

## Actinomycosis of the middle ear

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### Abstract

**Objective:** To present the first case of middle-ear actinomycosis in the UK in the last 60 years. The diagnosis and management of actinomycosis of the middle ear is also presented, as well as a review of the recent literature.

**Case report:** This paper reports the case of a nine-year-old girl who presented with recurrent otorrhoea caused by actinomycosis. Mastoid exploration with clearance of the infected tissue, in conjunction with six months amoxicillin, resulted in long-term disease remission. Histology revealed Gram-positive, silver avid (on Grocott staining) and diastase-resistant periodic acid-Schiff negative staining of colonies; this profile was in keeping with the characteristic 'sulphur granules' of actinomycosis.

**Conclusion:** Actinomycosis of the middle ear and mastoid is rare, with less than 45 cases presented in the worldwide literature. This case confirms that the disease should be treated with full surgical clearance and long-term antibiotics.

**Key words:** Ear, Middle; Mastoiditis; Actinomycosis

### Introduction

Actinomycosis is a bacterial disease caused by actinomycetes species. Actinomycetes are anaerobic or facultatively anaerobic Gram-positive bacteria from the actinobacteria genus. The disease commonly presents with granulomatous abscesses of the oral-cervicofacial region, but other sites including the thorax and abdomen have been described.<sup>1</sup>

The involvement of the temporal bone and middle ear is far less common. To date, approximately 43 cases of tympanomastoid actinomycosis have been presented in the literature. These cases have been distributed across the world; however, prior to the current paper, a case had not been reported in the UK for over 60 years.

### Case report

A nine-year-old female was referred for a second opinion because of a history of intermittent left otorrhoea since early childhood. She had been seen in another ENT department, and had received numerous courses of topical and systemic antibiotics without a definitive diagnosis. Her past medical history was unremarkable.

At the initial assessment her left ear was discharging, with granulation tissue visible on the tympanic membrane. An audiogram revealed a moderate conductive hearing loss in the affected ear and normal hearing in her right ear. In view of these findings, an examination of her left ear was undertaken under general anaesthetic. This revealed an inflammatory polyp arising from an area of myringitis on the pars tensa. The remaining tympanic membrane was featureless.

A myringotomy was performed and thick, glue-like secretions were aspirated. At this time there appeared to be a white plaque visible within the middle ear, which raised the possibility of a cholesteatoma. The polyp was removed

and sent for histological examination, and was reported as 'an inflammatory aural polyp'. A swab taken at the time of the examination cultured an antibiotic-sensitive *Staphylococcus aureus*. A computed tomography scan revealed complete opacification of the middle-ear cleft, with no evidence of ossicular or scutum erosion (Figure 1).

In view of these findings, a combined approach tympanoplasty was performed. This was conducted under anaesthetic,

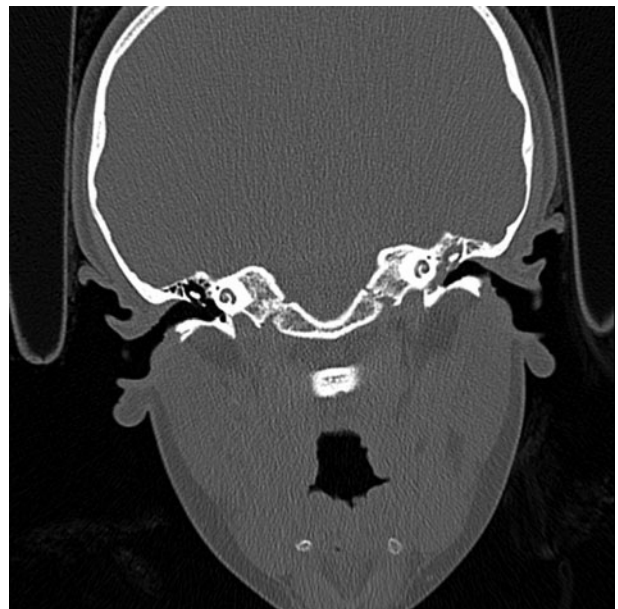


FIG. 1

Coronal (high-resolution, unenhanced) computed tomography image showing complete opacification of the left hypo-, meso- and epi-tympanum.

TABLE I  
CASES OF MIDDLE-EAR ACTINOMYCOSIS\*

Author	Age (y), sex	Disease extent	Treatment	Outcome	Location
Gazzano <i>et al.</i> , 2010 <sup>7</sup>	8, F	Middle ear, mastoid	Amoxicillin or pristinamycine, 3 mth po	Resolved	Dijon, France
Budenz <i>et al.</i> , 2010 <sup>8</sup>	12, F	Middle ear, mastoid, petrous apex, inner ear	Petrosectomy, clindamycin + ampicillin sulbactam IV, clindamycin 6 mth po	Resolved	New York, USA
Shishegar <i>et al.</i> , 2009 <sup>9</sup>	24, M	Middle ear & mastoid	Mastoid exploration, antibiotics not documented	Not documented	Shiraz, Iran
Mehta <i>et al.</i> , 2007 <sup>10</sup>	11, M	Middle ear, mastoid, inner ear	Mastoidectomy, partial labyrinthectomy, ampicillin sulfabactam 9 wks IV	Resolved	Pittsburgh, USA
Sivarajasingam & Rajan, 2007 <sup>11</sup>	78, M	Middle ear, mastoid	Mastoidectomy, ampicillin 6 wks po	Resolved	Malaysia
Sobol <i>et al.</i> , 2004 <sup>12</sup>	14, F	Middle ear, mastoid	Mastoidectomy + grommet, penicillin 6 mth po	Resolved	Pennsylvania, USA
Ajal <i>et al.</i> , 1997 <sup>3</sup>	10, M	Middle ear, mastoid	Mastoidectomy + grommet, penicillin 4 wks IV, penicillin 3 mth po	Resolved	Darlinghurst, Australia
Hoshino <i>et al.</i> , 1996 <sup>13</sup>	24, F	Middle ear, mastoid	Myringotomy, cefuroxime 4 d + unspecified on-going treatment	Resolved	Hamamatsu, Japan
Tarabichi & Schloss, 1993 <sup>14</sup>	7, F	Middle ear, mastoid	Mastoidectomy + grommet, penicillin 4 wks IV, penicillin 3 mth po	Resolved	Wisconsin, USA
Olson <i>et al.</i> , 1989 <sup>15</sup>	6, M	Middle ear, mastoid	Mastoidectomy, penicillin 6 mth po	Resolved	California, USA
Shelton & Brackmann, 1988 <sup>16</sup>	10, M	Middle ear, mastoid	Mastoidectomy, penicillin 6 mth po	Resolved	California, USA
Miglets & Branson, 1983 <sup>17</sup>	37, M	Middle ear, mastoid	Mastoidectomy, penicillin 3 mth po	Resolved	Ohio, USA
Leek, 1974 <sup>18</sup>	8, F	Middle ear, mastoid	Mastoidectomy, penicillin 3 mth po	Resolved	Chicago, USA
Fletcher, 1956 <sup>19</sup>	73, M	Middle ear, mastoid, petrous apex, foramen magnum	Mastoidectomy, aureomycin 1 week, chloromycetin 10 d, gantrisin 2 wks	Resolved	California, USA
Townrow & Barrie, 1945 <sup>20</sup>	64, M	Middle ear, mastoid, sigmoid sinus, jugular bulb, subarachnoid, subdural	Mastoidectomy, sulphanilamide 2 d po	Death	Sheffield, UK
Ouidi <i>et al.</i> , 2005 <sup>21</sup>	58, M	Middle ear, mastoid	Mastoidectomy, penicillin 5 mth po	Resolved	Fes, Morocco <sup>†</sup>
Skladzien <i>et al.</i> , 2002 <sup>22</sup>	–	–	–	–	Poland <sup>†</sup>
Boor <i>et al.</i> , 1998 <sup>23</sup>	–	–	–	–	Slovakia
Lester & Juhasz, 1990 <sup>24</sup>	–	–	–	–	Ethiopia
Gattaz & Naujoks, 1984 <sup>25</sup>	–	–	–	–	Germany <sup>†</sup>
Gladkii <i>et al.</i> , 1983 <sup>26</sup>	–	–	–	–	Russia <sup>†</sup>
Kunel'skaia <i>et al.</i> , 1977 <sup>27</sup>	–	–	–	–	Russia <sup>†</sup>
Sinel'nikov & Novikova, 1970 <sup>28</sup>	–	–	–	–	Russia <sup>†</sup>
Szlezak & Sowinski, 1965 <sup>29</sup>	–	–	–	–	Poland <sup>†</sup>
Kulina, 1958 <sup>30</sup>	–	–	–	–	Russia <sup>†</sup>
Preobrazhenskii, 1957 <sup>31</sup>	–	–	–	–	Russia <sup>†</sup>

\*Cases presented in the worldwide literature since 1945. <sup>†</sup>Foreign language articles. Y = years; F = female; mth = months; po = per os (by mouth); IV = intravenous; M = male; wks = weeks; d = days

seven months after the initial examination (there was a three month delay due to inter-departmental consultant referral and a four month operation waiting list delay).

At operation, the middle ear was found to be full of granulation tissue and there were two yellow coloured 'plaques' in the middle ear. The long process of the incus was eroded, but the head of the stapes was intact. The mucosa of the mastoid air cells was oedematous, but no cholesteatoma was found. The granulation tissue and plaques were removed and sent for histology, and an ossiculoplasty (incus transposition) was performed.

Histological examination revealed a large colony of microorganisms surrounded by a film of necrotic, fibrinous debris and neutrophil polymorphs. The central portion of the colony was basophilic and eosinophilic, with club-shaped filaments radiating at the peripheries in a rosette pattern into which neutrophils were enmeshed.

Histochemical testing of the colonies revealed Gram-positive, silver avid (on Grocott staining) and diastase-resistant periodic acid-Schiff negative staining. This histochemical profile was deemed most in keeping with actinomycosis, wherein the colonies represented the characteristic 'sulphur granules'. Fungal infection was ruled out on the basis of the negative diastase-resistant periodic acid-Schiff test.

On the advice of the microbiologist, the patient was started on a six-month course of oral amoxicillin and referred for a paediatric assessment to exclude an underlying immune deficiency. This assessment did not reveal any abnormality.

Following surgery, the patient had no further ear discharge. The tympanic membrane appeared normal and her hearing was normal in both ears. She was followed up for a period of one year (with no further symptoms) before being discharged.

## Discussion

Actinomycosis is a subacute or chronic granulomatous disease, which typically presents with abscesses and tissue necrosis in the oral-cervicofacial region. The infective agent is a non-spore forming anaerobic or facultative anaerobic actinobacteria, most often of the *Actinomyces israelii* species.<sup>1</sup> This species was first described as a human pathogen by Israel in 1878.<sup>2</sup>

Actinomyces are known to be a commensal flora of the oral cavity; hence, it is postulated that entry to the middle ear is most likely the result of seeding from the nasopharynx via the eustachian tube. Direct infections of the external auditory canal or haematogenous spread are possible but less likely alternatives.<sup>3</sup> In our case, the initial presentation of an aural polyp and featureless tympanic membrane prompted a myringotomy. After the aspiration of thick, glue-like secretions, a white plaque was found, which filled the middle ear. This was initially suspected to be a cholesteatoma; however, in retrospect this was almost certainly a primary plaque of actinomyces. We are unable to speculate further on the mechanism of primary actinomyces colonisation of the middle ear in our case. There is, however, a possibility that actinomyces could have been introduced into the middle ear at the time of the initial myringotomy, which would therefore constitute a secondary superadded infection of a middle-ear plaque of uncertain aetiology.

Identification of actinomyces bacteria is difficult due to their recalcitrance to culture and their exquisite sensitivity to oxygen. However, the identification of Gram-positive filamentous organisms with sulphur granules is strongly

suggestive of actinomycosis.<sup>4</sup> The differential diagnosis for culture-insensitive chronic otitis media includes nocardiosis and tuberculosis. Newer diagnostic techniques such as polymerase chain reaction, 16S rRNA sequencing and mass spectrometry hold promise, but are not routinely used in clinical practice.<sup>5,6</sup>

Our review of the worldwide literature revealed 26 cases of actinomycosis of the middle ear and mastoid since 1945. These are distributed broadly across the world, with a clustering in the USA (see Table 1).<sup>7–31</sup> We speculate that this clustering is an artificial effect due a higher publication rate from this region. We present the first case in the UK in over 60 years.

- **Actinomycosis is a bacterial disease caused by actinomyces species**
- **Middle-ear and mastoid actinomycosis is rare, presenting as recurrent otorrhoea with granulation tissue**
- **This paper presents the first case of middle-ear actinomycosis in the UK for 60 years**
- **Actinomyces form basophilic and eosinophilic colonies with club-shaped filaments radiating into rosette patterns ('sulphur granules')**
- **Histochemical profiling of colonies revealed silver avid, diastase-resistant periodic acid-Schiff negative staining**
- **Our recommended treatment is full surgical clearance of diseased tissue with six months amoxicillin**

In the last documented case from the UK, Townrow and Barrie (1945) reported a case of a 64-year-old horseman with actinomycosis of the middle ear.<sup>20</sup> The patient had presented with a 57-year history of ear discharge, which had worsened over the previous 8 days. He underwent mastoid exploration, but suffered a large venous bleed. He developed meningitis which was unsuccessfully treated with a sulphoamide antibiotic, ultimately resulting in his death. The authors reviewed the literature up to 1945 and noted that 16 out of 17 reported cases were fatal, with meningitis being the leading cause of death. Diagnosis in the pre-antibiotic age was based on post-mortem assessment. Following the introduction of penicillin, prognosis radically improved: the condition, previously associated with a near 100 per cent mortality rate, became a non-fatal and eminently treatable pathology.

It is our recommendation that complete surgical removal and histological examination of all infected tissue should be undertaken in similar cases. If actinomyces are found, we recommend six months oral amoxicillin as the first-line treatment. Other antibiotics including erythromycin, chloramphenicol and tetracycline have also been used successfully.<sup>1</sup>

## References

- 1 Wong VK, Turmezei TD, Weston VC. Actinomycosis. *BMJ* 2011;**343**:d6099
- 2 Schultheiss D. James Israel (1848–1926). Discoverer of actinomycosis and pioneer of kidney surgery [in German]. *Aktuelle Urol* 2008;**39**:105–8
- 3 Ajal M, Turner J, Fagan P, Walker P. Actinomycosis oto-mastoiditis. *J Laryngol Otol* 1997;**111**:1069–71

- 4 Smego RA Jr, Foglia G. Actinomycosis. *Clin Infect Dis* 1998;**26**:1255–61
- 5 Patel JB. 16S rRNA gene sequencing for bacterial pathogen identification in the clinical laboratory. *Mol Diag* 2001;**6**: 313–21
- 6 Welker M. Proteomics for routine identification of microorganisms. *Proteomics* 2011;**11**:3143–53
- 7 Gazzano E, Chanteret C, Duvillard C, Folia M, Romanet P. A case of actinomycosis of the middle ear and a review of the literature. *Int J Pediatr Otorhinolaryngol Extra* 2010;**5**:70–3
- 8 Budenz CL, Tajudeen BA, Roehm PC. Actinomycosis of the temporal bone and brain: case report and review of the literature. *Ann Otol Rhinol Laryngol* 2010;**119**:313–18
- 9 Shishegar M, Hosseini SH, Varedi P, Mahmoodi S, Ashraf MJ, Faramarzi A. Actinomycosis of the middle ear. *Otol Neurotol* 2009;**30**:686–7
- 10 Mehta D, Statham M, Choo D. Actinomycosis of the temporal bone with labyrinthine and facial nerve involvement. *Laryngoscope* 2007;**117**:1999–2001
- 11 Sivarajasingam M, Rajan P. Temporal bone actinomycosis: a histopathologic report. *Otol Neurotol* 2007;**28**:869–70
- 12 Sobol SE, Samadi DS, Wetmore RF. Actinomycosis of the temporal bone: a report of a case. *Ear Nose Throat J* 2004;**83**:327–9
- 13 Hoshino T, Amano H, Tanaka K. Actinomycosis of the middle ear and mastoid. *Eur Arch Otorhinolaryngol* 1996;**253**:378–80
- 14 Tarabichi M, Schloss M. Actinomycosis otomastoiditis. *Arch Otolaryngol Head Neck Surg* 1993;**119**:561–2
- 15 Olson TS, Seid AB, Pransky SM. Actinomycosis of the middle ear. *Int J Pediatr Otorhinolaryngol* 1989;**17**:51–5
- 16 Shelton C, Brackmann DE. Actinomycosis otitis media. *Arch Otolaryngol Head Neck Surg* 1988;**114**:88–9
- 17 Miglets AW, Branson D. *Arachnia propionica* (Actinomyces propionicus) as an unusual agent in tympanomastoiditis. *Arch Otolaryngol* 1983;**109**:410–12
- 18 Leek JH. Actinomycosis of the tympanomastoid. *Laryngoscope* 1974;**84**:290–301
- 19 Fletcher R. A rare case of chronic otorrhea with intracranial complications. *Laryngoscope* 1956;**66**:702–10
- 20 Townrow V, Barrie HJ. Fatal Actinomyces infection of the middle ear. *J Laryngol Otol* 1945;**60**:329
- 21 Oudidi A, Hachimi H, Ridal M, El Alami MN. Actinomycosis of the middle ear: case report [in French]. *Rev Laryngol Otol Rhinol (Bord)* 2005;**126**:29–32
- 22 Skladzien J, Trabka-Zawicki P, Wierzchowski W, Maciejak K. Actinomycosis of the middle ear [in Polish]. *Otolaryngol Pol* 2002;**56**:369–71
- 23 Boor A, Jurkovic I, Friedmann I, Benicky M, Dudrikova K, Krajcar R. Actinomycosis of the middle ear. *J Laryngol Otol* 1998;**112**:800–1
- 24 Lester FT, Juhasz E. Actinomycosis of the ear. *Ethiop Med J* 1990;**28**:41–4
- 25 Gattaz G, Naujoks JH. Actinomycosis of the middle ear [in German]. *HNO* 1984;**32**:65–8
- 26 Gladkii NI, Lisovets VT, Shevchenko AM. Actinomycosis of the ear [in Russian]. *Vestn Otorinolaringol* 1983:76–7
- 27 Kunel'skaia V, Chkannikov AN, Kovalenko EE. Actinomycosis of the ear [in Russian]. *Vestn Otorinolaringol* 1977:78–9
- 28 Sinel'nikov MM, Novikova RK. Case of actinomycosis of the middle ear [in Russian]. *Zh Ushn Nos Gorl Bolezn* 1970;**30**: 109–10
- 29 Szlezak L, Sowinski H. Primary actinomycosis of the middle ear [in Polish]. *Otolaryngol Pol* 1965;**19**:191–5
- 30 Kulina TV. Actinomycosis of the middle ear and mastoid process [in Russian]. *Vestn Otorinolaringol* 1958;**20**:94
- 31 Preobrazhenskii NA. Actinomycosis of the ear [in Russian]. *Vestn Otorinolaringol* 1957;**19**:72–8

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Mr P Kullar takes responsibility for the integrity of the content of the paper

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