

Salivary Gland Choristoma of the Middle Ear and Review of the Literature

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ABSTRACT

Conductive hearing loss due to middle ear masses is uncommon and usually diagnosed after biopsy. We present a case of a permanent facial palsy occurred following an uneventful biopsy during an exploratory tympanotomy in a salivary gland choristoma of the middle ear. Most salivary gland choristomas have been found in the head and neck. Its location in the ear is extremely rare, and thus we present the 38th case in English and non-English literature from the first publication by Taylor in 1961. Complete surgical removal of salivary gland choristomas of the middle ear is indicated when may not result in permanent damage to the facial nerve. Only biopsy and observation are recommended when the mass is intimately associated with the facial nerve or there are unsafe facial nerve abnormalities. Although the facial nerve is involved in 40% of cases, transient or even permanent facial palsies are exceptional. The reactivation of latent herpes virus in the facial canal may be involved in facial palsy' etiology following minimal and uneventful middle ear surgery like a biopsy rather than nerve injury related to facial canal malformations.

Keywords: Salivary Gland; Choristoma; Middle Ear; Facial Palsy; Herpesvirus Reactivation

1. Introduction

Choristomas are not tumors but normal mature tissue masses of a single histological type that are at an abnormal anatomical location [1].

We present a rare condition, the 38th case of salivary gland choristomas of the middle ear in literature from the first publication in 1961 [2-22]. Only one case was bilateral, and described postmortem [23] (see **Table 1**). Our case was diagnosed by biopsy following an exploratory tympanotomy due to unilateral conductive hearing loss and showed the third reported permanent facial palsy. This article may contribute to the limited clinical knowledge of this condition and assist clinicians in its diagnosis and management.

2. Case Report

A 32-years-old Caucasian woman was referred to the ENT Department complaining of hearing loss increased after a delivery and coincident with left ear's acute otitis media (AOM) 3 months before. History of a self-limited ipsilateral facial paresis diagnosed as Bell's palsy 13 months before and a treatment with rifampicin, myam-

butol and isoniazid for 6 months treating lymph node tuberculosis 1.5 years before. Otomicroscopy was normal. Pure tone audiometry demonstrated a left ear conductive hearing loss with 40 dB pantonal air thresholds and normal bone thresholds in low frequencies shifting to 30 dB for frequencies above 2000 Hz. Tympanogram showed a reduction of compliance, pressures centered at 0 daPa and on-off stapedial reflexes, designating a decreased mobility of the ossicular chain. Preoperative assessment, examination and tests were normal. Exploratory tympanotomy of the left ear was performed under local anesthesia and sedation. Surgical findings included normal tympanic membrane and stringy mucus in the tympanic cavity, occupied by a large polyp-like lesion involving the mesotympanum and the incudo-stapedial joint. Ossicular chain was complete and mobile. We only performed a biopsy of the polypoid mass for diagnostic purposes and a transtympanic drainage was placed. The sample collected was reported as a salivary gland choristoma (**Figures 1-3**). Immunohistochemical staining for S-100 protein disclosed the presence of nerve fibers.

An immediate Computed Tomography (CT) scan was performed, showing only a mild hypertrophic middle ear

Table 1. Salivary gland choristomas of the middle ear reported in the literature (n = number of patients).

Race	Caucasian people 97.4% (n = 37)
	N/A 2.6% (n = 1)
Gender	Women 1.6:1 (female n = 23; male n = 14; N/A n = 1)
Side	Left ear 1.6:1 (left n = 23; right n = 14)
	Bilateral n = 1 (postmortem)
Presentation	Unilateral hearing loss > 1 year 87.2% (n = 33)
	Neonatal screening 5.3% (n = 2)
	School screening 7.9% (n = 3)
	Tinnitus 10.5% (n = 4)
Type of hearing loss	Conductive type 55.3%, moderate in 74%, before age 25 (n = 21)
	Sensorineural type 2.6% (n = 1)
	Mixed 21%, older than 15 years (n = 8)
	N/A 15.8% (n = 6)
Otoscopic visualization of a mass in the tympanic cavity	37% (n = 14)
Causes of misdiagnosis	Seromucous otitis and repetitive AOM 24% (n = 9), undergoing myringotomy and ventilation tube placement in half of these cases (n = 5)
	Suspicious of fixation of the ossicular chain 15.8% (n = 6): normal otoscopy, conductive hearing loss
Bone malformations	Affecting portions of the incus and the stapes 60.5% (n = 23), 50% simultaneous (n = 19)
	Malleus 10.5% (n = 4)
	Oval and round windows 15.8% (n = 6)
Facial nerve abnormalities	Dehiscent 31.6% (n = 12)
	Altered position 7.9% (n = 3)
	Mass very attached to bony canal or to dehiscent facial nerve 36.8% (n = 14)
	Mass preventing the visualization of the facial nerve 7.7% (n = 3)
Salivary gland choristomas location	Posterior region of the tympanic cavity, involving the incudo-stapedial joint 63.2% (n = 24)
	The mass was separated from the medial wall of the tympanic cavity and adjacent to the tympanic membrane 5.3% (n = 2)
	Pedicles have been observed 10.5% (n = 4)
Treatment	Transcanal tympanotomy 76.3% (n = 29)
	Retroaural tympanotomy 5.3% (n = 2)
	Mastoidectomy 15.8% (n = 6)
	Ossicular chain reconstruction surgery 18.4% (n = 7)
Outcomes	Complete removal 57.9% (n = 22)
	Incomplete removal 7.7% (n = 3)
	Only biopsy 28.9% (n = 11)
	N/A 7.9% (n = 3)

mucosa without destruction of bone structures (**Figure 4**).

Left facial paresis appeared one day following surgery, grade II according to the House-Brackman scale. Electroneuronography test (ENog) was performed at day 5th showing 36.9% on the affected side compared to the healthy side, consistent with neuropraxia, so we decided corticosteroid therapy and rehabilitation [24]. IgG antibodies against HSV-1 were not high. Grade II facial pa-

resis persisted despite medical treatment, home exercises and electrotherapy, withdrawn a few days later due to earache and hypogeusia. Six months following surgery the patient was admitted for immediate delivery after 31 weeks of gestation and a cesarean section was carried out. The pregnancy was unknown at the time of surgery. Fifteen months following surgery the patient was admitted to the Emergency Room with high fever and low level of consciousness with pustular skin blistering “in starry

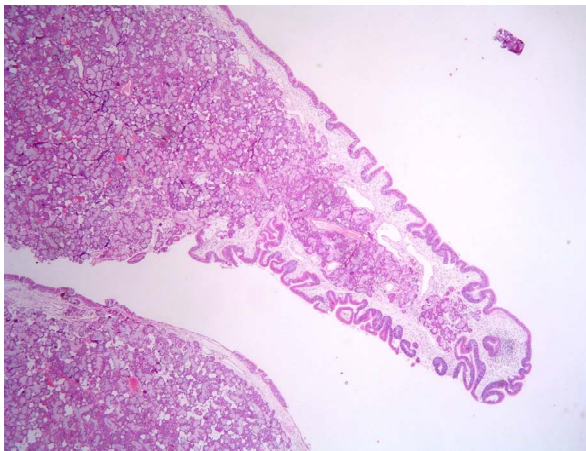


Figure 1. Overview showing polypoid appearance of the lesion. (HE, $\times 20$).

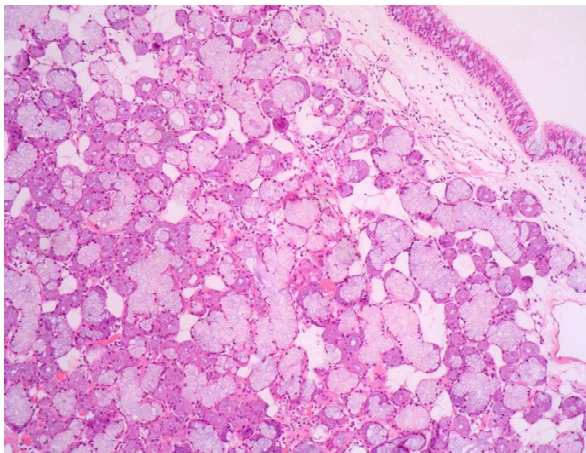


Figure 2. The lesion is covered by a pseudostratified ciliated epithelium with goblet cells and consisting of mucous acini, serous and mixed. (HE, $\times 100$).

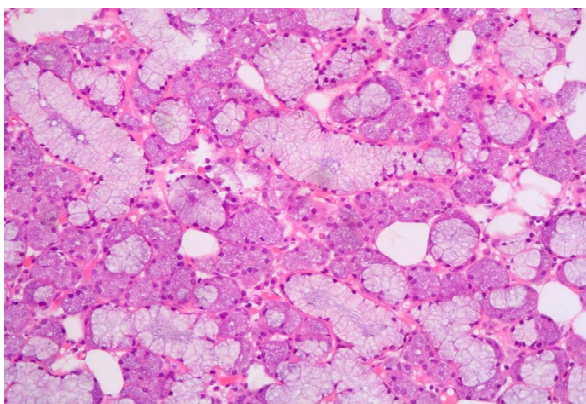


Figure 3. Detail of mucous acini, serous and mixed. (HE, $\times 200$).

sky” around the upper body consistent with chickenpox. Lab tests of immune status were normal: complete blood cell count, C-reactive protein (CRP), immunoglobulins

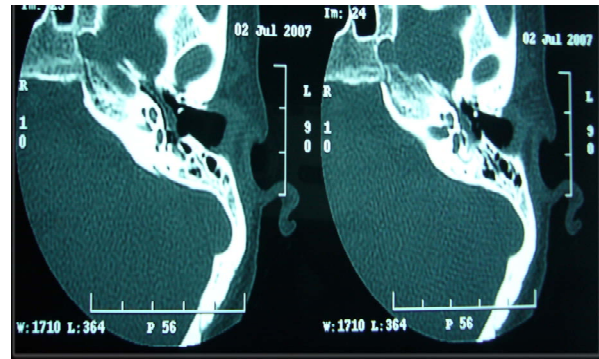


Figure 4. Left-sided temporal bone scan reveals soft tissue density in the middle ear emerging over the promontory. There are no osseous erosions.

(Ig) G, M, and A, antinuclear antibody, and total protein. Blood and CSF culture and serum PCR for herpes-simplex virus were negative. With the diagnosis of chicken-pox encephalitis treatment with acyclovir was prescribed [25]. CT scans performed 3 and 4 years after surgery didn't show any increase in the middle ear hypertrophic mucosa or destruction of structures. A Grade I-II facial palsy was the permanent sequela.

3. Discussion

Choristomas are also known as heterotopic tissue, ectopia, congenital heterotopic remnants or aberrant debris. Most salivary gland choristomas have been found in head and neck. The most common locations are the paraparotid area, the upper neck, mandible, external ear canal, vulva, mediastinum, cerebellopontine angle, thyroid gland, pituitary gland, rectum, thyroglossal duct, parathyroid capsule, tongue, tonsil, and tonsillar fossa. Most of choristomas are non-neoplastic lesions that do not require excision, unless recent growth is observed in adults or disproportionate growth occurs in childhood, but biopsy is needed for diagnosis [26]. We updated the review conducted by Rinaldo [27] and recent reports [28-37] of middle ear location.

Salivary gland choristomas are the most common type of heterotopic tissue found in the middle ear. Pathogenesis of salivary gland choristomas of the middle ear remains unknown. Willis [38] has suggested 3 possibilities: 1) abnormal persistence and development of vestigial structures; 2) dislocation of a portion of a definitive organ rudiment during mass movement; and 3) heteroplasia, which is abnormal differentiation of local tissues. Abadir [39] suggested that the persistence of remnants of salivary glands in the middle ear must occur before the fourth month of embryonic development. We agree with Nassar [40] that the criteria described by Buckmiller [41] for the definition of a syndrome are present in less than a third part of the reported cases and this condition would not

constitute a syndrome. Incidental lesions are not consistent enough to describe a syndrome: preauricular fistulae/cysts, branchial cysts, absence of malleus/stapes muscles, oval and round windows and ossicular anomalies, periauricular alopecia, hemifacial atrophy, situs inversus. Most injuries associated to middle ear salivary gland choristomas are probably due to the sole presence and action of the choristomatous mass and not resulting from abnormal development of the first and second branchial arches. Most ossicular defects described resemble lytic lesions found in other chronic middle ear conditions.

Low otoscopic visualization of a mass in the tympanic cavity (37%) and high prevalence of conductive hearing loss in patients' first decades of life misdiagnoses the condition. Antecedents of female, hearing loss presentation following a delivery and a Type As Tympanogram directed us to the preoperative diagnosis of ossicular chain fixation.

CT images started to be used in cases published after 1992 and only in 12 patients, when hearing loss was combined with a remarkable image of otoscopic tympanic mass and cases of normal otoscopy and striking high audiometric thresholds. The appearance of the lesions has been described in several colors (yellowish, reddish, brownish or pinkish) and consistency (soft and elastic or rubbery), with smooth or lobulated surfaces. Sizes vary from 2 mm up to occupy almost the whole tympanic cavity. At any case its length has exceeded the limits of the tympanic cavity, the entrance of the tympanic orifice of the Eustachian tube, the facial recess and the tympanic sinus.

A relatively straightforward differential diagnosis can be formulated with epidermal lesions like granuloma (nodular inflammatory lesion) or cholesteatoma (accumulation of keratin debris within a sac of squamous epithelium in absence of otorrhea), paragangliomas or vascular lesions (vascularized appearance, pulsatile). But the look, feel, location and CT images make the differential diagnosis more challenging with benign or malignant tumors (adenoma, schwannoma, endolymphatic sac tumor, rhabdomyosarcoma, metastatic disease, histiocytosis X). Only histopathological investigations can establish the definite diagnosis of salivary gland choristoma, differentiating it from hamartoma (various mature tissue normally found in the site of lesion), dermoid cysts (ectodermal tissue only with epidermal adnexae), epidermoid cysts (ectodermal tissue with no epidermal adnexae) and teratomas (benign/malignant mature/immature tissues of several embryological origin) [1].

One objective criterion to indicate removal of the lesion when diagnosed is the treatment of conductive hearing loss and avoids the probable course of the hypoacusia to an increase in bone thresholds. Complete surgical removal is indicated when may not result in permanent

damage to the facial nerve. Only biopsy and observation are recommended when the mass is intimately associated with the facial nerve or there are unsafe facial nerve abnormalities. Facial nerve electrical monitoring and the use of KTP laser have allowed choristomas removal without neurological sequelae. Involvement of facial nerve or round/oval windows prevents a proper reconstruction of the frequently affected ossicular hearing mechanism (71%). Incomplete removal of salivary gland choristoma of the middle ear have not ever evolved to malignization, while monitoring of all reported cases has not been long enough to predict a possible malignancy. Rinaldo [27] points to a prominent benign behavior of ear's salivary gland choristomas in her literature review, including 4 malignant salivary gland tumors originating from the middle ear and speculatively evolving from choristomas.

Despite the frequent involvement of the facial nerve (40%), only two transient facial palsies and three permanent ones (10.5% of patients) have been described following surgery. Surgical damage may appear as the immediate cause of the facial paresis/palsy, but herpesvirus implication could be considered even with lower HSV-1 titers: our patient had suffered a previous transient episode of facial palsy and subsequently presented a rare herpetic encephalitis (1.7 cases of encephalitis per 100,000 cases of chickenpox). Clinical practice guidelines recommend treatment with acyclovir even in undiagnosed herpetic encephalitis but suspected. Unknown host immune factors and the unsuspected pregnancy may be responsible to suffer facial palsy even before 2.5 days described in Bonkowsky series [42]. This supports the hypothesis of facial palsy by reactivation of latent herpes viral particles stationed in the geniculate ganglion when surgery is limited to a non-damaging simple biopsy [43].

4. Conclusion

Complete surgical removal or rare salivary gland choristomas of the middle ear is indicated when may not result in permanent damage to the facial nerve. Only biopsy and observation are recommended when the mass is intimately associated with the facial nerve or there are unsafe facial nerve abnormalities. Although facial nerve is involved in 40% of middle ear's choristomas, transient or even permanent facial palsies are exceptional. The reactivation of latent herpes virus in the facial canal may be involved in facial palsy's etiology following minima and uneventful middle ear surgery like a biopsy rather than nerve injury related to facial canal malformations.

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