CASE REPORT

A case of a rapidly progressing, recurrent orbital mass

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ABSTRACT

Background and aim: We present a case of fast-growing, recurrent orbital mass which was ultimately diagnosed as sarcoidosis in a patient who lacked systemic symptoms.

Methods: A case report was conducted.

Results: A 58-year-old woman from Pakistan presented with a ortbial mass that is hyperintense orbital mass on MRI. Excisional biopsy and pathology revealed non-necrotizing granulomatous inflammation. Negative AFB culture, GMS, and flow cytometry ruled out TB and lymphoma. Chest X-ray was unremarkable. ACE and Lysozyme levels were marginally elevated. She presented with symptomatic recurrence of mass requiring repeat excision. Chest CT confirmed the presence of pulmonary nodules and hilar lymphadenopathy. She was started on corticosteroids and methotrexate with resolution of local inflammation and residual mass effect.

Conclusions: Chest CT has higher sensitivity to aid definitive diagnosis in the setting of negative chest X-ray. Surgical treatment may be useful for diagnostic and therapeutic purposes. Subsequent long-term treatment with corticosteroids and antimetabolites has shown great response.

Key words: orbital mass, granuloma, sarcoidosis, lymphoma, recurrent

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Introduction

The differential diagnosis of a fast-growing orbital mass is broad, including orbital tumors such as lacrimal gland tumor, rhabdomyosarcoma, orbital lymphoma, inflammatory conditions such as thyroid orbitopathy, sarcoidosis, or igG-4 related disease (1), and infectious disease etiologies include bacterial, fungal, and parasitic (2). Sarcoidosis is a systemic inflammatory condition which can have a variable local presentation. The percentage of ocular involvement ranges from 10% to 71%. The most common manifestation of ocular sarcoidosis is uveitis followed by conjunctival nodules. Less common manifestations include eyelid granulomas, conjunctivitis, episcleritis/scleritis, peripheral ulcerative keratitis, glaucoma, cataracts, retinitis, choroiditis, lacrimal gland granuloma, extraocular muscle granuloma, or optic nerve disease. We present a unique case of a fast-growing, recurrent primary orbital sarcoidosis (3).

Case presentation:

A 58-year-old female patient from Pakistan presented to the emergency department with right eye tearing, itching, pain, and swelling, and refered to an oculoplastic surgeon. Exam showed a firm, tender, non-mobile mass was palpated over the supranasal area (Figure 1A). Visual acuity was 20/30 in the right eye and 20/25 in the left eye. Eye pressures and pupils were normal without afferent pupillary defects.

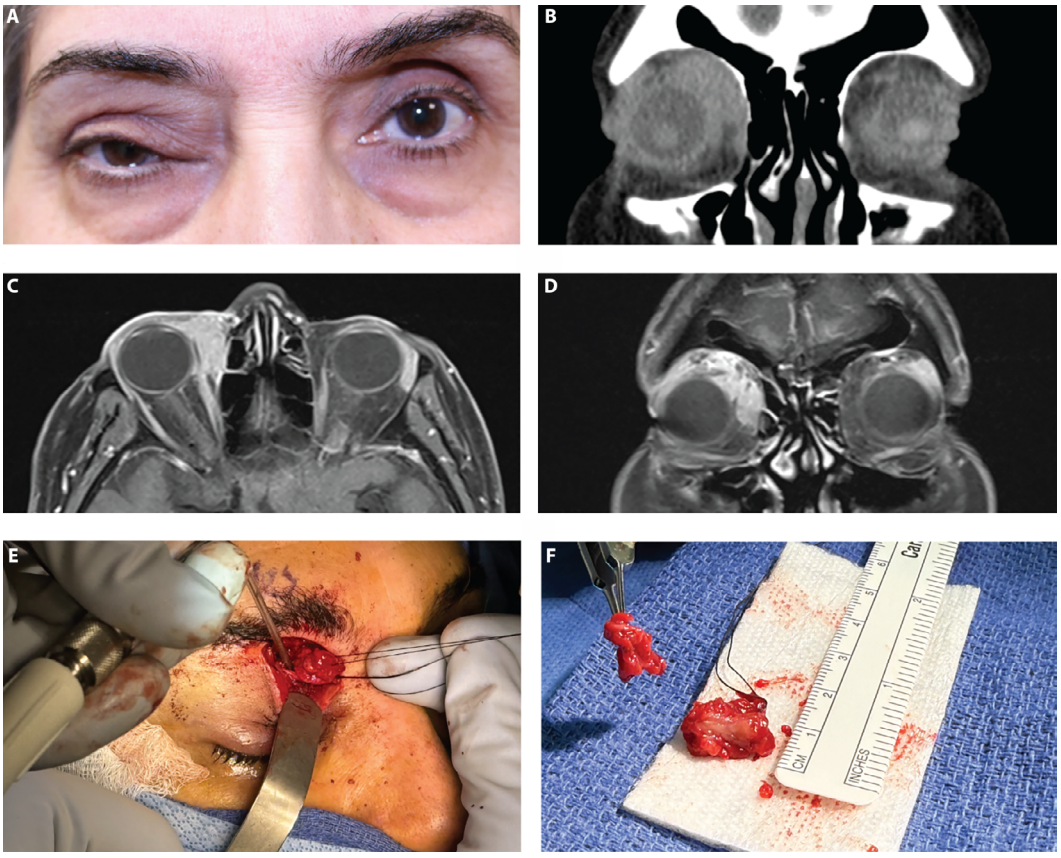

Figure 1. A) Exterior photograph of the patient presented with ptosis and orbital mass. B) Coronal view of CT orbit showing the orbital mass in the right orbit. C) Transverse view and D) Coronal view of MRI orbit showing the orbital mass in the right orbit. E) Intraoperative photograph of the mass during orbitotomy procedure. F) Exterior specimen photograph of the bi-sected orbital mss removed from the patient.

Visual fields and extraocular motion were full. Fundus exams were normal. Initial MRI imaging showed a homogeneously hyperintense mass with irregular borders involving the medial rectus and superior oblique muscles (Figure 1B, C, D). The patient did not report any systemic symptoms or relavent past medical history. Social history was notable for recent travel to Pakistan.

A diagnostic and therapeutic orbitotomy was performed in the operating room due to the patient's increasing discomfort and eye displacement, revealing a grey-white mass, rubber-like in texture (Figure 1E, F) which measured 25mm x 30mm. Hematoxylin and Eosin (H&E) stain showed confluent noncaseating granulomas with multinucleated giant cells. Acid-Fast Bacillus (AFB) culture, Ziehl-Neelson and Grocott's methenamine silver (GMS) stains were negative for tuberculosis and fungal etiologies, respectively. Flow cytometry revealed 90% lymphocytes with no aberrant antigen expression and a polyclonal kappa:lambda ratio (1.68:1), ruling out a diagnosis of non-hodgkin lymphoma and acute leukemia. Rheumatology was consulted for additional recommendations. Chest X-ray was unremarkable. The following laboratory workup was within normal limits: Erythrocyte Sedimentation Rate (ESR), Anti-Neutrophil Cytoplasmic antibody (ANCA), Myeloperoxidase Antibody (MPO), anti-Proteinase 3 antibody (PR3), Gold Quantiferon test for tuberculosis, and Fungitell. Angiotensin-Converting Enzyme (ACE) level was mildly elevated to 83 (normal range for age 14-82). Lysozyme was elevated to 8.8 (normal range 3.2 - 7.6). While results of the laboratory tests ordered by Rheumatology were pending, the patient returned to the Ophthalmology clinic with recurrence of painful mass within three months of the excisional biopsy at the same location. Due to the severity of the pain and rapid recurrence of the mass, a repeat orbitotomy was performed. Repeat of pathology showed same results from prior. A chest CT was obtained and showed mediastinal and hilar adenopathy as well as multiple bilateral pulmonary nodules highly suggestive of sarcoidosis. Rheumatology initiated treatment with Prednisone 60 mg daily and Methotrexate 15 mg weekly. Prednisone was slowly tapered off over six months without recurrence of the orbital mass.

Discussion

Our case

We describe an atypical case of a rapidly progressing, recurrent orbital mass in a patient without systemic symptoms, inconclusive lab work, and a negative chest X-ray which was ultimately diagnosed as sarcoidosis on Chest CT. Chest X-ray has a lower sensitivity as compared to chest CT (4). The sensitivity of ACE and lysozyme in diagnosing systemic sarcoidosis varies substantially within the literature. The sensitivities of ACE and lysozyme are estimated to be between 40-80% and 42-60%, respectively (5, 6).

Orbital involvement in sarcoidosis

Ocular involvement is the presenting symptom in 20% to 30% of cases of sarcoidosis (2). Ocular sarcoidosis manifesting as orbital mass is more common in patients above the age of fifty and in women (7). Orbital masses are most commonly located in the lacrimal gland, followed by orbit, eyelid, and lacrimal sac (7). Among the patients with an orbital location, the anterior inferior quadrant is the most common. Our patient's mass was located in the anterior superomedial quadrant, which is less common. The superior quadrant location was only reported in 1/26 patients in a review of orbital and adnexal sarcoidosis. Our patient presented with involvement of the medial rectus and superior oblique muscles. In the literature, involvement of the extraocular muscles was seen in 6/26 cases (7).

Treatment and prognosis

Literature review shows that surgical intervention with excisional biopsy is useful in making a diagnosis of sarcoidosis, and a suitable option for patients who have pain and rapid recurrence. Surgical excision may also be warranted in masses involving structures such as the optic nerve and extraocular muscles. Surgical excision should not be performed without subsequent treatment with oral steroids, as the risk of recurrence is high (8, 9). Antimetabolites including azathioprine and methotrexate have been used in cases refractory to oral steroids. The treatment duration may

vary from months to years and requires close follow-up. Our patient responded well to the addition of methotrexate (10).

Conclusion

This case highlights key factors in early detection and prompt management of ocular sarcoidosis. A chest CT should be ordered when clinical suspicion of ocular sarcoidosis is high and chest X-ray is indeterminate or negative. Serum ACE or Lysozyme can be used as adjuncts that are neither sensitive nor specific. Surgical management is helpful in diagnosis and can provide rapid symptomatic relief. Prompt treatment with steroids and immunosuppressants should be considered to prevent recurrence.

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Declaration on the Use of AI: No generative AI was used during the preperation of this manuscript

Consent for Publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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