Hypertrophic recurring lichen planus of the external auditory canal

Košec, Andro; Kostić, Mirjana; Ajduk, Jakov; Ries, Mihael

Source / Izvornik: **European Annals of Otorhinolaryngology, Head and Neck Diseases, 2019, 136, 123 - 126**

Journal article, Accepted version
Rad u časopisu, Završna verzija rukopisa prihvaćena za objavljivanje (postprint)

https://doi.org/10.1016/j.anorl.2017.12.013

Permanent link / Trajna poveznica: https://urn.nsk.hr/urn:nbn:hr:105:847769

Hypertrophic Recurring Lichen Planus of the External Auditory Canal

Andro Košec, MD, PhD[a], Mirjana Kostić, MD, PhD[b, c], Jakov Ajduk, MD, PhD[b], Mihael Ries, MD, PhD[b]

[a] Department of Otorhinolaryngology and Head and Neck Surgery, University Hospital Center Sestre milosrdnice, Vinogradska cesta 29, Zagreb, Croatia

[b] School of Medicine, University of Zagreb, Šalata 3b, Zagreb, Croatia

[c] Croatian Institute for Health Insurance, Zagreb, Croatia

Author correspondence:

Andro Košec, MD, PhD

Department of Otorhinolaryngology and Head and Neck Surgery, Clinical Hospital Center Sestre milosrdnice, Vinogradska cesta 29, Zagreb, Croatia

Phone: +385 1 3787108, Fax: +385 13769067

e-mail: andro.kosec@yahoo.com

Abstract

Introduction: We report a case of unilateral progressive primary hypertrophic lichen planus of the external auditory canal requiring several surgical interventions to deal with constant pruritus, otorrhoea, stenosis and conductive hearing loss.

Case summary: A 58-year-old woman was initially treated with meatoplasty for suspected chronic obliterating otitis externa. She remained symptom-free for 5 years, before the disease recurred, affecting other body surfaces as well. Otorrhoea, conductive hearing loss and pruritus worsened, and a canal wall down tympanomastoidectomy was performed, removing the skin of the external auditory canal and the tympanic membrane completely. Lichen planus was confirmed histopathologically.

Discussion: Very few surgical results have been published on stenosis of the external auditory canal caused by lichen planus. Complete medial external auditory canal skin elevation and removal with postoperative split-skin grafting is advised for initial treatment. We discuss treatment options and surgical outcome after initial surgical failure.

Key-words:

Lichen planus; tympanomastoidectomy; hearing loss; conductive; ear canal

1. Introduction

Lichen planus is a chronic disease caused by a T cell-mediated immune response of unknown origin. It may be found with other diseases of altered immunity, such as ulcerative colitis, alopecia areata, vitiligo, dermatomyositis, morphea, lichen sclerosis, and myasthenia gravis, usually affecting middle-aged adults. [1] Lesions initially develop on flexural surfaces of the limbs, with a generalized eruption marked by pruritus of varying severity, depending on the type of lesion and the extent of involvement. It can affect any area of the skin, alongside the oral mucosa. In up to 85% of patients, the disease resolves within 18 months. [2] Primary cutaneous lichen planus involving the external auditory canal is extremely rare. After performing a literature search of PubMed and Google Scholar databases using varying search queries (otic, external auditory meatus, ear canal lichen planus), we identified one individual case report and a case series encompassing 19 patients. [3, 4, 5, 6]

Here, we report a case of primary hypertrophic lichen planus of the external auditory canal requiring several surgical interventions to deal with constant pruritus, otorrhoea, stenosis and conductive hearing loss. We discuss treatment options and surgical outcome after initial surgical failure.

2. Case Report

A 58-year-old woman was examined due to intense pruritus of her left external auditory canal that had lasted for several months. No mechanical manipulation or medical treatment of the ear canal was noted in the patient's medical history. An otoscopic examination of the left ear revealed smooth, firm, whitish papules and granulation tissue partially covering the medial third of the external auditory meatus causing inflammatory stenosis. The right ear and external auditory canal were completely normal in appearance. She was diagnosed with chronic obliterating otitis externa and was scheduled for meatoplasty in general anesthesia.

The procedure went smoothly, and all excess inflammatory granulation tissue was removed from the distal third of the external auditory canal up to the tympanic membrane annular ligament. The skin defect was reconstructed with a split-skin transplant from the retroauricular area, while Silastic and soft Gel-foam packing were applied after surgery. Postoperative healing was uneventful. Excised tissue was not sent for additional histopathological testing.

After 5 years without any apparent symptoms, the condition recurred. Similar cutaneous areas of lichenoid papules on her lower and upper extremities and genitalia were then noted for the first time and a dermatologist was consulted. Lichen planus was suggested as a possible cause, and expectative treatment was advised. Otorrhoea and conductive hearing loss were soon noted, with a significant progression of fibrosis and granulation obliterating the distal third of the external auditory canal and obscuring the tympanic membrane. No other conditions of altered immunity were diagnosed, and the patient was not taking any medication that could aggravate the disease affecting her external auditory canal. Disease progression in other affected areas was noted as well. Topical and oral steroids (prednisone 1 mg/kg/day) proved ineffective in controlling the symptoms, the patient was unwilling to pursue topical tacrolimus treatment, and after a year of

unsuccessful treatment, surgical therapy in order to reduce otologic symptoms was advised. Multi-slice computed tomography showed normal temporal bone anatomy on the right side, and a normal, well pneumatized mastoid cavity on the left side. However, the medial half of the external auditory canal was filled with soft tissue corresponding to chronic inflammation, ending with an intact tympanic membrane and an intact ossicular chain and facial nerve. (Figure 1) Her preoperative pure tone audiogram showed a left-sided mixed hearing loss with a hearing threshold ranging from 60 do 105 dB, and an air-bone gap ranging up to 50 dB in the lower and middle frequencies. (Figure 2)

Surgery entailed complete removal of the skin lining the external auditory canal alongside the tympanic membrane. Subsequently, a canal wall down tympanomastoidectomy was performed through a retroauricular incision, and the cavity was lined with temporalis fascia grafting. Every precaution was made to avoid leaving any lichenoid tissue inside the tympanic cavity or the external auditory canal. Split-thickness skin grafts were also avoided, and the area of denuded bone in the external auditory canal was allowed to heal *per secundam intentionem.*

Histopathology showed attenuated orthokeratotic and hyperkeratotic epidermis with abundant lymphocyte infiltration, exocytosis and vacuolar degeneration in the basal layers, confirming the diagnosis of hypertrophic lichen planus. (Figure 3)

The patient has been in regular otologic postoperative follow-up for the last 6 months. Her postoperative pure tone audiogram showed a left-sided mixed hearing loss with a hearing threshold ranging from 35 do 90 dB, and an air-bone gap ranging up to 20 dB in the lower and middle frequencies. (Figure 4) She has no signs of disease recurrence in the external auditory meatus, (Figure 5) while other affected areas have shown slow, but steady improvement.

3. Discussion

Acquired conductive hearing loss may occur due to a large number of conditions, both inflammatory, post-surgical and neoplastic. [3] Differential diagnoses may include psoriasis, seborrheic dermatitis, cicatricial pemphigoid and contact dermatitis. In some instances, chronic idiopathic inflammatory medial meatal fibrotising otitis may occur simultaneously with oral lichen planus, but there are histopathlogical differences between the two entities. [4] All of these diagnoses were excluded after histopathologic examination and testing, and confirmatory biopsies from other loci confirmed the diagnosis.

This case was marked by a long period of disease inactivity after the first surgery, with limited secondary fibrosis in the immediate postoperative follow-up. However, after several years of inactivity, the inflammatory process was rekindled and soon after metal fibrosis and otorrhoea were noted, a generalized outbreak of lichen planus was observed.

Cases of external auditory canal stenosis and conductive hearing loss associated with lichen planus are extremely rare, with one individual previous case and a case series encompassing 19 patients reported in literature. The management, outcome and follow-up of known cases is displayed in Table 1. [3, 4, 6] Our case is interesting in that the disease was initially present only in the left external auditory canal, involved the outer epithelial layer of the tympanic membrane and that its first presenting symptom was conductive hearing loss. There were no signs of middle ear involvement. The disease started to affect other typical areas after a 5-year latency. In addition, it is one of few cases that was resolved through surgery.

The disease pathogenesis leading to fibrosis and stenosis is well documented, especially in the oesophagus. [1, 2] Treatment options are few, with immunosuppressive therapy recommended in refractory disease. [3] Unlike other inflammatory medial meatal fibrotising conditions, antibiotic

and antiseptic droplets and ointments have little or no effect, and granulations and hypertrophic tissue extend beyond the medial third of the external auditory meatus. [4] Our patient was treated with prednisolone both topically and orally, but no other immunosuppressant was tried, since the patient declined further medical therapy and was interested in pursuing surgical treatment options.

Surgery may be used if the disease does not respond to medical treatment in select areas, such as the external auditory canal, in order to improve hearing. Although all surgical options are straightforward, there is a specific added risk of inducing non-specific skin trauma leading to skin changes identical to the original disease (Koebner phenomenon) that is characteristic for lichen planus. [7]

4. Conclusion

Very few surgical results have been published on postinflammatory acquired fibrous stanosis of the external auditory canal. Most authors recommend an initial transcanal approach, complete medial external auditory canal skin elevation and removal with postoperative split-skin grafting and ear packing. However, there are no guidelines on treating recurring disease, and no published cases on treating recurring lichen planus in the external auditory canal. In this instance, radical skin removal through a canal wall down procedure resulted in symptom improvement, but also left the patient with a permanent conductive hearing loss.

Disclosure of interest

The authors have no conflicts of interest to disclose.

5. References

[1] Boyd AS, Neldner KH. Lichen planus. J Am Acad Dermatol 1991;25:593-619.

[2] Sugerman PB, Savage NW, Walsh LJ, Zhao ZZ, Zhou XJ, Khan A et al. The pathogenesis of oral lichen planus. Crit Rev Oral Biol Med 2002;13:350–65.

[3] Martin L, Moriniere S, Machet MC, Robier A, Vaillant L. Bilateral conductive deafness related to erosive lichen planus. J Laryngol Otol 1998;112:365-6.

[4] Hopsu E, Pitkäranta A. Idiopathic, inflammatory, medial meatal, fibrotising otitis presenting with lichen planus. J Laryngol Otol 2007;121:796-9.

[5] Park K, Taniyama T, Kabashima K, Miyachi Y. Primary cutaneous lichen amyloidosis of the external auditory canal, possibly due to scratching with a metal nail head for severe pruritus. Eur J Dermatol 2014;24:95-6.

[6] Sartori-Valinotti JC, Bruce AJ, Krotova Khan Y, Beatty CW. 10-Year Review of Otic Lichen Planus: The Mayo Clinic Experience. JAMA Dermatol 2013;149:1082-6.

[7] Becker BC, Tos M. Postinflammatory acquired atresia of the external auditory canal: treatment and results of surgery over 27 years. Laryngoscope 1998;108:903–7.

Tables

Table 1. Management, outcome and follow-up of cases previously reported in literature.

Study	Age	Sex	Symptoms	Duration of symptoms	Therapy	Outcome	Follow-up (years)
Sartori-Valinotti et al. [6]	21	Female	Otorrhoea, pain	2 years	Tacrolimus, topical	Subjective and objective improvement	9.5
	73	Female	Otorrhoea, pruritus, hearing loss	2 years	Tacrolimus, topical	Subjective and objective improvement	6
	19	Male	Otorrhoea, hearing loss	Few days	Tacrolimus, topical	Subjective and objective improvement	2.5
	55	Female	Otorrhoea, hearing loss	2 years	Tacrolimus, topical	Subjective and objective improvement	3
	66	Female	Otorrhoea, hearing loss	2 years	Tacrolimus, topical	Subjective and objective improvement	8.5
	57	Female	Otorrhoea, hearing loss	Unknown	Tacrolimus, topical	Subjective and objective improvement	11
	68	Female	Otorrhoea, hearing loss	Few days	Tacrolimus, topical	Subjective and objective improvement	5
	65	Male	Hearing loss, pruritus	6 months	Clobetasol propionate, topical	Unknown	None
	62	Female	Otorrhoea, hearing loss	Unknown	Tacrolimus, topical	Objective improvement, no change in symptoms	6
	69	Female	Otorrhoea, hearing loss, tinnitus	1 year	Tacrolimus, topical	Subjective and objective improvement	9
	21	Female	Otorrhoea	14 years	Tacrolimus, topical	Subjective and objective improvement	1
	66	Female	Otorrhoea, pruritus	8 years	Tacrolimus, topical	Unknown	None
	72	Female	Otorrhoea, pruritus, pain	2 years	Tacrolimus, topical	Unknown	None
	78	Female	Hearing loss	2 years	Tacrolimus, topical	No change in symptoms, worsening objective findings	0.5
	52	Male	Otorrhea, hearing loss, bleeding, crusting	10 years	Tacrolimus, topical	Unknown	None
	52	Female	Otorrhoea, pruritus	2 years	Tacrolimus, topical	No subjective or objective improvement	4
	68	Male	Otorrhoea, pruritus	Unknown	Tacrolimus, topical	Objective improvement, no change in symptoms	0.8
	49	Female	Hearing loss	Unknown	Ciprofloxacin and dexamethasone, topical	Unknown	4
	72	Female	Otorrhoea, hearing loss	4 years	Tacrolimus, topical	No subjective or objective improvement	3
Martin et al. [3]	69	Female	Otorrhea, hearing loss, pain, crusting	7 years	Surgery Acitretine, oral Prednisone, oral	Subjective and objective improvement	1
Our patient	58	Female	Otorrhoea, pruritus, hearing loss	4 months	Surgery Acitretine, oral Prednisone, oral	Subjective and objective improvement	0.5

Figures and Labels

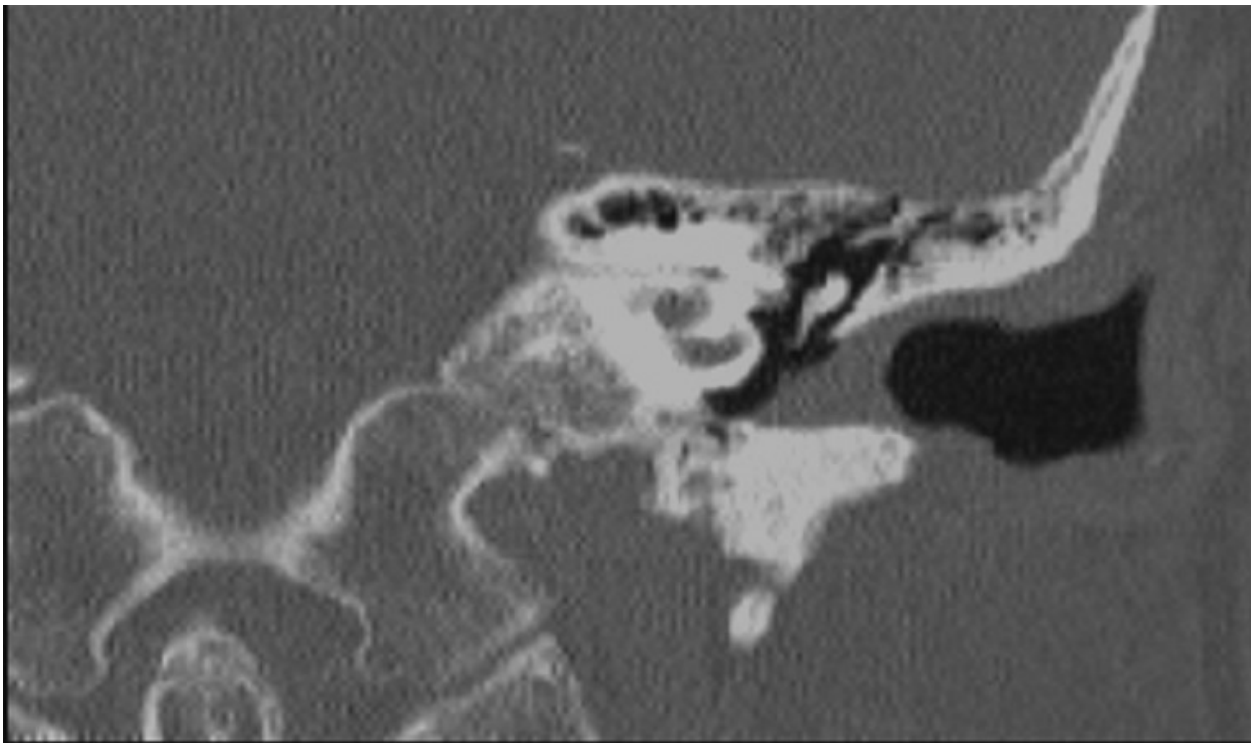
Figure 1. Multi-slice computed tomography, axial plane. The medial half of the external auditory canal filled with chronic inflammation tissue, ending with an intact tympanic membrane. No disease involvement of the middle ear was noted, and the ossicular chain and epitympanum are intact.

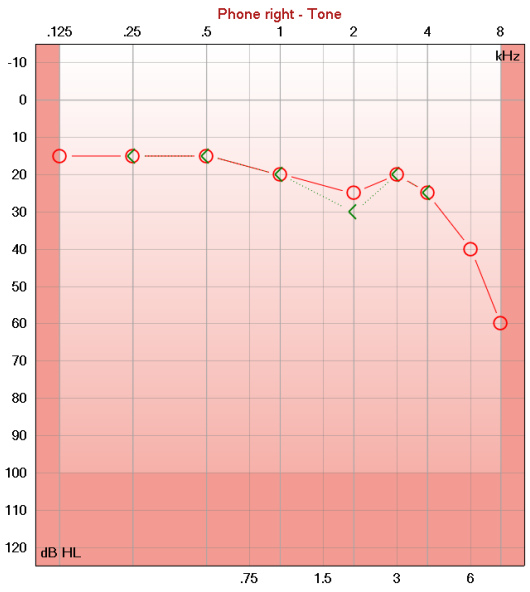
Figure 2. Preoperative pure tone audiogram showing left sided conductive hearing loss with a hearing threshold ranging from 60 to 105 dB, and an air-bone gap ranging up to 50 dB.

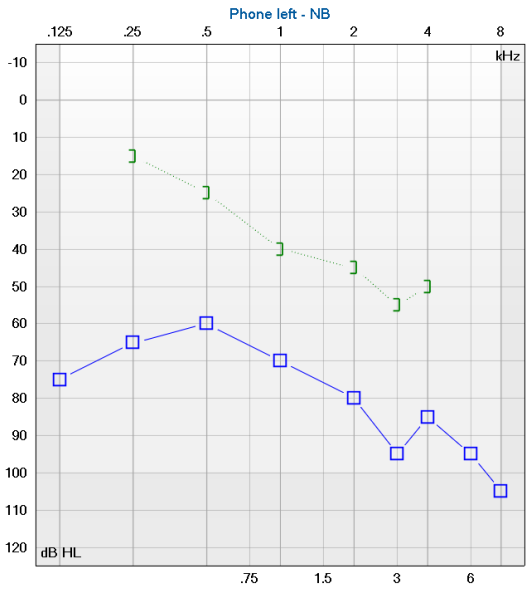
Figure 3. Haemotoxylin and eosin staining showing attenuated orthokeratotic and hyperkeratotic epidermis with abundant lymphocyte infiltration, exocytosis and vacuolar degeneration in the basal layers, typical of lichen planus.

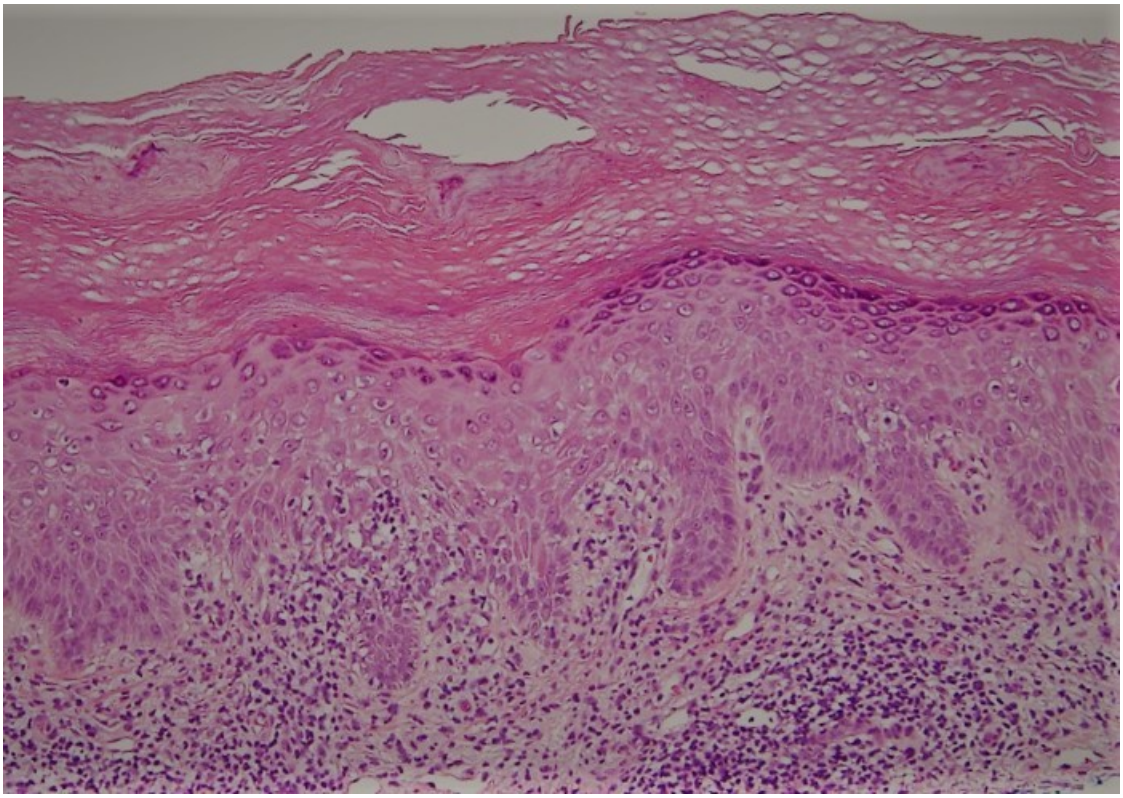
Figure 4. Postoperative pure tone audiogram showing left sided conductive hearing loss with a hearing threshold ranging from 35 to 90 dB, and a reduced air-bone gap measuring up to 20 dB.

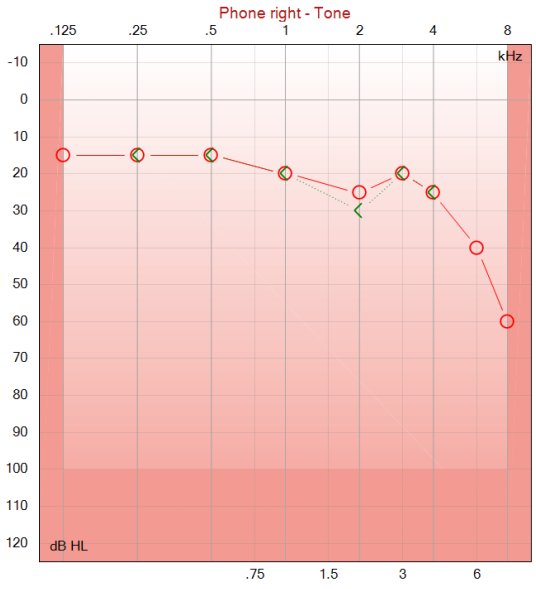
Figure 5. Postoperative appearance of the left external auditory meatus and radical cavity after 6 months of follow-up.

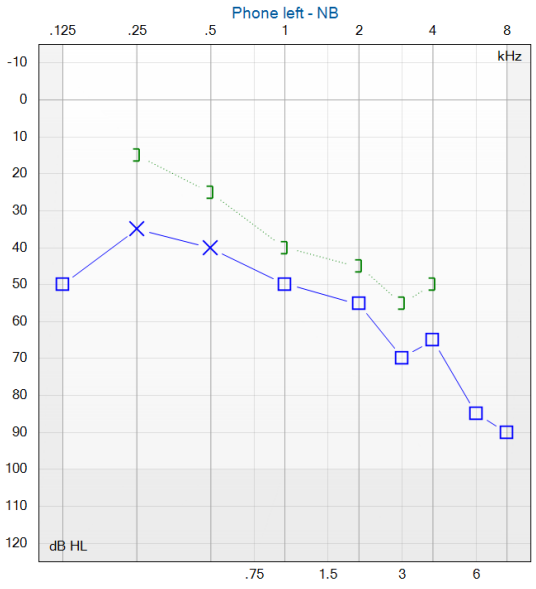



Phone right - Tone
.125
.25
.5
1
2
4
8
kHz
-10
0
10
20
30
40
50
60
70
80
90
100
110
120
dB HL
.75
1.5
3
6


Phone left - NB
.125
.25
.5
1
2
4
8
kHz
-10
0
10
20
30
40
50
60
70
80
90
100
110
120
dB HL
.75
1.5
3
6




Phone right - Tone
.125
.25
.5
1
2
4
8
kHz
-10
0
10
20
30
40
50
60
70
80
90
100
110
120
dB HL
.75
1.5
3
6


Phone left - NB
.125
.25
.5
1
2
4
8
kHz
-10
0
10
20
30
40
50
60
70
80
90
100
110
120
dB HL
.75
1.5
3
6