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Case Report

Optic nerve tuberculoma in an immunocompetent patient: Case report [☆]

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ARTICLE INFO

Article history:

Received 21 December 2020

Revised 1 January 2021

Accepted 2 January 2021

Keywords:

Tuberculosis

Optic nerve

MRI

Immunocompetent

ABSTRACT

Optic nerve tuberculoma is a rare extra-pulmonary manifestation of mycobacterial infection. It is usually associated with HIV infected patients. Due to its clinical polymorphism, the diagnosis and management of the disease either in immunocompetent patients and immunocompromised ones remains challenging. Herein, we discuss a case of a 32-year-old patient with no history of immunodeficiency admitted for multifocal tuberculosis with a reduced visual acuity as an extra-pulmonary manifestation, and radiologically revealed the presence of tuberculoma in the optic nerve in the cerebral MRI.

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Introduction

Tuberculosis is a public health problem worldwide, especially in the developing countries. It remains the most common single cause of morbidity and mortality worldwide, causing nearly 3 million deaths each year [1–3].

The causative organism is mycobacterium tuberculosis. It is an airborne mycobacteria transmitted by droplets that remains suspended in the air. Therefore, pulmonary tuberculosis is the most common form. However, there are many extra-pulmonary tuberculosis manifestations, including the ocular involvement that remains exceptional.

Tuberculosis is latent in immunocompetent individuals. It only reactivates if the immunity is compromised. Regarding ocular manifestations, it is still unknown if it results from a direct mycobacterium infection or hypersensitivity reaction reflected on the management of tuberculous uveitis.

Case presentation

A 32-year-old patient with a family history of tuberculosis was admitted in the emergency room for a right iliac fossa pain associated with meteorism and diarrhea defining the

[☆] Competing interests: None.

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<https://doi.org/10.1016/j.radcr.2021.01.006>

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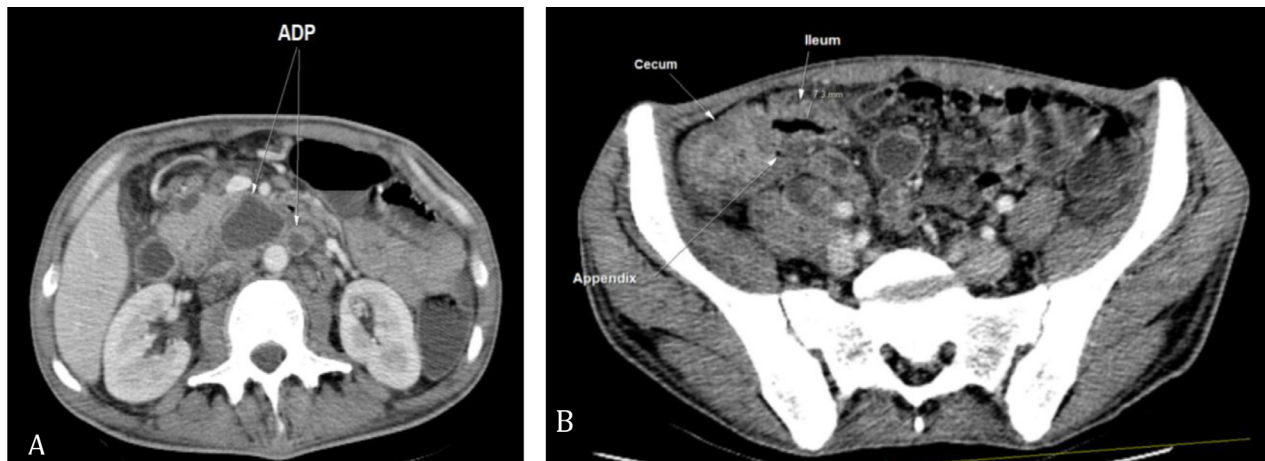


Fig. 1 – Abdominal CT with axial thin cuts showing the wall thickening of the ileocecal region (B) associated with intra and retro peritoneum adenopathies = ADP (A) (white arrows).



Fig. 2 – Chest CT with axial and coronal thin cuts showing multiples micronodules with hematogenous spread confirming the miliary tuberculosis.

König's syndrome also known as the ileocecal valve syndrome. An abdominal ultrasound was done and showed a parietal wall thickening of the ileum. Thereafter, an abdominal and chest CT scans were performed that confirmed the parietal wall thickening associated with intraperitoneum necrotic adenopathy (Fig. 1) and miliary tuberculosis (Fig. 2).

A colonoscopy revealed an ulcerative aspect of both ileal and cecal mucosa. A biopsy showed a granulomatosis ileitis. The patient was then put under antituberculosis drugs: Isoniazid, Rifampicin, Pyrazinamide, and Ethambutol. Two weeks later, the patient developed a complete visual loss in the left eye associated with a meningeal syndrome. The clinical examination found a tachycardia, polypnea, and meningeal stiffness. The ophthalmologic examination has shown a second degree papillary edema and no light perception in the left eye with no uveitis. The patient underwent a cerebral CT that revealed multiple tuberculous granulomas in the right parietal and frontal lobes. Afterward, a cerebral MRI confirmed the parenchymal tuberculomas (Fig. 3) and showed an additional optic nerve lesion (Fig. 4) that has the same characteristic of the cerebral lesions. Therefore, the diagnosis of optic nerve tuberculoma was retained. The decision to add oral corticotherapy in association with antituberculosis therapy for a period

of 9 months was then made. Unfortunately the evolution of our patient was unfavorable; he developed a hepatic cytolysis then died 4 months later.

Discussion

Mycobacterium tuberculosis is primarily an airborne infection that spreads from one contaminated person to another by expelling infected droplets. The development of the disease depends on the body's innate defenses [4–6]. The majority of people (90%) infected remain asymptomatic throughout their lifetime. Whereas in about 5% of patients, the disease appears within the first few years after the exposure [4–6]. The remaining 5% keeps the infection latent and may develop the symptoms whenever their immunity is compromised, defining the reactivation of the latent infection [6,7].

For a better understanding of the intraocular manifestations, we shall briefly mention the essential steps in the disease pathogenesis. Essentially there are 5 stages in the pathogenesis of tuberculous infection [8–11]. Throughout the stages, the bacilli undergo phagocytosis by the alveolar

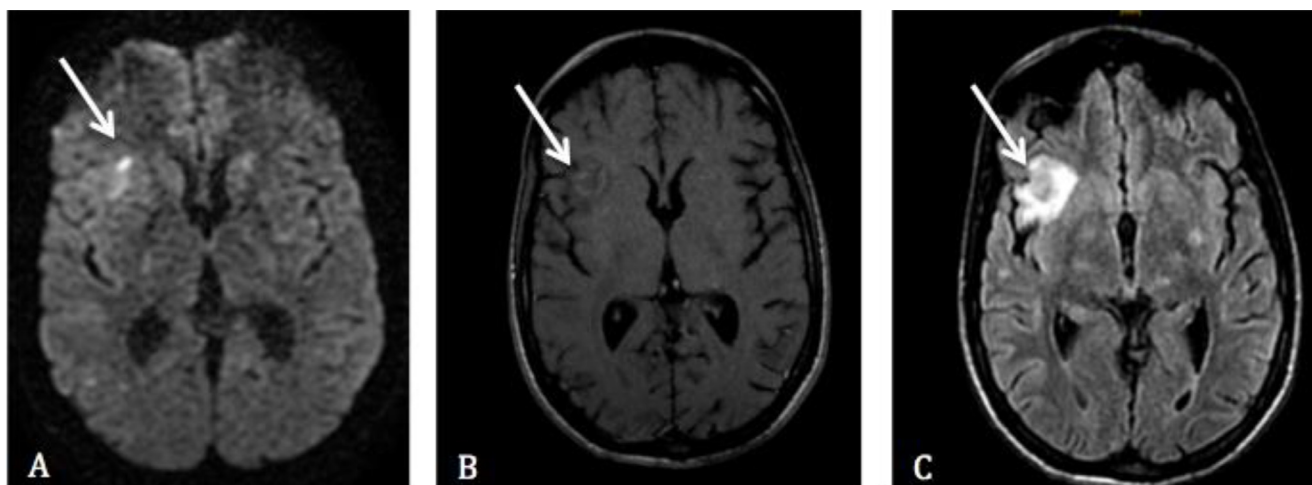


Fig. 3 – A (Diffusion), B (FLAIR C+), C (T2): brain MRI with axial cuts showing multiples tuberculomas (white arrows) in the right parietal lobe with a restriction in diffusion (A) hyper signal in T2 (C) and annular enhancement in FLAIR C+ (B).

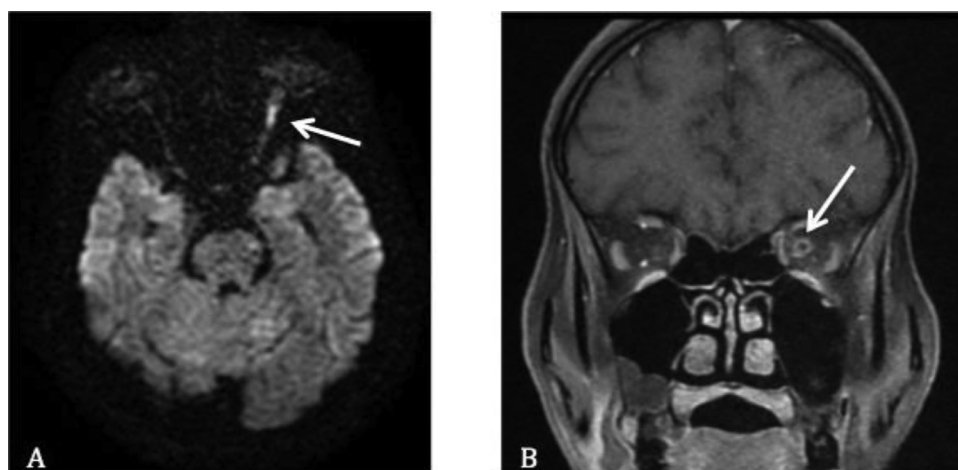


Fig. 4 – A (Diffusion), B (coronal FLAIR C+): brain MRI showing a tuberculoma of the left optic nerve with a restriction in diffusion (white arrow A) and annular enhancement in FLAIR C+ (white arrow B).

macrophages that are destroyed resulting in an initial nidus, and then a delayed type of hypersensitivity response develops destroying the bacteria laden macrophages resulting in tissue damage, which manifests as caseous necrosis. If, by this time, cell-mediated immune response (CMI) is quite poor, bacilli escaping from the edge of caseous necrosis start re-multiplying in nonactivated and partly activated macrophages, thus carrying bacteria to the different parts of the lung or to the various other organs, including the eye. If, on the other hand, good CMI develops, highly activated macrophages surround the caseous necrosis destroying those bacilli that are escaping, thus halting the progression of lesion at a subclinical stage.

The bacilli in the ocular tissue, as well as in the optic nerve, may remain dormant for years before they reactivated. In most adults who are not HIV positive, it appears that the ocular tuberculosis develops through hematogenous dissemination which is the case in our patient. The second mechanism involves a hypersensitivity reaction secondary to antigen re-

lease [12]. This immune reaction causes tissue and vascular damage [13].

The optic nerve location produces various pictures: Papillitis, tuberculoma of the optic nerve, retro bulbar optic neuropathy, compressive optic neuropathy, anterior ischemic optic neuropathy and optico-chiasmatic arachnoiditis. Therefore, the differentiation from other inflammatory optic neuropathies may be initially difficult, especially in the absence of systemic involvement. The lack of uniformity in diagnostic criteria, the large variations in clinical presentations, as well as the unclear natural history of optic nerve tuberculoma make the diagnosis difficult [14]. In most cases the diagnosis requires both corroborative evidence, such as a positive Purified Protein Derivative and chest x-ray, and the exclusion of other causes. The evidence could consist of the demonstration of acid-fast organisms under the microscope or the detection of bacterial genome by nucleic acid amplification procedures. However, in our case orbital MRI played an important

role in confirming the diagnosis. In fact, the optic nerve tuberculoma had a pathognomonic aspect: in T1-weighted images, it appears iso-intense to gray matter with a hyper-intense central region, in T2-weighted images, it appears iso-intense to gray matter but with a central region of hypo-intensity representing gliosis and abundant monocyte infiltration [15]. In T1-weighted gadolinium-enhanced magnetic resonance there is a ring enhancement with a restriction in the central region; the diffused-weighted imaging represents the liquid necrosis.

Even if the MRI has specific appearances, the diagnosis can be misleading with other inflammatory diseases such as sarcoidosis or infectious ones especially in immunocompromised hosts (toxoplasmosis, cryptococcosis, lymphoma, and less likely cytomegalovirus) [16]. In fact, the lack of parenchymal brain lesions can make the optic neuropathies diagnosis difficult. Sometimes extensive tuberculous optic nerve involvement with radiological appearance simulate an optic nerve glioma [17].

Antitubercular drugs for optic nerve tuberculoma are similar to those for pulmonary or extra pulmonary tuberculosis. In fact, few studies have described a course of chemotherapy consisting of isoniazid and rifampicin for 9 months [18–21]. Whereas the Centers for Disease Control recommend the use of all four drugs (isoniazid, Rifampicin, Pyrazinamide, and Ethambutol) for an initial 2 months period followed by a choice of different options over the next 4–7 months. In our case, the patient was given all 4 drugs for a period of 9 months. Besides the antitubercular drugs, the utility of adjunctive corticosteroid therapy for 4–6 weeks has been proven by various clinical and experimental studies, especially for cases of tubercular meningitis and pericarditis [22–24]. It may indeed limit the damage to ocular tissues caused from delayed type hypersensitivity.

Conclusion

The optic nerve tuberculoma remains a rare extra-pulmonary manifestation of tuberculosis that can be seen in immunocompromised patients as well as immunocompetent ones. The diagnosis is primarily clinical with the need often for corroborative evidence from other organ systems. Indeed imaging findings, such as the presence of extra-pulmonary tuberculomas or a miliary tuberculosis as well as the typical aspect of the tuberculoma located in the optic nerve in the MRI support the diagnosis, and therefore the decision of therapy. It is a multidisciplinary decision that should be discussed by the radiologist, the ophthalmologist and the pulmonologist in order to contribute more effective care to the patient.

Patient consent

The patient first was discharged against medical advice, then was reported deceased by the family. We tried to reach the family for a consent statement but there were no further answer.

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