

Hydatidosis of the orbit in Turkey: results from review of the literature 1963–2001*

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Received 31 December 2001; accepted 11 November 2004

Key words: hydatid cyst, echinococcus granulosus, orbita, review, Turkey

Abstract

Hydatidosis can involve almost every organ or tissue in the body via the systemic circulation, but hydatid infestation of the orbit comprises far less than 1% of the total incidence. This study investigated the data on all patients of orbital hydatid disease reported in Turkey since 1963. In this meta-analysis, a total of 25 patients were included and slowly progressive unilateral proptosis, with or without pain, was the most frequent clinical manifestation (80%). The other presenting symptoms were visual loss (48%), periorbital pain (24%), chemosis (16%), and headache (12%). The presumptive diagnosis of hydatid cyst was made on the images obtained from ultrasonography (US), computed tomography (CT) and/or magnetic resonance imaging (MRI) examinations. These diagnostic studies demonstrated a well-circumscribed cystic mass in almost all patients. Total surgical removal without rupture remains the best mode of therapy for orbital hydatid disease. The study indicates that in Turkey hydatidosis remains a serious problem, causing blindness. Orbital hydatid cyst should be included in the differential diagnosis of unilateral proptosis and visual handicap in patients from countries where hydatidosis is endemic.

Introduction

Hydatidosis is an infestation caused by tapeworms of the genus *Echinococcus*, most commonly *E. granulosus*. It has a characteristic geographic distribution, occurring in sheep-raising regions in the Middle East, Mediterranean countries, Eastern Europe, Australia, New Zealand, East and North Africa, India, and Latin America [1–4]. At the present, it is still a serious public health problem in Turkey. The definitive hosts are dogs and other carnivora, domestic or wild [1, 5, 6]. Hydatid cysts (metacestode stage) develop in sheeps and cows, while the larval stage develops in humans [1, 4, 7]. Although it can involve almost every organ or tissue in the body via the systemic circulation,

hydatid infestation of the orbit comprises far less than 2% of the total incidence [1, 3, 4, 8, 9]. In this retrospective study, a review of data from the literature since 1963 was conducted to present all patients reported from Turkey with emphasis on the clinical features, radiological characteristics, and method of management of hydatid disease affecting the orbit.

Patients and methods

We have done a review of the literature going back to 1963 and found more than 70 patients of orbital hydatid disease from Turkey. Information on all patients were collected from published studies and publications regarding hydatidosis of other neighbouring structures (brain, infratemporal fossa, etc.) were excluded. If a case appeared in

*This material was used in part for a poster presentation at the XXth International Congress of Hydatidology, 4–8 June 2001 in Kuşadası, Turkey.

more than one paper on this subject, the earlier report was omitted. Overall, 20 reports giving sufficient information on 25 patients were selected from a total of 31 reports in the Turkish literature since 1963 [2, 4–7, 10–35]. In this meta-analysis, data regarding location in the orbital cavity, clinical presentation, diagnostic study, method of management, and outcome were extensively evaluated.

Results

All patients had undergone surgical treatment and histological confirmation of orbital hydatid cyst was available in all patients except 1 patient reported by Akhan et al. [10] in the Turkish literature. Based on the presumptive diagnosis of hydatid cyst, they described a patient aspirated percutaneously under ultrasonographic (US) guidance without histopathological examination. In this series, hydatid disease was caused by *E. Granulosus* in all patients. Data regarding these patients according to year of publication is shown in Table 1. Seventy-two percent of the patients were 20 yrs of age or younger at the time of initial examination. The average age was 20 yrs in the series (range, 4–60 yrs). There was no sex predilection. In this meta-analysis, slowly progressive unilateral proptosis, with or without pain, was the most frequent clinical manifestation (80%). All patients presented with this clinical finding varying from 3 weeks to 2 yrs in duration. There was complete or partial visual loss in 12 patients (48%) at presentation. Out of these patients, one had sudden complete loss of vision due to the spontaneous rupture of an unoperated hydatid cyst. Other presenting symptoms were periorbital pain (24%), chemosis (16%), and headache (12%). On physical examination, a palpable mass was found in 5 patients (20%) and initial neuro-ophthalmological examination showed complete visual loss in all but 1 patient in this series.

The presumptive diagnosis of hydatid cyst was made on the diagnostic imaging techniques such as computed tomography (CT), US and magnetic resonance imaging (MRI). They demonstrated a well-circumscribed intraorbital mass in all but 3 patients (extraocular in 23 patients and intraocular in 2 patients). Out of 23 patients with extraocular lesion, 7 (28%) were located within the muscle

cone. Most cysts were located in the posterior part of the orbit, 10 in the medial quadrants, five in the superior quadrants, five in the inferior quadrants, and three in the lateral quadrants. In three patients, there was extension of the cystic lesion to the neighbouring structures such as maxillary sinus, intracranial cavity or superior palpebra. In one out of these patients with orbital hydatid cyst, ocular and orbital hemodynamics had been evaluated by color Doppler imaging and the blood flow in the ophthalmic artery had been decreased owing to compression by the mass. Furthermore, the blood flow in the posterior ciliary artery had been increased at the proximal side owing to constriction, but it had been found to be normal at the distal side.

Seven patients in this series had other organ involvement: liver in 5 patients, lung in 2, brain in 2, spleen in 1, and thyroid gland in 1. As seen in Table 1, results of serologic tests (Casoni's intradermal test and specific precipitin complement-fixation tests) were positive in only 7 patients (28%).

The kinds of surgical procedure performed are stated in 23 of 25 patients, as seen in Table 1. In the majority of patients, surgical excision or extirpation of the cystic lesion was performed and different surgical approaches for orbital hydatid cysts were used, with transcranial approach the most frequent. Out of these patients, 9 patients were treated with frontoorbitotemporal craniotomy, five with lateral orbitotomy (Kronlein–Berke approach), four with anterior direct approach, two with simple enucleation, and one with inferior orbitotomy. Percutaneous surgical treatment of the cyst was performed in 2 patients. Surgical intervention was complicated by the rupture of the cyst in 8 patients and it unfortunately resulted in blindness in 1 patient.

On surgical exploration rupture or puncture was reported in a total of 16 patients (64%). Out of these patients, 8 patients had surgical rupture because of its adherence to the surrounding tissues. To prevent anaphylaxis or dissemination, it was traditionally followed by aspiration and irrigation with different solutions including 15% hypertonic saline (nine patients), 30% hydrogen peroxide (2 patients), and 37% formalin (1 patient). As seen in Table 1, 12 patients received chemotherapy after the operation (albendazole in 7 patients and mebendazole in five). Follow-up

Table 1. Twenty-five patients of orbital hydatid disease in Turkey described in the literature since 1963

| Author(s) [Ref. #] | Year | Age/sex | Side of the cyst | Location in the orbital cavity | Presenting symptom or signs | Diagnostic study | Other organ involvement | Serological finding | Form of surgical intervention | Antihel treatment | Outcome |
|---------------------------------|------|----------|---------------------|--------------------------------------|---|---------------------|-------------------------------|------------------------|-------------------------------------|----------------------|--|
| Abadan and Hasanreisoglu [6] | 1973 | 32 yrs/M | L | Intraocular | Orbital pain, visual loss | None | ua | ua | simple enucleation | (-) | ua |
| Dinçer et al. [14] | 1973 | 12 yrs/F | R | Infero- postero- medial | Proptosis, diplopia, chemosis | X-ray | ua | (-) | lateral orbitotomy | (-) | ua |
| Slem et al. [18] | 1976 | 10 yrs/F | L | Supero- lateral ^a | Prosis, Palpebral swelling, chemosis | None | (+) | (+) | subiliary incision | (-) | ua |
| İlcayto et al. [16] | 1979 | 18 yrs/M | L | Supero- medial | Painful proptosis, headache | None | ua | ua | subiliary incision | (-) | no recurrence ^e |
| | | 7 yrs/F | L | ua | Proptosis, pain, visual loss | None | ua | ua | puncture | (-) | ua |
| Kars et al. [2] | 1982 | 7 yrs/M | L | Postero- medial | Proptosis | CT | (-) | ua | transcranial | ua | No recurrence at 2 yrs ^f |
| | | 11 yrs/F | R | Postero-medial | Proptosis | CT | (-) | ua | transcranial | ua | no recurrence at 6 mos |
| Engin et al. [15] | 1988 | 8 yrs/M | L | Posterior, intraconal | Diplopia, proptosis | US, CT | (-) | (-) | lateral orbitotomy | MEB | ua |
| İnal and Andi [17] | 1989 | 20 yrs/F | R | Postero- superior ^b | Proptosis, visual loss, headache | X-ray, US | ua | (+) | subiliary incision | (-) | no recurrence |
| Alparslan et al. [7] | 1990 | 11 yrs/M | L | Supero- medial | Proptosis, visual loss | CT | (-) | (+) | transcranial | (-) | no recurrence at 6 mos |
| | | 16 yrs/F | R | Lateral | Proptosis | CT | (-) | (-) | ua | ua | no recurrence at 4 mos |
| Sinav et al. [13] | 1991 | 13 yrs/F | R | Intraocular | visual loss, pain | CT, US | (-) | (-) | enucleation | (-) | no recurrence at 18 mos |
| Turgut et al. [4] | 1992 | 5 yrs/M | L | Medial, intaconal | Proptosis | CT | (+) | ua | transcranial | MEB | no recurrence at 3 yrs ^g |
| Özek et al. [33] | 1993 | 52 yrs/F | R | Superior, intraconal | Painful proptosis, visual loss | CT, MRI | (-) | (+) | Kronlein- Berke | MEB | No recurrence at 7 mos |

Table 1. Continued

| Author(s) [Ref. #] | Year | Age/sex | Side of the cyst | Location in the orbital cavity | Presenting symptom or signs | Diagnostic study | Other organ involvement | Serological finding | Form of surgical intervention | Antihel treatment | Outcome |
|--------------------------------|------|----------|---------------------|--------------------------------------|---|---------------------|-------------------------------|------------------------|-------------------------------------|----------------------|----------------------------|
| Diren et al. [19] | 1993 | 10 yrs/F | R | Postero- medial | Proptosis, headache | CT | (+) | (-) | Transcranial | ua | ua |
| Gelişken et al. [30] | 1994 | 20yrs/F | ua | ua | Proptosis | CT | ua | (+) | ua | MEB | ua |
| Hanioglu et al. [5] | 1997 | 4 yrs/M | L | Postero- medial ^c | Proptosis, chemosis | CT, US | (-) | (-) | Lateral orbitotomy | MEB | No recurrence at 6 mos |
| Karakaş et al. [31] | 1997 | 54 yrs/M | R | Infero- posterior intraconal | Proptosis, visual loss | CT, US | (+) | (+) | Transcranial | ALB | ua |
| | | 6 yrs/M | L | Supero- medial, intraconal | Proptosis, visual loss | CT, US | (+) | (+) | Transcranial | ALB | ua |
| Gökçek et al. [12] | 1997 | 60 yrs/F | L | Postero- inferior, intraconal | Visual loss, orbital swelling | CT, MRI | (-) | (-) | Inferior orbitotomy | ALB | ua |
| Ergün et al. [11] ^d | 1997 | 5 yrs/F | L | Postero- medial | Proptosis | CT | (+) | ua | Transcranial | ALB | No recurrence at 5 yrs |
| | | 13 yrs/M | L | Postero- medial | Proptosis, visual loss | CT | (-) | ua | Transcranial | ALB | No recurrence at 6 mos |
| Akhan et al. [10] | 1998 | 21 yrs/M | R | Infero- posterior, intraconal | Proptosis, diplopia | US, MRI | (-) | ua | PAIR | ua | No recurrence at 21 mos |
| Özkaya et al. [21] | 2001 | 43 yrs/M | R | Lateral | Proptosis, diplopia, visual loss, chemosis | CT, MRI | ua | ua | Lateral orbitotomy | ALB | No recurrence at 5 yrs |
| Öner et al. [20] | 2001 | 33 yrs/F | R | Infero- posterior | Painful proptosis, visual loss | US, MRI | (+) | ua | Subiliary incision | ALB | No recurrence |

M, male; F, female; dys, days; mos, months; yrs, years; L, left; R, right; ua, unavailable; CT, computed tomography; US, ultrasonography; MRI, magnetic resonance imaging.

MEB, mebendazole; ALB, albendazole; Antihel, antihelminthic; PAIR, puncture-aspiration-injection-reaspiration.

^a Secondary infected cyst patient with involvement of superior palpebra.

^b A giant orbital cyst with intracranial extension.

^c An intraorbital cyst with extension to the maxillary sinus.

^d Because two of four patients reported by the authors had already been described by Karakaş et al. [31] before, we considered 2 patients to avoid an overlap.

^e This patient underwent a second operation at 2 mos after first one because of recurrence.

^f Re-exploration was performed because of recurrence at 2 years after first surgical intervention.

^g Medical treatment was given because of recurrence at 7 months after surgery.

information was available in 15 of the 25 patients because some patients had not come to control examination or etc. There was a total of three recurrences; 2 patients were asymptomatic after the second surgical intervention and another patient had interestingly regressed into a calcified residue with antihelminthic therapy.

Discussion

The causative organism of orbital hydatid cyst is *E. granulosus*, which is rarely seen in organs other than the liver (60%) and lungs (20%) [1, 11]. On the other hand, orbital hydatid cysts account for 20% of orbital tumors in Iraq, 6% in Lebanon, and 5% in Argentina [1, 3, 8]. Indeed, limited clinical data regarding orbital hydatidosis exist in the literature. Undoubtedly, it is therefore difficult to diagnose that the cause of the unilateral proptosis is a hydatid cyst of the orbit in the absence of other organ involvement in the body. This study indicates that hydatid cysts should be included in the differential diagnosis of the unilateral proptosis in patients from countries where hydatidosis is endemic.

In Turkey, patients with orbital hydatid cyst are treated surgically by neurosurgeons or ophthalmic surgeons. As far as we know, the study is the first meta-analysis study which reviewed the patients with orbital hydatidosis in Turkey. Meanwhile, the present series is one of the large series of orbital hydatidosis in the world literature (Table 2). In our series, all but 1 patient had complete visual loss at the time of initial neuro-ophthalmological examination and a total of 9 patients (36%) had surgical (in 8 patients) or spontaneous (in 1 patient) rupture of the cyst. The incidence of complete visual loss in orbital hydatid disease varies from series to series: 20 and 44% in

our comprehensive review [1, 3]. The high incidence of visual loss and rupture of the cyst in the Turkish population is probably due to the advanced stage of the disease in most patients.

Clinically, the majority of patients with intra-orbital involvement present with unilateral proptosis and in some patients, there is mechanical restriction of ocular movements and visual impairment [36]. According to their anatomical locations, they may erode the orbital walls towards different directions as observed in some patients in our series. Only 7 patients (28%) in this series had other organ involvement. In a review of the literature, we found that intraocular hydatid cyst is extremely rare and is localized in the vitreous cavity, retina or anterior chamber [1, 22].

The orbital hydatid cysts situate in the superolateral and supero-medial angles of the orbit, either within or outside the muscle cone; inferiorly located cysts are rare [3, 7, 8]. In the present series, 16% of the patients were treated before the introduction of modern imaging techniques to clinical practice. On orbital ultrasonography (US), hydatid cyst appears as unilocular and well-circumscribed anechoic lesion [5]. Today, CT and MRI have become the radiological modalities of choice in the diagnosis of orbital hydatid cyst. On CT, it is described as a hypodense, unilocular, well-defined, thin-walled, homogenous mass with a hyperdense rim [2, 4, 5, 7, 11, 19, 30, 31, 36]. The densitometric values of the cysts range from +23 to +68 HU and the retrobulbar cystic lesions have a higher densitometric values due to the bone effect of the orbit [9]. Following contrast material injection, the cystic lesions show peripheral rim enhancement in their fibrous capsule (Figure 1) [2, 4, 5, 8, 12, 19, 21, 35]. Considering the radiological characteristics, a diagnosis of hydatid cyst of the orbit may be strongly suggested on CT scans and its

Table 2. Clinical series on orbital hydatid cyst in the world literature

| Author(s) [Ref. #] | Year | No. of patients | Percent of patients with | | |
|--------------------------|------|-----------------|--------------------------|-------------|---------|
| | | | Complete visual loss | Enucleation | Rupture |
| Talib [3] | 1972 | 21 | ua | 0 | ua |
| Gomez Morales et al. [1] | 1987 | 35 | 20 | 9 | ua |
| Present series | 2002 | 25 | 44 | 8 | 36 |

ua, unavailable.



Figure 1. Postcontrast axial computed tomography scan showing an enhanced well-defined cystic unilocular lesion located posteromedially along the medial wall in the left orbit [4].

relationship to the surrounding structures may be defined for the proper management of orbital hydatidosis. At present, orbital MRI has proved to be a very useful noninvasive diagnostic tool to rule out other cystic lesions with less water content in spite of its limited use due to high cost in endemic areas [5]. It shows a cystic lesion with a low-intensity signal on T1-weighted images and a high-intensity signal on T2-weighted images [5, 12, 20, 21]. On MRI, the capsule is seen as a hypointense rim surrounding the mass on T2-weighted images [10, 12].

Color Doppler study of the orbit is a new imaging modality for making a correct diagnosis. Doppler spectral analysis is helpful in studying ocular and orbital hemodynamics [5]. It may give more detailed information about the changes in blood flow in the central retinal artery and vein, posterior ciliary artery and ophthalmic artery owing to compression by the cystic mass [5]. Unfortunately, there are not enough cases in the literature yet to describe the specific features of orbital hydatid cysts on Doppler imaging. There is no doubt that a pre-operative correct diagnosis is very important since misdiagnosis may lead to serious consequences.

The differential diagnosis includes any well-circumscribed, nonpulsatile, and nonreducible lesion such as abscess, (epi)dermoid cyst, haematocele, teratoma, encephalocele and mucocele [5, 7, 9, 31]. Careful radiological evaluation coupled with the clinical findings usually allows

differentiation among these lesions. However, in some patients, a correct diagnosis can only be confirmed by the histopathological examination of the materials taken during surgical intervention. Histopathological confirmation had been obtained in all patients except 1 patient recorded in the Turkish literature. In such patients, the cyst wall typically consists of an inner layer of syncytial cells and an outer acellular layer (cuticular membrane) [1, 5, 7, 12–14]. There are numerous microtriches towards the lumen of the invagination and superficial epithelium with vacuolated cytoplasm on electron microscopic examination [1].

The treatment of choice is surgical, with removal of the cyst. Unfortunately, delay in establishing diagnosis and management causes complete visual loss. Whenever an intraorbital lesion is suspected, ophthalmologists generally advocate orbital exploration, while neurosurgeons recommend a primary transcranial procedure. As a result of this preference, frontal craniotomy approach was usually used by neurosurgeons in most of the patients in this review because of underdevelopment of the orbital surgery in the ophthalmology clinics in Turkey until the 1980s. Technically, intraorbital hydatid cysts can be extirpated easily by various orbital surgical methods unless there is any extension of the lesion to the cranial cavity. From the literature and from our own observations surgical removal without rupture is the only cure for hydatid cyst, but this is not possible in some patients because of the anatomical complexity of orbital cavity, the restricted surgical area, and a time consuming procedure [3, 11]. For this reason, surgical decompression and irrigation with local disinfectant solutions have been used as an alternative approach to conventional surgical extirpation in the literature [3, 16]. Recently, Akhan et al. [10] described the percutaneous treatment of an orbital hydatid cyst which is more satisfactory to both the patient and the physician. There is no question that preoperative diagnosis is very important for planning a medico-surgical treatment protocol in avoiding complications, because the rupture and dissemination of the cyst may result in catastrophic events. In these patients, postoperative chemotherapy with mebendazole or albendazole has been shown to be effective in recurrent cases of orbital hydatid cysts for regression or arrest of their growths [4]. We believe that the coming decade will possibly solve

this problem in certain rural areas of the world including Turkey.

Conclusions

This study is a meta-analysis of the Turkish literature on hydatid disease of the orbit over the last four decades. It provides an overview on clinical symptoms, diagnostic studies, and surgical treatments in 25 patients with intraorbital hydatidosis. In this series, there was a predilection for this lesion to affect children and young adults. Therefore, we think that hydatidosis should be a cause of etiological factor for visual handicap in children and young adults. It was concluded that:

1. Turkey is one of countries where hydatidosis is still encountered as a serious cause of complete loss of vision;
2. Hydatid disease of the orbit should be suspected when patients present with unilateral progressive proptosis with/without pain, especially in the nations where echinococcosis is endemic;
3. Current imaging modalities such as CT and MRI have added detailed information in the diagnosis of intraorbital hydatidosis;
4. Although intraocular hydatid cysts are reported occasionally, the cysts are usually located in retrobulbar region, the superior and medial quadrants are more frequently involved.
5. Accidental rupture of the cyst may lead to serious consequences such as an anaphylactic reaction or a spread of infection to neighbouring tissues.
6. Most of the patients with intraorbital hydatid cyst are treated with transcranial approach, while an orbital approach may be used in some patients.

Acknowledgements

We wish to thank our colleagues reporting the patients from Turkey in the literature for giving the necessary data for this study. We also thank the anonymous reviewers of International Ophthalmology for their constrictive comments and help with the manuscript.

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