ORIGINAL ARTICLE

# Orbital Hydatid Cyst: An Interventional Case Series

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## ABSTRACT

**Purpose:** To find out the demographics, presentation, and outcome of surgical treatment in patients of orbital hydatid cyst.

**Study Design:** Interventional case series.

**Place and Duration of Study:** Khyber Teaching Hospital Peshawar, Pakistan from 2009 to 2019.

**Methods:** This study included 11 patients with orbital hydatid cyst who presented in Khyber Teaching Hospital, Peshawar. Detailed history, ocular examination and Orbital imaging (Ophthalmic B-Scan, CT scan and/or MRI) was performed. The patients underwent Orbitotomy, cyst extirpated and sent for histopathology. Albendazole was given to the patients for 12 weeks after surgery. The preoperative and postoperative data until last follow-up was analyzed.

**Results:** Male to Female ratio was 5:6 and the mean age of the patients was 18.17 ± 17.4 years. Mean amount of proptosis was 26.27 ± 2.05mm and visual acuity was 0.23 ± 0.33 decimal in the affected eye at presentation. Eight patients (72.8%) had Relative Afferent Pupillary Defect with swollen discs. After imaging studies, presumptive diagnosis of hydatid cyst was made. Histopathology confirmed the diagnosis of hydatid cyst in all cases. Mean proptosis at the last follow up improved to 19.04 ± 1.45mm (P value = 0.00) and visual acuity to 0.47 ± 0.22 decimals (P value = 0.048). Only one patient (9.1%) had an associated hydatid cyst in the lung. There was no recurrence until last follow-up.

**Conclusion:** Hydatid cyst should be considered in differential diagnosis of proptosis in patients under 20. Surgical excision followed by a course of oral Albendazole is effective for the treatment of orbital hydatid cyst.

**Key Words:** Orbital hydatid cyst, Proptosis, Orbitotomy.

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## INTRODUCTION

Orbital hydatid cyst is a rare disease accounting for about 1% of the total cases of hydatid disease.[1] It is caused by a tapeworm Echinoccocus granulosus and human is the accidental intermediate host. Orbital hydatid cyst may occur alone or as a part of systemic Disease where cysts may be found in liver, lungs or other parts of the body.[2] It is usually located in intraconal compartment of the orbit mostly occupying the superomedial or superolateral part of the orbit. Rarely, it occupies the floor of the orbit, pushing the eyeball forward and superiorly.[2]

The most common presentation of orbital hydatid cyst is progressive unilateral painless proptosis. Mechanical effect of large cyst in a limited space of bony orbit may cause compression of the optic nerve and limitation in ocular movements. Imaging studies especially the Magnetic Resonant Imaging (MRI) can help in diagnosis but definitive diagnosis is done by histopathology. The Ultimate treatment of orbital hydatid cyst is surgical excision, followed by oral

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Albendazole (10mg/kg) for 12 weeks. The surgical approach depends upon size and location of the cyst.

In this article, we present a series of 11 patients of orbital hydatid cyst, who presented in our department between 2009 and 2019.

## METHODS

It is a retrospective case series where record of 11 cases of orbital hydatid cyst was evaluated and analyzed. These patients were admitted in "Orbit and Oculoplastics" department of Khyber Teaching Hospital Peshawar, Pakistan between 2009 to 2019. After detailed history and examination, each patient underwent imaging study (B-Scan, CT Scan and/or MRI Orbit), chest X-Ray, abdominal ultrasound and complete blood examination. Pulmonologist and general surgeon were also consulted. Depending upon presumptive diagnosis of orbital hydatid cyst on imaging studies, all patients underwent orbitotomy and cyst extirpated. All cysts ruptured during removal and field irrigated with hypertonic saline. The cyst walls were sent for histopathological examination and results came positive for hydatid cyst in all cases. Postoperative follow-up of the patients was between 03 months and one year. Nine out of eleven patients came from Afghanistan which is a war-affected area and patients could not come back for long term regular follow-up.

Various variables including age, gender, preoperative and postoperative amount of proptosis, visual acuity, extraocular movement, pupillary reaction and surgical techniques were recorded and analyzed using SPSS – Version 25. Mean with standard deviations were derived for numerical variables like age, amount of proptosis and visual acuity (in decimals) and frequencies were calculated for categorical variables like gender and pupillary reaction. Paired samples t-test was applied to calculate p-value to compare preoperative and postoperative proptosis and visual acuity.

## RESULTS

In the period from 2009 to 2019, eleven cases of orbital hydatid cysts were admitted and operated in our department. Male to female ratio was 5:6. The mean age of the patients was 18 ± 17.4 years and 9 out of 11 patients had age 20 years or below. All patients presented with moderately progressive unilateral painless proptosis. Mean amount of proptosis was 26.27 ± 2.05mm in the affected eye, while mean difference of proptosis between the affected and non-affected eye was 7.25 ± 2.05mm. Mean visual acuity in the affected eye was 0.23 ± 0.33 decimal at presentation. Eight patients (72.8%) had relative afferent pupillary defect (RAPD) with swollen optic discs and one (9.1%) had afferent pupillary defect (APD) with no perception of light. Fundus was not visible in this case due to hazy media. Rest 3 (27.3%) eyes had normal pupillary reaction. Imaging study of all patients was performed including MRI in 9 (81.8%) cases. Preliminary diagnoses of hydatid cyst was made on peculiar features on MRI and other imaging studies in all cases. In 9 (81.8%) cases cyst was intraconal and in 2 (18.2%) it was extraconal. All patients underwent surgical orbitotomy to excise the cyst, the approach depending upon cyst location. At one week postoperatively mean proptosis improved to 19.82 ± 2.14mm from 26.27 ± 2.05mm (P value = .000) and mean visual acuity improved to 0.28 ± 0.17 decimals (P value = 0.513). Mean proptosis at last follow-up was 19.04 ± 1.45mm (P value = 0.000) while difference of proptosis between affected and unaffected eye improved to 0.91 ± 0.14mm from 7.25 ± 2.05mm (P value = 0.000). All patients took Albendazole 10 mg/Kg for 12 weeks. The mean visual acuity at last follow up in the affected eye improved to 0.47 ± 0.22 decimals (P value = .048). All of our patients had isolated orbital involvement except one (9.1%) case in which a hydatid cyst was found concomitantly in the right lung. The excised cysts were sent for histopathology and all were confirmed to be hydatid cysts. One cyst from the old man (case 11) was calcified. The individual details of the cases are shown in table 1.

**Table 1:** *Demographics, Clinical presentation and post-operative findings.*

| S/N | Age | Gender | Laterality | Pre-operative Visual Acuity | Pre-operative Proptosis | Pupil | Cyst Location | Visual Acuity at Last Follow-up | Proptosis At Last Follow-up | Pupil At Last Follow-up |
|---|---|---|---|---|---|---|---|---|---|---|
| 1 | 15 | F | L | 0.1 | 30 | RAPD | Intraconal | 0.67 | 22 | N |
| 2 | 15 | M | R | 0.1 | 27 | RAPD | Intraconal | 0.69 | 20 | N |

| | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| 3 | 3 | F | L | Un-cooperative | 30 | RAPD | Intraconal | Un-cooperative | 18 | N |
| 4 | 17 | F | L | 0.08 | 28 | RAPD | Intraconal | 0.5 | 18 | N |
| 5 | 28 | F | R | 0.05 | 26 | RAPD | Intraconal | 0.5 | 19 | N |
| 6 | 19 | M | L | 0.05 | 29 | RAPD | Intraconal | 0.17 | 19 | RAPD |
| 7 | 20 | F | L | 0.67 | 26 | N | Extraconal | 0.67 | 19 | N |
| 8 | 6 | M | R | 1.00 | 22 | N | Intraconal | 0.5 | 18 | N |
| 9 | 6 | M | R | 0.1 | 25 | RAPD | Intraconal | 0.5 | 19 | N |
| 10 | 5 | M | L | 0.1 | 21 | RAPD | Intraconal | 0.5 | 17 | N |
| 11 | 65 | M | L | 0 | 25 | APD | Extraconal | 0 | 21 | APD |

APD = Afferent Pupillary Defect, F = Female, L = Left, M = Male, N = Normal, R = Right, RAPD = Relative Afferent Pupillary Defect

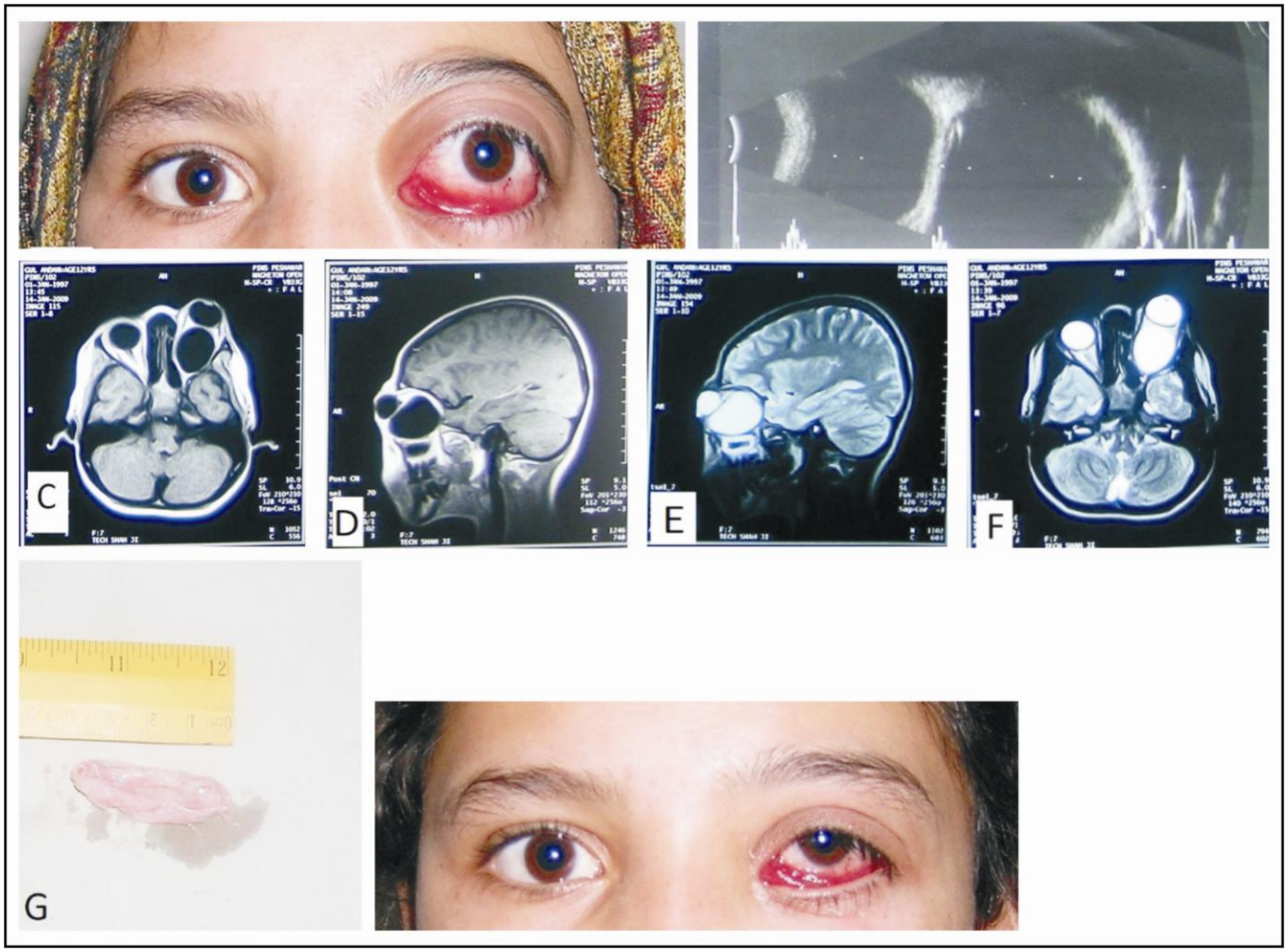


**Figure: 1A.** Left proptosis at presentation. **B.** B-Scan left eye. **C.** T1 weighted axial MR Scan. **D.** T1 weighted sagital MR Scan. **E.** T2 weighted sagital MR Scan. **F.** T2 weighted axial MR Scan. **G.** Excised cyst wall. **H.** First postop day.

## DISCUSSION

Orbital hydatid cyst is caused by a tapeworm called Echinococcus granulosus. This disease is endemic in certain areas such as in Australia, New Zealand, Middle East, South America and Mediterrian region.[3] Dog is a definitive host and adult tapeworm lives in dog's intestine. Eggs are shed infeces, which are taken-up by cattle (Sheep, cows, etc.). Cattle is the intermediate host and hydatid cysts are formed in various organs of the cattle. Dogs get infection by ingesting the cyst containing organs of intermediate host. Human is an aberrant intermediate host and gets infection by ingesting raw vegetables contaminated with E. granulosus eggs. Oncospheres hatch from the

eggs in the human intestine. These penetrate the intestinal wall and enter the circulation to reside in variety of organs including liver, lung, brain, bone and rarely in the orbit.[4] Orbital involvement comprises less than 1% of the total incidence.[1] In endemic areas orbital hydatid cyst comprised 5 – 18% of orbital space occupying lesions.[5] In a study on cystic lesions in the orbit from an endemic area, hydatid cyst comprised 25.8% of the cystic orbital lesions, second to dermoid cysts which was 29.7%.[6] In literature, it is mentioned that it affects mainly the left orbit, which can be explained by path of left carotid artery.[7] In our case series, the ratio of left and right orbit involvement is 7:4 i.e. two third involving the left orbit. In our study, the youngest patient was of 3 years age and median age was 15 years (Figure: 1). It is consistent with the age found in international literature.[8] No gender predilection is reported so far which is consistent with our case series.

Unilateral progressive proptosis is the most frequent presenting feature and proptosis is usually quite significant (Figure: 2A). Due to progressive enlargement of the cyst in a limited space of bony orbit, there is increasing intra orbital pressure and compression of the optic nerve which leads to swollen optic disc, decrease in vision and RAPD. In our study, 9 (81.8%) eyes presented with compressed optic nerve with RAPD and decreased visual acuity (mean visual acuity 0.23 ± 0.33 decimals) in affected eyes. In a study from Morocco on 10 patients, 90% of patients had visual loss at presentation.[9] In a meta-analysis from Turkey, 48% patients had visual impairment.[10] Imaging studies including B-scan, CT-scan and MRI of the orbit help in presumptive diagnosis. B-scan shows a characteristic "double wall" sign in orbital hydatid cyst.[11] (Figure:2B). CT-Scan shows a well-defined hypodense mass with perilesional rim of hyperdensity.[5] MRI demonstrate a homogeneous lesion, which is hypo-intense on T1 weighted images and hyper-intense on T2 weighted images.[5] (Figure:2C-F). In our study, presumptive diagnosis of hydatid cyst was accurate in 10 (90.9%) cases on imaging study. In one (9.1%) case, the cyst was calcified and diagnosis was made on histopathological examination. Calcification of the cyst wall when present indicate dead organism.[12] Total calcification of the cyst in liver can also occur.

Regarding location of the hydatid cyst in the orbit, it was intraconal in 9(81.9%) cases and extraconal in 2 (18.2%) cases. Even in the intraconal location, the main bulk of the cyst was occupying different locations in the orbit. In literature it is mentioned that most of the cysts are intraconal.[13] In a study from Iran on 8 patients, 2 cysts were extraconal, 3 intraconal, 1 in lacrimal gland, 1 in medial rectus and 1 intraosseous in lateral orbital wall.[14] Kiratli H. reported a case of intramuscular hydatid cyst in medial rectus muscle.[15] It shows that although more common site in the orbit is intraconal, almost any part of the orbit can be involved. There are also some case reports on subretinal hydatid cyst.[16]

Different approaches and excision methods are described in the literature. Neurosurgeons approach through craniotomy and orbitotomy and the orbital surgeons perform orbitotomy.[17]The classical surgical method for removal of hydatid cyst is described in "Mansons tropical diseases". The contents of the cysts are sucked out with a wide bore needle. Then 10% formalin is injected in the cyst, left for 5 minutes and then aspirated. Then the cyst wall is excised.[18] Into to excision of the cyst without rupture is another way of cyst removal. Aspiration of the cyst, followed by removal of the germinal layer of cyst by holding it with cryo probe is another procedure.[19] Some authors described a simpler technique by draining the cyst percutaneously under ultrasound guidance, refilled it with 15% hypertonic saline and re-aspirated after 10 minutes.[20]

Limitation of this study is the small sample size and lack of long term follow-up. Majority of the participants were from Afghanistan and they could not make up follow-up visits.

## CONCLUSION

Although rare, orbital hydatid cyst should be considered in differential diagnosis of unilateral proptosis, especially in children. Imaging studies including B-scan, CT-scan and MRI can give presumptive diagnosis of hydatid cyst in more than 90% of cases. Treatment of choice is surgical excision and postoperative results are excellent, provided surgical excision in done earlier followed by oral Albendazole for 12 weeks.

## Ethical Approval

The study was approved by the Institutional review board/ Ethical review board. **(667/DME/KMC).**

## Conflict of Interest

Authors declared no conflict of interest.

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## Authors' Designation and Contribution

Tajamul Khan; Professor: *Concepts, Literature search, Data acquisition, Data analysis, Statistical analysis, Manuscript editing, Manuscript review.*

Ibrar Hussain; Professor: *Concepts, Design, Literature search, Data acquisition, Data analysis, Statistical analysis, Manuscript preparation.*

Zaman Shah; Associate Professor: *Literature search, Manuscript editing, Manuscript review.*