

## Case report

## Orbital alveolar echinococcosis: A case report

Mohammad Taher Rajabi<sup>a</sup>, Saeed Mohammadi<sup>a</sup>, Narges Hassanpoor<sup>a,\*</sup>,  
Seyedeh Simindokht Hosseini<sup>a</sup>, Narges Shahbazi<sup>b</sup>, Seyed Ziaeddin Tabatabaie<sup>a</sup>,  
Mohammad Bagher Rajabi<sup>a</sup>

<sup>a</sup> Eye Research Center, Farabi Eye Hospital, Tehran University of Medical Sciences, Tehran, Iran

<sup>b</sup> Cancer Institute, Imam Khomeini Hospital, Tehran University of Medical Sciences, Tehran, Iran

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## Abstract

**Purpose:** To report a rare case of orbital alveolar echinococcosis in Iran.

**Methods:** A 23-year-old woman with multi-lobular mass lesion in the right orbit underwent excisional biopsy via a deep lateral orbitotomy approach. The pathologic investigation of the lesion was a multilobulated cystic lesion shown where the cyst wall structure was compatible with alveolar Hydatid cyst in histopathology. Clinical course, surgical, and medical management of the disease is noted briefly in the case of orbital involvement of the alveolar echinococcosis.

**Results:** Patient was treated with anti-fungal medication, and the cysts were successfully removed by a surgical excision.

**Conclusion:** Although orbital alveolar echinococcosis is extremely rare, it is noteworthy to study its clinical manifestations and radiological examinations to be able to make a true diagnosis.

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**Keywords:** Orbit; Echinococcosis; Alveolar; Hydatid cyst

## Introduction

Hydatid disease rarely involves the orbits, but the orbital involvement can occur in less than 1% of the population.<sup>1</sup> Similar to most of the orbital mass lesions, it presents itself as a painless unilateral proptotic eye.<sup>2</sup> Alveolar echinococcosis is a chronic zoonotic infection which is mostly caused by larval stage of *Echinococcus multilocularis* (*E. multilocularis*). The most common organ affected is the liver, and involvement of orbit is very rare.<sup>3,4</sup> Almost all mentioned

cases of orbital echinococcosis in the literature are uniloculated. Multiloculated cyst or alveolar echinococcosis is extremely rare in orbital region. Alveolar cysts are more likely to cause complications and are harder to treat in comparison with other hydatid cysts.<sup>5,6</sup> Patients should be diagnosed by clinical symptoms and orbital imaging to avoid complications.

## Case report

A 23-year-old woman came to Farabi Eye Hospital, Tehran, Iran complaining of progressive proptosis of right eye for the past 3 months (Fig. 1). The patient had no history of trauma, convulsion or loss of consciousness. She has also had no history of orbital surgery.

Although, visual acuity of both eyes was 10/10, the slit-lamp examination of anterior segment was normal and funduscopy showed no pathological alteration, the patient had

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Authors obtained informed consent from the patient to publish the photos.

\* Corresponding author. Eye Research Center, Farabi Eye Hospital, Qazvin Square, Tehran, 1336616351, Iran.

E-mail address: [nargeshassanpoor@gmail.com](mailto:nargeshassanpoor@gmail.com) (N. Hassanpoor).

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Fig. 1. The patient with proptosis and inferior displacement of the globe of right eye.

severe proptosis (13-mm difference in exophthalmometry) and infero-nasal globe displacement.

Magnetic resonance imaging (MRI) was done to investigate the cause of proptosis. There was a multiloculated cystic structure which had low intensity signal in T1 (Fig. 2: A and B) that displaced globe inferiorly. In T2 images, this cystic structure had high intensity signal (Fig. 2: C and D), and peripheral rim enhancement was seen after gadolinium injection (Fig. 2B). Due to the mentioned features of the lesion, the most probable diagnosis was hydatid cyst. The complete blood count (CBC) and erythrocyte sedimentation rate (ESR) were in normal range and Casoni test, ELISA and indirect haemagglutination (IHA) showed no sign of echinococcus infection.

Surgical en-bloc excision of the cyst was performed on the patient to prevent cyst-rupture and dissemination of the cyst's content into the orbit (Fig. 3). Since the MRI showed that the lesion is extended up to orbital apex, we decided to use “deep lateral orbitotomy” in this patient. The surgeon was able to access the superior orbital fissure with a deep lateral orbitotomy.

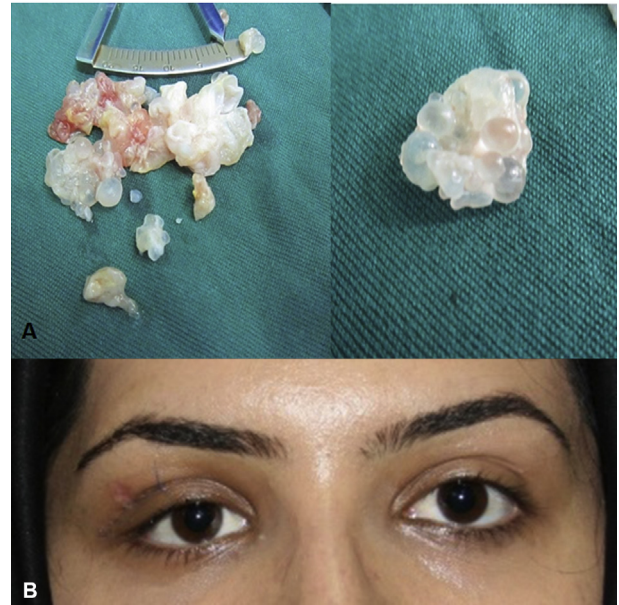


Fig. 3. The completely removed multiloculated hydatid cyst (A). Follow-up of the patient one week later (B). No recurrence was seen also at 1-year follow-up.

In addition, because our patient had no systemic involvement in clinical examination in both first and last follow-ups, and nothing abnormal was found in investigation of the internal viscera such as liver and spleen by ultrasonography, this presentation is considered the primary orbital presentation of the *E. multilocularis*. Systemic Albendazole was administered in a standard dosage (10 mg/Kg) for 3 months to prevent

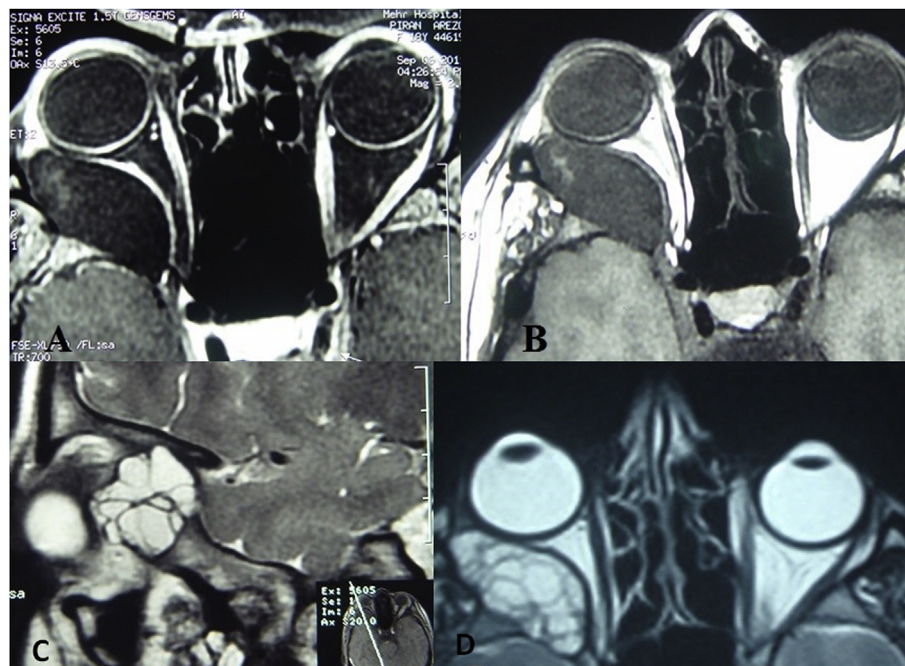


Fig. 2. T1-weighted axial magnetic resonance imaging (MRI) with fat suppression (A) showing an extraconal fusiform lesion that is hypointense in nature and has eroded the lateral wall of the orbit. (B) showing “ring” enhancement of the lesion after gadolinium injection. T2-weighted sagittal (C) and axial (D) MRI of the orbit shows well-defined multiloculated cystic lesion with high-intensity signal and hypointense ring as fibrotic capsule around the lesion.

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