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Unusual Case of Fishhook Lodged in the Neck:
A Case Report and Literature Review

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Original Investigations

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Publisher Contact
Address: Molla Gürani Mah. Kaçamak Sk. No: 21/1
34093 İstanbul, Türkiye
Phone: +90 (530) 177 30 97 / +90 (539) 307 32 03
E-mail: info@galenos.com.tr/yayin@galenos.com.tr
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Contents

Editorial Self-Publication in Scientific Journals: Insights and Implications for Otorhinolaryngology Practice

Editorial

Ali Bayram

University of Health Sciences Türkiye, Kayseri City Hospital, Clinic of Otorhinolaryngology, Kayseri, Türkiye

Editorial board members play a pivotal role in the publication process of scientific journals, such as assessing the suitability of manuscripts for review, selecting appropriate reviewers, and making final decisions regarding acceptance or rejections. Since publishing in scientific journals is a key means for researchers to gain academic promotion and recognition, journal editors have a considerable influence on the academic careers of scientists. Journal editorial board members are typically chosen from experts in their field, many of whom are not full-time professionals and manage their editorial responsibilities alongside their academic work. As active and productive researchers, they may prefer to publish their studies in their journals. Editorial self-publication denotes the practice whereby members of an editorial board publish their own research in the journals for which they hold editorial responsibilities (1). Although there are no specific restrictions on this practice, being an editor-as-author (EAA) can raise significant concerns, including potential conflicts of interest, bias, and favoritism.

Leading international publication ethics guidelines, including those from the International Committee of Medical Journal Editors and the World Association of Medical Editors, have outlined general frameworks and precautions concerning editorial self-publication (2,3). In summary, the guidelines do not impose any restrictions on serving as an EAA, provided that a transparent review process is maintained, ensuring no undue advantages are granted to the editorial members. Additionally, the guidelines suggest that editorial members should avoid submitting articles to their journals when suitable alternatives are available (2,3).

There is limited data in the literature regarding the self-publication practices of journal editors. Liu et al. (4) conducted a comprehensive analysis of editorial publication patterns among 20,000 journal editors and found that 12% of editors published at least 20% of their papers in their own journals, while 6% published at least 33%. Helgesson et al. (5) performed a systematic review on the prevalence of editorial self-publication

ORCID IDs of the authors:
A.B. 0000-0002-0061-1755

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Corresponding Author:
Ali Bayram, MD;
dralibayram@gmail.com

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and identified significant variation across academic fields, with self-publication ranging from negligible to extensive.

Research on editorial self-publication remains scarce in medical journals, particularly within specific specialties. In otorhinolaryngology, Bayram et al. (1) investigated the prevalence and characteristics of editorial self-publication, analyzing 795 editorial board members from 12 Science Citation Index-Expanded journals in 2023. They found that 185 editors (23.3%) had at least one instance of EAA, with rates ranging from 11.3% to 41.5% across different journals. The editorial members were listed as an EAA in 290 of the 2106 articles (13.8%), with a range of 5.7% to 54.5%, and original articles were the most common type of editorial self-publication. The authors concluded that the prevalence of editorial self-publication varies widely among otorhinolaryngology journals, similar to findings in other fields.

As a conclusion, editorial self-publication is a significant issue that stakeholders in the scientific community should recognize. The review process for such articles must be transparent, ensuring objective and unbiased evaluation. EAAs should refrain from participating in the review and decision-making processes of their submissions and adhere to the recommendations outlined in international publication ethics guidelines regarding editorial self-publication. Further studies investigating editorial self-publication practices across different scientific disciplines, including otorhinolaryngology, could help in establishing mechanisms for self-regulation and addressing ethical concerns effectively. The Turkish Archives of Otorhinolaryngology's editorial board is resolutely dedicated to maintaining the highest ethical standards in academic publishing. Our board diligently follows international rules and best practices to ensure that editorial self-publication is carried out with honesty, fairness, and accountability.

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Investigation of Keratinized Squamous Epithelium from Mastoid Cortical Bone Dust in Patients with or without Cholesteatoma

Original Investigation

Halil Orkan Orman[1], Muharrem Dağlı[1], Tuğba Taşkın Türkmenoğlu[2]

[1]Dışkapı Yıldırım Beyazıt Research and Training Hospital, Clinic of Otorhinolaryngology and Head and Neck Surgery, Ankara, Türkiye
[2]Dışkapı Yıldırım Beyazıt Research and Training Hospital, Clinic of Pathology, Ankara, Türkiye

Abstract

Objective: This study aimed to investigate squamous metaplasia in mastoid cells of patients undergoing surgery for chronic otitis media (COM) with or without cholesteatoma. Bone dust was stained with hematoxylin and eosin (H&E) for squamous cells and keratin and immunohistochemically for p63. Additionally, the feasibility of routine pathological examination of bone dust via H&E staining was evaluated for cost-effectiveness and for identifying patient groups needing advanced follow-up.

Methods: Thirty-one patients with COM were enrolled: 14 with cholesteatoma (study group) and 17 without cholesteatoma (control group). Mastoid bone dust obtained during surgery was examined specifically for the presence of squamous cells, keratin, and p63, with evaluation performed using H&E and immunohistochemical staining techniques. Findings were compared between the study and control groups.

Results: Keratin was significantly more frequent in the study group than in controls (43% vs. 6%, p=0.01). No significant differences were observed for squamous cell (p=0.43) or p63 expression (p=0.20). However, when any of the three markers were positive, a statistically significant difference was found between the groups (43% vs. 12%, p=0.049).

Conclusion: These findings suggest that the mastoid air cell systems of patients with cholesteatoma may be affected differently prior to cholesteatoma spreading to the mastoid system. This could be linked to microcirculation of inflammatory proteins, impaired aeration, and the formation of retraction pouches. These results align with the metaplasia theory as a possible explanation for the etiopathogenesis of acquired cholesteatoma.

Keywords: Cholesteatoma, otitis media, mastoid bone, squamous metaplasia, p63 protein, immunohistochemistry

ORCID IDs of the authors:
H.O.O. 0000-0003-3260-0468
M.D. 0000-0003-2099-9395
T.T.T. 0000-0001-6314-2928

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Corresponding Author:
Halil Orkan Orman, MD;
orkanorman@gmail.com

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Introduction

Cholesteatoma, a disease studied for centuries, remains enigmatic. The most discussed theories on the etiopathogenesis of cholesteatoma include invagination, basal hyperplasia, migration, and squamous metaplasia. The squamous metaplasia theory, introduced by von Tröltsch in 1864, posits that middle ear mucosa transforms into squamous epithelium under pus pressure. Wendt

(1) later suggested that this metaplasia results from chronic inflammation, and Sadé et al. (2) demonstrated pluripotent middle ear mucosal cells transforming under inflammation. Support for the theory includes biopsies showing keratinized squamous epithelium in pediatric patients with otitis media with effusion. Despite these findings, histological proof or experimental confirmation of these mechanisms contributing conclusively to cholesteatoma formation is lacking (3-7). In our study, we examined mastoid bone dust samples histopathologically and immunohistochemically (IHC) with the hypothesis that squamous epithelial tissue undergoing metaplasia within the mastoid air cell system could be a factor for recurrences in cases where cholesteatoma is limited to the middle ear. Therefore, more emphasis was placed on this theory. Moreover, if mastoid bone dust obtained during mastoidectomy is routinely subjected to histopathological examination using only hematoxylin and eosin (H&E) staining as a cost-effective and time-efficient method, patients with positive findings could be identified earlier and directed toward closer monitoring or more advanced diagnostic evaluation.

Methods

Our study is an original research designed prospectively and conducted at Dışkapı Yıldırım Beyazıt Research and Training Hospital. Ethical approval was received from Dışkapı Yıldırım Beyazıt Research and Training Hospital (decision no: 90/18, dated: 22.06.2020).

Files of all patients who were operated on due to chronic otitis media (COM) and met the inclusion criteria for the study were scanned, and the data was recorded. The content of this data included age, sex, type of surgery performed, the side of the operation, and previous otological surgical history. Informed consent, both verbal and written, was obtained from patients before surgery. Additionally, pathologies identified in the middle ear and mastoid air cell system during the operation were recorded, and histopathological evaluations of tissue samples taken during the operation were also documented.

Inclusion Criteria: Patients with no previous history of otologic surgery and who were planned to be operated on for COM with cholesteatoma (COMC) were accepted in the study group. The cholesteatoma in the patients was classified according to the STAM grading system, with involvement restricted to the T and A subzones (8). Patients with no previous history of otologic surgery and who were planned to be operated on for COM with no cholesteatoma were accepted in the control group.

Exclusion Criteria: Patients with a history of previous otologic surgery, patients who were planned to be operated on for COM without mastoidectomy, patients with cholesteatoma in mastoid air cells, patients with congenital cholesteatoma, patients who had acquired cholesteatoma due to traumatic or iatrogenic tympanic membrane perforation were excluded from the study.

Bone dust was collected during mastoidectomy using an endotracheal suction system held vertically with an exit filter to prevent dust loss (Figures 1,2). Mastoidectomies were performed as canal wall-up (CWU) or canal wall-down (CWD) procedures without cavity obliteration. Collection began only after the mastoid cortex had been washed with saline and the infusion removed via a separate suction-to avoid contamination, and it was ceased before the external

Figure 1. An illustrative image of the tracheal aspirate kit used for sample collection

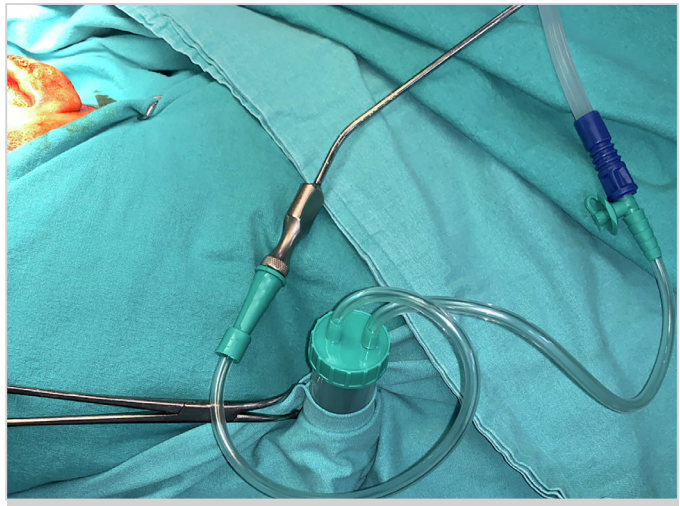

Figure 2. An illustrative image showing the stabilized position of the tracheal aspirate kit during the procedure, with the Fergusson aspirator attached at the tip

ear canal (EEC) skin incision or any intervention to the middle ear or cholesteatoma sac. The obtained bone dust was fixed in 10% neutral buffered formaldehyde and processed routinely: tissues were embedded in paraffin, sectioned at 3.5 µm, and stained with H&E using an automatic tissue staining device. All specimens were examined under a light microscope for keratin, squamous epithelium, and granulation tissue. To enhance detection of squamous epithelium, IHC staining for p63 was performed using the 4A4 clone (Ventana Benchmark XT, Roche Diagnostics, Switzerland). All mastoidectomy procedures were carried out by the same experienced surgeon. Following staining, the bone dust samples were evaluated for the presence of squamous cells (Figure 3) and keratin on H&E sections (Figure 4) and for p63 expression on IHC sections (Figure 5). Squamous cells were identified as isolated or clustered formations, while keratin was recognized by its acellular appearance and measured in micrometers. p63 expression was observed in the nuclei of squamous cells, and all parameters were recorded as either present or absent.

Statistical Analysis

Tested variables were evaluated for both groups as age, sex, IHC staining of p63, keratin presence and squamous cell presence. All the statistical analysis were run on IBM SPSS Statistics for Windows Version 26.0 software. Ordinary variables were summarized as mean ± standard deviation, median value (minimum-maximum). Groups were compared with student's t-test in ordinal variables after homogeneity tests were run and with chi-square test in nominal variables. A p-value of <0.05 was considered statistically significant.

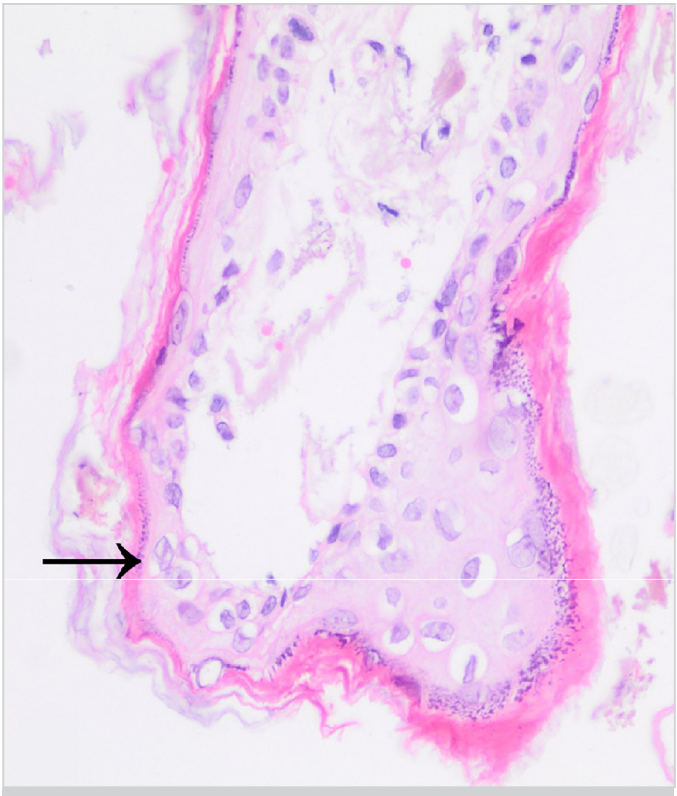

Figure 3. The images of squamous cells under a light microscope at 400 magnification field, marked with black arrow, following H&E staining
H&E: Hematoxylin and eosin

Results

Our study included 18 male and 13 female patients, totaling to 31 individuals. Mastoid bone dust was collected from all participants and subjected to histopathological examination. Based on the inclusion criteria, 14 individuals were assigned to the study group, while the remaining 17 formed the control group. The patients' ages ranged from 14 to 76 years, with a median age of 37.32±15.694 years (Figure 6).

In the study group, there were seven male patients (50%) and seven female patients (50%), with a median age of 35.14±17.244 years (Figure 6). Patient ages in this group ranged between 15 and 76 years. In the control group, there were 11 male patients (64.7%) and six female patients

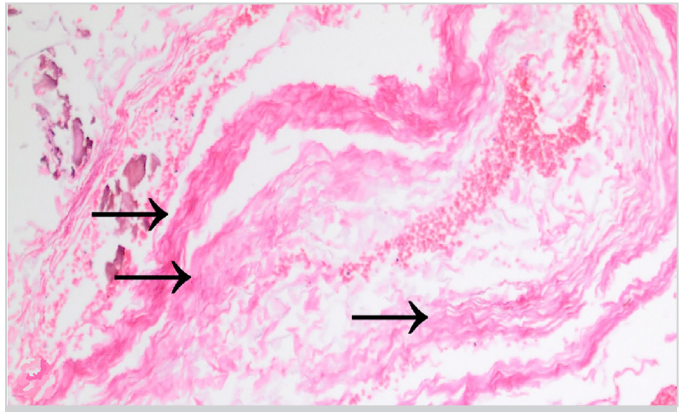

Figure 4. The image of keratin lamellae after H&E staining observed at a magnification field of ×200 under a light microscope, marked with black arrows
H&E: Hematoxylin and eosin

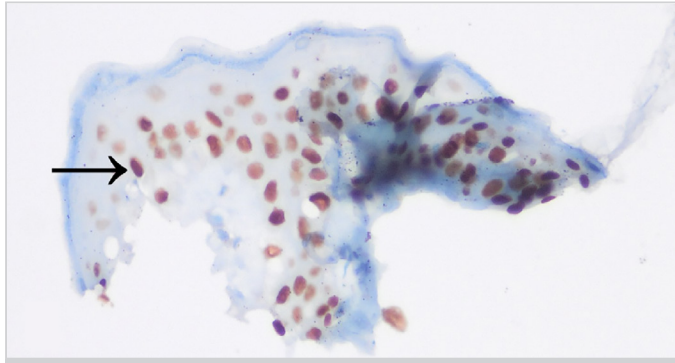

Figure 5. The image of p63 immunohistochemical staining of squamous cell nuclei at a magnification field of ×400 under a light microscope. The arrow indicates the squamous cell nucleus

Table 1. The table shows the frequency of keratin and squamous cell presence following H&E staining in the study and control groups, as well as the frequency of p63 presence after immunohistochemical staining, along with the statistical significance of intergroup comparisons

Frequency distributions of the examined parameters by groups				
Parameter		**Study group**	**Control group**	
p63	Positive	3	1	
	Negative	11	16	p=0.20
Keratin	Positive	6	1	
	Negative	8	16	p=0.014
Squamous cell	Positive	2	1	
	Negative	12	16	p=0.43
p63, keratin and squamous cell	Any one of the parameters is positive	6	2	
	All negative	8	15	p=0.049

H&E: Hematoxylin and eosin

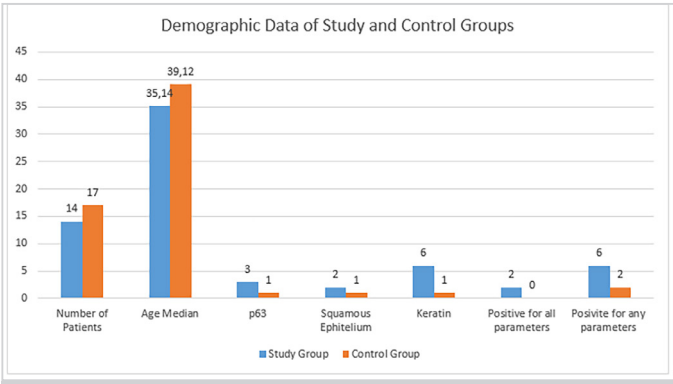

Figure 6. Comparison of demographic features and positivity rates for keratin, squamous cells, and p63 between study and control groups. Medians and positive case numbers are shown. Significant differences were found in keratin positivity and positivity for any parameter (p<0.01 and p<0.049, respectively)

(35.3%), with a median age of 39.12±14.581 years (Figure 6). Patient ages in this group ranged between 14 and 65 years. The groups were homogeneously distributed in terms of age and sex. No significant differences were observed between the groups regarding these two variables, with p-values of 0.70 and 0.41, respectively.

In the study group, 10 patients (79%) underwent CWD mastoidectomy, and four (21%) underwent CWU mastoidectomy out of a total of 14 patients. In the control group, CWD mastoidectomy was performed in five patients (29%), while CWU mastoidectomy was performed in 12 patients (71%) out of 17. A significant difference between the groups was noted (p=0.02). However, as there was no contamination during bone dust collection, we did not perform a multivariate analysis between the groups regarding surgical methods and group allocation.

In the study group, operations were performed on eight (57%) right ears and six (43%) left ears. In the control group, operations were performed on seven (41%) right ears and 10 (59%) left ears. No significant difference was found in terms of operation sides between the groups (p=0.38).

Upon evaluating the study group in terms of p63 IHC staining, while three patients (21.4%) out of 14 tested positive, 11 (78.6%) tested negative. In the control group, one patient (6%) tested positive, and 16 (94%) tested negative. There was no statistically significant difference between the groups (p=0.20, Table 1, Figure 6).

When analyzing the study group for keratin staining, six patients (43%) out of 14 had positive results, while eight (57%) had negative results. In the control group, one patient (6%) had positive staining, and 16 (94%) had negative results. Statistical analysis revealed a significant difference between the groups (p=0.01, Table 1, Figure 6).

In terms of squamous cell presence, while two patients (14%) in the study group were positive, 12 (86%) were negative. In the control group, one patient (6%) tested positive, and 16 (94%) tested negative. Statistical analysis showed no significant difference between the groups (p=0.43, Table 1, Figure 6).

If any assessed parameter (e.g., p63 IHC staining, keratin, or squamous cell presence) was positive, indicating metaplasia, six patients (43%) in the study group and two patients (12%) in the control group showed positivity for at least one parameter. Statistical analysis indicated a significant difference between the groups (p=0.049). Furthermore, two patients (14%) in the study group exhibited positivity for all parameters simultaneously, while no control group patients tested positive for all parameters (Table 1, Figure 6).

Discussion

Modern research focuses on molecular and stem cell studies to better understand the pathophysiology of cholesteatoma. In our study, we examined mastoid bone dust samples pathologically and IHC, hypothesizing that in cases where

cholesteatoma is limited to the middle ear, squamous epithelial tissue potentially undergoing metaplasia could be present in the mastoid air cell system.

In our study, a significant difference in the types of surgery performed was found between the study and control groups (p=0.02). Among the 14 patients in the study group, four underwent CWU mastoidectomy and 10 underwent CWD mastoidectomy, reflecting the more extensive surgeries required for COMC cases. To minimize epithelial contamination during CWD mastoidectomy, we avoided collecting samples while lowering the EEC's posterior wall. In CWU procedures, posterior wall incisions and drilling were deferred until the mastoid antrum was reached or mastoidectomy was completed. Consequently, mastoid bone dust collected during both types of procedures was obtained from the same sites at the same stages, eliminating contamination risk. Thus, the significant difference in surgery types did not affect study outcomes.

Cavity obliteration is a common modification in mastoid surgery, with recurrence rates of up to 19% reported in meta-analyses, possibly due to residual epithelial tissue or cholesteatoma. However, recent studies show lower recurrence in obliteration groups compared to CWU procedures, likely due to better disease clearance. While no studies have assessed bone dust for squamous epithelium or keratin in recidivism, our findings (14% squamous cell positivity, 43% keratin positivity) suggest a potential role. One study reported a 13.4% residual cholesteatoma rate with bone paté but did not confirm whether obliteration material contributed to recurrence (9).

Histological studies indicate that while all epithelial cells can synthesize keratin, only squamous transformation enables its secretion. Environmental factors and inflammation are key drivers of keratin accumulation, with disrupted homeostasis often linked to inflammatory processes (10). Supporting the metaplasia theory, Sadé et al. (2) detected keratin in 19 of 101 patients with limited cholesteatoma-and even behind intact, non-retracting tympanic membranes in 3.7% of cases. Similarly, Viswanatha et al. (11) reported three cases of cholesteatoma arising primarily in the mastoid air system without middle ear involvement.

In our study, H&E staining revealed keratin positivity in 43% of the study group versus 6% in the control group (p=0.01). Although no study has conclusively shown that keratin alone causes cholesteatoma, experimental models by Hinohira et al. (12,13) demonstrated that introducing keratin debris into epithelial cysts produced perimatrix-like granules, vascularization, and inflammatory changes, suggesting a contributory role for keratin in disease development. Notably, the absence of squamous cells or p63 staining in some keratin-positive samples may reflect sampling limitations, implying that additional sections might reveal a more complete picture of metaplastic changes.

Current theories do not fully explain the etiopathogenesis and clinical features of cholesteatoma, prompting further molecular investigation. p63, encoded by the TP63 gene, is a crucial transcription factor for epidermal development whose overexpression has been linked to squamous cell carcinomas (14). Studies have consistently shown that p63, localized in the basal layer of epithelial cells, is more prominently expressed in cholesteatoma tissues and may influence both keratin production and epithelial proliferation (15-17). Takahashi et al. (16)- Yamamoto-Fukuda et al. (17) demonstrated that keratinocyte growth factor-a key factor in cholesteatoma pathogenesis-upregulates p63, reinforcing its potential role in disease progression.

Our study is among the first to assess p63 expression in mastoid bone dust samples, offering unique insights into metaplastic changes within the mastoid air cell system that may precede cholesteatoma development. These findings highlight p63's potential as a biomarker for cholesteatoma progression and recurrence, and they open avenues for further research into its mechanistic role in this pathology.

Cholesteatoma development is widely associated with the presence of squamous cells in the middle ear, which produce keratin. However, our study revealed notable findings. Squamous cell staining was observed in two study group patients and one control, while p63 staining was positive in three study group patients and one control. Keratin positivity was more common, detected in six study group patients compared to one in the control group. Interestingly, some patients exhibited keratin without squamous cells, possibly due to sampling limitations in random sections after paraffin blocking.

When considering positivity for any parameter as an indicator of epithelial metaplasia, six study group patients and two controls tested positive, showing a statistically significant difference (p=0.049). However, simultaneous positivity for all parameters was rare, observed in only two study group patients, with none in the control group (p=0.11). These findings support the hypothesis that metaplasia can occur in mastoid air cell regions independent of cholesteatoma.

Given the potential recurrence risk in patients with any positive parameter, routine histopathological examination of mastoid bone dust using cost-effective H&E staining could help identify those requiring closer monitoring and advanced follow-ups.

A key limitation of our study is the small sample size, comprising 31 patients (14 in the study group and 17 in the control group). Larger cohorts are needed for more robust and generalizable results. Additionally, the lack of recent studies on this topic in the literature, despite older publications providing more extensive insights, highlights a

notable gap in contemporary research. This shift in research focus has limited comparisons with the current data, making it challenging to fully support our discussion. No support was received from any individual or organization for this study.

Conclusion

We evaluated p63 IHC staining, keratin, and squamous cells to identify squamous cell metaplasia in the mastoid air cell system. Our findings revealed significant differences between the study and control groups when positivity for any of these parameters was considered (p=0.049). These results indicate that the mastoid air cell system in patients with cholesteatoma undergoes alterations independent of middle ear cholesteatoma or its spread to the mastoid system.

Our findings strongly support the metaplasia theory, emphasizing its potential as a key mechanism in cholesteatoma pathogenesis. This challenges the traditional view that keratin presence is incidental and instead positions it as a possible contributing factor to recurrence risk.

Given the significant keratin positivity (p=0.01), routine histopathological examination of mastoid bone dust using H&E staining emerges as a cost-effective and time-efficient approach. This could facilitate the identification of high-risk patients requiring closer monitoring or advanced diagnostic assessments.

While our study is limited by a small sample size, the observed trends highlight the need for further research. Larger cohort studies should aim to validate these findings and elucidate the molecular mechanisms underlying squamous metaplasia and keratin accumulation. Moreover, exploring p63's role as a potential biomarker for cholesteatoma progression and recurrence could offer new insights into its invasive and recurrent nature.

In conclusion, our study underscores the importance of understanding the alterations in the mastoid air cell system to improve surgical outcomes and long-term patient management in cholesteatoma cases. Routine histopathological evaluation, alongside molecular investigations, could pave the way for tailored therapeutic approaches and enhanced recurrence prevention strategies.

Ethics

Ethics Committee Approval: Ethical approval was received from Dışkapı Yıldırım Beyazıt Research and Training Hospital (decision no: 90/18, dated: 22.06.2020).

Informed Consent: Informed consent was obtained from all patients or, in the case of minors, from their parents or legal guardians, both verbally and in writing, prior to surgery.

Authorship Contributions

Surgical and Medical Practices: H.O.O., M.D., Concept: H.O.O., M.D., Design: H.O.O., M.D., T.T.T., Data Collection and/or Processing: H.O.O., T.T.T., Analysis and/or Interpretation: H.O.O., T.T.T., Literature Search: H.O.O., M.D., Writing: H.O.O., M.D., T.T.T.

Conflict of Interest: The authors declare that they have no conflict of interest.

Financial Disclosure: The authors declare that this study has received no financial support.

Main Points

- We found that squamous epithelial tissue or fragments, such as keratin, thought to undergo metaplasia within the mastoid air cell system could be a reason for recurrences in cases where cholesteatoma is limited to the middle ear.
- In this introductory study we aimed to investigate whether mastoidectomy dust samples should routinely undergo histopathological evaluation using hematoxylin and eosin staining alone, with a focus on cost-effectiveness and time efficiency. The goal is to identify patient groups who may require more frequent or advanced follow-up examinations.
- The study highlights the potential role of p63 as a biomarker for cholesteatoma progression, suggesting that its expression in mastoid bone dust samples could indicate early metaplastic changes and aid in risk assessment for recurrence.

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Evaluation of Bone-Anchored Hearing Instruments in Chronic Otitis Media: A Multicenter Prospective Study

Original Investigation

Fazıl Necdet Ardıç[1], Mehmet İhsan Gülmez[2], Şemsettin Okuyucu[3], Mehmet İlhan Şahin[4], Ayça Başkadem Yılmazer[5], Ömer Afşin Özmen[6], Mehmet Ekim[6], Ece Altınöz Baykal[1], Emel Aslantaş[7], Serap Uçar[8], Ali Tanrıöven[5], Mahmut Tayyar Kalcıoğlu[7]

[1]Pamukkale University Faculty of Medicine, Department of Otorhinolaryngology, Denizli, Türkiye
[2]Hatay Mustafa Kemal University Faculty of Medicine, Department of Otorhinolaryngology, Hatay, Türkiye
[3]Medicana International Hospital, Department of Otorhinolaryngology, Ankara, Türkiye
[4]Erciyes University Faculty of Medicine, Department of Otorhinolaryngology, Kayseri, Türkiye
[5]University of Health Sciences Türkiye, Prof. Dr. Cemil Taşçıoğlu Hospital, Department of Otorhinolaryngology, İstanbul, Türkiye
[6]Uludag University Faculty of Medicine, Department of Otorhinolaryngology, Bursa, Türkiye
[7]İstanbul Medeniyet University Göztepe Süleyman Yalçın City Hospital, Department of Otorhinolaryngology, İstanbul, Türkiye
[8]Erciyes University Halil Bayraktar Vocational School of Health Sciences, Department of Otorhinolaryngology, Kayseri, Türkiye

Abstract

Objective: This study aims to assess the effectiveness of bone-anchored hearing instruments (BAHIs) in patients with chronic otitis media (COM), using objective and subjective measures. It is a multicenter, prospective trial involving patients with COM who have undergone surgical treatment and have been rehabilitated using BAHIs.

Methods: COM Questionnaire-12 (COMQ-12), Speech, Spatial, and Qualities of Hearing Scale (SSQ), World Health Organization (WHO) Quality of Life-BREF questionnaire, and audiometric tests were used for assessment.

Results: Twenty-eight patients were included. The average duration of COM was 20.1±13.32 years. Among the patients, 60.7% (17) were using hearing aids, with a mean usage duration of 10.8±10.7 years (ranging from 1 to 36 years). Seven patients received the PONTO device, two received the BAH aid (BAHA) connect system, and 19 were implanted with the BAHA attract system. COMQ-12, SSQ, WHO questionaries, and audiometric tests showed significant improvement, and the results were found stable during follow-up.

Conclusion: This study reinforces the effectiveness of BAHIs in improving hearing thresholds and quality of life for patients with COM.

Keywords: Chronic otitis media, hearing loss, bone-anchored hearing aids, hearing rehabilitation, quality of life

ORCID IDs of the authors:

F.N.A. 0000-0003-4230-3141
M.İ.G. 0000-0003-0462-6353
Ş.O. 0000-0001-8552-2403
M.İ.Ş. 0000-0002-9576-1448
A.B.Y. 0000-0001-9967-8046
Ö.A.Ö. 0000-0002-9698-0546
M.E. 0000-0003-1745-3711
E.A.B. 0000-0002-4163-9586
E.A. 0000-0001-6460-7459
S.U. 0000-0002-0511-1928
A.T. 0000-0001-9478-7219
M.T.K. 0000-0002-6803-5467

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Corresponding Author:
Fazıl Necdet Ardıç, Prof. MD;
fnecdetardic@gmail.com

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Introduction

Chronic otitis media (COM) is one of the most common ear conditions characterized by symptoms such as ear fullness, tinnitus, pruritus, otalgia, recurrent ear discharge, and, most significantly, hearing loss (1). In addition to the social burden of hearing impairment, patients also face the risk of chronic infections in the cranial bone. The global incidence of COM is 4.76% (2). It is estimated that 31 million new cases are diagnosed every year (3). Most of these are in less developed countries and one-fifth are children aged under five years (3). Typically, surgery is recommended to most patients to eradicate infection and restore hearing function. Approximately 70% of surgical cases were successful, with patients achieving an air-bone gap of less than 20 dB. However, 29% of patients had an air-bone gap greater than 20 dB (4). Bone conduction thresholds are also crucial, as these patients are at risk of developing sensorineural hearing loss (5). These two factors together indicate that a significant proportion of patients require hearing rehabilitation.

The first-line option for hearing rehabilitation is the use of hearing aids. Although patient compliance with hearing aid usage has improved, many individuals with hearing loss still do not use their hearing aids (6). Thirty-eight percent of patients were reported as a non-user in a recent meta-analysis (7). The reasons were lack of awareness of their condition, low perceived benefits, finding the device uncomfortable, social stigma, insufficient income, lack of social support and older age. Additionally, patients with COM, even those who have been adequately treated, may experience challenges such as acoustic feedback, improper fitting molds, background noise, sound distortion, discomfort in the ear canal, irritation, infection, and persistent discharge. Bone-anchored hearing instruments (BAHIs) offer a valuable alternative for these individuals. It is reported that there was no significant difference between hearing aids and BAHIs on audiometric test parameters (8).

This study aims to assess the effectiveness of BAHIs in patients with COM using objective and subjective measures.

Methods

This study is a multicenter, prospective trial involving patients with COM who have undergone surgical treatment and have been rehabilitated using BAHIs. Written informed consent was obtained from all participants or, in the case of pediatric patients, from their parents or legal guardians, and ethical approval was granted by the Pamukkale University Non-Interventional Clinical Research Ethics Committee (number: 14, date: 28.07.2020). The study is registered at clinicaltrials.gov, NCT06047639.

The observed effect size in a reference study was strong (dz=2.447). A power calculation showed that including a minimum (min.) of 24 participants would yield a study power of 80% at a 95% confidence level (9).

All patients were recruited from the otolaryngology departments. After careful ear examination, computed tomography, pure tone audiometry, and speech discrimination tests were performed. The subjects who volunteered to join the study completed the Clinical Service Recipient Inventory (18 questions) (10).

Inclusion Criteria:

- Adults
- No difficulty in attending follow-up visits
- Sufficient communication skills to interact with researchers
- Indication for BAHIs due to COM

Exclusion Criteria:

- Severe systemic illnesses (e.g., cancer, human immunodeficiency virus)
- Patients with compliance issues regarding hearing tests
- Use of medications or medical devices that could interfere with the study outcomes
- Inability to adhere to regular follow-ups or non-compliance with device usage

Parameters

The following tools were used for the subjective evaluation of BAHIs: the COM Questionnaire-12 (COMQ-12), the Speech, Spatial, and Qualities of Hearing Scale (SSQ) (48 questions), and the World Health Organization (WHO) Quality of Life-BREF questionnaire (27 questions) (11,12).

Surgical notes and middle ear risk index were also recorded (13). Hearing performance was measured using pure tone audiometry, speech discrimination scores (%), and free-field audiometry. The mean of 500, 1000, 2000, and 4000 Hz was used to compare pure tones.

All assessments were conducted preoperatively and postoperatively in the third and twelfth months. All different brands of bone-anchored devices were accepted. The specific surgical manuals for every device were used for surgical implantation. Transcutaneous [PONTO (Oticon Medical) and BAHA connect (Cochlear Inc.)] or percutaneous [BAHA attract (Cochlear Inc.)] were included.

Statistical Analysis

Data were analyzed using IBM SPSS Statistics for Windows version 25.0. Continuous variables were reported as mean ± standard deviation, median (25^{th} and 75^{th} percentiles), and min.-max. values, while categorical variables were presented as frequency and percentage. The Shapiro-Wilk test was used to study the normality of the data distribution.

If parametric test assumptions were met, the independent sample t-test was applied for comparisons between independent groups. Otherwise, the Mann-Whitney U test was used. For comparisons within dependent groups, the paired sample t-test and repeated measures analysis of variance (with post hoc Bonferroni correction) were employed when parametric assumptions were met; otherwise, the Wilcoxon signed-rank test and the Friedman test were used. A p-value of <0.05 was considered statistically significant.

Results

A total of 28 patients, 16 male and 12 female, participated in the study. Their average age was 43.46±13.9 years, ranging from 11 to 64 years. The average duration of (COM was 20.1±13.32 years. Of the patients, 60.7% (17) were using hearing aids, with a mean usage duration of 10.8±10.7 years (ranging from 1 to 36 years).

Recurrent infections were reported in 72% of the patients, and 84% had middle ear problems. Additionally, 45% had other chronic conditions such as hypertension or diabetes, and 76% had undergone multiple ear surgeries. On average, patients visited their physician 3.42±3 times in the past six months and missed 2.29±6.73 workdays over the same period.

Regarding the implants, seven patients received the PONTO device (Oticon Medical), two patients received the BAHA Connect system (Cochlear Inc.), and 19 patients the BAHA Attract system (Cochlear Inc.). Fifteen patients had the implant on the left side and 13 on the right side.

All patients completed the COMQ-12 questionnaire during follow-up. Clinical improvements COM were observed in the 3rd month (p<0.05), with further improvements noted by the 12th month (p<0.01). There was no statistically significant difference between the 3rd- and 12th-month evaluations (p>0.05) (Figure 1).

According to the results of the WHO questionnaire, there was significant improvement in the general health of the patients (p<0.01), though other parameters showed no significant changes (Table 1).

The SSQ showed marked improvements across all measured parameters, with significant increases observed and sustained over the 12 months (p<0.001) (Table 1, Figure 2).

Audiometric evaluations revealed that all patients had benefitted from the implant. A statistically significant improvement was found when the mean air conduction threshold of the implanted side was compared with the mean postoperative free-field audiometric thresholds (p<0.001). This improvement persisted at the 12-month follow-up (p<0.001), with no significant difference between the 3rd and 12th-month evaluations (p>0.05) (Figure 3).

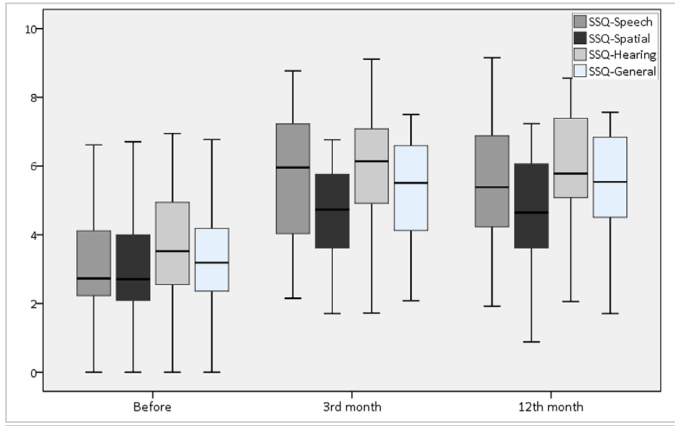

Figure 2. Changes in the Speech, Spatial, and Quality of Hearing Scale parameters were observed during the follow-up. There was a statistically significant change (p<0.001) in all parameters in the 3rd month, and it was stable during the 12 months

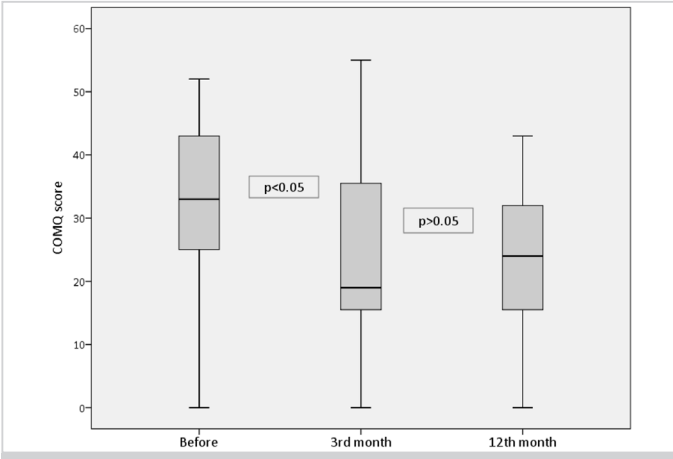

Figure 1. Chronic Otitis Media Questionnaire-12 continued to improve during the follow-up. There was a statistically significant decrease in the 3rd month (p<0.05) and the 12th month (p<0.01). But there was no difference between the 3rd and 12th months (p>0.05)

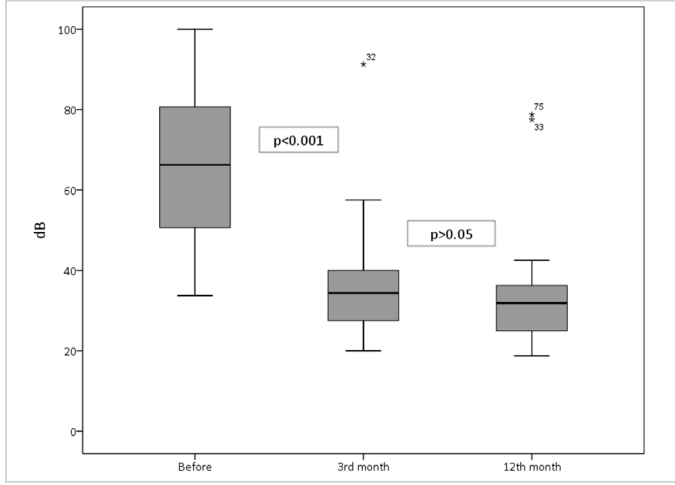

Figure 3. The mean air conduction threshold measured before the operation was compared with the mean free field thresholds after the implantation

Table 1. WHO Quality of Life-BREF Questionnaire was filled at the study's beginning and end. Speech, Spatial, and Qualities of Hearing Scale and audiometric tests were done before implantation and 3rd and 12th months after the operation

	Before			3rd month			12th month			
	Mean±SD	Median (IQR)	Min.-max.	Mean±SD	Median (IQR)	Min.-max.	Mean±SD	Median (IQR)	Min.-max.	p-value
WHO-general	40.62±24.44	43.75 (25)	0-100				59.72±22.28	62.5 (25)	12.5-100	0.001
WHO-physical	58.16±11.81	58.92 (10.7)	25-82.1				58.2±14.24	57.15 (25)	35.7-85.7	0.77
WHO-psychologic	58.33±15.12	58.3 (15.6)	20.8-83.3				60.03±15.64	58.3 (16.6)	25-100	0.44
WHO-social	55.95±22.32	58.3 (31.3)	16.7-100				62.96±23.71	66.66 (33.33)	8.3-100	0.15
WHO-environmental	60.6±16.6	62.5 (21.1)	25-96.9				66.66±12.73	65.62 (25)	46.9-90.6	0.14
COMQ-12	32.54±13.52	33 (19)	0-52	23.57±15.75	19 (55)	0-55	23.22±11.52	24 (43)	0-43	0.01
SSQ-speech	3.02±1.45	2.73 (1.9)	0-6.6	5.7±1.85	5.96 (3.32)	2.15-8.8	5.43±1.94	5.38 (3)	1.9-9.2	0.001
SSQ-spatial	3.01±1.53	2.7 (1.9)	0-6.7	4.67±1.4	4.73 (2.25)	1.7-6.8	4.73±1.64	4.64 (2.7)	0.9-7.2	0.001
SSQ-hearing	3.66±1.82	3.52 (2.4)	0-6.9	5.9±1.86	6.14 (2.27)	1.72-9.1	5.93±1.64	5.77 (2.44)	2.1-8.6	0.001
SSQ-total	3.26±1.51	3.19 (1.9)	0-6.8	5.41±1.57	5.51 (2.56)	2.08-7.5	5.37±1.58	5.54 (2.48)	1.7-7.6	0.001
Mean AC (dB)	66.38±17.44	66.25 (30.6)	33.8-100	65.58±18.18	66.25 (25)	30-100	65.69±17.09	653.75 (23.75)	32.5-100	0.97
Mean BC (dB)	30.52±16.09	27.5 (21.9)	3.8-67.5	31.65±15.61	29.37 (21.56)	6.3-61.3	32.45±16.21	32.5 (23.75)	7.5-63.8	0.90
FF w/implant (dB)				31.35±6.97	32.5 (12.5)	21.3-41.3	28.75±7.45	29.37 (15.63)	18.8-37.5	
WD w/o implant (%)				90±9.7	90 (19)	76-100	87.6±14.5	90 (19)	56-100	
WD w/implant (%)				96.8±3.1	96 (5)	92-100	96±4.2	96 (8)	88-100	

WHO: World Health Organization, SSQ: Speech, Spatial, and Qualities of Hearing Scale, AC: Air Conduction threshold of implanted side and BC: Bone conduction threshold of implanted side, FF: Free field audiometry, WD: Word discrimination, COMQ-12: Chronic Otitis Media Questionnaire-12, w/: with, w/o: without, Min.: Minimum, Max.: Maximum, SD: Standard deviation, IQR: Interquartile range.

There were no statistical differences between transcutaneous and percutaneous instruments regarding free-field implant gain and SSQ parameters (p>0.05).

Discussion

This study's findings show the positive impact of BAHIs in patients with COM. With an average duration of 20 years living with COM, the patient cohort reflects the challenges faced by those who suffer from recurrent infections, middle ear problems, and other chronic conditions. Notably, 60.7% of the patients had been using hearing aids, yet many still experienced limitations in hearing function, suggesting a need for more effective interventions like BAH systems.

These results align with previous studies that reported similar improvements in patients with conductive or mixed hearing loss using bone-anchored devices. The stability of these improvements between the 3rd and 12th months (p>0.05) further highlights the long-term effectiveness of BAHIs in improving quality of life. Lewis et al. (14) also reported improved both hearing and health-related quality of life.

The WHO questionnaire results indicated significant improvement in the general health of the patients (p<0.01), although other parameters did not show statistical changes. This can be attributed to the fact that the scope of BAHIs is primarily auditory. Nevertheless, improved hearing often leads to better communication, social interaction, and emotional well-being, which may explain the overall improvement in the general health perception of the patients. Twenty-seven patients were asked about their preference after using BAHIs for seven years, and 89% of them stated to prefer BAHIs over hearing aids (15). This preference for BAHIs over air-conduction hearing aids was reported as 58% in 1996 (16). In a follow-up study, 34 patients were asked about their BAHI and previous air-conduction hearing experiences. Majority of them stated to prefer BAHIs over air-conduction hearing aids; albeit the preference was primarily influenced by the decrease in ear infections rather than improved speech recognition (17). This finding was supported by significantly less hospital visits during a five-year follow-up period in COM patients using BAHIs (18).

The SSQ results were highly encouraging, demonstrating substantial improvement across all parameters at both the 3rd and the 12th months ($p<0.001$). The result was comparable with previous studies (14). This highlights that patients experienced better sound localization, speech understanding in various environments, and overall sound quality. This improvement was also reported by the partners of the patients (19).

Audiometrically, all patients showed measurable improvements in hearing thresholds post-implantation. The significant decrease in air conduction thresholds, evident in free-field audiometry at both the 3rd and the 12th months ($p<0.001$), provides strong support for the functional benefits of BAHIs. Notably, the absence of significant differences between the 3rd and the 12th months suggests that the auditory benefits offered by the implants are immediate and stable over time.

There was no statistical difference between transcutaneous or percutaneous instruments on free field implant gain and SSQ parameters ($p>0.05$) in our study. Pure tone thresholds at 3 kHz and 4 kHz were reported to be better in percutaneous systems than in transcutaneous systems. This advantage did not affect sentence recognition in silence but was effective in noise (20). On the other hand, patients with percutaneous systems made more visits to outpatient clinics (21).

Cost-effectiveness arguments come to the fore when deciding the indication and selecting the implant type in patients with COM. Since COM patients using implants were observed not to benefit from conventional hearing aids, it is meaningless to discuss the economic issues between hearing aids and BAHIs. Comparison of different implants has nevertheless been a popular topic in the literature. Transcutaneous implants were reported as more cost-effective than the percutaneous implants (22). But the cost effectiveness of the latter became comparable in the long-term follow-up (23). Recent advancements in technology are likely to take this discussion further. Novel active implants will be a breakthrough in the hearing rehabilitation of COM patients (24). Especially better outcomes at higher frequencies compared to passive implants may increase patient compliance to BAHIs (25).

Ostevik et al. (26) showed that open-fit hearing aids could be an alternative solution in patients with mild to moderate conductive hearing loss who were unwilling to undergo a BAHI surgery.

Our study's strength lies in its longitudinal follow-up and use of multiple assessment tools, including the COMQ-12, WHO questionnaire, and SSQ, to comprehensively assess the impact of BAHIs on clinical symptoms and quality of life. However, some limitations should be acknowledged. The relatively small sample size and the predominance of BAHA Attract users (19 out of 28) may limit the findings' generalizability to other BAHI systems.

Conclusion

This study supports the effectiveness of BAHIs in improving hearing thresholds and quality of life in patients with COM. The significant clinical improvements, enhanced auditory perception, and stable long-term results suggest that BAHIs are valuable intervention strategy in patients who may not fully benefit from conventional hearing aids. Further research with larger cohorts and diverse BAHI systems are warranted to confirm these findings and explore the broader implications of BAHIs in improving overall patient well-being. Large-scale studies assessing the efficacy of BAHI s across various demographic groups are needed. Long-term assessments of device-related complications based on device type should be conducted.

Ethics

Ethics Committee Approval: This study was approved by the Pamukkale University Non-Interventional Clinical Research Ethics Committee (number: 14, date: 28.07.2020).

Informed Consent: Written informed consent was obtained from all participants.

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We remember with respect our colleague and expected co-writer of this article, Assoc. Prof. Elif Tuğba Saraç, Ph.D., died in an earthquake on 6th February 2023.

Footnotes

Authorship Contributions

Surgical and Medical Practices: F. N.A., M. İ. G., Ş.O., M.İ.Ş., A.B.Y., Ö.A.Ö., M.T.K., Concept: F.N.A., M.İ.G., Ş.O., M.İ.Ş., A.B.Y., Ö.A.Ö., M.E., E.A., S.U., A.T., M.T.K., Desing: F.N.A., M.İ.G., Ş.O., M.İ.Ş., A.B.Y., Ö.A.Ö., M.E., E.A., M.T.K., Data Collection and/or Processing: F.N.A., M.İ.G., Ş.O., M. İ.Ş., A.B.Y., Ö.A.Ö., M.E., E.A., S.U., A.T., M.T.K., Analysis and/or Interpretation: F. N.A., Literature Search: F. N.A., Writing: F. N.A.

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Main Points

- Hearing rehabilitation is essential in chronic otitis media.
- Bone-anchored hearing aids are good alternatives.
- They improved hearing performance and quality of life significantly.
- This effect was stable for 12 months.

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Is There a Relationship Between Tonsillolith Formation and Nasal Septal Deviation or Chronic Sinusitis, with or without Nasal Polyps?

Original Investigation

Hasan Yasan[1], Mehmet Emre Sivrice[1], Yusuf Çağdaş Kumbul[1], Bekir Büyükçelik[2], Mustafa Kayan[3]

[1]Süleyman Demirel University Faculty of Medicine, Department of Otorhinolaryngology and Head and Neck Surgery, Isparta, Türkiye
[2]Isparta City Hospital, Clinic of Otorhinolaryngology and Head and Neck Surgery, Isparta, Türkiye
[3]Süleyman Demirel University Faculty of Medicine, Department of Radiology, Isparta, Türkiye

Abstract

Objective: The aim of this study is to assess the prevalence of tonsilloliths and investigate the potential role of nasal septal deviation (NSD) and/or chronic rhinosinusitis (CRS), with or without nasal polyps, in their development.

Methods: A retrospective analysis was conducted on computed tomography (CT) images of 3,516 patients obtained between January 2017 and December 2020. The presence of tonsilloliths was recorded along with NSD and CRS, with or without nasal polyps. The effects of age, gender, NSD, and CRS (with or without nasal polyps) on tonsillolith formation were analyzed. All CT images were re-evaluated for tonsilloliths, CRS, NSD and other pathological findings. The relationships between tonsillolith presence and NSD and CRS were statistically analyzed.

Results: A significant difference in age was observed between individuals with and without tonsillolith ($p<0.001$). Additionally, the prevalence of tonsillolith was markedly higher in patients diagnosed with NSD compared to those without this condition ($p<0.001$). Similarly, patients diagnosed with CRS exhibited a significantly increased incidence of tonsillolith when compared to individuals without CRS ($p<0.001$).

Conclusion: The incidence of tonsilloliths in this study was 27.6%. Tonsillolith prevalence increased with age, while no significant difference was observed based on gender. The presence of NSD, a condition associated with nasal obstruction, or CRS, an inflammatory disorder, was linked to a higher incidence of tonsilloliths.

Keywords: Tonsillolith, nasal polyps, rhinosinusitis, nasal septum, tomography

ORCID IDs of the authors:
H.Y. 0000-0002-5470-6784
M.E.S. 0000-0002-2396-6794
Y.Ç.K. 0000-0002-0713-2933
B.B. 0000-0002-3620-2443
M.K. 0000-0001-5255-7132

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Corresponding Author:
Hasan Yasan, MD;
dryasan@hotmail.com

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Introduction

Tonsilloliths (TLs) are white- or yellow-calcified concretions within the palatine tonsils (1). They are composed of phosphate and/or carbonate salts along with calcium and a protein matrix. Although TLs are thought to form as a result of recurrent tonsillitis, the exact mechanism remains unclear (2). Several studies in the literature suggest a relationship between TL formation and inflammatory conditions such as tonsillitis and periodontal diseases (2,3).

If an association exists between inflammation and TLs, it is reasonable to consider the potential impact of other upper respiratory tract inflammatory conditions, such as sinusitis. However, the roles of sinonasal inflammatory diseases and nasal obstruction in the etiology of TLs have not been investigated. This study aims to explore the possible role of chronic rhinosinusitis with/without nasal polyps (CRS and CRSwNP), in TL formation. Additionally, we sought to evaluate the potential influence of nasal obstruction, specifically nasal septal deviation (NSD), on TL development.

Methods

Study Design

Computed tomography (CT) images of 3,516 patients obtained between January 2017 and December 2020 were analyzed and 3,259 images met the inclusion criteria for this study. The study was conducted at Süleyman Demirel University Research and Training Hospital and was approved by the Ethical Committee for Clinical Studies of Süleyman Demirel University (date: 12.02.2021, number: 75). Additional informed consent was not required, as patients had already been informed that their radiographic imaging data could be used for future research purposes.

CT scans were conducted using a 128-slice CT scanner (Somatom Definition; Siemens Healthcare, Forchheim, Germany), generating approximately 120-180 images per study. Imaging parameters included a tube voltage of 80 kVp, a tube current of 120 mA, a slice thickness and reconstruction interval of 1 mm, a pitch factor of 1, a matrix size of 512×512, and a field of view of 14cm×17cm. Window settings were defined as a width of 2000 Hounsfield unit (HU) and a level of 400 HU. Coronal and sagittal plane images were reconstructed using a dedicated workstation.

The indications for CT evaluation included suspected rhinologic headache, chronic and/or acute sinusitis with or without complications, NP, sinonasal neoplasia, and preoperative assessment for rhinoplasty and/or nasal reconstruction. However, patients with a history of multiple CT scans, maxillofacial trauma, sinonasal neoplasia, prior sinonasal surgery, congenital anomalies, significant anatomic variations obstructing the nasal passages (e.g., large concha bullosa) or previous adeno/tonsillar surgery were excluded from the study.

After reviewing each patient's medical records, paranasal sinus CT data were independently assessed by an otolaryngologist and a radiologist in a blinded manner. The results were compared, and discrepancies were resolved through joint re-evaluation before finalizing the findings. The presence and laterality of NSD, the presence and laterality of chronic sinusitis, and the presence and laterality of NP were systematically recorded. Additionally, the presence and the number of TLs were documented. Following CT scan evaluation, findings were cross-checked with endoscopic nasal examination reports from patients' medical records. The relationship between TLs and the presence and laterality of NSD and CRS was then analyzed (Figures 1,2).

Statistical Analysis

Statistical analyses were done using SPSS version 24.0 (IBM Corp. IBM SPSS Statistics for Windows, Version 24.0. Armonk, NY). Categorical variables were summarized as frequencies and percentages, while continuous variables

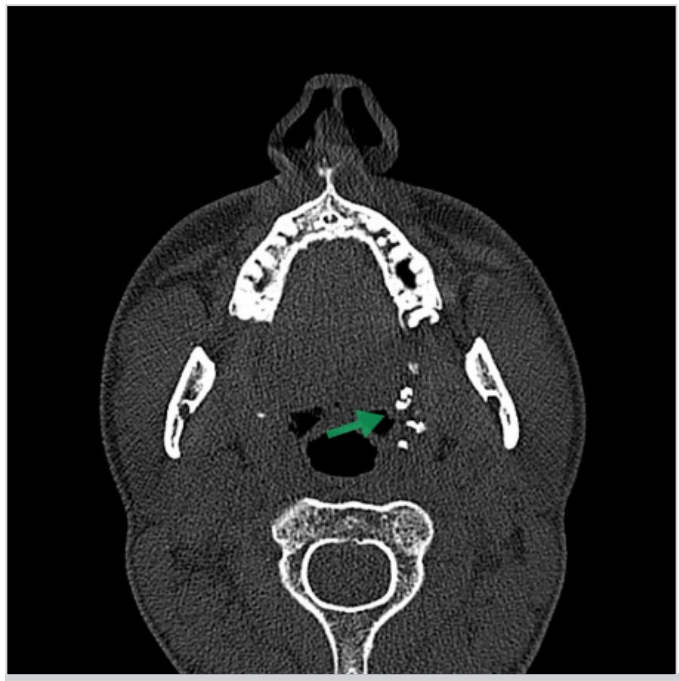

Figure 1. Axial CT image of multiple TLs in the left palatine tonsil (axial)
CT: Computed tomography, TLs: Tonsillolithiasis

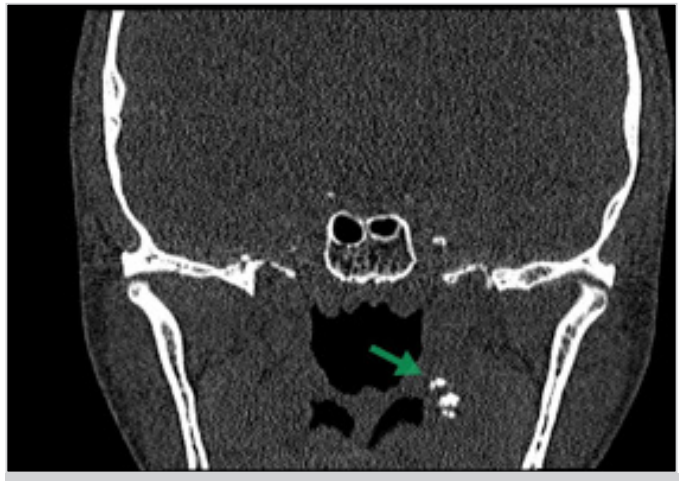

Figure 2. CT image of multiple TLs in the left palatine tonsil (coronal)
CT: Computed tomography, TLs: Tonsillolithiasis

were expressed as mean±standard deviation, along with their minimum and maximum values. The independent samples t-test was employed to assess differences in continuous variables, whereas the chi-square test was used for categorical data. To investigate the effect of nasal pathologies on the presence of TLs, logistic regression analysis was done within a multivariate logistic model. Results were reported as odds ratios (ORs) with 95% confidence intervals (CIs), and a p-value <0.05 was considered statistically significant.

Results

This study included paranasal sinus CT scans from 3,259 patients, namely, 1,411 females (43.3%) and 1,848 males (56.7%). The mean age of the patients was 38.85±15.84 years (range=18-91). Patients with TL had a mean age of 42.39±16.03 years, while those without TL had a mean age of 37.50±15.56 years. A statistically significant difference in age was observed between patients with and without TL (p<0.001); however, no significant difference was found in terms of gender (p=0.177). TL was absent in 2,358 patients (72.4%), while it was present on the right side in 312 patients (9.6%), on the left side in 301 patients (9.2%), and bilaterally in 288 patients (8.8%). In total, 901 patients (27.6%) presented with TL. Table 1 summarizes the relationship between TL and variables such as age and gender. The average number of TLs per patient was 2.63±2.18 (range=1-18).

The distribution of NSD among patients was as follows: 1,131 patients (34.7%) had no NSD, 896 patients (27.5%) had right-sided NSD, 845 patients (25.9%) had left-sided NSD, and 387 patients (11.9%) had bilateral NSD. In total, 2,128 patients (65.3%) had NSD. While 617 (29%) of 2,128 patients with NSD had TL, 284 (25.1%) of 1,131 patients without NSD had TL. The incidence of TL was significantly higher in patients with NSD than in those without (p<0.001). The presence of TL was 1.3 times more common in patients with NSD than in those without NSD (OR=1.302, p=0.002, 95% CI=1.102-1.538). Additionally, in patients with NSD, the side of deviation tended to correspond with the side of TL (Tables 2,3).

Of the total patients, 2,278 (69.9%) did not have CRS, 136 (4.2%) had right-sided CRS, 173 (5.3%) had left-sided CRS, and 672 (20.6%) had bilateral CRS. In total, 981 patients (30.1%) had CRS. While 342 (34.9%) of 981 patients with CRS had TL, 559 (24.5%) of 2278 patients without CRS had TL. The incidence of TL was significantly higher in patients with CRS than in those without (p<0.001). The presence of TL was 1.58 times more common in patients with CRS than in those without CRS (OR=1.588, p<0.001, 95% CI=1.333-1.892). Furthermore, in patients with CRS, the side of CRS tended to correspond with the side of TL (Tables 2,3).

While 90 (38.5%) of 234 patients with CRSwNP had TL, 811 (%26.8) of 3,025 patients without CRSwNP had TL. The incidence of TL was significantly higher in patients with CRSwNP than in those without (p<0.001). The incidence of TL was 1.35 times more common in patients with CRSwNP than in non-CRSwNP patients (OR=1.588, p<0.001, %95 CI=1.333-1.892). In patients with CRSwNP, the side with CRSwNP tended to be the same as the side with TL (Tables 2,3).

Discussion

TLs are calcifications, either single or multiple, within the palatine tonsils. Although they are typically asymptomatic, they can sometimes cause significant symptoms, occasionally necessitating tonsillectomy. TLs are known to be composed of phosphate and/or carbonated salts of calcium and a protein matrix. Potential symptoms associated with TLs include halitosis, cough, and throat discomfort. The incidence of TL has been reported in a wide range in the literature, varying from 0.43% to 30.3% across different studies (2,4-7). These discrepancies may be attributed to differences in evaluation

Table 1. The relationship of TL with age and gender

	TL (-) (n=2358)	TL (+) (n=901)	p-value
Female (n=1411)	1038 (73.6%)	373 (26.4%)	0.177
Male (n=1848)	1320 (71.4%)	528 (28.6%)	
Age	37.50±15.56	42.39±16.03	<0.001*

*Statistically significant, TL: Tonsillolith

Table 2. The relationship between TL and NSD/paranasal sinus pathologies

	TL (-)	TL (+)	p-value	OR (95% CI)	p-value
Nasal septal deviation (R)					
(-): (n=1131)	847 (74.9%)	284 (25.1%)	0.018*	1.302	0.002*
(+): (n=2128)	1511 (71%)	617 (29%)		1.102-1.538	
CRS with/without nasal polyps (R)					
(-): (n=2278)	1719 (75.5%)	559 (24.5%)	0.001*	1.588	<0.001*
(+): (n=981)	639 (65.1%)	342 (34.9%)		1.333-1.892	
CRS with nasal polyps (R)					
(-): (n=3025)	2214 (73.2%)	811 (26.8%)	0.001*	1.351	0.048*
(+): (n=234)	144 (61.5%)	90 (38.5%)		1.003-1.820	

The aspect of TL and nasal pathology tend to be on the same side

*Statistically significant, R: reference variable, NSD: Nasal septal deviation, OR: Odds ratio, CRS: Chronic rhinosinusitis, TL: Tonsillolith, CI: Confidence interval

Table 3. The directional relationship between TL and nasal pathologies

Nasal septal deviation	TL absent	TL right	TL left	TL bilateral	p-value
NSD (-) (n=1131)	847 (74.9%)	96 (8.5%)	81 (7.2%)	107 (9.5%)	
Right sided (+) (n=896)	662 (73.9%)	116 (12.9%)	64 (7.1%)	54 (6%)	
Left sided (+) (n=845)	602 (71.2%)	64 (7.6%)	118 (14%)	61 (7.2%)	<0.001*
Bilateral (+) (n=387)	247 (63.8%)	36 (9.3%)	38 (9.8%)	66 (17.1%)	
CRS with/without nasal polyps					
CRS (-) (n=2278)	1719 (75.5%)	201 (8.8%)	193 (8.5%)	165 (7.2%)	
Right sided (+) (n=136)	86 (63.2%)	33 (24.3%)	7 (5.1%)	10 (7.4%)	
Left sided (+) (n=173)	112 (64.7%)	10 (5.8%)	33 (19.1%)	18 (10.4%)	<0.001*
Bilateral (+) (n=672)	441 (65.6%)	68 (10.1%)	68 (10.1%)	95 (14.1%)	
CRS with nasal polyps					
CRSwNP (-) (n=3025)	2214 (73.2%)	289 (9.6%)	269 (8.9%)	253 (8.4%)	
Right sided (+) (n=12)	6 (50%)	5 (41.7%)	0 (0%)	1 (8.3%)	
Left sided (+) (n=11)	8 (72.7%)	0 (0%)	3 (27.3%)	0 (0%)	<0.001*
Bilateral (+) (n=211)	130 (61.6%)	18 (8.5%)	29 (13.7%)	34 (16.1%)	

Italic characters indicate the direction in which the presence of TL is highest, according to the direction of nasal pathology

*Statistically significant, TL: Tonsillolith, NSD: Nasal septal deviation, CRS: Chronic rhinosinusitis, CRSwNP: Chronic rhinosinusitis with/without nasal polyps

techniques (e.g., slice thickness in imaging), racial variations, and even dietary habits. The case numbers, inclusion and exclusion criteria of the studies can also be a reason for these differences. In our study, the incidence of TL was 27.6%. This value is within the range given in the literature and our study is also one of the largest series. TLs are a multifactorial condition that has gained increasing public attention, even on social media platforms (7).

Several studies in the literature have investigated the association between TLs and various factors. One such factor is patient age. There are contradictory results in the literature about the effect of patients' age. While Fauroux et al. (8) reported no significant association, Aragoneses et al. (9) found a higher prevalence of TLs in younger individuals, whereas Oda et al. (3) observed a greater incidence in older patients.

In our study, the mean age of participants was 38.85±15.84 years (range=18-91). Patients with TLs had a mean age of 42.39±16.03 years, whereas those without TLs had a mean age of 37.50±15.56 years. This age difference was statistically significant (p<0.001), indicating that TLs are more prevalent in older individuals.

Regarding the relationship between TL prevalence and gender, conflicting results exist in the literature. While some studies suggest that TLs are more common in women, others report a higher incidence in men (6,9,10). Additionally, several studies have found no significant gender-based difference in TL prevalence (3,8,11). In our study, 26.4% of female and 28.6% of male patients had TLs (p=0.177).

Aragoneses et al. (9) also studied racial differences in TL prevalence and reported that TLs were less common in Black individuals compared to Caucasian and Asian populations. However, our study did not assess racial differences.

It is known that TLs are formed by phosphate and/or carbonated salts of calcium and a protein matrix. While they are believed to result from recurrent tonsillitis, the exact mechanism of their formation remains unclear (2). TLs are also considered to be associated with chronic cryptic tonsillitis (12). Additionally, there is evidence suggesting a relationship between TLs and periodontal disease (3). If inflammation plays a role in the formation of TLs, it is plausible that other upper respiratory tract inflammations, such as sinusitis, could contribute to their development.

Among the 981 patients with CRS, 342 (34.9%) had TLs, compared to 559 (24.5%) of the 2,278 patients without CRS (p<0.001). In patients with CRS, the side affected by CRS was often the same side as the TL. Furthermore, TLs were found to be 1.35 times more common in patients with CRSwNP compared to those without CRSwNP, with a similar tendency for TLs to occur on the same side as CRSwNP. These findings suggest a potential relationship between chronic inflammatory diseases of the upper respiratory tract and TL formation. However, prospective clinical studies are needed to confirm these results with greater certainty. To our knowledge, there is no existing study directly comparing nasal obstruction (NSD) with TL formation.

In our study, the presence of TLs was 1.3 times more common in patients with NSD than in those without NSD. Moreover, in patients with NSD, the side of deviation often corresponded to the same side with TL. Prospective controlled clinical studies are necessary to validate these findings and strengthen the evidence supporting this relationship.

Our study has some limitations. Since this was not a prospective study and records of patient complaints regarding TLs were not consistently maintained, a clear comparison between complaints and findings could not be made. Since TLs tend to fall through the crypts from time to time, the records of cases with single TL in such studies appear to have limited data. The lack of rhinomanometry and/or acoustic rhinometry results, which provide objective

data about nasal obstruction, limits the effects of NSD on TL formation. Another limitation of our study is the absence of smoking, oral hygiene, and dietary habits in the inclusion and exclusion criteria.

Conclusion

This study represents one of the largest series studying TLs. The incidence of TL in this study was 27.6%, indicating that it is not a rare condition among adults. The incidence of TL was found to be higher with increasing age. The incidence of TL increases under conditions that cause nasal obstruction such as NSD and under inflammatory conditions such as CRS and/or NP. In addition, the incidence of TL increases on the side where there is a nasal obstruction or nasal inflammatory condition. In terms of cause-and-effect relationship, the development of TL with inflammatory conditions and nasal obstruction controlled prospective studies are needed.

Ethics

Ethics Committee Approval: The study was conducted at Süleyman Demirel University Research and Training Hospital and was approved by the Ethical Committee for Clinical Studies of Süleyman Demirel University (date: 12.02.2021, number: 75).

Informed Consent: Additional informed consent was not required, as patients had already been informed that their radiographic imaging data could be used for future research purposes.

Footnotes

Authorship Contributions

Surgical and Medical Practices: H.Y., M.E.S., Y.Ç,K,, B,B., M,K., Concept: H.Y., M.E.S., Y.Ç,K., B,B., M,K., Design: H.Y., M.E.S., Y.Ç,K., B,B., M,K., Data Collection and/or Processing: H.Y., M.E.S., Y.Ç,K., B,B., M,K., Analysis and/or Interpretation: H.Y., M.E.S., Y.Ç,K., B,B., M,K., Literature Search: H.Y., M.E.S., Y.Ç,K., B,B., M,K., Writing: H.Y., M.E.S., Y.Ç,K., B,B., M,K.

Conflict of Interest: There is no conflict of interest to disclose.

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Main Points

- The incidence of Tonsillolith (TH) in this study was 27.6%.
- TL was more common in patients with increased age.
- TL incidence increased in conditions causing nasal obstruction, such as nasal septal deviation, as well as in inflammatory conditions like chronic rhinosinusitis and/or nasal polyps.

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Extracranial Head and Neck Schwannomas

Original Investigation

Mehmet Birinci[1], Gökçe Aydın Balaban[2], Oğuzhan Okcu[3], Oğuz Gül[4], Tuğba Yemiş[1], Metin Çeliker[1], Özlem Çelebi Erdivanlı[1]

[1]Recep Tayyip Erdoğan University Faculty of Medicine, Department of Otorhinolaryngology, Rize, Türkiye
[2]Rize State Hospital, Clinic of Otorhinolaryngology, Rize, Türkiye
[3]Recep Tayyip Erdoğan University Faculty of Medicine, Department of Pathology, Rize, Türkiye
[4]Haçkalı Baba State Hospital, Clinic of Otorhinolaryngology, Trabzon, Türkiye

Abstract

Objective: Schwannomas are benign tumors originating from the nerve sheath. Extracranial schwannomas account for 25-45% of schwannomas in the head and neck region. This study aimed to evaluate the clinical presentation, diagnostic modalities, and postoperative outcomes of extracranial non-vestibular head and neck schwannomas and to discuss the findings in the context of the literature.

Methods: Medical records of patients who underwent surgical treatment for extracranial schwannomas between 2014 and 2022 were retrospectively reviewed. A total of 25 patients met the inclusion criteria and were included in the study.

Results: The mean age of the patients was 49 years, with a male-to-female ratio of 16/9. The most common presenting symptom was painless swelling. The face was the most affected site, followed by the oropharynx and scalp. Preoperative imaging was performed in nine patients, with ultrasound being the preferred modality. Preoperative biopsy was conducted in only two patients. The mean follow-up duration was 54 months, with no reported recurrences.

Conclusion: This study represents the largest national series of extracranial head and neck schwannomas, providing valuable insights into their clinical presentation, diagnostic approach, and long-term outcomes.

Keywords: Head and neck neoplasms, schwannoma, benign neoplasms, peripheral nervous system neoplasms, surgical procedures, treatment outcome

ORCID IDs of the authors:
M.B. 0000-0001-8184-7454
G.A.B. 0000-0002-2042-3874
O.O. 0000-0001-7481-4718
O.G. 0000-0003-4626-0820
T.Y. 0000-0001-8713-0251
M.Ç. 0000-0002-9833-402X
Ö.Ç.E. 0000-0001-9245-1551

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Corresponding Author:
Mehmet Birinci, MD;
mehmet.birinci@erdogan.edu.tr

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Introduction

Schwannomas are tumors that originate from Schwann cells. They typically arise from peripheral, cranial, and autonomic nerves, except for the olfactory and optic nerves (1). However, recent case reports have described schwannomas in these two nerves, challenging the long-standing belief that they do not develop in this location (2,3). This underscores the need to consider schwannomas in the differential diagnosis of lesions in all areas of the head and neck region where nerve cells are present, including the cranial nerves.

The clinical signs and symptoms of schwannomas depend on the tumor's location, size, and nerve of origin. Surgical excision is the primary treatment modality; however, complete removal may

not always be feasible without compromising the function of the affected nerve (4).

This study aimed to collect clinical findings, radiological, and pathological characteristics of extracranial non-vestibular head and neck schwannomas. By focusing on this specific subgroup, this study provides valuable insights into their diagnosis, management, and surgical outcomes, contributing to a better understanding of these rare tumors.

Methods

Ethical approval for this study was obtained from the Ethics Committee of Recep Tayyip Erdoğan University Non-Interventional Clinical Research (approval no: 2022/234, date: 22.12.2022). The study was conducted in accordance with the principles outlined in the Declaration of Helsinki. Medical records of patients with histopathologically confirmed extracranial head and neck schwannomas, treated at the Department of Otorhinolaryngology of Recep Tayyip Erdoğan University Training and Research Hospital between 2014 and 2022, were retrospectively reviewed. Patients were excluded if they had intracranial schwannomas, schwannomas located outside the head and neck region, or if surgery was performed in a non-ear, nose, and throat (ENT) department such as plastic and reconstructive surgery. All patients provided informed consent for the use of their anonymized data in this study.

In our clinic, preoperative evaluations are guided by the following algorithm: superficial, regularly shaped lesions with a benign appearance are not subjected to radiologic imaging. Deep-seated lesions, those with suspected malignancy, or unilateral lesions in the nasal cavity are assessed using appropriate imaging modalities. Ultrasound is the first-line imaging technique for neck lesions, while magnetic resonance imaging (MRI) is preferred for lesions suspected of malignancy or those located in the nasal cavity. Biopsy is performed only for lesions demonstrating radiologic features suggestive of malignancy. Therefore, in addition to patient demographics, presenting symptoms, and anatomical tumor location, we recorded the imaging modality or biopsy procedure, if performed. Finally, follow-up duration and recurrence status were documented.

Statistical Analysis

Statistical analysis was performed with IBM SPSS Statistics, Version 22.0 (IBM SPSS Statistics for Windows, Armonk, NY: USA). Descriptive statistics were presented as median (minimum-maximum) for age and follow-up duration, or number (percent -%) for gender, complaints, localization, imaging and preoperative biopsy.

Results

Records of 143 patients diagnosed with schwannoma were reviewed for the study. Of these, 73 had schwannomas located outside the head and neck region, 41 had acoustic (vestibular) schwannomas, and 4 had undergone surgery in non-ENT departments. Thus, 118 patients who did not meet the inclusion criteria were excluded. Demographic characteristics of the included patients are summarized in Table 1. Briefly, two patients were pediatric cases (aged 9 and 13 years), and four patients were over 65 years of age. Two-thirds of the lesions were located on the face, followed by the oropharynx and the scalp. Among facial lesions, four were located around the right eyebrow, one around the left eyebrow, and one on the right chin. In the oropharynx, four lesions were located on the tongue and one on the palate. Neck lesions were situated in the submandibular region and at level IV. In the oral cavity, two lesions were found on the lower lip and one on the upper lip.

Figure 1 presents schwannomas at various anatomical sites, emphasizing the importance of appropriate preoperative imaging. A well-circumscribed, superficial mass on the tongue (Figure 1A) and an encapsulated lesion on the upper lip (Figure 1B) were completely excised without the need for preoperative imaging or biopsy. A left-sided sinonasal schwannoma was excised following MRI and biopsy (Figure 1C).

Table 1. Demographic and clinical characteristics of the study population. Data presented as mean (range) or number (%)

Parameter	Patients (n=25)
Age, years	49 (9-90)
Gender, (n%)	Male: 16 (64%)
	Female: 9 (36%)
Complaints and localizations, (n%)	
Painless swelling (n=22)	Face (n=6, 24%)
	Oropharynx (n=4, 16%)
	Scalp (n=5, 20%)
	Neck (n=4, 16%)
	Oral cavity (n=3, 12%)
Nasal obstruction (n=2)	Nasal cavity (n=2, 8%)
Dysphagia (n=1)	Oropharynx (n=1, 4%)
Preoperative imaging, (n%)	Performed: 9 (36%)
Modality, (n%)	USG: 5 (20%)
	MRI: 4 (16%)
Preoperative biopsy, (n%)	Performed: 2 (8%)
	Not performed: 23 (92%)
Follow-up period, months	54 (6-98)

MRI: Magnetic resonance imaging, USG: Ultrasonography

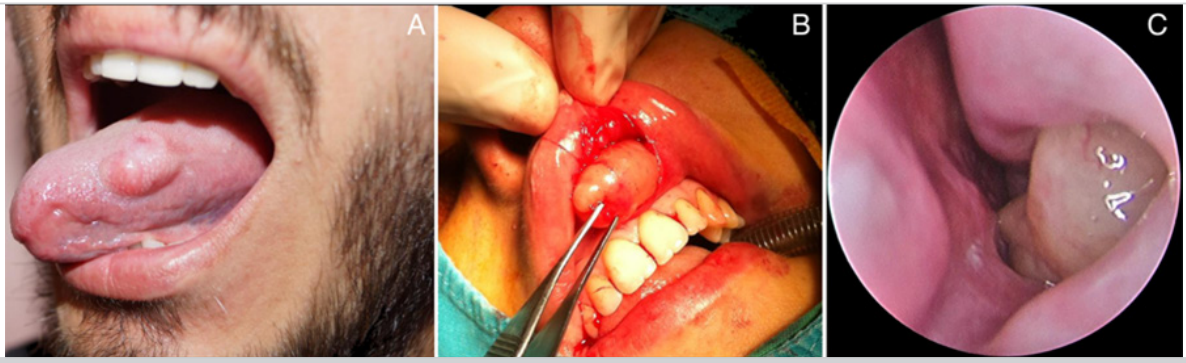

Figure 1. Clinical and intraoperative images of schwannomas located in various regions of the head and neck. **A) w**ell-circumscribed lingual schwannoma presenting as a submucosal mass on the tongue. **B)** intraoperative view of an encapsulated upper lip schwannoma during surgical excision. **C)** endoscopic image showing a left-sided sinonasal schwannoma located in the inferior meatus

Preoperative imaging was performed in approximately one-third of the patients to support differential diagnosis based on lesion localization. Ultrasound was utilized in three patients with neck lesions and in two patients with scalp lesions due to suspicion of malignancy; all lesions were reported as benign. MRI was preferred in cases requiring deeper tissue evaluation, including two patients with unilateral nasal cavity lesions, one with a tongue lesion, and one with a facial mass in the temporoorbital region. All MRI findings were consistent with benign pathology (Figure 2).

Two patients required preoperative biopsy for definitive diagnosis. One patient with a sinonasal lesion underwent incisional biopsy, while another patient with a neck lesion had a tru-cut biopsy. All patients underwent complete surgical excision, and the specimens were submitted for histopathological analysis. Figure 3 demonstrates the two routinely employed histological techniques. Hematoxylin and eosin staining (Figure 3A) revealed the characteristic biphasic architecture of schwannomas, with Antoni A areas consisting of densely packed spindle cells, and Antoni B areas showing a looser, myxoid stroma. Immunohistochemical staining for S-100 protein (Figure 3B) showed strong and diffuse positivity, confirming the neural crest origin of the tumors.

The mean follow-up duration was 54.3 months, with the longest follow-up period being 98 months and the shortest six months. No clinical, radiological, or histopathological evidence of recurrence was observed during the follow-up period.

Discussion

This case review demonstrated that extracranial non-vestibular head and neck schwannomas most commonly present as painless, well-circumscribed masses, with the face being the most frequently affected anatomical region. Surgical excision remains the standard treatment, and no recurrences were observed during long-term follow-up.

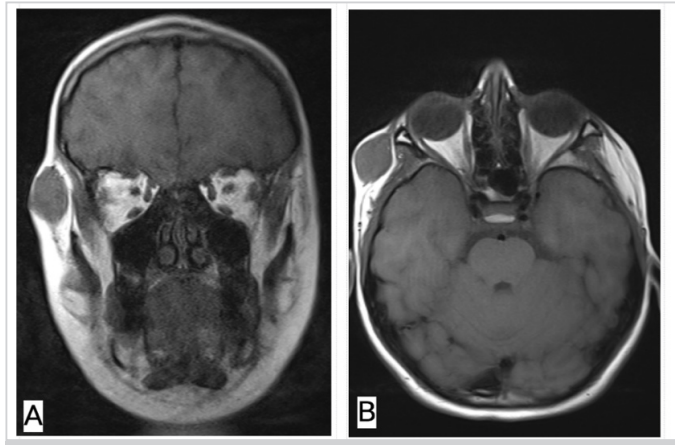

Figure 2. Preoperative magnetic resonance imaging reso of a schwannoma in the right temporoorbital region, demonstrating a well-defined 32×16×26 mm mass with low signal intensity on T1-weighted sequences **A)** coronal view **B)** axial view

The median age of patients in our study was consistent with previously published literature. Schwannomas are known to occur most frequently between the 3rd and 4th decades of life, with a slight female predominance reported in some studies (5).

In line with our findings, most extracranial schwannomas lack specific findings on physical examination and are often incidentally discovered as painless masses. However, depending on the tumor's size and anatomical location, symptoms such as dysphagia, cough, Horner's syndrome, dyspnea, and hoarseness may occur (6). Similarly, in our study, nasal obstruction was the predominant symptom in patients with nasal cavity involvement, whereas dysphagia was reported in one patient with a tongue base lesion. In rare cases, schwannomas may present as giant masses (7). It has also been noted that these tumors tend to be fixed along the axis of the originating nerve and more mobile in a direction perpendicular to it, a feature that may aid in diagnosis (8).

Extracranial schwannomas account for 25-45% of all schwannomas, with the cervical region and oral cavity frequently cited as the most common locations in different studies (9,10). In our study, the face was the most affected site, followed by the oropharynx and the scalp, which slightly differ from international series reporting the cervical region as the predominant site (11).

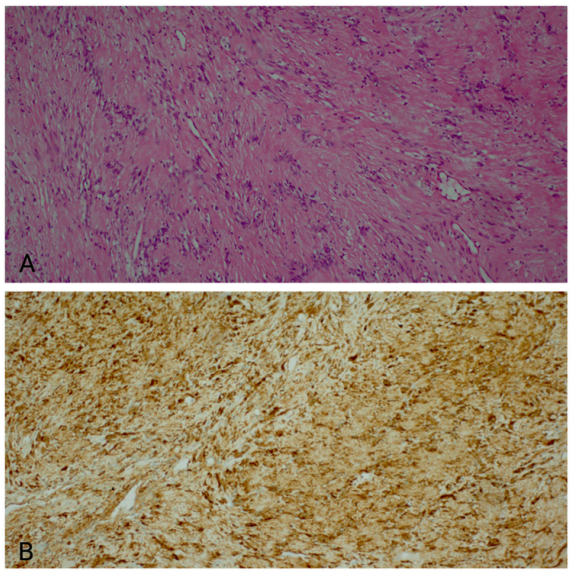

Figure 3. Histopathological findings characteristic of schwannomas **A)** hematoxylin and eosin staining at 200× magnification showing spindle-shaped Schwann cells arranged in Antoni A and Antoni B patterns **B)** S-100 immunohistochemical staining at 100× magnification demonstrating strong and diffuse cytoplasmic positivity, confirming Schwann cell origin

Intraoral schwannomas constitute 1-12% of the schwannomas in the head and neck region. In terms of specific subregions, the tongue, the floor of the mouth, the buccal mucosa, the lips, the palate, and the jaw are listed in order of frequency (12). Our findings align with this pattern, though we did not observe any buccal mucosa involvement in our cohort.

Neurogenic tumors in the head and neck region arise from neural crest cells that differentiate into Schwann cells and sympathoblasts. Schwann cells are the main cells of both schwannomas and neurofibromas. Neurofibromas originate from the perineurium and are therefore intimately connected to the nerve from which they arise. Schwannomas, on the other hand, assume a spindle-shaped appearance while longitudinally growing along the nerve without disrupting its structural and functional identity, and can be surgically separated from the nerves from which they originate (13). We could not identify the originating nerve in any of our cases. This may be attributed to the absence of neurological symptoms, which could have provided diagnostic clues. Additionally, no identifiable nerve trunk was observed intraoperatively. All dissections were performed according to good surgical practice.

Similar studies were conducted in our country with smaller case numbers (Table 2). The series by Altuntaş et al. (5) in 2012 is the first one and included 6 cases. The largest series was reported by Balcı et al. (14) consisting of 23 cases. In a series of 31 cases reported from Öztürk et al. (15), 18 patients

Table 2. Summary of extracranial schwannoma studies in Türkiye: localization, neurological deficits, and postoperative outcomes

Study	Period	Cases (n)	Localization					Preoperative neurologic deficit	Mean follow-up period (months)	Postoperative sequelae
			Neck	Oral cavity	Face and scalp	Nasal cavity	Middle ear cavity			
Altuntaş et al. (5)	2004-2008	6	3	1	-	1	1	N/A	N/A	Facial paresthesia
Balcı et al. (14)	2008-2016	23	15	3	4	1	-	Unilateral vocal cord paralysis (1)	17.6	Hypoesthesia (8)
										Facial nerve injury (3)
										Vagus injury (2)
										Hypoglossal nerve injury (1)
										Glossopharyngeal nerve injury (1)
Öztürk et al. (15)	2007-2018	18	12	3	-	-	3	Facial paralysis (3)	21	Seven patients (not described)
Çakır et al. (16)	1995-2015	14	11	1	-	2	-	None	14	Neural deficit (6) not described
Gülşen and Kurt (17)	2015-2019	14	9	4	-	1	-	N/A	12.3	Motor loss in brachial plexus 1
										Taste loss 1
										Ptozis 1

had an extracranial tumor. Recently, a series of 14 cases by Çakır et al. (16) and another series of 14 cases by Gülşen and Kurt (17) have been published.

Our study stands out as the largest series of extracranial head and neck schwannomas conducted in Türkiye, with longer follow-up compared to previous national studies (5,14-17).

Its retrospective nature is a limitation, as this may potentially introduce selection bias. Additionally, the lack of preoperative neurological deficits in our cohort limited the ability to identify the originating nerves.

Conclusion

Compared to international studies, our cohort had fewer imaging evaluations, which may reflect differences in institutional protocols and access to imaging modalities. Future prospective, multicenter studies are needed to establish standardized imaging protocols, optimize nerve preservation strategies, and assess long-term functional outcomes in patients with extracranial schwannomas.

Ethics

Ethics Committee Approval: This study was approved by the Non-Interventional Clinical Research Ethics Committee of Recep Tayyip Erdoğan University (approval no: 2022/234, date: 22.12.2022).

Informed Consent: All patients provided informed consent for the use of their anonymized data in this study.

Footnotes

Information: The authors acknowledge the use of a large language model (ChatGPT-4o, OpenAI) to refine the language, improve readability, and enhance the academic style of the manuscript following the completion of the initial draft. All modifications were carefully reviewed and validated by the authors to ensure scientific accuracy and adherence to ethical standards.

Authorship Contributions

Surgical and Medical Practices: M.B., Ö.Ç.E., Concept: M.B., G.A.B., Ö.Ç.E., Design: M.B., G.A.B., O.O., T.Y., M.Ç., Ö.Ç.E., Data Collection and/or Processing: M.B., O.O., O.G., M.Ç., Ö.Ç.E., Analysis and/or Interpretation: M.B., G.A.B., O.G., Literature Search: M.B., O.G., T.Y., M.Ç., Writing: M.B., O.O., O.G., T.Y., M.Ç.

Conflict of Interest: The authors declare that they have no conflict of interest.

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Main Points

- Extracranial schwannomas mostly present as painless, well-circumscribed masses, with the face being the most commonly affected region in this study.
- Surgical excision remains the definitive treatment, with no recurrences observed during long-term follow-up.
- Future prospective, multicenter studies are needed to standardize imaging protocols and assess long-term functional outcomes.

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Development and Evaluation of the Cadaver Dissection Training Program for the Resident School

Original Investigation

Hülya Eyigör[1], Murat Öztürk[2]

[1]University of Health Sciences Türkiye, Antalya Training and Research Hospital, Department of Otorhinolaryngology, Antalya, Türkiye
[2]Kocaeli University Faculty of Medicine, Department of Otorhinolaryngology, Kocaeli, Türkiye

Abstract

Objective: This study aims to evaluate and improve the cadaver dissection training program given by the Turkish Otorhinolaryngology and Head and Neck Surgery Association's resident school. The program addresses goals, outcomes, content, implementation sequence, educational strategies, and the steps for evaluation and regulation.

Methods: The Kern educational program development model was used by the 2023-2025 ENT Resident School Management for cadaver dissection training: both qualitative and quantitative research methods were employed. A needs analysis was conducted with structured focus interviews with the 12 students accepted into the course. Dissection steps, educational resources, and dissection videos were shared with residents before the training. A personalized cadaver dissection training plan, aligned with the Curriculum Development and Standardization System, was developed and shared with the instructors. Post-training, the Kirkpatrick program evaluation model was used to conduct first and second-level program evaluations.

Results: The first-level evaluation revealed high satisfaction with the training program, and the second-level evaluation indicated a statistically significant increase between pre-test and post-test scores (p=0.015). There was a significant positive correlation between the total mastoidectomy success scale scores and the difference scores (pre- and post-test) (p=0.019; r=0.663), while no significant correlation was found for the endoscopic sinus surgery success scale scores (p=0.996).

Conclusion: The Resident School, developed by the members of the Turkish Otorhinolaryngology and Head and Neck Surgery Association, is a high-participation, skill-intensive training program. The program, conducted with great dedication, received high participant satisfaction and assessed knowledge and skill learning levels.

Keywords: Cadaver, otorhinolaryngology, education, program evaluation

ORCID IDs of the authors:
H.E. 0000-0001-5317-431X
M.Ö. 0000-0002-3340-9975

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Corresponding Author:
Hülya Eyigör, MD;
hulinar@yahoo.com

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Introduction

Program development in medical education involves the systematic process of designing, developing, testing, implementing, evaluating, and refining a program. Evaluating the effectiveness and success of programs implemented in postgraduate education is crucial for program improvement (1). Program evaluation involves collecting, analyzing, and interpreting data to assign value to a specific educational activity (2). It determines the program's purpose, model, process, management, and principles and identifies factors that may affect the evaluation process and outcome. Stakeholders must carefully review these factors to make informed decisions about the program (3,4).

The Resident School, established under the roof of the Turkish Society of Otorhinolaryngology and Head and Neck Surgery [Turkish Ear, Nose, Throat (ENT) and HNS], has been contributing to the education of ENT residents for many years. As with any educational program, it is necessary to evaluate whether the assistant school training meets its objectives, and to conduct evaluation studies to develop the program according to the needs changing over time (5). This study aims to develop, implement, and evaluate a cadaver dissection training program for the Resident School.

Methods

The Resident Shool cadaver dissection training program was developed by the school administration (also the authors) following the Kern curriculum development steps and the program was evaluated. The data of the study were evaluated retrospectively. This retrospective study was conducted following the approval of the Kocaeli University Ethics Committee (decision: KÜ GOKAEK-2024/13.16, date: 22.08.2024).

Program Development

The cadaver dissection training program for the ENT Resident School was developed using the Kern program development steps (6).

1. Problem Identification: Cadaver dissection training is a crucial step for achieving interventional competencies in specialty education. The Turkish ENT and HNS Association has been supporting ENT resident education through cadaver dissection courses for many years. However, the rapid increase in the number of residents in recent years poses a significant threat to the standardization of skill training. Therefore, it was necessary to revisitthe training provided in the Resident School, define its outcomes, and evaluate whether the desired goals were achieved after the training.

2. Needs Assessment: A needs assessment was planned after the Resident School was announced and applications were received. Since the participants come from different training centers and have different seniority, students' needs and expectations for school education may also differ. A focus group discussion was conducted with the residents prior to the training during the interview, the researcher first introduced himself, shared information about the purpose of the interview and how long the interview would last, and verbal approval was obtained for recording. Using the Curriculum Development and Standardization System (CDSS) as a basis, qualitative data were obtained through online structured focus group interviews to identify each resident's educational deficiencies (Table 1).

3. Goals and Learning Objectives: The goals and learning objectives of the Resident School were defined following the 2019 version 2.4 TUKMOS interventional competency levels, considering the seniority levels of the participants.

4. Educational Strategies: Before the cadaver training, the Resident School management shared step-by-step surgical dissection guides, related books, online educational materials, and master dissection video recordings with the accepted students.

5. Program Implementation: The cadaver dissection was conducted at the TORLAK Surgical Anatomy and Distance Education Center on dates scheduled by the school management and the Turkish ENT and HNS Association. The managerial aspect of the program involved internal Resident School Management and external support (ENT School Instructors) at every stage. Expert instructors provided one-on-one dissection training to students during the dissection sessions.

6. Program Evaluation: The first- and second-level evaluations of the cadaver dissection training program were conducted using the Kirkpatrick program evaluation model (7). At the end of the training, all participants were asked to anonymously complete an online satisfaction survey. At the end of the training program, all participants were asked to evaluate the program in every aspect according to the propositions A multiple-choice test was administered before and after the training. The results of pre-test and post-test comparisons from online and face-to-face training sessions were used for the second-level evaluation. Additionally, the endoscopic sinus surgery (ESS) and mastoidectomy success scales were administered to assess skill training (8,9).

Statistical Analysis

Qualitative data were collected during focus group interviews, and video recordings were transcribed. Researchers coded the data, combined statements with similar meanings, and created themes. These themes were verified through participant feedback. Data analysis was performed using IBM SPSS Statistics for Windows version 25.0. Qualitative variables were presented as frequency and percentage.

Table 1. Structured focus group interview questions

Primary question	Question at the end	Explanation
What are your reasons for applying to the Resident School?	Do you consider the training you received in your clinic to be insufficient?	What do you think the school's contribution to you should be?
What surgeries are you currently able to perform related to endoscopic sinus surgery?	What are the surgical interventions that you think you have received inadequate training in your field in your residency training?	Please explain the topics you need and want to be included in the school program.
What surgeries are you currently able to perform related to temporal bone dissection?	What are the surgical interventions that you think you have received inadequate training in your field in your residency training?	Please explain the topics you need and want to be included in the school program.
What do you think about the inclusion of dissections related to head and neck-facial plastic in the Resident School program?	Which surgeries would you like to do?	Please explain the reasons.

Normal distribution of quantitative variables was tested using the Shapiro-Wilk test. For variables with normal distribution, arithmetic mean and standard deviation values were presented; for non-normally distributed variables, median, minimum, and maximum values were presented. The paired sample t-test was used to compare pre-test and post-test results. Pearson correlation was used to compare difference scores with total scores on the mastoidectomy and ESS success scales. A Type I error rate of 0.05 was considered.

Results

Participant Focus Group Results

Twelve students were invited to the focus group interviews, conducted in two sessions. Demographic characteristics of the students are summarized in Table 2. Qualitative data examples summarizing the students' expectations from the training are provided.

Examples of qualitative data summarizing students' expectations from education in needs assessment analysis:

- *"It would be good to see complications in the cadaver and intervene."*
- *"I am coming to the end of my residency; I attended the course to see different approaches schools of thought."*
- *"It will be good for understanding 3D anatomy."*
- *"Facelift will be very popular in the future; I think it will be great to receive this training on cadavers at the resident level."*
- *"I have not performed experienced mastoidectomy yet, I will do it on a cadaver first."*
- *"There were subjects that I missed a lot due to Covid and the earthquake, I plan to complete them in this course."*
- *"It will be useful for clinics where the addition of an endoscope is not performed in cadaveric ear surgery."*

Table 2. Demographic characteristics of participating resident cadaver dissection school students and trainers who gave feedback

Residents (n, %)	
Gender	Female (3, 25%)
	Male (9, 75%)
Type of hospital	University hospital (6, 50%)
	Training and research hospital (5, 41.7%)
	Overseas participation (1, 8.3%)
Seniority of resident	4th year (6, 50%)
	5th year (6, 50%)
Age	27-34 years (average=29.7)
Trainers (n, %)	
Gender	Female (4, 33.3%)
	Male (8, 66.7%)
Title	Professor (3, 25%)
	Associate professor (6, 50%)
	Doctor lecturer (1, 8.3%)
	Specialist (2, 16.7%)
Age	36-69 years (average=40.8)
Dissection	Temporal (5, 16.7%)
	ESS (4, 33.3%)
	Facial (2, 16.7%)
	Head and neck (1, 8.3%)

ESS: Endoscopic sinus surgery

Kirkpatrick Model Program Evaluation Results

First Level (Reaction): The participants' responses indicating that they were generally satisfied with the training provided are shown in Table 3.

According to the students, the aspects of education that need to be improved are:

- *"If more time was allocated for fascial plastic surgery, we could have performed do more procedures."*
- *"When performing temporal bone dissection, there may be a transfer to the tower or a second microscope eye. Computed tomography images of the cadavers could have been made available."*

Table 3. Evaluation of student satisfaction after cadaver dissection training program

		1	2	3	4	5
Achieving course objectives	1. The objectives of the course are clearly explained				1 (8.3%)	11 (91.7%)
	2. The content of the course met my learning goals				4 (33.3%)	8 (66.7%)
	3. The course was in line with my seniority in terms of content				1 (8.3%)	11 (91.7%)
	4. There was an effective communication environment in the course				1(8.3%)	11(91.7%)
	5. The course duration was sufficient to achieve the learning objectives		2 (16.7%)	2 (16.7%)	4 (33.3%)	4 (33.3%)
Program content	6. The topics included in the program addressed the areas I need in the clinic				1 (8.3%)	11 (91.7%)
	7. The allotted time was enough for the course content		2 (16.7%)	3 (25%)	5 (41.7%)	2 (16.7%)
Performance of trainers	8. Training was held with a competent trainer in the field				2 (16.7%)	10 (83.3%)
	9. Throughout the program, I was able to communicate effectively with the trainer				3 (25%)	9 (75%)
	10. The trainer took into account the needs of the participants			1 (8.3%)	2 (16.7%)	9 (75%)
Performance of trainers	11. The course was well organized				2 (16.7%)	10 (83.3%)
	12. The course was designed to improve my knowledge of surgical anatomy				2 (16.7%)	10 (83.3%)
	13. The course improved my surgical skills				2 (16.7%)	10 (83.3%)

Likert scale= 1: Strongly disagree, 2: Disagree, 3: Undecided/neutral, 4: Agree, 5: Strongly agree

- *"I just think that allocating a weekend to each subspecialty branch and teaching theoretical lessons in the presentation room beforehand will increase the benefit of the course for us."*
- *"Course date could have been notified earlier."*
- *"As ENT physicians, we see that in recent years there has been a trend towards rhinoplasty in ENT practice and good results of this trend along with education. However, unfortunately, I see that the focus on head and neck surgery and reconstruction is decreasing, and sometimes we have to cooperate with different departments or be dependent on different departments. I know that the number of clinics performing head and neck reconstruction (regional and free flaps), which is also included in our Specialty Board in Medicine core training program, is few compared to abroad. In this respect, I believe that organizing training (zoom lectures, meetings, live surgeries) and encouraging reconstruction will improve our healthcare service delivery as ENT physicians."*
- *"The course program was very detailed. It was very instructive for us. Having two residents per cadaver was very valuable in completing all the steps. Surgical sets, microscopes and endoscopes were perfectly equipped. In order for the program to be more complete, the course duration could have been longer, or it could have been done in three separate departments: Functional sndoscopic sinus surgery, otology and facial plastic. If the course program and content had been announced earlier, it would have been more beneficial for us in terms of studying and preparing. But even in its current state, it was incredibly educational and productive for residents."*

Instructor Feedback

Sixteen instructors took part in the training, with 12 providing feedback post-training. Demographic characteristics of the instructors are shown in Table 2. Satisfaction survey results are presented in Table 4.

Qualitative feedback results from trainers:

- *I really liked that the residents were trained, ready and motivated, and that they had determined their own expectations from the dissection.*

Table 4. Satisfaction survey results of instructors involved in the cadaver dissection training program

	1	2	3	4	5
I was adequately informed about the content of the pre-training and the learning objectives					12 (100%)
The course was conducted according to the program given at the beginning of the training				1 (8.3%)	11 (91.7%)
The level of knowledge of the residents was adequate for the course			1 (8.3%)	5 (41.7%)	6 (50%)
I was able to communicate effectively with the residents				1 (8.3%)	11 (91.7%)
The educational environment and infrastructure were sufficient				1 (8.3%)	11 (91.7%)
Surgical instruments were sufficient for dissection			1 (8.3%)	5 (41.7%)	6 (50%)

Likert scale= 1: Strongly disagree, 2: Disagree, 3: Undecided/neutral, 4: Agree, 5: Strongly agree

- *Everything was very systematic, there were no problems.*
- *Resident expectations were determined correctly, and regular and planned dissection could be performed.*
- *The course was very well-organized, and it is obvious that care was taken: in order to include more people, the number of hands-on trainee participants can be increased, and additional trainees can be recruited for free/a symbolic fee to watch the dissection without touching the cadaver, and to participate in all other trainings: theoretical, video, etc. (2 per cadaver) (e.g., +2 trainees).*
- *Adding missing surgical instruments (Scissors, curettes and rongeur tips should be renewed).*

Second Level (Learning): Significant improvement was observed between pre-test and post-test scores (pre-test 5.75±1.91, post-test 6.83±1.9, t=-2,862, p=0.015).

Skill Evaluation Results: The mastoidectomy and endoscopic sinus surgery (ESS) success scales were used to assess skill levels (Tables 5 and 6). There was a significant positive correlation between the total mastoidectomy success scale scores and the difference scores (pre-test and post-test). No significant correlation was found for the ESS success scale scores (p=0.996) (Table 7).

Discussion

In this study, we discuss the development steps of the Turkish ENT and HNS Association, Resident School cadaver dissection training program and its evaluation results. After the training, the program evaluation using the Kirkpatric model concluded that satisfaction was high and that it was a training that contributed to the resident's education in terms of knowledge and skills.

A training program should be planned following a targeted purpose and be evaluated for whether it has achieved its goal (1,2). When planning a training program, the needs of the participants should be determined first. The needs assessment analysis we conducted in our study showed that although the residents were similar in terms of seniority, their skill competency levels were different from each other. Such that, while some residents had performed all the steps of mastoidectomy and expected advanced surgery from this training, there were others who expected "I have not experienced mastoidectomy yet, I will do it on a cadaver first." We determined each resident's skill level and informed the trainers in advance and tried to implement the individualized education model.

Evaluation of a program provides very important feedback for the development and sustainability of the program. According to the Kirkpatrick program evaluation model, in the first stage, data can be collected to evaluate the program based on the participants' perceptions, that is, their satisfaction or dissatisfaction with the educational training program (7). In our study, we applied a satisfaction survey that questioned every aspect of the program to understand whether the residents and trainers found the training program useful, and when we evaluated their answers to the propositions, results showed that they were satisfied with the program. Moreover, their comments at the end of the program showed that participants found the training very useful and that they wanted to participate in such a program again-indicating that the program was successful for the first level evaluation. In qualitative data, deficiencies in the training program (such as duration of the training program, program content, surgical instruments) were recorded as important data sources for the development and improvement of future programs.

Table 5. Mastoidectomy checklist assessment

Student no	Cortical mastoidectomy	Revealing anatomical boundaries	Opening of the antrum	Digastric dissection	Thinning of the back wall of external ear canal	Opening of the facial recess	Posterior atticotomy
1	5	5	5	5	5	5	5
2	3	4	3	3	3	5	3
3	3	5	5	5	5	3	5
4	3	4	3	2	3	3	3
5	4	4	4	N/A	3	5	4
6	5	4	5	N/A	5	2	5
7	5	5	5	2	4	4	4
8	3	3	3	3	4	3	3
9	5	5	5	2	4	3	3
10	1	1	1	N/A	2	N/A	N/A
11	3	3	4	N/A	4	3	3
12	5	5	5	4	5	4	5
Average	3.5 (1-5)	4 (1-5)	4.5 (1-5)	2.6 (2-5)	3.9 (2-5)	3(2-5)	3.9(3-5)
Total	67±22.07						

Unable to perform 1, performs with minimal prompting 3, performs easily with good flow 5

Not applicable (N/A)

Table 6. Endoscopic sinus surgery checklist assessment

Student No	Sinus Endoscopy	Uncinectomy	Maxillary antrostomy	Anterior ethmoidectomy	Posterior ethmoidectomy	Sphenoidotomy	Frontal sinusotomy
1	4	4	4	4	5	4	3
2	5	5	5	5	5	5	3
3	5	5	5	4	5	3	3
4	4	4	5	4	4	4	3
5	5	4	4	4	4	4	3
6	5	5	5	5	4	4	4
7	5	5	5	5	5	5	4
8	5	5	5	5	5	3	3
9	5	5	5	5	5	5	3
10	5	5	5	5	5	5	5
11	5	5	5	5	5	5	5
12	5	5	5	5	5	5	4
Average	5 (4-5)	5 (4-5)	5 (4-5)	5 (4-5)	5 (4-5)	4.5 (3-5)	3 (3-5)
Total	79.75±10.75						

Unable to perform 1, performs with minimal prompting 3, performs easily with good flow 5

Not applicable (N/A)

Table 7. Comparisons between difference scores mastoidectomy and endoscopic sinus surgery total scores for total scores

		Difference score
Mastoidectomy total score	r	0.663*
	p	0.019
Endoscopic sinus surgery-total score	r	0.002
	p	0.996

*p<0.05

A second level evaluation is carried out shortly after the end of the program to investigate the changes in knowledge, skills and attitudes. Objective evaluation methods, performance tests, and attitude scales can be used at this stage (7). It is recommended to conduct measurement and evaluation in both cognitive and skill areas at every stage of the program following the goals (1,2). The Turkish ENT and HNS Association has been organizing pre- and post-graduation training programs under the name of ENT schools for more than ten years, but these have no systematic program evaluation. Regarding this subject, only Ecevit et al. (10) evaluated the two-year rhinology training program in terms of staff satisfaction and knowledge. In their study, the authors drew attention to the school's learning objectives, appropriate educational models and measurement and evaluation deficiencies. In our study, in addition to cognitive evaluation, we evaluated the skill levels of the residents using surgical skill scales, unlike the existing literature. Cognitively, statistical success was achieved in the post-test analysis conducted before the start of the training and after the training. However, as stated by the students in the qualitative evaluation form, it was considered restrictive due to the delay in determining the student admission list and the short duration of the surgical dissection training. Therefore, we recommend paying attention to the duration of the program. Secondly, although a standard educational resource was offered to all students, it was up to the students to prepare for them or not. As stated by the students in the qualitative data results, extending the training over a long period of time and providing additional theoretical courses in the future will contribute to standardization.

Objective measures in skills-based training courses allow trainees and trainers to evaluate performance and monitor progress. For this purpose, we tried to measure skill training with success scales. Francis et al. (9) developed the Global Evaluation Scale and the mastoidectomy evaluation scale to evaluate ear surgery in a real environment. Kara (11) reviewed the validity and reliability of various tools, including the mastoidectomy evaluation scale, used in evaluating the skill training of residents. In our study, we evaluated the competency of the residents in cadaver dissection steps by using these validated scales. At the end of the dissection training, one trainer evaluated the two students with whom they performed the dissection. Therefore, the fact that six different trainers evaluated 12 residents was considered the most important limitation of the study in terms of objectivity and standardization. Again, the fact that there were residents with different levels of surgical skills (seniority, facilities of the clinic where they received training, interest of resident, etc.) also led to different scores. In general, as expected, while the residents were successful in the first steps of mastoidectomy, they showed the lowest score (2.6) in the opening of the digastric region (recognition of the digastric muscle, recognition of the stylomastoid foramen). Although demonstration of proficiency achievements has not yet become a requirement for graduation in otolaryngology specialty, we think that this stage of defining surgical goals will become mandatory in the coming years. Therefore, scales that describe and objectively evaluate surgical steps will be needed. In their article published in 2024, Jayaraman Patnaik et al. (12) reported a scale study evaluating mastoidectomy in cadaver dissection for residents. They conducted the study with 16 residents at a tertiary care teaching hospital and showed the internal consistency of the Likert scale assessment. Mowry et al. (13) evaluated residents with the scale they developed for temporal bone dissection. Residents participated in weekly dissections for nine months every year. Individual student scores for each dissection skill were monitored over time. The authors reported that this criterion they developed was easy to use and that scoring was consistent among evaluators. They also emphasized that this criterion was successful in distinguishing between those who know and those who do not. Further to the above-described, another important limitation of our study is that we did not monitor the development process of the residents in terms of skills or evaluate their immediate proficiency.

The residency training program should provide residents with the skills and competence to perform surgery safely. However, education opportunities in this field are not standard and equal for every education center. For this reason, residents can experience difficulties in improving their surgical skills in this field. Cadaver dissection courses offer the most realistic skill training opportunity to meet this need. Reports on objective measurement tools used for assessment and evaluation of skills are limited in the literature. Laeeq et al. (14) evaluated 17 residents in the Johns Hopkins ENT residency program while performing ESS in the operating room. In their study, the authors evaluated three steps, including maxillary antrostomy+anterior ethmoidectomy, posterior ethmoidectomy+sphenoidostomy, and frontal sinusotomy. Between 2009 and 2011, eight evaluators completed a total of 73 evaluations for 17 residents (seniority 2-5). As a result of the evaluation, they stated that the residents showed the lowest score in the ESS steps in the frontal sinusotomy. In our study, residents received higher scores, and the lowest score was for frontal sinusotomy, which is consistent with the literature.

Cadaver dissection training requires knowledge of surgical anatomy as well as skill in the use of surgical instruments

and surgical technique. In our program evaluation, we found that the residents achieved significant success in the post-test results after training. When we evaluated the skill levels of the residents who improved themselves in terms of knowledge, we found that there was a high level of positive correlation between the results of the mastoidectomy success chart and the difference scores (pre- and post-test difference) (p=0.019; r=0.663), meaning that the students who achieved significant success in the post-test. They also demonstrated a high level of skill, and we observed that they received high scores. In general, ESS success scale scores were seen to be higher than mastoidectomy success scale scores. The reason for this may be that students perform this surgery more during their continuing education in their clinics or that the trainers working at ESS make more optimistic evaluations in scoring.

Another most important limitation of our study is that skill evaluations could not be performed because skill training regarding head and neck, and facial plastic is not implemented as a standard. Yet another limitation is that the results of the training program are according to the Kirkpatrick program evaluation model 3 (impact) and 4th (results) step could not be evaluated.

Conclusion

This report presents the first cadaver dissection training program, which was developed for ENT residents under the roof of the Turkish ENT and HNS Association and was evaluated using the Kirkpatrick model. Statistically significant results at each evaluation level indicated the program's success. We believe that there is a need for long-term, standardized training program development and evaluation studies for the Resident School, where skills are evaluated with objective measurement and evaluation methods and the reflections of the training program on the field can be closely monitored in the long term.

Ethics

Ethics Committee Approval: This retrospective study was conducted following the approval of the Kocaeli university Ethics Committee (decision no: KÜ GOKAEK-2024/13.16, date: 22.08.2024).

Informed Consent: Since this study was a retrospective one, patient consent was not required.

Footnotes

Authorship Contributions

Concept: H.E., M.Ö., Design: H.E., M.Ö., Data Collection and/or Processing: H.E., Analysis and/or Interpretation: H.E., Literature Search: H.E., M.Ö., Writing: H.E., M.Ö.

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Main Points

- Training programs should be planned in line with the targeted purpose and whether these goals have been achieved should be evaluated at the end of the training program.
- The Resident School, developed under the roof of the Turkish Otorhinolaryngology and Head and Neck Surgery Association by its members, is a high-participation, skill-intensive training program.
- Skills should be evaluated with objective measurement and evaluation methods, and the reflections of the training program on the field should be closely monitored in the long term.

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Infected Thyroglossal Duct Cyst with an Emergent Airway Threat

Case Report

Teng Huei Lee[1,2], Shangeetha Anbalagan[2], Santhi Kalimuthu[2], Norsyamira Aida Mohamad Umbaik[1]

[1]Department of Otorhinolaryngology-Head and Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia, Kelantan, Malaysia.
[2]Department of Otorhinolaryngology-Head and Neck Surgery, Hospital Tengku Ampuan Rahimah, Selangor, Malaysia

Abstract

Thyroglossal duct cyst (TDC) is a common congenital neck anomaly that typically presents as a midline neck mass and is most often diagnosed in the second decade of life. Although TDCs are generally painless and asymptomatic, infection of the cyst can lead to significant airway obstruction. We report the case of a 31-year-old male who experienced upper airway obstruction due to an acute TDC infection. We performed a curative modified Sistrunk procedure to relieve the obstruction during the acute episode. This case underscores the critical need for prompt diagnosis and intervention to prevent morbidity and mortality associated with airway emergencies in infected TDCs. We also discuss the challenges of managing an acutely infected TDC with emergent airway involvement, necessitating a more complex surgical approach.

Keywords: Thyroglossal cyst, airway obstruction, Sistrunk procedure, neck abnormalities, case report

ORCID IDs of the authors:
T.H.L. 0000-0002-3975-725X
S.A. 0000-0002-8886-7447
S.K. 0000-0001-8514-6815
N.A.M.U. 0000-0003-4556-7601

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Corresponding Author:
Norsyamira Aida Mohamad Umbaik, MD;
syamira@usm.my

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Introduction

Thyroglossal duct cyst (TDC) is a common congenital anomaly that typically presents as a painless anterior neck swelling during the second decade of life (1). During embryological development, the thyroid gland descends from the foramen cecum to its adult position via the thyroglossal duct, which normally obliterates by the tenth week of gestation (1). Failure or incomplete obliteration of this duct results in the formation of a TDC (1,2). Due to its anatomical relationship with the thyroid gland and foramen cecum, the swelling characteristically moves upward with swallowing and tongue protrusion (3,4).

TDC can become complicated by infection, owing to its proximity to the oral cavity, with infection rates ranging from 10% to 70% (1,5). An infected TDC may be life-threatening due to significant airway obstruction (6). Comprehensive surgical excision is the definitive treatment for TDC. The standard technique, described by Sistrunk, involves removing the cyst, the central portion of the hyoid bone, and the thyroglossal duct, as well as resecting tongue base tissue up to the foramen cecum, achieving a recurrence

rate of less than 6% (7). This procedure is typically performed in a non-infected state, as an infected TDC increases the risk of bleeding and residual tissue (7).

Case Presentation

A 31-year-old male with a history of atrial flutter presented with fever, painful anterior neck swelling, and purulent discharge from the swelling. The anterior neck swelling had been present for five years; however, this was the first episode of infection. The discharge was thick, milky, foul-smelling, and expressible upon palpation. He also reported a sensation of globus on swallowing and worsening dyspnea when lying flat. There was no noisy breathing or shortness of breath when sitting upright.

On examination, the patient's voice was breathy, but he could speak in full sentences, with no stridor observed. The midline anterior neck swelling measured 15x15 cm, extending laterally to involve levels II to IV bilaterally, with overlying skin appearing stretched and inflamed (Figure 1). A punctum was noted at the lower left aspect of the swelling. Needle aspiration yielded 10 cc of thick, seropurulent fluid. Flexible nasopharyngolaryngoscopy revealed a globular lesion at the left base of the tongue, obliterating the left vallecula and left aryepiglottic fold, with posterior displacement of the epiglottis causing supraglottic airway narrowing (Figure 2). Vocal cord mobility was asymmetric, with left vocal cord paresis on phonation, though fully compensated by the right vocal cord.

A contrast-enhanced computed tomography scan of the neck demonstrated a large, well-defined, unilocular cystic mass in the left paramedian anterior neck, measuring 9.3x11.6x10.8 cm. The mass involved both the suprahyoid and infrahyoid regions, exerting a local mass effect that narrowed the airway, suggestive of a TDC. A multidisciplinary team (MDT) discussion, involving the radiology, anesthesiology, and otorhinolaryngology departments, was held to determine the optimal approach for airway management and surgical timing. The MDT agreed to secure the airway via awake fiberoptic nasal intubation and proceed immediately with surgical intervention to excise the infected TDC.

After successfully securing the airway, we performed a direct laryngoscopy and a modified Sistrunk procedure. Intraoperatively, the cystic mass measured 15x18 cm (Figure 3). The midline mass was adherent to the body of the hyoid bone, extending laterally to the left, and was attached to the pharyngeal mucosa. It exhibited superior lateral extension to the vallecula and pyriform fossa, and inferior extension to the suprasternal notch. The mass was decompressed intraoperatively via needle aspiration and meticulously dissected from the underlying pharyngeal mucosa. The body of the hyoid bone was divided, and dissection continued to the tongue base, where a cuff-of muscle was excised along with the specimen. Direct laryngoscopy confirmed an intact pharyngeal mucosa with no evidence of airway edema.

Postoperatively, the patient was extubated in the operating theater after careful assessment of his respiratory effort and airway patency. He was monitored in a high-dependency unit for one-day before being transferred to the general ward for two-days, remaining stable throughout his hospital stay. His recovery was uneventful. At a two-week follow-up visit,

Figure 1. Large anterior neck swelling measuring 15×15 cm, with a punctum (blue arrow) visible in the lower left portion of the mass

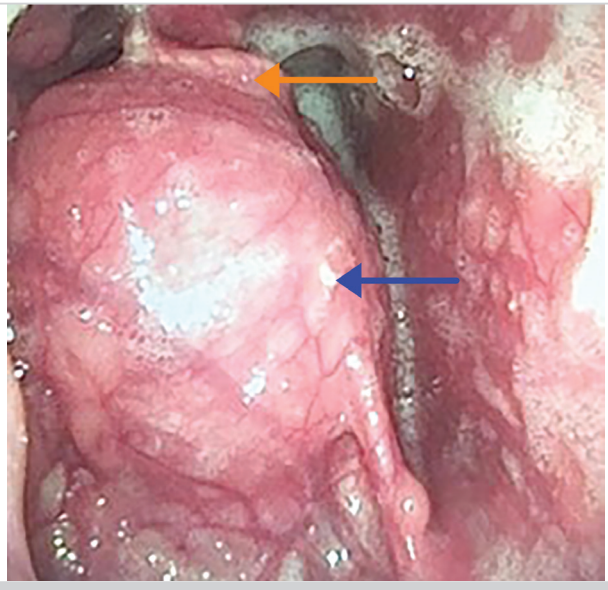

Figure 2. Flexible endoscopic examination revealed a large cystic, globular mass at the left base of the tongue, causing significant airway narrowing (blue arrow) and posterior deflection of the epiglottis (orange arrow)

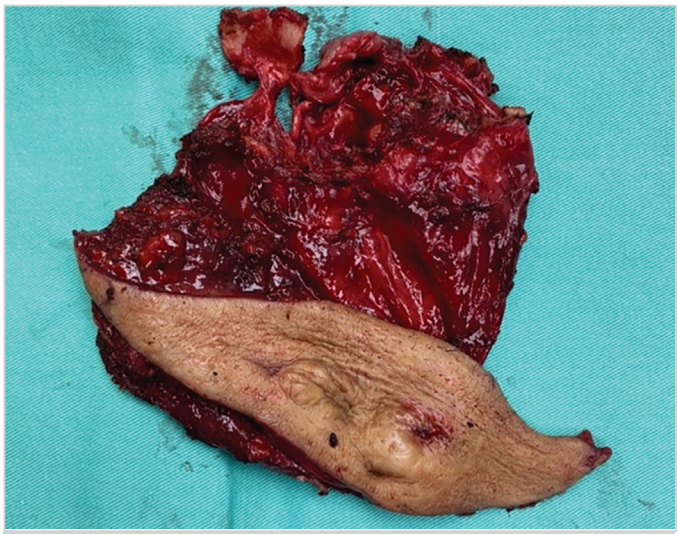

Figure 3. Postoperative specimen showing en bloc removal of the lesion, including part of the skin, tract, and body of the hyoid bone

the previously noted left vocal cord paresis had resolved. He was followed up for one-year with no evidence of recurrence. Informed consent was obtained from the patient for this case report.

Discussion

TDCs typically present as soft, cystic masses in the cervical region, particularly along the midline from the hyoid bone to the level of the thyroid gland. They are most commonly located between the hyoid bone and thyroid gland (60%), but may also arise at any point along the thyroglossal duct tract, including the suprahyoid region (24%), suprasternal region (13%), and, rarely, the intralingual area (1-2%) (2). Due to their anatomical proximity to the oral cavity, TDCs are predisposed to secondary infections, which often manifest as painful anterior neck swelling, sometimes accompanied by a discharging fistula, fever, and dysphonia, as seen in this patient (3). Common bacterial pathogens involved in TDC infections include *Haemophilus influenzae, Staphylococcus aureus,* and *Staphylococcus epidermidis* (5).

This case highlights the atypical presentation and management challenges of a long-standing, large anterior neck mass complicated by airway compromise, an uncommon feature of TDCs. Only a few cases of TDC-related airway involvement have been reported in the literature, and in this instance, the intralaryngeal extension of the cyst was a key contributing factor. The presence of a significantly narrowed supraglottic airway, exacerbated by infection, posed a substantial management challenge. Given the risk of failed intubation under general anesthesia and the difficulty of performing a tracheostomy due to the large anterior neck swelling obscuring the surgical field, awake fiberoptic nasal intubation was chosen as the safest approach for securing the airway. This case underscores the critical importance of a meticulous, tailored anesthetic approach to maintain adequate oxygenation, further highlighting the necessity of a MDT team strategy in managing complex airway obstructions.

The traditional Sistrunk procedure, first described by Sistrunk in 1920, involves en bloc resection of the TDC along with the body of the hyoid bone and the midline root of the tongue up to the foramen cecum (8). The suprahyoid portion of the thyroglossal duct contains multiple branches at the tongue's root, and incomplete excision of this segment may lead to recurrence. However, Sistrunk himself noted that the suprahyoid ductal tissue is extremely fragile and can easily break during surgery (8). While the Sistrunk procedure is the gold standard for TDC excision, it carries risks of injury to the lingual artery, oropharyngeal breach leading to pharyngocutaneous fistula, and infection (9). Recognizing these risks, Sistrunk modified his technique in 1928, limiting resection to the tongue base muscle without extending it to the foramen cecum (9). Since then, various adaptations of the modified Sistrunk procedure have been proposed, including a muscle-sparing approach, which has demonstrated favorable outcomes (10).

In this patient, a modified Sistrunk procedure was performed with an extended dissection in the neck region due to the cyst's large size and adherence to laryngopharyngeal structures. The surgery was particularly challenging due to strong adherence of the mass to the pharyngeal mucosa, requiring meticulous dissection to prevent mucosal tearing. Additionally, the overlying unhealthy skin had to be excised, contributing to a longer operative time and increased intraoperative bleeding.

Conclusion

This case underscores the critical importance of early diagnosis and timely surgical intervention in managing TDCs complicated by acute infection and airway obstruction. The successful use of the modified Sistrunk procedure during an acute episode highlights its efficacy, even in emergency settings, reinforcing its role in preventing morbidity and mortality. Furthermore, this case illustrates the challenges associated with managing infected TDCs in airway emergencies, emphasizing the need for heightened clinical awareness and a multidisciplinary approach to optimize patient outcomes. Finally, long-term follow-up remains essential to monitor for TDC recurrence and ensure complete resolution of the condition.

Ethics

Informed Consent: Informed consent was obtained from the patient for this case report.

Footnotes

Authorship Contributions

Concept: T.H.L., S.A., S.K., Design: T.H.L., S.A., S.K., N.A.M.U., Data Collection and/or Processing: T.H.L., S.A., S.K., Analysis and/or Interpretation: T.H.L., S.A., S.K., N.A.M.U., Literature Search: T.H.L., N.A.M.U., Writing: T.H.L., N.A.M.U.

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Main Points

- Thyroglossal duct cysts (TDCs) are typically painless and asymptomatic. However, acute infection can lead to significant airway obstruction.
- If not managed promptly, airway obstruction caused by an infected TDC can result in severe morbidity and mortality. A multidisciplinary team approach is crucial for optimal outcomes.
- Even in the acute stage, an infected TDC can be effectively treated with the Sistrunk procedure, providing definitive management while simultaneously improving airway obstruction.

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Cervicofacial Emphysema: A Rare and Potentially Fatal Complication of Tonsillectomy

Case Report

Vural Akın[1], Hasan Yasan[1], Çağlar Irmak[2], Erdinç Tanlak[3], Bekir Büyükçelik[4]

[1]Süleyman Demirel University Faculty of Medicine, Department of Otorhinolaryngology and Head & Neck Surgery, Isparta, Türkiye
[2]Yüksekova State Hospital, Department of Infectious Diseases and Clinical Microbiology, Hakkari, Türkiye
[3]Tomokay Medical Imaging Center, Clinic of Radiology, Bursa, Türkiye
[4]Isparta City Hospital, Department of Otorhinolaryngology and Head & Neck Surgery, Isparta, Türkiye

Abstract

Tonsillectomy is commonly performed in otorhinolaryngology clinics and is a relatively reliable procedure. Nevertheless, several complications, most notably postoperative bleeding, have been identified. Cervicofacial emphysema following tonsillectomy is exceedingly rare. This complication can lead to high-mortality outcomes, such as necrotizing fasciitis, mediastinitis, and pneumomediastinum. In this case report, we present the diagnosis and treatment of cervicofacial emphysema that developed on the third postoperative day in a 25-year-old female patient, along with a review of the relevant literature.

Keywords: Tonsillectomy, postoperative complications, cervicofacial emphysema, pneumomediastinum, case report

ORCID IDs of the authors:
V.A. 0000-0002-0050-4837
H.Y.0000-0002-5470-6784
Ç.I.0000-0002-7901-3757
E.T.0000-0001-7242-5698
B.B. 0000-0002-3620-2443

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Corresponding Author:
Vural Akın, MD;
vuralakin92@gmail.com

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Introduction

Tonsillectomy is among the most commonly performed procedures in otorhinolaryngology clinics. Although tonsillectomy is generally considered a safe operation, serious complications can occasionally occur (1,2). According to a study analyzing post-tonsillectomy complications in adult patients, the rate of any complication within the first 30 days postoperatively was found to be 21.46% (3). The most clinically significant complication after tonsillectomy is postoperative bleeding. Additionally, other well-defined complications include dental injuries, postoperative oropharyngeal infections, Grisel's syndrome, mandibular condyle fracture, and velopharyngeal insufficiency (1,4).

Cervicofacial emphysema is an extremely rare but potentially life-threatening complication of tonsillectomy (1,5). Cervicofacial emphysema can lead to infection in deep neck spaces, upper airway obstruction, pneumomediastinum, mediastinitis, or pneumothorax (5). In this case report, we present the diagnosis and treatment of cervicofacial emphysema following tonsillectomy in an adult patient, supported by a review of the literature.

Case Presentation

A 25-year-old female patient underwent tonsillectomy due to recurrent tonsillitis under general anesthesia in the otorhinolaryngology clinic. The tonsillectomy was performed extracapsularly, using bipolar cautery for dissection. Dissection of the right palatine tonsil was challenging due to significant adhesion of the tonsillar capsule to the adjacent muscle tissue. Pus flow was intermittently noted between the capsule and the muscle during dissection. After the tonsillectomy was completed, a defect approximately 2 mm in size was noted in the muscle tissue on the right side. This defect was primarily sutured and repaired using 4-0 Vicryl sutures. Positive pressure ventilation via mask was not applied after extubation. The patient was not discharged on the day of surgery due to the observed pus flow during the procedure and was started on intravenous ampicillin-sulbactam therapy at a dose of 4×1.5 g. On the third day of the antibiotic therapy, the patient reported swelling in her right cheek after coughing. On examination, crepitus was palpated extending from the superior border of the mandible, following the sternocleidomastoid muscle down to its mid-level. There was no increase in temperature or hyperemia. Endoscopic examination of the larynx was normal. Cervical and chest radiographs obtained showed that emphysema was confined to the neck. No defect was observed in the air column of the larynx or the trachea (Figure 1). Blood test results are presented in Table 1.

Upon the consultation of an infectious diseases and clinical microbiology specialist, intravenous ciprofloxacin at a dose of 2×400 mg was added to the treatment regimen due to its efficacy against Gram-negative bacteria. An antitussive was administered to prevent coughing. During inpatient follow-up, the patient's temperature, pulse, blood pressure, respiratory rate, and oxygen saturation remained stable. The facial swelling and palpable crepitus subsided, and no additional complaints were noted. Follow-up radiographs showed no increase in the extent of emphysema. On the seventh postoperative day, the patient was discharged with oral antibiotic therapy (amoxicillin-clavulanate 2×1 g and ciprofloxacin 2×500 mg). Antibiotic therapy was discontinued on the 14th day, as follow-up tests revealed no significant infectious findings and emphysema had resolved (Figure 2).

Informed consent for this case report was obtained from the patient.

Discussion

In a literature review conducted by Assiri et al. (6) in 2022, 32 cases of cervicofacial emphysema following tonsillectomy were identified. Of these cases, 11 were pediatric and 21 were adult patients. The most commonly reported symptom was swelling. Conventional radiographs were most commonly

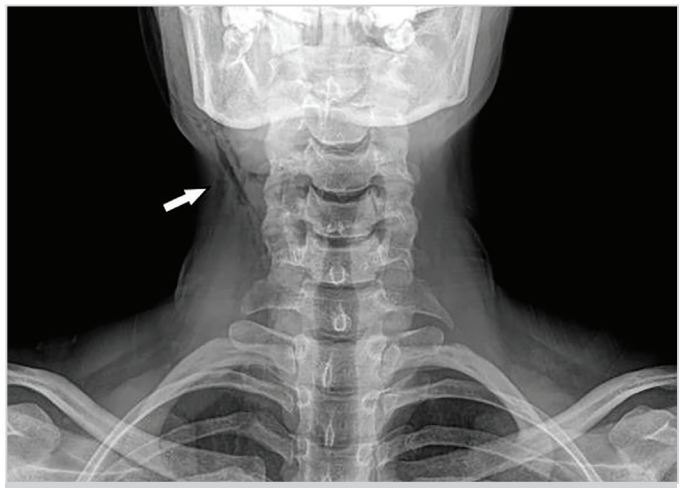

Figure 1a. Appearances compatible with emphysema predominantly concentrated in the right submandibular area of the neck (white arrow). No defect observed in the tracheal air column

Figure 1b. No defect observed in the laryngeal and tracheal air column

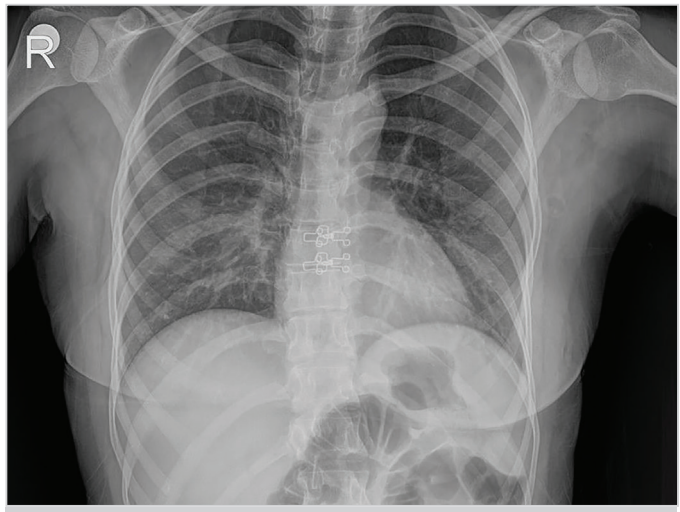

Figure 1c. No significant pathology observed in the chest on posteroanterior chest radiograph

used for diagnosis, followed by computed tomography. Antibiotic therapy was initiated in 25 patients. Additional treatment options included antitussives, steroids, laxatives, oxygen, and intravenous fluid support. Among the 32 reported cases, three required intubation, two patients underwent thoracotomy, and one required tracheotomy. The length of hospital stay ranged from 4 hours to 15 days. The authors noted that adult patients were at greater risk for emphysema, and that emphysema tended to occur later in adult cases (6).

The pathophysiology of this complication remains unclear, with several theories proposed (5,6). The first theory suggests that it may occur secondary to laryngeal or tracheal injury during intubation or high-pressure ventilation (1,6). The second theory posits that air leakage into deep neck spaces occurs as a result of damage to the tonsillar bed during the surgical procedure (1,6). Coughing, straining, and vomiting increase upper airway pressure, which predisposes for the development of emphysema (5). A third theory proposes that gases released by microorganisms may contribute to the development of emphysema (5).

The capsule of the palatine tonsils is a part of the pharyngobasilar fascia. The loose connective tissue between the capsule and the muscle facilitates easy dissection. The pharyngobasilar fascia covers the superior constrictor muscle, and deeper within, the buccopharyngeal fascia, which is part of the visceral layer of the middle layer of the deep cervical fascia, surrounds the pharyngeal muscles and separates the neck spaces (1). In extracapsular dissection, trauma to the buccopharyngeal fascia may allow air to enter the parapharyngeal space (1,2). A history of recurrent tonsillitis and peritonsillar abscess leads to fibrosis and adhesions in the tonsil tissue and underlying muscular layers. This condition can complicate dissection and increase the risk of subcutaneous emphysema (7). To prevent the development of subcutaneous emphysema, traumatic dissection should be avoided, and activities that increase pharyngeal pressure, such as coughing, sneezing, blowing the nose, or engaging in physical activities that raise pressure, should be avoided. Manual ventilation after extubation should also be avoided (1). In our case, the development of emphysema is thought to be consistent with the second theory due to difficulty during tonsil dissection on the affected side and the formation of a defect in the muscle tissue. The absence of difficulty during intubation and the development of emphysema on the third day following coughing further supports this mechanism.

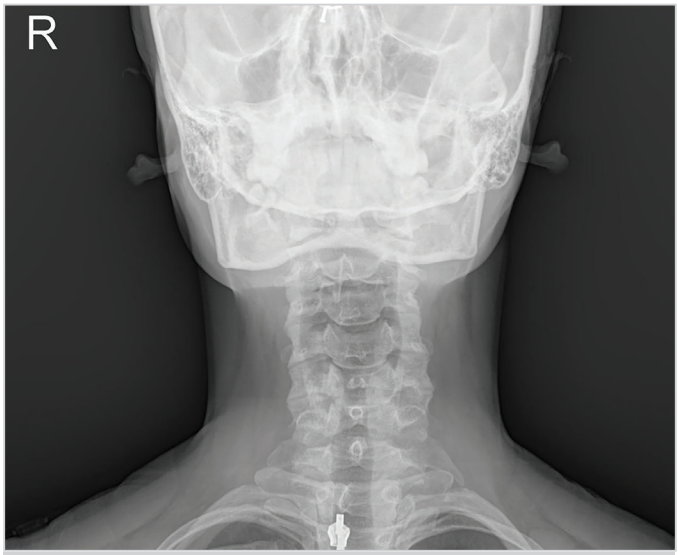

Figure 2. No findings supporting emphysema observed in the cervical radiograph

The most important finding from physical examination is the palpation of crepitus. Conventional radiographs are often used for diagnosis. The extent of emphysema can be thoroughly evaluated with computed tomography (1,2,5,8). The most important differential diagnosis is necrotizing fasciitis, which can be differentiated by the presence of infectious signs (1). In our case, since vital signs remained stable during hospitalization and radiographs showed limited emphysema, a computed tomography scan was not performed. Necrotizing fasciitis was not considered for primary differential diagnosis because of the absence of infectious signs.

The treatment of cervicofacial emphysema depends on its severity. Treatment is generally conservative as emphysema often resolves spontaneously (1,2,5). The extent of emphysema and whether the airway is affected should be regularly monitored (5). The patient should be closely followed up from a cardiopulmonary perspective. Broad-spectrum antibiotic therapy should be initiated due to the risk of mediastinitis and necrotizing fasciitis caused by oral cavity contamination (1). Patients are advised to avoid actions that increase pressure on the affected area, such as coughing, vomiting,

Table 1. C-reactive protein and complete blood count results during the follow-up period

Test	Postoperative day 3	Postoperative day 5	Postoperative day 7	Postoperative day 14	Reference range
Leukocyte (10^3 µ/L)	9.17	8.72	5.86	3.68	3.5-9.5
Neutrophil (10^3 µ/L)	6.28	5.67	3.79	1.98	1.8-6.3
Lymphocyte (10^3 µ/L)	2.24	2.58	1.65	1.40	1.1-3.2
Monocyte (10^3 µ/L)	0.33	0.36	0.38	0.22	0.1-0.6
C-reactive protein (mg/L)	25.5	30.3	20.4	9.7	0-5

and straining, until the emphysema resolves. If necessary, anti-tussives, laxatives, and antiemetics may be added to the treatment (2). In cases where respiration is affected, intubation or tracheotomy may be required. Although rarely, thoracotomy has been reported (1). If a significant defect is present in the surgical area, it is also recommended to repair it (1,5). In the presented case, intravenous antibiotic therapy was initiated before the development of emphysema due to the presence of pus during the intraoperative process. After the development of emphysema, the therapy spectrum was expanded based on the consultation of an infectious diseases and clinical microbiology specialist. An anti-tussive was added to treatment due to the development of emphysema following coughing. Because no new defect was observed, no additional surgical intervention was considered.

Conclusion

Tonsillectomy is a common procedure in otolaryngology clinics. Cervicofacial emphysema is a rare but potentially fatal complication of tonsillectomy. It is important for otolaryngologists to be aware of this complication, as early diagnosis and treatment are crucial.

Ethics

Informed Consent: Informed consent for this case report was obtained from the patient.

Footnotes

Authorship Contributions

Surgical and Medical Practices: V.A., Ç.I., E.T., Concept: V.A., H.Y., Ç.I., E.T., B.B., Design: V.A., H.Y., Ç.I., E.T., B.B., Data Collection and/or Processing: V.A., E.T., Analysis and/or Interpretation: V.A., H.Y., E.T., Literature Search: V.A., H.Y., Ç.I., E.T., B.B., Writing: V.A., H.Y., Ç.I., E.T., B.B.

Conflict of Interest: There is no conflict of interest to disclose.

Financial Disclosure: The authors declared that this study has received no financial support.

Main Points

- Cervicofacial emphysema is a rare but potentially fatal complication of tonsillectomy.
- Patients with cervicofacial emphysema require close monitoring.
- It is important for otolaryngologists to be aware of this complication, as early diagnosis and treatment are crucial.

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Unusual Case of Fishhook Lodged in the Neck: A Case Report and Literature Review

Case Report

Anne Marie Daou[1], Omar Aboul Hosn[1], Jad Hosri[1], Afif Mufarrij[2], Zeina Korban[1]

[1]American University of Beirut Medical Center, Clinic of Otolaryngology-Head and Neck Surgery, Beirut, Lebanon
[2]American University of Beirut Medical Center, Clinic of Emergency Medicine, Beirut, Lebanon

Abstract

Fishing constitutes a widely practiced sport considered pleasant and harmless. Yet, there are specific risks associated with fishhook injuries specifically in the neck. To the best of our knowledge, only few articles on fishhook injury in the neck were reported in the literature. We present the case of a 57-year-old previously healthy male coming to the emergency department with a J-type fishhook accidentally lodged in his neck. Prompt management with bedside removal of the sharp foreign body from the neck was achieved with no subsequent complications. In this case report, literature review of fishhook types and injuries as well as management options of such penetrating injuries will be detailed.

Keywords: Neck injuries, penetrating trauma, penetrating wounds, foreign bodies, fishhooks, wound management, case report

ORCID IDs of the authors:
A.M.D. 0009-0004-2794-7795
O.A.H. 0009-0000-0891-2395
J.H. 0000-0002-7781-3486
A.M. 0000-0001-7323-4359
Z.K .0000-0003-3233-9642

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Corresponding Author:
Zeina Korban, MD;
zk27@aub.edu.lb

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Introduction

Fishing is a widely practiced and seemingly harmless sport, yet fishhook incidents present specific risks. These injuries most commonly affect the hands but can also impact the head and neck, with the eyes and nose being the most frequently involved areas. Notably, fishhook injuries to the neck, which can be classified as penetrating anterior neck injuries, have not been documented in the literature. The neck's intricate anatomy includes essential, relatively unprotected structures, rendering it susceptible to severe vascular injuries such as occlusion, dissection, pseudoaneurysm, blood extravasation, or arteriovenous fistula formation. Approximately 25% of penetrating neck injuries involve arterial damage (1). Currently, no international consensus guidelines exist for managing these injuries, and published guidelines typically emphasize traditional zonal approaches.

Case Presentation

The authors present the case of a 57-year-old previously healthy male who arrived at the emergency department with a fishhook injury to the neck. The injury occurred while the patient was walking by the seaside, when a fisher accidentally cast his rod in the wrong direction, lodging the hook in the patient's neck. The patient arrived at the hospital ambulating and breathing comfortably, with stable

vital signs. He reported no neck pain, drooling, dysphonia, respiratory distress, dysphagia, or odynophagia.

Physical examination revealed a palpable fishhook on the right side of the neck, with its shank external to the skin and the sharp, curved part palpable in the subcutaneous plane Figure 1. There was no palpable collection or rapidly expanding hematoma. A bedside flexible laryngoscopy showed no evidence of a foreign body in the upper airway.

Neck X-ray and ultrasound (US) were performed Figures 2-3. The US revealed a J-type fishhook traversing the deep subcutaneous fat planes medially, with its tip oriented anteriorly and near a small vein, likely a branch of the anterior jugular vein. No major vessels were in proximity to the hook, and no hematoma was detected Figure 4.

The hook was removed in the radiology room under local anesthesia with lidocaine injections to the anterior neck. The hook was advanced to pierce the skin, externalizing its tip. The sharp tip and barb were cut and separated from the rest of the hook using a ring cutter and forceps, and the remaining smooth part was then retrieved Figures 5-6. A post-procedural US confirmed no acute injury to major vessels and no hematoma. The patient tolerated the procedure with minimal discomfort. He received a tetanus vaccine and was discharged home the same day with a course of amoxicillin-clavulanic acid.

Figure 1. Fishhook in the neck upon patient presentation to the emergency department

Figure 2. Neck X-ray in sagittal view of the fishhook stuck in the anterior subcutaneous fat of the neck, midline position. Note that the upper airways are intact

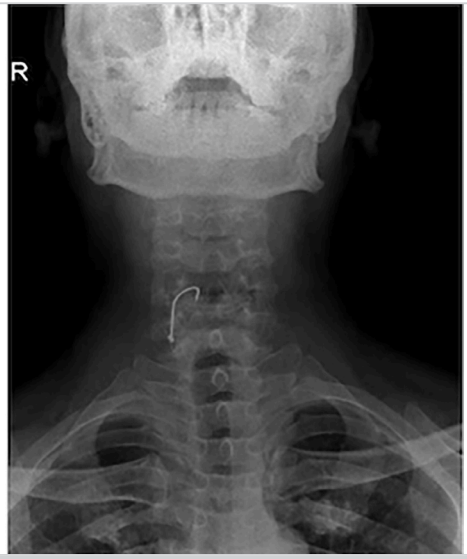

Figure 3. Neck X-ray in coronal view of the fishhook stuck in the anterior subcutaneous fat of the neck, midline position. Note that the upper airways are intact

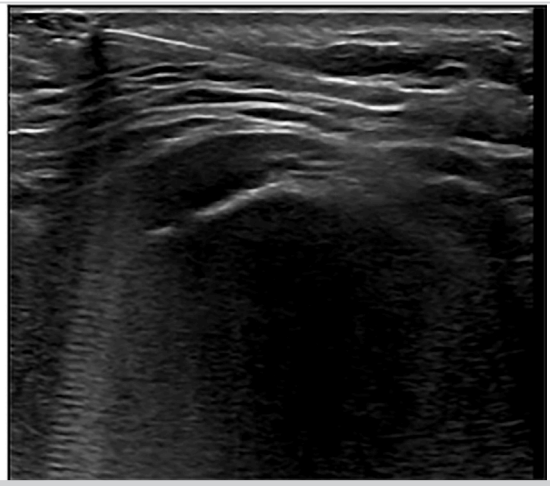

Figure 4. Ultrasound of the neck done prior to removal of fishhook showing the sharp foreign body penetrating in the subcutaneous plane with its tip inside

Figure 5. Successful advancement and cutting technique of the fishhook

Figure 6. J-type fishhook with its barb

Discussion

Fishing carries inherent risks particularly due to the sharply curved metal hooks used. There are various types of fishing hooks, including bait hooks, J hooks, circle hooks, and treble hooks, which can be equipped with or without barbs near the tip to secure the fish on the line. Barbless fishhooks reduce the likelihood of severe injuries (1). In our case, the neck was injured by a barbed J hook. Despite their superficial appearance, barbed hooks introduce a significant risk of dangerous internal injuries, especially when located near vital structures such as vessels, nerves, or tendons (2).

Historical reports like the one by Townsend (3) in 1848 highlight the challenges of removing fishhooks from sensitive areas such as the neck. Townsend's (3) description of extracting a fishhook from the neck underscores the persistent need for effective removal techniques when hooks embed near critical structures. The most commonly affected body part is the hand, followed by the head and eyes (2, 4). Rare instances of fishhook injuries to the nose and pinna have been reported only twice and once, respectively (4-6). An unusual case reported by Sa'adudeen Idris et al. (7) in 2023 involved the unintentional ingestion of a fishhook by an elderly woman, with the hook becoming entangled over the left greater horn of the hyoid bone, necessitating surgical removal.

Although there are no established guidelines for safe fishhook removal, the literature describes four main techniques: retrograde, string pull, needle cover, and advance/cut techniques. The latter involves advancing the hook through the skin surface, clipping the barb with ring cutters or flattening it with pliers, and then performing a smooth retrograde removal of the fishhook. The choice of removal method depends on factors such as the type of fishhook, its location, and the depth of tissue penetration (4). A single study assessing success rates identified the advance and cut technique as the most effective (8). Remarkably, fishhook injuries involving the anterior neck remain unreported.

Penetrating neck injuries, accounting for 5% to 10% of all trauma cases, have a mortality rate up to 10%, with vascular injuries being the leading cause of death (9). Injuries breaching the platysma layer are considered more severe. Anatomically, these injuries are categorized into three zones, each requiring different management strategies. Zone I, spanning from the clavicles to the cricoid cartilage, is the most serious and potentially lethal. Zone II, extending from the cricoid cartilage to the angle of the mandible, is the most commonly injured and is the easiest to access surgically, with a low risk of adverse outcomes. Zone III, covering the area between the angle of the mandible and the skull base, is challenging to examine and surgically explore. Management of these injuries depends on the zone involved and the specific vascular and anatomical structures within each zone (10).

Our case involved a zone II penetrating injury that reached the subcutaneous plane. Although the tip of the hook was near a small branch of the anterior jugular vein, there was no vascular injury or expanding hematoma. The patient remained asymptomatic during serial examinations. Given the absence of expanding or pulsatile hematoma, bleeding, neurological deficits, hemoptysis, or dysphonia, we opted for bedside fishhook removal. The advance and cut technique was chosen due to its documented success rates in the literature. The decision was supported by the patient's hemodynamic stability, the superficial positioning of the fishhook, and the use of duplex ultrasonography for guidance.

Conclusion

Fishhook lodged in the neck is an extremely rare injury with only few previously reported cases. Prompt recognition and appropriate management of such injuries are crucial to prevent complications such as bleeding, pulsatile hematoma, or neurological injury. In our case, US-guided removal was performed, with pre- and post-procedural radiologic evaluation of the neck. In severe cases with a higher risk of major vessel injury, surgical removal in a controlled setting is recommended.

Ethics

Informed Consent: Since patient-identifying information were not presented in this case report, only verbal informed consent was obtained from the patient's caregivers to participate in this study.

Footnotes

Authorship Contributions

Surgical and Medical Practices: A.M., Z.K., Concept: A.M., Z.K., Design: A.M., Z.K., Data Collection and/or Processing: A.M.D., Analysis and/or Interpretation: A.M.D., O.A.H., Literature Search: A.M.D., O.A.H., J.H., Writing: A.M.D., O.A.H., J.H.

Conflict of Interest: There is no conflict of interest to disclose.

Financial Disclosure: The authors declared that this study has received no financial support.

Main Points

- Fishing carries rare but serious risks, particularly related to fishhook injuries in the neck.
- The fishhook can be safely extracted at the bedside without the need for more invasive procedures.
- Careful removal was crucial in avoiding complications such as infection, damage to surrounding structures, or further injury.
- Monitoring for any delayed complications and providing appropriate wound care are essential to ensure the patient's full recovery.

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