AGRESYVI ABIPUSĖ SMILKINKAULIŲ TUBERKULIOZĖ. KLINIKINIS ATVEJIS

AGGRESSIVE BILATERAL TEMPORAL BONE TUBERCULOSIS. A CASE REPORT

Liubov Kornilenko[1], Irina Arechvo[2]

[1]Vilniaus universiteto Medicinos fakultetas
[2]Karoliniškių poliklinikos Otorinolaringologijos konsultacijų skyrius

[1]Faculty of Medicine, Vilnius University
[2]Outpatient Otorhinolaryngology Department, Karoliniskiu Polyclinic

SANTRAUKA

Reikšminiai žodžiai: smilkinkaulio tuberkuliozė, persistuojanti otorėja, veidinio nervo parezė.

Smilkinkaulio tuberkuliozė – reta būklė, jai būdingas specifinių simptomų nebuvimas, todėl laiku nenustatoma diagnozė. Dėl ligos retumo ir klinikinių simptomų įvairovės diagnozuodami tuberkuliozinį vidurinį otitą otologai turi būti itin budrūs. Kad atkreiptume gydytojų klinicistų dėmesį į šią patologiją, pristatome unikalų agresyvios eigos vidurinės ausies ir gretimų struktūrų tuberkuliozės atvejį. Iš pradžių ši būklė pasireiškė nespecifinio pūlingo vidurinės ausies uždegimo simptomais. Buvo taikomas įprastas gydymas sisteminiais ir vietiniais priešbakteriniais ir priešgrybeliniais preparatais, tačiau liga progresavo: pasireiškė būgninės ertmės gleivinės ir klausomųjų kauliukų staigus irimas, atsirado pilkų blyškių granuliacijų vidurinėje ausyje. Didelės skiriamosios gebos smilkinkaulių kompiuterinė tomografija, atlikta ligos pradžioje, parodė nespecifinį abipusį otomastoiditą, normalios pneumatizacijos spenines ataugas abipus ir nepakitusias atiko lateralines sienas. Ligoniui išsivystė veidinio nervo paralyžius ir smilkinkaulio osteomielito požymiai. Diagnozuojant vidurinės ausies tuberkuliozę, buvo remiamasi klinikinių simptomų, kompiuterinės tomografijos duomenimis ir pulmonologo patvirtinta recidyvuojančios plaučių tuberkuliozės diagnoze. Kruopščiai surinkta anamnezė, atkreipiant dėmesį į buvusią ar esamą plaučių tuberkuliozę, būtina teisingai diagnozei patvirtinti.

ABSTRACT

Key words: temporal bone tuberculosis, persistent otorrhea, facial nerve palsy.

Temporal bone tuberculosis is a rare condition with a lack of disease-specific signs. Due to the rarity and variety of clinical manifestations, tuberculous otitis media is requiring a high index of suspicion among otologists. To increase awareness of this condition, we report the unique recent case of bilateral aggressive tuberculous otitis media. The disease presented as an unspecific purulent otitis media. In spite of the conventional treatment with systemic and topic antibacterial and antifungal preparations more suspicion symptoms occurred: sudden disappearance of the ossicular and tympanic cavity mucous membrane, nodular granulations in the middle ear cleft. Unspecific otomastoiditis without sclerotic changes and scutum destruction was radiologically diagnosed at the beginning. Facial palsy and osteomyelitis of the temporal bone with sequestration were estimated. The diagnosis was put forward basing on the clinical symptoms, computed tomography data and reactivated pulmonary tuberculosis confirmed by pulmonologist. Thorough history with special emphasis to the previous lung tuberculosis is crucial for establishing of the correct diagnosis.

INTRODUCTION

Tuberculosis (TB), a multisystemic disease with myriad presentations and manifestations, is the most common cause of infectious disease – related mortality worldwide [1]. The disease can involve any organ. Approximately 85 % of reported TB cases are limited to the lungs and the remaining 15 % involve extrapulmonary or both extrapul-

Irina Arechvo
Karoliniškių poliklinikos
Otorinolaringologijos konsultacijų skyrius
L. Asanavičiūtės g. 27a, Vilnius
arechvo@gmail.com

monary and pulmonary sites [2]. Tuberculous otitis media (TOM) is a rare disease, accounting for between 0,04 % and 0,9 % of chronic suppurative otitis media cases [3]. Most cases of tuberculous otitis media are secondary, transmitted from primary focus [4]. Clinical presentation of the TOM is non-specific. Furthermore, there is a diminishing awareness amongst physicians of the threat still posed by tuberculosis. Due to the rarity and variety of clinical manifestations, tuberculous otitis media is requiring a high index of suspicion. To increase awareness of this condition, we describe the recent case of aggressive bilateral temporal bone tuberculosis.

CASE REPORT

A 40-year-old man presented to an outpatient otorhinolaryngology department with a 3-week history of hearing loss, ear pain and otorrhea bilaterally. Otoscopy revealed a subtotal perforation with mucous-purulent secretion in the tympanic cavity on the left and central perforation in posterior superior quadrant on the right side (Fig. 1A). The puretone audiogram revealed moderate-to-severe mixed hearing loss up to 78 dB with air-bone gap bilaterally.

Initially Neomycine followed by Cyprofloxacine drops were prescribed. The patient came back in several weeks without any improvement. Swab test showed the presence of Candida species. Clotrimazole cream was prescribed with the same negative response. Two weeks later the denuded ossicles with incus and malleus dislocations and pathological movements (especially of the malleus manubrium) of the both ossicles were detected with the pale pink attic and tympanic cavity granulations on the left (Fig. 2). There was the second marginal perforation in the anterior quadrants on the right side (Fig. 1B).

At the same time a facial palsy grade IV according to House-Brackmann scale developed. A high resolution computed tomography (CT) scans were obtained (Fig. 3). The course of the intratemporal facial nerve on the both sides was evaluated without obvious erosion of the Fallopian canals (Figs. 3B and 3C). The otoscopically detected fracture of the malleus manubrium on the left was confirmed radiologically (Fig. 3D). Tubercolous inflammation of the both temporal bones was suspected.

A history of the previous pulmonary tuberculosis and chronic alcoholism was obtained from the patient relatives. The results of the chest X-ray obtained earlier showed advanced tuberculous foci in the apex of the lung on the right side. The granulations from the attic and tympanic cavity were biopsied with negative results for Mycobacterium. Histology revealed a chronic granulomatous suppurative otitis media with mixed inflammatory cell infiltration. Candida species were detected microscopically as well. The patient was referred to the pulmonologist and the antituberculous treatment was started. One year later due to the severe hepatic dysfunction specific management was discontinued and the patient visited our department for

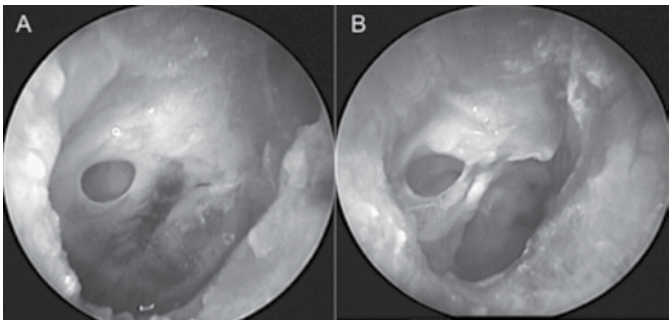

Fig. 1. Central tympanic membrane perforation on the right side (A). Progressive eardrum destruction – the second anterior marginal perforation two weeks later (B)

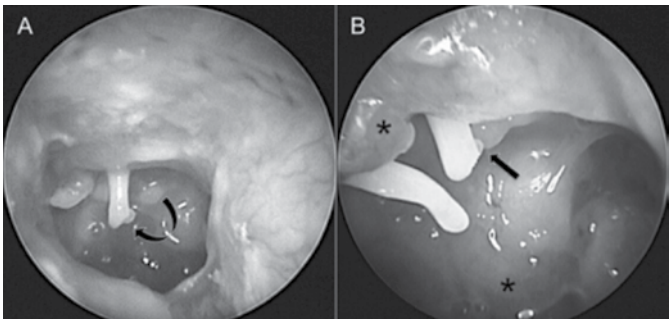

Fig. 2. Otoscopically the denuded pathologically mobile ossicles (curved arrow) were detected on the left side (A). Disarticulation of the ISJ (arrow) with pale pink tympanic cavity and attic granulations (asteriks) (B). Pathologically mobile the lower one third of the malleus manubrium was detected during gentle palpation of the ossicles

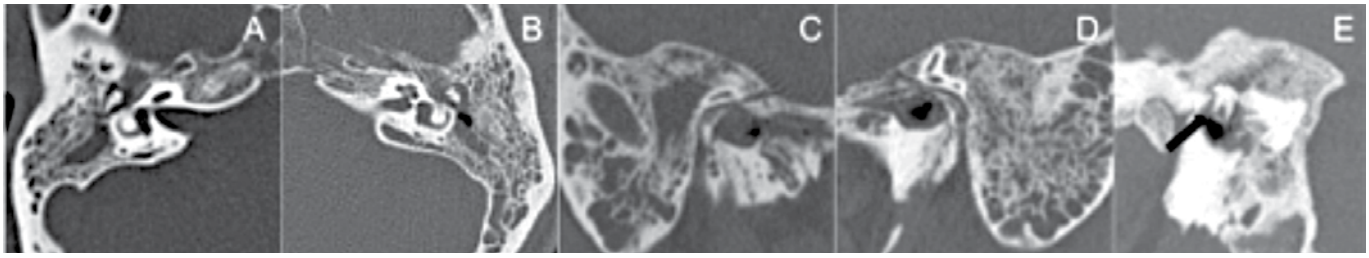

Fig. 3. Tuberculous otomastoiditis on both sides without obvious temporal bone erosion (A, B). The intratemporal course of the facial nerve on the right (C) and on the left side (D) without bone canal erosion. Oblique sagittal reformat (E) with MIP technique showing a malleus handle fracture (arrow).

further evaluation. His facial dysfunction improved with House-Brackmann grade II (Fig. 4).

However, some new complaints added. He started to feel a painful opening of the mouth as well as movements of the lower jaw. He also reported an abnormal click of the right side with opening of the mouth. Otoscopy revealed a movable bone sequester in the medial anterior wall of the external ear canal appearing with closed mouth and disappearing with opened (Fig. 5).

For one year after the initial diagnosis was proposed purulent otorrhea was still persisted from both ears. During otoscopy there were multiple eardrum perforations and granulations within the tympanic cavity. There was more denuded bone on the promontorium. One year later patient died due to the multiple organ dysfunction syndrome.

DISCUSSION

Temporal bone tuberculosis (TBT) is a rare otologic condition. To date, fewer than 200 (≈ 1 % of all tuberculosis cases) cases can be found in the English-language literature. Although tuberculous involvement of the temporal bone was described early in the 18th century [5] there are still diagnostic and therapic challenges with TOM. The lack of experience managing this rare condition and high index of suspicion may cause serious diagnostic difficulties among otologists. In the onset of the disease the TBT may manifest as a common bacterial otitis externa and media resistant to routinaly used antibiotics. Long-term usage of the local antibiotic drops can lead to fungal overgrowth that can delay correct diagnosis even longer.

Previous authors put a special attention on early TOM diagnosis establishment and as soon as possible beginning of specific antituberculous treatment because of destructive nature of disease [6].

According to literature biopsy from granulation tissue is the most reliable diagnostic method of TOM [7]. We have faced the problem with confirmation of the definite diagnosis. In our case the culture and biopsy from the tympanic cavity and EEC were negative for M. tuberculosis. More accurate histologic confirmation of the true granulomas with caseous foci surrounded by epithelioid and multinucleated inflammatory cells were described with intraoperative biopsies [7]. A diagnosis of the bilateral temporal bone tuberculosis was based on the clinical picture, CT data and reactivated pulmonary tuberculosis confirmed by pulmonologist.

Described earlier and well-known to the otologists of the previous generations the symptoms of TOM nowadays should be re-emphasized and revised in accordance with the imaging data. The clinicians should be especially alert in case of atypical destruction of the ossicles (denuded "bald" ossicles) and/or in cases of fast loss of mucous lining of the tympanic cavity as well as the presence of the the nodular pale pink granulations in it. Unsuccessful treatment with standard antibacterial and antifungal medications should raise a further suspicion. Perhaps, in our case a Candida species superinfection occurred following long-lasting treatment with Neomycine and Ciprofloxacine drops.

To our knowledge a destruction of the anterior walls of the tympanic cavity and EEC with a moving sequester and typical click with opening of the mouth was described for the first time. Probably, this was a consequence of the progressive osteitis, describied in such patients [8] with further sequestration. It should emphisized that the CT images obtained in the initial phase of the disease didn't show any destructive changes. Unspecific otomastoiditis without sclerotic changes and scutum destruction was radiologically diagnosed at the beginning. These finding are in the agreement with the recent results of Rho et al. [9]. The history of the previous active pulmonary tuberculosis was undetected and some diagnostic delay occurred.

Recently, the tuberculosis infections are described in association with AIDS or other immunosupressive conditions [10]. In our case the patient had a history of chronic alcoholism, which in association with his uncooperatie behavior were the main causes of treatment failure and death of the patient.

Mycobacterial infection can achieve middle ear by three ways – through the EEC, Eustachian tube or blood

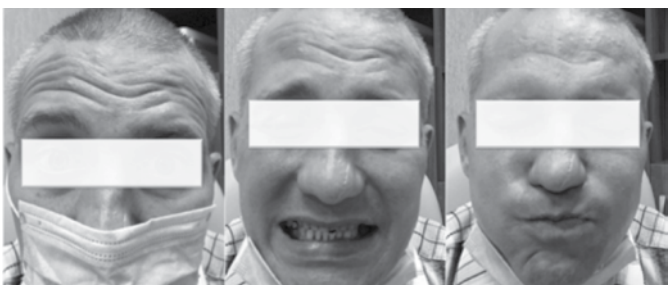

Fig. 4. Improvement of the facial nerve function (House-Brackmann grade II) with conservative antituberculous treatment

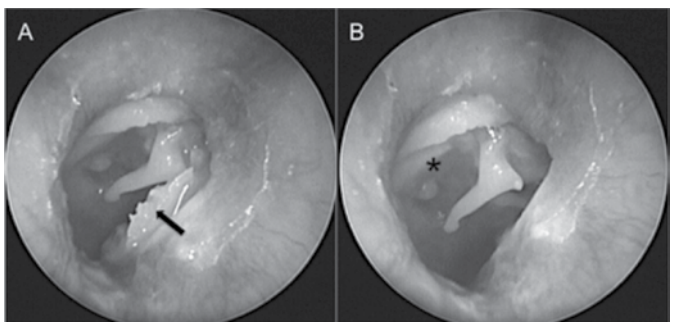

Fig. 5. A bone sequester (arrrow) within anterior wall of the EEC appearing with closed mouth (A) and disapperaring with closed (B). A resorption of the incus occurred as well (asterisk)

[11]. We suppose that in our patient two latter ways were possible – hematogenously from the active pulmonary loci or via Eustachian tube from the nasopharynx as the both temporal bones were involved.

Previous investigators proposed that surgical treatment might be useful in case of facial nerve paralysis [6]. In our case the symptoms of the latter condition improved with antituberculous therapy. The surgery is also useful to obtain sufficient tissue for histologic confirmation of the disease. In our case a biopsy from the tympanic cavity through the tympanic membrane perforation was negative.

CONCLUSION

Initially temporal bone tuberculosis may appear as an unspecific purulent otomastoiditis. However, the rapidly progressive destructive soft tissue and bony changes, unresponsiveness to the standard treatment protocols, typically appeararing denuded ossicles, pale granulations in the tympanic cavity, hearing loss disproportionate to otoscopic appearance [12] should rase a suspicion of temporal bone tuberculosis. In the latter stages of the disease facial palsy and osteomyelitis of the temporal bone with sequestration may appear. Thorough history with special emphasis to previous lung tuberculosis is crucial for establishing of the correct diagnosis.

REFERENCES

1. Herchline TE, Amorosa JK. Tuberculosis. 2015 Oct [cited 2016 Jan 3]. http://emedicine.medscape.com/article/230802-overview.
2. Diagnostic standards and classification of tuberculosis in adults and children. Am J Respir Crit Care Med., 2000; 161(4): 1376–95.
3. Awan MS, Salahuddin I. Tuberculous otitis media: two case reports and literature review. Ear Nose Throat J., 2002; 81: 792–4.
4. Hwang KE, Kim SY, Chung JS, Lee SH, Choi KH, Park DS, et al. Tuberculous otitis media with endobronchial tuberculosis. J Infect Chemother, 2012; 18(6): 951–4.
5. Simon Schaaf H, Zumla A. Tuberculosis: a comprehensive clinical reference. 1st ed. Philadelphia, PA Saunders Elsevier, 2009.
6. Chmielik LP, Ziolkowski J, Koziolek R, Kulus M, Chmielik M. Ear tuberculosis: clinical and surgical treatment. Int J Pediatr Otorhinolaryngol., 2008; 72(2): 271–4.
7. Nicolau Y, Northrop C, Eavey R. Tuberculous otitis in infants: temporal bone. Histopathology and clinical extrapolation. Otol Neurotol., 2006; 27(5): 667–71.
8. Aremu SK, Alabi BS. Tuberculous otitis media: a case presentation and review of the literature. BMJ Case Rep., 2010 Dec 1.
9. Rho MH, Kim DW, Kim SS, Sung YS, Kwon JS, Lee SW. Tuberculous otomastoiditis on high-resolution temporal bone CT: comparison with nontuberculous otomastoiditis with and without cholesteatoma. AJNR Am J Neuroradiol., 2007; 28(3): 493–96.
10. Kahane J, Crane BT. Tuberculous otitis media. Otol Neurotol., 2009; 30(6): 865–66.
11. Gonzales OY, Adams G, Teeter LD, Bui TT, Musser JM, Graviss EA. Extrapulmonary manifestations in the large metropolitan area sith a low incidence of tuberculosis. Int J Tuber Lung Dis., 2003; 7(12): 1178–86.
12. Abes GT, Abes LB, Jamir JC. The variable clinical presentation of tuberculosis. Otitis media and the importance of early detection. Otol Neurotol., 2011; 32(4): 539–43.

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