CASE REPORT
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# Middle ear Actinomycosis: a rare cause of acute otitis media with labyrinthine fistula

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**ABSTRACT**

Actinomycosis is a very rare pathology of the middle ear and mastoid, with less than 50 cases reported worldwide. This paper reports the case of an 11-year-old girl presenting with acute otitis media and acute vertigo owing to labyrinthine fistula. Surgical tympanomastoid exploration yields granulomatous disease of middle ear and mastoid, and histopathology shows actinomycosis. The authors report this case, which was treated by surgical debulking of the granulomatous tissue and closure of the fistula, followed by 2 weeks of intravenous treatment with penicillin and at least 3 months of oral amoxicillin. The authors discuss the challenges in managing this case in view of recent literature. Our case confirms that diagnosis of this very rare condition is only made through histopathological diagnosis after surgical exploration and requires long-term medical treatment thereafter.

**Keywords:** Actinomyces, actinomycosis, mastoid, middle ear, otitis media

## Introduction

Actinomyces spp., gram-positive filamentous bacteria, are commensal bacteria found in the oropharyngeal, intestinal, and genitourinary regions. These rarely act as opportunistic pathogens, causing saprophytic infections called actinomycosis. The most frequent (60%) location of actinomycosis is the orocervicofacial region, where it is sometimes seen after minor trauma. It usually presents as a slowly progressive granulomatous infection in anaerobic conditions. Actinomycosis of the tympanomastoid region is very rare, with less than 50 cases reported worldwide. We report the first Belgian case of middle-ear actinomycosis in an 11-year-old girl, presenting as acute otitis media with labyrinthine fistula.

## Case Presentation

An 11-year-old girl without otologic history was sent from the outpatient pediatrics department, with a sudden onset of a left-sided acute otitis media and spontaneous grade 3 horizontal nystagmus with fast phase toward the right. Microscopic examination of the ear showed an intact red eardrum and a nonaerated middle ear. No edema or erythema of the mastoid region was observed. Pure-tone audiometry showed an ipsilateral conductive hearing loss with a pure-tone average (PTA) of 60dB HL. Video head impulse test showed left-sided reduced gain (0.44) of the vestibulo-ocular reflex of the horizontal semicircular canal with many overt saccades and a normal gain on the right side (0,8). Tentative diagnosis of a bacterial labyrinthitis was made. Serum levels of immunoglobulin G, immunoglobulin A, and immunoglobulin M were normal. Myringotomy with ventilation tube insertion was performed on the same day, and a treatment with intravenous (IV) amoxicillin clavulanate and IV methylprednisolone was started. Subsequent temporal bone computed tomography (CT) showed complete opacification of the middle ear with bony erosion of the ossicles and the facial nerve and a fistula of the horizontal semicircular canal (Figures 1 and 2). No bony erosion of the skull base was apparent on CT. Temporal bone magnetic resonance imaging (MRI) showed signs of inflammatory debris in the middle ear and mastoid and slight diffusion restriction in the middle ear, but not the typical image of a congenital cholesteatoma (Figures 3-5). There were no signs of sigmoid sinus thrombosis on MRI. Differential diagnosis at this preoperative stage, with an opacified middle ear and bony erosion, together with only atypical findings on MRI, was relatively broad and contained some rare causes (Table 1). Because there was continuing ear discharge after 1 week despite systemic and local antibiotics, in the absence of any bacteriologic identification and with the aforementioned ra-

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diological findings, the decision for an exploratory tympanomastoidectomy was made. This surgery showed granulomatous tissue filling the middle ear and the attic (Figure 6). The horizontal semicircular canal fistula was found and covered immediately, first with temporalis fascia and then followed by bone paté. Granulation tissue was removed where possible but was left partly in place at the level of the facial nerve (because peroperatively the diagnosis remained unclear but was obviously not a cholesteatoma). An allograft incus ossiculoplasty was performed. The ear drum including the ventilation tube was left untouched. Microscopic evaluation showed numerous neutrophil granulocytes intimately intermingled

**Table 1. Differential diagnosis of nontraumatic middle-ear opacification with bony erosion of the labyrinth**

| Chronic otitis media | Cholesteatoma (acquired or congenital) |
|---|---|
| Uncommon Infections | Cholesterol granuloma |
| | Tuberculous otitis media |
| | Actinomycosis |
| | Temporal bone osteomyelitis |
| | Invasive fungal otomastoiditis |
| Benign tumors | Facial nerve schwannomas |
| | Glomus tympanicum |
| Malignant tumors | Squamous cell carcinoma |
| | Adenoid cystic carcinoma |
| | Adenocarcinoma |
| | Rhabdomyosarcoma, chondrosarcoma |
| | Melanoma |
| | Lymphoma |
| | Langerhans cell histiocytosis and other histiocytoses |
| Autoimmune disorders | Wegener's granulomatosis |
| | Granulomatosis with polyangiitis (GPA) |
| | Sarcoidosis |
| | Eosinophilic GPA |

Note: Table 1 shows a nonexhaustive list of causes of a middle-ear opacification in a nontraumatic patient. The list includes some rare pathologies potentially causing bony erosion of the labyrinth, as in our described case

**Main Points:**

- Actinomyces are commensal bacteria that can rarely cause pathologic infections.
- In the middle ear, actinomycosis is a very rare cause of acute otitis media, possibly leading to bony erosion of ossicles and labyrinth.
- Histopathology after surgical exploration yields the diagnosis.
- Postoperative treatment consists of several months of antibiotics, usually penicillin.

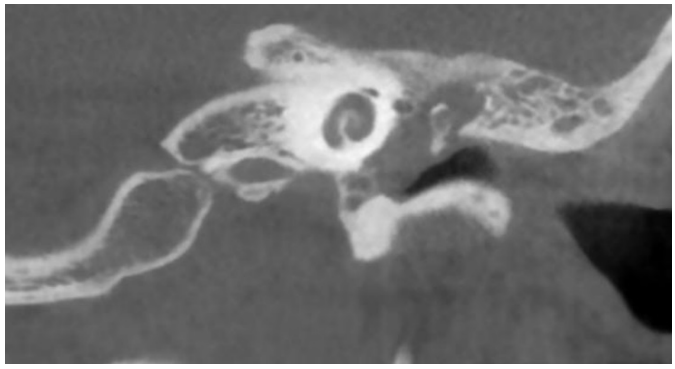

**Figure 1.** Coronal reformatted CT shows partial osteolytic destruction of the left ossicular chain and soft tissue infiltration of the tympanic cavity, antrum, and epitympanic recess

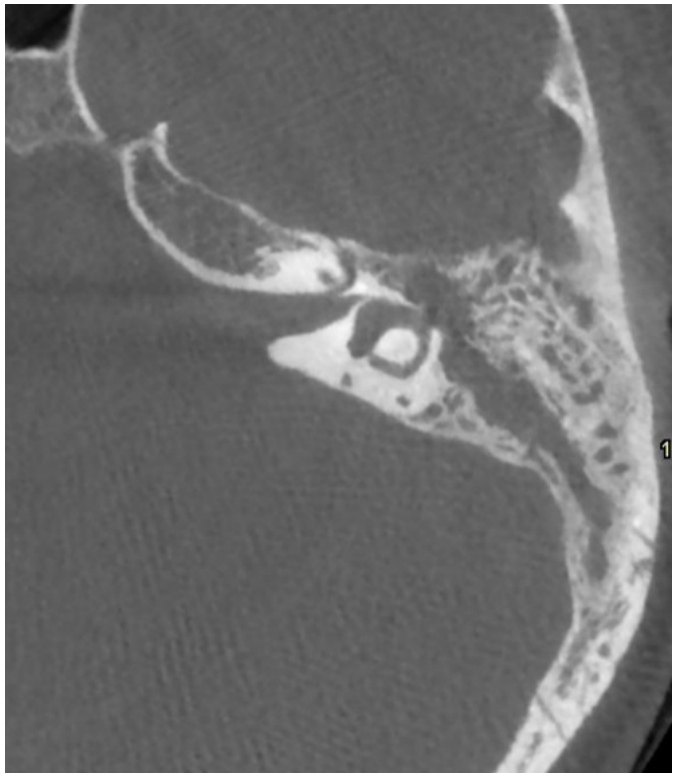

**Figure 2.** Axial reformatted computed tomography shows partial osteolytic destruction of the left lateral semicircular canal and soft tissue obliteration of the otomastoidal air spaces

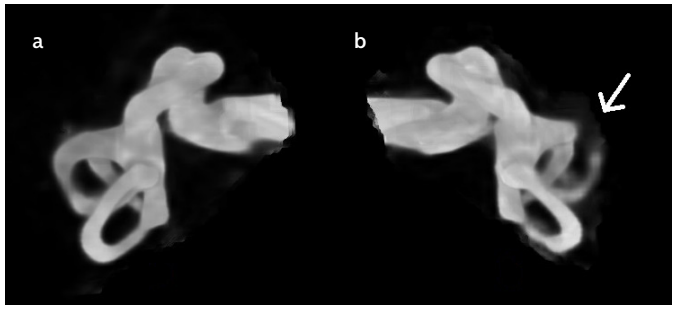


**Figure 3. a, b.** Three-dimensional Sampling Perfection with Application optimized Contrasts using different flip angle Evolution (SPACE) reconstructed magnetic resonance image shows a comparison of right (a) and left (b) sided inner ear structures. The arrow indicates the signal loss and defect in the left lateral semicircular canal, corresponding to the labyrinthine fistula found in our patient

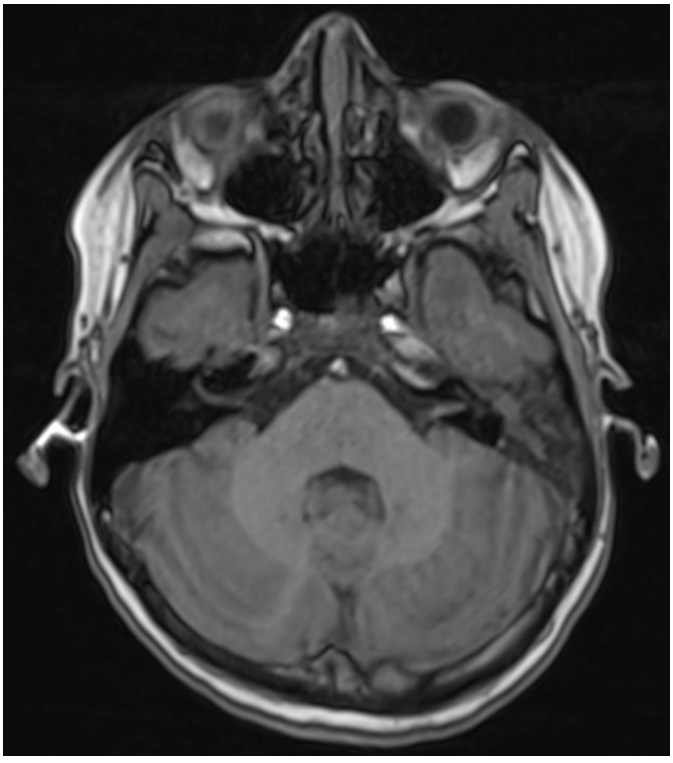

Figure 4. T1-weighted image demonstrates soft tissue signal intensity owing to filling of the air cells of the left mastoid and tympanic cavity with inflammatory debris

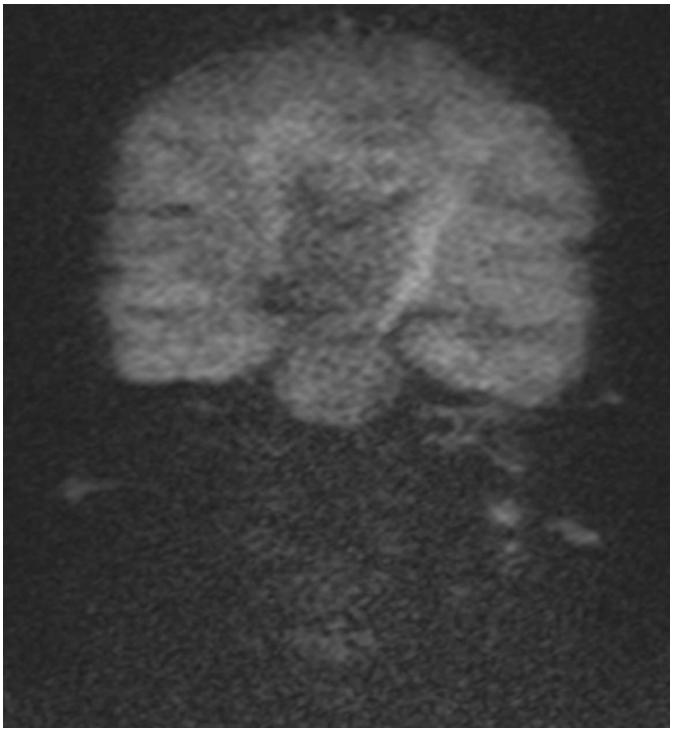

Figure 5. Coronal turbo spin echo diffusion-weighted image shows some slight hyperintensity in left otomastoidal region, albeit not as hyperintense as the "light bulb sign," as would be seen in case of a cholesteatoma

with basophilic colonies (Figure 7). These colonies reveal club-shaped filaments arranged into a radiating rosette pattern, compatible with the diagnosis of actinomycosis. There were no signs of malignancy.

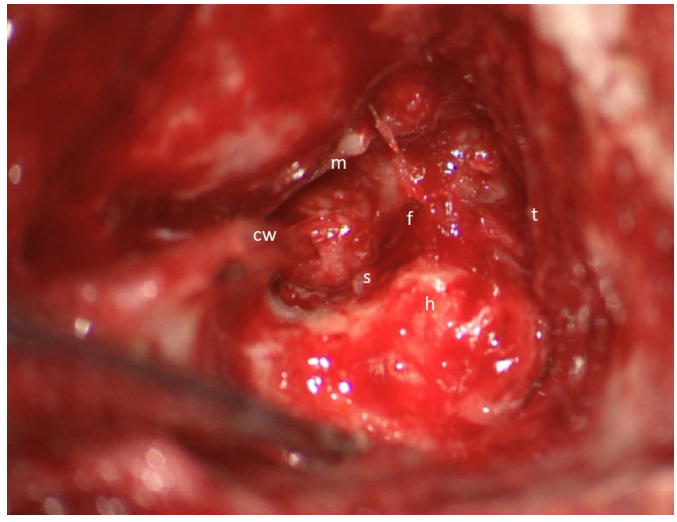


Figure 6. A peroperative situation with temporary removal of the canal wall (cw) (which was put back in place with hydroxyapatite cement at the end of the surgery), removal of the incus, and with granulomatous tissue covering all the medial wall of the middle ear, horizontal semicircular canal (h), and facial nerve (f). Malleus (m), stapes (s), and tegmen tympani (t) are also shown

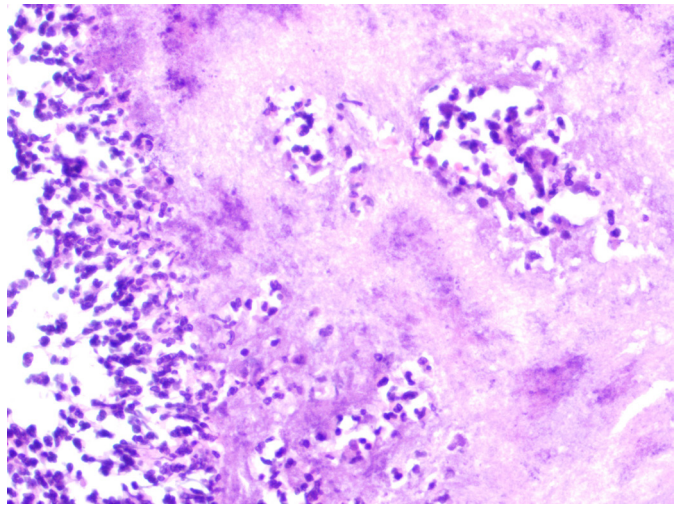

Figure 7. The edge of an Actinomyces colony surrounded by neutrophil granulocytes (hematoxylin-eosin stain; ×400 magnification)

Postoperatively, the patient recovered well, and the ear remained dry with the ventilation tube still in place. Ipsilateral hearing improved to almost normal hearing thresholds of PTA of 22dB HL. After confirmation of the diagnosis, patient received an IV treatment with penicillin $6 \times 10^6$ units per day, followed by amoxicillin 100 mg/kg/day for 3 months. Currently, she is without ear complaints and on follow-up. The patient and her parents gave their informed consent for publication of this case report.

## Discussion

We present the first case of middle-ear actinomycosis in Belgium. For the management of this pathology, we had to rely on the recently published literature (1-7). There are currently approximately 50 cases reported worldwide, at least 9 of whom are children. The work of Kullar et al. (5) was particularly helpful because it offers an overview of all known cases up to

2013. Actinomyces spp. are commensal flora of the oral cavity, and studies have shown that up to half of the children have Actinomyces colonies in the tonsils. The presence of Actinomyces is not correlated to having recurrent tonsillitis episodes but is slightly more common in children older than 5 years than younger children (8-10). 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 (where it is also commensal) or hematogenous spread are less likely. Cases usually present as chronic suppurative otitis media, contrary to our case that was an acute onset with labyrinthitis. Mehta et al. (7) reported on a similar case of acute labyrinthitis and facial palsy in a 10-year-old patient with primary ciliary dyskinesia (PCD). Most probably, PCD acted as a predisposing condition by enhancing spread of Actinomyces from oronasal cavity to the middle ear. Our patient did not have any ear, nose, and throat history or predisposing factors whatsoever.

Management of the cases in literature is predominantly surgical by an intact canal wall tympanomastoidectomy, essential for histopathologic diagnosis and debulking, followed by prolonged antibiotic treatment. Microbiologic confirmation seems to be a challenge, because cultures are negative in up to 70% of infections and differentiation between *Actinomyces meyeri*, *Actinomyces turicensis,* or *Actinomyces israelii* is, thus, not possible. At the time of surgery, we found a small white plaque in the middle ear and sent it for microbiological examination. Direct microscopic examination revealed typical branched network of filaments covering the surface, correlating to Actinomyces; however, prolonged cultures remained negative. However, the identification to the species level is of little relevance because all Actinomyces spp. are reported to be sensitive to penicillin.

Treatment duration varies from 6 weeks to 6 months in the reported cases. Another challenge is to evaluate the therapeutic effect that might help to guide toward longer or shorter therapy duration. Follow-up imaging with CT and MRI can rule out progressive bony erosion and indicate amount of residual granulation tissue and aeration of the middle ear. Topical therapy has unfortunately no place in the treatment and is not mentioned in literature because Actinomyces are resistant to ciprofloxacin, and other topical antibiotics pose the risk of ototoxicity especially when used for such long durations. There is no mention of a second-look surgery, which is somewhat surprising because it would provide direct visual evaluation and control of the pathology. In addition, the clinical and/or tympanometric evaluation of middle ear aeration is useful because Actinomyces is an anaerobic organism. Placement, maintenance, and, if necessary, replacement of a functional ventilation tube will be part of the follow-up in our patient for at least 1 year.

Our case confirms that the diagnosis of this very rare condition is usually made after surgical exploration and histopathological diagnosis. The long-term medical treatment after surgery is based on literature and the experience of more common Actinomyces-related pathologies outside the middle ear, with the known susceptibility to penicillin.

**Informed Consent:** Verbal informed consent was obtained from the patients who agreed to take part in the study.

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**Conflict of Interest:** The authors have no conflict of interest to declare.

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