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Hydatid Cyst Presented With Orbital Cellulitis Manifestation: A Case Report and Brief Literature Review

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Abstract

Background: This study aimed to report on the case of a child with the manifestation of orbital cellulitis which was finally diagnosed as an orbital hydatid cyst.

Case report: The case is a 5-year-old girl with an isolated orbital hydatid cyst presented with slowly progressive proptosis, deterioration of vision, chemosis, periorbital swelling, and palpebral edema. Evaluations for other organ involvements including liver and lung were negative. A cystic orbital mass lesion with enhancing walls was found in orbital Computed Tomography (CT) scanning. The cyst was removed completely through the upper lid crease incision and treatment continued with systemic albendazole. The patient was followed for three months with a favorable outcome.

Conclusion: We should consider the diagnosis of a hydatid cyst in a child with the signs and symptoms of orbital cellulitis, specifically in endemic areas.

Key Words: Cystic echinococcosis, Orbital cellulitis, Pediatrics.

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1- INTRODUCTION

Hydatid cyst also called cystic echinococcosis, is a parasitic infectious disease caused by a tapeworm named Echinococcus granulosus. It is more common in Africa, Asia, the Middle East, Central and South America, and Southern Europe. The prevalence is low in North America (1). Hydatid Cyst is endemic in the Middle East as well as India, Turkey, and Africa (2).

Hydatid cyst is one of the most frequent zoonoses in Iran (3). The prevalence rate of human hydatidosis in Iran has been reported to be 0.61-2 per 100000 population (4). Definitive hosts include dogs and other Canidae in which adult worms reside in the small intestine (1, 3). The disease is transmitted to the intermediate hosts like sheep, goats, camels, humans, etc. via contamination of food or water with definite host feces containing eggs released by gravid proglottides (1, 5). The most common sites of involvement in humans are the liver and lungs (5). Hydatidosis in unusual body sites such as the heart, brain, muscle, salivary gland, bone, urinary tract, and pancreas have been reported in Iran (5).

Orbital Hydatid cyst is a rare presentation of cystic ecchinococcois (6). The signs and symptoms may include pain, periorbital swelling, chemosis, ocular motility limitations, decreased vision, and progressive proptosis (7, 8, 9). The primary treatment is surgery but systemic antiparasitic therapy should be considered if there is a cyst rupture or for prophylaxis (9). Here, we introduce the case of a 5-year-old girl with signs and symptoms of acute orbital inflammation, finally diagnosed with an orbital hydatid cyst.

2- CASE PRESENTATION

A 5-year-old girl was referred for progressive painful proptosis and deteriorated vision two weeks ago. On external examination, there was diffuse periorbital swelling with warmness and tenderness, chemosis, ocular motility limitation in all directions, and proptosis of the left eye (**Fig. 1**).

Fig. 1: A 5-year-old girl with diffuse periorbital swelling, chemosis, inferolateral displacement, and proptosis of the left eye

Anterior segment examinations including cornea, anterior chamber, lens, and iris were not remarkable in both eyes. Intraocular pressure was normal. In the fundus examination, there was grade 1 optic nerve head swelling of the left eye. RAPD was weakly positive on the left side. Differential diagnoses included orbital cellulitis, tumors, vascular disorders, and nonspecific orbital inflammation. The patient was admitted to the emergency department and orbital imaging (axial and coronal orbital CT scan) was requested. In the orbital CT

scan, an anteroposteriorly situated elliptical well-defined superonasal cystic mass with thick irregular margins and significant globe displacement was detected (**Fig. 2**).

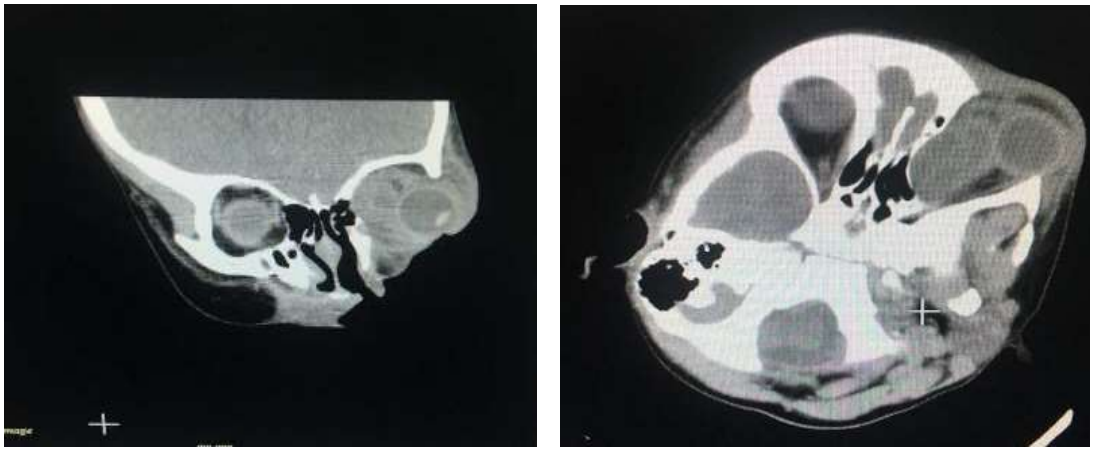


Fig. 2: In axial and coronal orbital CT scan, there is an anteroposteriorly situated elliptical well-defined superonasal cystic mass with dense margins and significant globe displacement.

There were also some small densities in the mass lesion remembering the daughter cyst in hydatid cysts. The child's parents signed the consent form, after being informed about all possible intra- and postoperative complications. The patient, then, underwent surgery with the possible diagnosis of an isolated orbital hydatid cyst. To explore the lesion, supraorbital space was approached through an upper lid crease incision, and after dissection of the surrounding tissue, the lesion was removed completely. The surgical field was irrigated with the saline solution due to a small rupture within the cyst and some leakage. The specimen was sent for histological evaluation. The hydatid cyst was confirmed in the histopathology report (**Fig. 3**).

In postoperative workups, no cystic lesion was found in chest radiography and abdominal ultrasonography. To reduce the spread and recurrence of the disease, the treatment continued with systemic albendazole of 15 mg/Kg per day for three months. There was no recurrence and the patient recovered with a favorable outcome.

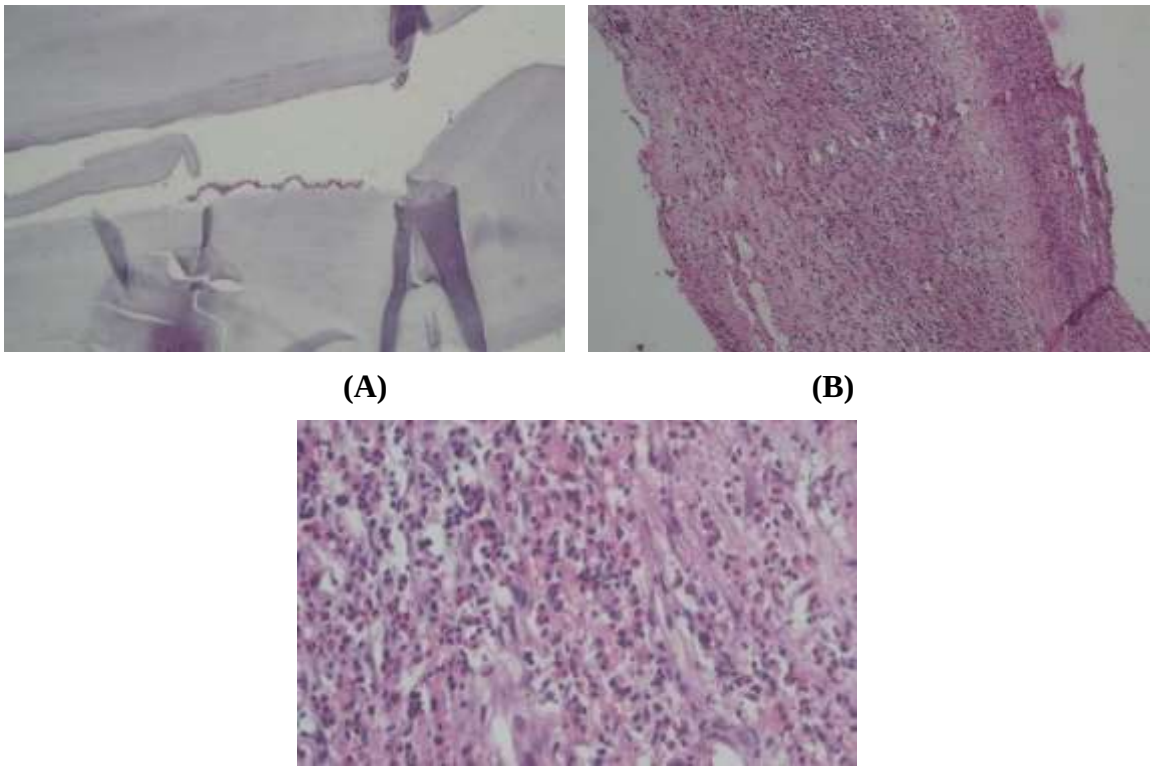
3- ETHICAL CONSIDERATIONS

Informed consent for the publication of the patient's clinical images and other anonymized data was obtained from the legal guardian of the patient.

4- DISCUSSION

Orbital involvement is extremely rare and accounts for less than 1% of all cystic involvements. The presentation is usually as orbital masses in a patient with slow-growing proptosis in endemic areas (10). Orbital involvement is typically unilateral, intra and/or extra conal, and presented with a variety of signs and symptoms such as slowly progressive proptosis, deterioration of vision, chemosis, periorbital swelling, and palpebral edema (3, 4, 6). It can cause compressive optic neuropathy (4). The orbital presentation as orbital cellulitis is very rare. As demonstrated in **Table 1**, it seems that the most frequent presentation of orbital hydatidosis is slowly progressive painless proptosis with a wide range of visual impairment. Our case was a 5-year-old girl, presented with manifestations of orbital cellulitis including diffuse

periorbital swelling with warmness and tenderness, chemosis, ocular motility limitation in all directions, and proptosis which is extremely rare.



(A) **(B)**

(C)

Fig. 2: A) Germinal layer (center) and laminar layer (upper and lower areas) of hydatid cyst (H&E, *100), B) Hydatid pericyst consists of inflamed fibrous tissue. (H&E, *100), C) Eosinophil-rich inflammatory infiltrates in pericyst (H&E, *400)

Table-1: A brief literature review table in the field of the primary orbital hydatid cyst.

Authors	Type of the study	Year	Gender/ Age at the onset of symptoms/ The main clinical manifestations
Assimakopoulos SF et al.(6)	Case report	2020	F/ 31 YO/ unilateral painless proptosis and visual decrement
Abouassi M et al. (13)	Case report	2020	F/ 21 YO/ unilateral painless visual impairment and proptosis
Chitra K et al. (14)	Case report	2019	F/ 3 YO/ unilateral painless slow-growing proptosis and vision loss
Rajabi MT et al. (15)	Case report	2018	F/ 23 YO/ unilateral progressive painless proptosis
Fasina O et al. (16)	Case report	2017	F/ 33 YO/ unilateral painless slow-growing proptosis and vision loss
Lentzsch AM et al. (17)	Case report	2016	F/ 5 YO/ unilateral painless proptosis and downward displacement of the left eye
Berradi S et al. (7)	Case report	2015	M/ 46 YO/ unilateral painless proptosis for 3 months

Sendul SY et al. (18)	Case report	2015	F/ 24 YO/ unilateral progressive painless proptosis, papilledema, macular edema, and tortuosity of retinal blood vessels
Rajabi MT et al. (19)	Case series	2014	Eight cases, 4 males and 4 females, with primary orbital hydatid cyst were introduced. The age range was from 13 to 62 years. Two cases were under 18 years old. The main symptom was painless slow-growing proptosis.
Mathad VU et al. (20)	Case report	2013	F/ 80 YO/ unilateral painless slow-growing proptosis
Al-Muala HD et al. (8)	Case report	2012	F/ 42 YO/ unilateral painful lid swelling, conjunctival injection, and slow-growing proptosis
Somay H et al. (21)	Case report	2012	M/ 10 YO/ unilateral painless progressive proptosis
Limaiem F et al. (22)	Case report	2010	M/ 74 YO/ unilateral painless progressive proptosis and visual loss
Benazzou S et al. (23)	A review of 10 cases	2010	Ten cases, 6 males and 4 females, with primary orbital hydatid cyst were introduced. Seven cases were less than 12 years of age. The main symptoms were unilateral painless slowly progressive proptosis.
Ghosh A et al. (24)	Case report	2008	F/ 35 YO/ unilateral painless progressive proptosis and vision loss
Ciurea AV et al. (12)	A report of 2 cases	2006	Two children presented with painless non pulsatile proptosis
Turgut AT et al. (25)	Meta-analysis	2004	A total of 25 patients were included. The most frequent clinical manifestations were slowly progressive unilateral proptosis (80%), visual loss (48%), and periorbital pain (24%).
Kıratlı H et al. (26)	Case report	2003	F/ 20 YO/ periocular pain induced with ocular movements
Xiao A et al. (27)	Research article	1999	Eighteen cases of primary orbital hydatid cyst, including 7 males and 11 females were assessed. The age range was 3 to 55 years old, and 15 cases were under 16 years of age. The main presentation was painless proptosis.
Sami A et al. (28)	A review of 10 cases	1995	Ten cases, 5 males and 5 females, of primary orbital hydatid cyst were assessed. The mean age was 25 years. The main presenting symptom was progressive unilateral painless exophthalmia.
Morales AG et al. (10)	A review of 35 cases	1988	Thirty-five cases with primary orbital hydatid cyst were reviewed. The age range was 2-57 years with an average of 16 years old. The main presentation was unilateral slow-growing proptosis.

F: Female, M: Male, YO: Years old.

Orbital imaging modalities such as CT scanning and magnetic resonance imaging (MRI) are useful methods for diagnosis and surgical planning. On CT scanning image, a low-density homogenous well-defined cystic lesion, which shows isointense signaling on T1- and T2-weighted images on MRI, is highly suggestive for orbital hydatidosis. In both CT and MR imaging, peripheral rim

contrast is seen after the injection of a contrast medium (28, 29, 30).

Surgical excision of the cyst with special attention to avoid cyst rupture is the treatment of choice (13). Because of the risk of disease dissemination following cyst rupture, a three-month course of medical treatment with albendazole or mebendazole has been recommended (13, 31, 32). In a clinical trial, researchers showed the superiority of albendazole at 15mg/Kg per day for three months compared to mebendazole (33).

5- CONCLUSION

Orbital hydatid cyst in children has a variety of non-specific symptoms and can even manifest as orbital cellulitis. Radiological assessments including orbital CT-scan and MRI are helpful in making the proper diagnosis. We should consider this entity in the differential diagnosis of any case with slow-growing proptosis, with or without signs and symptoms of acute orbital inflammation, in endemic areas.

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