• Letter to the Editor •

Unilateral solitary ciliary body mass: a case report and review of the literature

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Dear Editor,

Ciliary body masses are diagnostically challenging due to their hidden location, diverse pathologies, and limited examination methods[1]. We report a case of a ciliary body inflammatory mass treated with trans-scleral excision and anti-inflammatory therapy, preserving functional vision.

Ethical Approval This study was approved by the Ethical Committee of the First Affiliated Hospital, Zhejiang University School of Medicine. Written consents were obtained from the patient.

A 53-year-old male presented with 3mo of recurrent left eye (OS) redness and acute vision blur. Initial examination revealed nasal sectoral anterior scleritis. Oral methylprednisolone (16 mg/d) and topical anti-inflammatory drops (tobramycin-dexamethasone and pranoprofen four times a day) for 1mo showed poor response. Visual acuity was 20/100, with an intraocular pressure (IOP) of 13 mm Hg, accompanied by nasal scleritis, anterior chamber and vitreous cells, and a superonasal yellow-white ciliary body mass (3.2×5.5 mm^2) associated with retinal edema and hemorrhage (Figure 1A). B-scan revealed a dome-shaped solid mass with vitreous opacities (Figure 1B). Empirical intravitreal dexamethasone (400 μg), vancomycin (1 mg), and ceftazidime (2 mg) were administered with intravenous dexamethasone (5 mg/d) and cefuroxime (1.5 g/d). After one week, scleritis/uveitis improved (Figure 1C, 1D), but the mass persisted. Vitreous cultures were negative. Ultrasound biomicroscopy (UBM) showed a 2.4×5.2 mm^2 ciliary body mass (9–11 o'clock) with medium reflectivity (Figure 1E, 1F). Magnetic resonance imaging (MRI) demonstrated a T1-isointense and T2-hypointense superonasal enhancement (Figure 1G, 1H). Serological testing, including C-reactive protein, rheumatoid factor, antinuclear antibody, and immunoglobulin levels, as well as chest computed tomography, were unremarkable except for positivity for hepatitis B surface antigen. The patient had no history of tuberculosis, rheumatic disorders, or sarcoidosis. Notably, he had prior leg trauma with fever/rash preceding ocular symptoms, though causality remained unclear.

Trans-scleral excision biopsy was shown in Figure 2A, 2B. Microscopic examination showed inflammatory cell infiltration located in the episclera, made up of small lymphocytes with fibrous tissues, and chronic purulent inflammation with lymphocytes hyperplasia located in the ciliary body biopsy (Figure 2C, 2D). Immunohistochemical (IHC) examination demonstrated infiltration of CD3-positive T lymphocytes and CD20-positive B lymphocytes into the episclera. In the ciliary body, immunomarkers for melanoma, including S-100 protein, human melanoma black-45 (HMB-45), and tumor protein P53, were all negative, and no well-formed lymphoid follicles or granulomatous inflammation were observed.

Based on these findings, pathological diagnosis was non-specific inflammatory of the sclera and chronic purulent inflammation of the ciliary body tissue. Postoperative treatment included tapered oral methylprednisolone (16 mg/d for 5d and 8 mg/d for 5d) and topical anti-inflammatories/antibiotics. At 4-month follow-up, vision improved to 20/50 with resolved inflammation (Figure 2E, 2F).

In our case, there was a histological difference between the local sclera and the ciliary body, with the former presenting as non-specific inflammation and the latter as chronic purulent inflammation. Determining which tissue was the first to be affected, the sclera or the ciliary body, seems challenging. This difficulty arises because changes in these two tissues may interact with each other, and lesions in the ciliary body are often too hidden to be detected. However, we tend to believe that the onset occurred in the ciliary body, while the scleral inflammation was a reactive response, based on the histological

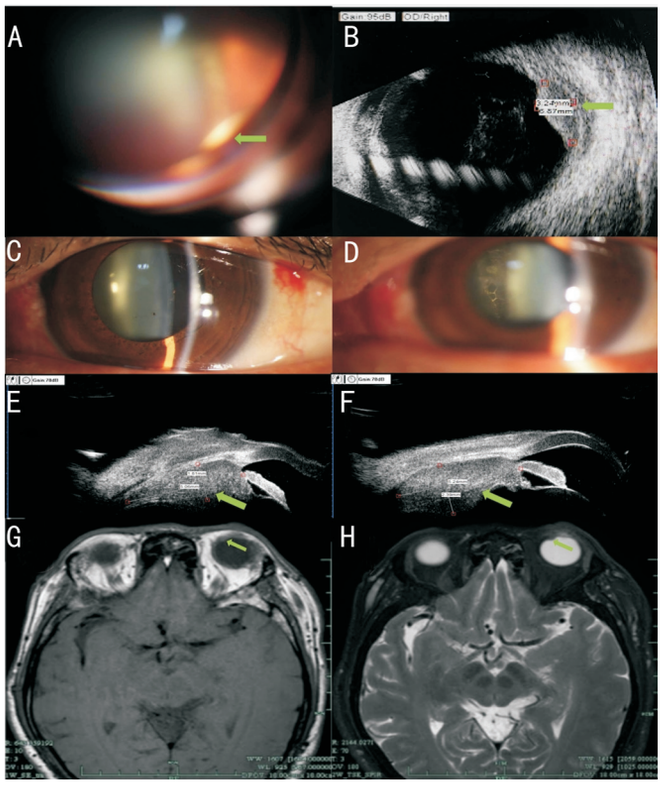

Figure 1 Slit-lamp examination and imaging of the patient's left eye before surgery A: Slit-lamp photography (with digital wide field) showed a ciliary body mass in yellow-white color on the supero-nasal side; B: B-scan ocular ultrasound revealed a dome-shaped, acoustically solid mass with vitreous opacities, especially near the mass; C and D: Slit-lamp photography showed anterior chamber cell and vitreous cell one week after vitreous aspiration and empirically intravitreal injection of dexamethasone, vancomycin, and ceftazidime from the temporal conjunctiva; E and F: Ultrasound biomicroscopy demonstrated a ciliary body mass with middle internal reflectivity, extending from 9 to 11 clock hour; G and H: Magnetic resonance imaging revealed the mass with isointensity in T1-weighted imaging and hypointensity in T2-weighted imaging. The green arrows indicate the location of the mass in each image.

characteristics and the relatively long course of the disease. Reports suggesting that when a persistent episcleral lesion is present, a ciliary body lesion should be sought lend credibility to this hypothesis[2]. The second question is what caused the ciliary body inflammation. Considering the patient had no history of systemic autoimmune diseases, the OS disease might be related to the systemic reaction to his stabbed leg, which caused a whole-body rash and fever. Disappointingly, no microorganisms were identified in our case, whether through systemic or local examination. However, the limitations of laboratory and histopathologic examinations should also be considered under these circumstances[3].

As known, the pathological types of ciliary body mass are various and complicated. Apart from benign and malignant tumors, some cases have reported a rare type: idiopathic

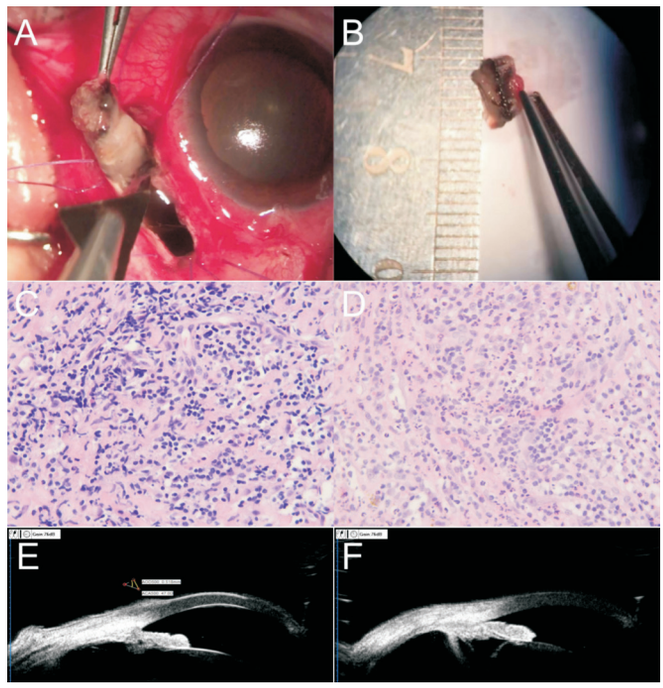

Figure 2 Trans-scleral excision biopsy and UBM imaging after surgery A: Trans-scleral excision biopsy of the ciliary body mass was performed; B: Excisional biopsy showed the size and the appearance of the lesion; C and D: Hematoxylin and eosin stain showed the mass being mainly consisted of small lymphocytes and fibrous tissues in the sclera (C), and chronic purulent inflammation with lymphocytes hyperplasia in the ciliary body biopsy (D), magnification ×400; E and F: Resolution of ciliary body mass after trans-scleral excision biopsy demonstrated by UBM. UBM: Ultrasound biomicroscopy.

solitary granuloma of the uveal tract[3-4]. However, to our knowledge, this is the first reported case of a non-granulomatous inflammatory mass of the ciliary body. Among the previous reports, only one eye retained a final visual acuity of 20/25, while the others all resulted in profound vision loss within months and necessitated enucleation shortly thereafter[3-4]. By comparison, there are some similarities between our case and the solitary granuloma[4]: both exhibited long-term redness, blurry vision, and were found to have sectoral anterior scleritis, anterior and intermediate uveitis, and a ciliary body mass; both UBM examinations showed middle internal reflectivity of the mass; and no infectious evidence or relevant systemic diseases were found. However, in our case, the pathological examination revealed mainly lymphohistiocytic hyperplasia but no fibrosis, which failed to form a granuloma. As described above, we suppose that our case may be an early and moderate stage of idiopathic solitary granuloma. Besides, differentiation from Immunoglobulin G4 (IgG4)-related intraocular inflammation and ciliary body melanoma is also needed. IgG4-related disease is a relatively new and uncommon clinical entity seen in various organs and tissues of the body, associated with elevated serum levels

of IgG4[5-6]. One recent case reported the first instance of an intraocular IgG4-associated inflammatory mass in the ciliary body mimicking melanoma in a 23-year-old female[7]. In that case, there was profound vision loss: counting fingers at 3 m and also severe anterior uveitis: 360° posterior synechiae with anterior chamber flare and cells (+++) with a 1 mm hypopyon. Microscopic description of the ciliary body showed diffuse plasmacytoid cells, with reactive lymphoid follicle and other inflammatory cells, and immunohistochemistry for IgG4 was positive in occasional cells, which also expressed CD138 (plasma cells). Serum IgG4 levels were also elevated. Thus, this seemed not to be the same case as our patient, either in symptoms or histopathologic characteristics. Another similar report described an IgG4-related ophthalmic disease with an inflammatory mass in the ciliary body and scleritis; however, that case showed sequential bilateral involvement, with enucleation of the misdiagnosed left eye, histopathology and IHC results supporting probable IgG4-related, and the right eye responding well to long-term corticosteroid therapy as demonstrated by cytokine profiling and multimodal imaging[8]. Ciliary body melanomas are less commonly seen compared to those of either the iris or choroid but have the worst prognosis of all intraocular melanomas[9], so all ciliary body masses should be considered potential melanomas. They can present when they are quite large, in part because they are hidden from view (behind the iris). When they become large enough to cause symptoms, they have grown to displace the iris, lens, or extend into the visual axis[1,10]. In our case, although the mass had a yellow-white color, amelanotic melanoma could not be excluded because the UBM, B-scan ocular ultrasound, and MRI all had similarities to melanoma, necessitating a histological biopsy to achieve an exact diagnosis. Likewise, although rare, malignant ciliary body medulloepithelioma must be ruled out due to its predominance in children (median age 9y), neuroepithelial origin with neuron-specific enolase (NSE)/S-100 positivity, and characteristic primitive neuroblastic foci on histopathology[11]. Ocular sarcoidosis should also be excluded, as it is characterized by uveitis and a diffuse or segmental ciliary body mass, with histopathological findings of granulomatous inflammation[12].

In conclusion, when ophthalmologists noticed persistent sectoral scleritis, ciliary body mass such as melanoma, medulloepithelioma, ocular sarcoidosis and idiopathic solitary granuloma as well as IgG4-related disease should be included as differential diagnoses. Careful observation, especially UBM, is necessary to avoid missing the ciliary body lesions. As if it can be found in a relatively small size, timely trans-scleral excision combined with systemic and local anti-inflammatory and anti-infective drugs (if histopathologically indicated as inflammatory lesion) may receive good effect.

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