

CASE REPORT

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Plasma cell type Castleman's disease of lacrimal gland: a case report and literature review

Liangyuan Xu¹, Jing Li¹, Xiaolin Xu¹, Tingting Ren¹ and Jianmin Ma^{1*}

Abstract

Background Orbital Castleman's disease is a rare benign lymphoproliferative disorder of extranodal origin.

Case presentation A 72-year-old man presented with bilateral upper eyelid swelling. Radiology revealed enlarged lacrimal glands in both eyes. Castleman's disease (plasma cell type) was confirmed by histo-immunochemical examinations after excision of the left orbital mass. No extraorbital involvement was found in systemic evaluation. Postoperatively, corticosteroid therapy was given. After 1 year of follow-up, there was no recurrence in the left orbit, and the swelling of the right eyelid was improved.

Conclusions Here, we report a case of orbital Castleman's disease involving the lacrimal gland bilaterally and summarize previously reported cases. For bilateral orbital masses, the possibility of Castleman's disease should be considered. Before the diagnosis, it is necessary to fully distinguish from other diseases such as IgG4-related eye diseases.

Keywords Castleman's disease, Lacrimal gland, Orbit, Plasma cell type

Background

Castleman's disease (CD), also known as giant lymph node hyperplasia or angiofollicular lymph node hyperplasia, is a rare and uncertain chronic lymphoproliferative disease with high heterogeneity [1–3]. Orbital CD is an extranodal lymphoproliferative disease of benign lymphoid tissue, which is caused by abnormal proliferation of lymphoid cells in orbit [4, 5]. We describe a case of plasma cell type CD involving bilateral lacrimal glands and summarize the reported cases of orbital CD confirmed by pathology in order to increase the experience of diagnosis and treatment of orbital CD.

Case presentation

A 72-year-old man presented with bilateral upper eyelid swelling for one year. The patient reported a palpable, progressively enlarged mass on the left upper eyelid and lacrimation for nearly one month without visual deterioration, diplopia, proptosis. He had no systemic symptoms such as weight loss, fever, chills and night sweats, and denied any history of trauma, systemic illness or familial hereditary disease. The best-corrected visual acuity for both the right eye and left eye was 20/20. Intraocular pressure was 16 mmHg and 15 mmHg in the right and left eye, respectively. Both eyelids were swollen, and the left side was more noticeable. A local painless mass was palpable under the left temporal orbital rim, which extended into the orbit, with well-defined, non-movable and firm in consistency. The patient was orthophoric in primary gaze, and there was no restriction on movement. No associated ocular involvement was found in the anterior segment of both eyes, and no changes such as choroidal folds were detected in the fundus examination.

*Correspondence:

Jianmin Ma

jmma@sina.com

¹ Beijing Institute of Ophthalmology, Beijing Tongren Eye Center, Beijing Tongren Hospital, Capital Medical University, Beijing 100730, China



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The test results of human herpesvirus 8 (HHV-8), hepatitis B virus (HBV), hepatitis C virus (HCV) and human immunodeficiency virus (HIV) were all negative. Orbital computed tomography (CT) showed bilateral lacrimal glands enlargement without invasion to the bone (Fig. 1). Orbital magnetic resonance imaging (MRI) revealed solid masses in the bilateral lacrimal fossa, which suggested the possibility of lymphoproliferative lesions (Fig. 1).

The patient underwent excisional biopsy via lid crease approach in the left eye. The well-encapsulated, pink mass with a size of 2.5 cm×1.6 cm×1.2 cm was examined by histopathology and immunohistochemistry, and it was found to be consistent with the characteristics of plasma cell type Castleman's disease. Microscopically, numerous plasma cell infiltrates were seen between atrophic lymphoid follicles (Fig. 2A-C). The immunohistochemical results were: CD3+, CD20+, CD21+, CD38+, Bcl2(+), and Ki-67 (germinal center+); and IgG4/IgG ratio < 40% following pathological immunofluorescence investigation, with 15 IgG4+ plasma cells per high power field (HPF) and 40 IgG-positive plasma cells per high-power field (HPF) on microscopy (Fig. 2D-I).

After the operation, the patient was treated with methylprednisolone 80 mg infusion for 3 days, and then taken methylprednisolone 24 mg orally, with reducing 4 mg every 7 days. He also had improvement of the eyelid

swelling in his right eye a few weeks after starting steroid therapy. The patient was referred to the Hematology and Oncology Department for a comprehensive evaluation, which confirmed the absence of systemic disease. After one month's oral treatment, the therapy stopped after the patient had a resolution of symptoms at the outpatient review. At the 12-month follow-up, there was no evidence of recurrence.

Discussion and conclusions

CD is a rare idiopathic lymphoproliferative disease, which belongs to benign lesions, but tends to be malignant and can be transformed into Hodgkin's lymphoma or non-Hodgkin's lymphoma [6]. It was first reported and defined in mediastinal lymph nodes by Castleman et al. in 1956 [7]. CD can be classified into unicentric CD (UCD) and multicentric CD (MCD). MCD is subdivided into HHV-8 associated MCD and HHV-8 negative/idiopathic MCD (iMCD) [8]. HHV-8 associated MCD is common in immunosuppressed people, especially HIV positive patients. HHV-8 can escape the host immune response and is obviously related to the occurrence of lymphoproliferative diseases [5, 9]. In different studies, the proportion of the two clinical types is quite different. UCD accounts for 47.6%-79.7% of CD in many domestic research reports [10-13].

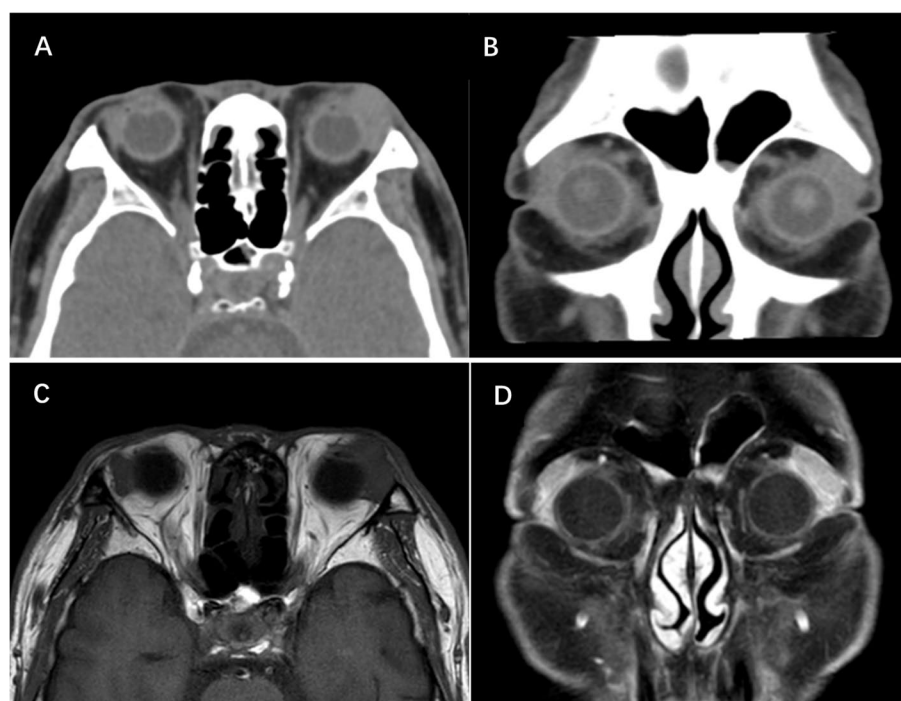


Fig. 1 Computed tomography (CT) and magnetic resonance imaging (MRI) findings of orbits. CT images in axial view (A) and coronal view (B) revealed middle-density masses involving bilateral lacrimal glands. T1-weighted images in axial view (C) showed slightly hypointense signal. T1-weighted contrast-enhanced images in coronal view (D) showed homogeneous enhancement of the lesions

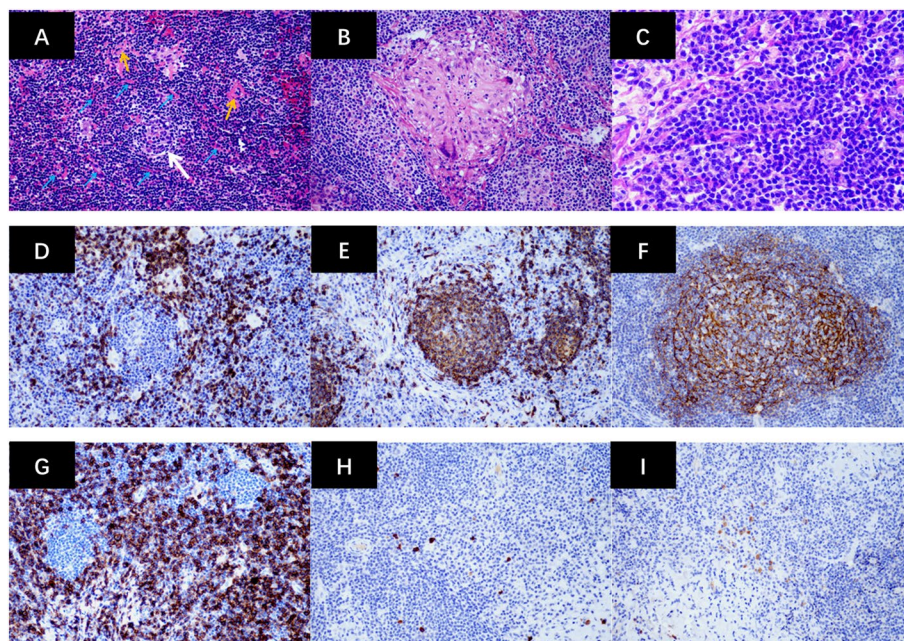


Fig. 2 Histopathology showing typical characteristics of Plasma cell type in Castleman's disease. **A** Showed atrophy and shrinkage of germinal centers and lymphoid follicles (white arrow), proliferation of blood vessels in the interfollicular area (blue arrow), and hyalinization of blood vessel walls (orange arrow) (HE, $\times 200$). **B** Showed epithelial-like cell granuloma reaction (HE, $\times 200$). **C** Showed infiltration of many plasma cells (HE, $\times 400$). **D** T lymphocytes positive for CD3 (IHC, $\times 200$). **E** B lymphocytes positive for CD20 (IHC, $\times 200$). **F** Follicular dendritic cells net positive for CD21 (IHC, $\times 200$). **G** Plasma cells positive for CD38 (IHC, $\times 200$). **H** Plasma cells scattered positive for IgG4 (IHC, $\times 200$). **I** Plasma cells scattered positive for IgG (IHC, $\times 200$)

American research reports that UCD accounts for 75% of CD, while Japanese research reports that UCD only accounts for 30% of the incidence of CD [8, 14].

CD mainly occurs in lymph nodes, the most common sites involved are abdominal, mediastinal and cervical lymph nodes, and various extranodal sites can also be involved, including those lacking lymphatic vessels and lymphoid tissues such as orbit [6, 15]. In 1989, Gittinger first reported a case of MCD with intraocular and orbital involvement, but no eye biopsy was performed [16]. In 1993, Snead et al. reported the first case of orbital UCD confirmed by pathology [17].

Most cases of orbital UCD present as a solitary mass [18]. The common clinical manifestations are exophthalmos, eyelid swelling, ptosis, strabismus, diplopia, and limited eye movement, while some tumors are visible or palpable [8]. MCD is associated with diffuse lymphadenopathy and may present with more severe systemic symptoms and signs, including hepatomegaly, splenomegaly, edema, rash, arthralgia, myalgia, and neurological abnormalities, and is often associated with abnormal laboratory findings such as anemia, elevated erythrocyte sedimentation rate, proteinuria, hypergammaglobulinemia, and abnormal liver function [19].

Histopathological examination combined with immunohistochemical examination is the gold standard for diagnosis [20]. The histopathological types of CD can be divided into three types: hyaline-vascular type, plasma cell type, and mixed cell type with the characteristics of the first two types [21]. UCD is mostly hyaline-vascular type, accounting for 70%~90% of UCD patients [3, 10]. Hyaline-vascular type CD is mainly characterized by proliferation of scattered lymphatic follicles and hyalinization of small blood vessels between follicles, with the appearance of a 'lollipop on a stick' [22, 23]. Plasma cell type is the most common type of MCD, accounting for 86.0% of MCD [10, 14]. The characteristic histopathological finding of plasma cell type CD is the presence of solid confluent plasma cell sheets in the interfollicular zone [22]. Follicular centers are usually enlarged compared to the atrophic follicular centers observed in hyaline-vascular type CD [22]. Paracortical hypervascularity and onionskin mantle zones are usually absent [22]. The mixed cell type is the coexistence of the above two types, which is manifested as hyaline vascular lesions with numerous plasma cells [18]. Other diagnoses need to be excluded before the diagnosis of plasma cell type CD, especially when bilateral lacrimal glands are involved, it

needs to be differentiated from IgG4-related eye diseases [4]. In our case, the patient's pathological examination did not meet the criteria for IgG4-related eye diseases. In addition, the epithelioid granulomatous changes in the histopathological staining of this case are not generally observed in IgG4-related eye diseases.

UCD mostly has a good prognosis and can usually be treated by surgical excision of the focal lymphoproliferative mass, which is curative in most cases [21]. MCD often requires active systemic therapy and predicts a poor outcome [22]. The treatment of MCD include

surgical excision, chemotherapy, radiotherapy, corticosteroids, immunomodulators, and monoclonal antibodies [21].

Table 1 summarized the clinical data of cases of orbital CD confirmed by surgical excision or biopsy pathology retrieved by PUBMED [4, 5, 15, 17, 18, 21–30]. The clinical subtypes of CD patients with bilateral orbital masses and no systemic involvement are still controversial. The author categorized this condition as multicentric CD. In the Table 1, Male patients accounted for 55% (11/20) and female patients accounted for 45% (9/20). The age

Table 1 Orbital involvement in Castleman's disease confirmed by surgical excision or biopsy: A summary

Case report	Year of publication	Sex	Age	Laterality	Involved site	Clinical subtype	Histopathological subtype	Treatment	Outcome
the present case	2024	M	72	bilateral	lacrimal gland	MCD	PC	SE + C	no recurrence in a year
Liu LC [4]	2023	M	67	bilateral	lacrimal gland without systemic evaluation	MCD	Mixed	SE	no recurrence in a month
Li D [5]	2020				lacrimal gland				follow-up for 2 to 5 years:
		M	76	unilateral		UCD	PC	SE + C	no recurrence
		F	61	bilateral		MCD	HV	SE + C	no recurrence
		F	76	unilateral		UCD	PC	SE + C	no recurrence
		F	46	unilateral		UCD	PC (transformation to plasma cell tumor)	SE	recurrence
		F	44	bilateral		MCD	PC	SE + C	recurrence
Goel R [24]	2020	M	62	unilateral	lacrimal gland	UCD	Mixed	SE	no recurrence in 3 years
Kang D [25]	2015	M	53	unilateral	intraconal space	UCD	HV	SE	no recurrence in 13 months
Mukherjee B [26]	2014	M	48	bilateral	extraconal space without systemic evaluation	MCD	HV	SE	unknown
Jones NW [21]	2013	F	17	unilateral	extraconal space	UCD	HV	SE	no recurrence in 10 months
Brubaker JW [23]	2011	M	53	unilateral	intraconal space	UCD	HV	SE	unknown
Venizelos I [15]	2010	M	70	bilateral	intraconal space lymph nodes	MCD	PC (biopsy)	C	no recurrence in 6 months
Jáñez L [27]	2010	M	69	unilateral	extraconal space	UCD	HV	SE	no recurrence in a month
Rivas DM [28]	2004	M	52	unilateral	intraconal space	UCD	HV	SE	unknown
Koppens JM [29]	2004	F	84	unilateral	lacrimal gland	UCD	HV	SE	unknown
Ide M [30]	2003	M	61	bilateral	orbital soft tissue lymph nodes	MCD	HV (biopsy)	RT + I + C	no recurrence in 10 months
Park KS [22]	2002	F	18	bilateral	extraconal space	MCD	HV	SE	no recurrence in a year
Alyahya GA [18]	2002	F	19	unilateral	extraconal space	UCD	Mixed (biopsy)	C + I + RT	stable condition in 7 years
Snead MP [17]	1993	F	26	unilateral	lacrimal gland	UCD	HV	SE	no recurrence in 5 years

M Male, F Female, UCD Unicentric CD, MCD Multicentric CD, Mixed Mixed cell type, HV, Hyaline-vascular type, PC Plasma cell type, SE Surgical excision, C Corticosteroids, I Immunosuppressives, RT Radiotherapy, SEU Systemic evaluation was unclear

of onset ranged from 17 to 84 years (mean 53.7 years). Among them, UCD accounted for 60% (12/20) and MCD accounted for 40% (8/20). The pathological types of UCD patients were hyaline-vascular type in 7 cases (58.33%), plasma cell type in 3 cases (25%) and mixed cell type in 2 cases (16.67%). The pathological types of MCD patients were hyaline-vascular type in 4 cases (50%), plasma cell type in 3 cases (37.5%) and mixed cell type in 1 case (12.5%). The prognosis of CD patients was generally good. One case of recurrence occurred in each of the UCD and MCD patients. Both recurrent patients were involved in lacrimal gland, and the pathological type was plasma cell type.

Among the patients with eye involvement in MCD who did not undergo eye biopsy, Barile et al. reported a case of ocular involvement of MCD with initial symptoms including visual acuity changes with exudative retinal detachment in both eyes, which resolved during treatment with high-dose systemic corticosteroids [19]. Samoszuk et al. reported a case of MCD with unilateral involvement that progressed to exophthalmos in both eyes after 7 months of local radiotherapy [31]. Kurokawa et al. reported a case of MCD with bilateral lacrimal gland involvement who was treated with corticosteroids, relapsed after 10 months, and eventually died due to systemic infection with Cytomegalovirus and *Aspergillus* [32].

In this paper, a case of plasma cell type CD with bilateral lacrimal gland involvement was reported. For patients with plasma cell type CD, the diagnosis should be made after full differentiation from other diseases, and it is imperative to utilize additional postoperative treatment modalities for consolidation and ensure follow-up.

Abbreviations

CD	Castleman's disease
UCD	Unicentric Castleman's disease
MCD	Multicentric Castleman's disease
CT	Computed tomography
MRI	Magnetic resonance imaging

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Not applicable

Authors' contributions

XLY drafted the manuscript, collected the data and reviewed the literature. LJ interpreted the data, reviewed the literature and helped to draft the manuscript. XXL and RTT helped the manuscript and follow-up data collection. MJM performed surgery, made diagnosis, and critically reviewed the manuscript. All authors read and approved the final manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Competing interests

The authors declare no competing interests.

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