BMC Ophthalmology

CASE REPORT Open Access

Orbital migration of schistosome eggs: a case report

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Abstract

Background: Ocular damage, including damage to the conjunctiva, lacrimal gland, eyelids, and orbit, caused by *Schistosoma haematobium* is sporadic. We report a clinical case of orbital migration of schistosome eggs.

Case presentation: A 14-year-old boy of Malian nationality presented with a painless swelling of the upper right eyelid, which had been gradually increasing for approximately 3 months. Visual acuity was logMAR 0.10 and 0.00 in the right and left eye, respectively. External examination revealed a right palpebral mass, pushing the globe slightly downward and inward. Computed tomography revealed a mass of the right lacrimal gland. Total excision of the mass was performed by transpalpebral orbitotomy. Pathological examination revealed an inflammatory granulomatous infiltrate of the lacrimal gland consisting of lymphocytes, eosinophils, giant cells, epithelioid cell, histiocytes and calcified Schistosoma eggs with terminal spine.
Urine examination revealed eggs of *S. haematobium*. Praziquantel 40 mg/kg was administered to the patient. The hematuria stopped after 1 week. After 3 years of follow-up, no recurrence was noted.

Conclusions: The bilharzian granuloma of the lacrimal gland is an ectopic site of the parasite. In this case, the granuloma was cured by surgical excision followed by a course of Praziquantel.

Keywords: Orbit, Schistosoma, Neglected tropical disease, Lacrimal gland, Case report

Background

Neglected tropical diseases (NTDs) are a diverse group of bacterial, viral, parasitic, fungal, and non-communicable origin diseases [1]. These diseases affect mainly tropical areas, in the neglected and resource-poor communities. Currently, the control efforts for these diseases focus on approximately 20 NTDs, including schistosomiasis [1, 2].

Schistosoma mansoni (*S. mansoni*) and *Schistosoma haematobium* (*S. haematobium)* are the two most common types of schistosomes in Africa. The preferred site for *S. mansoni* and *S. haematobium* is the digestive tract and urogenital tract, respectively. Ectopic forms of schistosomiasis, most of which are secondary to *S. mansoni* and *Schistosoma japonicum,* are found in the digestive, urogenital, central nervous, cardiovascular, pulmonary, dermatological, and ophthalmological systems [3–6].

Ocular damage, including damage to the conjunctiva, lacrimal gland, eyelids, and orbit, caused by *S. haematobium* is sporadic [7–14]. Ocular manifestations are dominated by inflammation and granuloma [8, 10]. Granulomatous involvement of the lacrimal gland was first described in 1977. Herein, we report a clinical case of orbital migration of schistosome eggs in a 14-year-old boy.

Case presentation

A 14-year-old boy of Malian nationality accompanied by his parents presented at IOTA, Bamako, Mali, with a

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painless swelling of the upper right eyelid, which had been gradually increasing for approximately 3 months. Visual acuity was logMAR 0.10 and 0.00 in the right and left eye, respectively. External examination revealed a right orbito-palpebral mass, pushing the globe slightly downward and inward (Fig. 1a). Slit lamp examination of the anterior segment and fundus was normal in both the eyes; intraocular pressure was 13 mmHg on both eyes. Computed tomography revealed a mass of the right lacrimal gland with low tissue density and heterogeneity that was enhanced by iodine contrast. The right palpebral mass measured 29 × 13 mm (Fig. 1b).

Total excision of the mass was performed by transpalpebral orbitotomy. Pathological examination revealed an inflammatory granulomatous infiltrate of the lacrimal gland consisting of lymphocytes, eosinophils, giant cells, epithelioid cell, histiocytes and calcified schistosome eggs with terminal spine (Fig. 2).

We questioned the patient and his parents regarding a possible history of schistosomiasis. The patient revealed that he has been experiencing terminal hematuria for at least 3 months. Urine examination revealed eggs of *S. haematobium*. An abdominopelvic ultrasound found irregular thickening of the bladder wall with cystitis and homogeneous splenomegaly. No other ectopic locations of the eggs were found. Praziquantel 40 mg/kg was administered to the patient. The hematuria stopped after 1 week. The dose of the prescribed medication was renewed 1 month later. After 3 years of follow-up, no recurrence was noted (Fig. 3).

Discussion and conclusions

In Mali, the estimated prevalence of *S. haematobium* is 38.4% among school-aged children [15]. The areas of endemic bilharzian diseases are mainly along the Niger River. Our case was residing in this area (Macina) at the time of diagnosis.

Adult male and female worms of *S. haematobium* are found in the vascular system in humans [6]. They lay eggs that are then released into the vesical venous plexus before being expelled in the urine.

Ophthalmic damage caused by *S. haematobium* was first described in 1927 in the conjunctiva [13]. Other localizations, particularly in the eyelids, orbit, and optic nerve, have also been described [7, 10, 12, 13]. Granulomatous involvement of the lacrimal gland was first described in 1977 by Jakobiec [10]. The patient reported was a 11-year-old boy from Sierra Leone. The pathological description of our patient's surgical specimen was similar to that as described by Jakobiec.

In the case reported by Jakobiec, adult worms were found simultaneously along with the eggs [9, 16]. In our case, no worm was found.

Only 20 to 55% of the eggs are successfully excreted, while the rest are inevitably trapped in the host tissues. Schistosome eggs eventually pass through the mesenteric vessels [6, 16, 17].

Consequently, the infiltrating eggs are focal points for the host immune system, which sets up a distinct attack and sequestration strategy in response: the granuloma. The formation of granulomas around the eggs is a consequence of a delayed hypersensitivity reaction [18].

Granulomas are highly organized multicellular structures that are enriched with a range of immune cells, including T helper type 2 cells, macrophages, and eosinophils, with mast cell infiltration and accumulation of type 2 cytokines [16, 18].

The newly laid eggs are immediately covered with cells and proteins from the hemostatic system. These haemostatic constituents promote their anchoring to the endothelium and prevent them from being transported by the bloodstream [16].

The granuloma is both beneficial and harmful to the host. On one hand, granulomas protect host tissues from toxins released by the eggs. On the other hand, fibrous sequelae following granuloma resolution are the major cause of morbidity and mortality in schistosomiasis [16].

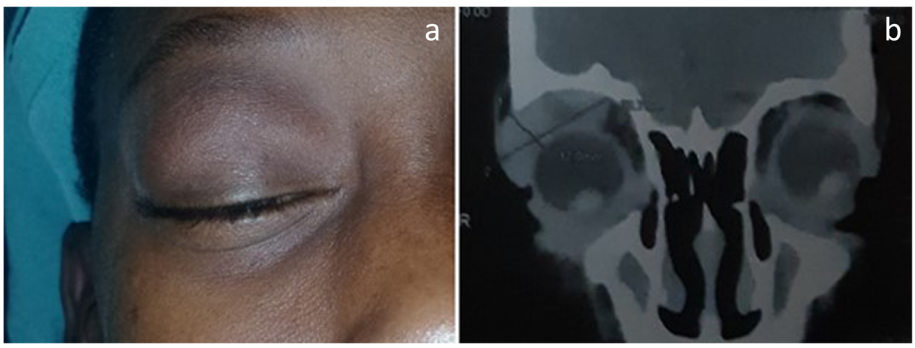

Fig. 1 **a** orbito-palpebral mass, pushing the eyeball slightly downward and inward, with grade 1 exophthalmos (patient under GA). **b** Computed tomography: Right lacrimal gland mass measuring 29 × 13 mm with low tissue density and heterogeneity that was enhanced by iodine contrast

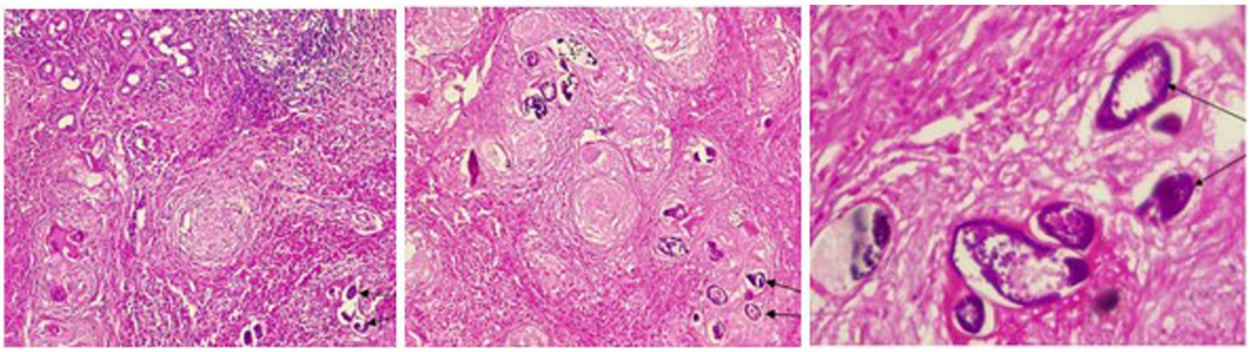

Fig. 2 Lacrimal gland granuloma with lymphocytes, eosinophil, giant cells, epithelioid cell, histiocytes and calcified schistosome eggs (arrows) with terminal spine (magnification 100–200-300 X)

The World Health Organization recommends the use of praziquantel for the treatment of *S. haematobium* [17]. In our case, two subsequent doses of praziquantel resulted in a complete cure.

Orbital migration of *S. haematobium* eggs is infrequent, especially to the lacrimal gland. The bilharzian granuloma of the lacrimal gland is an ectopic site of the parasite. In this case, the granuloma was cured by surgical excision followed by a course of Praziquantel.

Abbreviations

NTDs: Neglected tropical diseases

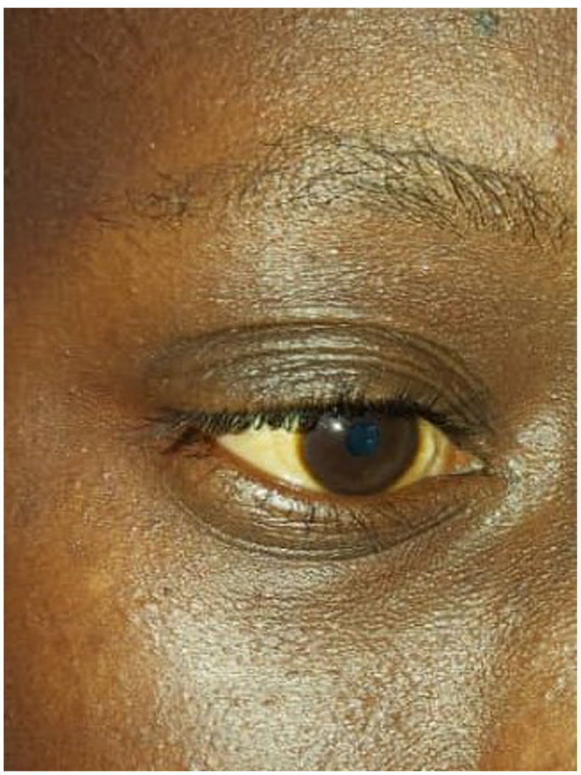

Fig. 3 No recurrence after 3 years follows up

Acknowledgements

Not applicable.

Authors' contributions

NG, AKH and MG drafted and wrote the manuscript, NG and SR and LT and RDR revised it critically for important intellectual content. SB, AN, ERR and FS analyzed and interpreted patient clinical details and CT findings. All authors read and approved the final submitted manuscript.

Funding

No grants or funds were received for this study.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Declarations

Ethics approval and consent to participate

The study was approved by the scientific review comity of CHU-IOTA. Written informed consents were obtained from the parents.

Consent for publication

Written informed consents were obtained from the parents.

Competing interests

The authors declare that they have no competing interests.

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Received: 20 January 2021 Accepted: 21 April 2021
Published online: 27 April 2021

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