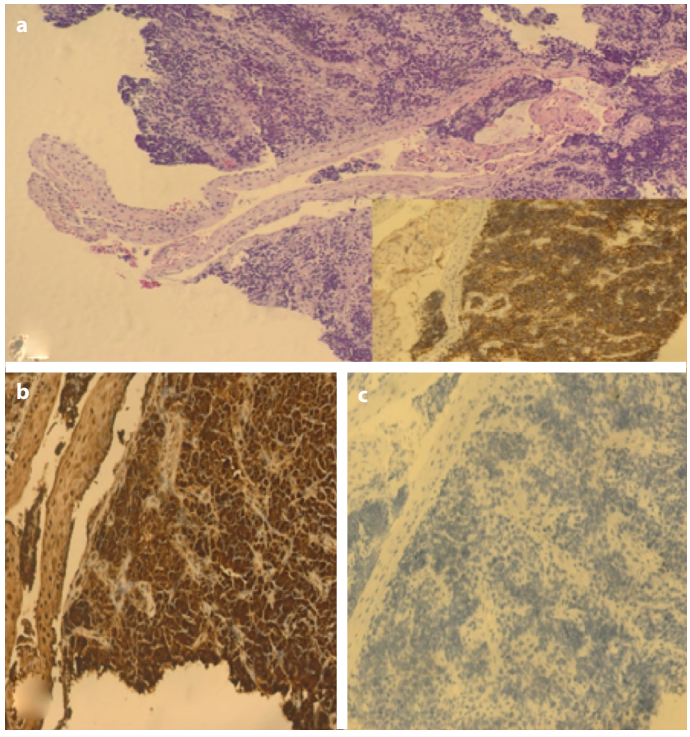

Fig. 3. Pathological sections of the tumor. **(a)** Histopathological examination shows dense infiltrate of plasma cells in the upper conjectival epitelium (Hematoxylin and Eosin × 200) immünohistopathologically positive CD38 (small figure). **(b)** It is showed in immunohistochemistry: Kappa (B × 200). **(c)** It is showed in immunohistochemistry: Lambda (B × 200).

involvement may be in the orbit, MM cases with atypical involvement should be kept in mind for early diagnosis and prognosis.[2]

Ophthalmological findings in patients with MM are caused by infiltration of tissues with plasma cells and hyperviscosity syndromes. Orbital plasmacytomas have a locally aggressive course and it has been suggested that they are frequently seen in the upper temporal quadrant.[7] Involvement in this region is common due to the rich vascular network of the lacrimal gland and the large bone marrow of the sphenoid bone.[5] In our case, contrary to expectations, plasmacytoma located in the caruncle suggests that extramedullary localization may be atypical.

The incidence and prevalence of caruncle malignancies are unknown; however, they are estimated to be rare, accounting for 1% of all ophthalmological biopsy specimens.[8] Delays in diagnosis and treatment can lead to morbidity and mortality.[9] Caruncle lesions are usually enlarged within months, and redness and pigmentation can be observed.[9] As seen in our case, lesions with rapid growth, irregular surface and vascularization are suspicious for malignancy. Lymphoma, basal cell carcinoma, squamous

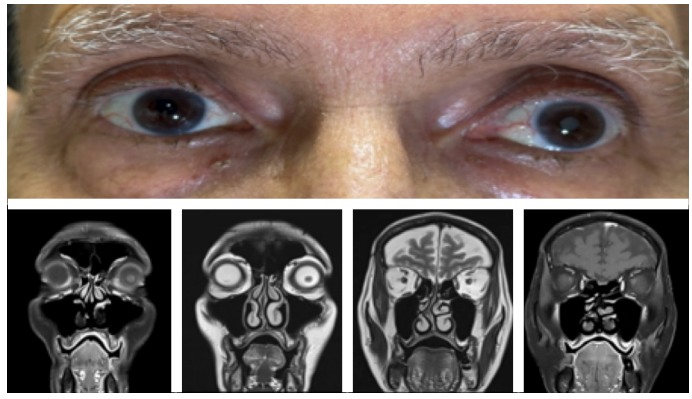

Fig. 4. External view of the patient after treatment. In the MRI taken after the treatment, it was observed that the size of the lesion located anteriorly in the medial left orbit had significantly regressed, and the lesion located posteriorly in the medial left orbit had completely disappeared.

cell carcinoma, sebaceous carcinoma, and melanoma have previously been reported in the caruncle.[9-11] MM may involve orbital and intraocular structures, either primary or metastatic.[3] Although one case of plasmacytoma in the lower eyelid, conjunctiva, fornix, and caruncle has been reported in the literature, we did not encounter an isolated case of caruncle metastasis.[12]

Conclusion

Orbital plasmacytoma may be the first manifestation of MM or indicate recurrence. Urgent imaging and aggressive treatment after biopsy to confirm the diagnosis are important for the survival of patients with MM with orbital metastases. Atypically located orbital plasmacytomas should be considered during early diagnosis and treatment.

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