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Review

Hearing loss due to urate deposition in the middle ear: A case report and literature review

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ABSTRACT

Gout is the most common cause of monoarthritis in men occurring classically in the great toe and the knee. Extra-articular gout manifestations are rare. Only a few cases of head and neck urate crystals deposits have been described in the literature. Precipitations in the middle ear cause conductive hearing loss with common otoscopic anomalies and difficult imaging diagnosis.

We report a case of a healthy 58-years-old man with a middle ear urate deposit causing a progressive hearing loss as the very first symptom of gout. The nature of the deposit was unsure on computer tomography (CT) due to atypical density. The final diagnosis was revealed after surgical procedure and histologic examination.

A review of the literature is also presented. Seven cases of middle ear urate deposit as the first symptom of gout were found and compared.

Progressive conductive hearing loss in middle-aged patients with abnormal otoscopy and middle ear atypical density mass on CT scan must lead to a minimal surgical procedure with a histologic examination to exclude urate crystals deposits.

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1. Introduction

Gout is a common metabolic disease affecting 1–2% of adults in developed nations, mostly middle-aged men and postmenopausal women (Gaffo, 2019). This inflammatory disease is characterized by monosodium urate crystal deposit in joints and connective tissues due to hyperuricemia. Urate tends to precipitate when plasma

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concentration exceeds 6.8 mg/dL (Braun et al., 2016). Precipitation usually occurs in extremities due to anatomic and environmental factors such as lower body temperature, poorly perfused distal tissues, repeated joint microtrauma, or history of surgery. The most frequent localizations are the first metatarsophalangeal joint and the knee with well-known clinical signs of painful acute monoarthritis. Precipitations occur in joints but serum diffusion in tissues is also enough to create deposits. Other medical risk factors include hypertension, renal insufficiency, diuretic use, alcohol consumption, a purine-rich diet, immunodepression, and obesity (Chabra and Singh, 2013).

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Fig. 1. Left ear otoscopy.

Head and neck localization are rare. Only about ten cases have been described in the literature such as helix subcutaneous nodules, larynx, nasal septum, and middle ear deposit.

We report a case of middle ear urate deposit causing a progressive hearing loss as the first symptom of gout and a literature review.

2. Case presentation

A 58-year-old man presented with a two-year progressive leftsided hearing loss. In his medical history, he underwent at 20years-old a left tympanoplasty with complete ossicular allograft for chronic otitis. He did not consult for hearing problems since then. In his relevant medical history, he was treated for 2 kidney stones and had cardiac auricular fibrillation under medical treatment He had no hypertension or renal insufficiency, and he had a healthy weight and no history of alcohol abuse.

Otoscopy revealed a left cicatricial tympanic membrane with a white granular deposit (Fig. 1). Pure tone audiometry illustrated a left mixed hearing loss with a conductive component around 30 dB (Fig. 2). His temporal bone computed tomography (CT) showed a mineral mass, surrounding the incus (Figs. 3 and 4). The stapes was normal, but the mass was in close contact with the facial nerve. The average density mass was around 852 Hounsfield Units (HU)

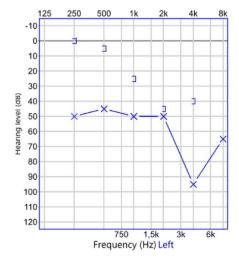


Fig. 3. Siemens SOMATOM Definition AS CT scanner, 0.4 mm non contrast enhanced spiral acquisition with 0,5 mm multiplanar post processing (Siemens Syngovia software). Transversal section of the left middle ear showing urate deposit adherent to the malleus and uncus.

corresponding to the range between ossification and cholesteatoma. (Fig. 4). The heterogeneous process was a challenging radiologic diagnosis and differential diagnosis would be atypical cholesteatoma or chondroma. In this context, a surgical excision was proposed.

He underwent a left middle ear exploration. During the surgery, a white wet sugar-like material was arising from the incus and fulfilled the attic region (Fig. 5). The material could easily be removed from the ossicular chain with micro curette, ear hook and forceps. The ossicular allograft was completely functional after removal so no other procedure was performed on the chain. The facial nerve canal was not dehiscent (Fig. 6).





Post-operative left side

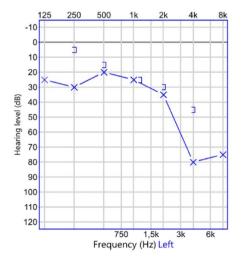


Fig. 2. Pre and post -operative pure tone audiometry.

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Fig. 4. Siemens SOMATOM Definition AS CT scanner, 0.4 mm non contrast enhanced spiral acquisition with 0,5 mm multiplanar post processing (Siemens Syngovia software). Coronal section of the left temporal bone: hyperdense urate deposit = 852.7 Hounsfield units.



Fig. 5. 0° endoscope intra-operative view. Isj, incudo-stapedial joint; pe, pyramidal eminence; ct, chorda tympani.

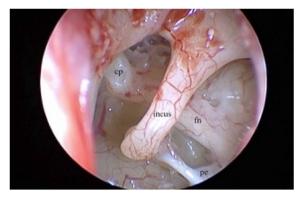


Fig. 6. 30° endoscopic intra-operative view after removal of material. Complete ossicular chain homograft remains intact. cp, cochleariform process; fn, facial nerve; pe, pyramidal eminence.

Histologic examination with polarized microscopy revealed an amorphous negative birefringent material compatible with urate crystals deposit, such as found typically in gout tophi.

Rheumatologist advice was taken, and the patient had a biology and urine test, revealing a hyperuricemia (8,6 mg/dL serum uric acid level) but no uricosuria. The patient didn't have other clinical

signs of gout. Because of hyperuricemia, he started a progressive urate-lowering therapy (allopurinol 50 mg/day) to prevent new tophi formation. One year after surgery, his conductive hearing loss was recovered and has no consequence of the surgery and no sign of recurrence (Fig. 2).

Consent of the patient was obtained to publish his atypic clinical case.

3. Discussion

Gout is the most common cause of monoarthritis in middle-aged men. Only a few cases of head and neck localization have been described in the literature. Ears are the most cited site, and it usually appears as a hard-yellowish subcutaneous nodule on the helical rim. Other localizations are the larynx (hyoid bone, cartilages, false vocal cords.), dorsal nasal septum, temporomandibular joint, and middle ear (Saliba et al., 2019).

You can find a resume of the middle ear cases found in the literature in Table 1. None of the patients had a history of gout or previous ear surgery. The sex ratio was 1/1 and the pathology is more frequent for patients over 55-years-old. The major clinical symptom was a simple progressive hearing loss (Gargula et al., 2019). Abnormal otoscopy occurred in 7 out of 8 patients (Mutlu et al., 2016) with 50% of white sclerotic plaque under the normal tympanic membrane.

CT scan is the recommended examination and show abnormal heterogeneous mass in the middle ear (Reineke et al., 2009). Even if urate deposits have a theoretical different mineral density in conventional CT (750–900 HU) in our case vs > 1000 HU for cortical bone structure and <100 for cholesteatoma (Kim, 2014)), middle ear gout was never diagnosed in pre-operative images. The lack of a previous history of gout and the atypical presentation make the diagnostic nearly unthinkable for the radiologist as well as for the otologist. The heterogenicity of the mass can be explained by punctiform calcification (Saliba et al., 2019).

Surgical procedures were performed in each case, finding the wet sugar-like or semolina-like white mass (Tausch-Treml and Berghaus, 1990). Negatively birefringent monosodium urate crystals within phagocytes examined under compensated polarized light microscopy confirmed the histologic diagnostic. Surgical procedures allowed a hearing recovery with a gap closure if the ossicular chain remains intact.

Differential diagnosis is cholesteatoma, otitis media, osteoma, or chondroma. Clinical symptoms such as otorrhea or pain are usually more frequent in otitis media and cholesteatoma.

When the diagnosis of gout is confirmed, we recommend a rheumatological examination with a blood test (Towiwat et al., 2019). In case of hyperuricemia, an uricemia-lowering therapy can be started. Without treatment, gout can become a chronic condition and increase the risk of developing other tophi and joints damages. There are four treatments used in gout disease (CBIP, 2021): nonsteroidal anti-inflammatory drugs, corticosteroid, Xanthine oxidase inhibitors (XOIs) known as allopurinol and uricosuric. Because middle ear tophi do not generate pain or inflammation, only XOI's should be used as initial therapy in case of hyperuricemia.

Theoretically, the clinical and radiological diagnosis could avoid surgery procedures in some small cases. Urate lowering therapy allows a resolving of small gout tophi in the helix (Chabra and Singh, 2013) but it may take several months to observe a size reduction. There is no information available regarding the middle ear medical treatment, the recurrence rate, or the dietary recommendations. We do not have any data on possible hearing recovery after treatment by XOIs, and it would be interesting to carry out further studies in this respect.

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Table 1Comparative review of middle ear urate deposition cases found in the litterature.

	Age	Sex	Symptoms	Otoscopy	Hyperuricemia	History of gout	Non contrast CT of the temporal bone	Per op aspect	URATE LOWERING THERAPY
Tausch- Treml et al.	68	F	Pain and pruritus	"Red rough polyp in the external canal".	No	No	"Filling of the epitympanum"	"A whitish, solid and partially crystalline material"	Not specified
Gargula et al.	64	F	Conductive hearing loss	"White calcified mass under the tympanic membrane"	Not specified	No	"Non-erosive nodular calcified mass"	"White calcified but friable, semolina-like, mass"	Not specified
Mutlu et al.	34	M	Otorrhea Mixed hearing loss	"Subtotal perforation"	Yes	No	"Increase of soft tissue in the middle ear"	"White tympanosclerotic plaques"	Yes
Reineke et al.	66	F	Mixed hearing loss	"White sclerotic plaque under the tympanic membrane	No	No	"Partly opacified middle ear"	"Semolina pudding-like middle ear mass"	Not specified
Saliba et al.	83	F	Mixed hearing loss	"White mass in the anterior middle ear"	No	No	"Heteroge neously hyperdense lesion"	«Granular white mass»	Yes
	67	M	Conductive hearing loss	"White-colored polypoid plaque"	Not obtained	No	"Heterogeneously hyperdense lesion»	"The white mass was of chalky consistency"	Referred to primary care provider
Braun et al.	63	M	Mixed hearing loss	normal	No	No	"Irregular bone structure"	"Crumbly whitish mass of chalky consistency"	No
Current case	57	M	Mixed hearing loss	Cicatricial tympanic membrane	Yes	No	"Irregular mineral mass"	"White wet sugar-like material"	Yes

In conclusion, the present case is a rare clinical manifestation of gout disease. Tophi gout deposit should be considered in conductive hearing loss with abnormal bone density mass on CT scan, especially if heterogeneous mass. At this point, only histological diagnosis makes it decisive. If feasible, minimal surgical procedures should be performed without modification of the ossicular chain.

Conflicts of interest and source of funding

No conflicts of interest.

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