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# Late Sudden Deafness in Unoperated Ear of Patients with Otospongiosis: Case Studies

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***Authors' contributions***

*All authors contributed directly to the development of this article. All authors read and approved the final manuscript.*

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***Case Study***

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## ABSTRACT

**Background:** It is not known whether there is etiological relationship between otospongiosis and sudden deafness. The most common etiologies that explain sudden deafness are viral infections, vascular and autoimmune process.
**Methods:** Two cases of patients with otospongiosis that have developed sudden deafness are presented.
**Results-Case report:** We report two cases of late sudden deafness in the unoperated ear of patients with otospongiosis. Both patients underwent previous surgical treatment in the contralateral ear with a a short period of hearing improvement and poor hearing outcome some days after. In both reported cases the patients with bilateral otospongiosis had sudden deafness affecting both ears. The first episodes had the onsets postoperatively at the operated ears. The second episodes were at the contralateral ears in the late follow-up, especially considering that in both cases the patients showed satisfactory results regarding hearing improvement postoperatively although temporarily we therefore question the existence of the association between otospongiosis and sudden deafness.

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**Discussion:** Sudden deafness in patients with otospongiosis is a rare occurrence and because of the few cases reported, it is not possible to establish any relationship. Maybe there is a possible immunological cause for the association of otosclerosis and sudden deafness.
**Conclusion:** The association between sudden deafness and otosclerosis can be considered.

*Keywords: Otospongiosis; otosclerosis; stapedotomy; stapes surgery; sudden hearing loss.*

## 1. INTRODUCTION

Sudden deafness is hearing loss of at least 30dB in at least three consecutive frequencies in tonal audiometry pattern. This loss settles abruptly within three days or less [1]. It is estimated the occurrence between five and 30 cases in 100.000 people per year [2-4].

Several etiologies have been considered to explain its etiology. The most frequent have been viral infections, vascular changes and autoimmune processes [5]. The etiology is not identified despite adequate investigation in approximately 88% of cases [6].

Otospongiosis is a change in bone remodeling of the otic capsule resulting in progressive conductive or sensorineural hearing loss due to stapes footplate fixation at oval window and bone resorption of the cochlea [7]. The stapedotomy is the main surgical treatment for otospongiosis aiming to restore the vibration of the fluid in the cochlea [8].

There are not many reports of patients with otospongiosis that developed sudden deafness. However, considering that otospongiosis is also a relatively rare disease, we believe that the incidence of sudden deafness may be higher in patients with otospongiosis comparing to the general incidence.

We describe two cases of late sudden deafness in the unoperated ear of patients with otospongiosis. We suggest that a possible association between these diseases may exists.

## 2. METHODS

This is a retrospective descriptive study of two cases of patients with bilateral otospongiosis that developed sudden deafness.

All authors hereby declare that all experiments have been examined and approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

## 3. CASE REPORTS

### 3.1 Case 1

Female, 56 year old, white, with bilateral otospongiosis diagnosed in 1998. She had undergone left stapedotomy with use of Teflon prosthesis without postoperative complications. The preoperative audiometry is illustrated in Fig. 1. On the eighth postoperative day (PO) she presented intense vertiginous crisis with vomiting, without change in hearing thresholds. She referred improvement of hearing in the operated ear. Rinne's Test was positive bilaterally and Weber's Test showed lateralization to the right ear with the use of the tuning forks of frequency 240 Hz and 520 Hz.

She had improvement of vertigo after the use of Dimenhydrinate and favorable resolution with oral Prednisone. Audiometric testing performed on the 14$^{th}$ postoperative day showed satisfactory auditory gain (Fig. 1).

On the 42$^{th}$ postoperative day, she came to medical consultation reporting sudden hearing loss and severe tinnitus in the operated ear. Audiometry then demonstrated left deafness (Fig. 2) irreversible, despite therapy with Prednisone. Were requested serology, rheumatologic tests and autoimmune and RM cerebellopontine angle, with no positive result.

Eleven and half years after the sudden deafness in left ear, the patient complained of worsening of hearing at right ear. Fig. 2 illustrates the audiometry made at this medical visit and demonstrated worsening in audiometric thresholds in right ear. She underwent on oral treatment with Prednisone, recovering the hearing levels as observed in Fig. 3.

The hearing is stable so far, five months later.

### 3.2 Case 2

Female, 56 year old, white, with bilateral otospongiosis diagnosed in 2004. She underwent left stapedotomy with prosthesis of Fluoroplastic without postoperative complications.

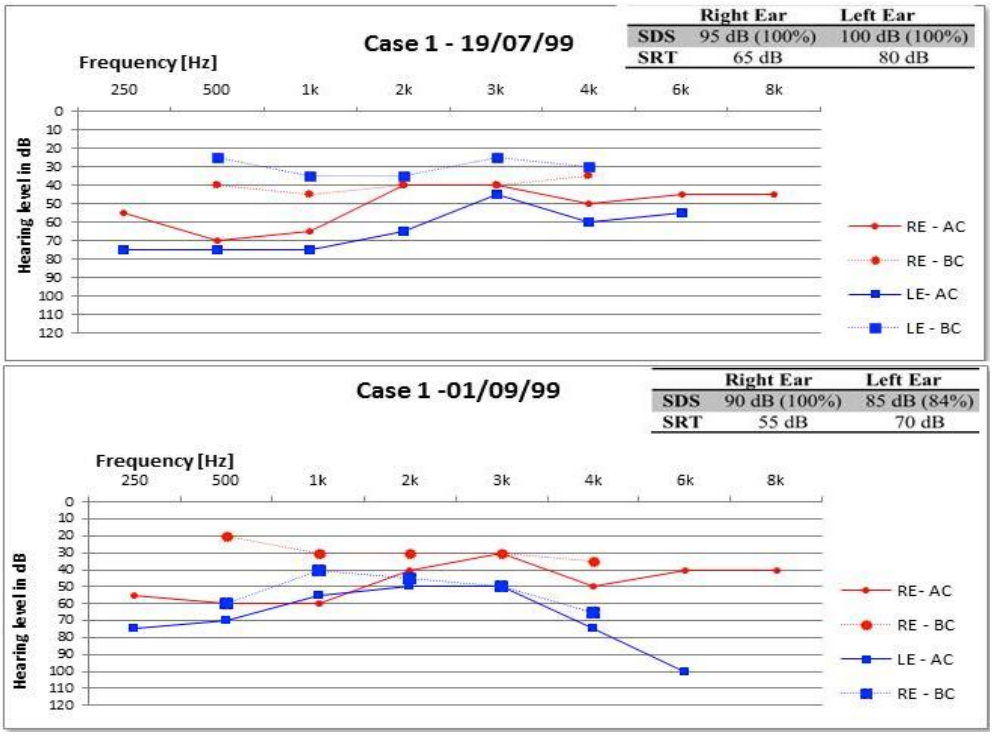


**Fig. 1. Preoperative audiometry (19.07.1999) and 14th postoperative day of left Stapedotomy audiometry with reduction of the gap and improving of hearing pattern at the left ear**

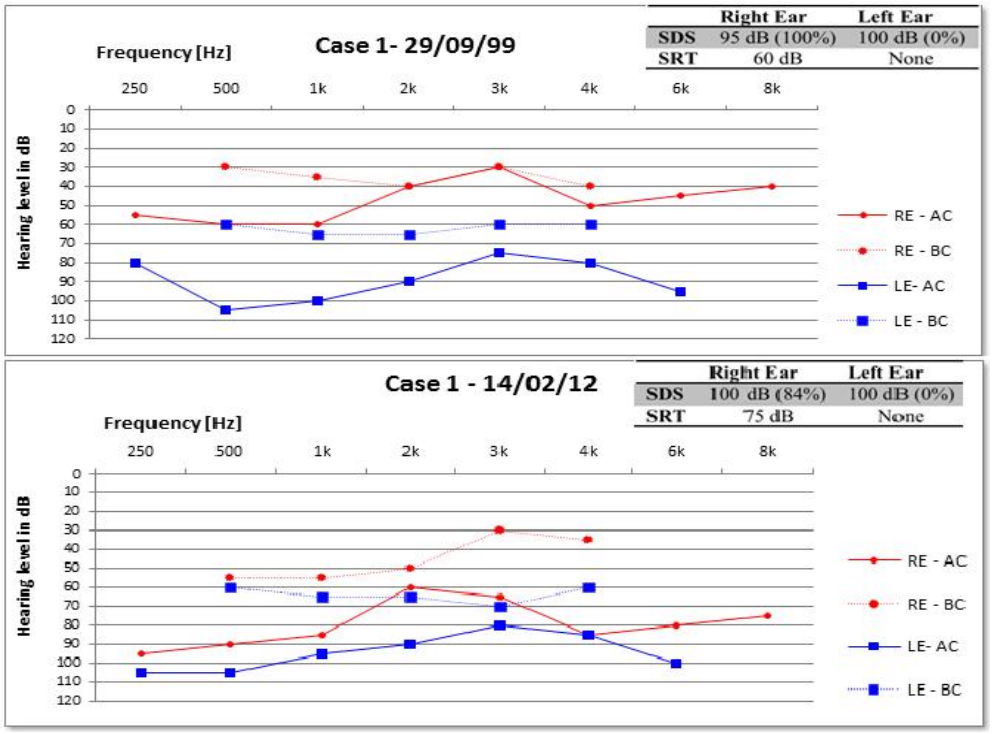


**Fig. 2. Audiometry 42 days after surgery, when the patient came to the clinic complaining of hearing loss and tinnitus at the left ear and audiometry made 11 ½ years later, with the patient complaining of sudden deafness at right**

Fig. 4 illustrates the preoperative audiometry. She returned eight days later, reporting intense vertigo.

Audiometry depicted deafness in the left ear (Fig. 4), refractory to treatment with Prednisone and Dimenhydrinate orally. Seven and a half years later, she came to medical consultation complaining of sudden deafness in the right ear for few hours, depicted in Fig. 5. She was put on oral treatment with Prednisone and had hearing improvement as shown in Fig. 5. The study with serology, rheumatologic tests and autoimmune and RM cerebellopontine angle were normal.

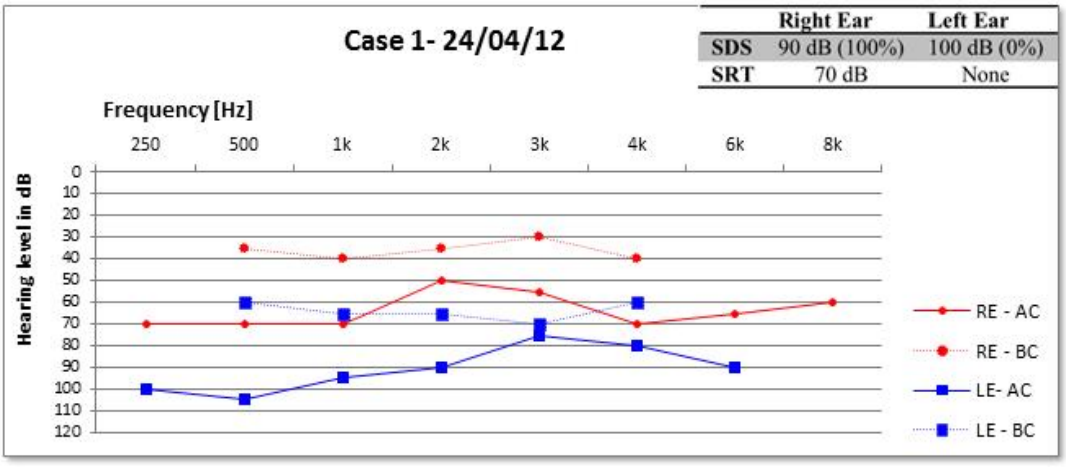


**Fig. 3. Audiometry made two months after treatment with Prednisone orally**

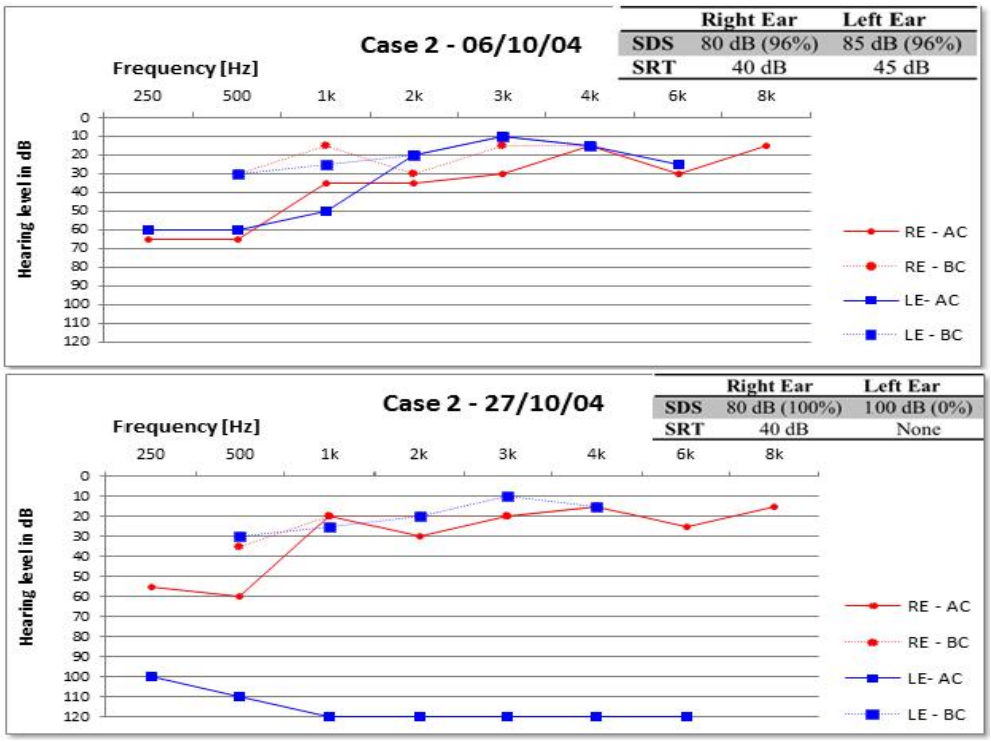


**Fig. 4. Preoperative audiometry in 2004 and audiometry on the eighth postoperative day, showing deafness in the operated ear (left)**

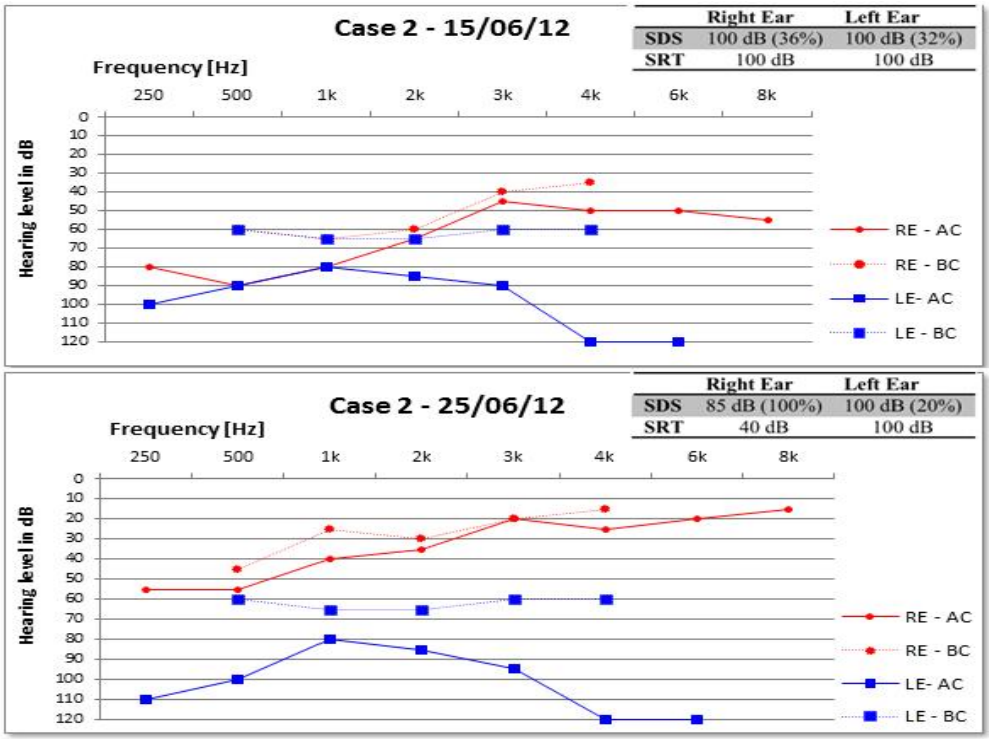


**Fig. 5. Audiometry seven and a half years after surgery, showing the sudden hearing loss in the right ear (unoperated) and audiometry performed 10 days after treatment with Prednisone, showing improvement the hearing pattern of the right ear**

## 4. DISCUSSION

Sudden deafness in patients with otospongiosis is a rare occurrence. We report two cases of patients with otospongiosis previously submitted to stapedotomy with transient success for a short time, followed by sudden total hearing loss in the operated ear and late sudden deafness in the unoperated ear.

Many years after surgical treatment both patients evolved with sudden deafness also in the remaining contralateral ear. In the first case, the sudden deafness has affected the unoperated ear eleven and a half years after contralateral surgery, with complete recovery of the standard hearing levels with oral Prednisone.

In the second case, sudden deafness in the unoperated ear occurred seven and a half years after contralateral surgery, with total recovery of standard hearing levels with oral Prednisone also.

The reports of sudden deafness in patients with otosclerosis are scarce in literature. Although this article describes only two cases, other similar cases have been reported. The studies point to a possible immunological cause for the association otosclerosis and sudden deafness.

Duvall et al. [9] reported six cases of sudden deafness in the unoperated ear of patients with otosclerosis who had undergone stapedotomy in the contralateral ear. According to the authors, the estimated incidence of sudden deafness in the unoperated ear of patients with otosclerosis is no larger than incidence in the general population, but with such a small number of reported cases, it is not possible to establish any relationship.

Felisati et al. [10] reported a case of a patient with Fabry disease and otosclerosis who presented sudden deafness. Two cases of sudden deafness have been described in patients with otosclerosis who developed deafness immediately after craniotomy made by unrelated disease to otologic [11].

## 5. CONCLUSION

Reports on the association of sudden deafness and otospongiosis are scarce. In both reported

cases the patients with bilateral otospongiosis had sudden deafness affecting both ears. The first episodes had the onsets postoperatively at the operated ears.

The second episodes were at the contralateral ears in the late follow-up, especially considering that in both cases the patients had satisfactory results regarding hearing improvement postoperatively although temporarily we therefore question the existence of this association.

Would sudden deafness in these patients, both postoperative and late, be expression of the same clinical phenomenon? Would the sudden deafness in these patients have otospongiosis as etiology?

## CONSENT

It is not applicable.

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## COMPETING INTERESTS

The authors declare that they have no competing interests.

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