

Ocular cysticercosis in pediatric patient

Cisticercose ocular em paciente pediátrico

Mateo Gasca-Sánchez¹ , Daniela Jaramillo¹ , Nathalia Pachón-Blanco² , Natalia Marín-Núñez² , Tatiana Urrea-Victoria³ ¹ Pontificia Universidad Javeriana, Hospital Universitario San Ignacio, Bogotá, Colombia.² Pontificia Universidad Javeriana, Bogotá, Colombia.³ Hospital Universitario San Ignacio, Bogotá, Colombia.

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Corresponding author:

Mateo Gasca Sanchez
Hospital Universitario San Ignacio
Kra 7 #40-62, 7th Floor, Bogotá, Colombia
mateogasca@javeriana.edu.co
+57 3113063675

Institution:

Pontificia Universidad Javeriana, Hospital
Universitario San Ignacio, Bogotá,
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ABSTRACT

Ocular cysticercosis is a parasitic infection caused by *Taenia solium*. Its early diagnosis and treatment decreases the possibility of visual morbidity. It can either compromise the anterior chamber or the posterior segment, which translates into a very variable and interspecific presentation that changes depending on the site of the infection. It is important to report this case due to its low presentation rate and the fact that a high suspicion index is required to make an assertive and timely diagnosis.

This is especially important in geographical areas that are endemic to this parasite due to the direct relationship between an early diagnosis and treatment and better visual outcomes. In this case report, we will discuss the multidisciplinary interventions of a pediatric patient in a high complexity hospital.

RESUMO

A cisticercose ocular é uma infecção parasitária causada pela *Taenia solium*. O diagnóstico e tratamento precoces diminuem a possibilidade de morbidade visual. Ela pode comprometer a câmara anterior ou o segmento posterior, o que se traduz em uma apresentação muito variável e interespecífica, que muda dependendo do local da infecção. É importante relatar esse caso devido à sua baixa taxa de apresentação e ao fato de que é necessário um alto índice de suspeita para fazer um diagnóstico assertivo e oportuno. Isso é especialmente importante em áreas geográficas endêmicas para esse parasita, devido à relação direta entre diagnóstico e tratamento precoces e melhores resultados visuais. Neste relato de caso, discutiremos as intervenções multidisciplinares de um paciente pediátrico em um hospital de alta complexidade.

INTRODUCTION

Cysticercosis is the most common infection caused by *Taenia*. It is specifically caused by *Cysticercus cellulosae*, the larva of the porcine *Taenia soleum*.⁽¹⁾ Although it can present worldwide, it is much more common in Latin America, India, and Sub-Saharan Africa.^(2,3) Ocular symptoms can be present in up to 46% of patients.⁽⁴⁾ Clinical presentation depends on the number, size, and stage of the life cycle of the larvae, as well as the inflammatory response caused by cyst degeneration.⁽²⁾

The diagnosis can be suspected through fundus examination in which the cystic lesions can be seen; however, ocular ultrasound and brain and orbit computed tomography (CT) scans should be done to discard compromise of the nervous system, as well as possible differential diagnosis.

Clinical presentation varies depending on the location of the cyst, the most common one being the orbital presentation (extraocular) in which patients present with alteration of ocular movements, diplopia, and strabismus.⁽⁴⁾ The most common intraocular presentation is subretinal and vitreous. In these cases, patients present with decreased visual acuity and myodesopsias; however, panuveitis, ocular hypertension, retinal detachment, leukocoria, optic disc edema, and intraretinal hemorrhage have also been reported.⁽⁵⁾

The longer time the patient has an active infection correlates directly with visual outcomes due to the fact that longer time translates into the growth of the cyst that starts to cause mass effect on nearby structures disrupting the normal anatomy of the eye, as well as with liberation of antigenic toxins that upregulate the local inflammatory response.⁽⁴⁾

Twenty-four to 72 hours after ingestion, eggs transform into embryos that penetrate the intestine wall entering the systemic circulation and travelling to different organ systems before turning into larvae.⁽²⁾ The cysts usually reach the eye through the choroidal vessels with posterior migration to the subretinal space and vitreous cavity. If the cyst acquires a subretinal localization, there can be hemorrhage, vasculitis, exudates, and vascular tortuosity of the overlying retina.⁽²⁾

Histologically, the larvae cause a granulomatous reaction associated with a severe inflammatory response that encapsulates by surrounding itself with a fibrous tunica. The parasites can usually be seen with amoeba-like movements inside the cyst.⁽²⁾

The differential diagnosis of ocular cysticercosis changes depending on the age of the patient and the

localization of the cyst in adults hydatid cysts. Thyroid eye disease and Inflammatory pseudotumor should be suspected (with the orbita presentation). In pediatric patients, retinoblastoma needs to be considered and discarded as the first diagnostic possibility due to its high mortality and visual morbidity. Infection by toxocara is another important differential diagnosis. Endophthalmitis should be considered in all age groups in patients with a history of trauma, surgery, or systemic infection.

In patients with an intraocular presentation, surgical removal of the cyst must be considered, as well as the systemic antiparasitic therapy and the use of corticosteroids to downregulate the inflammatory response.⁽⁴⁾ Cyst removal is done with a posterior vitrectomy via pars plana with retinal detachment, vitreous hemorrhage, postoperative uveitis being the most common complications. Surgical management is preferred over antiparasitic medication due to the fact that the death of the parasite causes antigen release and upregulation of the inflammatory response with a higher risk of visual morbidity.

The use of focal laser photocoagulation of the cyst has also been described; however, it requires expertise and precision of the surgeon considering the fact that damage of the healthy retina can cause scotomas and increase inflammatory response.⁽²⁾

We will discuss a case of a 3-year-old female patient, who came from a rural area, presented with decreased visual acuity and absence of retinal red reflex. Discarding an intraocular neoplasia, mainly retinoblastoma was imperative due to clinical presentation, age group. Due to the diagnostic challenge, the patient needed interdisciplinary management in a high complexity hospital in Bogotá, Colombia.

We obtained informed consent from the patient's family in order to use information from the minor medical record, without including personal data, identification or history number, adhering to the 1975 Helsinki declaration and the 2013 revision.

CASE REPORT

Female patient, 3 years and 9 months old, with no relevant history, born at 37 weeks by vaginal delivery, with adequate birth weight, who did not require management in the neonatal intensive care unit. The patient presented trauma due to a fall from her own height with a non-fixing wound in the eyelid and consulted with the optometry, where a decreased visual acuity associated with loss of retinal red was detected, and the patient was referred to an ophthalmology evaluation.

The Ophthalmology Service of her hometown hospital found evidence of a whitish, avascular, cotton wool-like mass in the posterior pole of the right eye, with the presence of retinal red in the periphery associated with ipsilateral exotropia. They performed an ocular ultrasound scan documenting an anteroposterior diameter preserved with a high-density image on the posterior pole. In addition, a cranial CT scan showed a 7x9mm hyper-dense nodular image in the right eyeball projection, in its posterior segment, in contact with the retina, with no CNS alterations. Given the findings and because of the age group, a diagnosis of retinoblastoma was a differential diagnostic to be considered. They referred the patient to a more complex center for comprehensive evaluation.

At the time of the initial evaluation, the patient was in good general condition, hemodynamically stable, without obvious compromise of other systems. On the ophthalmological examination, right exotropia was clear, with no rejection to light. Healthy anterior segments and transparent crystalline lens were shown, intraocular pressure within normal limits and right fundus with dense vitreitis that made it impossible to visualize details were registered. In view of these findings, the diagnosis of intraocular mass workup and right sensory exotropia was considered, so simple and contrasted magnetic resonance imaging (MRI) of the orbits and brain and a new ocular ultrasound were requested.

During hospitalization, the patient did not present deterioration of her general condition. MRI showed a nodular lesion on the right eye, apparently dependent on the retina, with an intermediate signal on T1 and high on STIR, with a hypointense periphery in the same sequence and with sequential artifact because of bleeding or calcification, without enhancement with the application of gadolinium, with dimensions of 7mmx5mmx10mm, with hypointensity in the inferior aspect of the vitreous chamber suggestive of hemorrhage, diffuse alteration in the intensity of the vitreous and nonspecific increase in the enhancement of the posterior segment, without alterations in the orbit, optic nerve chiasm, or in the contralateral eye. Considering the first diagnostic possibility retinoblastoma without signs of intraocular extension, the brain MRI was within normal limits. However, the ocular ultrasound showed a cystic lesion on the optic disc, with opacities in its interior simulating Scolex with associated vitreitis, considering a lesion compatible with ocular cysticercosis on the right eye (Figure 1)

In view of these findings, a medical meeting was held with retina and oncological Ophthalmology Services,

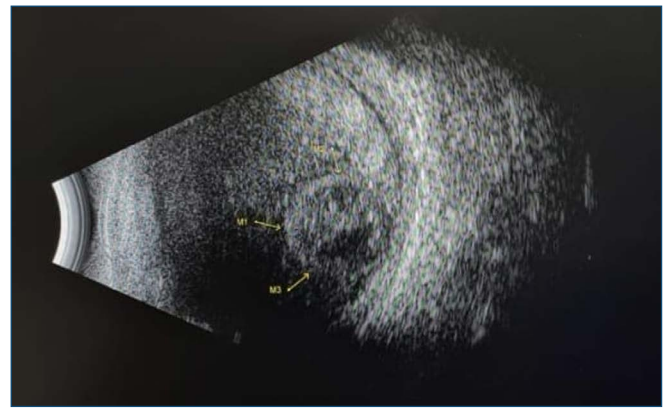


Figure 1. Ocular B-Scan ultrasonography showing a cystic lesion compatible with ocular cysticercosis.

considering the performance of diagnostic and therapeutic posterior vitrectomy with intraoperative cytologic evaluation. Depending on the result of the latter, additional conducts were defined. If negative for malignant cells, posterior vitrectomy would be completed and, if the result were positive, enucleation would be performed. Visual evoked potentials were requested with a report of a severe lesion of the right optic pathway at the prechiasmatic level.

During the procedure, marked inflammation of the posterior segment was evidenced, with presence of inflammatory membranes and fibrin, with a dome-shaped lesion, violaceous, located over the optic nerve, of approximately four disc diameters of solid consistency, associated with whitish subretinal perilesional content (Figure 2). Lensectomy was performed and a frozen cytological sample was taken and evaluated without evidence of malignant cells. Anterior and middle vitrectomy were performed exposing the cyst area, which was removed without evidence of parasitic forms inside. The vitrectomy was completed, and retinal tearing was evidenced, which was managed with endothermy. Retinotomy and subretinal material aspiration were performed, later laser blocking of the retinotomy was carried out, ending with silicone injection and inferior iridectomy.

The patient presented an adequate postoperative evolution with modulated pain, intraocular pressure (IOP) within normal limits, in aphakia, with no signs of endophthalmitis. It was established that the patient could continue ambulatory follow-up.

In the ambulatory control, the patient showed visual acuity of hand movement at 20cm with retina applied, traces of laser photocoagulation properly healed, without signs of infection or inflammation in the anterior chamber or posterior segment, with whitish peripapillary inferior fibrotic tissue that is directed towards inferior and temporal retina without traction (Figure 3). Pediatric ophthalmology

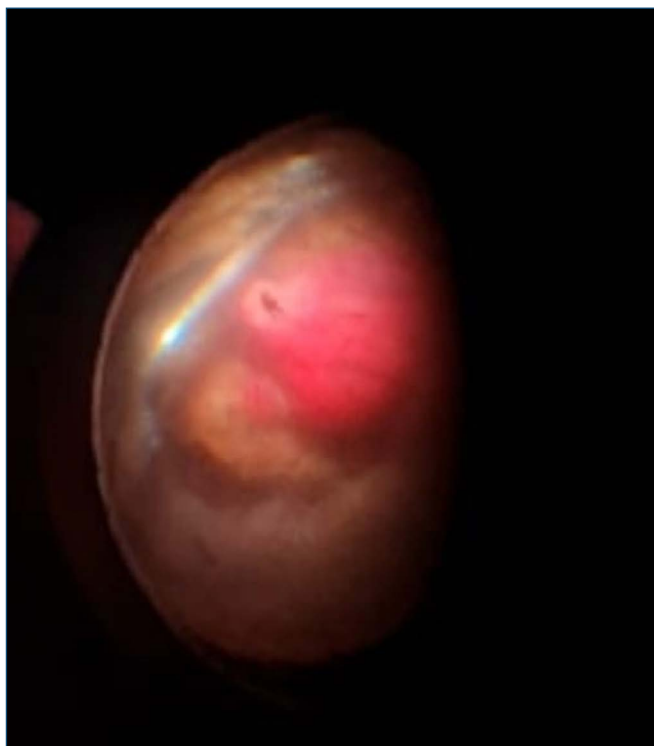


Figure 2. Intraoperative fundus image of a cystic lesion, located above the optic nerve, compatible with a cysticercosis cyst.



Figure 3. Postoperative fundus photo showing fibrotic tissue that is directed towards inferior and temporal retina without traction.

evaluated the patient, and it was considered that there was no requirement of surgical management of exotropia secondary to neurosensory deprivation.

DISCUSSION

Ocular cysticercosis is an important pathology due to the high visual morbidity, which can be prevented with

timely management. However, its low index of suspicion usually delays diagnosis. The presented case shows the context of a Latin American country, where cysticercosis is endemic, as well as a multidisciplinary approach of a pediatric patient in a high complexity center.

There are different factors associated with the case presented that show unique aspects of diagnostic and therapeutic behavior of a patient with ocular cysticercosis. First, the initial consultation of the minor was not associated with symptoms of long evolution. On the contrary, it was related to an acute condition in the context of a trauma, which exemplifies the barriers in the ophthalmologic controls. Adequate clinical management at the place of origin and a timely referral that allows multidisciplinary management are rescued.

Through the fourth level institution, joint management by pediatrics allows the suspicion of systemic alterations that cannot be assessed through the ophthalmologic consultation. Up to 24% of patients with ocular cysticercosis present neurocysticercosis even without systemic manifestations.⁽⁵⁾ Joint management with oncologic ophthalmology and pediatric oncology allows timely interventions in case the patient presents retinoblastoma, which was an important differential diagnosis regarding the age of our patient. In addition, the evaluation by an infectious disease service can clarify whether the patient requires adjuvant management with antiparasitic drugs, the evaluation by an ocular ultrasound specialist allows characterizing the lesion and ruling out differential diagnoses. Finally, the presence of a retinologist is essential for an adequate surgical approach that allows resection of the lesion and management of possible intraoperative complications.

We reiterate the importance of recognizing and suspecting of intraocular cysticercosis in the pediatric population, of periodic visual controls by a specialist physician to favor early detection, and education of patients and family members on warning signs since the increase in size and evolution of cysts are related to worse visual prognosis.

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