CASE REPORT

Non-tuberculous mycobacterial infection of the parotid gland in an immunocompetent elderly patient

Toshiaki Yamanaka, Hideyuki Okamoto, Hiroshi Hosoi

Department of Otolaryngology-Head and Neck Surgery, Nara Medical University, Nara, Japan

Correspondence to

Dr Toshiaki Yamanaka,
toshya@naramed-u.ac.jp

SUMMARY

We experienced an extremely rare case in which combined antibacterial therapy for a non-tuberculous mycobacteria (NTM) infection of the parotid gland achieved a favourable outcome in an elderly immunocompetent patient. Although a 79-year-old man, who presented with swelling and fistula formation in the left parotid gland region, initially received combined antituberculous therapy due to a positive result of acid-fast staining, the lesion did not respond to these agents. Thereafter, since the culture examination did not detect *Mycobacterium tuberculosis* or NTM, we excluded tuberculosis and considered the possibility of an NTM infection caused by a rare mycobacterial species. Therefore, we switched to the clarithromycin-based antibacterial treatment for eight consecutive months without a surgical intervention, resulting in the complete disappearance of the lesion and no evidence of recurrence detected for 4 years. This conservative chemotherapy might be a feasible alternative to a surgical intervention for treatment against NTM infections of the parotid gland.

BACKGROUND

Non-tuberculous mycobacteria (NTM) is the generic name given to mycobacteria other than *Mycobacterium tuberculosis* and *Mycobacterium leprae*. NTM infections are considered to be acquired from environmental exposure. Unlike tuberculosis, NTM infections do not exhibit human-to-human transmission. The incidence of NTM infections affecting the lungs has gradually increased in recent years.¹

On the other hand, NTM infections originating from the parotid region are very rare, and most reported cases have involved children.²⁻³ In previous reports, surgical incision and/or treatment with multiple antibacterial agents were selected. However, no effective treatment strategy has yet been established for such infections. Moreover, although technical improvements have been made in the method used to diagnose NTM infections, it is still difficult to precisely diagnose them. We report a rare case of an NTM infection of the parotid gland in an elderly immunocompetent patient.

CASE PRESENTATION

In August 2008, a 79-year-old man who presented with swelling and tenderness in the left parotid gland region received antibiotic treatment (flomoxef sodium, piperacillin sodium and clindamycin) at a

primary clinic for 2 weeks. Nevertheless, since the lesion continued to increase in size, he was referred to our tertiary hospital.

The lesion was found to be a tumorous mass in the left parotid gland, which measured 75×55 mm in size, and exhibited violaceous discolouration, fistula formation and skin breakdown in the swollen region (figure 1). An enhanced CT scan showed a low-density region that displayed rim enhancement indicating that parts of the parotid gland were necrotic (figure 2). The patient had no history of any remarkable findings including either tuberculosis or intrapulmonary lesions. Blood examinations revealed a white blood cell count 8525/μL, a C reactive protein level 4.72 mm/h, and normal blood glucose levels and liver and renal function. A test for HIV antibody was negative. Radiographic and CT examinations did not detect any pulmonary lesions that were suggestive of tuberculosis or an NTM infection. A cytological examination revealed severe inflammation, but not malignancy. The cultures for aerobes, anaerobes and fungi were negative. Although a Gaffky test detected acid-fast bacteria being classified as class V, the PCR analysis did not detect *M. tuberculosis*, *Mycobacterium avium* or *Mycobacterium intracellulare*. Specific methods including high-performance liquid chromatography for mycolic acids and molecular assays using DNA hybridisation were not performed for the identification of any minor mycobacteria because the patient was unable to afford it and due to the lack of necessary facilities in our laboratory services.

Figure 1 A photograph showing the violaceous discoloured lesion in the left parotid gland and the associated skin breakdown.

To cite: Yamanaka T, Okamoto H, Hosoi H. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-200990

Figure 2 An enhanced CT scan showing the subcutaneous necrotic tissue and the rim enhancement exhibited by the lesion (arrow) in the left parotid gland.

We initially applied the antituberculous agents involving rifampicin (RIF) 450 mg/day, isoniazid (INH) 300 mg/day and ethambutol hydrochloride (EB) 750 mg/day and continued this treatment for 6 weeks since tuberculosis could not be completely excluded. However, the lesion did not respond to these antituberculous drugs. Thereafter, the culture examination did not detect *M tuberculosis* or any NTM species and the positive result (class V) remained in another Gaffky test. Thus, we considered the possibility of an NTM infection caused by a rare mycobacterial species, and hence, added two antibiotic drugs, clarithromycin (CAM) 800 mg/day and levofloxacin (LVFX) 300 mg/day, to the three initially applied drugs. Alternatively, we could also have considered surgical excision, for example, parotidectomy, if the antibacterial therapy had only achieved limited results. The administration of the 5-drug antibacterial therapy for three consecutive months reduced the mass and ameliorated the patient's fistula formation. Since the lesion showed an improvement after the addition of CAM and LVFX to the antituberculous therapy, we switched to a combined 3-drug antibacterial therapy involving CAM 400 mg/day, RIF 450 mg/day and LVFX 300 mg/day, without INH and EB. This combination therapy was continued for the next 5 months and resulted in complete remission.

OUTCOME AND FOLLOW-UP

The lesion completely disappeared (figure 3), and MRI did not show any lesion in the left parotid gland (figure 4). No recurrence of the NTM infection has been detected for more than 4 years.

DISCUSSION

NTM infections of the parotid gland are very rare. There are some reports about such infections in children,^{2 4-7} but very few adult cases have been reported.^{3 8-10} In adults, the NTM infections of the parotid gland have only been reported in patients who had a background of immunosuppression induced by HIV infection or steroid maintenance therapy for rheumatism,^{3 8 9} except in one report describing the case of a 35-year-old middle-aged immunocompetent woman.¹⁰ This is the first report of a parotid gland NTM infection to involve an elderly (79-year old) immunocompetent man.

In general, most NTM infections are caused by *M avium* or *M intracellulare*. Some cases with NTM of the parotid gland caused by *Mycobacterium fortuitum*,⁵ *Mycobacterium chelonae*,^{6 10} or *Mycobacterium scrofulaceum*^{2 8} have been reported in addition to these major mycobacteria. In our case, NTM was not detected

Figure 3 A photograph showing the disappearance of the lesion from the left parotid gland.

in culture. However, we considered that the tumorous lesion observed in this case was caused by rare mycobacterial species such as *M fortuitum*⁵ or *M chelonae*,^{6 10} because the PCR analysis did not detect *M avium* or *M intracellulare*. It is difficult to detect various minor mycobacterial species as their detection is influenced by conditions such as the culture period, temperature and humidity.¹¹ This might explain why NTM were not detected in the present case.

NTM have been isolated environmentally from soil, dust, water and food and they were recently isolated from bathtub inlets and showerhead biofilm.¹²⁻¹⁴ It was suggested that most NTM infections of the parotid gland are caused by *Mycobacterium* ascending via Stensen's duct in the oral cavity.^{3 5} As the patient liked the garden, the NTM in the present case might have originated from soil on the patient's hand. NTM infections of the parotid gland usually result in the production of a tumour-like mass within a short period. In an untreated case,

Figure 4 An MR image of the left parotid gland taken after the combined antibacterial therapy, which does not show any lesions.

the mass softened and its form fluctuated.¹⁵ Moreover, skin breakdown and pus discharge from a fistula were observed during the development of the lesion.¹⁶ Our patient displayed similar clinical features to these previous reports.^{15 16}

It has been reported that surgical treatment is more effective than antibiotic treatment alone for NTM infection-induced parotid lymphadenitis in younger patients.^{4 13} In our elderly patient, who possessed a large progressively developing mass, surgical intervention with parotidectomy would have necessitated the reconstruction of a large skin defect and might have caused facial palsy. In addition, it has been reported that CAM-based multiple antibacterial drug regimens are recommended for the treatment of NTM infections of the parotid gland before surgery.^{6 7 17 18} Therefore, we decided to employ combined antibacterial therapy as a first-line treatment. Eventually, the conservative therapy alone without surgical intervention produced a successful result. The effect of antituberculous drug treatment against NTM infections in the parotid gland is unclear. However, the favourable result achieved in the present study suggests that CAM-based antibiotic therapy with multiple antituberculous drugs might be a feasible treatment against NTM infections in cases in which specific mycobacterial species cannot be identified, as was experienced in the present case.

In conclusion, an elderly immunocompetent patient suffering from an NTM infection of the parotid gland was successfully treated with combined antibacterial therapy, in the present case. Therefore, CAM-based multiple antibacterial drug therapy might be a feasible first-line treatment against intractable infectious lesions in the parotid gland in cases in which the characteristic symptoms of an NTM infection are present and tuberculosis has been excluded, even if mycobacterial species cannot be detected.

Learning points

- ▶ This is the first report of a parotid gland non-tuberculous mycobacteria (NTM) infection to involve an elderly immunocompetent adult.
- ▶ The clarithromycin-based antibacterial treatment achieved a favourable outcome in a 79-year-old man who was suffering from NTM infection of the parotid gland.
- ▶ Combined antibacterial therapy might be a feasible alternative to a surgical intervention for treatment against NTM infections of the parotid gland, especially in elderly patients.

Contributors TY was involved in the data collection and analysis and in the writing of the manuscript, and took responsibility for the integrity of the content of the manuscript. HO has analysed the data and edited the manuscript. HH has revised the manuscript.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Griffith DE, Aksamit T, Brown-Elliott BA, *et al.* An official ATS/IDSA statement: diagnosis, treatment, and prevention of nontuberculous mycobacterial diseases. *Am J Respir Crit Care Med* 2007;175:367–416.
- 2 Cox HJ, Brightwell AP, Riordan T. Non-tuberculous mycobacterial infections presenting as salivary gland masses in children: investigation and conservative management. *J Laryngol Otol* 1995;109:525–30.
- 3 Gittinger FS, Raible A, Kempf VA. Non-tuberculous mycobacterial infection of the parotid gland in an immunosuppressed adult. *J Med Microbiol* 2008;57:536–9.
- 4 Lindeboom JA, Kuijper EJ, Bruijnesteijn van Coppenraet ES, *et al.* Surgical excision versus antibiotic treatment for nontuberculous mycobacterial cervicofacial lymphadenitis in children: a multicenter, randomized, controlled trial. *Clin Infect Dis* 2007;44:1057–64.
- 5 Chen CC, Chen SY, Chen YS, *et al.* *Mycobacterium fortuitum*-induced persistent parotitis: successful therapy with clarithromycin and ciprofloxacin. *Head Neck* 2007;29:1061–4.
- 6 Shyur SD, Chu SH, Wu YL, *et al.* Bilateral parotitis caused by *Mycobacterium chelonae* in an immunocompetent child. *J Microbiol Immunol Infect* 2009;42:530–3.
- 7 Padovani D, Aimoni C, Grasso DL, *et al.* Non tuberculous mycobacteria infection of the parotid region: two familiar cases. *Auris Nasus Larynx* 2007;34:577–9.
- 8 Lawn SD, Checkley A, Wansbrough-Jones MH. Acute bilateral parotitis caused by *Mycobacterium scrofulaceum*: immune reconstruction disease in a patient with AIDS. *Sex Transm Infect* 2005;81:517–18.
- 9 Benharrats I, Jacob L, Taulera O. Parotitis due to *Mycobacterium* and HIV infection. *Rev Med Interne* 1998;19:676–7.
- 10 Shaaban HS, Bishop SL, Menon L, *et al.* *Mycobacterium chelonae* infection of the parotid gland. *J Glob Infect Dis* 2012;4:79–81.
- 11 Piersimoni C, Scarparo C. Extrapulmonary infections associated with nontuberculous mycobacteria in immunocompetent persons. *Emerg Infect Dis* 2009;15:1351–8.
- 12 von Reyn CF, Waddell RD, Eaton T, *et al.* Isolation of *Mycobacterium avium* complex from water in the United States, Finland, Zaire, and Kenya. *J Clin Microbiol* 1993;31:3227–30.
- 13 Nishiuchi Y, Tamura A, Kitada S, *et al.* *Mycobacterium avium* complex organisms predominantly colonize in the bathtub inlets of patients' bathrooms. *Jpn J Infect Dis* 2009;62:182–6.
- 14 Feazel LM, Baumgartner LK, Peterson KL, *et al.* Opportunistic pathogens enriched in showerhead biofilms. *Proc Natl Acad Sci USA* 2009;106:16393–9.
- 15 Robson CD, Hazra R, Barnes PD, *et al.* Nontuberculous mycobacterial infection of the head and neck in immunocompetent children: CT and MR findings. *AJNR Am J Neuroradiol* 1999;20:1829–35.
- 16 Panesar J, Higgins K, Daya H, *et al.* Nontuberculous mycobacterial cervical adenitis: a ten-year retrospective review. *Laryngoscope* 2003;113:149–54.
- 17 Shah MB, Haddad J Jr. Nontuberculous mycobacteria-induced parotid lymphadenitis successfully limited with clarithromycin and rifabutin. *Laryngoscope* 2004;114:1435–7.
- 18 Tunkel DE. Atypical mycobacterial adenitis presenting as a parotid abscess. *Am J Otolaryngol* 1995;16:428–32.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit

<http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow