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Orbital Loiasis masquerading as orbital cellulitis: A Case Series

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Abstract

Background: Orbital loiasis is a rare ocular disease which is sparsely reported in the literature. It is caused by the human filarial parasite, *Loa loa,* which is rarely found in other continents except in Africa and among African immigrants. The ocular presentation of orbital loiasis is similar to orbital cellulitis, thus, a high index of suspicion is required to make a diagnosis.
Methods: A retrospective description of the patients diagnosed and treated for orbital loiasis in a tertiary health facility in Ogun State, Nigeria between 1998 and 2013 was done. Data on the demographic characteristics, place of residence of the patients, symptoms, signs, and results of ancillary investigations were retrieved from the records.
Case findings: Three cases of presumed orbital loiasis were seen within the study period. All the patients presented with sudden onset of ocular pain with proptosis without the history of shifting body or facial swellings or visible worm in their eyes. Other features recorded in all the three patients included severe axial proptosis, eyelid oedema with mechanical ptosis, conjunctival injection with chemosis and restriction of ocular motility in all positions of gaze. Full blood count revealed eosinophilia while the radiological investigation was neither in keeping with sinusitis, thyroid-related orbital disease or orbital pseudotumor. Treatment was switched to Diethylcarbamazine when there was no satisfactory clinical response to the initial antibiotics and all the patients had a good outcome.
Conclusion: Orbital loiasis should be suspected when orbital cellulitis cases appear to be recalcitrant to treatment with antibiotics, particularly when there is eosinophilia on peripheral blood film.

Keywords: *Diethylcarbamazine, Eosinophilia, Loa loa, Orbital cellulitis, Proptosis, Vision threat*

Introduction

Loiasis is commonly found in Africa but orbital loiasis is rare and is sparsely reported in the literature. This infestation, is caused by the human filarial parasite known as *Loa loa*, is prevalent in Africa and among African immigrants or travellers on other continents. [1 – 3] *Loa loa* infections have been reported in all the geo-political zones in Nigeria. In Ogun State of Nigeria, the prevalence of *Loa loa* infestation was reported to be 2.5% and 15-61% in Egbado (Yewa) and Ijebu divisions respectively, but the exact prevalence in Remo area of Ogun State is presently unknown. [4]

Orbital loiasis as a disease has been aptly described in the literature by Sandford Smith. [5] It has also been described as worm underneath the conjunctiva associated with conjunctivitis and orbital oedema. [6] Neither of these documentations of orbital loiasis described any systemic feature in the disease.

Orbital cellulitis is a rather uncommon but serious bacterial infection of the contents behind the

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orbital septum which may be associated with loss of vision and life. [7] Orbital loiasis seems to be a close differential diagnosis of orbital cellulitis. The two conditions share similar ophthalmic characteristics in clinical presentations (symptoms and signs) whereas systemic features such as high-grade fever and other manifestations of acute systemic illness caused by pyogenic infection are typically absent in orbital loiasis.

Although orbital loiasis is not life threatening like orbital cellulitis, it poses a threat to vision and presents a diagnostic challenge to the attending ophthalmologist. The aim of this study was to describe the clinical features of orbital loiasis and highlight the challenges its diagnosis and treatment may be associated with in a low-resource setting.

Method

This was a retrospective description of cases of *Loa loa* infestation affecting the orbit which were managed at the Olabisi Onabanjo University Teaching Hospital, Sagamu, Ogun State, Nigeria between January 1998 and December 2013. The clinical details were retrieved from the patients' hospital records.

The data collected included the demographic characteristics and their places of residence. The presenting symptoms and duration of illness were noted. History of fever, malaise, presence of catarrh, preceding trauma to the affected eye and possible history of thyroid eye disease were retrieved from the records. The clinical findings as documented in the patients' clinical notes were recorded for analysis. Full blood count and blood culture were also requested. Blood film for microfilaria was done when marked eosinophilia was elicited from the peripheral blood film and the patients did not respond or deteriorate while on antibiotic treatment. Radiological investigations requested included plain sinus X-rays and orbital ultrasonography.

The patients were admitted with a working diagnosis of orbital cellulitis and commenced on empirical intravenous cefuroxime, metronidazole and gentamicin prior to the availability of laboratory results. After the sixth day of intravenous antibiotics, the diagnosis of Orbital loiasis was entertained in the presence of marked eosinophilia on peripheral blood film, negative blood culture and lack of clinical response to parenteral antibiotics. Two of the patients were commenced on oral Diethylcarbamazine (DEC) 50mg daily for three days while antibiotics were discontinued. The dose was increased to 150mg for 3 weeks after no adverse reaction to the drug was documented.

Case Findings

During this period, three cases of presumed orbital loiasis and 63 cases of orbital cellulitis were managed in the unit. Two of the presumed cases of orbital loiasis were males while one was a female. The age range was 35 to 67 years. The three patients lived in Remo area of Ogun State which is not a known *for Loa loa* endemicity. One of the patients was a farmer while the other two were artisans.

The right eye was affected in two patients. Watering and eye pain/ache were present in all the three patients, photophobia in one patient and foreign body sensation in one patient. Shifting facial or body swellings or visible worm in the eye, history of trauma or features suggestive of thyroid ophthalmopathy were absent in the three patients. None of the three patients had systemic symptoms. Examination of these patients revealed visual acuities of NPL, 6/36 and 6/24 in the affected eye. Peri-orbital fullness, eyelid oedema with mechanical ptosis, conjunctival injection with chemosis and corneal staining (which was described as exposure keratopathy) were present in the three patients. One patient had optic atrophy and imminent corneal perforation. There was no anterior neck swelling or signs of thyrotoxicosis and neither were there signs of sinonasal diseases. The initial diagnosis in all the three cases was orbital cellulitis.

The packed cell volume was normal in the three patients but with marked eosinophilia of 20-40%. Blood culture yielded no growth in the cases. Plain sinus X-rays did not show evidence of sinusitis and orbital ultrasound was neither supportive of thyroid eye nor orbital pseudotumour. Deterioration of symptoms and signs were noted in the three patients despite consistent intravenous antibiotics therapy. Presumed diagnosis of orbital

loiasis was retrospectively made in one patient who was discharged against medical advice before the diagnosis could be made because there was no significant improvement despite treatment.

The two patients who were treated with DEC improved and were discharged for follow-up care in the clinic. The proptosis eventually resolved completely with the resumption of ocular movement. Figures 1, 2 and 3 show one of the patients with progressive disease while on antibiotics and following the commencement of DEC.

Figure 1: One of the patients: two days after commencement of antibiotics

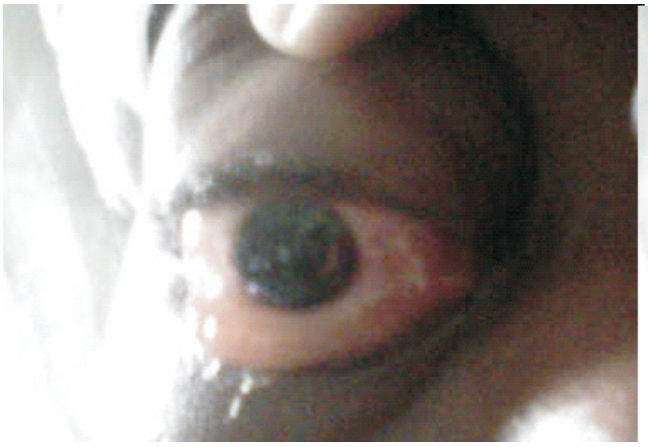

Figure 2: The same patient six days on antibiotics

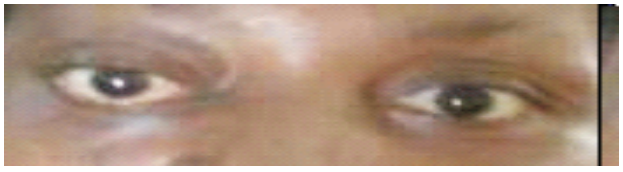

Figure 3: The same patient after one week on DEC

Discussion

Loa loa infection is a common problem in sub-Saharan Africa and it is one of the neglected tropical diseases. [8] The patients with orbital loiasis in our study lived in Remo which was not previously known to be endemic for loiasis [4] although, it is possible to find sporadic cases of ocular loiasis outside the areas of endemicity. [9, 10] None of the patients in this study admitted to the history of travel to areas of *Loa loa* endemicity. We are aware that other microfilariae such as *Wuchereria bancrofti* can cause diseases in the eye and one of the challenges of this retrospective study was that effort made to confirm *Loa loa* infections in these patients was not adequately documented and our suspicion of *Loa loa* was based purely on clinical grounds.

All the patients with presumed orbital loiasis presented acutely within 72 hours of the onset of proptosis and pain without past history of shifting localised body (subcutaneous) swellings, pruritus or passage of visible worm in the eye. Some authors reported that the disease may remain asymptomatic with the clinical signs appearing years after infection. [9, 11 - 14] This pattern of presentation was similar to that of orbital cellulitis - which is a life threatening condition but the toxic look, absence of eosinophilia and good response to systemic antibiotics found in orbital cellulitis are the major differences between the two diseases.

Loa loa occupying the anterior chamber of patients was earlier reported in Ibadan by Osuntokun *et al*. [15] Omolase *et al*, [16] in Owo, reported a case of subconjunctival worm in an adult female but the literature on orbital involvement are rare. The blood film examination for microfilaria was negative in all the patients; this pattern of negative smear in the patients who were certain to have *Loa loa* infection had been noted in other studies. [9,17]

In our study, the diagnosis of orbital loiasis was based on disease progression, the presence of marked eosinophilia on the peripheral blood film typical of parasitic infections, [18] non-response to potent intravenous antibiotics despite compliance. A high index of suspicion with the therapeutic trial of DEC was also helpful as suggested by the patients' good response. Therefore, full blood count is recommended for all patients with proptosis and a detailed attention should be paid to all the components of the result.

Some of the major challenges in this study included lack of access to computerised tomographic scan which is a gold standard radiological investigation for proptosis and lack of facilities for serological

studies which might have been of help in differentiating *Loa loa* from the other parasites. In a resource-limited facility, emphasis should be placed on the result of full blood count among other factors to make a diagnosis of orbital loiasis. The challenge of making the appropriate diagnosis of orbital loiasis can also occur in the best-equipped centres in African and non-African countries where facilities exist for all the needed investigations. Ophthalmic clinicians should, therefore, keep an open mind and always remember that this disease exists.

Conclusion

Due to human dynamics, orbital loiasis should be suspected in patients presenting with ophthalmologic features similar to orbital cellulitis, with marked eosinophilia on peripheral blood film and who are not responsive to antibiotic treatment, particularly with the history of recent travels to *Loa loa* endemic areas. The retrospective nature of the study and lack of facility to carry out serological tests to confirm the *Loa loa* microfilaria infestation are some of the limitations of this study.

Consent: One of the patients gave consent for the use of her images.

Authors' Contributions: OTO conceived and designed the study and drafted the manuscript. All the authors participated in the literature search, reviews and contribution of intellectual content to the manuscript.

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