DOI: 10.5603/oj.96261

Extrapulmonary tuberculosis presenting as diplopia: a case report of an atypically located tuberculoma in an immunocompetent patient in a non-endemic region

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ABSTRACT

BACKGROUND: Central nervous system tuberculosis occurs due to blood spread, with meningitis being more common than tuberculomas. In non-endemic areas, tuberculomas are more frequent in immunocompromised patients.
CASE PRESENTATION: Our case report presents a young girl who presented to the University and Polytechnic Hospital La Fe, Valencia, Spain. After examination, she was diagnosed with a cerebellar tuberculoma, despite having no other risk factors besides her mother having tuberculosis during her pregnancy. Medical treatment alone, without surgery, was sufficient to treat her condition.
CONCLUSION: Diagnosing tuberculomas can be challenging in non-endemic regions and patients without high-risk factors, particularly since these lesions can present as the initial manifestation of tuberculosis without fever or leukocytosis. Surgical intervention is generally not required for their diagnosis, management, or treatment.

KEY WORDS: diplopia; neurophthalmology; tuberculosis; tuberculoma

Ophthalmol J 2023; Vol. 8, 122–125

INTRODUCTION

Lung affection remains the most common clinical presentation of tuberculosis. The central nervous system is one of the most aggressive forms of extrapulmonary tuberculosis. However, it is not the most common tissue affected in cases of extrapulmonary tuberculosis, except for individuals with acquired immunodeficiency syndrome [1].

Intracranial involvement in tuberculosis is typically the result of hematogenous spread from an extracranial focus. Meningitis is a more common manifestation than tuberculoma. Tuberculomas are more frequently located in the cerebral and cerebellar hemispheres due to a higher blood supply in these areas, while the brain stem is an uncommon location for tuberculomas [2].

In countries where tuberculosis is endemic, tuberculomas are a differential diagnosis to be considered due to their high prevalence, accounting for up to 40% of space-occupying intracranial masses [3]. In areas where tuberculosis is not endemic, this presentation is more common in patients with immune suppression [1].

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CASE PRESENTATION

An 18-year-old girl presented to the Emergency Department of the University and Polytechnic Hospital La Fe, Valencia, Spain, with complaints of double vision. The patient denied any significant medical history. She had been well until the last week before the admission when she experienced paraesthesia in her right limb that lasted for 2 days.

Vital signs were in the normal range, and no abnormalities were detected upon examination of the lungs, heart, abdomen, and extremities. She was alert and oriented. On neurological examination, left abducens nerve (CN VI) palsy was noted. An examination of the ocular motility revealed left-eye esotropia and horizontal diplopia in the primary gaze. Her diplopia disappeared on closing either eye, and it became more pronounced at the left lateral gaze with restricted left eye adduction. Other extraocular movements were normal, and there was no pain or other cranial nerve involvement. There were no concomitant symptoms or other focal neurological deficits on clinical examination.

On the ophthalmological examination, pupils and eyelids were normal. Visual acuity was 20/20 for both eyes. Anterior segment and funduscopic examination were unremarkable.

Blood tests, electrocardiogram, and head computed tomography (CT) scan were performed during her stay in the emergency department but did not reveal any abnormalities.

The patient was admitted for further workup, and an magnetic resonance imaging (MRI) was performed. Neuroimaging revealed a 1.8 cm focal lesion in the left medullary bulbar area with cranial extension to the pons. The lesion had an oval morphology, was hypointense on T2 sequence, and showed intense homogeneous contrast uptake. There was also an extensive perilesional vasogenic edema affecting the entire medullary bulb and dorsal pontine area (Fig. 1). Considering the imaging results and the patient's age, the most likely diagnosis were cerebral lymphoma or pseudotumoral diseases, while brain metastases or atypical infectious processes were considered less likely.

Lumbar puncture did not provide any cytobiochemical or microbiological evidence of relevance. Flow cytometry of the cerebrospinal fluid did not provide any evidence of lymphoma infiltration. The immunophenotypic study of the cerebrospinal fluid showed a predominantly CD4 T lymphocyte population (CD4+: 79.2%, CD8+: 20.8%), with isolated polyclonal B lymphocytes accounting for the remaining 2.5%.

Within a few days, results from complementary tests performed at the emergency department were available. The results were unremarkable, except for a positive Quantiferon, which, in the absence of risk factors or exposure to tuberculosis, was not considered relevant at that moment.

It was decided to start treatment with a megadose of corticosteroids, but there was no improvement in the size of the lesions in the control image study. Hence, a positron emission tomography computed tomography (PET-CT) scan was performed. The lesion in the left brainstem had high glycidic metabolism, with a contralateral substance lesion index of 2.7, indicating a high-grade lesion in the case of primary tumors of the central nervous system. Metabolically, it was not possible to differentiate between lymphoma versus inflammatory lesions. Right supraclavicular hypermetabolic adenopathies were also noted, with a short axis of 9 mm and maximum standardized uptake value (SUVmax) of 5.6 and another with a short axis of 7mm and SUVmax of 2.4. A histological study was advised to rule out malignancy.

Ultrasound-guided fine needle aspiration (FNA) biopsy was performed on the lymphadenopathy, obtaining material with granulomatous structures with minimal cellularity. The little cellularity was made up of lymphocytes, polymorphonuclear

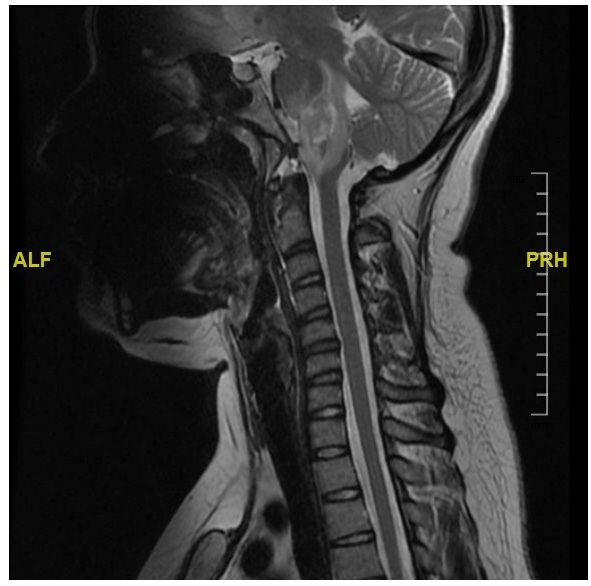

FIGURE 1. Magnetic resonance imaging (MRI) T2 sequence: hypointense oval 1.8 cm lesion located in the left medullary bulbar area with cranial extension to the pons

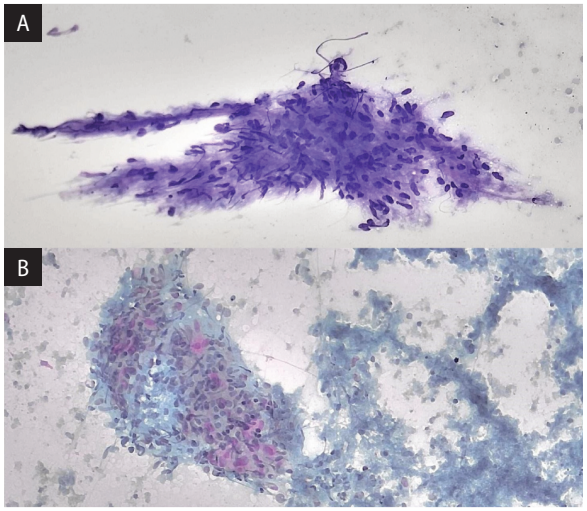

FIGURE 2. Lymph node cytology: Epithelioid histiocytic aggregates, with a spindled morphology, composed of cells with abundant eosinophilic cytoplasm and rounded to oval nuclei on a background composed of cellular debris. **A.** Panoptic staining; **B.** Papanicolau staining

leukocytes, and occasional formations with a granulomatous appearance (Fig. 2).

Upon insistence, the patient's mother reported that she had tuberculosis during the patient's pregnancy. Following the diagnosis, the patient started a quadruple antituberculous therapy, with doses adjusted to weight (65 kg). Drugs included: isoniazid 300 mg/day, pyridoxine 300 g/day, rifampicin 600 mg/day, pyrazinamide 1850 mg/day, ethambutol 1100 mg/day, dexamethasone 10 g/6 hours. On the first day, the patient presented headache and nausea, which were interpreted as a favourable response to the treatment.

She was then discharged and continued her treatment at home for one year. One year later, the clinical symptoms were remitted, and the imaging showed a complete resolution of the lesion.

DISCUSSION

The treatment of tuberculoma is usually medical, and in most cases, there is a great response to medical therapy alone. As in our case, which was entirely resolved with medical treatment only, surgical intervention is not usually required. Surgery is not typically required for diagnosis or treatment, even in cases where there is doubt [4].

As in our case, clinical scenarios and radiological findings are usually sufficient to start medical treatment. The use of steroids is controversial and may be indicated in cases of extensive perilesional edema. However, in our case, the initial megadose did not have any effect on the lesion [4, 5].

In endemic areas, tuberculomas should be considered in the differential diagnosis of any intracranial space-occupying process [3]. In our environment, it should be considered in patients with immunodeficiency or the presence of tuberculosis risk factors1. Identifying patients at risk can be challenging in a world where people move between different continents. Immunocompetent patients, like the one in our case, may present with intracranial tuberculoma as the initial manifestation of tuberculosis. Diagnosing these patients can be challenging, especially since fever and leukocytosis may not be present during the initial presentation of intracranial tuberculomas.

Tuberculomas can mimic a variety of entities depending on their location. They may present in a wide variety of ways, from asymptomatic with a chronic course to subacute presentations with severe intracranial hypertension. The limited available literature and the diversity of clinical manifestations, when tuberculoma is located in the brainstem tuberculoma, hinder the recognition of a typical syndrome.

CONCLUSION

In non-endemic regions and patients without high-risk factors, diagnosing tuberculomas can be a challenge, especially since these lesions can be the initial manifestation of tuberculosis without fever or leukocytosis.

Surgical intervention is typically unnecessary for the diagnosis, management, or treatment of central nervous system tuberculomas.

Ethics statement

The authors certify that they have obtained all appropriate patient consent.

Author contributions

All the authors have made a substantial contribution to the concept or design of the article; or the acquisition, analysis, or interpretation of data for the article.

Conflict of interest

The Authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

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