Case Report

Tuberculous otomastoiditis complicated by thrombosis of the sigmoid sinus in an infant

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A B S T R A C T

Otomastoiditis is a rare and exceptional complication of tuberculosis; its pathogenesis has been debated since its recognition as a distinct condition. The evolution and prognosis in general are modified after the end of antituberculosis treatment. In our observation, we report the case of an infant with tuberculous otomastoiditis complicated by sigmoid sinus thrombosis.

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I n t r o d u c t i o n

Morocco is a tuberculosis endemic country, as are other developing countries. Pulmonary involvement is the most common location, about 85% of cases of tuberculosis, while 15% are extrapulmonary locations [1].

The ear localization remains exceptional, and is manifested by otitis media and chronic suppurative mastoiditis whose tuberculous origin remains rare [2].

We report a rare observation of an infant with a tuberculous otomastoidal disorder complicated by sigmoidal sinus thrombosis.

O b s e r v a t i o n

This is a 15-month-old infant with no recent tuberculosis infection, who presented for a 3-month-old symptomatology of left ear otalgia. A month later, the symptomatology was complicated by the appearance of a left retro-auricular swelling gradually increasing in volume, as well as wheezing dyspnea associated with a dry cough at night and during the day. All evolving in a context of unencrypted fever and alteration of the general condition made of anorexia, and weight loss of 3 kilos over 1 month. The patient was put on antibiotic treatment twice but without any improvement. This motivated the

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parents to consult with an otorhinolaryngologist doctor who performed an ultrasound of the swelling having shown a left retro atrial cystic formation measuring $18 \times 12 \text{ mm}$ rather well limited to thin walls, with finely echogenic content, with next bone cortical continuity solution. A mass puncture was made bringing back SCC of purulent and hematic fluid whose cyto-bacteriological study was negative, but the gene Xpert on the puncture fluid showed the presence of *Mycobacterium tuberculosis*. The patient was referred to us for treatment.

The interrogation was repeated and objectified the notion of administration of unpasteurized goat’s milk 6 months ago.

Clinical examination objectified a hemodynamic and respiratory stable infant, apyretic at $36.9^\circ\text{C}$, eupneic at $37 \text{ cc/min}$, and Normocard at $99 \text{ bat/min}$.

Its weight was $10 \text{ kg (-3DS)}$ and its height was $74 \text{ cm (-3DS)}$.

The ENT examination objectified a soft retro auricular swelling, painful to palpation measuring $4 \text{ cm}$ without inflammatory signs in front (Fig. 1). The remainder of the review was non-specific.

A chest X-ray indicated a deviation of the trachea with bilateral chest distension (Fig. 2).

**BK sputum and gene Xpert in sputum were negative**

A CT scan of the petrous bone showed an appearance in favor of an aggressive left otomastoiditis (bone lysis) (Fig. 3) complicated neighborhood collection with homolateral sigmoid sinus thrombosis (Fig. 4).

The patient was put on antituberculosis treatment, $2\text{RHZE/10RH}$ because of the bone lysis with anticoagulant treatment low molecular weight heparin: $100 \text{ IU/kg/12 h}$ and antivitamin K: $0.2 \text{ mg/kg/jr}$ with INR monitoring each week.

The evolution was satisfied by the regression of the retroauricular swelling as well as the disappearance of the otalgia and the swelling with a weight gain and recovery of the appetite.

**Discussion**

Tuberculous otitis media and tuberculous mastoiditis form a pathological process and are here called tuberculous otomastoiditis [3]. *M tuberculosis* the pathogen involved in 0.05%-0.9% of chronic middle ear infections [9]. This agent can reach
the middle ear by blood or by direct implantation in the external ear canal and perforation of the tympanic membrane [4]. The introduction of unpasteurized goat’s or cow’s milk is now a risk factor in infants as in our case [5]. According to the literature, a considerable delay before diagnosis is often reported ranging from 12 months to several years due to the rarity of this infection [6]. Concerning the clinical picture, a triad was mentioned associated persistent otorrhea despite antibiotic treatment, facial paralysis which can alone evoke the tubercular origin, and multiple tympanic perforation [6].

Our case had the particularity of presenting otalgia that testify to the mastoid extension which joins the case reported in Turkey by Sahbudak et al. in 2012 [2] as well as in the series of Yang [1] which recovered pain in only 13% of cases.

The possible diagnosis of tuberculous otomastoiditis is based either on biological confirmation by the presence of M tuberculosis at the fluid level of the discharge or by gene Xpert which is a real-time PCR test and can in itself even identify the M tuberculosis on an ear swab such as ours, or by histological confirmation that highlights typical granulomatous lesions [1].

Although diagnosis is based on microbiological identification, radiological studies such as CT may suggest specific signs of aggression that may suggest early rare causes of middle ear and mastoid cavity damage [7]. Assessment of pulmonary involvement is essential as up to 50% of patients may have clinical and radiological pulmonary tuberculosis as the case of our patient. Concerning complications, venous thrombosis is a classic complication of acute otitis media and mastoiditis. One published case of an 11-year-old child who had venous thrombosis on otomastoiditis following a prothrombin deficit [2].

After putting the diagnostic of tuberculous otomastoiditis, a medical treatment based on antituberculosis must be established, based on isoniazid, rifampicin, pyrazinamide and ethambutol, the total duration is 6-9 months [3].

Surgical treatment plays an important role in treatment by removing infected debris [3]. Complications requiring a surgical approach include facial nerve palsy, sub-periosteal abscess, labyrinthitis, persistent post-tauricular fistula and extension of central nervous system infection [8].

Ototogenic thrombophlebitis remains a serious infectious complication with high morbidity potential for the children concerned. Anticoagulation can be added safely and could reduce the lasting side effects of the blocked sinus. Thus, it is not necessary to look for high prothrombotic factors in the absence of an appropriate clinical history, as they appear to play a minor role in the development of otogenic thrombophlebitis. The duration of anticoagulation do not need necessarily to be correlated with sinus recanalization and, finally, 3 months of treatment is sufficient and well tolerated without major complications [9].

High awareness is necessary for early diagnosis of tuberculous otomastoiditis. His late diagnosis has been shown to be associated with more complicated treatment procedures and fewer good results. On the other hand, early detection requires simpler treatment procedures and promotes better results. Therefore, early detection of tuberculous otomastoiditis is not only of high diagnostic value, it can also have high prognostic value [10].

Conclusion

Although tuberculosis remains a rare cause of otitis media, the clinician must have a high index of suspicion especially in the developing country where tuberculosis remains a major health problem. As well as it is always necessary to think of the tuberculous origin in front of any otomastoiditis with no improvement under antibiotic treatment.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient’s parent.

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