Case report

Post-surgery cholesteatoma complicated by facial nerve paralysis: A case report from Afghanistan

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ABSTRACT

Introduction: Cholesteatoma is a benign tumoral lesion of squamous epithelial cells in middle ear that can exist as congenital or acquired forms.

Presentation of cases: A 35-year-old housewife presented to ENT clinic of a private hospital in Kabul, Afghanistan, with a complete facial nerve paralysis in the right side. In her antecedents, there is a tympanomastoidectomy due to chronic middle ear infection. First symptom was right side earache without any discharge. She started to notice a progressive nodule in the posterior-inferior side of her right ear. The patient was taken to the operating room. She underwent general anesthesia, an extensive cholesteatoma was removed, and a limited area of the fallopian canal in which facial nerve oedema or redness was evident. Post-operative House Brackmann grade was 1 on day 15 after the surgery.

Discussion: Cholesteatoma is primarily managed surgically and currently there is no suitable medical substitute treatment strategy for cholesteatoma. Hearing improvement, making the ear dry and total omission of cholesteatoma are primary goals of surgical interventions in cholesteatoma management.

Conclusion: Cholesteatoma after surgical manipulations of middle ear is a rare complication with notable morbidity that has been reported almost from all around the world but our patient is the first reported case of cholesteatoma formation after surgical management of COM from Afghanistan that presented with facial nerve paralysis and hear decline.

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1. Introduction

Cholesteatoma is a benign tumoral lesion of squamous epithelial cells in middle ear that can exist as congenital or acquired forms [1]. The existing theories about the natural history of acquired cholesteatoma are squamous metaplasia triggered by middle ear inflammation, squamous cell migration via a perforation in drum, and basal cell hyperplasia [2]. Another notable mechanism of cholesteatoma formation, is secondary triggering by trauma, infection or surgical interventions [1]. Cholesteatoma is a rare but important complication after otologic surgical interventions as it can become infected or have compressive effects on adjacent structures such as facial nerve [3,4]. Facial nerve compression can result in facial nerve paralysis, and surgical decompression can mitigate the situation [4]. Based on the Surgical Case REport, 2020 (SCARE) guidelines [5], in this article we report Afghanistan’s first confirmed case of facial nerve paralysis resulted from cholesteatoma formation after surgical management of chronic otitis media (COM).

2. Case presentation

A 35-year-old housewife female referred to ENT clinic of a private hospital in Kabul, Afghanistan with a complete right side facial nerve paralysis. The facial muscles weakness started about 6 months before her referral; the weakness was progressive and during this time she did not visit any health facilities and did not receive any treatments. In her antecedents, there is a tympanomastoidectomy due to chronic middle ear infection dating back in December 2019 and otherwise in good health.
Family history, drug history and social history were unremarkable. The first symptom was right side earache without any discharge, she started to notice a progressive nodule in the posterior-inferior side of her right ear. The facial weakness has progressed to a nodule size. Later she presented progressive weakness around her mouth and forehead muscles (Fig. 1). The patient denied any taste impairment and/or deafness. The examinations were done on the patient in good state. In inspection, signs of right complete peripheral facial paralysis, with a giant nodule in the back of the ear were seen (Fig. 2). Pre operation House-Bracmann grade was four (Table 1). The patient was transferred to ENT ward for further interventions. Computed Tomography (CT) scan and Magnetic Resonance Imaging (MRI) were not available in the center. Blood investigations were in normal ranges; a total leucocyte count of 6000/mcL (Normal 4000/mcl to 11,000/mcl), Hemoglobin (HB) 11 g/dL (Normal: 13 g/dL to 18 g/dL), Platelet 224 × 10³ (Normal: 140 10³ to 400 10³), Partial Thromboplastin Time (PTT) 33 (Normal: 23 s to 35 s), International Normalized Ration (INR) 1 (Normal: 0.8 to 1.1). The patient was taken to the operating room. She underwent general anesthesia, an extensive cholesteatoma was removed, and a limited area of the fallopian canal in which facial nerve oedema or redness was evident. The epineural sheet was opened for nerve decompression. The surgery had been carried out by attending ENT surgeon. The patient was discharged after 24 h after surgery and visited on day 15 after surgery, post-operative House Brackmann grade was 1 on day 15 after surgery, (Fig. 3), (Table 1) [6].

3. Discussion

Cholesteatoma is a benign lesion in middle ear with local invasion and potential destructive effects due to its excessive abnormal growth

| Grade | Results | Definition of recovery |
|-------|---------|------------------------|
| I     | Super   | Excellent with minimal mass movement |
| II    | Excellent | Mass movement; can close eyes, smile |
| III   | Good    | Tone and symmetry without ability to smile and close eyes simultaneously |
| IV    | Fair    | Incomplete eyelid closure or very weak mouth movement |
| V     | Poor    | Symmetry only, tone intact, no movement |
| VI    | Failure | Flaccid, tone lost |

Fig. 1. The photos show the peripheral weakness around the mouth and eyebrow in the right side.

Fig. 2. The photo show a giant nodule in the inferior-posterior of the right side ear.

Table 1
Scale of facial muscle function after repair.
Cholesteatoma is categorized into two subgroups; congenital and acquired which the acquired subtype consists of retraction pocket and non-retraction pocket forms [1,7]. Congenital anomalies, trauma, surgical manipulations and infections of middle ear are some of the risk factors of cholesteatoma development [8,9]. Our patient had a history of COM that was managed surgically. Cholesteatoma can cause symptoms such as otorrhea, progressive conductive hearing loss and compressive effects on adjacent structures such as facial nerve, resulting in facial nerve paralysis [4,10]. Our patient experienced hearing decline and facial nerve paralysis after she underwent tympanotomy for management of COM. Cholesteatoma is primarily managed surgically and currently there is no suitable medical substitute treatment strategy for cholesteatoma [7]. Hearing improvement, making the ear dry and total omission of cholesteatoma are primary goals of surgical interventions in cholesteatoma management [11]. In our patient, surgical removal of cholesteatoma resulted in improvement of hearing and restored the facial nerve function. Cholesteatoma after surgical manipulations of middle ear is a rare complication with notable morbidity that has been reported almost from all around the world but our patient is Afghanistan’s first reported case of cholesteatoma formation after surgical management of COM that presented with facial nerve paralysis and hearing decline.

Finally, we should highlight that although cholesteatoma is a benign tumoral lesion of middle ear, it can cause serious morbidity and be annoying to patients as it can have compressive effects on adjacent structures such as facial nerve. Thus, facial nerve paralysis after otologic surgical interventions should make physicians suspicious of cholesteatoma formation and appropriate surgical interventions should be considered as there is not any medical substitution for surgery in management of cholesteatoma. We hope that reporting this case would make our colleagues more sensitive to surgical complications of middle ear surgery and improve patients’ quality of life after surgical management of COM.

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Ethical approval
This is a case report paper.

Consent
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CRedit authorship contribution statement
SH, SQ, SHM and YF conceptualized the study. YF, AAMN and SQ acquired the data, and drafted the manuscript. FN and SH revised the manuscript for critical intellectual concept and approved the final draft.

Declaration of competing interest
Author declare no conflict of interest.

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Registration of research studies
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References
[1] K.L. Kennedy, A.K. Singh, Middle Ear Cholesteatoma, StatPearls [Internet], 2020.
[2] L. Louw, Acquired cholesteatoma: summary of the cascade of molecular events, J. Laryngol. Otol. 127 (6) (2013) 542.
[3] M. Nejadkazem, et al., Intratympanic membrane cholesteatoma after tympanoplasty with the underlay technique, Arch. Otolaryngol. Head Neck Surg. 134 (5) (2008) 501–502.
[4] C.-L. Kuo, et al., Updates and knowledge gaps in cholesteatoma research, Biomed. Res. Int. 2015 (2015).
[5] R.A. Agha, et al., The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[6] W. House, Facial nerve grading system, Otolaryngol. Head Neck Surg. 93 (1985) 184–193.
[7] J.T. Castle, Cholesteatoma pearls: practical points and update, Head Neck Pathol. 12 (3) (2018) 419–429.
[8] A.L. James, B.C. Papsin, Some considerations in congenital cholesteatoma, Curr. Opin. Otolaryngol. Head Neck Surg. 21 (5) (2013) 431–439.
[9] B.A. Jessnings, et al., The genetics of cholesteatoma. A systematic review using narrative synthesis, Clin. Otolaryngol. 41 (1) (2016) 55–67.
[10] J.E.A.P.d. Aquino, N.A. Cruz Filho, J.N.P.d. Aquino, Epidemiology of middle ear and mastoid cholesteatomas: study of 1146 cases, Braz. J. Otorhinolaryngol. 77 (3) (2011) 341–347.
[11] A.G. Pusalkar, Cholesteatoma and its management, Indian J. Otolaryngol. Head Neck Surg. 67 (3) (2015) 201–204.