CASE REPORT

Conjunctival Mycobacteriosis

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Abstract. A 6 1/2- year old Indonesian girl with 30 kg of body weight was consulted to the Department of Child Health, Cipto Mangunkusumo Hospital - Medical School University of Indonesia, Jakarta, for eye surgery tolerance. At 3 1/2 years old, she came for the first time with a granulomatous tumor on her left eye. Excision was done, and histopathologic examination revealed granulation tissue corresponding to tuberculosis. The patient was referred to the Department of Child Health for further exploration. Sensitin test was positive for *Mycobacterium intracellulare*, not for *Mycobacterium tuberculosis*. The tumor reappeared twice and the last histopathologic examination revealed nonspecific granulation tissue. **[Paediatr Indones 1993; 33:182-90]**.

Introduction

Mycobacteriosis is a disease caused by mycobacteria. The most common mycobacteriosis is due to *M. tuberculosis,* usually called tuberculosis.[1,2] Mycobacteria other than *M. tuberculosis* and *M. leprae* are called atypical mycobacteria,[1] which usually live as saprophyte and is not pathogenic for human.[3] However, they also have an opportunistic character. When local resistance is decreased, the infection may progress.[1] Among atypical mycobacteria, the most pathogenic ones are *M. intracellulare* and *M. avium.*[3]

Mycobacteriosis can cause two forms of lesions in the conjunctiva. First, is the *phlyctenular conjunctivitis,* a hypersensitivity reaction to bacilli protein. Bacteria can not be found at the lesion. The second is infection at the conjunctiva, where bacteria can be found at the lesion. Other than conjunctiva, any part of the eye can be infected, such as the palpebrae, cornea, lacrimal glands, retina, uvea (iris, cilliary body, and choroid) and sclera.[4-7] In children, the conjunctiva and cornea are the most frequent affected sites.[5] Conjunctival mycobacteriosis is a very rare disease.[5-7] The first case was reported by Koester in 1873.[8] Since that time only few cases were reported. Con-

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junctival mycobacteriosis usually is caused by *M. tuberculosis*, but it could also be due to atypical mycobacteria.[1,2] Although less common, mycobacteriosis due to atypical mycobacteria usually becomes a problem since the management and clinical course of the disease varied.[9]

Case report

A 6 1/2 year old girl was consulted to the Department of Child Health, Cipto Mangunkusumo Hospital - Medical School University of Indonesia, Jakarta, for preoperative evaluation for eye surgery. When she was 1 1/2 years old, a red small lump appeared on her left eye. Her parents took her to a general practitioner. She was given some medicine and the lump healed. One year later, on the same site a lump reappeared. She began to have night sweat and mild fever. There were no other complaints. She was taken to the same GP and was given similar medicine but there was no improvement.

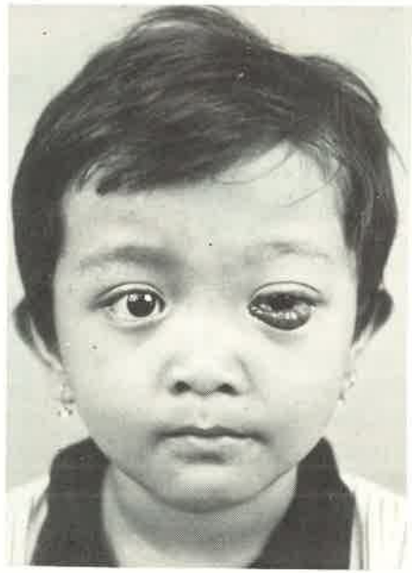
The lump enlarged and one year later it had covered all the white part of her left eye. Her parents took her to the Department of Ophthalmology, Cipto Mangunkusumo Hospital. Excisional biopsy was done and histopathologic examination showed tuberculous granulation tissue. The patient was then referred to the Department of Child Health for further exploration.

The patient's nutritional status was adequate. Her growth and development were normal. The patient had had no basic immunization. A neighbor suffered from chronic cough and had been treated at Persahabatan Hospital, but her parents did not know what the diagnosis was. There was no pet, the water source was from a hand pump well, and the patient had no hobby of swimming.

On her first visit to the Department of Child Health she was 3 1/2 years old, with a body weight 14.5 kg. Her vital signs were normal. On her eyes there was no phlyctene. On her left eye there was a red granulation tissue, which covered the bulbar conjunctiva at the nasal and temporal side, and the inferior tarsal conjunctiva (Figure 1a and 1b). There was lymph node enlargement, but no abnormality of other organs.

The peripheral blood was normal. Mantoux test with PPD RT 23 2TU gave negative reaction. Blood sedimentation rate was 22 mm/hour. Culture for ccid fast bacilli of gastric aspirate was negiative. Chest X-ray disclosed infiltration in both lungs and enlargement of perihilar lymph nodes (Figure 2). Histopathologic examination showed caseation, necrotic area, epitheloid cells, and Langhans giant cells surrounded by lymphocytes (Figures 3 and 4) consistent with tuberculosis.

The diagnosis at that time was conjunctival and pulmonary tuberculosis. The patient was given isoniazid, rifampicin, and ethambutol. On the third month of treatment, the lump enlarged and pedunculated, so there was a part of the tumor protruding between her palpebrae when she closed her eyes. Excisional biopsy was



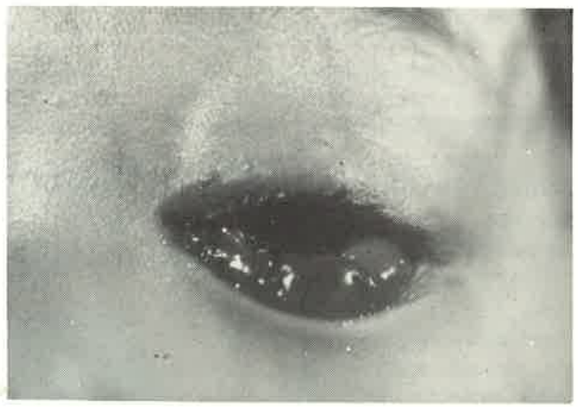


Fig 1a and 1b.

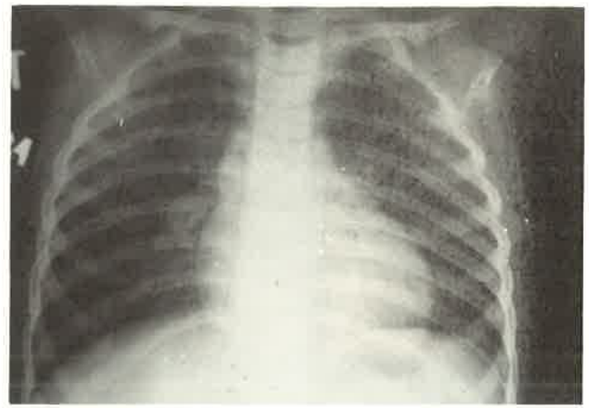


Fig 2.

Fig 3.

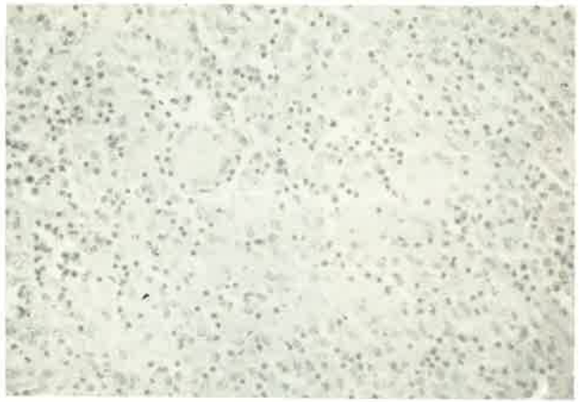


Fig 4.

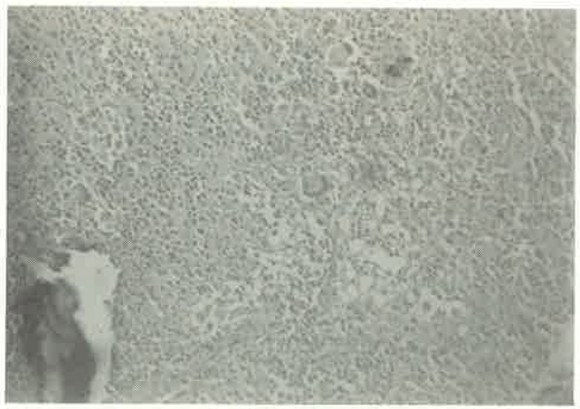


Fig 5.

revealed nonspecific granulation tissue (Figure 5). At that time, the possibility of atypical mycobacteria as the etiology was considered, but unfortunately sensitins for atypical mycobacteria were not available. On the ninth month of treatment, the body weight decreased, chest X-ray showed mediastinal pleurisy (Figure 6), and Mantoux test with PPD RT 23 2TU gave a dubious result. Treatment was continued, and follow up chest X-ray showed improvement (Figure 7).

Sensitin test was done on the sixteenth month of treatment and it gave positive result (14 mm) for *M. intracellulare;* the diagnosis of intracellular Mycobacteriosis was established. The treatment was discountinued after 24 months. Two months later, the lump reappeared. Ten months after reappearance of the lump, once again excision was done, and the result of histopathologic examination was nonspecific granulation tissue (Figure 8).

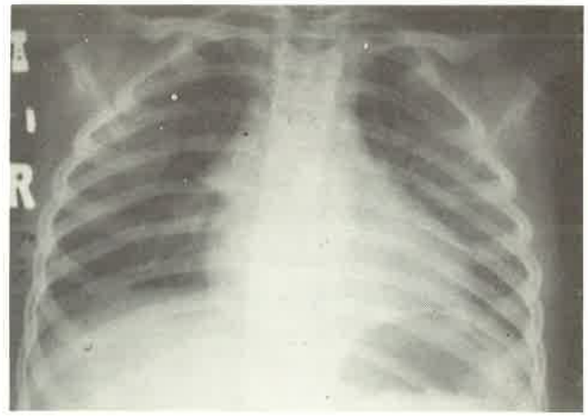


Fig 6.

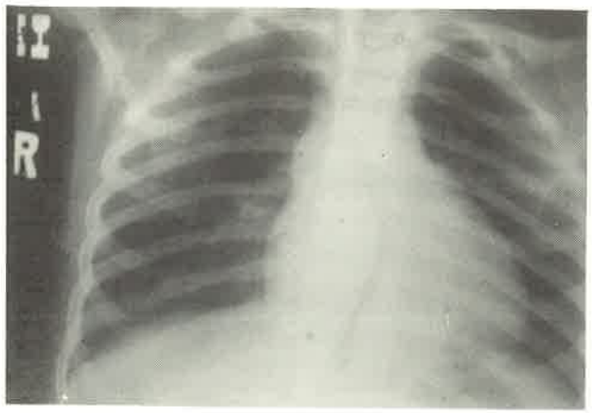


Fig 7.

Fig 8.

Discussion

This case is a very rare one, as we can conclude from the literatures.[5-8] Up till now the exact incidence of this disease is still unknown. To give some pictures, at Baltimore ENT and EYE Hospital there was no case of conjunctival mycobacteriosis among 41,730 patients. Among 139,607 patients at New York Institute of Ophthalmology, there were only 1 case of atypical mycobacteriosis found in 1.000 patients with primary tuberculosis. In Bellevue Hospital, Ghon and Kudlich mentioned only 0.05% cases located at conjunctiva among 2,114 TB patients. At the Department of Ophthalmology Cipto Mangunkusumo Hospital, this case seemed to be the third case of conjunctival mycobacteriosis. The previous two cases were due to *M. tuberculosis.*

When the patient was 1 1/2 years old a small lump appeared on her left eye which was relieved by nonspecific treatment. It seemed that the small lump was a nonspecific hyperplasia. The lesion did not heal completely and became a *locus minoris resistantiae.* One year later the locus was infected by *Mycobacterium intracellulare* where the source of infection maybe something in her environment.

On her first admission to the Department of Ophthalmology, Cipto Mangunkusumo Hospital, it had not been considered that mycobacterium was the etiology of the disease.. Conjunctival tuberculosis was diagnosed when the histopathologic examination revealed specific appearance, since tuberculosis is the most common etiology giving that appearance. Different kinds of mycobacterium cannot bee differentiated by the histopathologic appearance.[11]

The possibility of atypical mycobacteria as the etiology was considered when the tuberculin test was negative, her general condition was good, and the granulation reappeared on her left eye. Sensitin test confirmed that the disease was caused by *M. intracellulare.* Saitz[1] and Huebner[12] stated that sensitin test to be reliable to confirm the diagnosis. Clinical complaints of the patient was rather mild, therefore the parents did not seek further medical help sooner. As Miller[6] has stated, mycobacteriosis of the conjunctiva is characterized by the absence of pain and the slowness of onset.

Clinical classification of conjunctival mycobacteriosis could be differentiated by the route of infection and morphologic appearance.[8] In this case, clinical manifestation was matched to two forms of morphological classification. At first, it appeared as a hypertrophic granulation tissue, and then developed as a pedunculated tumor.

There are two routes of infection, the primary and secondary infection. Primary infection occurs when there is a direct inoculation of bacilli at the conjunctiva of a previously healthy person. Secondary infection happens in a person with mycobacteriosis in other parts of the body, infects him/herself by endogen or exogenous route.[2,8] Different from tuberculosis, infection by atypical mycobacteria is not transmitted from human to human.

The source of infection could be from water, soil, animal, even from plants.[1,3,11]

Most likely, this case was a primary infection. This assumption is based on the age of the patient, absence of basic immunization, the site of lesion at conjunctiva, and unilateral lesion. The literatures mentioned that this primary infection is the most frequent one. In this case, lung involvement was secondary.The lungs were involved, as detected by radiological examination which showed alteration during treatment. The prognosis of conjunctival mycobacteriosis has not been specifically discussed in the literature. Most of the atypical mycobacteria are resistant to antituberculosis drugs. So surgery is a strong indication.[13]

As we can see in this patient, after being given antituberculosis treatment and excision had been done, the granulation reappeared until twice. But the next histopathologic examination did not show specific appearance anymore. It seemed that the granulation had changed from specific to nonspecific process, similar to keloid formation on a skin wound. If it is right the prognosis of this patient is good, since it can be cured by corticosteroid.

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