Case report

A rare case of asymptomatic Leptospirosisneuroretinitis

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Abstract

Leptospirosis is an endemic disease worldwide and its incidence is increasing with outbreaks and mortality. It is caused by *Leptospire*s *interrogans* which are very thin, spiral-shaped, tightly coiled, gram-negative aerobic, spirochetes. It can cause several infection to human including organs such as the kidneys, liver, lungs, heart and the central nervous system. The organism can give rise to several ocular manifestations and when it affect the eyes, it causes Leptospirosisneuroretinitis. The present case illustrated a case of 9 year-old boy succumbed withleptospirosisinfection presenting as neuroretinitis. The unique of the case is that the boy had noobvious systemic manifestation of leptospirosis which caused a challenge to a physician to arrive at the diagnosis. Other important differentials hadbe entertained namely toxoplasmosis and ocular tuberculosis. Further management had been discussed until the boy was discharged and being followed up as an outpatient. This is followed by the discussion on leptospirosis neuroretinitis.

Keywords: Leptospirosis; Neuroretinitis; Macular star; uveitis

Bangladesh Journal of Medical Science Vol. 22 No. 01 January'23 Page : 225-228
DOI: https://doi.org/10.3329/bjms.v22i1.63082

Introduction

Leptospirosis is an endemic disease occurring in urban as well as in developing countries and rural area throughout the world. Its incidence is increasing with outbreaks and mortality[1]. The organism enter humans through an intact mucosa or abraded skin causing bacteraemia leading to infection of various organs such as the kidneys, liver, lungs, heart and the central nervous system. Eyes are not exceptional organ to be involved. Eye complications are frequent in leptospirosis and may develop in both the acute (bacteraemic) and the second (immunologic) phases of the disease[2]. The eye signs include externally as conjunctival congestion, chemosis, sub-conjunctival haemorrhage and fundoscopicallyas optic disc oedema, retinal vasculitis, retinal haemorrhage and hard exudates. Neuroretinitis is a rare complication of leptospirosis. Neuroretinitis is a focal inflammation of the optic nerve and peripapillary retina or macula. It can be either infectious or idiopathic in aetiology and is characterized by acute unilateral vision loss. This eye condition could also be triggered by other bacterial, viral and parasitic agents[3-4]. It is a potentially treatable problem hence the cause should be determined by detailed history and examination.

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Case report

A 9-years-old Malay boy, with no past medical illness presented at a primary care centre in Nilai, Negeri Sembilan, Malaysia complained of headache especially when reading small printing for one month. It was associated with a single episode of vomiting the night before presenting to the ophthlamologist. For the past one week, he claimed that he felt headache when reading small prints during the school hours. His parents thought that the problem might be due to increment of refractory error as the son had never revisited the optometrist for the refractory problem. The boy had been using glasses and was told to have short-sightedness with significant astigmatism since one year ago. He then developed sudden severe generalised headache to the point of vomiting. Worried about serious illness, he was brought by his parents to a primary care doctor for evaluation the next day. He admitted to experiencing bad flu a few weeks earlier. Otherwise there were no obvious fever, joint pains and diarrhoea to suggest infective causes. He had not come into contact with cats or other pets recently nor had any history of jungle trekking or swimming in the river in the past 3 months.

On background history, he has short sightedness and high astigmatism of both eyes which required him to wear high powered glasses for one year. At that point, he was referred to an ophthalmologist for which serious pathology causing high power and high astigmatism had been ruled out. Since then he wore glasses appropriate to his eye condition.

On examination, a relative afferent pupillary defect with best-corrected visual acuity of 20/120 in the left eye and 20/20 in the right eye was noted. A slit-lamp examination disclosed that the anterior segment was unremarkable in the right eye. Fundus examination of the left eye revealed an optic disc with indistinct margins, disc oedema and partial star-shaped exudates distributed in the macula. There was also dilated and tortuous vessels at peripapillary area. The retinal periphery, however, was normal in the right eye (Figure 1). The ability to perceive colour was impaired in the left eye. The right eye was unremarkable in all aspects.

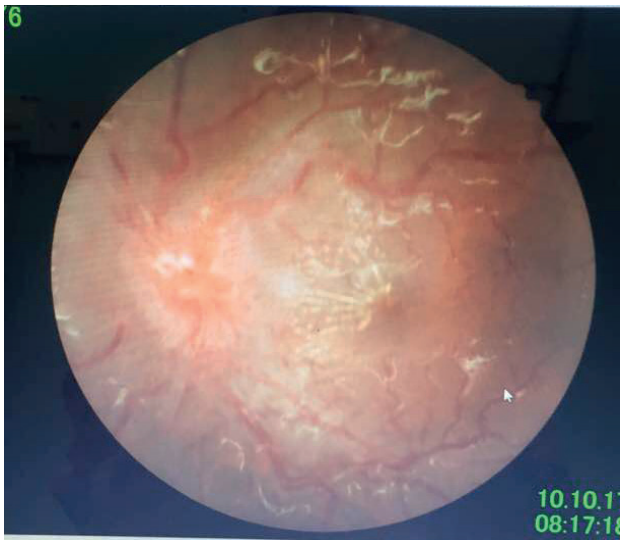

Figure 1: 1st funduscopic photo of left eye (before treatment)

He was then referred urgently to a hospital for CT brain in order to rule out any pathology causing increased intracranial pressure. At the hospital he was further evaluated thoroughly. CT scan was performed, and the finding was normal brain with mucosal plug at the maxillary sinus.

The clinical findings of optic disc oedema in the left eye, coupled with subretinal fluid and a macular star, were consistent with the diagnosis of neuroretinitis. Blood investigation revealed mild leucocytosis with predominantly neutrophilia and the erythrocyte sedimentation rate of 15 mm at the end of 1st hour. Blood biochemistry and urine examination were normal.

The patient then was treated as neuroretinitis and warded for empirical antibiotics. Septic workout was performed and was positive for leptospirosis *Ig M*. Other results came back negative for Mantoux test, ANA, VDRL, HIV, serologic tests for hepatitis B and C, toxoplasmosis. In view of these results, he was treated for leptospirosis. A complete regime to eradicate the organism was given. The regime was intravenous (IV) penicillin for 1 week, followed by oral amoxicillin 500mg three times a day for 6 weeks. Oral prednisolone 0.5mg/kg was started one week after the I/V antibiotic and tapered fast 10mg/kg followed by improvement of the sign and symptoms. A week after IV antibiotic treatment in the ward, he was discharged, and oral amoxicillin as described above was prescribed. He had multiple follow up sessions assessing his vision and fundus. The prednisolone was tapered accordingly and after about 6 weeks, prednisolone and antibiotics

werestopped. His left eye swollenoptic disc subsided substantially. The diagnosis of left eye neuroretinitis secondary to leptospirosis remained although the MAT (Microscopic Agglutination Test) showed inconclusive result which was available only after few weeks from the initiation of treatment.

Discussion

The classical feature of neuroretinitis is optic disc oedema with a macular star, whereby these changes start to disappear after 1 month, but in some cases it can take 6 - 12 months for total healing[5].In Leptospirosis, the ocular findings such as conjunctival hyperaemia with engorged vessels (clinical hallmark) was not demonstrated in the present case. Uveitis is an important late complication of leptospirosis which manifests usually around six months of systemic infection[2]. Other eye signs which can be found externally include conjunctival congestion, chemosis and sub-conjunctival haemorrhage. Whereas in the acute phase of leptospirosis infection funduscopic finding include optic disc oedema, retinal vasculitis, retinal haemorrhage and hard exudates[6].

In the present case, at the initial presentation, the boy was treated empirically with broad spectrum antibiotics since the serological result of leptospirosis was only available after few days later. A Microscopic Agglutination Test (MAT) was used to aid diagnosis. MAT is currently considered a gold standard test although it can give rise to false positive result. Like in this case where the result was inconclusive. Nonetheless for the benefit of doubt he was still treated as leptospirosis. Other differential diagnoses include toxoplasmosis and tuberculosis need to be considered as it could mimic similar presentation.

In this case, it was difficult to obtain the diagnosis of neuroretinitis caused by leptospirosis. This is due to no obvious contact with rats or swimming in recreational water areas, withabsenceof classical systemicsymptoms. Hence, ocular manifestation with neuroretinitis alone without systemic manifestation makes the diagnosis of leptospirosis more perplexing. Therefore, physicians especially ophtalmologists need to be aware that leptospirosis can manifest atypically and infective causes such as leptospirosis need to be considered.

Leptospirosis presenting as neuroretinitis, has recently been reported in the literature from Malaysia[7] as well as other countries[8,3,9]. In Albania, a retrospective analysis of ocular manifestation of leptospirosis had been undertaken emphasizing that leptospirosis frequently causes ocular pathology[8]. In the early course of the disease, treatment with appropriate anti-microbial agents for example, penicillin, amoxycillin, doxycycline or ceftriaxone, is indicated in systemic leptospirosis[10,11]. However, steroids are the mainstay of treatment for uveitis[2].

Conclusion

The present case report highlights a case of leptospirosis presenting as neuroretinitis alone, unaccompanied by systemic manifestation of leptospirosis. Without the positive serological test for leptospira, the case might have been treated differently which could lead to further eye complication such as cataract. Physicians especially ophthalmologists in leptospirosis endemic areas should consider the diagnosis of leptospirosis when encountering neuroretinitis. Other differentials such as toxoplasmosis and tuberculosis should also be excluded, especially in tropical regions.

Source of fund: Nil

Conflict of interest: None

Ethical clearance: Taken

Authors's contribution:

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Study design:MRR,FBSM,SNSM

Data gathering: MRR, FBSM,SNSM

Writing and submitting manuscript: MRR, FBSM, SNSM

Editing and approval of final draft: MRR FBSM, SNSM, NAAT, MAM

References:

1. Costa F, Hagan JE, Calcagno J, et al. Global Morbidity and Mortality of Leptospirosis: A Systematic Review. *PLoS Negl Trop Dis*. 015;9(9):e0003898. Published 2015 Sep 17. doi:10.1371/journal.pntd.0003898
2. Rathinam SR. Ocular manifestations of leptospirosis. *J Postgrad Med*. 2005;51(3):189-194..
3. Narayan SK, Kaliaperumal S, Srinivasan R. Neuroretinitis, a great mimicker. *Ann Indian Acad Neurol*. 2008;**11(**2):109-113. doi:10.4103/0972-2327.41879
4. Walse FB HW. Clinical neuroopthalmology. In: 3rd ed Baltimore Md: Williams and Wilkins Co. 1982. p. 234–5.
5. Gass JD. Diseases of the optic nerve that may simulate macular disease. *Trans Sect Ophthalmol Am Acad Ophthalmol Otolaryngol*. 1977;**83**(5):763-770.
6. Martins MG, Matos KT, da Silva MV, de Abreu MT. Ocular manifestations in the acute phase of leptospirosis. *Ocul Immunol Inflamm*. 1998**;6(**2):75-79. doi:10.1076/ocii.6.2.75.4045
7. Subramaniam S, Hafiz A, Norliza R. Leptospira , the ocular predator. *Malaysian Journal of Ophthalmology*. Vol **2** No 2 (2020):151–9. https://myjo.org/index.php/myjo/article/view/86
8. Ghosh S, Das R, Saha M, Das D. Neuroretinitis as an Unusual Manifestation of Leptospirosis: A Case Report. *J Clin Exp Ophthalmol.* 2011;**2**(1):1–2.
9. Basumatary LJ, Das S, Das M, Goswami M, Kayal AK. Leptospirosis presenting as neuroretinitis. *J Neurosci Rural Pract*. 2012;**3**(2):224-226. doi:10.4103/0976-3147.98264
10. Bharti AR, Nally JE, Ricaldi JN, et al. Leptospirosis: a zoonotic disease of global importance. *Lancet Infect Dis*. 2003;**3**(12):757-771. doi:10.1016/s1473-3099(03)00830-2
11. Panaphut T, Domrongkitchaiporn S, Vibhagool A, Thinkamrop B, Susaengrat W. Ceftriaxone compared with sodium penicillin g for treatment of severe leptospirosis. *Clin Infect Dis*. 2003;**36(**12):1507-1513. doi:10.1086/375226