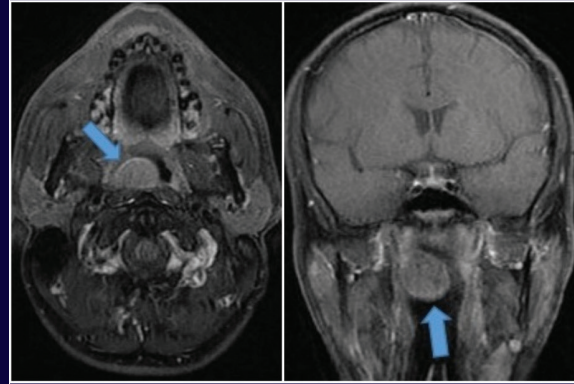


Turkish Archives of Otorhinology



Official Journal of the
Turkish Otorhinology
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Türk Otorinolarenoloji Arşivi



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Turkish Archives of Otorhinology

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Aims and Scope

The Turkish Archives of Otorhinology (Turk Arch Otorhinology) is the scientific, peer-reviewed, open-access journal of the Turkish Otorhinology-Head and Neck Surgery Society since 2001. The journal comprises four issues as March, June, September and December in a volume, and it is published quarterly every year. The journal's publication language is English.

The aim of the journal is to publish qualified original clinical, experimental and basic researches on ear, nose, throat, head and neck diseases and surgery, reviews that contain sufficient amount of source data conveying the experiences of experts in a particular field, case reports, video articles and original images of rare clinical pictures which would shed light on the clinical practice and which were not previously published, letters from the readers and experts concerning the published studies, articles about general practice and subject of the journal with historical content, memories of scientific significance, educative and catechetical manuscripts about medical deontology and publication ethics.

The target audience of the journal includes academic members, specialists, residents and other relevant health care professionals in the field of ear, nose, throat, and head and neck disorders and surgery.

The editorial and publication processes of the journal are shaped in accordance with the guidelines of the International Committee of Medical Journal Editors (ICMJE), World Association of Medical Editors (WAME), Council of Science Editors (CSE), Committee on Publication Ethics (COPE), European Association of Science Editors (EASE), and National Information Standards Organization (NISO). The journal is in conformity with the Principles of Transparency and Best Practice in Scholarly Publishing.

Turkish Archives of Otorhinology is indexed in PubMed, PubMed Central, Web of Science (Emerging Sources Citation Index), ULAKBIM TR Index, EBSCO, GALE, CINAHL, J-Gate and ProQuest.

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Our journal's Abstracting/Indexing services store essential information about articles. In addition, some of our journals' Abstracting/Indexing services archive metadata about the article and electronic versions of the articles. In this way, copies of articles are presented to the scientific

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community through these systems as an alternative to journals. This journal's archive has been backed up by PubMed Central (PMC) as from 2015 publications.

Author Self-Archiving Policy

Authors are permitted and encouraged to post their articles on personal and institutional websites after publication (while providing full bibliographic details and a link to the original publication).

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Turkish Archives of Otorhinolaryngology

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Instructions to Authors

CONTEXT

The Turkish Archives of Otorhinolaryngology (Turk Arch Otorhinolaryngol) is a scientific, open access periodical published by independent, unbiased, and double-blinded peer-review principles. The journal is the official publication of the Turkish Otorhinolaryngology Head and Neck Surgery Society, and published quarterly in March, June, September and December. The publication language of the journal is English.

The aim of the journal is to publish qualified original clinical, experimental and basic research on ear, nose, throat, head and neck diseases and surgery, reviews that contain a sufficient amount of source data conveying the experiences of experts in a particular field, case reports and original images of rare clinical pictures which would shed light on the clinical practice and which were not previously published, letters from the readers and experts concerning the published studies, articles about general practice and subject of the journal with historical content, memories of scientific significance, educative and catechetical manuscripts about medical deontology and publication ethics.

EDITORIAL AND PUBLICATION PROCESS

The editorial and publication process of the Turkish Archives of Otorhinolaryngology are shaped in accordance with the guidelines of the International Committee of Medical Journal Editors (ICMJE), World Association of Medical Editors (WAME), Council of Science Editors (CSE), Committee on Publication Ethics (COPE), European Association of Science Editors (EASE), and National Information Standards Organization (NISO). The journal is in conformity with the Principles of Transparency and Best Practice in Scholarly Publishing.

Originality, high scientific quality, and citation potential are the most important criteria for a manuscript to be accepted for publication. Manuscripts submitted for evaluation should not have been previously presented or already published in an electronic or printed medium. The journal should be informed of manuscripts that have been submitted to another journal for evaluation and rejected for publication. The submission of previous reviewer reports will expedite the evaluation process. Manuscripts presented in a meeting should be submitted with detailed information on the organization, including the name, date, and location of the organization.

PEER REVIEW PROCESS

Manuscripts submitted to the Turkish Archives of Otorhinolaryngology will go through a double-blind peer-review process. Each submission will be reviewed by at least two external, independent peer reviewers who are experts in their fields in order to ensure an unbiased evaluation process. The editorial board will invite an external and independent editor to manage the evaluation processes of manuscripts submitted by editors or by the editorial board members of the journal. The Editor in Chief is the final authority in the decision-making process for all submissions. For more detailed information, please read Ethical Policy page of the Journal.

Preprint

The Turkish Archives of Otorhinolaryngology does not consider preprint publications as prior publications. In other words, authors are allowed to present and discuss their findings on a non-commercial preprint server before submission to a journal.

Authors must provide the journal with the preprint server deposition of their article accompanying its DOI during initial submission. If the article is published in the Turkish Archives of Otorhinolaryngology, it is the responsibility of the authors to update the archived preprint and link it to the published version of the article.

AUTHORSHIP

Each person listed as an author should fulfil the authorship criteria recommended by the International Committee of Medical Journal Editors. The ICMJE recommends that authorship is based on the following four criteria:

Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; AND

Drafting the work or revising it critically for important intellectual content; AND

Final approval of the version to be published; AND

Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

In addition to being accountable for the parts of the work he/she has done, an author should be able to identify which co-authors are responsible for specific other parts of the work. Also, the authors should have confidence in the integrity of the contributions of their co-authors.

All those designated as authors should meet all four criteria for authorship, and all who meet the four criteria should be identified as authors. Those who do not meet all four criteria should be acknowledged in the title page of the manuscript.

Author Affiliations

Authors are expected to state the institutions which they affiliated in the time of the study. Their current affiliation can be added to the article as the corresponding address. Change of affiliation requests will not be implemented after submission. The Turkish Archives of Otorhinolaryngology requires corresponding authors to submit a signed and scanned version of the Authorship Contribution Form during the initial submission process to act appropriately on authorship rights and to prevent ghost or honorary authorship. If the editorial board suspects a case of "gift authorship", the submission will be rejected without further review. As part of the submission of the manuscript, the corresponding author should also send a short statement declaring that he/she accepts to undertake all the responsibility for authorship during the submission and review stages of the manuscript.



Instructions to Authors

Change of Authorship

The Turkish Archives of Otorhinolaryngology reviews the authorship according to the author's declaration in the Title Page; thus, it is the authors' responsibility to send the final order of the complete author names. Requests in the change of authorship (e.g. removal/addition of the authors, change in the order etc.) after submission are subject to editorial approval. Editorial Board will investigate these kind of cases and act following COPE flowcharts.

Change of authorship requests should be submitted to the Editorial Office with an official letter stating the change's reasons. The letter must be signed by all authors and include their approval on the change in authorship. If the request is approved by the Editorial Board, authors need to submit a new Copyright Agreement Form according to the final order list.

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SUBMISSION REQUIREMENTS

- Cover Letter,
- ICMJE Conflict of Interest Statement Form for all contributing authors,
- A separate title page (Title Page should be submitted with all manuscripts and should include the title of the manuscript, name(s), affiliation(s), major degree(s) and ORCID ID of the author(s). The name, address, telephone (including the mobile phone number) and fax numbers and e-mail address of the corresponding author should be clearly listed. Grant information and other sources of support should also be included. Individuals who contributed to the preparation of the manuscript but did not fulfil the authorship criteria should also be acknowledged on the title page),
- Abstract divided into appropriate sections,
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- Article divided into appropriate sections,
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- The Copyright Agreement and Acknowledgement of Authorship Form (Please submit a wet-signed and scanned copy of the Copyright Transfer Form with your submission),
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- Figures (Figures should be submitted as standalone images through the submission system in .JPG or .TIFF format),
- Ethics Committee Approval Statement (with decision/file no, date and name of the institution, for original articles),

MANUSCRIPT PREPARATION

The manuscripts should be prepared in accordance with ICMJE Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals. Authors are required to

The presentation of the article types must be designed in accordance with trial reporting guidelines:

Human research: Helsinki Declaration as revised in 2013



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Instructions to Authors

Systematic reviews and meta-analyses: PRISMA guidelines

Case reports: the CARE case report guidelines

Clinical trials: CONSORT

Animal studies: ARRIVE and Guide for the Care and Use of Laboratory Animals

Diagnostic accuracy: STARD Guidelines

Non-randomized public behaviour: TREND

Manuscripts can only be submitted through the journal's online manuscript submission and evaluation system, available at www.turkarchotolaryngol.net. Manuscripts submitted via any other medium and submissions by anyone other than one of the authors will not be evaluated.

Manuscripts submitted to the journal will first go through a technical evaluation process where the editorial office staff will ensure that the manuscript has been prepared and submitted in accordance with the journal's guidelines. Submissions that do not conform to the journal's guidelines will be returned to the submitting author with technical correction requests.

Authors are required to submit the following:

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ICMJE Potential Conflict of Interest Disclosure Form (should be filled in by all contributing authors) during the initial submission. These forms are available for download at turkarchotolaryngol.net.

Preparation of the Manuscript

Title page: A separate title page should be submitted with all submissions, and this page should include:

The full title of the manuscript, as well as a short title (running head) of no more than 50 characters,

Name(s), affiliations, highest academic degree(s), and ORCID IDs of the author(s),

Grant information and detailed information on the other sources of support,

Name, address, telephone (including the mobile phone number), and e-mail address of the corresponding author,

Acknowledgement of the individuals who contributed to the preparation of the manuscript but who do not fulfil the authorship criteria.

Abstract: An abstract should be submitted with all submissions except for Letters to the Editor. The abstract of Original Articles should be structured with subheadings (Objective, Methods, Results, and Conclusion). Please check Table 1 below for word count specifications.

Keywords: Each submission must be accompanied by a minimum of four to a maximum of eight keywords for subject indexing at the end of the abstract. The keywords should be listed in full without abbreviations.

The keywords should be selected from the National Library of Medicine, Medical Subject Headings database.

Main Points: All submissions except letters to the editor and clinical images should be accompanied by 3 to 5 "main points" which should emphasize the most noteworthy results of the study and underline the principle message that is addressed to the reader. This section should be structured as itemized to give a general overview of the article. Since "Main Points" target the experts and specialists of the field, each item should be written as plain and straightforward as possible.

Manuscript Types

Original Articles: This is the most essential type of article since it provides new information based on original research. The main text of original articles should be structured with Introduction, Methods, Results, Discussion, and Conclusion subheadings. Please check Table 1 for the limitations for Original Articles.

Statistical analysis to support conclusions is usually necessary. Statistical analyses must be conducted in accordance with international statistical reporting standards (Altman DG, Gore SM, Gardner MJ, Pocock SJ. Statistical guidelines for contributors to medical journals. *Br Med J* 1983; 7; 1489-93). Information on statistical analyses should be provided with a separate subheading under the Methods section, and the statistical software that was used during the process must be specified.

Units should be prepared in accordance with the International System of Units (SI).

Clinical Trials

Turkish Archives of Otorhinolaryngology adopts the ICMJE's clinical trial registration policy, which requires that clinical trials must be registered in a publicly accessible registry that is a primary register of the WHO International Trials Registry Platform (ICTRP) or in ClinicalTrials.gov.

Instructions for the clinical trials are listed below:

A clinical trial registry is only required for the prospective research projects that study the relationship between a health-related intervention and an outcome by assigning people.

To have their manuscript evaluated in the journal, the author should register their research to a public registry at or before the time of first patient enrollment.

Based on most up to date ICMJE recommendations, the Turkish Archives of Otorhinolaryngology accepts public registries that include a minimum acceptable 24-item trial registration dataset.

Authors are required to state a data sharing plan for the clinical trial registration. Please see details under "Data Sharing" section.

For further details, please check ICMJE Clinical Trial Policy.

Data Sharing

As of 1 January 2019, a data-sharing statement is required for the registration of clinical trials. Authors are required to provide a data



Instructions to Authors

sharing statement for the articles that reports the results of a clinical trial. The data sharing statement should indicate the items below according to the ICMJE data sharing policy:

Whether individual de-identified participant data will be shared

What data, in particular, will be shared

Whether additional, related documents will be available

When the data will be available, and for how long

By what access criteria will be shared

Authors are recommended to check the ICMJE data sharing examples at <http://www.icmje.org/recommendations/browse/publishing-and-editorial-issues/clinical-trial-registration.html>

While submitting a clinical trial to Turkish Archives of Otorhinolaryngology:

Authors are required to make registration to a publicly accessible registry according to ICMJE recommendations and the instructions above.

The name of the registry and the registration number should be provided in the Title Page during the initial submission.

Data sharing statement should also be stated on the Title Page even the authors do not plan to share it.

The clinical trial and data sharing policy of the journal will be valid for the articles submitted from 1 January 2021.

Editorial Comments: Editorial comments aim to provide a brief critical commentary by reviewers with expertise or with a high reputation in the topic of the research article published in the journal. Authors are selected and invited by the journal to provide such comments. Abstract, Keywords, Tables, Figures, Images, and other media are not included.

Review / Systematic Review Articles: Reviews prepared by authors who have extensive knowledge on a particular field and whose scientific background has been translated into a high volume of publications with a high citation potential are welcomed. These authors may even be invited by the journal. Reviews should describe, discuss, and evaluate the current level of knowledge of a topic in clinical practice and should guide future studies. The main text should contain Introduction, Clinical and Research Consequences, and Conclusion sections. While submitting your Review, please confirm that your manuscript is a systematic review and include a statement that researchers have followed the PRISMA guidelines.

Please check Table 1 for the limitations for Review / Systematic Review Articles.

Video Article: Videos should be up to 30 minutes in duration. The video must include audio narration explaining the procedure. All text and audio in the video must be in English. Audio must include narration in clear, grammatically correct English. Videos must be clear, in focus, and without excessive camera movement. Radiographs and other material must not contain any patient-identifiable information. Limited number

of slides incorporated into video may be included to provide details of patient history, clinical and laboratory findings.

Video articles should include:

1) Copyright Transfer and Author Declaration Statement Form: This form must indicate that "Patients' Informed Consent Statement" is obtained.

2) Title Page

3) **Summary:** Summary should point out critical steps in the surgery up to 500 words. This part was published as an abstract to summarize the significance of the video and surgical techniques. The author(s) may add references if it is required.

5) **Video:** Please upload your video to turkarchotolaryngol.net using online submission system. Accepted video formats are Windows Media Video (WMV), AVI, or MPEG (MPG, MPEG, MP4). High-Definition (HD) video is preferred.

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SL, Barlett JG, Blacklow NR, editors. Infectious Diseases. Philadelphia: Lippincott Williams; 2004.p.2290-308.

Books with a Single Author: Sweetman SC. Martindale the complete drug reference. 34th ed. London: Pharmaceutical Press; 2005.

Editor(s) as Author: Huizing EH, de Groot JAM, editors. Functional reconstructive nasal surgery. Stuttgart-New York: Thieme; 2003.

Conference Proceedings: Bengissson S. Sothemin BG. Enforcement of data protection, privacy and security in medical informatics. In: Lun KC, Degoulet P, Piemme TE, Rienhoff O, editors. MEDINFO 92.

Proceedings of the 7th World Congress on Medical Informatics; 1992 Sept 6-10; Geneva, Switzerland. Amsterdam: North-Holland; 1992. pp.1561-5.

Scientific or Technical Report: Cusick M, Chew EY, Hoogwerf B, Agrón E, Wu L, Lindley A, et al. Early Treatment Diabetic Retinopathy Study Research Group. Risk factors for renal replacement therapy in the Early Treatment Diabetic Retinopathy Study (ETDRS), Early Treatment Diabetic Retinopathy Study Kidney Int: 2004. Report No: 26.

Thesis: Yılmaz B. Ankara Üniversitesindeki öğrencilerin beslenme durumları, fiziksel aktiviteleri ve beden kitle indeksleri kan lipidleri arasındaki ilişkiler. H.Ü. Sağlık Bilimleri Enstitüsü, Doktora Tezi. 2007.

Manuscripts Accepted for Publication, Not Published Yet: Slots J. The microflora of black stain on human primary teeth. Scand J Dent Res. 1974.

E-pub Ahead of Print Articles: Cai L, Yeh BM, Westphalen AC, Roberts JP, Wang ZJ. Adult living donor liver imaging. Diagn Interv Radiol. 2016 Feb 24. doi: 10.5152/dir.2016.15323. [Epub ahead of print].

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Total Transcanal Endoscopic Ear Surgery for Cholesteatoma

Original Investigation

Abdullah Dalğıç, Gökçe Aksoy Yıldırım, Mehmet Ekrem Zorlu, Orçun Delice, Abdulhalim Aysel

Department of Otolaryngology and Head & Neck Surgery, İzmir Bozyaka Training and Research Hospital, University of Health Sciences Turkey, İzmir, Turkey

Abstract

Objective: This study aimed to evaluate the outcomes of total transcanal endoscopic cholesteatoma surgery.

Methods: Twenty-seven cholesteatoma patients that had undergone transcanal endoscopic ear surgery (TEES) were included in the study. Age, sex, operation date of patients, operated side, need for ossiculoplasty, graft material, and surgical technique were recorded. All patients were evaluated through otoscopic, endoscopic, and audiological examinations and followed up for at least five months after surgery. All patients were staged using the European Academy of Otolology and Neurotology/Japan Otological Society (EAONO/JOS) Staging System on Middle Ear Cholesteatoma.

Results: Mean age of the patients was 36.4 years (range, 4–67 years). According to the EAONO/JOS Staging System, 11 patients were stage 1, while 11 were stage 2, and five were stage 3. Two had lateral semicircular canal defect, one had facial canal dehiscence, and one had oval window defect. The average follow-up period was 19 months (range, 5–41 months), during which two patients experienced retraction pocket and hearing loss and one patient had perforation. One patient underwent revision surgery during follow-up and no recurrence or residual cholesteatoma was observed. The preoperative and postoperative air–bone gaps were 25.14±13.93 dB and 22.22±12.64 dB with no significant difference.

Conclusion: TEES is a minimally invasive and safe procedure with low complication and recurrence rates. As with all surgical procedures, experience is essential, and as experience increases, the capability to perform endoscopic otologic surgery on more complex cases may become possible.

Keywords: Cholesteatoma, endoscopic surgery, otologic surgical procedures, tympanoplasty, mastoidectomy

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Presented in: This study has been presented at the 42nd Turkish National Congress of Otolaryngology Head and Neck Surgery, November 3-7, 2021, Cyprus.



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Introduction

Cholesteatoma is defined as the growth of keratinizing squamous epithelium with the keratin debris and surrounding inflammatory reaction. Cholesteatoma is classified as congenital or acquired (1).

Primary acquired cholesteatoma occurs when the retraction pocket originating from the tympanic membrane reaches the tympanic cavity and then advances toward the sinus tympani, facial recess, and attic (2). In advanced cases, cholesteatoma extends into the mastoid cavity. Surgical

treatment primarily aims to eradicate the disease and ensure a healthy middle ear cavity and to restore hearing if possible.

The post-auricular microscopic approach with the canal wall-down (CWD) mastoidectomy has been one of the most popular techniques for the treatment of cholesteatoma. Recently, however, minimally invasive approaches are becoming more popular. Tympanoplasties with or without cholesteatoma can be performed with endoscopes in a minimally invasive manner. Endoscopes have been used in otology since the 1960s, although initially for diagnosis (3). In the 1990s, their use spread to ear surgeries in addition to microscopes as an auxiliary tool. Recently, many otologic surgeries such as myringoplasty, tympanoplasty, stapedectomy, and cholesteatoma surgery are being performed with endoscopes alone. Endoscopes provide wider visualization, and with 0° and angled telescopes that can reach hidden regions such as the facial recess, the sinus tympani, the hypotympanum or the anterior attic (4). Endoscopic ear surgery (EES) does not require soft tissue incisions like postauricular or endaural incisions aside from harvesting graft. However, this method requires intense training due to its disadvantages such as one-hand surgery, need for good hemostasis, absence of depth perception due to two-dimensional view, and risk of thermal injury (5). In this study, we aimed to evaluate the outcomes of transcanal endoscopic cholesteatoma surgery in our clinic.

Methods

Only cholesteatoma patients who had undergone transcanal endoscopic ear surgery (TEES) were included in the study. Ethical approval was obtained from University of Health Sciences Turkey, İzmir Bozyaka Training and Research Hospital Clinical Research Ethics Committee (decision no: 2021/108, date: 23.06.2021). All patients were informed about the objective of the study and signed the written consent form. Using the data from patient records, we reviewed their complaints and the results of their otoscopic and audiological examinations done after surgery. All patients were followed up for at least five months after surgery. All operations were performed by the senior author with a rigid endoscope (3 mm, 0°, 30°, 15-cm lens) under general anesthesia. We recorded the age, sex, operation date, side, need for ossiculoplasty, graft material and surgical technique, and audiogram findings before and five months after surgery. Air-bone gap (ABG) closure and recovery levels based on each frequency (500, 1000, 2000, 4000, 8000 dB) were evaluated in the audiological results.

In 2018, EES was classified by Cohen et al. (6) as follows: EES Class 1, endoscopic inspection without dissection; EES Class 2, mixed use of the endoscope; and EES Class 3, total transcanal endoscopic surgery. In our study, all patients were treated with EES Class 3 (TEES). All video recordings of

the surgeries were reviewed. The origin of cholesteatoma was identified as congenital, pars tensa, pars flaccida, or secondary to a tensa perforation. The European Academy of Otolaryngology and Neurotology/Japan Otological Society (EAONO/JOS) Staging System on Middle Ear Cholesteatoma was used to categorize the stages of cholesteatoma (7): Stage 1, cholesteatoma localized in the primary site [the site of cholesteatoma origin, i.e., the attic (A) for pars flaccida cholesteatoma; the tympanic cavity (T) for pars tensa cholesteatoma, congenital cholesteatoma, and cholesteatoma secondary to a tensa perforation]; stage 2, cholesteatoma involving two or more sites; stage 3, cholesteatoma with extracranial complications or pathologic conditions; and stage 4, cholesteatoma with intracranial complications (7).

During follow-up, all patients were monitored for postoperative complications, recurrence, and residual cholesteatoma. Diffusion-weighted magnetic resonance imaging (MRI) was used for monitoring recurrence in suspected cases after the otoscopic and endoscopic examinations.

Statistical Analysis

Data were analyzed using the SPSS software for Windows (v22.0; IBM Armonk, NY, USA). Individual and aggregate data were summarized using descriptive statistics including mean, standard deviations, and medians (minimum-maximum), as well as frequency distributions and percentages. Paired t-test was performed to compare the means. P values <0.05 were considered statistically significant.

Results

A total of 27 patients [14 females and 13 males with a mean age of 36.4 years (range, 4–67 years)] that had undergone TEES were included in the study (Table 1). Of them, 19 patients were operated on in the left ear and eight patients in the right ear. The origin of cholesteatoma was congenital cholesteatoma in two patients, secondary to a tensa perforation in three patients, pars flaccida cholesteatoma in 15 patients, and pars tensa cholesteatoma in seven patients (Figure 1). According to the EAONO/JOS Staging System on Middle Ear Cholesteatoma, 11 patients were in stage 1, 11 patients in stage 2, and five patients in stage 3 (Table 2).

Mean hospitalization was 2.4 days, ranging from one to seven days. Two patients had lateral semicircular canal fistula (LSCF) (Figure 2), one had facial canal dehiscence, one had oval window defect, and three had tympanosclerosis. In terms of perioperative complications, two patients had chorda tympani damage and one patient experienced bleeding due to a high jugular bulb. For tympanoplasty, perichondrium and cartilage graft were used in all patients. For ossicular reconstruction, type 1 tympanoplasty was performed in four patients, type 2 tympanoplasty in 15 patients, and type 3

tyimpanoplasty in eight patients. However, ossiculoplasty was not performed on four patients. Of all surgical operations, 23 were primary surgery and four were revision surgery. Ossicular reconstructions was performed using total ossicular reconstruction prosthesis (TORP) in six patients, incus in 13 patients, partial ossicular reconstruction prosthesis (PORP) in three patients, and bone cement in one patient (Figure 3).

Canal wall-up tympanoplasty (CWU) was performed in 26 patients, and CWD tympanoplasty in one patient. In CWU tympanoplasty, reconstruction was done with cartilage after atticotomy (Figure 4). Obliteration with cartilage and fascia was performed in only one patient who underwent CWD tympanoplasty. Average surgical time was 164 minutes (range, 116-minutes). The average follow-up period was 19 months (range, 5–41 months). In the follow-up period, retraction pocket and hearing loss were noted in two patients and a 2-mm size perforation in one patient. Diffusion-weighted MRI was used in two cases who were suspected of recurrence. However, there were no high-signal-intensity in diffusion-weighted MRI scans of these patients. Revision surgery was done in one patient who underwent TEES due to progressive conductive hearing loss two years after the first operation. There was only a dislocated TORP from the primary surgery and no recurrent cholesteatoma was identified during the surgery. Recidivism was not seen in any of the patients in the follow-up period.

Mean preoperative and postoperative ABG were 25.14 ± 13.93 dB and 22.22 ± 12.64 dB (Table 3). Mean preoperative and postoperative air-conduction thresholds,

bone-conduction thresholds, and ABG were not significantly different ($p=0.237$, 0.189 , and 0.417 , respectively).

Discussion

The use of surgical microscopes in ear surgery in the 1950s brought about significant developments in otology, thanks to their magnification and illumination features. Thereafter, the recent widespread use of endoscopes has led to similar developments in the field of EES. Endoscopes, which were initially used as auxiliary instruments in microscopic surgery, have replaced mainly microscopes, especially in transcanal surgeries. Although endoscopes have been used in ear surgery for more than 20 years, they have become increasingly popular with the development of those with narrower diameter, cold light sources, and high-resolution video recording systems (8, 9). The first procedure was endoscopic

Table 1. Demographic characteristics and surgical results of the patients

Gender	
Female	14 (51.86%)
Male	13 (48.14%)
Age	36.4 years (4–67)
Follow-up period	19 months (5–41 months)
Hospitalization time	2.4 days (1–7 days)
ABG (dB)	
Preoperative ABG	25.14 ± 13.93
Postoperative ABG	22.22 ± 12.64
Perioperative complications	
Chorda tympani injury	2 patients
Intraoperative bleeding	1 patient
Postoperative complications	
Retraction pocket	2 patients
Perforation	1 patient
Recurrence of cholesteatoma	No
Residual cholesteatoma	No

ABG: Air-bone gap, dB: Decibel

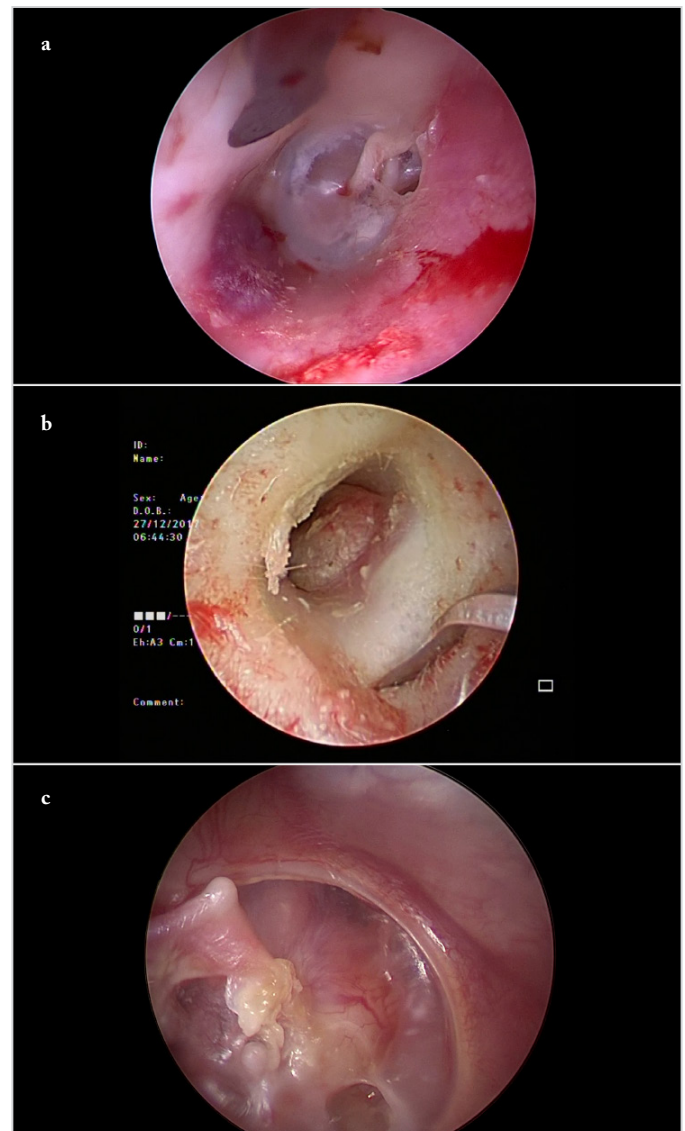


Figure 1. Different types of cholesteatoma: a) Attic cholesteatoma in the left ear, b) Congenital cholesteatoma in the left ear, c) Pars tensa cholesteatoma in the right ear

myringoplasty followed by limited attic cholesteatoma removal, ossicular chain reconstruction, and now all can be performed with TEES (10, 11).

Despite the success of the treatment techniques for cholesteatoma, there is no consensus on the optimal surgical technique that simultaneously eradicates the disease with a low recurrence rate and preserves the middle ear anatomy and physiology. Though good results can be obtained with a postauricular approach and microscopic surgery and recurrences originating from the mastoid cavity are low, recurrences occur due to the inability to sufficiently clear the cholesteatoma, especially from hard-to-reach areas such as sinus tympani and facial recess (12). CWD mastoidectomy is performed by lowering the posterior wall of the external auditory canal to reach these areas and results in handicaps such as the inability to create a self-cleaning cavity and the need for the patient to avoid contact with water (13). As an alternative, CWU mastoidectomy combined with posterior tympanotomy allows access to these areas while preserving the posterior wall, but this procedure requires postauricular incision and sometimes sacrificing the healthy bone (9, 13). On the other hand, the recurrence rate of the disease, which is approximately 5–7% in CWD, can increase to 20–25% when CWU is preferred (14, 15). Another disadvantage of the CWU techniques is the frequent need for second-

Table 2. Type of surgical technique and classification of cholesteatoma

Classification (EAONO/JOS Staging System on Middle Ear Cholesteatoma)	Number of patients n=27 (100%)
Stage 1	11 (41%)
Stage 2	11 (41%)
Stage 3	5 (18%)
Stage 4	None
Origin of cholesteatoma	
Congenital cholesteatoma	2 (7%)
Cholesteatoma secondary to a tensa perforation	3 (11%)
Pars flaccida cholesteatoma	15 (56%)
Pars tensa cholesteatoma	7 (26%)
Type of tympanoplasty	
Canal wall-up	26 (96%)
Canal wall-down	1 (4%)
Ossicular reconstruction	
No	4 (15%)
Incus interposition	13 (48%)
PORP	3 (11%)
TORP	6 (22%)
Bone cement	1 (4%)

TORP: Total ossicular reconstruction prosthesis, PORP: Partial ossicular reconstruction prosthesis, EAONO/JOS: European Academy of Otolaryngology and Neurotology/Japan Otolaryngology Society

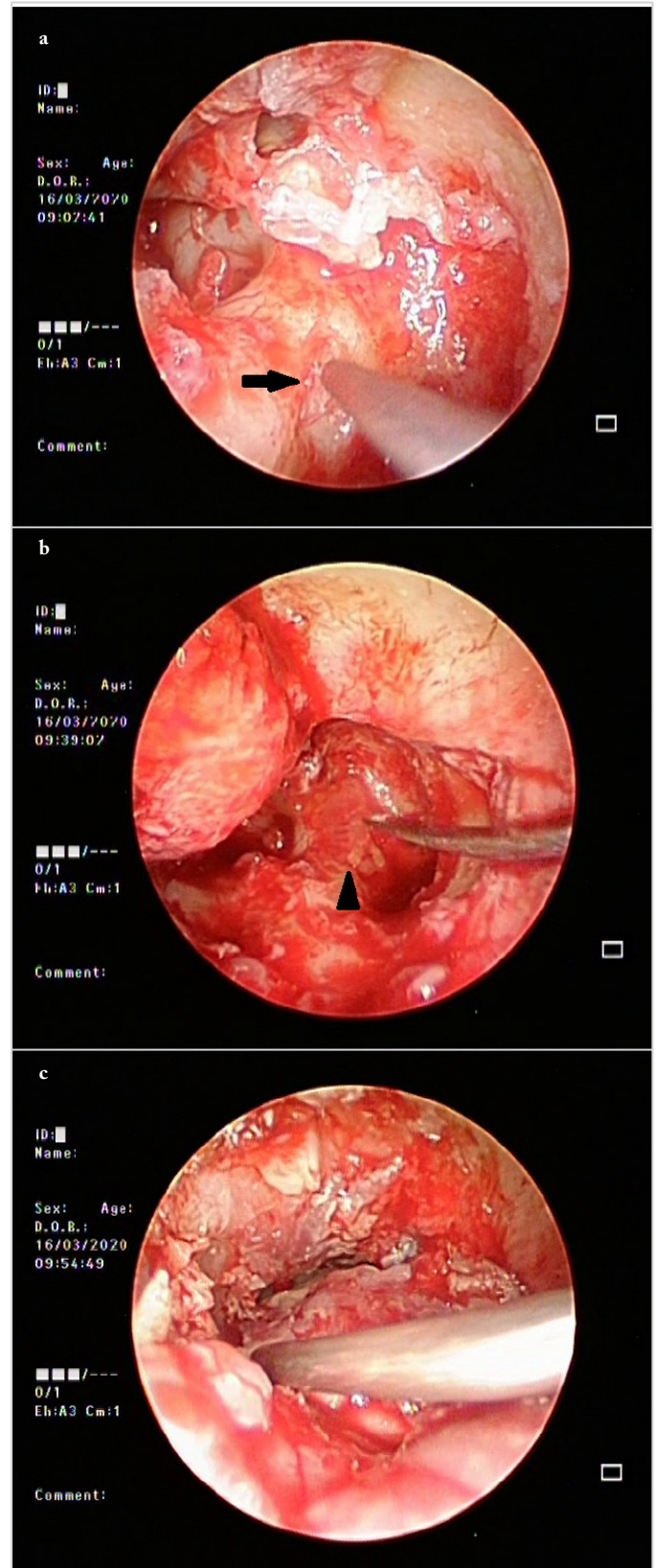


Figure 2. Lateral semicircular canal bone defect (LSCF): a) LSCF in the left ear, black arrow indicates bony defect, b) The fistula is covered by autologous fat and tragal perichondrium, black arrowhead indicates the defect covered by the tragal perichondrium, c) Final view of the atticotomy defect and tympanic membrane after the reconstruction

look surgery in terms of close follow-up of the recurrence. Another alternative is transcanal atticotomy, and it requires a certain amount of exposure, which can be difficult to provide in cases where the external auditory canal is narrow and the rate of recurrence of cholesteatoma may increase (14).

In their review published in 2020, Verma and Dabholkar (16) included 1685 patients who underwent exclusively endoscopic surgery or endoscopic surgery in combination with a microscope and found that in 267 (15.82%) patients who underwent microscopic surgery, residual cholesteatoma was detected most commonly in hidden areas such as the sinus tympani, the facial recess, and the anterior epitympanic regions when the endoscope was used for inspection after microscopic surgery. In endoscopic surgery, these areas can be seen quite well from a wider angle, even in patients with a narrow external auditory canal. When Ghadersohi et al. (17) compared EES 1, EES 2, and EES 3 in 68 pediatric patients who were operated on because of cholesteatoma, they found

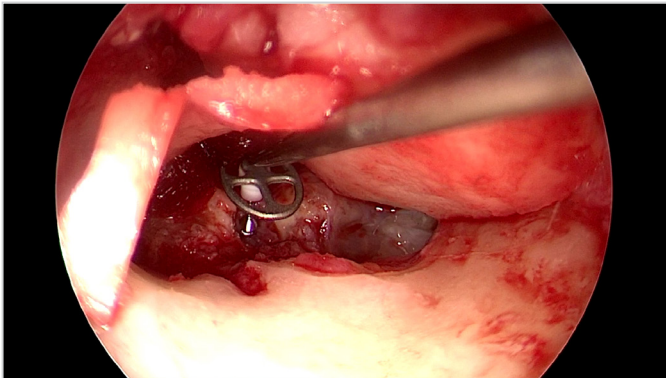


Figure 3. Ossiculoplasty with titanium prosthesis in the right ear

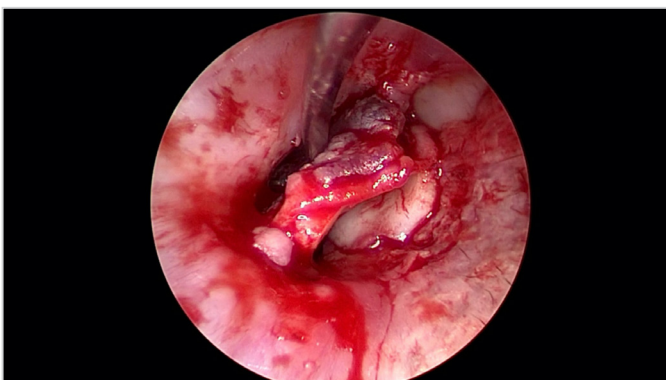


Figure 4. Repair of atticotomy defect with cartilage in the left ear

the lowest recurrence and residual rate in the TEES group (EES 3) with a recurrence rate of 4.5% and residual disease of 0%. One of the main advantages of using a microscope over endoscopes is that it allows the use of both hands. However, Dixon and James (18) compared the postauricular approach with TEES in pediatric cholesteatomas and found the rate of residual disease to be 6.3% in the TEES group and 10.9% in the postauricular approach, concluding that, microscopically, the two-handed approach did not provide any additional advantage over the endoscopic approach in clearing cholesteatomas in the middle ear and/or attic. Similarly, Li et al. (9), in their meta-analysis comparing microscopic ear surgery and EES, revealed that residual disease and recurrence were statistically significantly less in EES and that graft success, operation time, and auditory performance were not significantly different between the groups. In the presented study, no recurrence or residual disease was detected in any of the cases. However, while retraction pocket and hearing loss occurred in two patients, perforation occurred in one patient, and no residual cholesteatoma was detected in one patient who underwent revision surgery.

One disadvantage of microscopic surgery is the access to only the downstream corridor of ventilation through mastoidectomy. Thanks to the vision it provides, endoscopic surgery allows surgeons to reach the upstream parts of the ventilation system, such as the protympanum, anterior mesotympanum, eustachian tube isthmus, and tympanic isthmus (19). A postauricular incision and an excessive mastoidectomy is not needed for TEES. Therefore, TEES is a good option especially in patients with small and sclerotic mastoid cavities.

Although TEES is generally not recommended for cholesteatomas extending beyond the lateral semicircular canal, our experience shows that cholesteatoma can be reliably excised by TEES even in patients in stage 3 patients (18, 20). However, a postauricular approach and mastoidectomy may be required in cases with a large mastoid since it would be difficult to follow the cholesteatoma sac. Two patients in our study had LSCF and were operated on with TEES. In one patient, after the removal of the bone with curettes and burrs in the attic part of the external auditory canal, the LSCF was managed to be visualized by angled endoscopy and handled with curved instruments. Subsequently, the cholesteatoma was removed from the fistula and the endosteum was

Table 3. Preoperative and postoperative comparison of pure-tone audiometry results

	Preoperative (n=27) (mean ± SD)	Postoperative (n=27) (mean ± SD)	p-value
Air-conduction PTA	50.48±24.22	46.29±26.45	0.237
Bone-conduction PTA	25.34±18.53	24.09±17.23	0.189
Air-bone gap	25.14±13.93	22.22±12.64	0.417

PTA: Pure-tone audiometry, SD: Standard deviation

intact. The bony defect of 2-mm diameter on the lateral semicircular canal was covered with autologous fat and tragal perichondrium. Villari et al. (21) showed a similar approach with TEES in their study regarding LSCFs on the ampullar arm apart from the graft material. Only one of our patients was operated on with transcanal endoscopic canal-wall-down procedure. We concluded that endoscopic surgery could be performed in complicated cases, but more studies are needed to determine the utility of TEES in complicated cases.

In terms of auditory performance, we found that our patients' average ABG had decreased from 24 dB to 21 dB, which could be due to the lower pre-operative ABG of our patients compared to those reported in other studies that showed slightly higher values. Glikson et al. (14) compared EES and CWU tympano-mastoidectomy and found that air conduction pure-tone average had increased from 37.2 preoperatively to 39.6 postoperatively in the EES group. Similarly, it had increased from 41.5 to 42.2 in the microscopic group, and the difference was not significant. In contrast, Hunter et al. (22) achieved improvement in all groups in a similar study, but they found no difference between microscopic, endoscopic, and combined approach groups, although there was more improvement in the endoscopic patient group. In the present study, the incus was used most frequently in ossicular reconstruction with a rate of 48%. On the contrary, Hunter et al. (22) used PORP in three of their patients who underwent ossicular chain reconstruction in the TEES group.

We used the closed technique in all our patients that were operated on with TEES and repaired the defect in those who underwent atticotomy with tragal cartilage. No complications were reported in these patients, and the normal external auditory canal anatomy was seen intact in the postoperative follow-up period. Thus, we can say that TEES has the advantage of avoiding additional morbidity from a postauricular incision. Postoperative pain is less in patients who undergo EES compared to that of those who undergo classical microscopic surgery (23,24). This could be due to the absence of the need for a postauricular incision or the drilling of the mastoid bone. Magliulo and Iannella (24) compared EES and microscopic surgery in attic cholesteatomas in 80 patients and found that graft success, postoperative ABG, and taste sensation were not significantly different between the groups, while the mean recovery time and postoperative pain were less in the EES group. Moreover, mean recovery time was 36.3 days in the EES group and 69.9 days in the microscopic surgery group. The mean follow-up period in studies on TEES range from 4.96 to 31.2 months (9, 17, 22, 25). Our average follow-up period was 19 months, ranging from 5 to 41 months, similar to other studies.

Our study has some limitations. The first is its retrospective nature and the absence of a control group. More accurate results can be obtained in a prospective study conducted

in comparison to a control group undergoing microscopic cholesteatoma surgery. The second limitation is the low number of patients and the subsequent relatively short average follow-up period, albeit our follow-up periods are not short compared to those in the literature. Longer follow-up periods would enable more accurate results.

Conclusion

Although the superiority of TEES over the classical approaches in terms of recidivism rates has not been proven, the results obtained so far seem promising. While facilitating access to hidden areas such as sinus tympani, facial recess, and anterior epitympanic space, TEES also allows the preservation of normal middle ear physiology and stands out as a good alternative for microscopic surgery in suitable cases. Experience is essential in all surgical procedures, and as experience increases, endoscopic surgery may be performed on more complex cases. Further studies with longer follow-up periods and with large number of patients operated on with TEES are needed.

Ethics Committee Approval: This study was approved by University of Health Sciences Turkey, İzmir Bozyaka Training and Research Hospital Clinical Research Ethics Committee with decision number 2021/108 dated 23.06.2021.

Informed Consent: All patients were informed about the objective of the study and signed written consent forms.

Peer-review: Externally peer-reviewed.

Authorship Contributions

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

Conflict of Interest: The authors have no conflicts of interest to declare.

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Main Points

- Transcanal endoscopic ear surgery (TEES) stands out as a good alternative to microscopic surgery in suitable cases and facilitates access to hidden areas such as the sinus tympani and the facial recess.
- We found TEES to be a safe and highly reliable approach for treating cholesteatoma with low complication and recidivism rates.
- Endoscopic ear surgery may be performed on more complicated cases as experience increases.

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A New Technique for Use Instead of Lateral Crural Overlay for Reduction of Nasal Tip Projection in Revision Rhinoplasty

Original Investigation

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Abstract

Objective: Correction of nasal tip projection (NTP) deformities involve techniques for the lower lateral cartilage of the nose. Previously, it would have been surgically difficult to perform the lateral crural overlay (LCO) technique for the second time in revision rhinoplasty in patients who already had undergone rhinoplasty with the LCO technique because of the length of the NTP. In this study, we evaluated the lateral crural segmental excision (LCSE) technique in patients with overprojected nasal tip in revision rhinoplasty.

Methods: We retrospectively studied the cases of 19 patients who had initially undergone rhinoplasty with the LCO technique for overprojected nasal tip, and later underwent revision rhinoplasty with the LCSE technique after insufficient NTP was observed on facial analysis between 2018 and 2022.

Results: Of the patients, 12 (63%) were male, with an average age of 29.6 years, and 7 (17%) patients were female, with an average age of 25.3 years. Using Goode's formula, NTP indexes of patients measured 79.4 ± 1.8 preoperatively and 56.0 ± 1.3 postoperatively. Statically significant difference was observed between preoperative and postoperative values. None of the patients had malnutrition at the incision margins, and all patients recovered on time and without any problems. Granulation tissue was detected in the mucosa in only one patient.

Conclusion: The LCSE technique, with a short surgical time, recovery period without complications, and satisfactory nasal respiratory function, is preferred over a second LCO application in cases of NTP.

Keywords: Rhinoplasty, surgical revision, nasal tip, lower lateral cartilage

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Introduction

Evaluation and resolution of nasal tip (NT) length, projection, and rotation have an important role in rhinoplasty operations (1). Detailed facial analysis should be performed in the preoperative period. In the recent years, most rhinoplasty operations have been performed to correct NT disorders. NT projection (NTP) is the

distance from the NT to the vertical facial plane in a posterior-anterior direction, and an essential criterion in the aesthetic appearance of the face. Corrections of NTP deformities constitute an important part of rhinoplasty operations (2). However, given the increasing number of rhinoplasties in recent years, revision rhinoplasties (RRs) have also increased as a result of unsatisfactory results from

primary rhinoplasties. To achieve excellent results in NTP surgery, the surgeon should have comprehensive knowledge of the anatomical structures that make up the NT [i.e., the septal angle, the caudal septum, the medial crus (MC) and the lateral crus (LC) of the lower lateral cartilage, the nasal dome, and the nasal spine] and the surgical techniques (3-5).

In this study, we evaluated 19 patients who had previously undergone an operation with the lateral crural overlay (LCO) technique to correct an overprojected NT, but still complained of NT overprojection which was treated with the lateral crural segmental excision (LCSE) technique in RR.

Methods

This study was done retrospectively on 19 patients who underwent RR in the Otorhinolaryngology Department of the Yeni Yüzyıl University Faculty of Medicine between January 2018 and April 2022. Approval for the study was obtained from the İstanbul Yeni Yüzyıl University’s Ethics Committee for Non-Invasive Health Sciences Research (no: 2022/07-881, date: 04.07.2022). Also consent was obtained from the patients participating in the study.

All patients had previously undergone external rhinoplasty with the LCO technique to correct NT overprojection but required RR due to unsatisfactory results. Oedema that occurs after primary rhinoplasty can persist in the NT area; therefore, RR should be delayed for at least one year (6). In our patient group, this delay was 14 months on the average. Patients were followed-up for at least seven months after RR.

LCSE Surgical Technique

A columellar flap was created with the standard open rhinoplasty technique using an intercartilaginous transcolumellar reverse-V incision under general anaesthesia. Dissection was performed in the subperichondrial and subperiosteal planes up to the frontonasal junction. After revealing the bilateral LC, a 5-mm long cartilage segment from the NT to approximately 10 mm cranially was removed vertically together with the nasal vestibular mucosa (Figure 1). Cartilage ends were sutured with 5/0 round needle Trofilen (Doğsan, İstanbul, Turkey) and the nasal mucosa was sutured with 5/0 round needle egesorb Pegesorb (Doğsan, İstanbul, Turkey) (Figure 2). A new NT was created by removing the hemidomal and transdomal sutures

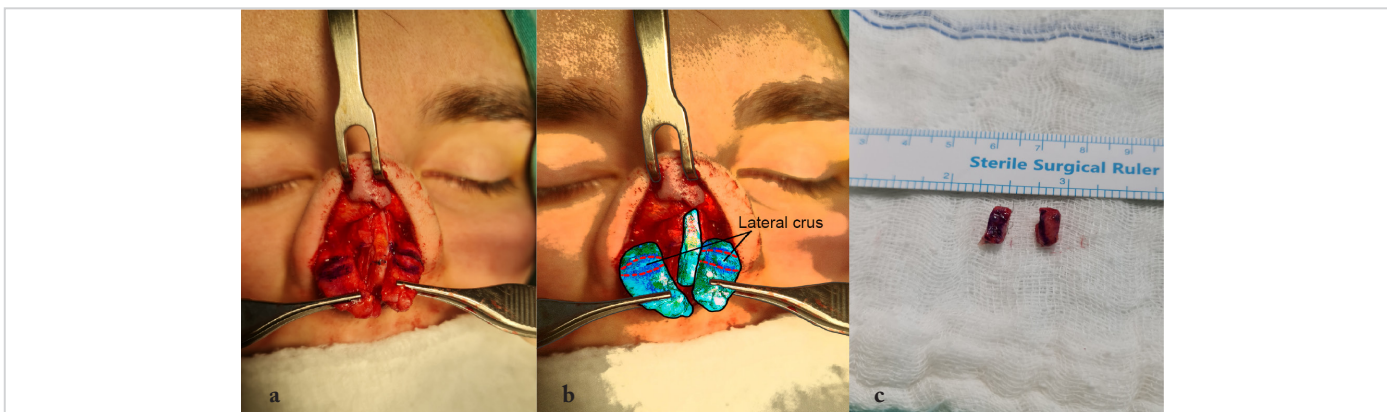


Figure 1. (a, b) Marking the segment to be resected from the LC, (c) Cartilage and mucosa segment resected from the LC
LC: Lateral crus

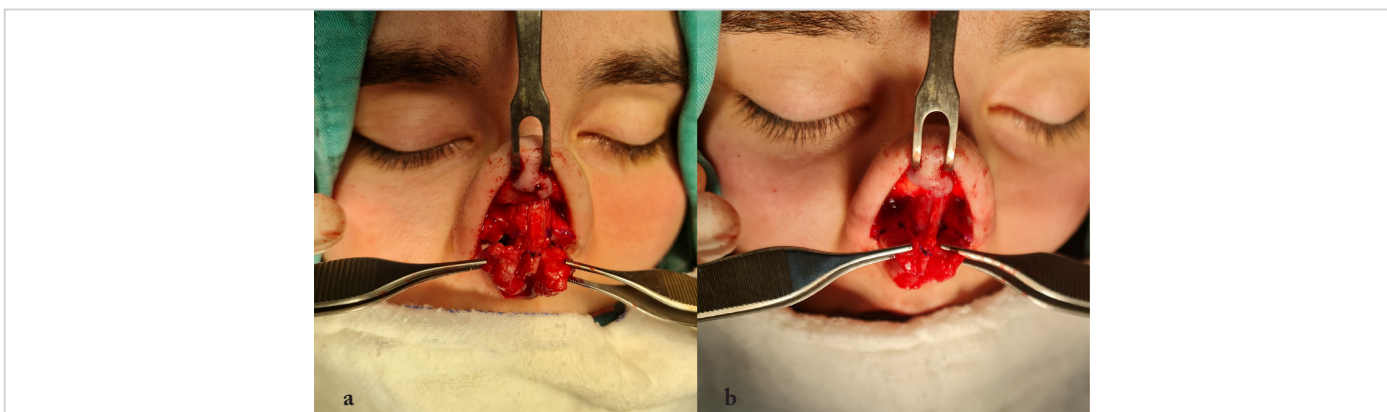


Figure 2. (a) LC with segmental resection, (b) Sutured LC
LC: Lateral crus

and sutured to the septum using the septal extension graft technique. The operation was completed by replacing the nasal flap and suturing it with 5/0 round needle pegesorb Pegesorb (Doğsan, İstanbul, Turkey) (Figure 3). The dorsum and the tip of the nose were carefully taped. A metal splint was placed on the dorsum for eight days. A mixture of epithelial and antibiotic cream (dexpanthenol/chlorhexidine hydrochloride and nitrofurazone) was applied to the nasal vestibule for 15 days.

NTP was assessed with Goode's formula by taking preoperative and postoperative facial photographs from a standard distance in the lateral planes (7). The nasal projection (TY) [distance between NT (T) and alar-facial junction (Y)] was approximately 55–60% of the nasal length [distance between nasion (N) and alar-facial junction (Y)] (Figure 4).



Figure 3. Postoperative internal LC mucosal appearance
LC: Lateral crus

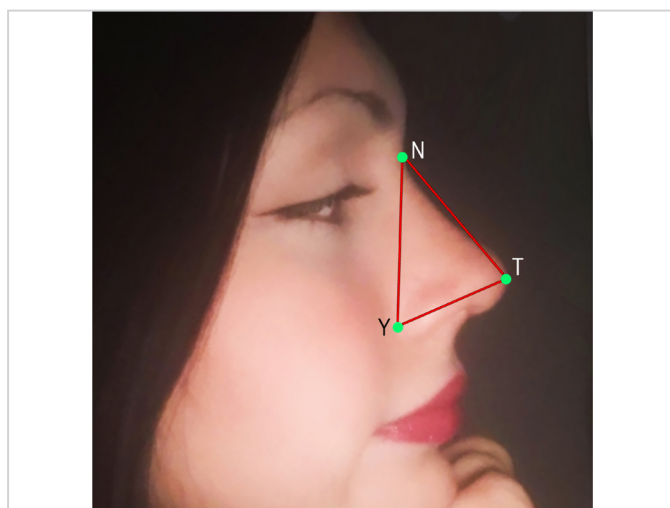


Figure 4. Facial analysis of nasal projection [Nasal tip (T), alar-facial junction (Y) and nasion (N) (YT/NTx100 ~ 55–60%)]

Statistical Analysis

Statistical analyses were done with SPSS v15.0 to compare measurements and repeated measurements. The Bonferroni method was used as a post hoc test in advanced paired comparisons. The Kolmogorov–Smirnov test was used to evaluate compliance with normal distribution. For statistical analyses, results were considered significant at $p < 0.05$.

Results

Nineteen patients with overprojected NT underwent open rhinoplasty using nasal septal caudal excision and the LCO technique; however, their results were found unsatisfactory on facial analysis done with Goode's formula. Of these patients, 12 (63%) were male, with an average age of 29.6 years, and 7 (17%) were female, with an average age of 25.3 years. LCSE was performed on all patients in RR.

A costal cartilage graft was used in five patients because of insufficient nasal septum cartilage reserve, and low NT and nasal dorsal hump were observed in four patients.

Significant difference was observed between the preoperative and postoperative nasal projection values ($p < 0.001$). Significant difference was observed between the measurements in all advanced paired comparisons ($p < 0.001$).

Preoperative and postoperative projection indexes of the patients were 79.4 ± 1.8 and 56.0 ± 1.3 , respectively. There was significant difference between preoperative and postoperative values ($p < 0.001$) (Table 1, Figure 5).

Table 1. Statistical analyses of projection index

	Pre-operative	Post-operative	p-value
Projection index	79.4 ± 1.8	56.0 ± 1.3	< 0.001



Figure 5. (a) Preoperative NTP appearance, (b) Postoperative NTP appearance

On the average, RR was performed 14 months after the primary procedure. Patients were followed-up for at least seven months after the operation and none of the patients had non-healing tissue at the incision margins, and all patients recovered promptly and without complications. Granulation was observed in the mucosa in only one patient.

Discussion

A successful rhinoplasty depends on adequate control of the overprojected NT and its supporting elements that control it. The elements that directly affect NTP are the caudal nasal septum, the LC, the MC, the nasal dome, and the nasal spine (8). In the tripod concept recently introduced by Anderson (9), the tripod structure-with one leg formed by the MCs in the middle and two legs formed by the LCs on the sides-provides nasal projection and rotation. LC and MC lengths play important roles in the overprojection of the NT. Therefore, surgical techniques for NTP reduction were designed for tripod elements (2, 10). However, there are many alternative methods for reducing NTP that are generally divided into two main groups. One of them is shortening the long lateral crura including the LCO (11), lateral crural steal (12), vertical dome division (13) and dome truncation (13) techniques. The other one is shortening the long medial crura including the septocolumellar or tongue-in-groove sutures (14), medial crural steal (15), footplate resection (2), Lipsett (2), medial crural overlay (16), vertical dome division (17), and the dome truncation (18) techniques.

The techniques that are described for NT overprojection in the literature generally are procedures for shortening the length/s of the LC and/or the MC. This technique is especially suitable for use in cases related to the LC length of the nasal over-projection.

In the LCO technique, the lateral crura are elevated from the mucosa and typically divided vertically at 10 mm lateral to the dome. Then, the medial segment is placed over the lateral segment and sutured. Although this technique is effective in shortening the length of the LC, it is difficult to achieve bilateral symmetry. As a result of the overlapping of the cartilage segments, this technique causes mucosal swelling at the nasal vestibule entrance and narrows nasal air entry (Figure 6) (11). Reduction of the nasal air passage occurs because the external nasal valve at the level of the nasal rim and the internal nasal valve at the junction of the septum upper lateral cartilage are affected. There is a risk that patients will develop nasal congestion in the postoperative period as the use of the LCO technique in the same patient group for a second time will decrease the size of the nasal vestibule, increase nasal vestibular swelling, and further decrease nasal airflow (18). However, because nasal vestibular swelling does not occur in the LCSE technique, the decrease in nasal airflow is limited.

Although the approach to NTP is difficult in primary rhinoplasty, elevation of the LC from the nasal mucosa is difficult or even impossible in RR due to fibrotic scar tissue, tissue healing complications, deterioration of anatomical structures, and synechia. Because the cartilage and the mucosa are not elevated in LCSE, fibrotic scar tissue does not represent a problem in the application of the technique (19). Moreover, the ease of the procedure allows for shorter surgery time compared to LCO.

In the LCSE technique, segmental resection was performed between the external nasal artery and the lateral nasal artery branches to preserve the nasal arterial blood supply. Therefore, deterioration of tissue vascularization was not observed in any of the patients in the postoperative period (Figures 3, 7).

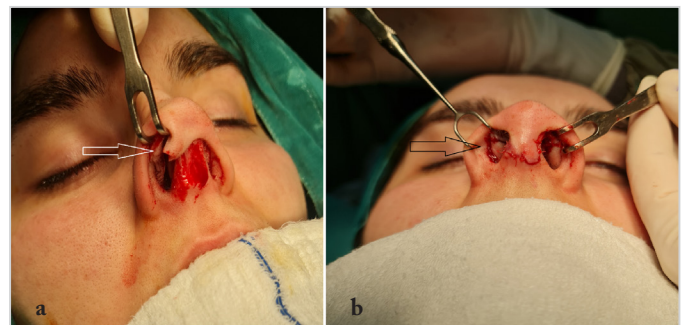


Figure 6. (a) LC cartilage mucosal swelling in preoperative LCO technique, (b) LC mucosal appearance in the postoperative LCSE technique

LC: Lateral crus, LCO: Lateral crural overlay, LCSE: Lateral crural segmental excision

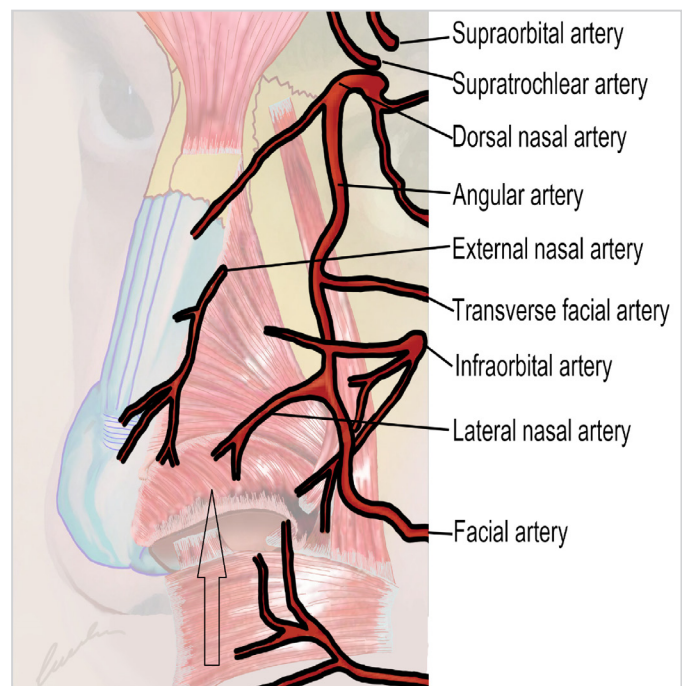


Figure 7. Nasal arterial nutrition: arrow (LC excision area)

LC: Lateral crus

Considering the possibility that our technique could weaken the resistance of the lateral crura and cause nasal alar collapse, the tension of the LCs was increased by fixing the NT to the septum using the septal extension graft technique. Our aim was to increase the stability of the tripod elements. Nasal collapse was not seen in any of the patients during the seven-month postoperative follow-up (20, 21). Granulation tissue was seen in the nasal vestibule mucosa only in one patient.

Considering that LCSE and end-to-end suturing may cause weakness and collapse in the alar region, a lateral crural strut graft can be applied. However, we did not apply an alar support graft in any of our cases. Alar collapse, asymmetry, or deformity in the nostrils was not detected in any of the patients during the postoperative follow-up. Nevertheless, the technique should be studied in larger cohorts with longer follow-up times for more rigorous evaluation.

Conclusion

Ideal NTP was achieved with the LCSE technique in patients with overprojected NT, after facial analysis showed unsatisfactory results with primary open rhinoplasty using the LCO technique. The LCSE technique offers the benefits of short surgical time, uncomplicated recovery period, and satisfactory nasal respiratory function.

Ethics Committee Approval: Approval for the study was obtained from the İstanbul Yeni Yüzyıl University's Ethics Committee for Non-Invasive Health Sciences Research (no: 2022/07-881, date: 04.07.2022).

Informed Consent: Consent was obtained from the patients participating in the study.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: A.S.R., F.Ö., Concept: A.S.R., F.Ö., Design: A.S.R., Data Collection and/or Processing: A.S.R., Analysis and/or Interpretation: A.S.R., Literature Search: A.S.R., Writing: A.S.R.

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Main Points

- Nasal tip projection (NTP) is an important aspect of facial esthetics. The creation of proportional NTP basically involves techniques for the lower lateral cartilage of the nose.
- The most important of these is the lateral crural overlay (LCO) technique. In patients who already have had an LCO procedure, however, it is difficult to apply this technique in revision rhinoplasty (RR) because of synechia and fibrosis in the surgical area.
- For this reason, the lateral crural segmental excision (LCSE) technique was developed.
- The main advantages of the LCSE technique are that it is easy to apply in RR, shortens operation time, and provides satisfactory surgical results.

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Functional and Aesthetic Outcomes of Asymmetric Dorsal Preservation for Correction of I-Shaped Crooked Nose Deformity

Original Investigation

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Abstract

Objective: The study was designed to evaluate the effectiveness of the asymmetric dorsal preservation technique for correcting I-shaped crooked nose deformity (CND).

Methods: Patients with I-shaped CND who underwent asymmetric dorsal preservation in the period from September 2020 to September 2021 were included in this retrospective study. The Rhinomanometry and Standardized Cosmesis and Health Nasal Outcomes Survey (SCHNOS) was used to assess the outcomes. Deviation angle (DA) measurements were used to evaluate the degree of crookedness. The results were recorded both preoperatively and 12 months postoperatively.

Results: Twenty-three patients were included in the study. Total nasal airflow and long-side nasal airflow were significantly higher 12 months postoperatively ($p=0.001$ each). Total nasal resistance, long-side nasal resistance, SCHNOS scores and DA measurements were significantly lower 12 months postoperatively ($p<0.001$ each).

Conclusion: Asymmetric dorsal preservation is a successful alternative technique for correcting I-shaped CND and achieving optimal aesthetic and functional outcomes.

Keywords: Nose, facial asymmetry, rhinoplasty, rhinomanometry

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Introduction

Crooked nose deformity (CND) is one of the most difficult surgical challenges in rhinoplasty (1). The deviation of the nasal bone and the cartilage roof from the midline to any side is called a CND. There are three types of CND: I-shaped, C-shaped, and S-shaped. An I-shaped CND is a linear deviation to one side. In a C-shaped CND, there is concavity on one side and convexity on the opposite side. An S-shaped CND has more than one convexity and concavity. In I-shaped

CND, the nasal bone is usually short on the deviated side and long on the opposite side. The management of CNDs is still difficult and controversial, and complications are among the most common causes of revision rhinoplasty (1, 2).

Dorsal preservation is a technique in which the nasal dorsum and middle roof are preserved. In this technique, dorsal hump reduction is achieved by removing the cartilage and bony septal strip and using either the “let down” or “push down” technique (3).

In 1914, the let-down technique, which uses wedge resection and subdorsal cartilage resection, was defined by Lothrop (4). Cottle and Loring (5) described the push-down technique in 1946. In 1975, Huizing (6) applied Lothrop's technique while pushing down the nasal pyramid with wedge bone resection.

Successful results were obtained with the dorsal preservation technique by combining these two techniques in the correction of I-shaped CNDs. The push-down technique is applied to the side of the short nasal bone where there is deviation, and the let-down technique is applied to the opposite side. Thus, the nasal pyramid is brought to the midline. This technique is called "asymmetric dorsal preservation" (7).

This study aimed to evaluate nasal patency and cosmetic results obtained by rhinomanometry, deviation angle (DA) measurements and the Standardized Cosmesis and Health Nasal Outcomes Survey (SCHNOS) in patients undergoing asymmetric dorsal preservation.

Methods

This retrospective study included 23 patients with I-shaped CND who underwent surgery between September 2020 and September 2021 in our tertiary hospital. The study was approved by the Necmettin Erbakan University Ethics Committee (decision no: 2022/4004, date: 07.10.2022). Patients with S-shaped or C-shaped CND were excluded from the study. All surgeries were carried out by the senior author (M.A.A.), and all patients were followed up at least 12 months postoperatively. Informed consent was obtained from the patients depicted in Figures 1, 2 and 3 to use their photos for medical and academic purposes.

Surgical Technique

All patients included in the study had undergone closed rhinoplasty. In all patients the nasal dorsum was dissected via the subperichondrial and subperiosteal planes. A high septal strip was removed from the subdorsal septum. A bony strip was resected with a rongeur from the perpendicular plate of the ethmoid. Radix, transverse, and lateral osteotomies were carried out with a microsaw. The maxillary bony wedge was removed with a straight lateral microsaw from the longer non-deviated side. Complete release of the nasal pyramid was followed by the push-down technique on the shorter deviated side and the let-down technique on the longer non-deviated side. Radix transition was camouflaged with drilling, and the asymmetric tip was solved with an asymmetric lateral crural steal and medial crural overlay. A columellar strut graft was used, and the Pitanguy ligament was preserved in all cases. Septoplasty was carried out with conservative resections and scoring. After the septum was separated from the maxillary spine, a burr hole was made in the maxillary spine, and the septum was fixated to the non-deviated side with 5-0

Prolene® suture. Radiofrequency cauterization was applied to the inferior concha, and outfracture was performed bilaterally in all cases.

Rhinomanometry

Rhinomanometry was performed according to the international RIGA consensus dated 2017 (8). Nasal decongestant spray was used 15 minutes before each rhinomanometry (performed one day preoperatively and 12 months postoperatively). The results were recorded and evaluated as total nasal airflow (TNA), nasal airflow for the long side (NALS), nasal airflow for the short side (NASS), total nasal resistance (TNR), nasal resistance for the long side (NRLS) and nasal resistance for the short side (NRSS) at 150 Pascal.

Deviation Angle Measurements

The Adobe Photoshop 2021 software package was used for the measurements. The reference middle line was drawn from the midpoint of the nasion to the midpoint of the upper lip. The other line was drawn from the nasion to the most prominent point of the nasal tip. These two lines were used to measure the angle of deviation. The degree of the angle between these two lines was recorded as the DA pre- and postoperatively (Figure 1).

Standardized Cosmesis and Health Nasal Outcomes Survey

The SCHNOS evaluates the functional and aesthetic results of rhinoplasty (9). The SCHNOS scale includes 10 questions and 2 parts. Each question is scored on a 6-point scale (0= no trouble to 5= extreme trouble). SCHNOS-O includes 4 questions and evaluates obstruction (items 1-4, maximum score of 20). SCHNOS-C includes 6 questions and evaluates cosmesis (items 5-10, maximum score of 30). The Turkish validated SCHNOS scale was used one day preoperatively and 12 months postoperatively (10). Pre- and postoperative SCHNOS scores were used to evaluate nasal patency and cosmesis.

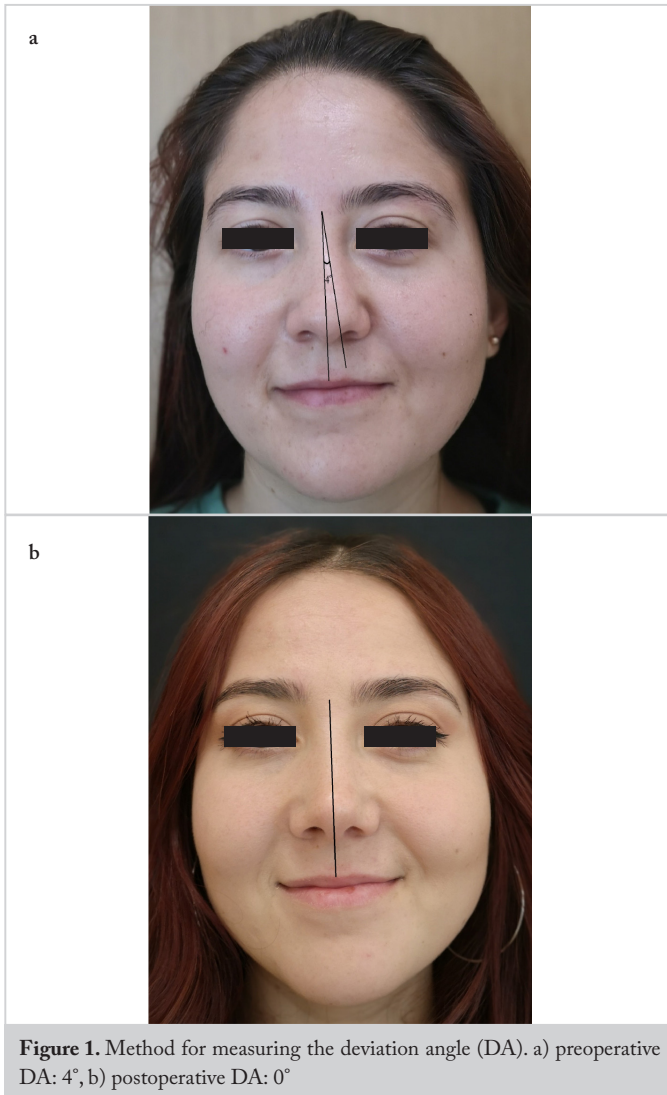
Statistical Analysis

Statistical analyses were done with SPSS 22.0 for Windows. All continuous variables were assessed for normality using the Shapiro-Wilk test. Nasal airflow, nasal resistance, DA and SCHNOS score changes were analyzed by the Wilcoxon signed-rank test. For all statistical analyses, results were considered significant at $p < 0.05$.

Results

This study was conducted with 23 patients (12 females and 11 males). Their mean age was 24.1 ± 2.1 years (range, 20-29). While all patients had I-shaped axis deviation,

nine had deviation to the right side and 14 had deviation to the left side. TNA, TNR, NASS, NALS, NRSS, NRLS, SCHNOS-O, SCHNOS-C and DA results preoperatively and 12 months postoperatively are shown in Table 1.



Photos of two patients preoperatively and 12 months postoperatively are shown in Figures 2 and 3.

None of the patients had severe complications perioperatively and/or postoperatively. Most of the patients were satisfied with the functional and cosmetic outcomes 12 months postoperatively.

Discussion

Various techniques have been described for the correction of CND (11–15); however, most of them are associated with high revision rates. Many surgeons have emphasized the importance of septal surgery in the management of CND (16, 17). A classification of septonasal deviations was reported by Guyuron et al. (18), who in their study, evaluated 93 septoplasty patients of whom 40% had I-shaped, 32% had C-shaped and 9% had S-shaped septal deviation. In the study of Stepnick and Guyuron (1), the authors suggested separation of the posterocaudal septum from the vomer, partial separation of the quadrangular cartilage from the perpendicular plate and midline repositioning of the caudal septum on the maxillary crest for the correction of I-shaped deformities. Additionally, correction of the septum and nasal pyramid with asymmetric osteotomies and complete separation of the osteocartilaginous structures, rather than correction with asymmetric spreader grafts, are performed (1, 2).

Demir (2) emphasized a new method using a unilateral spreader graft for the correction of CND. They used this approach for the long side, performed asymmetric osteotomy, separated the caudal septum from the nasal spine and fixed the neutral position. Only aesthetic outcomes were evaluated in their study using DA.

Kavuzlu and Şahin (19) used a cross-spreader graft for I-shaped CND in their retrospective study of 25 patients. They used the Nasal Obstruction Symptoms Evaluation

Table 1. Preoperative and postoperative rhinomanometry results, DA measurements and SCHNOS scores

	Preoperative		Postoperative 12th month		p-value*
	Mean	SD	Mean	SD	
TNA	642.5	83.6	765.0	57.2	0.001
NA(SS)	371.4	22.9	391.4	48.0	0.167
NA (LS)	271.0	85.3	373.1	44.6	0.001
TNR	0.23	0.03	0.19	0.01	<0.001
NR (SS)	0.39	0.02	0.37	0.03	0.017
NR (LS)	0.61	0.19	0.42	0.05	<0.001
SCHNOS-O	16.7	3.6	1.5	1.8	<0.001
SCHNOS-C	27.2	4.1	2.1	1.8	<0.001
DA	7.4	1.5	0.8	1.2	<0.001

*P-value for Wilcoxon signed-ranks test. TNA: Total nasal airflow, NA: Nasal airflow, SS: Short side, LS: Long side, TNR: Total nasal resistance, SCHNOS: Standardized Cosmesis and Health Nasal Outcomes Survey, O: Obstruction, C: Cosmesis, DA: Deviation angle, SD: Standard deviation



Figure 2. Twenty-one-year-old female who has severe I-shaped axis deviation and underwent closed approach preservation rhinoplasty. a, b) frontal and profile views (preoperatively), c, d) frontal and profile views (12th month postoperatively)

and Rhinoplasty Outcomes Evaluation scales and DA measurements for outcome evaluation, determining that the cross-spreader graft is an effective method for correcting I-shaped CND and promises good functional and aesthetic results.

In the recent years, the asymmetric dorsal preservation technique has come to the fore as a good alternative for I-shaped CND correction (7). Asymmetry in the nasal pyramid is corrected using the let-down technique with bony wedge resection on the long side and the push-down technique on the short side. Given that the main problem in these patients is usually nasal bone asymmetry, the nasal roof is not opened, which eliminates the need for a spreader graft. We think it is a conservative approach and a useful method for I-shaped CND.

Preservation of the natural nasal dorsum avoids complications associated with midvault reconstructions. In structural rhinoplasty, especially with the use of spreader grafts, some

patients experience widening of the roof. Preservation rhinoplasty avoids this widening, as it does not disrupt the natural roof (3).

One of the subtleties of preservation rhinoplasty is choosing the right patient. It is important that S-shaped and C-shaped CND cases be excluded, as done in our study. In severe septal deviations, low septal strip removal using the Cottle technique may be preferred instead of high septal strip removal. One of the common problems in preservation rhinoplasty is a residual hump—a major cause of patient dissatisfaction. This is less common in cases where the high septal strip is removed (3, 7).

Most studies have used DA measurements for CNDs (2, 7). We, too, used DA measurements but left the evaluation of the aesthetic results to the patients using the SCHNOS-C scale. Since rhinoplasty focuses on patient satisfaction, this is also subjective. Some studies have shown that the dorsal preservation technique provides successful aesthetic

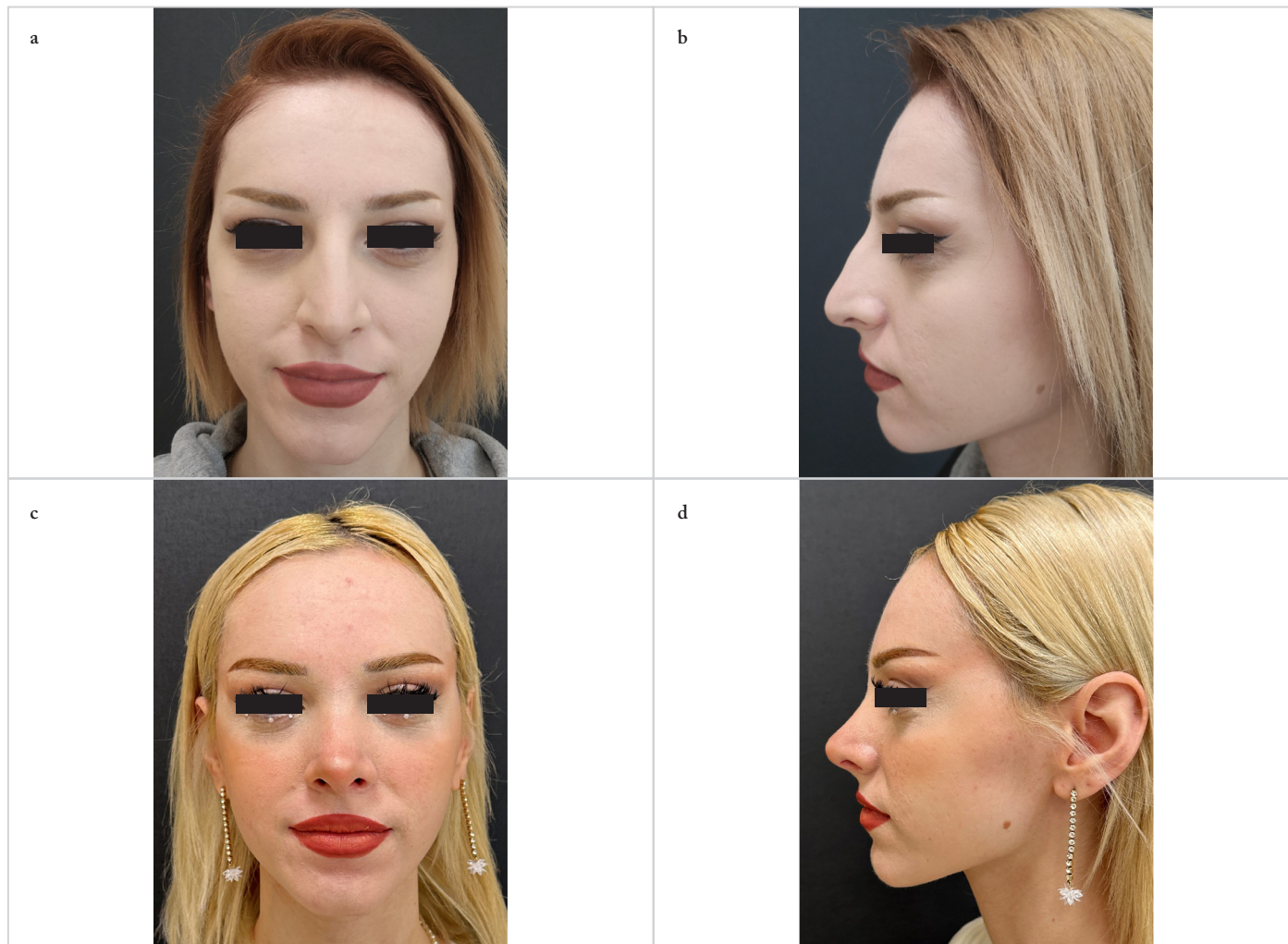


Figure 3. Twenty-four-year-old female who has mild axis deviation and underwent closed approach preservation rhinoplasty. a, b) frontal and profile views (preoperatively), c, d) frontal and profile views (12th month postoperatively)

results in I-shaped CNDs, but we observed that functional outcome assessment is incomplete in many studies on CND; and further noted that objective evaluations, such as rhinomanometry, are not often used in similar studies on CNDs (7).

Our study is unique in that rhinomanometry shows that asymmetric dorsal preservation provides good functional outcomes in I-shaped CND. At 12 months postoperatively, there was a significant increase in TNA, a decrease in TNR, an increase in NALS and a decrease in NRLS. The significant decrease in SCHNOS-O scores 12 months postoperatively shows that nasal patency increased to a degree that the patients could notice. In terms of aesthetic results, there was a significant decrease in SCHNOS-C scores 12 months postoperatively. This study is also unique as the first to evaluate the functional effects of the asymmetric dorsal preservation technique.

The limitations of our study are that it is retrospective and does not have a control group. More valuable results could

be obtained by designing a prospective study with a control group in which CND was corrected with the structural rhinoplasty technique.

Conclusion

Although few studies have evaluated the success of asymmetric dorsal preservation in I-shaped CND, especially regarding aesthetic results, this study, using objective and subjective evaluations, has shown that the asymmetric dorsal preservation technique is a good alternative for the correction of I-shaped CND, with successful aesthetic and functional outcomes.

Ethics Committee Approval: This study was approved by the Necmettin Erbakan University Ethics Committee (decision no: 2022/4004, date: 07.10.2022).

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Authorship Contributions

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

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Main Points

- Crooked nose deformity (CND) is one of the most difficult surgical challenges in rhinoplasty.
- The management of crooked nose is still difficult and controversial, and complications are among the most common causes of revision rhinoplasty.
- Successful results were obtained with the dorsal preservation technique in the correction of I-shaped CND.
- Asymmetric dorsal preservation technique is a good alternative for the correction of I-shaped CND, with successful aesthetic and functional outcomes.








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Perineural Invasion of Superior and Inferior Laryngeal Nerves in Advanced Stage Squamous Cell Carcinoma of the Larynx: A Case Series and Review

Original Investigation

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Abstract

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Objective: To evaluate bilateral superior and recurrent laryngeal nerves for tumor spread in patients of advanced-stage laryngeal carcinoma undergoing surgical resection.

Methods: A prospective study was conducted including biopsy-proven cases of laryngeal squamous cell carcinoma (SCC) that were planned for total laryngectomy. Patients with metachronous or synchronous SCC were excluded from the study. All patients underwent total laryngectomy, where both superior and recurrent laryngeal nerves were harvested along with the specimen, and the proximal ends of the nerves were marked for reference. Perineural invasion (PNI) was assessed in nerves within the tumor and in bilateral extra-laryngeal nerves.

Results: The study included 22 patients with a mean age of 58 years. Intra-tumoral PNI was found in 7 of the 22 cases (32%). The free nerve margins of superior and recurrent laryngeal nerves, which were examined from proximal to distal orientation, showed no tumor infiltration in any of the cases.

Conclusion: Perineural invasion of minor nerves constitutes a major pathway of spread. On the contrary, invasion of superior or recurrent laryngeal nerves does not constitute a route for tumor spread. Hence, there is no need to extend the surgical boundary for total laryngectomy to include these major nerves separately.

Keywords: Larynx cancer, squamous cell carcinoma, laryngectomy, recurrent laryngeal nerve, perineural spread

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Introduction

In India, laryngeal cancers account for 3–6% of all cancers in males. The age-adjusted incidence rates of laryngeal cancer vary from 1.26 to 8.18 per 100,000 populations (1). The inconspicuous symptomatology of laryngeal cancers is responsible for late presentation and diagnosis at advanced stages. The treatment

for laryngeal cancers has advanced over the last few decades, still, the 5-year survival rates are dismal. Loco-regional recurrence (LRR) rates vary between 30–50% for these advanced laryngeal cancers (2). Various risk factors have been cited for high LRR failure rates, including age, smoking index, initial T stage, tumor grade, nodal metastasis, surgical suitability, and residual disease post treatment (2).

The invasion of the perineural space by tumor cells is an important risk factor for LRR (3). The involvement of the nerve occurs at the terminal endings and may progress to involve the major trunks proximally. Tumor spread along the perineural space has been reported to occur for long distances, which may preclude obtaining a clear surgical margin (4). Perineural invasion (PNI) is an important component of the Brandwein–Gensler et al. (5) pathological risk assessment model for LRR and survival in patients of squamous cell carcinoma (SCC) of the head and neck. Involvement of the major nerves (>1 mm) has been given a score of three and directly places the patient in the high-risk category for LRR, hence, the recommendation of adjuvant therapy in such cases. The larynx is a site where nerve supply is through paired superior and recurrent laryngeal nerves, which are easily identified during surgical resection (6). The tumor may spread along these major nerves distant from the tumor micro-environment constituting perineural tumor spread. We hypothesized that recurrences may, in part, be attributed to the residual tumor along these major nerves.

We conducted a prospective study in a tertiary care hospital in India to find out the incidence of PNI in patients of advanced-stage laryngeal carcinoma undergoing surgical resection. We also aimed to evaluate bilateral superior and recurrent laryngeal nerves for perineural tumor spread in these patients.

Methods

A prospective study was conducted in the Department of Otorhinolaryngology and Head-Neck Surgery of All India Institute of Medical Sciences, New Delhi, India. The study received ethical clearance from the Institute Ethics Committee of All India Institute of Medical Sciences, New Delhi, India (Ref. No. IEC-287/02.06.2017, RP-58/2017, date: 04.07.2017). The study included biopsy-proven cases of laryngeal SCC that were planned for total laryngectomy. Informed written consent was obtained from the participants. The clinical and demographical details of all the patients were collected in the proforma. All patients underwent physical examination and radiological evaluation in the form of contrast-enhanced computed tomography of the cervical and thoracic regions with or without contrast-enhanced magnetic resonance imaging of the cervical region followed by biopsy from the lesion. All patients with biopsy-proven laryngeal SCC were referred to a multidisciplinary head and neck tumor team for treatment planning. Disease staging was done according to the staging system proposed by the American Joint Committee on Cancer, eighth edition. The patients who underwent total laryngectomy during the study period were included. The patients with metachronous or synchronous SCC were excluded from the study. All patients underwent total laryngectomy using a standardized protocol, where both superior and

recurrent laryngeal nerves were harvested along with the specimen, and the proximal ends of the nerves were marked for reference (Figure 1). The specimens were sent for histopathological examination. The tissues were fixed in 10% neutral formalin and then embedded in paraffin. The formalin-fixed paraffin-embedded specimen was sectioned using microtome to obtain 4–5 µm thick paraffin sections. These were stained with hematoxylin and eosin. Perineural invasion was defined as more than 33% of circumferential involvement of the nerve by malignant cells or the presence of tumor cells in the three layers of nerve sheaths. Similarly, the sections of both superior and inferior laryngeal nerves were also examined to investigate perineural tumor spread.

Statistical Analysis

Descriptive statistics were used to describe the data. Statistical analysis was done using Stata/MP 16 (StataCorp LLC, Texas, USA). A chi-square test was used to compare PNI between primary and residual/recurrent cases of advanced-stage laryngeal SCC and survival between PNI-positive and PNI-negative cases of laryngeal SCC.

Results

The study included 22 patients with advanced-stage laryngeal carcinoma. The clinical and demographical details are summarized in Table 1. The most common presentation was voice change (100%) followed by dysphagia (46%).

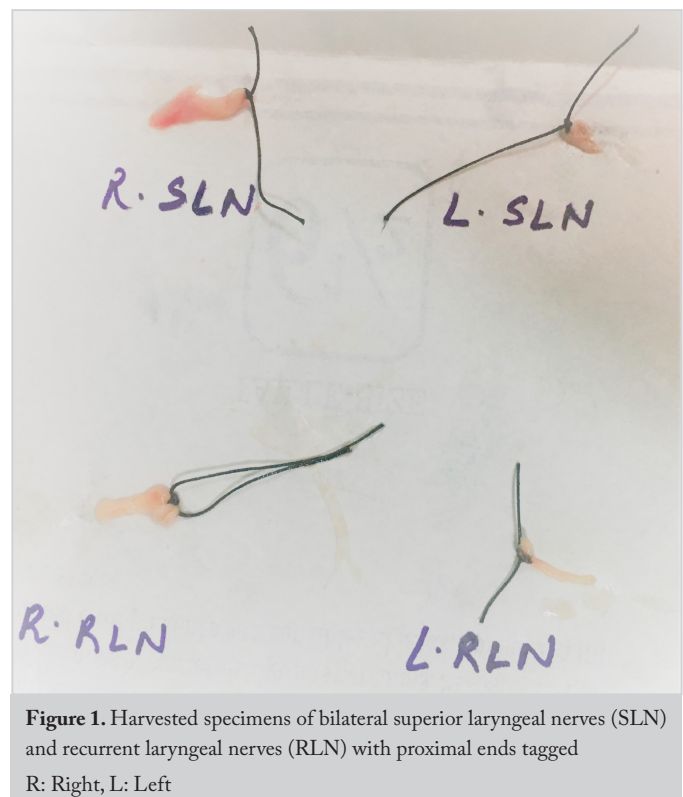


Figure 1. Harvested specimens of bilateral superior laryngeal nerves (SLN) and recurrent laryngeal nerves (RLN) with proximal ends tagged
R: Right, L: Left

Table 1. Clinical and demographical details of the cases

	Number (percentage)
Total patients	22
Age in years (mean, range)	58, 33 to 84
Sex	
Male	20 (90.9%)
Female	2 (9.1%)
Stage distribution	
Stage 3	4 (18.2%)
Stage 4	18 (81.8%)
Case distribution	
Primary cases	14 (63.6%)
Residual/recurrent cases	8 (36.4%)
Histopathological surgical margins	
Free of tumor	13 (59.1%)
Close	8 (36.4%)
Involved	1 (4.5%)
Post-operative adjuvant treatment	
Radiotherapy	18 (81.8%)
Chemo-radiotherapy	1 (4.5%)
None	3 (13.7%)
Recurrence	
Loco-regional	3 (13.7%)
Distant	1 (4.5%)

Ten patients had undergone preoperative tracheostomy as an emergency airway procedure. Preoperative biopsy showed that 19 patients had moderately differentiated SCC, while three patients had poorly differentiated SCC. TNM staging, subsite distribution, and PNI data are detailed in Table 2.

Intra-tumoral PNI was found in seven of the 22 cases (32%). Of these, five had primary laryngeal malignancy and two had residual/recurrent disease post radiotherapy/chemo-radiotherapy. The difference in PNI between primary and residual/recurrent cases was not found to be statistically significant ($p=0.49$). The free nerve margins of superior and recurrent laryngeal nerves, which were examined from proximal to distal orientation, showed no tumor infiltration in any of the cases.

The patients were followed-up for a median duration of 54 months (range, 2 to 74 months). Five patients were lost to follow-up (one in the PNI-positive group and four in the PNI-negative group). Overall survival at five years was 58.8% (10 out of 17). In the PNI-positive group, 2/6 (33.3%) patients were alive and disease-free, while in the PNI-negative group, 8/11 (72.7%) patients were alive and disease-free at five years. The difference was not found to be statistically significant ($p=0.14$).

Table 2. Details of the patients regarding TNM staging, tumor subsites, prior treatment and perineural invasion within the larynx and nerves

Case no.	Age/sex [#]	Stage	Subsite	Prior treatment	PNI larynx	PNI nerves [^]
1	61/M	T3N3bM0	Supraglottis	No	Yes	No
2	60/M	T3N0M0	Transglottis	No	No	No
3	45/F	T4aN0M0	Supraglottis	No	Yes	No
4	50/M	T4aN1M0	Transglottis	No	No	No
5	59/M	rT4aN0M0	Supraglottis	CT/RT	Yes	No
6	59/M	rT4aN0M0	Transglottis	CT/RT	No	No
7	65/M	rT3N0M0	Glottis	RT	No	No
8	84/M	T4aN0M0	Transglottis	No	No	No
9	57/M	rT4aN0M0	Transglottis	CT/RT	No	No
10	80/M	rT3N0M0	Glottis	RT	No	No
11	60/M	T4aN0M0	Transglottis	No	No	No
12	66/M	T4aN1M0	Supraglottis	No	Yes	No
13	42/M	T4aN0M0	Supraglottis	No	No	No
14	53/M	rT3N0M0	Glottis	RT	Yes	No
15	57/M	rT4aN0M0	Transglottis	CT/RT	No	No
16	59/F	T4aN3bM0	Supraglottis	No	Yes	No
17	33/M	T4aN0M0	Transglottis	No	No	No
18	59/M	T4aN2bM0	Supraglottis	No	No	No
19	58/M	rT4aN0M0	Transglottis	RT	No	No
20	48/M	T4aN0M0	Transglottis	No	Yes	No
21	62/M	T4aN0M0	Supraglottis	No	No	No
22	59/M	T4aN0M0	Transglottis	No	No	No

CT: Chemotherapy, F: Female, M: Male, PNI: Perineural invasion, RT: Radiotherapy

Discussion

Perineural invasion is difficult to assess preoperatively by clinical examination. Even though radiological evaluation may give some clue about the PNI of major nerves in the form of changes in signal intensity, contrast enhancement, or widening of the foramen, such is observed only where involved nerves are quite large (4). Mesolella et al. (7) have found the incidence of PNI at 15.8% in their retrospective study of 40 patients with advanced-stage laryngeal SCC. Zhu et al. (8), in their retrospective study, have evaluated 1,272 laryngeal SCC patients undergoing surgical treatment. The authors found that 118 patients (9.28%) had PNI. In their matched-pair analysis, they have concluded that PNI-positive laryngeal SCC had significantly worse overall survival compared to their PNI-negative group. Our data also shows a similar trend with 72.7% survival in PNI-positive group as compared to 33.3% survival in the PNI-negative group. However, the difference was not found to be statistically significant which may be because of the small sample size.

Ten out of the fifteen laryngeal SCC patients (66.6%) evaluated by Vural et al. (4) were found to have PNI in the histopathological evaluation. The study by Chirilă et al. (6) included 246 patients of laryngeal and hypopharyngeal SCC. The authors found that 35.59% of the patients had evidence of PNI on histological evaluation. Similar to the study by Chirilă et al. (6), we found the incidence to be 32% in our study. The wide variation in the reported values of PNI suggests the geographical differences in the population included in the studies.

The evaluation of PNI in superior and recurrent laryngeal nerves has been done in two previous studies, albeit, in a different geographical setup. Vural et al. (4), in their evaluation of 15 patients, did not find PNI of any of these nerves. Similarly, the study by Chirilă et al. (6) was unable to find evidence of PNI of these nerves in any of the cases of laryngeal SCC. Our findings were consistent with those of the previous studies suggesting that perineural tumor spread through superior and recurrent laryngeal nerves did not constitute a pathway of spread in laryngeal SCC, even in locally advanced stages. Hence, there is no need to extend the surgical resection boundaries of total laryngectomy to include the bilateral superior and recurrent laryngeal nerves.

Conclusion

Perineural invasion of minor nerves varies widely in laryngeal SCC but constitutes a major pathway of spread. On the contrary, invasion of superior or recurrent laryngeal nerves does not constitute a route for tumor spread. Hence, there is no need to extend the surgical boundary for total laryngectomy to include these major nerves separately.

Ethics Committee Approval: The study received ethical clearance from the Institute Ethics Committee of All India Institute of Medical Sciences, New Delhi, India (Ref. No. IEC-287/02.06.2017, RP-58/2017, date: 04.07.2017).

Informed Consent: Informed written consent was obtained from the participants.

Peer-review: Externally peer-reviewed.

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Surgical and Medical Practices: K.S., A.T., H.V., Concept: S.B., K.S., A.T., H.V., Design: S.B., Sh.B., K.S., A.T., H.V., Data Collection and/or Processing: A.S.J., S.B., Sh.B., S.K., Analysis and/or Interpretation: A.S.J., S.K., Literature Search: A.S.J., S.K., A.T., H.V., Writing: A.S.J., S.B., Sh.B., S.K., K.S., A.T., H.V.

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Main Points

- Risk of recurrence in advanced-stage laryngeal carcinoma varies between 30–50%.
- Perineural invasion is an important risk factor for loco-regional recurrence.
- Perineural invasion of minor nerves in the tumor micro-environment contributes a major pathway of spread for laryngeal cancers.
- However, invasion of superior or inferior laryngeal nerves does not form a pathway of spread for laryngeal squamous cell carcinoma.
- Hence, there is no need to extend the surgical boundary for total laryngectomy to include these major nerves separately.

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Evaluation of Factors Affecting Hearing Aid Use in Children

Original Investigation

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Abstract

Objective: The purpose of this study was to determine the factors affecting hearing aid use in children and investigate how these factors affected their subjective auditory performance.

Methods: The study was carried out with 34 children aged three to six years who had mild or moderate sensorineural hearing loss and had used a bilateral hearing aid for at least six months. The daily hearing aid usage times of children were collected with the help of data logging software. Parent-child interactions were assessed with the Maternal Behavior Rating Scale (MBRS) and Child Behavior Rating Scale (CBRS) scores. To assess the parents' levels of knowledge about hearing devices, the Hearing Aid Awareness Question Form for Parents, which was prepared by the researchers, was used. Finally, to evaluate the subjective hearing performance of the children, the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) survey was implemented.

Results: Strong and significant correlations were found between the MBRS and CBRS scores, the results obtained from some questions in the Hearing Aid Awareness Question Form for Parents, the overall PEACH score, the QUIET subscale score, and the NOISE subscale score ($p < 0.001$). According to the multivariate linear regression analyses, it was observed that the use of the pediatric clip hearing aids holder (question 17) had a significant effect in decreasing both the overall PEACH score ($\beta = -3.07$, $p = 0.008$) and the PEACH-NOISE subscale score ($\beta = -1.88$, $p = 0.012$). A unit increase in the score given to question 24 of the Hearing Aid Awareness Question Form for Parents (i.e., using the hearing aids longer) caused a 2.35-fold increase in the PEACH-NOISE subscale score, a 1.74-fold increase in the PEACH-QUIET subscale score, and a 4.06-fold increase in the overall PEACH score.

Conclusion: Parent-child interaction and parents' knowledge about hearing aid use are important factors affecting hearing aid use in children. These factors also affect the children's subjective auditory performance. Parents should be given detailed information about hearing aid use and be more sensitive and responsive in their interactions with their children.

Keywords: Hearing loss, hearing aids, parent child relations, pediatrics, hearing impairment, audiology

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Introduction

Hearing loss is a common condition in children and can lead to delays in language skills, as well as to learning problems and social integration disorders (1, 2). These adverse effects can be reduced with appropriate audiological intervention and auditory rehabilitation (2, 3). Hearing aids are one of the audiological intervention options (4).

In most studies that investigate the factors affecting the use of hearing aids, it was emphasized that daily hearing aid use and the parents' educational levels and socioeconomic statuses were effective at daily hearing aid use time (5, 6). In addition to these factors, parents' knowledge about hearing aid use and parent-child interactions were also thought to affect hearing aid use.

As is often mentioned in the literature, one of the factors affecting hearing performance in children is the duration of hearing aid use (7-10). Improved auditory performance and language development are observed in full-time users (8, 11).

Another important factor in hearing aid usage is the interaction between the parents and the child. As this interaction increases, the development in the child's cognitive, social, language and communication areas also increase (12, 13).

Another important factor affecting the use of hearing aids in children is the correct and effective use of these devices. Parents who encountered hearing aids for the first time reported that they had some difficulties in how to integrate new skills into daily life, had little knowledge and had deficiencies (14-16). However, no informative material have been found for parents to overcome these inadequacies.

Factors affecting hearing aid use also affect auditory performance in children (11, 14). There is limited information in the existing studies on how parent-child behaviors and parents' knowledge about hearing aid usage affect the subjective auditory performance of a child (17). Moreover, no study has been found on how parent-child interaction, parents' knowledge about hearing aid use and daily use of hearing aids affect subjective auditory performance. This study aimed to evaluate the factors affecting hearing aid usage of children in regard of the literature. The study further aimed to evaluate the effects of correct hearing aid use, the parent's levels of knowledge about the hearing aid, parent-child interaction and daily hearing aid use have on the subjective auditory performance of children diagnosed with hearing loss.

Methods

The study was approved by Hacettepe University Non-Interventional Clinical Research Ethics Committee on October 9, 2018 (decision no: GO 18/841-29). The research

was carried out on a voluntary basis. As this study was conducted for research purposes, participants and researchers were asked to sign two written informed consent forms, i.e., the audiologist's explanation and the parent's permission statement for the child.

Subjects

The study was carried out with 34 children (20 females, 14 males) aged three to six years (4.31 ± 1.06 years) who had mild or moderate sensorineural hearing loss, had used bilateral hearing aids for at least six months, and showed normal development. Individuals with hearing aids whose free-field hearing thresholds were in the speech banana were recruited. Individuals with unilateral or bilateral severe/profound hearing loss, inner ear anomaly, diagnosed with retrocochlear pathology, developmental delay or auditory neuropathy spectrum disorder were excluded from the study. One participant was excluded from the study because of a Down Syndrome diagnosis and another due to diagnosed developmental delay. Participants were randomly selected, regardless of their social level, and consisted of early diagnosed individuals who regularly received special education. The brands and models of hearing aids and the gender of the subjects were not taken into account.

The demographic characteristics of the participants are given in Table 1.

Procedure

The Denver II Developmental Screening Test was applied to rule out the gross motor, fine motor, and personal/social developmental retardation of the individuals participating in the study. Individuals who were found to have developmental retardation in any area other than language development were excluded from the study. The hearing aid gains were evaluated in the free field and the following steps were applied in the given order to those whose hearing aid thresholds were within the speech banana.

Determination of the Daily Use of Hearing Aids

Data logging is the process of collecting data about the participants' use of the hearing aid with the help of a computer software (Phonak Target version 4.2, Switzerland; Oticon Genie Fitting Software, Denmark) that records the environmental conditions (quiet/noisy) and time used (hours/day) (18).

Evaluation of Parent-Child Interaction

The Maternal Behavior Rating Scale (MBRS) consists of 12 items and three sub-factors aimed to evaluate the behavior of parents during their communication and interaction with their children. The scale was developed by Mahoney et al. (19) to evaluate parent-child interaction and adapted to Turkish by Diken et al. (20) as the Turkish version of the

MBRS, MBRS-TV. The scale consists of three subscales, namely, 'Being Sensitive and Responsive', 'Being Emotionally Expressive', and 'Being Success-oriented and Directive'. Scoring is done on a range of one to five using the five-point Likert-type scoring. For the nine items under the headings Being Sensitive-Responsive and Being Emotionally Exvepressive, a score of '1' indicates no interaction, while '5' indicates ideal interaction. In the three items under the heading Success-Oriented-Directive, 3 was accepted as ideal, and '4' and '5' as extremes.

The Child Behavior Rating Scale (CBRS), developed by Mahoney and Whedeen (21), was adapted to Turkish as

the Turkish version of the CBRS (CBRS-TV) (20). Here, child behavior is assessed with seven items and two factors as 'Attention' and 'Initiation'. The heading 'Attention', addresses the child's attention, continuity, participation, and cooperation. The heading 'Initiation' addresses the child's initiation, joint attention, and emotional state items. In scoring, '1' for each item indicates no behavior and '5' indicates ideal behavior. MBRS-TV and CBRS-TV scales were implemented by the lead author to assess parent-child interaction within the Practical Use Certificate. The sessions were recorded on video for 10–15 minutes with the consents of the parents and the children, in a way that would not

Table 1. Demographics of subjects

Subject	Gender	Age (months)	Etiology	Degree of hearing loss	Mother's education level
S1	M	52	Congenital	Moderate	Bachelor's or master's degree
S2	F	47	Congenital	Moderate	High school
S3	F	50	Congenital	Moderate	High school
S4	F	69	Congenital	Moderate	High school
S5	F	44	Congenital	Moderate	Bachelor's or master's degree
S6	F	42	Congenital	Moderate	Bachelor's or master's degree
S7	F	70	Congenital	Moderate	High school
S8	M	58	Congenital	Moderate	Bachelor's or master's degree
S9	M	67	Congenital	Moderate	Primary education
S10	M	36	Congenital	Moderate	High school
S11	F	70	Congenital	Moderate	High school
S12	F	48	Congenital	Moderate	Primary education
S13	F	38	Congenital	Moderate	High school
S14	M	42	Congenital	Moderate	Bachelor's or master's degree
S15	F	63	Congenital	Mild	High school
S16	M	41	Congenital	Moderate	Primary education
S17	F	70	Congenital	Moderate	Primary education
S18	M	53	Congenital	Moderate	Bachelor's or master's degree
S19	M	41	Congenital	Moderate	Primary education
S20	F	66	Congenital	Moderate	Primary education
S21	F	65	Congenital	Mild	High school
S22	F	60	Congenital	Mild	High school
S23	M	39	Congenital	Moderate	Primary education
S24	F	60	Congenital	Moderate	Bachelor's or master's degree
S25	M	46	Congenital	Moderate	High school
S26	F	66	Congenital	Moderate	Bachelor's or master's degree
S27	F	56	Congenital	Moderate	Primary education
S28	F	63	Congenital	Moderate	Bachelor's or master's degree
S29	M	53	Congenital	Mild	High school
S30	M	45	Congenital	Moderate	Bachelor's or master's degree
S31	F	51	Congenital	Moderate	Bachelor's or master's degree
S32	F	38	Congenital	Moderate	High school

S: Subject, M: Male; F: Female

affect the behavior, and participants who were distracted by the recording process were naturally observed.

Assessment of Parents' Levels of Knowledge About Hearing Aid Use

The Hearing Aid Awareness Question Form for Parents is a form created by the researchers for this study. In this form, parents were asked about their children's hearing aid use, hearing aid care, time used, battery tracking, mold tracking, etc. (Appendix A).

The Hearing Aid Awareness Question Form for Parents, which aims to evaluate hearing aid use, care and control information for parents, was filled. The clinician asked the questions and the form was completed by explaining the parts which the parent did not understand.

Assesment of Subjective Auditory Performance

The Parents' Evaluation of Aural/Oral Performance of Children (PEACH) scale was developed by Ching and Hill (22), and the Turkish validity/reliability studies were done by Eroğlu et al. (23). The scale aimed to assess the subjective hearing performance of children based on the observations of the parents. The PEACH scale consists of 13 items related to situations in quiet and noisy environments. A 4-point Likert scale is used for scoring. Three different scores were obtained as quiet environment, noisy environment and total score, and these scores were recorded as raw scores. During the interview, the parents were asked to answer the questions in the questionnaire, taking into account their child's behavior in the past week.

Statistical Analysis

The descriptive measures used were frequency and percentage values for categorical variables and mean and standard

deviation for normally distributed numerical variables, median and quartiles for measurements that did not fit to the normal distribution. The normality assumption of the data was studied with the Shapiro–Wilk's test and a histogram. Relationships between the non-normally distributed numerical variables were examined with Spearman's correlation coefficient (partial with age-adjusted), and the relationship between the dichotomous variables (0 or 1) and the numerical variables were examined with the point biserial correlation coefficient. The relationship between the ordered factors (taking values 0, 1, and 2) and the numerical variables was analyzed with the polyserial correlation coefficient. Some questions in the Hearing Aid Awareness Question Form for Parents, which were found to be significant in correlation analysis, data logging, MBRS-TV score, CBRS-TV score, and the effect of age on PEACH score were examined with a multivariate linear regression with backward variable selection. Correlation (r) value; 0.00–0.19 were taken as 'very weak', 0.20–0.39 as 'weak', 0.40–0.59 as 'medium', 0.60–0.79 as 'strong', and 0.80–1.00 as 'very strong' positive correlation. The significance level was accepted as <0.05. IBM SPSS Statistics version 22 (Armonk, NY: IBM Corp.) and R statistical programming language (version 3.4.3) were used in the analysis of the data.

Results

The demographic information and hearing aid daily use statistics of all participants are given in Table 2. No significant difference was observed between men and women in terms of age (p=0.117), data logging score (p=0.404), MBRS-TV (p=0.880), CBRS-TV (p=0.970), PEACH-QUIET score (p=0.287), PEACH-NOISE score (p=0.084) AND overall PEACH score (p=0.117) distribution.

Table 2. Demographic characteristics and summary statistics of participants

	Male	Female	All participants	p-value
Age, mean ± SD (median)	3.85±1.07 (3.00)	4.63±0.96 (5.00)	4.31±1.06 (4.00)	0.117
Maternal education				
Primary education	4 (30.8)	4 (21.1)	8 (25.0)	0.627
High school	4 (30.8)	9 (47.4)	13 (40.6)	
Bachelor's or master's degree	5 (38.5)	6 (31.6)	11 (34.4)	
Data logging, mean ± SD (median)	9.50±3.90 (10.80)	10.64±3.35 (11.20)	10.18±3.57 (10.95)	0.404
MBRS-TV, mean ± SD (median)	44.07±7.80 (46.00)	43.78±7.53 (46.00)	43.90±7.52 (46.00)	0.880
CBRS-TV, mean ± SD (median)	28.15±5.99 (29.00)	28.21±6.42 (28.00)	28.18±6.15 (28.50)	0.970
PEACH-QUIET score, mean ± SD (median)	22.07±3.66 (24.00)	21.78±2.34 (22.00)	21.90±2.90 (23.00)	0.287
PEACH-NOISE score, mean ± SD (median)	16.69±4.17 (18.00)	15.63±2.58 (15.00)	16.06±3.30 (16.50)	0.084
Overall PEACH score, mean ± SD (median)	38.76±7.72 (42.00)	37.42±4.37 (39.00)	37.96±5.88 (39.00)	0.117
N	13	19	32	

SD: Standard deviation, Data-logging: Hearing aid daily use (h), MBRS-TV: Turkish version of the Maternal Behavior Rating Scale, CBRS-TV: Turkish version of the Child Behavior Rating Scale, PEACH-QUIET: The Parents' Evaluation of Aural/Oral Performance of Children-QUIET, PEACH-NOISE: The Parents' Evaluation of Aural/Oral Performance of Children-NOISE, Overall PEACH: The Parents' Evaluation of Aural/Oral Performance of Children-Overall

The distribution of the answers obtained from the Hearing Aid Awareness Question Form is given in Figure 1. In the findings of the Hearing Aid Awareness Question Form created for parents, the best scores were seen in earmold change (96.9%), earmold tube change (96.8%) and regular hearing aids battery change (93.8%). The lowest scores were seen in the use of cleansing tablets for earmolds (21.9%), use of drying capsules for earmolds (28.1%), use of tamper-proof battery doors (53.1%) and use of pediatric clips hearing aid holder (53.1%).

No statistically significant relationship was found between the daily use of hearing aid and the Turkish version of the MBRS scores ($r_s=0.302, p=0.098$) (Table 3). There were no statistically significant correlations between individuals' daily use of the hearing aids and the CBRS-TV scores ($r=0.288, p=0.116$). High and statistically significant correlations were identified between the PEACH-QUIET subscale score, the overall PEACH score and the MBRS-TV scores ($r=0.734, p<0.001; r=0.704, p<0.001$, respectively). There were moderate and significant correlations between the MBRS-TV and the PEACH-NOISE subscale scores ($r_s=0.602, p<0.001$). Positive, moderate and significant correlations were also identified between the CBRS-TV and the PEACH-QUIET subscale scores, the PEACH-NOISE subscale score and the overall PEACH scores ($r=0.620, p<0.001; r=0.563, p=0.001; r=0.644, p<0.001$, respectively). Again, there was no significant correlation of data logging with the PEACH-QUIET subscale score ($p=0.096$), the PEACH-NOISE subscale score ($p=0.470$) and with the overall PEACH scores ($p=0.235$).

Positive and statistically significant correlation was found between the use of cleansing tablets for earmolds, which is the third item of the Parents' Hearing Aid Awareness Question

Form, and the CBRS-TV scores ($r=0.358, p=0.044$). Positive, moderate and significant correlation was identified between the scores obtained from question 9 'Do you check blockage or tear in the ear molds or tubes for clogs, tears, etc?' and MBRS-TV scores ($r=0.433, p=0.013$).

Similarly, positive and statistically significant correlations were found between the scores obtained from question 18 of the Hearing Aid Awareness Question Form 'Do you use a tamper-proof battery door against the risk of removing or swallowing the battery?' and the MBRS-TV ($r=0.350, p=0.049$) and CBRS-TV ($r=0.383, p=0.031$) scores.

Positive, moderate and statistically significant correlation was identified between the scores of question 20 in the Hearing Aid Question form, 'Does your child use the hearing aid willingly?' and the MBRS-TV ($r=0.503, p=0.003$). Positive, strong and statistically significant correlation was also found between the scores given to this question and the CBRS-TV ($r=0.700, p<0.001$). After comparing the daily use of hearing aid with the answers to questions asked, while moderate and significant correlations were found between the scores from questions 10, 11, 18, 20 and 24; positive, strong and statistically significant correlations were found with the scores from questions 19, 23 and 25 (Table 4).

There were statistically significant correlations close to a moderate level between question 19 of the Hearing Aids Awareness form 'Does your child wear the hearing aid during all waking hours?', the PEACH-QUIET subscale score ($r=0.378, p=0.033$), the noise subscale score ($r=0.379, p=0.032$), and with the overall PEACH score ($r=0.399, p=0.024$). Positive, strong and statistically significant correlations were identified between the scores of question 20 of the Hearing Aid Awareness form 'Does your child use the hearing aid willingly?', the PEACH-QUIET subscale score ($r=0.703, p<0.001$), the PEACH-Noise subscale score ($r=0.633, p<0.001$), and the overall PEACH score ($r=0.701, p<0.001$).

Question 25 of the Hearing Aids Awareness form 'How many hours on average does your child use the hearing aid?' was positively and significantly related to the PEACH-QUIET subscale score ($r=0.459, p=0.008$) and the overall PEACH score ($r=0.380, p=0.032$).

Results from the multivariate linear regression analyses (based on the backward stepwise variable selection method) for the overall PEACH score, the PEACH-QUIET subscale score, and the PEACH-NOISE subscale score are presented in Table 5. After removing the effects of all other variables included in the model, the scores from questions 17 and 24 of the Hearing Aid Awareness Question Form for Parents and the CBRS-TV score were found to be significant factors on the overall PEACH score. Findings from the analysis for the PEACH-QUIET subscale score alone revealed that

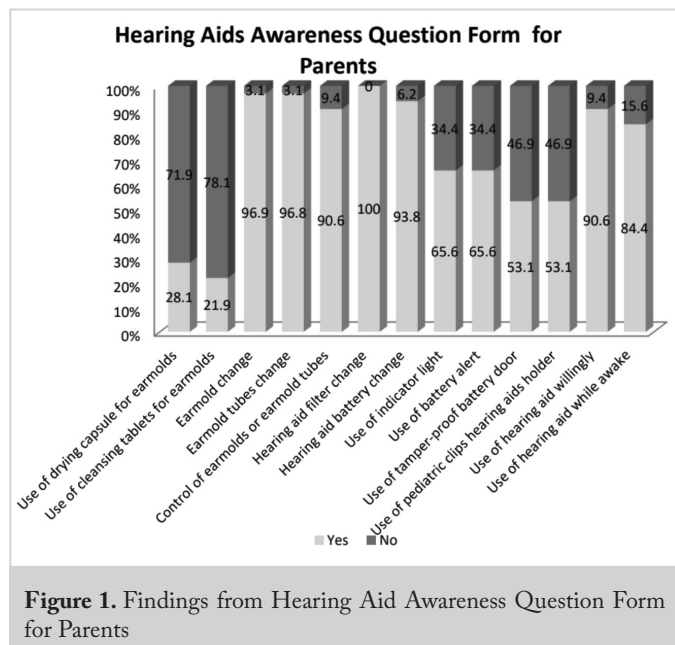


Figure 1. Findings from Hearing Aid Awareness Question Form for Parents

the scores from question 18 ($\beta=1.69, p=0.012$), question 24 ($\beta=1.74, p<0.001$), and the CBRS-TV ($\beta=0.27, p<0.001$) scores were positively associated with the PEACH-QUIET subscale performance. The results for the PEACH-NOISE subscale score alone showed that the scores from questions 17 and 24, and the CBRS-TV scores were associated with the PEACH-NOISE subscale score. A unit increase in the score given to question 24 of the Hearing Aid Awareness Question Form for Parents (i.e., using the hearing aids longer) caused a 2.35-fold increase in the PEACH-NOISE subscale score, a 1.74-fold increase in the PEACH-QUIET subscale score, and a 4.06-fold increase in the overall PEACH score. It was observed that the use of the pediatric clip hearing aids holder (question 17) had a significant effect in decreasing both the overall PEACH score ($\beta=-3.07, p=0.008$) and the PEACH-NOISE subscale score ($\beta=-1.88, p=0.012$) (Table 6).

Discussion

Auditory perception measurements and language development assessments are used to evaluate auditory performance in children using hearing aids. It has been reported that since children with hearing loss exhibit lower language skills and auditory performance compared to children with normal hearing, formal auditory test batteries used for young children did not match their real-life performances and parent observations would provide more accurate information (24, 25).

The sample group included in our study consisted of children who were diagnosed with hearing loss, started to use hearing aids in the early period, received regular auditory rehabilitation, did not have any additional disabilities, had hearing aids within the speech area, and whose receptive language developments were compatible with their peers. In the current study, we aimed to investigate how the correct use of the hearing aid, the parents' level of knowledge about the hearing aid, parent-child interaction and the daily use of the hearing aid affected the subjective auditory performance

of the children using hearing aids. To that end we used the MBRS, CBRS, PEACH and the Hearing Aid Awareness Question Form for Parents in our study.

Examination of the Relationship Between the Daily Use of Hearing Aids and Parent-Child Interaction

Moeller et al. (26) stated that parents should guide their children to continue wearing their hearing aids when they want to remove them, and it has been shown that both parent and child behaviors play a role in hearing aid use. Although the findings obtained from the current study are compatible with the referred study, it was thought that the reason why no significant relationship could be found between the parent-child interaction and the daily use of hearing aids may be due to the limited number of participants. Good interaction may make the parent think that the child is benefiting from the hearing aid and may encourage them to increase the wearing time. Parents should ensure that their children wear their hearing aids during all waking hours and increase the wearing time by showing the right behaviors.

Examination of Parents' Levels of Knowledge About Hearing Aid Use

Based on the results obtained from the Hearing Aid Awareness Questionnaire created by the researchers, it was concluded that parents had a lack of knowledge on some points about the use of hearing aids. In line with the responses given by the parents, earmold change, earmold tube change, battery change, checking the mold and tube for tearing/clogging while wearing the hearing aids were the most common controls by the parents. Parents were least informed about the uses of drying capsules, cleansing capsules, protective battery caps and pediatric clip hearing aids holders.

Our aim in creating a hearing aid awareness questionnaire was to reveal the issues that parents had difficulty with and were concerned about. Our findings revealed that parents had

Table 3. Correlation analysis of Hearing Aid Awareness Question Form for Parents with Hearing Aid daily use, and with other scale scores

Age adjusted correlations

	MBRS-TV	CBRS-TV	PEACH-QUIET subscale score	PEACH-NOISE subscale score	Overall PEACH score	Data logging
MBRS-TV	1.000	0.855 (<0.001)	0.734 (<0.001)	0.602 (<0.001)	0.704 (<0.001)	0.302 (0.098)
CBRS-TV		1.000	0.620 (<0.001)	0.563 (0.001)	0.644 (<0.001)	0.288 (0.116)
PEACH-QUIET subscale score			1.000	0.693 (<0.001)	0.881 (<0.001)	0.305 (0.096)
PEACH-NOISE subscale score				1.000	0.943 (<0.001)	0.135 (0.470)
Overall PEACH score					1.000	0.220 (0.235)
Data-logging						1.000

MBRS-TV: Turkish version of the Maternal Behavior Rating Scale, CBRS-TV: Turkish version of the Child Behavior Rating Scale, PEACH-QUIET subscale score: Parents' Evaluation of Children's Aural/Oral Performance in Quiet Environment, PEACH-NOISE subscale score: Parents' Evaluation of Children's Aural/Oral Performance in Noisy Environment, Overall PEACH score: Parents' Total Assessment of Children's Aural/Oral Performance, Data logging: daily use of hearing aid

difficulties in and lack of knowledge about the management of hearing aids, and this finding was supported by the information in the literature (16). In a study by Meibos et al. (27), most audiologists stated that parents should receive more training in hearing aid management. Given the consistency between our results and the literature, detailed information should be given to parents, both verbally and in writing, about hearing aid management.

Investigation of the Effect of Parents’ Knowledge About Hearing Aids on Regular Hearing Aid Use

Studies in the literature reported that parents lacked knowledge about using hearing aids, and that children could be encouraged to use their hearing aids for longer periods if and when parents are equipped with the knowledge and skills to support their children (10, 11, 17). Moreover, these studies reported that safety precautions (against swallowing the battery, putting the device in the mouth, etc.), the worry of losing their children’s hearing aids, the fear that the device

might break down as a result of the device getting wet or sweating, all negatively affect the duration of hearing aid use (26). In our study, we observed that children’s use of hearing aids for as long as they are awake, using them willingly, and using them with the necessary apparatus for safety measures increased daily usage time. In the light of these findings, we concluded that a parent with sufficient knowledge about the use of hearing aids increased the daily use of hearing aids by children.

Investigation of the Effect of Parents’ Knowledge About Hearing Aids on Parent-Child Interaction

It has been stated that children depend on their parents for managing their devices and parental attitudes were an important factor in the success of appropriate intervention (16, 17). The findings of our study revealed that families with better parent-child interaction also had better level of knowledge about hearing aids. It is thought that this could be because parents who are responsive to their child’s wishes

Table 4. Correlation analysis between the hearing aid awareness questions for parents and the MBRS-Turkish version, CBRS-Turkish version scores and daily use of hearing aids

Items	MBRS-TV	CBRS-TV	Data logging r (p-value)
Q1	0.249 (0.169)	0.227 (0.211)	0.108 (0.556)
Q2	0.258 (0.202)	0.225 (0.470)	0.263 (0.146)
Q3	0.333 (0.062)	0.358* (0.044)	-0.223 (0.219)
Q4	0.497 (0.069)	0.547 (0.287)	-0.192 (0.292)
Q5	-	-	-
Q6	0.203 (0.126)	0.099 (0.320)	-0.015 (0.934)
Q7	-	-	-
Q8	0.417 (0.089)	0.354 (0.625)	0.268 (0.138)
Q9	0.433* (0.013)	0.221 (0.224)	-0.199 (0.274)
Q10	-0.212 (0.243)	-0.290 (0.107)	0.513** (0.003)
Q11	-0.030 (0.265)	-0.272 (0.304)	0.379* (0.033)
Q12	0.046 (0.802)	0.035 (0.848)	0.112 (0.543)
Q13	0.198 (0.059)	0.103 (0.072)	0.070 (0.702)
Q14	0.093 (0.614)	-0.016 (0.929)	-0.116 (0.527)
Q15	0.188 (0.304)	0.137 (0.455)	0.028 (0.879)
Q16	0.287 (0.111)	0.302 (0.093)	0.252 (0.164)
Q17	-0.140 (0.444)	-0.153 (0.403)	-0.274 (0.130)
Q18	0.350* (0.049)	0.383* (0.031)	0.470** (0.007)
Q19	0.122 (0.505)	0.198 (0.277)	0.713** (<0.001)
Q20	0.503** (0.003)	0.700** (<0.001)	0.584** (<0.001)
Q21	0.118 (0.520)	0.021 (0.911)	0.064 (0.728)
Q22	-0.046 (0.242)	-0.273 (0.294)	0.261 (0.148)
Q23	0.314 (0.320)	0.176 (0.225)	0.686** (<0.001)
Q24	0.190 (0.116)	0.041 (0.395)	0.363* (0.041)
Q25	0.359 (0.198)	0.371 (0.290)	0.677** (<0.001)

Q: Question, Data logging: Daily use, MBRS-TV: Turkish version of the Maternal Behavior Rating Scale, CBRS-TV: Turkish version of the Child Behavior Rating Scale, r (p-value): correlation coefficient, *p<0.05, **p<0.001

Table 5. Correlations between the parent’s evaluation of auditory/oral performances of the children and the scores of the hearing aid awareness question form for parents

Items	PEACH-QUIET subscale score r (p-value)	PEACH-NOISE subscale score (p-value)	PEACH-total
Q1	0.116 (0.527)	0.330 (0.065)	0.242 (0.182)
Q2	0.053 (0.773)	0.272 (0.131)	0.176 (0.334)
Q3	0.203 (0.266)	0.106 (0.563)	0.159 (0.384)
Q4	0.172 (0.347)	0.049 (0.792)	0.103 (0.576)
Q5	-	-	-
Q6	-0.095 (0.605)	-0.146 (0.424)	-0.148 (0.419)
Q7	-	-	-
Q8	0.177 (0.333)	0.091 (0.622)	0.122 (0.507)
Q9	0.172 (0.345)	0.164 (0.370)	0.177 (0.333)
Q10	-0.054 (0.770)	-0.074 (0.685)	-0.068 (0.711)
Q11	0.106 (0.562)	0.000 (1.000)	0.062 (0.737)
Q12	0.057 (0.757)	0.059 (0.750)	0.061 (0.740)
Q13	-0.137 (0.453)	-0.173 (0.343)	-0.181 (0.321)
Q14	-0.049 (0.788)	0.231 (0.203)	0.105 (0.566)
Q15	0.032 (0.864)	0.167 (0.362)	0.109 (0.553)
Q16	0.212 (0.244)	0.139 (0.449)	0.182 (0.318)
Q17	-0.320 (0.074)	-0.384* (0.030)	-0.373* (0.035)
Q18	0.508** (0.003)	0.270 (0.134)	0.402* (0.023)
Q19	0.378* (0.033)	0.379* (0.032)	0.399* (0.024)
Q20	0.703** (<0.001)	0.633** (<0.001)	0.701** (<0.001)
Q21	0.088 (0.634)	0.115 (0.529)	0.108 (0.557)
Q22	0.157 (0.391)	0.157 (0.390)	0.163 (0.373)
Q23	0.345 (0.053)	0.251 (0.165)	0.291 (0.106)
Q24	0.362* (0.042)	0.402* (0.022)	0.397* (0.025)
Q25	0.459** (0.008)	0.291 (0.106)	0.380* (0.032)

Q: Question, r (p-value): correlation coefficient, PEACH-QUIET subscale score: Parents’ Evaluation of Children’s Aural/Oral Performance in Quiet Environment, PEACH-NOISE subscale score: Parents’ Evaluation of Children’s Aural/Oral Performance in Noisy Environment; *p<0.05, **p<0.001

Table 6. Predictors of Overall PEACH score, PEACH-QUIET subscale score, and PEACH-NOISE subscale score: findings of backward variable selection method

Response	Overall PEACH-score			PEACH-QUIET subscale score			PEACH-NOISE subscale score		
	Variables included in the model								
	Age, maternal education, data logging, Q17, Q18, Q19, Q20, Q24, Q25, MBRS-TV, CBRS-TV			Age, maternal education, data logging, Q18, Q19, Q20, Q24, Q25, MBRS-TV, CBRS-TV			Age, maternal education, data logging, Q20, Q24, Q17, Q19, MBRS-TV, CBRS-TV		
	Selected predictors according to the Backward Stepwise linear regression								
	Variable	β (SE)	p-value	Variable	β (SE)	p-value	Variable	β (SE)	p-value
	Q17	-3.07 (1.08)	0.008	Age	0.53 (0.31)	0.09	Q24	2.35 (0.52)	<0.001
	Q24	4.06 (0.80)	<0.001	Q18	1.69 (0.63)	0.012	Q17	-1.88 (0.70)	0.012
	CBRS-TV	0.60 (0.08)	<0.001	Q24	1.74 (0.42)	<0.001	CBRS-TV	0.29 (0.05)	<0.001
				CBRS-TV	0.27 (0.05)	<0.001			
R ² :	0.764			0.745			0.684		
SE of estimate	3.01			1.56			1.95		

Q: Question, PEACH-QUIET subscale score: Parents’ Evaluation of Children’s Aural/Oral Performance in Quiet Environment, PEACH-NOISE subscale score: Parents’ Evaluation of Children’s Aural/Oral Performance in Noisy Environment, Overall PEACH score: Parents’ Total Assessment of Children’s Aural/Oral Performance, SE: Standard error

(Evans JD. Straightforward Statistics for the Behavioral Sciences. 1st ed. Pacific Grove, Calif: Brooks/Cole Publishing; 1996. p.600.)

and needs can more easily solve the difficulties encountered in the use of hearing aids as a result of their responsive behavior. In line with the findings, we observed that children's fond use of their hearing aids were related to parent and child behavior.

Factors Affecting Auditory Performance

Studies have shown that children who used hearing aids full-time had better auditory performance and more vocabulary as the duration of auditory deprivation got shorter (5, 11, 14). The reason why no significant relationship could be found between the duration of daily use and subjective auditory performance is thought to be due to the limited number of samples. In addition, this situation suggests that the duration of daily use alone will not be a factor in subjective auditory performance. Therefore, in our study, we also assessed parent-child interaction, child behaviors and parents' knowledge levels about hearing aids along with their daily use times.

Janjua et al. (28) stated that parents of children with hearing loss should be extremely responsive to their children's wishes and behaviors and that a more child-centered interaction should be established by encouraging their children. In the presented study, we found that parent-child interaction had a significant effect on children's subjective auditory performances. We moreover observed that child behaviors along with parental behaviors had positive effects on auditory performance. Obtaining findings consistent with the previous study suggests that the literature is strengthened.

The questionnaire evaluating the parents' knowledge of hearing aid use revealed that better parental knowledge were associated with an improvement in subjective auditory performance. There are no resources available where parents can easily find sufficient information on hearing aid use and care after diagnosis and intervention (15, 16, 29, 30). In studies, parents stated that they removed the hearing aids when outside of their home because of concerns about losing the hearing aid (15, 16). In response to this information in the literature, the use of pediatric clip hearing aids holders for security measures has also been encountered with effective results on auditory performance. In a study conducted with preschool children, it was stated that most of the parents did not have the necessary knowledge and skills to understand whether or not the hearing aid was working properly (6). In our study, we observed that when the indicator lights of hearing aids were not used, parents had difficulty in understanding whether or not the device was working.

To summarize, in cases which the factors that can affect acoustic parameters are not taken into account (e.g., mold change, tube change, filter change, etc.), there may be a decrease in the subjective auditory performance scores, because consistent access to auditory stimuli cannot be achieved despite long-term use of hearing aids.

The present study boasts several notable strengths that contribute to its robustness, but besides the significant findings, this study is not without its limitations, which warrant careful consideration. The strongest aspect of the presented study is that it is the first study to assess the effects of parent-child interaction and hearing aid knowledge together on hearing aid use. While other studies focused on the factors affecting the use of hearing aids, such as education level, cognitive factors, starting age for hearing aid etc., in the current study, the factors that have a homogeneous distribution in terms of demographic characteristics and that have an effect on the auditory performance of the participants in the early childhood period have been assessed in detail.

In the study, the subjective auditory performance evaluation was made only in line with the information obtained from the parents in the evaluation of auditory performance. It was thought that the PEACH questionnaire alone would not suffice to evaluate the auditory performance of children with hearing loss. In future studies, objective assessment tools such as detailed auditory perception tests and cortical auditory evoked potential measurements can be used in addition to the scales to evaluate the auditory performance.

The Hearing Awareness Questionnaire created for the purposes of the study is a non-standard questionnaire since its validity and reliability has not yet been determined. It was used for the first time in this study as a continuation of the story form. It is planned to be expanded by standardizing in future studies.

The TEACH scale includes the evaluation of the auditory/verbal performance of children with hearing loss by their teachers. Since the Turkish validity/reliability study of the TEACH scale has not been performed yet, this scale was not included in our study. Only the effect of parental perspective on hearing aid use was investigated.

Conclusion

Our study has demonstrated a positive increase in auditory performance with the correct use of hearing aids. Since the resources available for parents to obtain information on hearing aid management are limited, the Hearing Aid Awareness Questionnaire was created, and thereby, comprehensive information was obtained to reveal the deficiencies in the use of hearing aids by parents. We concluded that the communication of parents with their children is quite effective on auditory performance and considering the positive effects of parental behavior on the child and vice versa, we suggest that it would be beneficial to add family education programs to auditory rehabilitation programs.

Ethics Committee Approval: The study was approved by Hacettepe University Non-Interventional Clinical Research

Ethics Committee on October 9, 2018 (decision no: GO 18/841-29).

Informed Consent: The research was carried out on a voluntary basis. As this study was conducted for research purposes, participants and researchers were asked to sign two written informed consent forms, i.e., the audiologist's explanation and the parent's permission statement for the child.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Concept: N.Ö., M.Ö.B., Design: N.Ö., M.Ö.B., Data Collection and/or Processing: N.Ö., M.Ö.B., Analysis and/or Interpretation: S.Y.I., Literature Search: N.Ö., M.Ö.B., Writing: N.Ö.

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Main Points

- Hearing loss is one of the most common types of childhood disorders and negatively affects language development, cognitive skills, social development, and educational process.
- After the diagnosis of hearing loss, these negative effects are reduced with appropriate audiological intervention and rehabilitation.
- Since children with hearing loss are dependent on their parents for the use of hearing aids, parents should be informed about the use of hearing aids.
- In addition, one of the factors affecting the auditory performance development of children with hearing loss is parent-child interaction, and parents should be informed about appropriate rehabilitative strategies.

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Appendix A. Hearing Aid Awareness Question Form for Parents and summary statistics

Item No	Hearing Aid Awareness Questions for Parents	Response		
Q1	1. Do you use a drying capsule for earmolds?	Yes 11 (34.4)		No 21 (65.6)
Q2	2. How often do you change the drying capsule for earmolds? (month)	0-2 10 (31.3)	2-3 -	3+ 22 (68.8)
Q3	3. Do you use a cleansing tablet for earmolds?	Yes 7 (21.9)		No 25 (78.1)
Q4	4. How often do you use earmold cleaning tablets in a week?	1 5 (15.6)	1-3 2 (6.3)	3+ 25 (78.1)
Q5	5. Do you change the earmolds regularly?	Yes 32 (100)		No 0 (0)
Q6	6. How often do you change your earmolds? (month/s)	0-6 28 (87.5)	6-12 2 (6.3)	12+ 2 (6.3)
Q7	7. Do you change the earmold tubes regularly?	Yes 32 (100)		No 0 (0)
Q8	8. How often do you change your earmold tubes? (month/s)?	0-3 16 (50)	3-6 12 (37.5)	6+ 4 (12.5)
Q9	9. Do you check the earmold or the earmold tubes for clogs, tears, etc.?	Yes 30 (93.8)		No 2 (6.2)
Q10	10. Do you change the hearing aid filter regularly, if it requires replacement?	Yes 30 (93.8)		No 2 (6.2)
Q11	11. If the hearing aids filter requires replacement, how soon do you change it? (month/s)	0-3 28 (87.5)	3-6 -	6+ 4 (12.5)
Q12	12. Do you replace the hearing aid batteries regularly?	Yes 31 (96.9)		No 1 (3.1)
Q13	13. How often do you replace hearing aid batteries? (says)	0-6 18 (56.3)	7-10 9 (28.1)	10+ 5 (15.6)
Q14	14. Do you keep the indicator lights on to monitor the working status of the hearing aid, battery warning, and program change, etc.?	Yes 19 (59.4)		No 13 (40.6)
Q15	15. Does the indicator light give a warning when the battery is low?	Yes 24 (75)		No 8 (25)
Q16	16. Do you use the buttons for volume/program control?	Yes 6 (18.8)		No 26 (81.2)
Q17	17. Do you use a pediatric clip hearing aids holder to prevent the hearing aid from falling off or getting lost?	Yes 15 (46.9)		No 17 (53.1)
Q18	18. Do you use a tamper-proof battery door against the risk of removing or swallowing the battery?	Yes 19 (59.4)		No 13 (40.6)
Q19	19. Does your child wear the hearing aid during all waking hours?	Yes 27 (84.4)		No 5 (15.6)
Q20	20. Does your child use the hearing aid willingly?	Yes 29 (90.6)		No 3 (9.4)
Q21	21. Does your child wear the hearing aid/s to kindergarten/preschool?	Yes 31 (96.9)		No 1 (3.1)
Q22	22. How many hours does your child use the hearing aid in kindergarten/preschool?	>5 27 (84.4)	2-5 3 (9.4)	<2 2 (6.2)
Q23	23. When does your child wear the hearing aid?	At wake up 21 (65.6)	At breakfast 4 (12.5)	After breakfast 7 (21.9)
Q24	24. When does your child take off the hearing aid?	At night 28 (87.5)	After dinner -	Before dinner 4 (12.5)
Q25	25. How many hours on average does your child use the hearing aid per day?	>8 26 (81.3)	6-8 1 (3.1)	<6 5 (15.6)



The Pitfalls and Perspectives of Assessing Olfactory Function via Optical Brain Imaging

Systematic Review

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Abstract

Olfaction is critical for maintaining daily life activities. It is crucial to measure olfactory performance for the diagnosis and treatment of certain neurodegenerative diseases. Moreover, impairments and a lack of quality in the olfactory system may indicate the early diagnosis of some diseases such as Parkinson's. In this context, there are several imaging methods available for evaluating olfactory function. In addition to the conventional methods used in measuring the brain's responsiveness to olfactory stimuli, this article presents a systematic review of the current applicability of optical brain imaging (i.e., functional near-infrared spectroscopy) in the evaluation of olfactory function. A database literature search was conducted in PubMed, Scopus, the Web of Science, and ScienceDirect. This review excluded animal studies, clinical studies, pathology- or neurodegenerative disease-related studies, newborn-related studies, cross-modal- and dual-task-related studies, and non-original research studies. Thus, seven studies were examined to discuss the pitfalls and perspectives of the use of optical brain imaging under olfactory stimulation. As for this conclusion, they can be used to evaluate olfactory performance in healthy individuals through the interpretation of hemodynamic changes. Further studies are needed to standardize the applicability of these optical imaging techniques.

Keywords: Near-infrared spectroscopy, olfaction, prefrontal cortex, hemodynamic brain response

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Introduction

Optical imaging technologies have gained a key role in the field of brain research in the last decade. These technologies involve non-invasive vascular-based neuroimaging methods such as functional near infrared spectroscopy (fNIRS) or near infrared spectroscopy (NIRS), and functional magnetic resonance imaging (fMRI). The basis of these methods is to map the changes in blood flow and blood hemoglobin changes in activated cortical regions (1, 2). When a particular cortical region is activated, cerebral blood flow transiently increases. The concentrations of oxyhemoglobin (Oxy-Hb), deoxyhemoglobin (HbR), or

the summed total hemoglobin (HbT) in the activated region can be indicative of the changes in blood flow (3). This condition is related to neuronal activity at the cortical capillary level over the scalp and can be measured with fNIRS and NIRS (4, 5). fNIRS is an effective and reliable imaging method for examining the relationship between hemodynamic changes and neural activity, providing consistent and parallel results with other hemodynamic imaging techniques such as fMRI and positron emission tomography (PET) (5, 6). Research with the fNIRS method has expanded rapidly as the fNIRS system has some advantages (compared to fMRI), such as relatively high temporal

resolution, portability, and insensitivity to electrical or magnetic devices (e.g., hearing aids, pacemakers, or cochlear implants) (7, 8).

In the last decade, with the development of its technology, the fNIRS method was used by multiple disciplines in various experimental designs to investigate cognitive functions as well as sensory systems under different conditions (4, 9). In addition to these studies, fNIRS was used in studies evaluating olfactory performance. Olfaction is one of the crucial perceptions for humans to avoid spoiled food or gas leaks and adds to the awareness of personal hygiene and provides the ability to enjoy the taste of food and beverages (10, 11). Impairment in the sense of smell has been associated with eating disorders (overeating or avoiding food), decreased quality of life, developing moderate to severe depression and resulting anxiety problems, social isolation, physical health problems, and increased mortality rate (12, 13). Olfactory dysfunction can occur due to various clinical reasons such as chronic sinusitis, viruses, nasal polyps, intranasal tumors, allergic rhinitis, pathological or congenital conditions (hypogonadotropic hypogonadism known as Kallmann syndrome) or presbyosmia (2, 11, 14). Idiopathic loss of smell can be a precursor symptom of Parkinson's disease. Patients with idiopathic olfactory loss have a high rate of getting Parkinson's disease in the following decade (10, 15).

An early diagnosis of olfactory dysfunction helps with successful treatment and opens the door to identifying the neurodegenerative disease before motor symptoms occur (16). Given the importance of olfaction, routine clinical evaluation of olfactory performance is essential. In clinical evaluation, MRI, fMRI, electroencephalography (EEG), and computed tomography are used to identify the structural and functional changes of the brain and to determine the etiology of olfactory disorders (16). In recent years, fNIRS/NIRS has become an alternative imaging technique for assessing olfactory performance. Despite the limited number of studies, optical imaging can offer promising techniques as they offer rapid evaluation of blood flow changes in the prefrontal cortex (PFC), and Oxy-Hb, HbR, and HbT parameters can be used to assess olfactory performance in healthy individuals and in patient groups (3).

With the aforementioned information, a question has arisen in recent years: can fNIRS be used as a reliable neuroimaging method to track olfactory changes? There are limited number of studies in the literature that have already shown that fNIRS is a useful technique in the evaluation of olfactory performance. In this review, we attempted to elucidate the current and potential applications of fNIRS and NIRS in olfaction-related studies.

Methods

Search Strategy and Resources

A systematic literature search in four databases (PubMed, Scopus, Web of Science, and ScienceDirect) was conducted in June 2022. All relevant studies using fNIRS/NIRS techniques for monitoring hemodynamic activity to assess olfactory performance are reviewed. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses diagram was used to choose articles. The following terms were used to search the three databases:

- NIR OR fNIRS OR "functional near-infrared spectroscopy" OR "near-infrared spectroscopy" OR "functional near-infrared spectroscopic" OR "optical imaging system".

- Olfaction OR smell OR olfactory perception OR sense of smell.

Inclusion and Exclusion Criteria

The relevant studies were selected for the PICOS [Participant(s), Intervention(s), Comparison(s), Outcome(s), and Study design] principle (17). The studies written in English and involving healthy subjects to measure hemodynamics with fNIRS/NIRS were included. The exclusion criteria were defined as follows: animal studies, clinical studies, pathology, or neurodegenerative diseases-related studies, newborns-related studies, cross-modal and dual-task studies, editorial letters, books, and book chapters.

Data Extraction

All potentially relevant studies (n=105) were retrieved. Their titles and abstracts were recorded in an Excel file, and duplicates (n=53) were removed by manual review. Articles that did not meet the inclusion criteria were excluded (n=40). Then, the full articles (n=12) were reviewed to identify whether the study met the eligibility criteria. Seven of the 12 eligible studies were used for this systematic review (Figure 1).

Results

Information (authors, title, methodology, and results) about the articles of seven studies are provided in Table 1. We analyzed the results section of all included articles by searching evaluating the Oxy-Hb, HbR, and HbT parameters. In these studies various odors such as B-phenyl ethyl alcohol, iso-valeric acid, γ -undecalactone, vanilla, strawberry, scatol, kyara, sumatra, samora, rakoku, managa, manaban, citral, human body odors and garlic were used to observe the hemodynamic changes in the prefrontal area of the brain. There were also mixed group characteristics in the evaluated studies, such as mothers, healthy subjects, and

“koh-do experts”. In five studies, NIRS were used while the other two studies employed the fNIRS as the monitoring tool. In four studies, Oxy-Hb, HbR and total Hb parameters were evaluated, while three studies evaluated general PFC without giving any details about the hemoglobin parameters.

In a study conducted with “koh-do” experts and non-experts, there was a significant difference in the right and left PFC during the discrimination odor task (18). Ishimaru et al. (19) conducted the study via NIRS, B-phenyl ethyl alcohol and iso-valeric acid exposure altered Oxy-Hb and HbT levels. Oxy-Hb correlated positively with odorant concentration. Oxy-Hb responses differed between odor and placebo stimulations (19). In another study using vanilla, scatol and strawberry odor as stimuli, found that scatol odor increased Oxy-Hb concentrations, notably in the frontal (orbitofrontal) region. During vanilla exposure, both frontal sides plateaued at 12–15 seconds. on the same time span, the strawberry odor changed somewhat but not statistically on either frontal side. These findings shed light on how different scents affect frontal brain regions, which may help us comprehend olfactory processing and sensory perception (20). An additional NIRS experiment conducted

by Kokan et al. (21), Participants correctly identified citral in six out of seven instances and rose in four out of seven throughout the olfactory identification task. Notably, participants who correctly recognized the odorant and those who did not, showed changes in HbT concentration in the left orbitofrontal cortex (OFC). However, neither between the groups exposed to citral and rose odorants nor between those exposed to pleasant and disagreeable odorants, there were any appreciable differences (21).

In another investigation employing fNIRS approach, researchers utilized strawberry odor stimulation to represent a pleasant odor, whereas garlic odor stimulation was employed to represent an unpleasant odor. The study’s findings indicate that sessions using odors, as opposed to sessions without odors, resulted in a significant reduction in PFC activation in both the left and right channels after exposure to olfactory stimulation (22). The findings of the study that used fNIRS method demonstrated a significant rise in Oxy-Hb concentrations following to the delivery of isovaleric acid. However, no significant changes in HbR levels were observed. It is notable that isovaleric acid demonstrated a significant increase in HbT levels in comparison to the control saline condition. The results of this study indicate that isovaleric acid produces unique changes in hemodynamic responses, particularly in terms of oxygenated hemoglobin levels (23).

In a study conducted with new mothers, a significant increase was detected in the PFCs of mothers compared to non-mother participants. It was thought that mothers who gave birth to a newborn may have an increased ability to recognize newborn odor compared to mothers who did not give birth to a newborn (24). The findings obtained from the reviewed studies reflect that different odor stimuli can cause a significant change in PFC activation and this activation can be captured by the fNIRS and NIRS methods.

Discussion

This review concluded that the fNIRS approach could be used to estimate hemodynamic changes in the brain in response to olfactory inputs. We provided evidence from the literature that fNIRS is an applicable method for measuring olfactory function in adults with no history of neurodegenerative disorders or any other pathological conditions. The scientific papers used for this review addressed various limitations and advantages.

Blood flow alterations in the PFC in response to olfactory stimuli are observable with fNIRS, the leading method for non-invasive optical imaging techniques. Because of the unique characteristics of olfaction, olfactory perception is not unidirectional. When a signal created by an odor molecule travels from the nose to the brain, activities such as identifying the odor, determining its intensity, recalling

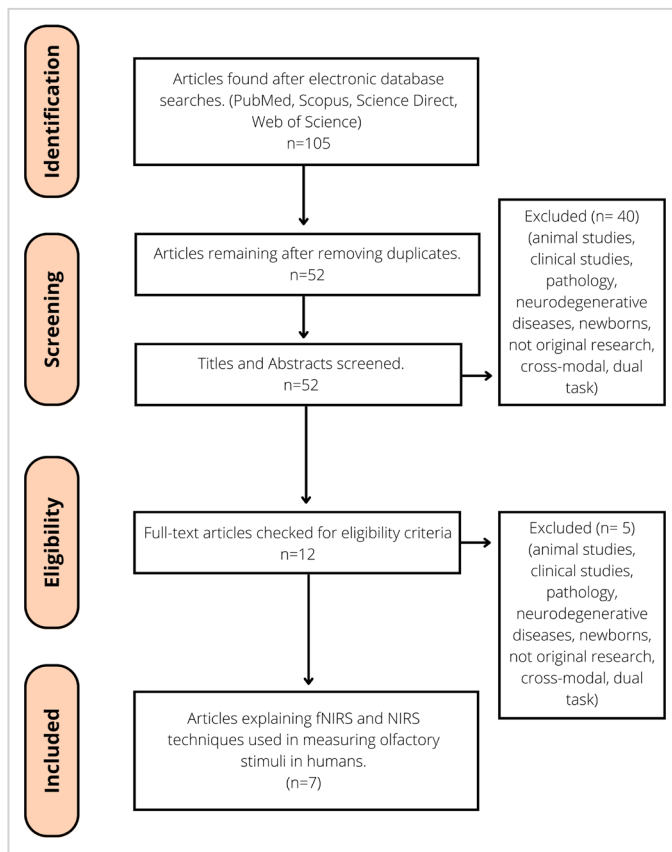


Figure 1. PRISMA flow diagram of the selection process of the studies

PRISMA: The Preferred Reporting Items for Systematic Reviews and Meta-Analyses, fNIRS: Functional near infrared spectroscopy, NIRS: Near infrared spectroscopy

memories triggered by the odorant, and perceiving the odorant occur simultaneously. Even if we cannot witness all these processes independently with fNIRS, it is crucial to be able to observe the hemodynamic changes in the brain generated by olfactory stimulation. Considering the seven articles that we selected based on our criteria, we discussed the results achieved using the fNIRS technique.

Fujii et al. (18) revealed that increased activation was observed in the PFC when distinguishing odor stimuli and representing multidimensional symbols of odor. Although there were differences in the responses of the right and left prefrontal regions, both hemispheres were involved in the odor-related reasoning process, and this universal effect could be demonstrated with NIRS. Different activations were observed in the right and left PFC, depending on

Table 1. A brief description of seven studies that used the fNIRS/NIRS method to evaluate olfactory performance

Studies	Optical imaging method	Sample	The type of stimuli	Results
Ishimaru et al. (19), 2004	NIRS	Twelve healthy subjects (eight males) with normal olfaction	- B-phenyl ethyl alcohol - Iso-valeric acid - γ -undecalactone	- Oxyhemoglobin (Oxy-Hb) and total hemoglobin (HbT) increased after the presentation of B-phenyl ethyl alcohol and iso-valeric acid. - Oxy-Hb increased in correlation with the odorant concentration. - The difference in Oxy-Hb responses between odor and placebo stimulation were significant ($p < 0.05$). - Significant periods of olfactory responses on the left side were shorter than on the right side.
Harada et al. (20), 2006	NIRS	Thirteen healthy subjects (seven men) with normal olfaction	- Vanilla - Strawberry - Scatol - Distilled water (negative control)	- Oxy-Hb concentration was increased in scatol odor in the frontal (orbitofrontal area). - A plateau is seen in 12–15 seconds on both frontal sides in vanilla odor. - There is a minor change seen in 12–15 s in strawberry odor but no significant changes on both frontal sides.
Fujii et al. (18), 2007	NIRS	Twenty healthy subjects (experts and beginners)	- Kyara - Sumatra - Samora - Rakoku - Managa - Manaban	- There is a significant difference between the right and left prefrontal cortexes (PFCs) of “koh-do” experts and beginners during the discrimination task.
Kobayashi et al. (23), 2007	fNIRS	Eight healthy subjects (four females)	- Isovaleric acid (cheesy odor) - Saline (as control)	- There is a significant increment in Oxy-Hb for isovaleric acid. - Besides, there are no significant changes in deoxyhemoglobin (HbR). - In HbT, isovaleric acid showed a significant increase compared to the saline.
Kokan et al. (21), 2011	NIRS	Four female college students with normal olfaction	- B-phenyl ethyl alcohol (rose-like odor) - Citral (lemon-like odor)	- Citral was detected correctly 6/7. - Rose was detected correctly 4/7. - HbT concentration in the left orbitofrontal cortex (OFC). - There are significant differences in HbT signal on the right OFC between participants who correctly identified the odorant or not. - There is no significant difference between the pleasant and unpleasant groups or citral or rose groups.
Nishitani et al. (24), 2014	NIRS	Thirty eight healthy female subjects (19 mothers and 19 non-mothers)	Human body odor: - Newborn infant odor - Adult male odor	- Mother subjects have significantly different PFC activation compared to non-mother subjects.
Moein et al. (22), 2020	fNIRS	Seventeen healthy subjects (seven males) with normal olfaction	- Strawberry - Garlic - Placebo (odorless)	- In odor sessions compared with odorless sessions, there was significantly decreased PFC activation in both left and right channels after odor stimulation.

fNIRS: Functional near infrared spectroscopy, NIRS: Near infrared spectroscopy

whether the subjects were expert or not. An increase was observed in the right PFC when discriminating and shaping symbols in the mind. An increase was also observed in the left PFC.

Ishimaru et al. (19) reported a significant increase in the right prefrontal region Oxy-Hb responses when comparing the olfactory stimulus (gamma-undecalactone) to placebo to assess olfactory performance. In another study, odor stimulation and water stimulus hemodynamic responses were observed to show no significant change against water stimulus, but an increase in Oxy-Hb was observed in the orbitofrontal region against the odor stimulus scatol. The increase observed in the orbitofrontal could be interpreted as increased blood flow and neural activation in this region. The increase in Oxy-Hb concentration only occurred against the olfactory stimulus, and no significant difference was observed in placebo and water stimulation (20). Moreover, hemodynamic changes to olfactory stimulation were observed in the left orbitofrontal region. When olfactory familiarity tasks were present, the right orbitofrontal region was activated beside the left OFC (21). Considering the aforementioned studies, fNIRS/NIRS technique can be used to show how olfactory stimulus changes blood flow in the prefrontal/orbitofrontal area.

In another study, hemodynamic changes were evaluated under the olfactory stimulus with a button press task. Participants were asked to press a button as soon as they detected any odor. The results showed a significant difference between odorless and odor-containing trials. The authors suggested that fNIRS could be a potentially useful technique for assessing olfactory performance (22). The study used saline (as control) and isovaleric acid (cheesy odor) and spotted a significant increment in Oxy-Hb for isovaleric acid odor. Besides, there were no significant changes in HbR. In HbT, isovaleric acid showed a significant increase compared to saline. The authors also suggested that multichannel near-infrared spectroscopy could provide the hemodynamic changes for odor stimulation (23). In another study, infant odor, which is known as a baby odor, significantly increased the bilateral prefrontal activity of female subjects who recently gave birth. In the same study, another interesting result was that male odors increased the prefrontal activity of female (both mother and non-mother) subjects similarly, but this increase was not significant (24). Based on the mentioned studies, it can be claimed that the hemodynamic activation of the OFC can be evaluated via the fNIRS system.

To understand the physiological effects of odor modulation on specific tasks, it is important to distinguish between simple olfactory stimulation and the effects of odor modulation on blood flow. Although finger-tapping was considered a control clue, whether the hemodynamic increase observed in the prefrontal regions of the participants who

were instructed to press the button when they perceived an odor was due to the odor stimulation or the decision-making process had to be determined (22). The activation of the PFC is accompanied by increased hemodynamic responses when stress is present. According to the researchers' findings, during mental arithmetic tasks that induce mental stress, the higher PFC activation and the stress factor shifted the activity of the PFC from the right to the left. Further research is required to determine whether the hemodynamic response observed with fNIRS is due to olfactory stimulus, cognitive function/cognitive load, or other emotional factors. Future studies should address questions such as: What distinguishes a pure odor stimulation from the hemodynamic changes caused by performing various tasks under odor modulation? Is the fNIRS-observed hemodynamic response due to olfactory stimulation or cognitive function/cognitive load? Are the hemodynamic responses to various odors (edible, unpleasant) identical? Which regions do we observe activation?

Conclusion

In this review, we compiled and summarized seven articles employing the fNIRS/NIRS techniques to measure olfaction-related activities. In the evaluation of olfactory performance, the fNIRS method appears more advantageous and promising than other neuroimaging techniques (EEG, PET, fMRI) based on the information presented in this review. Each method has its advantages and disadvantages, and we have detailed the disadvantages and limitations of the fNIRS method in the introduction and limitations sections. With further studies, it will be possible to complete the deficiencies in the literature fNIRS may establish itself as a standard procedure and become a reliable and valid technique. We hope that this review can encourage further studies that use the fNIRS method to investigate olfactory function. Considering the critical role of olfactory perception in human life, an early diagnosis of olfactory dysfunction will help successful treatment and open the door to identifying neurodegenerative diseases before motor or other symptoms occur. The fNIRS technique is expected to improve standard clinical assessments for early detection owing to its ease of use, portability, and lack of potential adverse effects.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Concept: G.A.Ö., G.E., E.E., Ç.G., Design: G.A.Ö., G.E., E.E., Ç.G., Analysis and/or Interpretation: E.E., Ç.G., Literature Search: G.A.Ö., G.E., E.E., Writing: G.A.Ö., G.E., E.E., Ç.G.

Conflict of Interest: The authors have no conflicts of interest to declare.

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Main Points

- This review investigates the possible and reliable usage of the non-invasive optical brain imaging method for assessing olfactory function.
- Optical brain imaging methods can be used to evaluate the olfactory performance in healthy individuals.
- Change in various parameters, such as oxyhemoglobin, deoxyhemoglobin, and total hemoglobin can be monitored in the prefrontal region via olfactory stimulation.

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Coexistence of Adenoid Cystic Carcinoma and Sialolithiasis in Submandibular Gland: Case Report

Case Report

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Abstract

Sialolithiasis is the primary cause of swelling in the major salivary glands. The etiopathogenesis is not clear. Adenoid cystic carcinoma is a slow-growing salivary gland malignancy with a poor prognosis. There are only a few cases in the literature reporting the coexistence of sialolithiasis and adenoid cystic carcinoma. In this report, we present a case that was thought to have sialolithiasis in the foreground because of the calcified image on computed tomography, but was diagnosed with adenoid cystic carcinoma after excision, together with a discussion of the relationship between sialolithiasis and carcinogenesis.

Keywords: Salivary glands, adenoid cystic carcinoma, sialolithiasis, malignancy, tumor, case report

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Introduction

Adenoid cystic carcinoma (ACC) is a slow-growing salivary gland malignant tumor with a poor prognosis. Its incidence is two per 100,000. It constitutes less than 1% of all head and neck cancers and less than 10% of all salivary gland neoplasms. Most of the cases are located in the major salivary glands. The 10-year survival is between 50–70%. Surgical excision and adjuvant radiotherapy (if necessary) are preferred for treatment (1).

Sialolithiasis is the primary cause of swelling in the major salivary glands. Its incidence has been reported between 1 in 30,000 and 1 in 10,000. It develops in the submandibular gland in 80–90% of the cases. Salivary gland stones are composed of a combination of organic and inorganic substances, including calcium

carbonates and phosphates, cellular debris, glycoproteins, and mucopolysaccharides. The pathogenesis of sialolithiasis is not clear (2).

In this article, we present the case of a patient who was thought to have sialolithiasis because of the calcified image on computed tomography (CT) but was diagnosed with ACC after excision together with a discussion of the relationship between sialolithiasis and carcinogenesis.

Case Presentation

A 65-year-old female patient was admitted to the Ear Nose and Throat Diseases Clinic with swelling under her left chin. She stated that the swelling had existed for five years and increased in size and pain over time. The patient had hypertension and was using antihypertensive drugs.

She had no history of smoking, alcohol, or previous surgery. The patient had no history of sialolithiasis or any other salivary gland disease. There was no feature in the family history. On physical examination, a solid, fixed, painless mass lesion measuring 3x2 cm was palpated in the left submandibular area. No additional finding was found in the routine ear, nose and throat examination. The lesion was evaluated with contrast-enhanced head and neck CT. On CT, a calcified lesion with a diameter of approximately 1.5 cm in the left submandibular gland and a hypodense area reaching 4 mm in the surrounding submandibular gland tissue of this lesion was observed (Figure 1). For this hypodense area, it was decided to perform an imaging-guided fine-needle aspiration biopsy to differentiate chronic inflammation and malignancy. The biopsy was reported as suspicious for malignancy. Submandibular gland excision was performed for definitive diagnosis. During the operation, it was observed that the submandibular gland adhered to the surrounding tissue and was difficult to dissect. The salivary stone was located intraparenchymal. Histopathological examination was reported as grade 2 ACC. The tumor, measuring 2.5x2.2x2.1 cm, showed tubular and solid features, extracapsular invasion, and diffuse perineural invasion (Figure 2). Surgical margins were positive for the tumor. As a result of these findings, it was decided to perform a left supraomohyoid neck dissection. The primary lesion area was also re-excised with the neck dissection. No metastatic lymphadenopathy was observed in the histopathological examination of the neck dissection specimen. A residual invasive tumor was observed in the 3 mm area of the scar tissue. Due to capsule invasion, perineural invasion, and grade 2 tumor, a total of 66 Gy radiotherapy in 2.2 Gy/30 fractions was applied to the primary tumor area with intensity modulated radiation therapy technique as adjuvant therapy. The department of medical oncology did not recommend adjuvant chemotherapy. The patient was closely followed. No pathological finding was observed in the magnetic resonance imaging (MRI) of the neck six months after the operation.

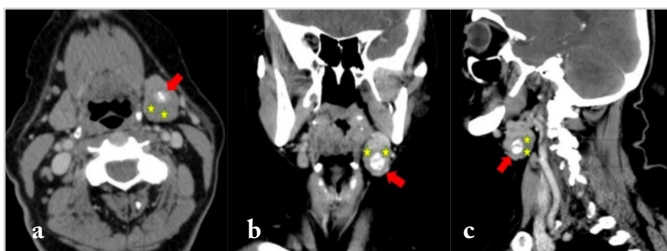


Figure 1. A calcified stone in the central part of the left submandibular gland (red arrow; AP×T×CC: 15×9×14 mm) in contrast-enhanced neck CT [a] axial, b) coronal, and c) sagittal planes]. A hypodense area (yellow stars) was observed that represent inflammatory-edematous changes caused by the stone. The left submandibular gland is larger than the right one
CT: Computed tomography

Informed consent was obtained from the patient for this report.

Discussion

ACC is a slow-growing salivary gland malignancy with a poor prognosis, the mean age at diagnosis is 57 years, and it is more common in women. Patients often present with complaints of swelling (1). Its association with sialolithiasis is very rare and there are only a few reported cases (3, 4). The relationship of sialolithiasis with salivary gland neoplasms is unknown (3). Some studies in the literature suggested that sialoliths might cause tumor formation or, conversely, that sialoliths might form in the presence of tumors (5). It is also stated that chronic inflammation due to obstruction by sialoliths could be related to oncogenesis (3-5). Another view suggests that sialolite and tumor formation could be caused by common predisposing factors rather than a cause-and-effect relationship (6). Hasegawa et al. (4) reported a case with the coexistence of ACC and sialolithiasis. They drew attention to the relationship between lithiasis and related inflammation and carcinogenesis in organs other than salivary glands with examples of cholangiocarcinoma-hepatolithiasis, pancreatic carcinomas-pancreatitis/pancreatolithiasis. The authors stated that dysplasia detected histopathologically in the bile duct epithelium of patients with hepatolithiasis could also be present between sialolithiasis and salivary gland carcinogenesis, although an objective relationship could not be demonstrated between them (4). The relationship between chronic inflammation and various epithelial malignancies has been known for many years. Increased risk of malignancy in patients with inflammatory bowel and esophageal carcinoma that may develop after reflux esophagitis are examples of this relationship (7). A case of squamous cell carcinoma occurring from the Wharton's duct epithelium in a patient with sialolithiasis has also been reported (8). In this case, squamous metaplasia of ductal epithelial cells surrounding the stones and histopathological indicators of malignant transformation foci suggested a relationship between carcinogenesis and chronic inflammation (7).

The duration of symptoms in some cases with the coexistence of sialolithiasis and salivary gland neoplasm in the literature is remarkable. The swellings duration was 10 years in Miyabe et al.'s (9) and Hasegawa et al.'s (4) cases, 3 years in Gallego et al.'s (7) case, and 1.5 years in Nanda and Mehta's (10) case. In our study, as in these cases, the swelling was present for a long time (5 years). This may support the hypothesis of malignant development as a result of prolonged exposure to inflammation.

Ultrasonography is frequently used as the first-line radiological examination of salivary gland lesions. CT and MRI provide more detailed imaging. CT can successfully show the features of the stone in a patient

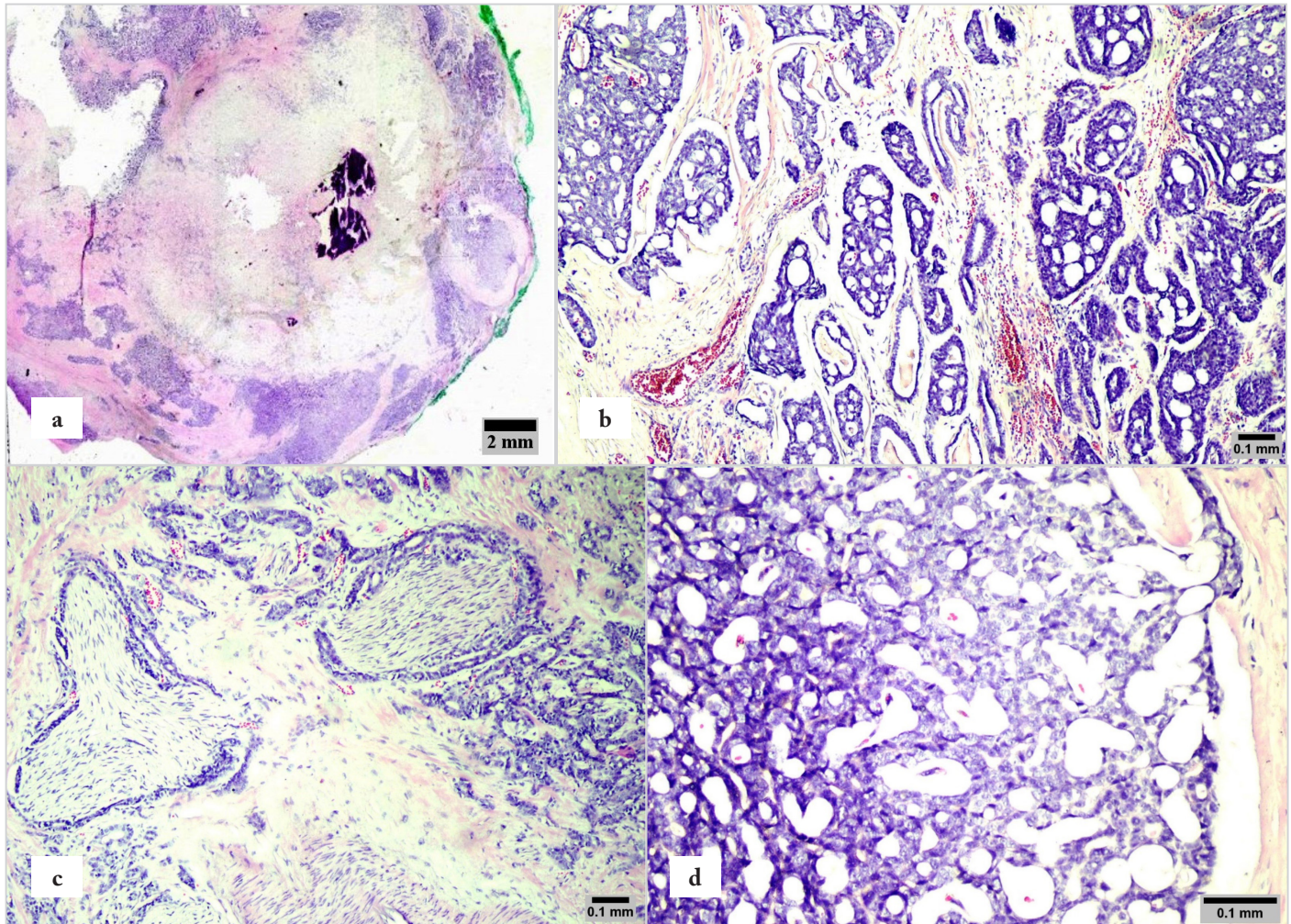


Figure 2. Images of the histopathological examination of the material after submandibular gland excision. a) Tumoral lesion consisting of centrally dystrophic calcification and hyalinization, peripheral solid islands, and scattered cells in between (x40, full slide scanning; H&E), b) Tubular cell groups with tubular and cribriform structure (x100; H&E), c) Perineural invasion areas (x100; H&E), d) Tumoral cells with large vesicular nuclei, prominent nucleoli, and extensive eosinophilic cytoplasm (x200; H&E)

with sialolithiasis. MRI, on the other hand, shows soft tissue lesions in more detail (2, 3). Batzakakis et al. (3) argued that ultrasonography alone was not sufficient in the planning of operations in patients with sialolithiasis and that such cases should be evaluated with MRI in the presence of suspected malignancy. However, contrast-enhanced CT can also successfully distinguish soft tissue lesions, as in our case. Therefore, we believe that MRI or contrast-enhanced CT may be preferred as an advanced imaging technique in these cases. If solid lesions other than stones are detected in the salivary gland in radiological examinations, we recommend performing a biopsy with suspicion of malignancy.

Conclusion

The coexistence of ACC and sialolithiasis is extremely rare. Especially in cases with long disease duration, the entire salivary gland should be carefully examined via imaging. In

the presence of suspicious solid areas, imaging-guided fine-needle aspiration biopsy can be performed in these areas before surgical treatment. More comprehensive studies are needed to clarify the cause-effect relationship in these cases.

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

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: V.A., M.E.S., Concept: V.A., M.E.S., V.A.A., M.K., Design: V.A., M.E.S., V.A.A., M.K., Data Collection and/or Processing: V.A., M.E.S., V.A.A., M.K., Analysis and/or Interpretation: V.A., M.E.S., V.A.A., M.K., Literature Search: V.A., M.E.S., V.A.A., M.K., Writing: V.A., M.E.S., V.A.A., M.K.

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Main Points

- The coexistence of adenoid cystic carcinoma and sialolithiasis is extremely rare.
- Especially in cases with long disease duration, the entire salivary gland should be carefully examined via imaging.
- In the presence of suspicious solid areas, imaging-guided fine-needle aspiration biopsy can be performed in these areas before surgical treatment.
- More comprehensive studies are needed to clarify the cause-effect relationship in these cases.

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Atypical Lipomatous Tumor Originating From the Nasopharynx in a Patient with Chronic Lymphocytic Leukemia

Case Report

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Abstract

Atypical lipomatous tumor, also known as well-differentiated liposarcoma, is rare in the head and neck region. The primary and most effective option in the treatment of this malignancy with a good prognosis is excision with clean surgical margins. Therefore, it is important to distinguish this malignancy from lesions that require more aggressive treatment. In this article, we present the case of an atypical lipomatous tumor originating from the nasopharynx and almost completely obstructing the oropharynx in a 38-year-old male patient with chronic lymphocytic leukemia. To the best of our knowledge, this is the first report in the literature of an atypical lipomatous tumor case originating from the nasopharynx.

Keywords: Nasopharynx, atypical lipoma, liposarcoma, chronic lymphocytic leukemia, neoplasms, case report

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Introduction

Liposarcoma is the most common type of sarcoma in adults. In the classification of head-neck tumors published by the World Health Organization in 2017, liposarcomas are histologically classified into three subgroups: well-differentiated, myxoid, and pleomorphic. The well-differentiated liposarcoma is also known as an atypical lipomatous tumor (1, 2). The head and neck location comprises only 5.6% of all liposarcoma cases (3). In this region, they can originate in the soft tissues of the pharynx, the larynx, the oral cavity, and the neck (1, 2, 4). There are

no reported cases of atypical lipomatous tumors localized in the nasopharynx. In this article, we present the case of a patient with chronic lymphocytic leukemia (CLL) with an atypical lipomatous tumor originating from the nasopharynx.

Case Presentation

A 38-year-old male patient presented to our clinic with a mass in his throat. He stated that he had noticed the mass for the first time two years ago and the mass had grown over time. He reported that he had increasing dyspnea, dysphagia, and obstructive sleep apnea and that in recent months, he could only swallow liquid

foods. He had a history of CLL. A well-circumscribed mass with yellow-pink-colored mucosa and local vascularity was observed. The mass was completely obstructing the passage between the soft palate and the base of the tongue and pushing the uvula to the anterosuperior. The pedicle of the mass protruded from the inferomedial region of the torus tubarius on the right side of the nasopharynx (Figure 1). The patient had contrast-

enhanced magnetic resonance imaging (MRI) performed 22 months ago and computed tomography performed 11 months ago (Figures 2, 3). The patient underwent orotracheal intubation with a video laryngoscope. The soft palate was suspended with a feeding tube inserted through the nostril. The area where the mass originated from the nasopharyngeal wall was also visualized by nasal endoscopy. The pedicle of

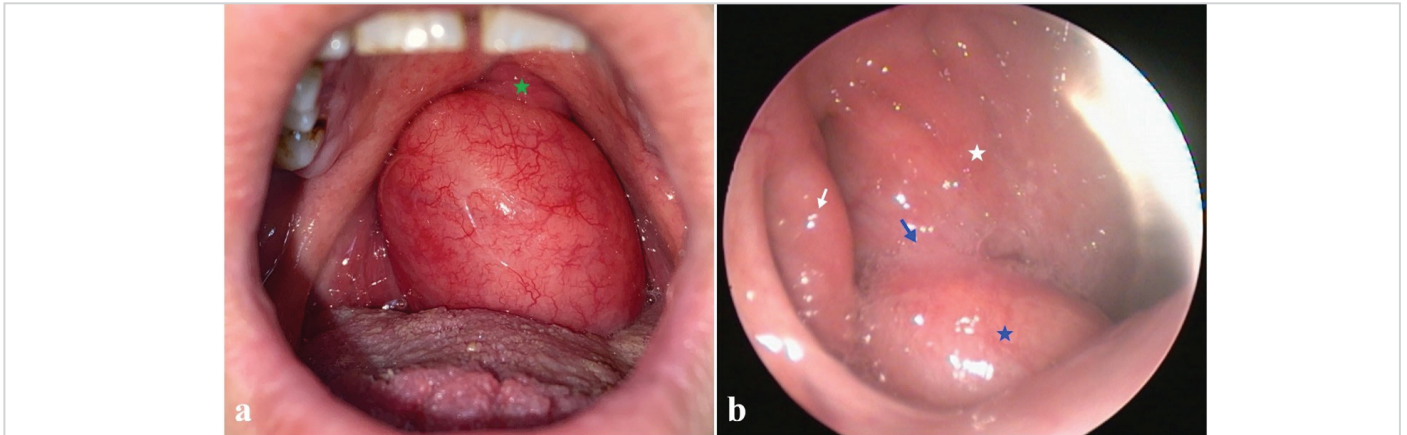


Figure 1. Gross appearance of the lesion: a) Oropharyngeal view, b) Nasopharyngeal view (green asterisk: uvula; white arrow: torus tubarius; white asterisk: posterior wall of the nasopharynx; blue asterisk: mass; blue arrow: peduncle of the mass)

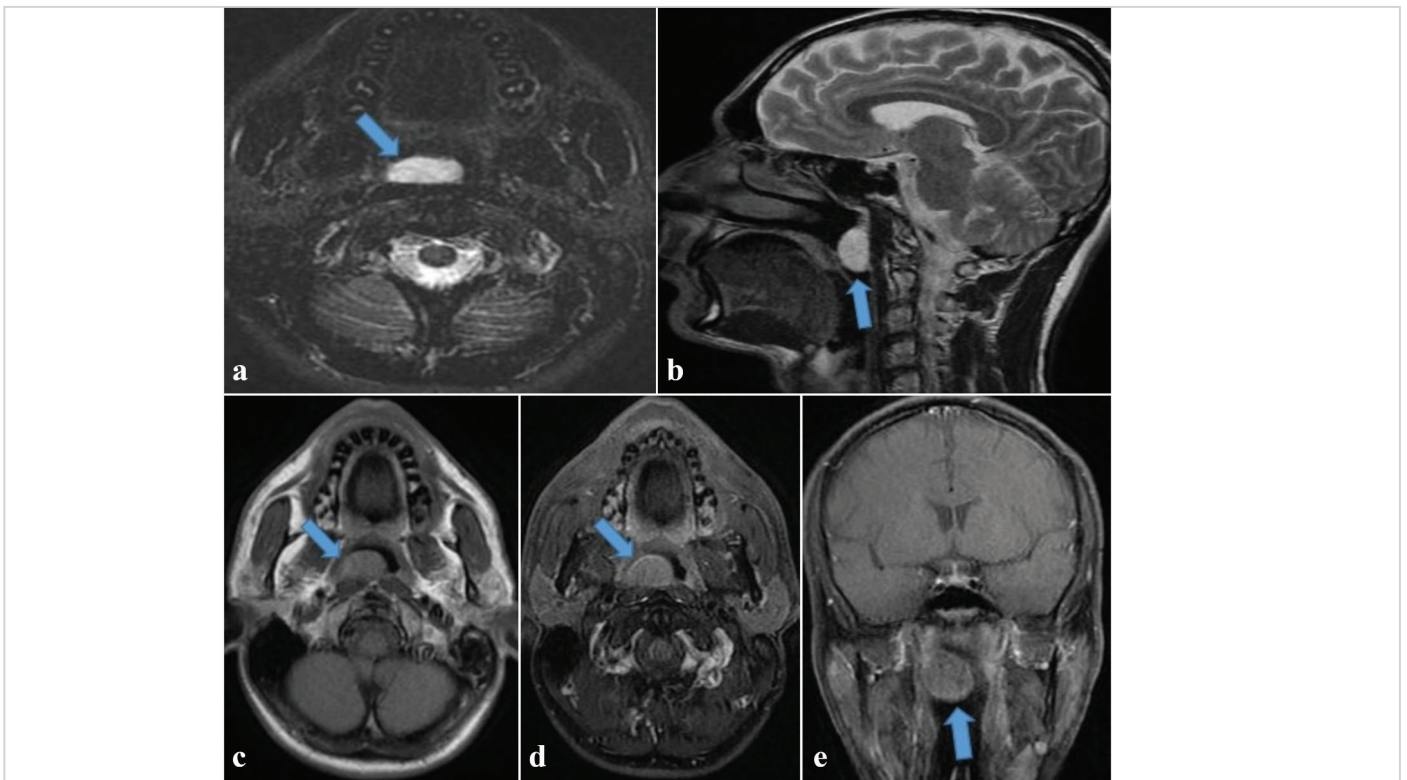


Figure 2. Contrast-enhanced nasopharyngeal MRI images taken 22 months before admission: a) Fat-suppressed T2W axial view, b) T2W sagittal view, c) T1W axial view, d) Contrast-enhanced T1 view, e) Fat-suppressed T1 coronal view (blue arrow: mass). MRI revealed a mass on the right side of the midline extending from the posterior inferior wall of the nasopharynx to the oropharynx, hyperintense in fat-suppressed T2W, hypointense in T1W, and without contrast enhancement on postcontrast fat-suppressed T1-weighted images. The MRI demonstrated that the mass was a 23×13×27 mm homogeneous lesion with an oval shape, smooth contours, and no contrast-enhancing septal structure

MRI: Magnetic resonance imaging

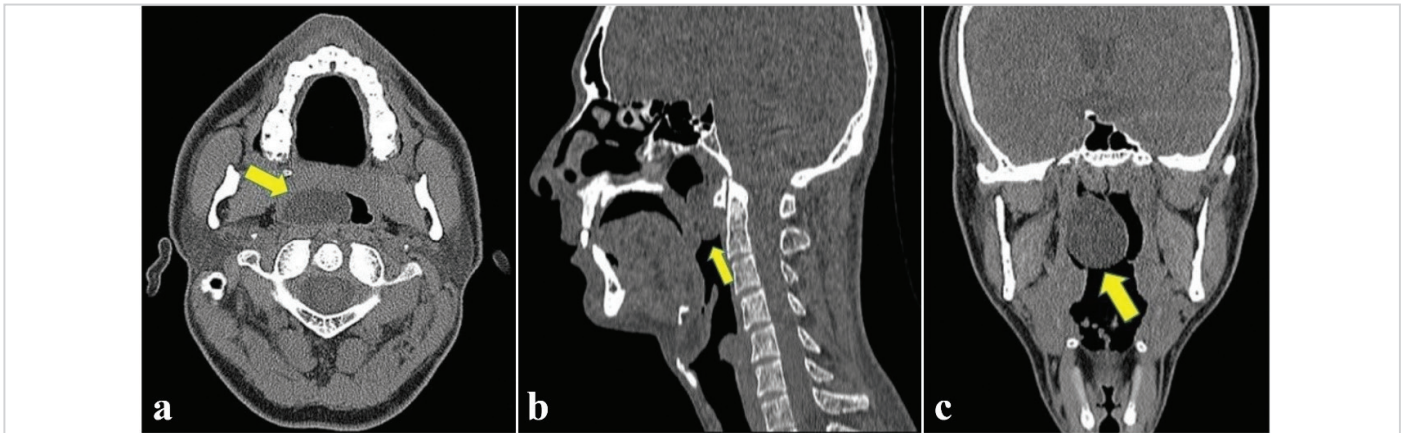


Figure 3. Non-contrast paranasal sinus computed tomography images taken 11 months before admission: a) axial, b) sagittal, and c) coronal (yellow arrow: Mass). The paranasal sinus CT report described the mass as a homogeneous lesion with dimensions of 29×14×30 mm and an average density of -5 Hounsfield Units. It was an oval-shaped, well-contoured capsulated mass structure extending from the posteroinferior wall of the nasopharynx to the oropharynx on the right side of the midline

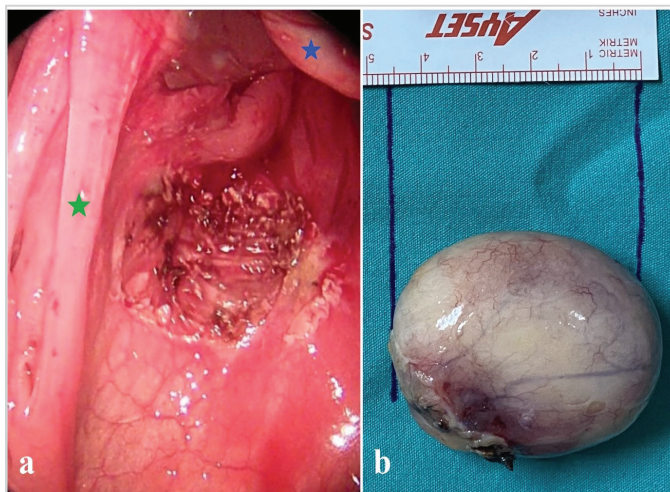


Figure 4. a) The location of the pedicle after removal of the mass (green asterisk: posterior plica; blue asterisk: uvula), b) gross specimen

the mass was dissected with a transoral approach using bipolar cautery (Figure 4). Histopathological examination of the mass reported an atypical lipomatous tumor. Surgical margins were clean, and lymphovascular invasion was not observed. The tumor had patchy staining with CD34. Ki-67 positive cells were 5% (Figure 5). At the one-month follow-up, the patient’s breathing and swallowing complaints were completely resolved (Figure 6). We asked for the results of the patient’s genetic examination tests performed for CLL in previous years. He was positive for 11q22.3 deletion. No recurrence was observed in the patient’s six-month follow-up examination. He was followed up clinically at three-month intervals.

Informed consent was obtained from the patient.

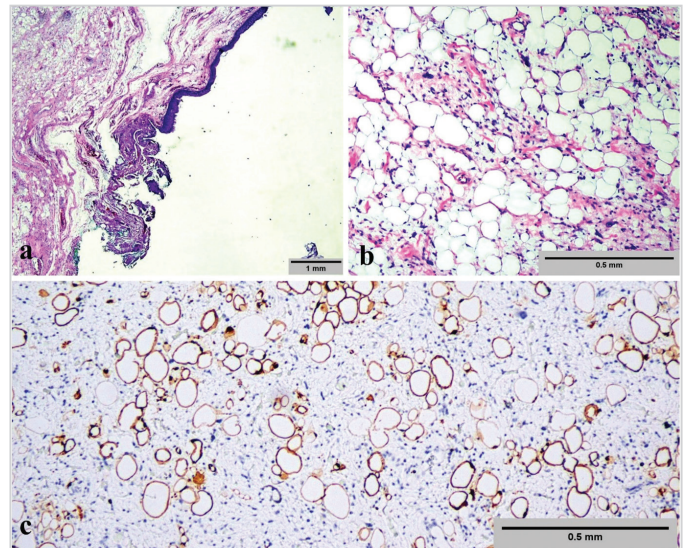


Figure 5. Histologic features of the mass: a) a well-circumscribed and thinly encapsulated lesion adjacent to the surface epithelium (H&E stain x20), b) variably sized adipocytes (H&E stain x100), c) positive staining with S100 was observed in adipocytes (S100 DAB, x100)

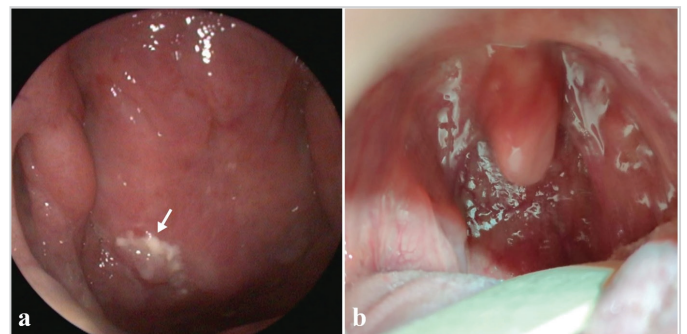


Figure 6. a) Nasopharynx and b) oropharynx view one month after the operation (white arrow: granulation tissue at excision site)

Discussion

Radiation, trauma, and genetic factors are blamed for the etiology of liposarcomas (2). Our patient did not have a history of radiation or trauma, but in his genetic tests for CLL, 11q22.3 (ATM) deletion was positive. This deletion has been reported as associated with a poor prognosis for CLL patients (5). No study has shown the relationship between this deletion and liposarcomas. 11q22.3 deletion has been associated with some solid cancers as well as hematological malignancies (6). This deletion, for which a direct relationship with the development of atypical lipomatous tumors could not be established, needs to be evaluated with further studies and case series. Moreover, the association of CLL with an atypical spindle cell lipomatous tumor was reported in one case (7). The fact that another case of atypical lipomatous tumor with CLL has been reported in the literature also makes our case interesting.

Liposarcomas develop more frequently in men, and the average age at diagnosis is 60 years (1, 2). These sarcomas are characterized by slow-growing, submucosal, well-circumscribed, and painless masses (1, 2). They are usually encapsulated (2). Symptoms vary by region of origin. Those located in the oral cavity, the larynx, and the pharynx may cause dysphagia and dyspnea, depending on their size (1, 2). In their article published in 2012, Papacharalampous et al. (8) reported a case of myxoid-round cell liposarcoma with moderate differentiation located in the nasopharynx. This was the case of a 58-year-old man, and chemoradiotherapy was applied to the patient because curative surgery was not possible. No recurrence was observed in the 14-month follow-up of the patient. In the same article, the authors stated that they had identified four more cases of nasopharyngeal localization in the English literature review. Two of these cases were female and two were male, the youngest was 12 years old and the oldest was 58 years old. In one of these cases, the histopathological subtype could not be determined, while two of them showed myxoid features, and one was a well-differentiated sclerosing subtype. In these five cases, cure could not be achieved with surgical excision alone. Radiotherapy was also applied in all cases, and chemotherapy was added to the treatment in two cases (8). In 2022, Nishith et al. (9) reported a case of low-grade dedifferentiated liposarcoma originating from the nasopharynx. This was a 36-year-old male patient and chemoradiotherapy was preferred. Unlike the other nasopharyngeal liposarcomas reported in the literature, our case could only be treated with excision. The reasons for this are that the lesion was pedicled and well-differentiated.

MRI is extremely helpful in the differential diagnosis of liposarcomas. In well-differentiated liposarcomas, the MRI image of the tumor is similar to that of normal adipose tissue. However, contrast enhancement indicates the level of

differentiation. Contrast enhancement is minimal in well-differentiated lesions, while more intense enhancement is observed in more aggressive subtypes (4).

Atypical lipomatous tumors have enlarged adipocytes of varying sizes, with hyperchromatic and enlarged nuclei (1, 2). They can be distinguished from benign lipomas because they contain multivacuolar lipoblasts. Immunohistochemical staining of lipoblasts with S-100 is essential for differential diagnosis (4). The tumor consists of lobules of adipose tissue separated by a thick fibrous band. Mild cytological atypia may be observed in a well-differentiated liposarcoma (10). Necrosis and mitotic activity are rare (4). More than 90% of liposarcomas are MDM2- and CDK4-positive. Likewise, 12q13-15 amplification is positive in atypical lipomatous tumors (1). Detection of MDM2 amplification is a sensitive and specific method for distinguishing well-differentiated liposarcoma from benign lipoma (10). The histopathological examination results of our case are also compatible with the literature. The mass consisted of well-circumscribed, encapsulated lesions with large nuclei and S-100 positive staining adipocytes.

Excision of the lesion with clean surgical margins usually provides effective treatment (1, 2). Localization and tumor grade are important factors in prognosis (1, 2). The rate of distant metastases in atypical lipomatous tumors is reported as less than 6%, and they most commonly metastasize to the lung. Regional nodal metastases are also extremely rare (2, 10). Atypical lipomatous tumors can recur at a rate of 30%, so long-term follow-up is essential (2). Radiotherapy may be preferred in unresectable tumors, although the effectiveness of radiotherapy is unclear, and there is no evidence of the efficacy of chemotherapy (2, 10). Regional lymph node dissection is not recommended (2). These tumors can sometimes differentiate or transform into high-grade sarcomas. This transformation occurs as a result of a delay in treatment or inadequate excision (4).

Conclusion

To our knowledge, this is the first reported case of an atypical lipomatous tumor originating from the nasopharynx. This pathology, with typical clinical and radiological features, should be included in the differential diagnosis of nasopharyngeal masses. The association with CLL and 11q22.3 deletion needs to be investigated in further studies.

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Authorship Contributions

Surgical and Medical Practices: M.E.S., V.A., B.B., H.Y., Concept: M.E.S., V.A., B.B., H.Y., M.K., V.A.A., Design:

M.E.S., V.A., H.Y., M.K., V.A.A., Data Collection and/or Processing: M.E.S., V.A., B.B., M.K., V.A.A., Analysis and/or Interpretation: M.E.S., V.A., M.K., V.A.A., Literature Search: M.E.S., V.A., B.B., H.Y., M.K., V.A.A., Writing: M.E.S., V.A., B.B., H.Y., M.K., V.A.A.

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Main Points

- An atypical lipomatous tumor, also known as well-differentiated liposarcoma, is rare in the head and neck region.
- This is the first case in the literature of an atypical lipomatous tumor originating from the nasopharynx.
- Excision of the lesion with clean surgical margins usually provides effective treatment.
- Further studies are needed to show the relationship of atypical lipomatous tumor with chronic lymphocytic leukemia and 11q22.3 deletion.

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