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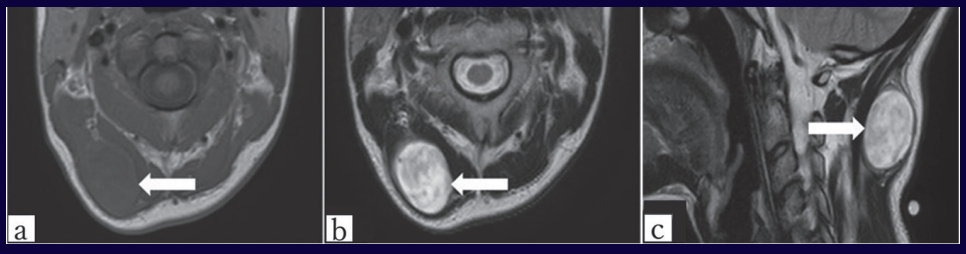

Posterior Cervical Intramuscular Schwannoma Within the Trapezius Muscle: A Case Report Page 105-8

Original Investigations

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Turkish Archives of Otorhinolaryngology
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Aims and Scope

The Turkish Archives of Otorhinolaryngology (Turk Arch Otorhinolaryngol) is the scientific, peer-reviewed, open-access journal of the Turkish Otorhinolaryngology-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readers and experts concerning the published studies, articles about general practice and subject of the journal with historical content, memories of scientific significance, educative and catechetical manuscripts about medical deontology and publication ethics.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 Otorhinolaryngology 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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Open Access Policy

This journal provides immediate open access to its content on the principle that making research freely available to the public supports a greater global exchange of knowledge.

The Turkish Archives of Otorhinolaryngology's Open Access Policy is based on the rules of Budapest Open Access Initiative (BOAI). By "open access" to [peer-reviewed research literature], we mean its free availability on the public internet, permitting any users to read, download, copy, distribute, print, search, or link to the full texts of these articles, crawl them for indexing, pass them as data to software. The only constraint on reproduction and distribution, and the only role for copyright in this domain, should be to give authors control over the integrity of their work and the right to be properly acknowledged and cited.

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All of the electronic content (website, manuscript, etc.) is stored in three different sources. Content on a server is online and accessible to readers. A copy of the same content is preserved as a backup on other servers. Should a server fail, other resources can be brought online, and the website is expected to be available in 24-36 hours.

Abstracting/Indexing Services

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

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
Türk Otorinolarengoloji Arşivi

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.

DECLARATION OF INTEREST

The Turkish Archives of Otorhinolaryngology requires and encourages the authors and the individuals involved in the evaluation process of submitted manuscripts to disclose any existing or potential conflicts of interests, including financial, consultant, and institutional, that might lead to potential bias or a conflict of interest. Any financial grants or other support received for a submitted study from individuals or institutions should be disclosed to the Editorial Board. To disclose a potential conflict of interest, the ICMJE Potential Conflict of Interest Disclosure Form should be filled in and submitted by all contributing authors. The journal's Editorial Board resolves cases of a potential conflict of interest of the editors, authors, or reviewers within the scope of COPE and ICMJE guidelines.

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The Editorial Board of the journal handles all appeal and complaint cases within the scope of COPE guidelines. In such cases, authors should get in direct contact with the editorial office regarding their appeals and complaints. The Editor in Chief is the final authority in the decision-making process for all appeals and complaints.

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Open access is an approach that supports interdisciplinary development and encourages collaboration between different disciplines. Therefore, the Turkish Archives of Otorhinolaryngology contributes to the scientific publishing literature by providing more access to its articles and a more transparent review process.

MATERIAL DISCLAIMER

Statements or opinions expressed in the manuscripts published in the Turkish Archives of Otorhinolaryngology reflect the views of the author(s) and not the opinions of the editors, the editorial board, or the publisher; the editors, the editorial board, and the publisher disclaim any responsibility or liability for such materials. The final responsibility regarding the published content rests with the authors.

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,

• Keywords (For indexing purposes, a list of 4–8 keywords in English is essential),

• Article divided into appropriate sections,

• List of references styled according to "journal requirements",

• A blinded main text (Please exclude all information that may indicate an individual or institution from the main document to ensure a blinded review process),

• The Copyright Agreement and Acknowledgement of Authorship Form (Please submit a wet-signed and scanned copy of the Copyright Transfer Form with your submission),

• Upload your title page and forms in the system to the Potential Conflict of Interest category to ensure a blinded review process,

• 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:

Copyright Agreement and Acknowledgement of Authorship Form

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

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.

6) "Acknowledgements From" should be uploaded separately.

Preparing video content

In order to provide reviewers with a convenient method of accessing video content online, we have restricted video file types to mp4, webM and Ogg format. This allows reviewers to view video content easily from all modern browser types without the inconvenience of downloading plug-ins and video players.

Mp4 is the most common online video format, and there are many converters available that will convert other file types to Mp4.

We can recommend using this free online converter to create a suitable mp4 file.

Video file size is limited to 50 Mbytes, and we suggest reducing file size for quicker upload times using this service Compress Mp4.

Please check Table 1 for the limitations for Video Article.

Case Reports: There is limited space for case reports in the journal and reports on rare cases or conditions that constitute challenges in diagnosis and treatment, those offering new therapies or revealing knowledge not included in the literature, and interesting and educative case reports are accepted for publication. The text should include Introduction, Case Presentation, Discussion, and Conclusion subheadings. Please check Table 1 for the limitations for Case Reports.

Letters to the Editor: This type of manuscript discusses important parts, overlooked aspects, or lacking parts of a previously published article. Articles on subjects within the scope of the journal that might attract the readers' attention, particularly educative cases, may also be submitted in the form of a "Letter to the Editor." Readers can also present their comments on the published manuscripts in the form of a "Letter to the Editor." Abstract, Keywords, Tables, Figures, Images, and other media should not be included. The text should be unstructured. The manuscript that is being commented on must be properly cited within this manuscript.

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Instructions to Authors

Clinical Image: These type of submissions should present a striking image that may challenge and inform readers and contribute to their education. Submissions can include high-quality clinical images, radiology results or surgical images. Please check Table 1 for the limitations for Clinical Images.

Please note that there are author limitations for some article types. Authors should provide a reason for the manuscripts that exceed author limitations. The exception of the articles that are above the author limits is subject to Editorial decision.

Tables

Tables should be included in the main document, presented after the reference list, and they should be numbered consecutively in the order they are referred to within the main text. A descriptive title must be placed above the tables. Abbreviations used in the tables should be defined below the tables by footnotes (even if they are defined within the main text). Tables should be created using the "insert table" command of the word processing software, and they should be arranged clearly to provide easy reading. Data presented in the tables should not be a repetition of the data presented within the main text but should be supporting the main text.

Figures and Figure Legends

Figures, graphics, and photographs should be submitted as separate files (in TIFF or JPEG format) through the submission system. The files should not be embedded in a Word document or the main document. When there are figure subunits, the subunits should not be merged to form a single image. Each subunit should be submitted separately through the submission system. Images should not be labelled (a, b, c, etc.) to indicate figure subunits. Thick and thin arrows, arrowheads, stars, asterisks, and similar marks can be used on the images to support figure legends. Like the rest of the submission, the figures, too, should be blind. Any information within the images that may indicate an individual or institution should be blinded. The minimum resolution of each submitted figure should be 300 DPI. To prevent delays in the evaluation process, all submitted figures should be clear in resolution and large in size (minimum dimensions: 100 × 100 mm). Figure legends should be listed at the end of the main document.

All acronyms and abbreviations used in the manuscript should be defined at first use, both in the abstract and in the main text. The abbreviation should be provided in parentheses following the definition.

When a drug, product, hardware, or software program is mentioned within the main text, product information, including the name of the product, the producer of the product, and city and the country of the company (including the state if in the USA), should be provided in parentheses in the following format: "Discovery St PET/CT scanner (General Electric, Milwaukee, WI, USA)."

All references, tables, and figures should be referred to within the main text, and they should be numbered consecutively in the order they are referred to within the main text.

Limitations, drawbacks, and shortcomings of original articles should be mentioned in the Discussion section before the conclusion paragraph.

References

Both in-text citations and references must be prepared according to the Vancouver style.

While citing publications, preference should be given to the latest, most up-to-date publications. Authors are responsible for the accuracy of references. If an ahead-of-print publication is cited, the DOI number should be provided. Journal titles should be abbreviated in accordance with the journal abbreviations in Index Medicus/ MEDLINE/PubMed. When there are six or fewer authors, all authors should be listed. If there are seven or more authors, the first six authors should be listed, followed by "et al." In the main text of the manuscript, references should be cited using Arabic numbers in parentheses. The reference styles for different types of publications are presented in the following examples.

Turkish Archives of Otorhinolaryngology does not acknowledge citations to preprints since preprints yet have not passed the reviewers' evaluation process and verified by experts in the field.

Journal Article: Erkul E, Cekin İE, Kurt O, Gungor A, Babayigit MA. Evaluation of patients with unilateral endoscopic sinus surgery. Turk Arch Otorhinolaryngol 2012; 50: 41-5.

Book Section: Suh KN, Keystone JS. Malaria and babesiosis. Gorbach

Table 1. Limitations for each manuscript type

Type of manuscript	Author limit	Word limit	Abstract word limit	Reference limit	Table limit	Figure limit
Original Article	N/A	3500	250 (Structured)	30	6	5 or total of 10 images
Review Article	4	5000	250	50	6	10 or total of 15 images
Systematic Review	N/A	5000	250 (Structured)	50	2	10 or total of 15 images
Video Article	N/A	1500	250 (Structured)	106	2	2
Case Report	6	1000	200	10	No tables	4 or total of 8 images
Letter to the Editor	3	500	No abstract	5	No tables	No media
Clinical Images	3	500	No abstract	5	No tables	3 or total of 7 images

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: Bengisson 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.

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Contents

CARE (CAse REport) Guidelines: A Recipe for More Transparent Case Reports

Editorial

Ali Bayram

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Case reports are the types of medical writing that are often described as the scientific documentation of a single clinical observation (1). The first examples of case reports probably date back to the BC era (2). Since then, case reports found themselves a permanent position in the medical literature for the purposes of informing about the findings of new diseases and modes of medical interventions, describing the adverse or beneficial outcomes of a procedure, and have also been used for teaching purposes (3). There are seminal examples of case reports in the medical literature that have antecedently addressed the beneficial or adverse effects of treatment protocols in specific clinical entities such as the relationship between phocomelia and thalidomide, or propranolol treatment for infantile hemangioma (4, 5). Despite, however, the numerous contributions of case reports to the medical literature, controversies have emerged among the scientific community regarding their varying quality and level of evidence, especially in the last 20 years (6).

In medical publishing, utilizing guidelines for any type of study design provides a framework for authors, reviewers, journal editors, and readers when writing, reviewing, or appraising a scientific study and improves the completeness of published scientific reports (7). In the medical literature, various guidelines have been reported for different study types including observational studies [Strengthening the Reporting of Observational studies in Epidemiology, (STROBE)], randomized-controlled trials [Consolidated Standards of Reporting Trials, (CONSORT)], and systematic reviews and meta-analyses [Preferred Reporting Items for Systematic Reviews and Meta-Analyses, (PRISMA)] (8-10). To improve the transparency and completeness of published case reports, an international expert panel

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consisting of a group of clinicians, researchers, and journal editors developed consensus-based guidelines for clinical case reports named the CAse REport (CARE) Statement and Checklist during 2011–2012 (7). In 2013, the CARE Statement and Checklist was presented at the International Congress on Peer Review and Biomedical Publication and simultaneously published in seven journals. Since then, the CARE guidelines have been widely endorsed and translated into multiple languages. The CARE Statement and Checklist is also adopted by the EQUATOR (Enhancing the QUAlity and Transparency of Health Research) Network, which is an umbrella organization composed of researchers, medical journal editors, developers of reporting guidelines, and other stakeholders who aim to improve the reliability and quality of health research publications (11). The CARE guidelines and related documents are available on the CARE website (www.care-statement.org) and the EQUATOR Network (www.equator-network.org). The checklist is composed of 13 items with multiple subitems descriptors which are assumed to be critical for reporting a case report with adequate details. The primary items of the checklist are title, keywords, abstract, introduction, patient information, clinical findings, timeline, diagnostic assessment, therapeutic interventions, follow-up and outcomes, discussion, patient perspective, and informed consent. In 2017, Riley et al. (12) reported an explanation and elaboration document for CARE guidelines that aimed to explain each item with illustrative published examples (the document is available at www.care-statement.org).

Although there were growing concerns among the scientific community against publishing case reports due to their low evidence levels and the citation potential, in my opinion, the CARE Statement and Checklist for case reports promise practical reporting standards to improve the scientific quality of case reports. Moreover, I believe that the wider adoption of the CARE guidelines may assist to accumulate high-quality data from case reports when systematically collected and combined into larger datasets. Therefore, we expect authors to use the CARE guidelines when submitting their case reports to the Turkish Archives of Otorhinolaryngology.

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Olfactory Neuroepithelium in Olfactory Cleft Polyps: Do They Have Any Effect on Olfaction Results After Endoscopic Sinus Surgery?

Original Investigation

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Abstract

Objective: How the presence of olfactory structures in olfactory cleft polyps (OCPs) affect olfaction function outcomes after surgical removal has not yet been investigated. In this study we aimed to assess the presence of olfactory structures in OCPs and correlate these findings with olfactory outcomes after endoscopic sinus surgery (ESS).

Methods: Twenty seven patients with OCP underwent preoperative topical and systemic steroid treatment and ESS. Biopsies from the middle meatal polyps (MMPs) and OCPs were immunohistochemically analyzed for olfactory marker protein (OMP). The smell diskettes olfaction test was applied to patients at baseline, after steroid treatment (AST) and after ESS.

Results: OCPs exhibited OMP staining more commonly and intensely compared to MMPs (p=0.008), however, there were no correlations between OMP staining scores and any of the olfaction scores (p>0.05). Steroid treatment increased smell function significantly (p<0.001), however, there were no significant differences between AST and after ESS smell scores (p=0.17). There were significant correlations between smell gains AST and final smell gains after ESS (r=0.665, p<0.001).

Conclusion: OCPs contain olfactory neuroepithelium more commonly and intensely than MMPs in nasal polyp patients. However, surgical importance of this finding is controversial because removal of these polyps did not decrease smell function postoperatively in our study. Nasal polyp patients who will take steroid treatment pre-operatively must be informed that the success of ESS on olfaction depends on the response of the steroid treatment and ESS AST might not have additional favorable effect on smell function.

Keywords: Chronic rhinosinusitis with nasal polyps, endoscopic sinus surgery, olfaction, olfactory neuroepithelium, olfactory cleft polyp

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Introduction

Chronic rhinosinusitis with nasal polyps (CRS w/NP) is the inflammation of the nasal mucosa and the paranasal sinuses with symptoms of rhinitis and the presence of middle meatal polyps (1). It affects 1–4% of the population and the underlying pathogenesis has not been fully understood. Infections, genetic predisposition, mucociliary dysfunction, vasomotor imbalance and neurogenic inflammation have all been suggested as the possible mechanisms of NP formation (2-4). The symptoms of CRS w/NP include nasal obstruction, headache, rhinorrhea, and olfactory dysfunction (5). Olfactory dysfunction affects 65–80% of the patients with CRS w/NP patients are more commonly affected than CRS patients without NPs.

The etiology of the olfactory problems in CRS w/NP patients is multifactorial. In addition to the conductive olfactory dysfunction by the mass effect of NPs, epithelial erosion, inflammatory infiltration, and cytokine related sensorineural damage of the olfactory epithelium (OE) are also important factors that contribute to the hyposmia/anosmia pathogenesis for CRS w/NP patients (6-8). Surgical removal of middle meatal polyps (MMPs) is the main surgical step during endoscopic sinus surgery (ESS) to relieve the nasal obstruction and restoring the smell deficit. However, removal of olfactory cleft polyps (OCPs) is a surgical controversy as these polyps are located between the nasal septum and the middle/superior turbinate adjacent to the OE and the smell function may be impaired while OCPs are removed during ESS. On the other hand, leaving these polyps in place may continue to block the OE and decrease the chance of olfactory recovery. Recent studies demonstrated that surgical excision of OCPs does not impair and even restore smell function after ESS (9-11). The OE has been demonstrated to be more anteriorly distributed than it was supposed to be before, and middle turbinate may also contain OE (12-14). The relationship between the presence of olfactory neuroepithelium in these polyps and olfactory function results has not yet been properly investigated.

In this study, we aimed to investigate the smell function of CRS w/NP patients with OCPs using the smell diskettes olfaction test pre- and post-operatively. We also aimed to investigate the olfactory structures in both MMPs and OCPs by using immunohistochemical analysis of olfactory marker protein (OMP), which is a marker widely used for detecting OE and olfactory structures in the nasal cavity, and we also correlated the staining results with the smell scores of the patients (6, 13-15).

Methods

Local ethical committee approval (decision no: 2010/350, date: 14.05.2010) and written informed consent from the patients were obtained for the study. The study group consisted of 27 CRS w/NP patients with OCPs. The diagnosis of CRS w/NP was made according to the recent European Position Paper on Rhinosinusitis and Nasal Polyps (1). All patients had presented with symptoms for more than 12 weeks before NPs were endoscopically visualized and diagnosed. Only CRS w/NP patients with OCP were included in the study. OCPs were diagnosed endoscopically and defined as a polyp between the nasal septum and the middle/superior turbinate (the ethmoid turbinate wall) (Figure 1) (10). All patients underwent detailed clinical examination including anterior rhinoscopy, nasal endoscopy with rigid telescopes (0 or 30°) and paranasal sinus computerized tomography. Lund–Mackay staging system was used to assess the extensiveness of the disease (16). Atopy of the patients were evaluated using skin prick test with a standardized allergen Prick test panel (Stallergenes®, Antony, France) (2). To eliminate the effect of allergic rhinitis on the study results, only patients whose skin Prick tests were negative were included. The exclusion criteria also included pediatric patients (age <18 years), patients who had aspirin intolerance, previous nasal surgeries, underwent septoplasty surgery during ESS, used topical or systemic steroid in the previous one month, or had mental and neurological disorders.

All patients received a three-week course of topical (mometasone furoate 0.05%, two sprays per nostril twice a day) steroid therapy. Additionally, systemic steroid therapy was given to all study patients. Oral methylprednisolone was started before the surgery, initially at a dose of 1 mg/kg and gradually tapered by ¼ of the initial dose every fourth day for a period of 16. All endoscopic sinus surgeries were performed by the same surgical team. Briefly, uncinectomy, maxillary antrostomy, total ethmoidectomy, sphenoidotomy, gentle excision of MMPs and OCPs were performed using through cutting forceps and microdebrider. Frontal sinusotomy was not performed in any of the patients. Biopsies were taken from both MMPs and OCPs. Topical steroid therapy was continued for three months after ESS.

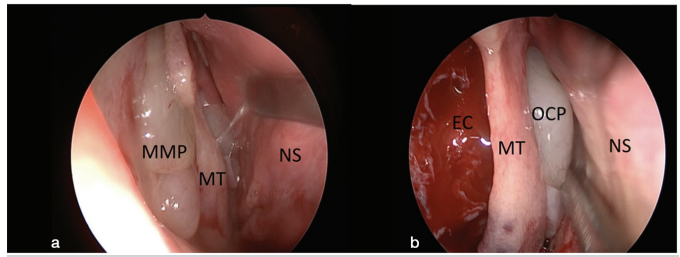

Figure 1. Intraoperative appearances of olfactory cleft polyps and middle meatal polyps were demonstrated: (a) Middle meatal polyp of the right nasal cavity; (b) Olfactory cleft polyp of the right nasal cavity

MMP: Middle meatal polyp, MT: Middle turbinate, OCP: Olfactory cleft polyp, NS: Nasal septum, EC: Ethmoid cavity

Smell Test

Smell diskettes olfaction test (Novimed, Dietikon, Switzerland) was used as the standardized and validated smell identification screening test. As previously described; eight odorants were used in a high suprathreshold concentration for the olfaction test. Patient were scored between 0 (no sense of smelling) and 8 (optimal sense of smelling) according to their olfaction performance (17). The smell test was performed three times for each patient: 1) Pre-treatment: at the initial evaluation of the patient, 2) after steroid treatment (AST), i.e., after topical and systemic steroid treatment before the surgery, and 3) after ESS, i.e., three months after the sinus surgery. To compare the effect of steroid treatment and ESS treatment on olfaction function; olfactory gains were also calculated. Olfactory gain AST was calculated by subtracting initial smell scores from AST smell scores. Final olfactory gain after ESS was calculated by subtracting initial smell scores from after ESS smell scores.

Immunohistochemistry

MMPs and OCPs were fixed in 10% formaldehyde, embedded in paraffin, and cut into 4 µm sections. One section was systematically stained for routine hematoxylin-eosin pathological examination. Sections processed for immunohistochemistry were deparaffinized and blocked with 4% bovine serum phosphate-buffered saline (PBS) for immunohistochemical analysis. Sections were incubated with antibodies against OMP (OMP Antibody, Abcam, ab62144). After 20 minutes of pretreatment, the sections were cooled at room temperature for 20 minutes and washed with distilled water. After another rinse with PBS solution, peroxidase block solution was applied for 10 minutes. Protein block solution and another rinse with PBS solution was done, then the primary antibodies against OMP were applied. Another rinse with PBS solution was done and sections were exposed to diaminobenzidine solution. Finally, counterstaining with hematoxylin was performed and sections were dehydrated in 96% alcohol and covered with Ultramount Labvision balsam for histopathological analysis (13).

All pathological specimens were evaluated by the same pathologist who was unaware of the group of the specimens during histopathological analysis. Briefly, the intensity of immunostaining was analyzed, and staining intensity was scored between 0 (no staining) and 4 (very intense staining) for each patient (18). The OE was regarded as positive control (score 4 staining) for OMP immunostaining. It was obtained from another CRS w/NP patient who was excluded from the study from the dorsoposterior part of the nasal cavity at the upper part of superior turbinate (Figure 2, 3) (6).

Statistical Analysis

Statistical analysis was performed using SPSS version 24.0 (IBM SPSS, New York, USA, 2016). Data were shown as mean ± standard deviation for continuous variables and the number of cases was used for categorical ones. Data were controlled for normal distribution using the Shapiro–Wilk test. Paired sample t-test was used to compare the mean initial, AST and after ESS smell scores of the patients.

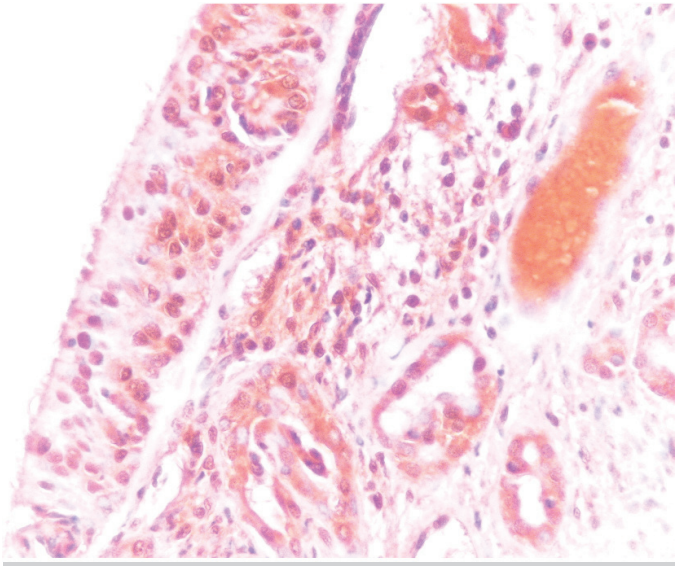

Figure 2. Very intense (+4) immunostaining of olfactory epithelium with olfactory marker protein (x400)

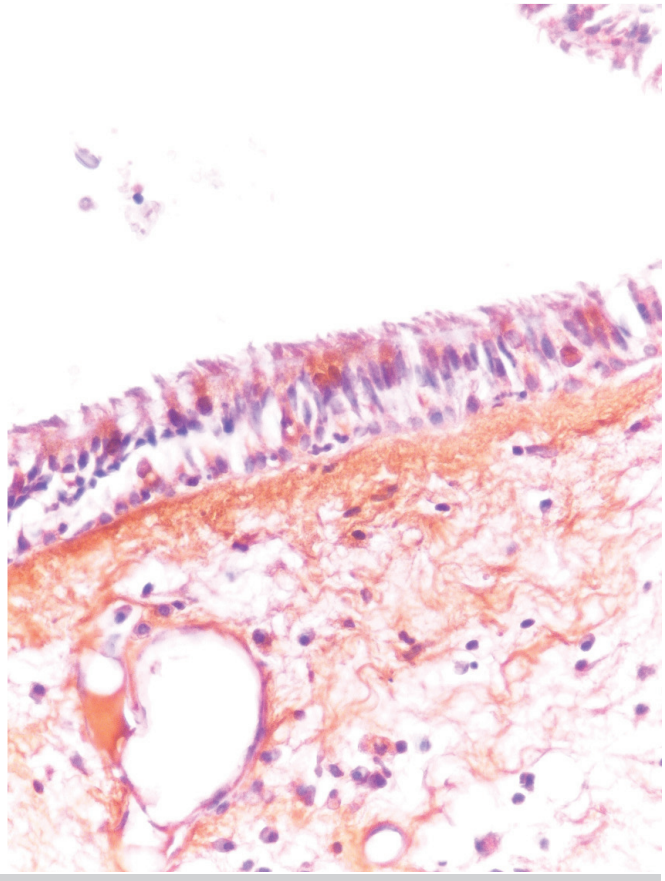

Figure 3. Very intense (+4) immunostaining of olfactory cleft polyp with olfactory marker protein (x400)

The Mann–Whitney U test was used to compare the mean OMP staining scores of OCPs and MMPs. Correlation coefficient was used to evaluate the correlation between smell scores Lund–Mackay scores (LMS) and smell scores-OMP staining scores. Correlation coefficient was also used to assess the correlation between smell gains AST and final smell gains after ESS. P-value of <0.05 was regarded as statistically significant.

Results

Initially 87 patients were diagnosed as CRS w/NP and evaluated according to the inclusion criteria. Thirty-two (36.8%) patients had OCPs. Five patients had atopy according to the skin prick test results and excluded from the study. Finally, 27 patients were included as the study group. There were 16 (59.2%) males and 11 (40.8%) female patients in the study group. Mean age of the patients was 45.58±13.03 years (minimum: 18, maximum: 68).

Histopathological diagnoses of the surgical specimens of the patients were reported as inflammatory NPs, and none of the patients were diagnosed as respiratory epithelial adenomatoid hamartomas (REAHs). Postoperative complications such as cerebrospinal fluid (CSF) leakage or bleeding was not seen in any of the study group patients.

OMP staining was present in 18 (66.6%) of the OCPs and 9 (33.3%) of the MMPs. Mean OMP staining score of OCPs (1.92±1.54) was statistically significantly higher compared to MMPs (0.85±1.29) (p=0.008) (Figure 4).

Mean AST smell scores (4.85±2.14) were statistically significantly higher when compared to the initial smell scores (2.62±3.12) (p<0.001). There was a slight decrease in the smell scores after ESS (mean: 4.44±2.15), but this decrease was not statistically significant (p=0.17). After ESS, smell scores were statistically significantly higher compared to the initial smell scores (p<0.001) (Figure 5).

There were significant correlations between smell gains AST and final smell gains after ESS when compared to the initial smell measurements. Thus, response to the steroid treatment directly correlated with the final smell gains after ESS (r=0.665, p<0.001) (Figure 6).

The mean of the Lund–Mackay scores of the patients was 14.59±4.59. There were no statistically significant

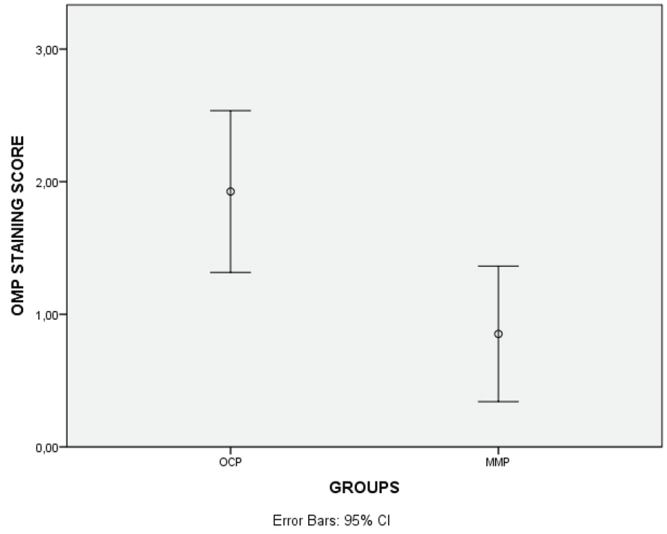

Figure 4. Comparison of mean olfactory marker protein staining scores between olfactory cleft and middle meatus polyps

OMP: Olfactory marker protein, OCP: Olfactory cleft polyp, MMP: Middle meatus polyp, CI: Confidence interval

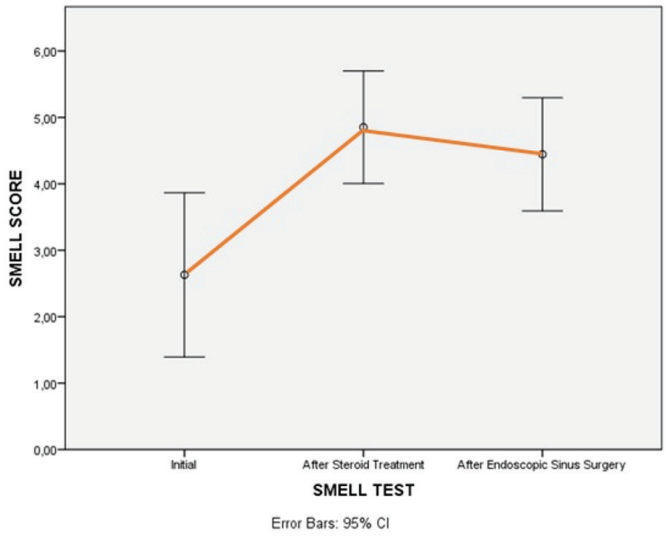

Figure 5. Comparison of mean smell test scores after application of smell diskettes olfaction test was illustrated

CI: Confidence interval

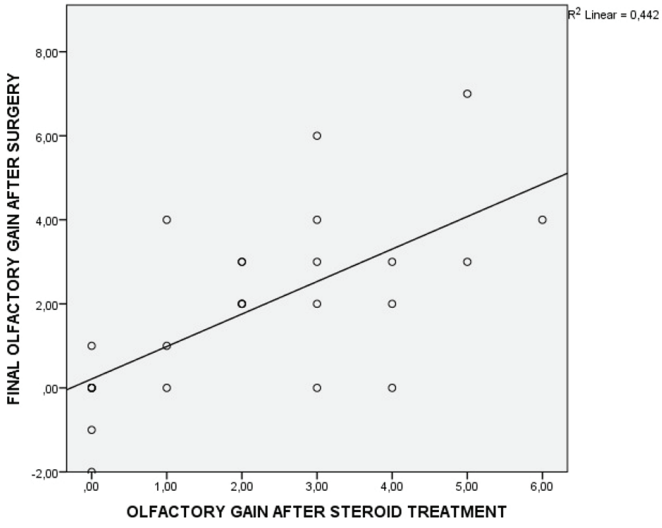

Figure 6. Correlation between olfactory score gains after steroid treatment and final olfactory score gain after endoscopic sinus surgery was demonstrated

correlations between the Lund–Mackay scores and the initial, AST and after ESS smell scores (p=0.585, 0.555 and 0.791, respectively).

There were no statistically significant correlations between the mean OMP staining scores of OCPs and the initial, AST and after ESS smell scores (p=0.519, 0.876 and 0.693, respectively).

Similarly, there were also no statistically significant correlations between the mean OMP staining scores of MMPs and the initial, AST and after ESS smell scores (p=0.76, 0.752 and 0.362, respectively).

Discussion

In this study, we found that steroid treatment significantly increased olfaction scores in CRS w/NP patients. Also, smell gains AST directly correlated with the final smell gains after ESS. Olfactory neuroepithelium in OCPs were observed to be more common and more intense compared to MMPs. However, this finding was not correlated with the preoperative and postoperative smell scores. Lund–Mackay scores of the patients were not related to olfactory outcomes.

The pathogenesis of olfactory dysfunction in patients with CRS w/NP is multifactorial. In addition to the obstructive smell loss by NPs, interleukin (IL) 2,5,6,10,13 related sensorineural loss, IL-5, eotaxin 1 and Charcot–Leiden crystal protein related inflammation by eosinophilic infiltration, inflammation related degeneration and disorganization in the OE were also demonstrated to increase the olfactory dysfunction (6, 8, 19). Thus, the effect of topical and systemic steroids on olfactory dysfunction were evaluated in patients with CRS w/NPs. Topical dexamethasone and fluticasone usage were proven to significantly improve smell function in patients with CRS w/NP (20, 21). Additionally, Bardaranfar et al. (22) reported that triamcinolone embedded gelatin coating application to the OC during ESS increased the smell scores significantly compared to the surgery alone in CRS w/NP patients. Decrease in smell function was demonstrated to be the most common improved symptom for CRS w/NP patients after systemic steroid treatment (23). Recently, two studies showed that the success of surgery on olfactory function was related to the response of the steroid treatment for CRS w/NP patients (24, 25). According to the results of Bogdanov et al. (24), ESS had no additional benefit on smell scores after steroid therapy. Similarly, in our study, significant smell gain was established after topical and systemic steroid treatment, however, surgery had no additional effect on the smell scores AST. Additionally, response to the steroid treatment directly correlated with the final smell gains after ESS in our study. Basing on our study results and the previous literature, it can be suggested that topical and systemic steroid treatment is effective in the recovery of the olfactory dysfunction for patients with CRS w/NP. Surgical removal of the polyps may overwhelm conductive smell loss, however, for the treatment of the OE inflammation, topical and/or systemic steroid treatment seems to be more useful for the recovery of smell function. CRS w/NP patients who take steroid therapy before surgery should be informed that ESS might not provide any additional benefit on the smell function compared to steroid treatment. Similarly, our findings suggested that the final olfactory outcome after ESS should be related to the response of the patient to the steroid treatment.

The OC region is surrounded laterally by the attachment of the middle and/or superior turbinate, medially by the superior part of the nasal septum, and posteriorly by the anterior face of the sphenoid sinus. The roof is formed by the cribriform plate and the floor is an imaginary line drawn one centimeter below the cribriform plate (26). OCPs and REAHs are among the most common benign lesions that block the OC region, impair airflow to the OE, and thereby diminish the olfactory function (10). Surgical procedures to remove these lesions in this critical area may result in major complications such as CSF leakage and permanent olfactory loss due to the surgical trauma (9). In fact, few contemporary studies have focused on the effect of surgical excision of OCPs on the olfactory function outcomes (9-11). Particularly, they have shown that the surgical removal of OCPs without preoperative steroid treatment did not impair and even improved olfactory function. Similarly, we also determined a significant final olfactory gain after excision of OCPs when compared to the initial smell scores in our study. Additionally, we encountered a statistically insignificant olfactory decrease after the excision of OCPs compared to steroid treatment. According to our study results, we might say that topical and systemic steroid treatment has a major effect on olfactory function improvement in patients with OCPs and surgical excision of these polyps AST does not impair olfactory function. The main difference of our study from the aforementioned studies is that we determined the olfactory structures in OCPs and compared these findings with the olfactory function (9-11). We demonstrated that OCPs contain olfactory structures more commonly and intensely as compared to the MMPs. However, surgery of these polyps did not deteriorate smell function. Bhutta et al. (15) showed that none of the polyps originating from the superior turbinate region contained olfactory structures immunohistochemically. Its difference from our study may be explained by the anti-inflammatory effect of steroid treatment on the OCPs. Steroid treatment may have decreased the polyp size and may have increased the chance of OE sampling intraoperatively in our study. Additionally, both light microscopy and electron microscopy studies demonstrated that there is no clear boundary of OE and respiratory epithelium. OE is distributed among the respiratory epithelium near the cribriform plate region in a patchy manner (12). These gaps among the OE may have caused false negative sampling of olfactory structures

in the study of Bhutta et al. (15). Also, Sasaki and Nakahara (27) demonstrated that NPs might contain nerve fibers and respond to neurogenic stimulation. Moreover, it was proven that even the middle turbinate contained olfactory structures in most of the concha bullosa patients (13-14). Hence, based on our study findings, both OCPs and MMPs may contain olfactory neuroepithelium. However, the surgical importance of this issue is controversial since removal of these polyps did not impair smell function in our study.

The correlation between olfaction and extensiveness of the disease on CT findings in CRS w/NP patients was investigated in various studies. According to the results of our study and the previous literature postoperative smell function was not correlated with the preoperative LMS values (11, 28, 29). However, some studies reported a negative correlation between LMS and preoperative olfactory function (29, 30). LMS values were not correlated with the preoperative olfaction test results in our study and in the study of Paksoy et al. (28).

Limitations of this study were firstly, the small sample size of the group. Secondly, we did not have a control group of patients without preoperative systemic steroid treatment or without removal of the OCPs to compare the results of different ESS techniques and steroid treatment on the olfactory function results. Lastly, we did not classify NPs according to the genotyping results of the histopathological specimens. Hence, we could not analyze the difference of the histological types of the NPs regarding the presence and intensity of the OE. Despite the limitations, we believe that our study exhibits meaningful results regarding the relationship between olfactory structures in the OCPs and olfactory function outcomes after ESS.

Conclusion

In conclusion, OCPs contain olfactory neuroepithelium more commonly and intensely than MMPs for CRS w/NP patients. However, surgical importance of this finding may be controversial because removal of these polyps did not decrease smell function postoperatively in our study. Future studies with larger patient groups should also investigate the impact of the presence of olfactory structures on postoperative smell function results in patients with OCPs. CRS w/NP patients who will take topical and systemic steroid treatment pre-operatively must be informed that the success of ESS on olfaction function depends on the response of the steroid treatment, and ESS AST might not have additional favorable effect on smell function.

Ethics Committee Approval: Mersin University Clinical Researches Ethics Committee approval was acquired for the current study (decision no: 2010/350, date: 14.05.2010).

Informed Consent: Written informed consent was taken from all patients who participated in the study.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: C.Ö., O.İ., F.M., Y.V., K.G., Concept: C.Ö., O.İ., F.M., İ.G., Y.V., K.G., Design: C.Ö., O.İ., F.M., İ.G., Y.V., K.G., Data Collection and/or Processing: C.Ö., O.İ., F.M., İ.G., Y.V., K.G., Analysis and/or Interpretation: C.Ö., O.İ., F.M., İ.G., Y.V., K.G., Literature Search: C.Ö., O.İ., F.M., İ.G., Y.V., K.G., Writing: C.Ö., O.İ., F.M., İ.G., Y.V., K.G.

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

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Main Points

- Olfactory cleft polyps (OCPs) contain olfactory neuroepithelium more commonly and intensely than middle meatal polyps (MMPs) in chronic rhinosinusitis with nasal polyp (CRS w/NP) patients.
- However, surgical removal of OCPs during endoscopic sinus surgery (ESS) does not decrease olfaction scores postoperatively, despite the existence of olfactory neuroepithelium in OCPs.
- The success of endoscopic sinus surgery on olfaction function depends on the response of the steroid treatment, and ESS after steroid treatment might not have additional favorable effect on smell function in CRS w/NP patients.

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Comparison of Treatment Outcomes Between Total and Near Total Glossectomy with Larynx Preservation and Flap Reconstruction: A Single Institution Experience with 23 Patients

Original Investigation

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Abstract

Objective: The purpose of this study was to evaluate the functional and oncological outcomes of total glossectomy with laryngeal preservation (TGLP) compared to near-total glossectomy with laryngeal preservation (nTGLP).

Methods: In this retrospective study, the data of 23 patients who underwent either TGLP or nTGLP between January 2010 and December 2020 in a tertiary university hospital were analyzed. The data including demographic findings, tumor stage, extent of surgery and technique, method of reconstruction, complications, overall survival (OS) and recurrence-free survival (RFS), and tracheostomy and gastric tube dependence during follow-up were assessed.

Results: Of the 23 eligible patients, 15 had undergone nTGLP (Group 1) and 8 had undergone TGLP (Group 2). Tracheostomy dependence and gastric tube dependence rates at the sixth month were 3/19 (15.7%) and 12/19 (63.1%), respectively, with no significant differences between the two groups. OS and RFS at one year were 47.6% and 27.8%, respectively, for the study population. OS rate showed significance with nodal positivity and extranodal extension (p=0.004 for both) only, but not within patient groups (p=0.734).

Conclusion: Both TGLP and nTGLP are feasible treatment options in patients with advanced tongue cancer with no differences in terms of functional and oncological results. Survival rates are still not satisfactory, and recurrences are high despite appropriate treatments. Proper selection of patients who are highly motivated and willing for long-term postoperative rehabilitation is essential.

Keywords: Cancer of tongue, glossectomy, laryngectomy, surgery, survival, tracheostomy

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Introduction

Total glossectomy (TG) or near-total glossectomy (nTG) and postoperative radiotherapy (RT) or chemoradiotherapy (CRT) are widely accepted as the treatments of choice in patients with massive infiltrating malignant tongue lesions (1-4). However, some drawbacks still remain for the extensive resection of the oral tongue, such as high morbidity, poor oral intake, aspiration problems, challenges in reconstruction, low speech intelligibility, tracheostomy dependence and overall reduced quality of life despite improved survival. Since the extent of the surgery involves either the whole or half of the tongue base, there is a high risk of aspiration in these patients. Total laryngectomy (TL) combined with TG is suggested in conventional teaching to overcome aspiration problems. With the emergence of successful reconstruction techniques with flaps, some studies have reported promising functional results by preserving the larynx (1, 5-7). They suggested that total glossectomy with laryngeal preservation (TGLP) provided better speech intelligibility and maintained swallowing ability (6-11).

Another issue is the high mortality rates, hence unsatisfactory survival rates, either because of locoregional recurrence or distant metastasis, despite radical surgical treatment combined with RT or CRT protocols. The oncologic outcomes and survival rates were found to be similar with or without TL combined with TG but were still low (12). To achieve better functional results, preservation of one functional unit of the tongue base, namely nTG (Type IVb) may be considered (13). However, some concerns such as surgical safety margin and recurrence-free survival can arise with Type IVb glossectomy instead of TG (Type V).

In this study, we aimed to assess and compare the functional and survival outcomes in patients who underwent TGLP or near-total glossectomy with laryngeal preservation (nTGLP).

Methods

This retrospective study was planned, and ethics approval was granted by the local ethics committee (Approval no. 2021-1/28). The study was conducted at the department of otolaryngology and head & neck surgery of a tertiary university hospital with patients who underwent TGLP or nTGLP with simultaneous bilateral neck dissection. Clinicopathological data of 27 patients diagnosed with an advanced stage malignant tongue lesion and who had TG or nTG between January 2010 and December 2020 were reviewed. Among these patients, four who had TL combined with primary surgery due to tumor extension beyond the base of the tongue and with distant metastasis were excluded. Thus, the data of 23 eligible patients included in the study population were retrieved from patient files. Data on demographic characteristics, pathological tumor stage, extent and technique of surgery, method of reconstruction, adjuvant treatment, complications, follow-up, survival, tracheostomy status and oral intake were recorded. All patients were evaluated by complete otolaryngological examination with endoscope, preoperative computed tomography (CT) and magnetic resonance imaging (MRI) of the primary site and neck, and positron emission tomography/CT for the presence of any distant metastasis.

In all cases, we performed surgery using lip-splitting median mandibulotomy approach combined with apron flap incision. The extent of dissection was determined according to accurate evaluation and analysis of preoperative CT and MRI images. We removed all anatomical compartments of the tongue and the floor of the mouth involved with tumor according to the imaging and clinical findings intraoperatively. A nTG (Type IVb) was used if the lesion exceeded the border of the hemilingual area of origin infiltrating the ipsilateral base of the tongue and the contralateral genioglossus muscle (13). Thus, one functional unit of the tongue base on one side could be spared (Figure 1). In cases where massive infiltrating tongue lesions bilaterally involved extrinsic genioglossus, hyoglossus, styloglossus and tongue base with impairment of the mobility of the tongue, then TG (Type V) was preferred (13). This involved removal of the entire tongue with the intrinsic and extrinsic muscles and the floor of the mouth above the hyoid bone (Figure 2). Simultaneous bilateral modified radical neck dissection, including levels from I to V, was performed in all cases. The reconstruction of the defect was performed immediately after tumor removal and frozen section examination confirmed negative tumor margins. Oral cavity reconstruction was performed by either free or pedicled myocutaneous flap. Tracheostomy cannula and nasogastric tube were placed in all patients.

Surgical margin positivity was accepted as the tumor being adjacent to the surgical margin or closer than five mm in histopathological examinations. Postoperative follow-up for functional outcomes was carried out by flexible endoscopic examination for both swallowing and airway edema. Patients who had satisfactory swallowing function without aspiration began oral feeding. Tracheostomy tube decannulation was performed when airway edema resolved, and flaps were secured in place. Functional assessment of oral intake and airway was made at the 6th month of surgery (9). Oncological outcomes such as overall survival (OS), disease-free survival (DFS) and local disease control were also recorded during the follow-up, and all prognostic variables were compared between the TGLP and nTGLP groups.

Statistical Analysis

The findings of the presented study were analyzed with the SPSS 23.0 software package (IBM SPSS® Statistics 23.0, Armonk, N.Y., USA). The results were analyzed with 95%

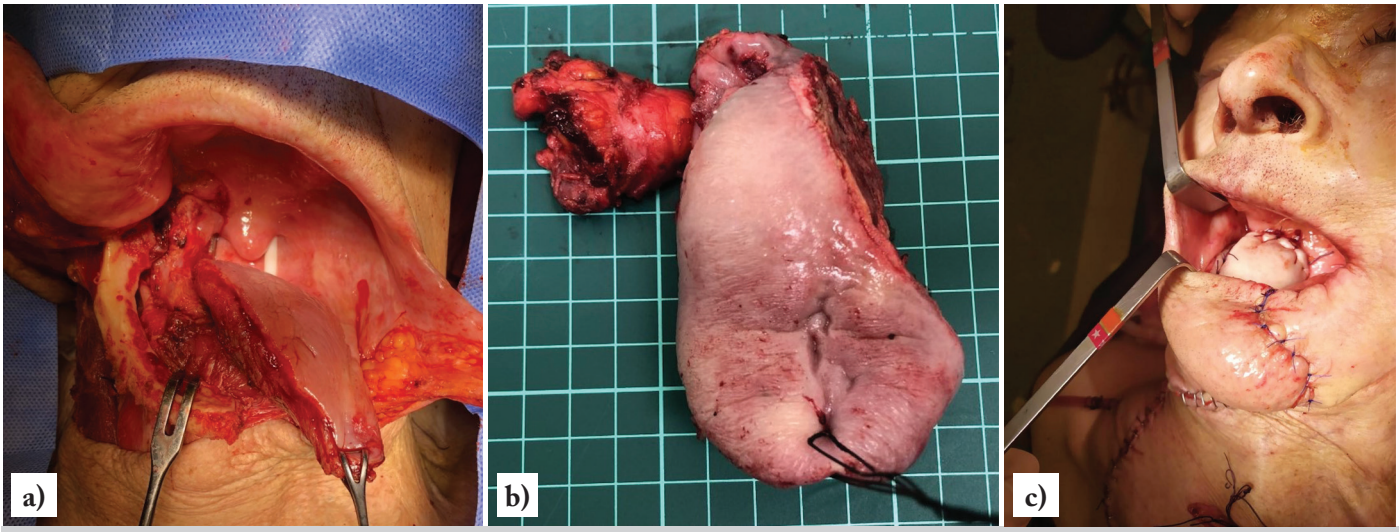

Figure 1. (a) Intraoperative; (b) specimen; and (c) postoperative photographs of a patient who underwent Type IVb glossectomy

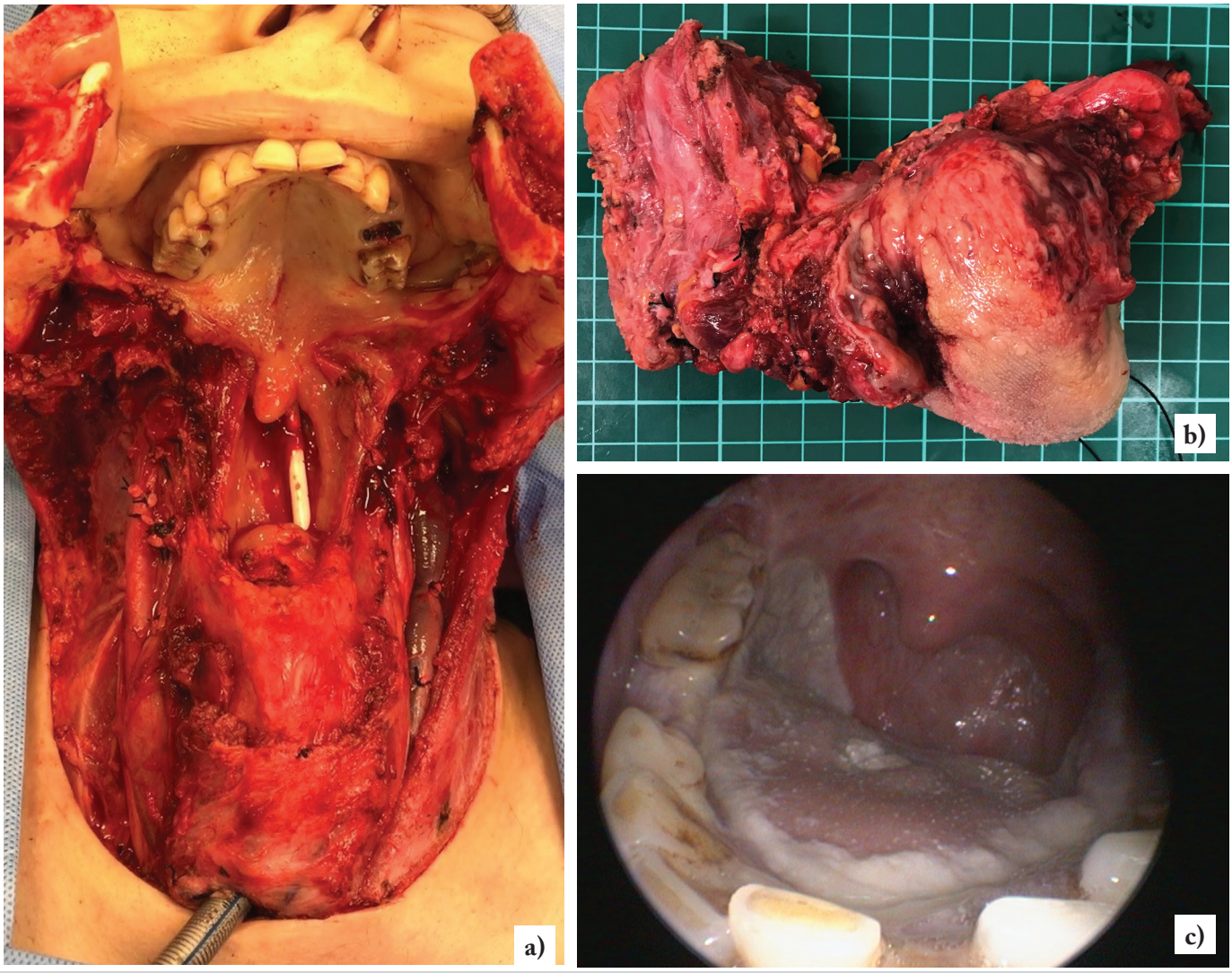

Figure 2. (a) Intraoperative; (b) specimen; and (c) postoperative endoscopic photographs of a patient who underwent Type V glossectomy

confidence intervals, and $p<0.05$ was considered statistically significant. Chi-square test was used to determine the predictors of categorical variables listed in Table 1. The Mann–Whitney U test was used to analyze continuous variables. OS and DFS rates were estimated, and survival distributions were analyzed with the Kaplan–Meier analysis with log-rank test.

Results

Patients' Profiles and Surgical Characteristics

The study population included 16 men and 7 women with a mean age at diagnosis of 52.87 years (range: 28 to 77). Eight patients underwent salvage surgery due to recurrence of oral cavity cancer following primary surgical treatment. The distribution of 23 patients regarding the operation technique was nTGLP in 15 patients (Group 1) and TGLP in 8 patients (Group 2). Squamous cell carcinoma was the histopathological diagnosis in all cases. Primary tumor site was the oral tongue in 16, the base of tongue in one and the floor of mouth in six cases. Segmental (n=4) or marginal mandibulectomy (n=5) was required in nine patients, and in four patients, resection included supraglottic structures; two had partial epiglottectomy, one had epiglottectomy with hyoid bone resection and another had supraglottic laryngectomy. We used pectoralis major myocutaneous flap in 15, radial forearm free flap in two, anterolateral thigh free flap in five and deltopectoral flap in one patient for defect reconstruction. Reexploration and reanastomosis were performed in one patient due to flap compromise. Revision reconstructive surgery was performed in four patients due to flap failure. Mandibular reconstruction plate was exposed in two patients, and orocutaneous fistula, which was treated conservatively, occurred in six patients. All patients underwent bilateral neck dissection except two who had neck dissection in their previous surgery with no sign of metastatic lymph node in imaging studies (Table 1).

Results of Histopathological Examinations

T stage was reported as T4 in all patients according to the 8th edition of the AJCC. Metastatic lymph nodes were found in 16 (76%, 16/21) patients, with extracapsular invasion in 13 patients. Nodal stages of the study population were as follows: N0 (n=5), N1 (n=2), N2b (n=3), N2c (n=5), N3 (n=4) and N3b (n=2). Surgical margin positivity in permanent pathologic examination was reported in three cases. Mean tumor dimension was found 59.30 mm (15-150 mm), while mean depth of invasion was 28.09 mm (8-50 mm). There was mandibular invasion in three patients and hyoid bone invasion in one patient. All patients were diagnosed with squamous cell carcinoma and tumors were well-/moderately differentiated (n=17) and poorly differentiated (n=6), respectively (Table 1).

Table 1. Clinical, surgical, and histopathological characteristics of patients in general and with respect to the type of glossectomy

	Total (n=23)	Group 1 (nTGLP) (n=15)	Group 2 (TGLP) (n=8)	p-value
Age (years)				
Min-max	28–77	28–77	30–58	0.047*
Mean	52.87	57.33	43.00	
Median	55	57	43	
SD	14.763	14.509	11.880	
Gender				
Female	7 (30.4%)	3 (20.0%)	4 (50.0%)	0.182**
Male	16 (69.6%)	12 (80.0%)	4 (50.0%)	
Follow-up time (month)				
Min-max	3–108	3–46	3–108	0.506*
Mean	21.48	16.53	30.75	
Median	11	11	8.5	
SD	27.799	12.755	44.190	
Primary/salvage surgery				
Primary	15 (65.2%)	10 (66.7%)	5 (62.5%)	1.000**
Salvage	8 (34.8%)	5 (33.3%)	3 (37.5%)	
Tumor localization				
Tongue	16 (69.6%)	11 (73.3%)	5 (62.5%)	0.443***
Floor of mouth	6 (26.1%)	4 (26.7%)	2 (25.0%)	
Tongue base	1 (4.3%)	0 (0.0%)	1 (12.5%)	
Supraglottic laryngectomy				
No	19 (82.6%)	14 (93.3%)	5 (62.5%)	0.103**
Yes	4 (17.4%)	1 (6.7%)	3 (37.5%)	
Mandibulectomy				
No	14 (60.9%)	8 (53.3%)	6 (75.0%)	0.396***
Marginal	5 (21.7%)	3 (20.0%)	2 (25.0%)	
Segmental	4 (17.4%)	4 (26.7%)	0 (00.0%)	
Neck dissection				
No	2 (8.7%)	1 (6.7%)	1 (12.5%)	1.000**
Yes	21 (91.3%)	14 (93.3%)	7 (87.5%)	
Tumor size				
Min-max	15–150	15–70	50–150	0.000*
Mean	59.30	44.13	87.75	
Median	52	45	80	
SD	30.598	15.090	32.697	
Depth of invasion				
Min-max	8–50	8–42	20–50	0.008*
Mean	28.09	23.07	37.50	
Median	26	20	40	
SD	11.874	9.316	10.690	
Differentiation				
Well-moderately	17 (73.9%)	11 (73.3%)	6 (75.0%)	1.000**
Poorly	6 (26.1%)	4 (26.7%)	2 (25.0%)	

Lymphovascular invasion				
No	19 (82.6%)	13 (86.7%)	6 (75.0%)	0.589**
Yes	4 (17.4%)	2 (13.3%)	2 (25.0%)	
Perineural invasion				
No	5 (21.7%)	3 (20.0%)	2 (25.0%)	1.000**
Yes	18 (78.3%)	12 (80.0%)	6 (75.0%)	
Bone invasion				
No	19 (82.6%)	13 (86.7%)	6 (75.0%)	0.589**
Yes	4 (17.4%)	2 (13.3%)	2 (25.0%)	
Lymph node metastases				
No	5 (23.8%)	3 (21.4%)	2 (28.6%)	1.000**
Yes	16 (76.2%)	11 (78.60%)	5 (71.4%)	
Extranodal extension				
No	8 (38.1%)	6 (42.9%)	2 (28.6%)	0.656**
Yes	13 (61.9%)	8 (57.1%)	5 (71.4%)	

*Mann–Whitney U test, **Fisher's Exact test, ***Fisher's Exact test (Monte Carlo)
SD: Standard deviation, min: Minimum, max: Maximum

Table 2. Functional and oncological results of Group 1 and Group 2

	Total (n=23)	Group 1 (n=15)	Group 2 (n=8)	p-value
Tracheostomy dependency				
No	16/19 (84.2%)	10/13 (76.9%)	6/6 (100%)	0.517*
Yes	3/19 (15.8%)	3/13 (23.1%)	0/6 (00.0%)	
Tube dependency feeding				
No	7/19 (36.8%)	4/13 (30.8%)	3/6 (50.0%)	0.617*
Yes	12/19 (63.2%)	9/13 (69.2%)	3/6 (50.0%)	
Recurrence				
No	9/23 (39.1%)	4/15 (26.7%)	5/8 (62.5%)	0.179*
Yes	14/23 (60.9%)	11/15 (73.3%)	3/8 (37.5%)	
Local-regional recurrence				
No	14/23 (60.9%)	8/15 (53.3%)	6/8 (75.0%)	0.400*
Yes	9/23 (39.1%)	7/15 (46.7%)	2/8 (25.0%)	
Mortality				
No	5/23 (21.7%)	2/15 (13.3%)	3/8 (37.5%)	0.327*
Disease free mortality	4/23 (17.4%)	2/15 (13.3%)	2/8 (25.0%)	
Disease specific mortality	14/23 (60.9%)	11/15 (73.3%)	3/8 (37.5%)	
Survival rate				
1 year overall survival rate	10/21 (47.6%)	7/14 (50.0%)	3/7 (42.9%)	1.000*
1 year recurrence free survival rate	5/18 (27.8%)	2/12 (16.7%)	3/6 (50.0%)	

*Chi-square test

Follow-up, Oncological and Functional Outcomes

One patient died due from early postoperative complications after the third month of operation. Mean follow-up duration was 21.5 months (range: 3 to 108) and for patients still alive was 51.8 months. All patients, except one, had adjuvant RT, chemotherapy, or CRT. There were locoregional recurrences in nine patients during follow-up and another five patients were diagnosed with metastatic lung cancer. Functional outcomes were assessed at the sixth month after surgery and three (15.7%) patients among the 19 who survived more than six months were tracheostomy dependent. An adequate swallowing function and good/acceptable oral intake were obtained in seven patients. OS rate was 50.0% for Type IVb and 42.9% for Type V glossectomy at one year. Disease-specific survival rates were 16.7% and 50.0% for Type IVb and Type V at one year, respectively. The comparison of functional outcomes and mortality rates between TGLP and nTGLP groups did not reveal any difference (Table 2). The extent of surgery and other clinical variables did not show any relation with survival rates, but we found significance for nodal positivity and extranodal extension with survival in the results of Kaplan–Meier analysis (Tables 3, 4, and Figures 3, 4).

Discussion

In patients with locally advanced tongue cancer, total or near nTG is sometimes the only possible treatment option. Unfortunately, the survival rate of patients with advanced stage tongue cancer is still very low despite extended surgical resection with adjuvant RT or CRT treatment. The adverse consequences of this mutilating surgery, such as chronic aspiration, poor speech, airway problems and social isolation, can be viewed negatively by many patients. However, the recent development of reconstruction techniques with flaps and laryngeal preservation can provide good functional outcomes. While considering the safe margins of resection oncologically, we should also consider maximum preservation of tumor-free tissues. Thus, our institutional approach was mainly targeted to sparing at least one functional unit of the tongue base if possible. In the last decade, we performed TGLP in eight cases and were able to preserve one half of the tongue base in the other 15 cases.

The tumor size and spread mainly determine the extent of resection in the oral cavity. In our study population, as would be expected, we performed TG rather than nTG in significantly larger tumor sizes and invasion depths. However, we had some concerns, because as the extent of resection increases, oral tongue functions deteriorate inevitably. Mazarro et al. (1) conducted a study with 12 patients who had undergone TGLP and reported that five of seven (71%) patients alive at one year were PEG dependent due to chronic aspiration. The study also revealed that in all seven patients, tracheostomy cannula was removed. Navach et al. (10) suggested that with proper postoperative feeding rehabilitation, 70% of these

Table 3. Results of overall survival analysis

	Total (n)	Death (n)	Death (%)	Survival mean (months)	Standard error	CI %95 lower	CI %95 upper	Log-rank test*	p-value
Glossectomy type									
Type IVb	15	13	86.7%	17.933	3.764	10.555	25.311	0.116	0.734
Type V	8	5	62.5%	36.429	17.133	2.848	70.010		
Overall	23	18	78.3%	26.602	7.596	11.714	41.490		
Lymph node metastases									
No	5	2	40.0%	72.000	18.344	36.046	107.954	8.490	0.004
Yes	16	14	87.5%	12.710	2.408	7.991	17.429		
Overall	21	16	76.2%	28.572	8.230	12.442	44.702		
Extranodal extension									
No	8	4	50.0%	57.925	16.757	25.082	90.768		0.004
Yes	13	12	92.3%	11.333	1.698	8.005	14.662	8.300	
Overall	21	16	76.2%	28.572	8.230	12.442	44.702		

*Chi-square value for Log-rank test, CI: Confidence interval

Table 4. Results of recurrence free survival analysis

	Total (n)	Recurrence (n)	Recurrence (%)	Survival mean (months)	Standard error	CI %95 lower	CI %95 upper	Log-rank test*	p-value
Glossectomy type									
Type IVb	15	11	73.7%	16.806	4.903	7.196	26.415	1.215	0.270
Type V	8	3	37.5%	66.175	18.971	28.991	103.359		
Overall	23	14	60.9%	36.073	10.980	14.553	57.593		
Lymph node metastases									
No	5	2	40.0%	71.333	19.552	33.012	109.655	4.961	0.026
Yes	16	11	68.7%	13.006	4.055	5.057	20.954		
Overall	21	13	61.9%	35.893	11.163	14.014	57.772		
Extranodal extension									
No	8	3	37.5%	64.190	18.313	28.296	100.085	6.533	0.011
Yes	13	10	76.9%	6.103	0.977	4.187	8.018		
Overall	21	13	61.9%	35.893	11.163	14.014	57.772		

*Chi-square value for log-rank test, CI: Confidence interval

patients could return to oral feeding. In the study by Keski-Säntti et al. (2), the authors evaluated the functional outcomes of 15 TGLP and 14 nTGLP patients at one year after surgery. They found that gastrostomy and tracheostomy tube dependence in all patients was 77% and 15%, respectively. However, the authors did not compare the outcomes between these two groups. In another study, Barry et al. (4) compared the oral intake outcomes of patients who had TGLP as first-line or salvage therapy and noted that the ability to eat soft food was 23/49 (47%) in the first-line group vs 25/60 (42%) in the salvage group with no significant difference. On the contrary, Rihani et al. (5) pointed out that sufficient oral intake without the need for a gastric tube was higher for primary surgery followed by postoperative RT compared with primary RT followed by surgical salvage: 56% vs 11% respectively. Overall, an important number of patients, 27/94 (29%) required prolonged gastrostomy tube in their study (5). Dziegielewski et al. (6) reported the gastrostomy tube dependence rate as 24% in a systematic review of the related literature. In another literature review, they concluded that gastric tube dependency after TGLP ranged from 30% to 44% and that tracheostomy tube removal was between 85% to 95% (11).

Some authors reported that even performing synchronous TL with TG does not prevent the risk of placement of a gastrostomy tube. Han et al. (7) found that 60% of patients who had total laryngoglossectomy (TGL) still required gastrostomy tube placement, but that this rate was 93% in those who had TGLP. They also reported that the eventual decannulation rate was 61%. Similarly, in another study, there was no difference (p>0.99) between feeding tube dependence rates in TGL and TGLP groups: 8/17 (47%) and 17/37

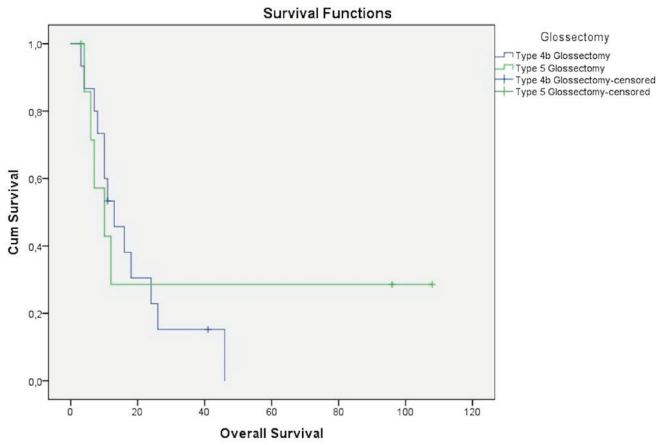

Figure 3. Kaplan–Meier curves for overall survival of groups regarding to glossectomy types. No significant difference was observed between the groups (p=0.734)

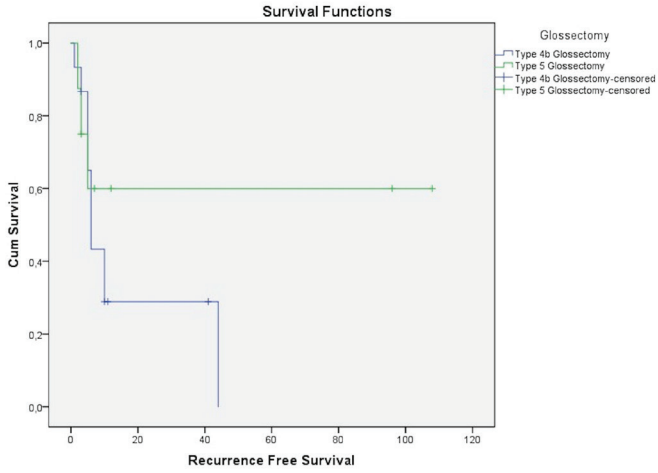

Figure 4. Kaplan–Meier curves for disease specific survival of groups regarding to glossectomy types. No significant difference was observed between the groups (p=0.270)

(45.9%), respectively (8). Sinclair et al. (12) revealed similar results in that although tube dependency (70%) was more common in TGLP patients, 30% of TGL patients were also tube dependent. In addition, 50% of TGLP cases were tracheostomy dependent too. The findings of our study are relevant to the previous literature in that about 63% of our patients were tube dependent at the 6th month of surgery but that this rate decreased to 50% at the end of one year follow-up. The tracheostomy removal rate was found to be 84%, which is similar to the literature.

To the best of our knowledge, the presented study is the first to evaluate and compare the functional results between TGLP and nTGLP in the literature. We concluded that there was no difference regarding adequate oral intake and tracheostomy removal rates between Type IVb and Type V glossectomy. None of our patients in the TGLP group was tracheostomy dependent compared to nTGLP (23.1%), but this did not reach significance. The rates of tube dependence were also similar for TGLP vs nTGLP: 50% vs. 69.2%, respectively. Based on our findings, we suggest that preserving one functional unit of the tongue base at one side does not have any additional functional benefit compared to total removal.

Survival rates and clinical variables that may have an impact on DFS are other important issues to consider in the management of advanced tongue cancers. Despite the appropriate treatment, the life expectancy is unfortunately still low in patients who have undergone TG. Navach et al. (10) found the actual five-year OS and DFS to be 54% and 47% respectively, and they reported that these rates decreased to 21% and 23% for patients who had TG as their second treatment. In another study, positive margins and rate of early recurrence were measured to be higher in patients who had preoperative RT (3). On the contrary, the study by Han et al. (7) reported no difference (p=0.45) regarding OS between primary treatment and salvage therapy, and one-year OS was 42% in the study population. In a study by Reiter and Harréus (9), three-year survival was found to be 57.1% but local disease control for patients with free resection margins reached 83.3% (10/12). Barry et al. (4) evaluated the prognostic risk factors in 109 patients who had undergone either first-line (n=49) or salvage (n=60) TGLP. They pointed out that shorter survival was significantly related to positive margins of resection (p=0.002) and tumor spread into the mandible (p=0.04), but not salvage surgery (p=0.09). Even so, 3-year and 5-year survival rates were higher in the first-line group compared to salvage surgery: 43% and 23% vs. 23% and 16%, respectively (4). However, tumor-free resection margins, pN class, extranodal extension and postoperative RT vs CRT did not reveal a significant relationship with OS in the study by Keski-Säntti et al. (2). An interesting finding was reported by Mazarro et al. (1), that one-year OS in patients ≥50 years old was higher than in those <50 years old: 71.4% vs 40% with an overall OS of 58.3%.

In our study, the OS for one year was similar to the previous literature as 47.6%. In the analysis of our data, we found a significant relation between survival and positive lymph node and extranodal extension but not with other clinical variables such as primary vs salvage, tumor size, depth of invasion, bone invasion, differentiation, margin positivity, and lymphovascular and perineural invasion. It is known that the status of regional lymph nodes is an important prognostic factor for locoregional tumor control, and that extranodal extension is a major cause of death affecting survival. In addition, there was no significant survival difference between the TGLP and nTGLP groups, although Type V glossectomy showed better results at one year: 57.1% vs 50.0%.

Conclusion

In conclusion, although this retrospective study included a limited number of patients, we suggest that both TGLP (Type V) and nTGLP (Type IVb) are feasible treatment options in patients with advanced tongue cancer. However, survival rates are not satisfactory and locoregional and distant recurrences are still high despite appropriate surgical and adjuvant treatments. Thus, proper selection of patients who are highly motivated and willing for long-term postoperative rehabilitation and follow-up is essential. This is the first study to demonstrate that TGLP provides similar oncological and functional outcomes to nTGLP, despite the presence of higher tumor volume and extensive tissue removal.

Ethics Committee Approval: This retrospective study was planned and ethics approval was granted by the local ethics committee of Bursa Uludağ University (approval no. 2021-1/28).

Informed Consent: Informed consent was obtained from the patients/caregivers before the study was initiated.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: U.L.D., M.A., H.C.İ., Concept: U.L.D., M.A., H.C.İ., Design: U.L.D., M.A., Data Collection and/or Processing: M.A., H.C.İ., Analysis and/or Interpretation: U.L.D., M.A., Literature Search: U.L.D., M.A., H.C.İ., Writing: U.L.D.

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

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Main Points

- Survival rates are still low due to locoregional recurrence and metastasis in advanced stage tongue cancers. In our study, overall and recurrence-free one-year survival rates were 47.6% and 27.8%, respectively, in patients who underwent total or near total glossectomy with laryngeal preservation due to advanced stage tongue cancer.
- Statistical analysis showed that nodal positivity and extranodal extension were the only factors negatively affecting overall survival and recurrence-free survival.
- Functional and oncological outcomes of total and near-total glossectomy with laryngeal preservation are similar in patients with advanced stage tongue cancer.
- Near-total glossectomy does not have a positive effect on nasogastric and tracheostomy tube dependence compared with total glossectomy.

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Prognostic and Clinical Role of Contrast Enhancement on Magnetic Resonance Imaging in Patients with Bell's Palsy

Original Investigation

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Abstract

Objective: To investigate the prognostic value of the magnetic resonance imaging in Bell's palsy patients.

Methods: Patients who were diagnosed and treated with Bell's palsy between October 2013 and March 2016 retrospectively selected. House–Brackmann grades, pre- and post-treatment pure tone audiograms, stapedial reflexes were analyzed and magnetic resonance imaging (MRI) scans with gadolinium-based contrast agents were evaluated. Contrast-enhanced segments of the facial nerve were determined. MRI findings were compared statistically with pre- and post-treatment grade, recurrence rate of Bell's palsy, MRI scanning timing, presence of stapes reflexes and posttreatment recovery data.

Results: No significant correlation was observed between pretreatment House–Brackmann grades and enhancement ($p>0.05$). Similarly, there was no significant correlation between clinical recovery and enhancement ($p>0.05$). Also, no significant correlation was observed between MRI scanning time, the recurrence rate of Bell's palsy and MRI findings ($p>0.05$). None of the MRIs showed neoplastic contrast enhancement.

Conclusion: The routine use of the contrast-enhanced temporal MRI is not recommended in the diagnosis and monitoring of Bell's palsy patients, because the contrast enhancement pattern of the facial nerve has no effect on the prognosis of Bell's palsy. MRI should be used in cases that do not heal despite treatment, for the differential diagnosis of facial nerve tumors and in patients who are candidates for surgical decompression.

Keywords: Bell's palsy, facial nerve, magnetic resonance imaging, gadolinium DTPA, prognosis, temporal bone, radiology, acoustic reflex

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Introduction

The most common cause of the sudden onset unilateral facial weakness is Bell's palsy (BP) (1). BP accounts for approximately 80% of all peripheral facial palsy cases (2). The incidence of BP is 20 to 30 cases per 100,000 adults per year (1). The etiology of BP is unknown, but viral infections, microvascular ischemic pathologies and autoimmune diseases have been proposed as possible mechanisms (3).

The facial nerve has a long and tortuous intratemporal course between the internal acoustic meatus and stylomastoid foramen. Therefore, the spread of inflammation and edema around the nerve is known to play a major role in the pathophysiology of BP as it leads to the blockage of axonal flow as a result of the compression within the bony canal (3).

Modern imaging techniques play an important role to evaluate the etiology of peripheral facial paralysis. Computerized tomography (CT) and magnetic resonance imaging (MRI) are the commonly used imaging techniques to evaluate patients with facial paralysis. While MRI is often used as the imaging modality in the diagnosis of possible inflammatory and tumoral causes, CT is often used to assess the temporal bone in trauma, otitis media, and preoperative evaluation (4). Compared with other imaging modalities, MRI provides more accurate results in detecting the neural damage. Gadolinium, a paramagnetic imaging agent, causes enhancement of T1-weighted signal intensity. Contrast material is primarily distributed in extracellular fluid and its effect is mostly notable in areas where an increased extracellular compartment is found such as neoplasm, inflammation, and edema (5). The most common pattern of facial nerve enhancement in patients with facial palsy has been reported to be at the distal intracanalicular and LS segments (2). Some studies report that the presence and spread of neural enhancement correlate with the duration and prognosis of the disease (2).

In the presented study we aimed to assess the clinical significance and the prognostic value of quantitative analysis of MRI in terms of contrast enhancement in patients with BP.

Methods

Patients with idiopathic peripheral facial nerve palsy (BP) who were treated between October 2013 and March 2016 in a tertiary referral center were retrospectively reviewed. Approval was obtained from the Ethical Committee of Dışkapı Yıldırım Beyazıt Training and Research Hospital before commencing the study (decision no: 33/16, date: 12.12.2016).

Complete neurotologic examination records of all patients were reviewed. Patients with otologic diseases, otologic and cranial operation history, autoimmune and rheumatologic diseases, acoustic trauma, and head trauma were excluded from the study. At first admission audiological tests (pure tone audiometry and stapedial reflex test) were performed and facial palsy was graded clinically according to the House–Brackmann (HB) grading system. All patients were treated with methylprednisolone therapy. On the first day of diagnosis, 250 mg methylprednisolone intravenous infusion was administered. In the following days, 1 mg/kg oral methylprednisolone was started, and the dose was decreased 16 mg every three days and the treatment continued for two weeks. We evaluated the progress of the disease and the final recovery for each patient. Recovery was accepted as at least one step improvement in the clinical grade of BP.

First, patients were classified clinically as primary and recurrent. Contrast enhancement rates between the two groups were compared. Then, patients with recurrent palsy were excluded, and other parameters were examined in primary patients. We compared MRI contrast enhancement patterns based on MRI examination time, HB grade, facial palsy, stapes reflexes and recovery rate after treatment.

All patients had been examined with 1.5-Tesla MRI device (Siemens, Magnetom Vision Plus VB33D, Erlangen, Germany). MRI was taken in coronal and axial planes using T1 sequences and T1-weighted postcontrast examination immediately after intravenous gadolinium-diethylenetriamine penta-acetic acid (Gd-DTPA) injection. On MRI scans specific anatomical segments of the facial nerve were determined and labeled as distal intrameatal (DIS), labyrinthine (LS), geniculate ganglion (GG), tympanic (TS), or mastoid (MS) segment of the facial nerve. Then each segment was assessed by the radiologist for the presence or absence of contrast enhancement (Figure 1). Enhancement results were recorded as positive or negative for each segment. All the images were examined by the same radiologist who was blind to the clinical grade of the facial palsy and the disease side.

Statistical Analysis

Standard deviation (SD), median, and rate values were used in the descriptive statistics of data. For qualitative data analysis, Pearson's chi-square test was used. A p-value less than 0.05 was considered significant. The Statistical Package for the Social Sciences for Windows, version 23.0 software program (Armonk, NY: IBM Corp.) was used.

Results

There were 81 female and 69 male patients with a mean age of 48 (18-85) years. Facial palsy was on the left side in 78 and on the right side in 72 patients. The average follow-up period was calculated as 7.1±1.9 (2 to 15) months. Mean time from the onset of the palsy to the Gd-MRI examination was 9.81 days (range: 1–60 days). Forty-three patients had enhancement of the facial nerve on postcontrast images (27.7%).

It was the first attack of facial palsy for 127 patients and 23 patients had previous history. Nine of the recurrences were on the ipsilateral side and 14 were on the contralateral side. Fifteen of the patients with recurrent palsy were female and eight were male, with a mean age of 51 (22–80) years. Patients with recurrent palsy had fully recovered from the previous paralysis. Their average recurrence time was three years. The relationship between the recurrence of facial

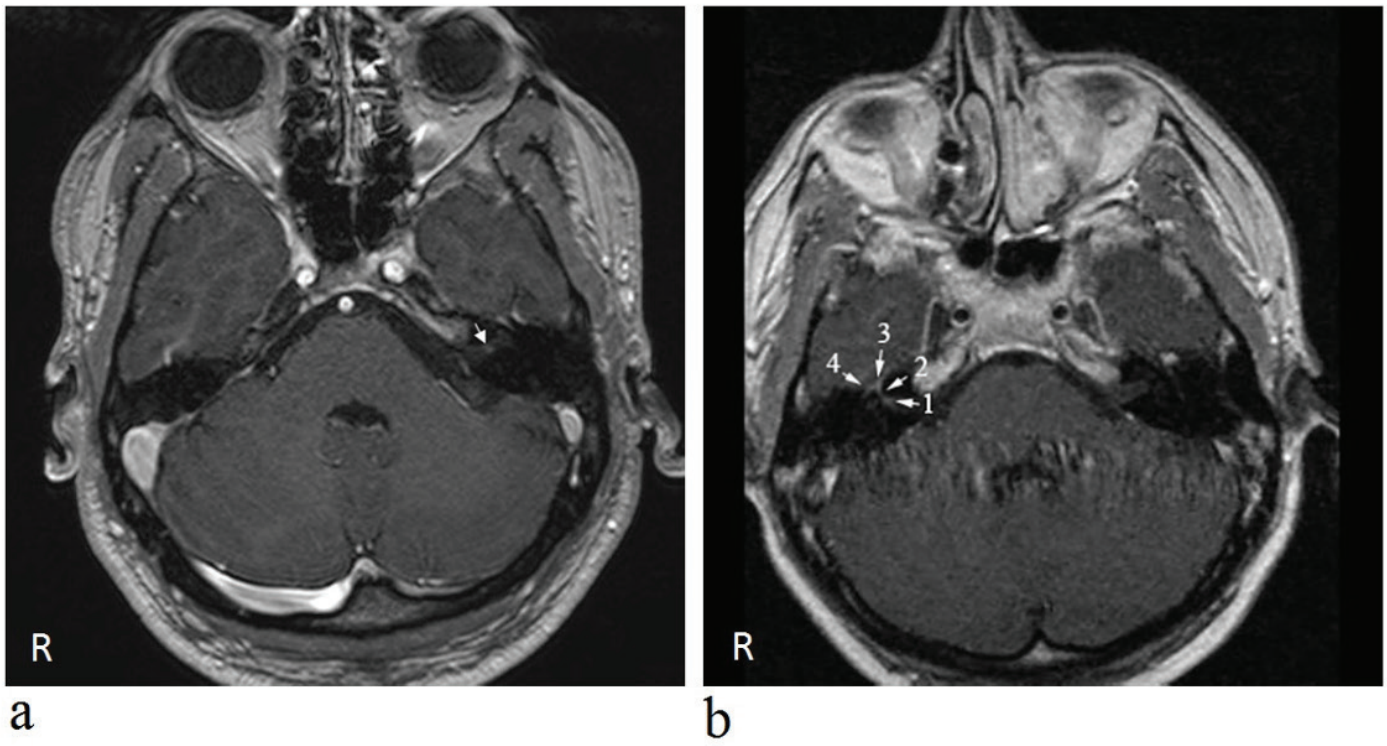

Figure 1. Contrast enhancement on T1-weighted axial MRI images a) Contrast enhancement of the left distal intrameatal segment, b) Contrast enhancement of right distal intrameatal (1), labyrinthine (2), geniculate ganglion (3) and tympanic segments (4) of another patient
MRI: Magnetic resonance imaging

paralysis and the facial nerve contrast enhancement on MRI was evaluated. Facial nerve enhancement was observed 29.1% in patients with first attack, whereas this rate was 26.1% in recurrent cases. The difference between the two groups was not statistically significant (p=0.963) (Table 1).

Patients with recurrent palsy were excluded and the enhancement distribution in the facial nerve segments of the primary patients was examined. Enhancement was observed in 37 of the 127 patients. While 18 of them had single segment involvement, 19 patients showed enhancement in more than one segment. The DIS segment was the most enhanced segment followed by LS, GG, TS, and MS, respectively (Table 2).

HB grade for primary cases were as follows: 54 patients grade II, 44 patients grade III, 12 patients grade IV, 13 patients grade V, and 4 patients grade VI. Pretreatment HB grade of the patients and facial nerve contrast enhancement rates on MRI were compared. There was no statistically significant relationship between HB grade and contrast enhancement (p=0.868) (Table 3). Pretreatment HB grade of disease was categorized as early and advanced. One hundred ten patients were in the early stage (HB II-III-IV) and 17 were in the advanced stage (HB V-VI). Then, the relationship between the early or late stage of the disease and the enhancement on MRI was evaluated. There was no statistically significant relationship between early or advanced stage disease and contrast enhancement (p=0.573) (Table 3). Further, we evaluated the relationship between the HB grade and the contrast enhancement of the single or multiple facial nerve segment. There was no statistically significant relationship between HB grade and single or multiple segment enhancement (p=0.794) (Table 4).

Table 1. Relationship between contrast enhancement and facial paralysis recurrence

			Contrast enhancement		p-value
			-	+	
Facial paralysis recurrence	First attack	n	90	37	0.784
		%	70.9	29.1	
	Same side recurrent	n	6	3	
		%	66.7	33.3	
	Opposite side recurrent	n	11	3	
		%	78.6	21.4	

At first admission of the patients, stapedial reflex test was performed and the relationship between stapedial reflex and contrast enhancement on MRI was evaluated. Stapedial reflex was positive in 71 (55.9%) patients and negative in 56 (44.1%). Contrast enhancement was observed in 31.0% of the stapes reflex positive cases, whereas this rate was 26.8% in stapes reflex negative cases. The difference between the two groups was not statistically significant (p=0.749) (Table 5).

Table 2. Contrast enhancement of primary cases according to facial nerve segments and distribution of contrast enhanced facial nerve segments

Contrast enhanced facial nerve segment	n (%)	Contrast enhanced facial nerve segment	n (%)
No enhancement	90 (70.9)	1	32 (86.5)
1	15 (11.8)	2	22 (59.5)
1,2	5 (3.9)	3	14 (37.8)
1,2,3	6 (4.7)	4	8 (21.6)
1,2,3,4	3 (2.4)	5	3 (8.1)
1,2,3,4,5	3 (2.4)	-	-
2	3 (2.4)	-	-
2,3,4	2 (1.6)	-	-

1: Distal intrameatal segment, 2: Labyrinthine segment, 3: Geniculate ganglion, 4: Tympanic segment, 5: Mastoid segment

Table 3. Relationship between contrast enhancement and pretreatment HB grade of primary cases

			Contrast enhancement		p-value
			-	+	
Pretreatment HB grade	II	n	38	16	0.868
		%	70.4	29.6	
	III	n	33	11	
		%	75.0	25.0	
	IV	n	8	4	
		%	66.7	33.3	
	V	n	9	4	
		%	69.2	30.8	
	VI	n	2	2	
		%	50.0	50.0	
	Early grade (HB II-III-IV)	n	79	31	0.573
		%	71.8	28.2	
	Advanced grade (HB V-VI)	n	11	6	
		%	64.7	35.3	

HB: House–Brackmann

The correlation between the post-treatment recovery rate of the patients and contrast enhancement on MRI was evaluated. During follow-up 119 patients showed improvement clinically and eight patients did not. While improvement was observed in 35 of the 37 patients with enhancement on MRI, improvement was observed in 84 of the 90 patients without enhancement. The difference between the two groups was not statistically significant (p=1.000) (Table 5).

The relationship between the time to MRI examination from the onset of the disease and the pathological facial nerve contrast examination was evaluated. Median time to MRI examination was seven days (mean: 9.81, SD: 9.62). We compared contrast enhancement on MRIs done in the first seven days and after the 7th day. There was no statistically significant difference between the two groups (p=0.329) (Table 6). We then examined the relationship of the contrast enhancement with pretreatment HB grade in patients who had their MRI in the first seven days. There was no statistically significant relationship between HB grade and contrast enhancement (p=0.658) (Table 6). The impact of contrast enhancement on the recovery rate was also analyzed in patients who had MRI in the first seven days. Recovery ratios were 88.9% and 92.6% in the enhancement positive and enhancement negative groups, respectively. The difference between the two groups was not statistically significant (p=0.636) (Table 6).

Table 4. Relationship between pretreatment HB grade and contrast enhancement of single or multiple facial nerve segment

			Contrast enhancement			p-value
			-	Single segment	Multiple segments	
Pretreatment HB grade	II	n	38	10	6	0.794
		%	70.4	18.5	11.1	
	III	n	33	4	7	
		%	75.0	9.1	15.9	
	IV	n	8	2	2	
		%	66.7	16.7	16.7	
	V	n	9	2	2	
		%	69.2	15.4	15.4	
	VI	n	2	0	2	
		%	50.0	0.0	50.0	

HB: House–Brackmann

Table 5. Relationship between pretreatment HB grade and contrast enhancement of single or multiple facial nerve segments on the MRIs done in the first seven days

			Contrast enhancement			p-value
			-	Single segment	Multiple segments	
Pretreatment HB grade	II	n	23	4	5	0.658
		%	71.9	12.5	15.6	
	III	n	16	3	1	
		%	80.0	15.0	5.0	
	IV	n	6	2	1	
		%	66.7	22.2	11.1	
	V	n	7	0	1	
		%	87.5	0.0	12.5	
	VI	n	2	0	1	
		%	66.7	0.0	33.3	
Total %		n	54	9	9	
		75.0	12.5	12.5		

HB: House–Brackmann, MRI: Magnetic resonance imaging

Table 6. Relationship between contrast enhancement and MRI examination time and relationship between contrast enhancement and pretreatment HB grade and recovery rates of patients who underwent MRI in the first 7 days

			Contrast enhancement		p-value
			-	+	
MRI examination time	0–7 days	n	54	18	0.329
		%	75.0	25.0	
	>7 days	n	36	19	
		%	65.5	34.5	
Patients with MRI in the first 7 days					
			Contrast enhancement		**p-value**
			-	+	
Pretreatment HB grade	II	n	23	9	0.658
		%	71.9	28.1	
	III	n	16	4	
		%	80.0	20	
	IV	n	6	3	
		%	66.7	33.3	
	V	n	7	1	
		%	87.5	12.5	
	VI	n	2	1	
		%	66.7	33.3	

		Contrast enhancement				p-value
		-		+		
		n	**%**	**n**	**%**	
		66.7	**75.8**	**33.3**	**24.2**	
Recovery after treatment	-	4	7.4	2	11.1	0.636
	+	50	92.6	16	88.9	

HB: House–Brackmann, MRI: Magnetic resonance imaging

Discussion

BP is a peripheral facial nerve palsy accompanied by any identifiable disease or external injury. It is the most frequently encountered type of peripheral facial nerve palsy and is thought to be mainly due to viral infections (1). The imaging method for the facial nerve disorders should be selected according to the suspected type and location of the pathology. Patients with BP generally have no abnormality in the bony canal of the facial nerve, therefore usually no pathological findings are seen on the temporal bone in CT (3, 6, 7). Thus, contrast enhanced MRI is mostly used by clinicians to evaluate any inflammation or tumorigenesis around the facial nerve for differential diagnosis (3, 8).

In MRI, gadolinium is used as the contrast agent to evaluate the facial nerve and the enhancement pattern is evaluated in T1-weighted series. Gadolinium mostly penetrates extracellular fluid. Although gadolinium cannot pass into the cranial nerves under normal conditions, it can penetrate through blood vessels in the presence of inflammation or edema, resulting in nerve enhancement (5, 8, 9). However, there are studies showing that there may be contrast enhancement in a facial nerve with normal functions (8, 10). In BP patients, MRI is used to gain insight into the severity and prognosis of the disease and to rule out possible tumoral pathologies by evaluating how much contrast enhancement each segment shows. Based on the hypothesis that facial nerve edema and inflammation increase the severity of facial paralysis, the increase in contrast enhancement on MRI may be correlated with the severity of facial paralysis (5, 9, 11). MRI can also reveal an enlargement of the facial nerve, a condition that is also seen in a neoplastic process. Facial nerve schwannomas appear as well-circumscribed round or fusiform soft tissue masses with low to intermediate signal intensity on T1 images, and after gadolinium injection show moderate to intense enhancement along the course of the facial nerve (cerebellopontine angle, internal auditory meatus, LS segment, GG, TS or MS segments and within the parotid gland) (6, 12).

In the light of this information, we investigated the effectiveness of temporal MRI in evaluating the severity and the prognosis of BP. For this purpose, we examined the facial nerve enhancement patterns on the temporal MRI scans of 150 patients who were treated for BP. Of these, 127 were primary and 23 were recurrent BP patients. There were no significant differences in contrast enhancement between primary and patients with recurrent palsy, but patients with recurrent palsy were excluded from the study to avoid bias. MRI scans of only 29.1% of the primary patients showed contrast enhancement. In fact, even this value alone raises suspicion about how suitable temporal MRI is for evaluating patients with BP; as contrast enhancement was seen in approximately one-fourths of the 127 patients, while there were no pathologies in the temporal MRI of the remaining 90 patients. None of the temporal bone MRIs showed a facial nerve tumor.

Recent studies on the benefit of temporal MRI in determining the prognosis of BP report confusing results. There are studies reporting that the presence and the spread of neural enhancement correlate with the duration and the prognosis of the disease (2, 10). According to the study of Yetiser et al. (2), there is a strong correlation between the presence of enhancement and the delay of recovery, and furthermore, involvement of more than one segment negatively affects the recovery. According to Sartoretti-Schefer et al. (10), the presence and the extent of enhancement are closely related to the duration and the outcome of the disease. It is worth noting that both studies have low numbers of cases. However, some studies also show that MRI did not have an association with the severity, the course, and the recovery of the disease (5, 8, 13, 14). In addition, there are studies in the literature that show that enhancement could occur even in normally functioning facial nerves. According to Gebarski

et al. (15) the geniculate, TS, and MS segments of the facial nerve have peri- and epineural venous plexus, so they can show enhancement under normal conditions. They found enhancement in 142 (76%) of 186 normal facial nerves in their study.

As known, prognosis is expected to be worse in advanced stage BP patients. We examined whether or not there were any differences in contrast enhancement in MRI according to the stages of the disease. We thought that there could be more contrast enhancement or enhancement in more segments in advanced stage patients. For this purpose, we first examined the contrast enhancement status in MRIs in all stages one by one and in early stage (HB II, III, IV) and advanced stage (HB V, VI) patients, and we did not see any statistically significant differences between the stages of the disease in terms of enhancement. In the literature, there are studies showing that involvement of multiple segments in MRI were related with the severity of the disease and prognosis in patients with BP (2, 10). In our study, we grouped patients as single segment and multiple segments involvement according to contrast enhancement on MRI and evaluated the relationship with the severity of the disease according to the HB staging. There were no statistically significant differences between single or multiple segments involvement and the severity of the disease. The relationship between post-treatment recovery rates and facial nerve enhancement seen on MRI was another issue we looked into in terms of the prognosis of BP.

In our study, facial nerve enhancement was observed in two of eight patients without clinical recovery and 35 of 119 patients with recovery after treatment. Thus, these results showed no statistically significant correlation between the enhancement of MRI and the recovery of BP. According to these results, we think that enhancement has no effect on prognosis and final outcome of the disease.

Time from the onset of palsy to MRI examination was considered to be another factor that could affect contrast enhancement on temporal MRI. It was deemed that there could be decreased contrast enhancement as the time from the onset of the disease increased, since the inflammation of the facial nerve would be alleviated by the treatment (5). There is also a study suggesting that enhancement could increase as the time to MRI is delayed (16). In our study, analysis of the time to MRI examination showed a median value of seven days. Presence of MRI enhancement was calculated as 25.9% in the first seven days after the onset of the disease and 32.3% after the seventh day. There was no statistically significant difference between the two groups, and it was concluded that the time to MRI examination had no correlation with facial nerve enhancement. Considering that the time to MRI could have an effect on enhancement and that the MRI times spanned a wide range, we wanted to analyze only those patients who had their MRIs done in the first seven days. There were no statistically significant differences between the clinical stages and the contrast enhancement rates of these patients. Moreover, there were no significant differences between the recovery rates and enhancement rates of this patient group. As a result, we do not recommend early detection with MRI since the time to MRI has no significant clinical benefits.

The stapes reflex is a diagnostic method that is frequently used in the clinical evaluation of BP patients, and it can be negative or positive. There are studies showing that this difference had an influence on prognosis, and prognosis is thought to be worse when stapes reflex is negative (17, 18). We could not find a study in the literature that investigated the relationship between stapes reflex and enhancement in MRI. We thought that in patients with negative stapes reflex, enhancement rate and spread could be higher because of the worse prognosis of the disease. Therefore, the relationship between stapes reflex and contrast enhancement on temporal MRI was evaluated. In our study, pretreatment stapes reflex was evaluated and found positive in 55.9% and negative in 44.1% of the patients. In cases with positive stapes reflex, 31.0% of the cases had contrast enhancement, whereas for those with negative stapes reflex it was 26.8%. The difference between the two groups was not statistically significant. We did not find that the stapes reflex had any effect on contrast enhancement. This result showed us that there were no relationships between enhancement on MRI and prognosis and the course of the disease. Two recently published clinical guidelines on the diagnosis, treatment and follow-up of BP, supporting our results, emphasize that routine MRI should not be performed in BP patients for clinical follow-up (19, 20).

There are some limitations of our study. The first and most important of these is that the imaging of the patients were made with a 1.5-Tesla MRI. More detailed and reliable results can be obtained in studies with the 3-Tesla MRI. Burmeister et al. (21) examined facial nerve enhancement in more detail in their studies using 3 Tesla MRI, but they showed that even this did not have a significant effect on predicting prognosis. In addition, contrast enhancement on MRI was evaluated by a single radiologist in our study. This evaluation may vary according to the experience of the radiologist (22). Lastly, this was a retrospective study and there was no standard MRI examination time. More accurate results may be obtained by conducting a prospective study with a larger patient group and standardized times to MRI.

Conclusion

In our study, the rate of contrast enhancement on MRI was extremely low and there were no statistically significant

relationships between contrast enhancement and the grade and the prognosis of the disease. Therefore, routine use of contrast-enhanced temporal MRI is not recommended in the initial phase of BP patients. Also, no facial nerve tumor was detected in our temporal bone MRIs. MRI should be used for nonhealing and progressive disease despite treatment, for the differential diagnosis of facial nerve tumors, and for patients who are candidates of surgical decompression.

Ethics Committee Approval: For this study ethics committee approval (decision no: 33/16, date: 12.12.2016) was obtained from the Ethical Committee of Dışkapı Yıldırım Beyazıt Training and Research Hospital, Ankara, Turkey.

Informed Consent: Informed consent was obtained from all individual participants included in the study.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

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

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

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Main Points

- Bell's palsy is the most common cause of sudden onset unilateral facial weakness.
- Magnetic resonance imaging is one of the most commonly performed examination in Bell's palsy.
- The aim of our study was to investigate the prognostic value of magnetic resonance imaging in patients with Bell's palsy.
- The routine use of contrast-enhanced temporal MRI is not recommended in the diagnosis and monitoring of Bell's palsy patients, because the contrast enhancement pattern of the facial nerve has no effect on the prognosis of Bell's palsy.
- MRI should be used in cases that do not heal despite treatment, for the differential diagnosis of facial nerve tumors, and in patients who are the candidates for surgical decompression.

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Use of Flexible Bronchoscopy in Foreign Body Aspiration

Original Investigation

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Abstract

Objective: Foreign body aspiration remains a serious health problem with a potential for severe consequences, and acute and chronic problems in children. It therefore demands immediate intervention. Rigid bronchoscopy has long been the method of choice for foreign body removal but is now being replaced by flexible bronchoscopy which offers reduced trauma and the ability to access distal bronchial regions. In the presented study we assessed the patients who underwent flexible bronchoscopy for foreign body removal in our clinic.

Methods: We reviewed the records of 20 patients who underwent flexible bronchoscopy due to suspected foreign body aspiration and had a foreign body removed in our clinic. Patients were analyzed in terms of sociodemographic data, foreign bodies removed, method of foreign body removal, foreign body location and time to diagnosis.

Results: Our study group included nine females and 11 males. The removed foreign body was organic in 19 of the 20 patients and inorganic in one patient. Hard organic food, such as hazelnuts, peanuts, seeds, almonds, and raw corn kernel were identified in 12 patients, a piece of fishbone in one patient and a piece of gelatin in another. The pieces of soft organic food identified were apple in one patient, egg in one patient, and boiled corn kernel in one patient and removed by suction. The foreign body was removed using forceps in nine patients, and a basket was used successfully in seven patients.

Conclusion: Foreign bodies can be removed with minimal complication using flexible bronchoscopy, basket, and forceps in children.

Keywords: Foreign bodies, airway, child, bronchoscopy, forceps

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Introduction

Flexible fiberoptic bronchoscopy (FB) was first used in pediatric patients in Germany in 1978, long after rigid bronchoscopy (RB) was used for foreign body removal in an adult patient in 1897. In general, bronchoscopy is helpful for the visualization of the airway anatomy, the assessment of airway dynamics, the treatment of obstructions and the collection of fluid samples, and brushing/biopsy procedures for microbiology and histopathology. It is an invasive procedure that requires sedo-anesthesia in children, and can lead to such

complications as desaturation, airway trauma, laryngeal spasm, and bleeding (1).

Foreign body aspiration (FBA) remains a serious medical issue that can lead to severe consequences and cause acute and chronic problems in children and thus requires immediate intervention (2). It is the fourth leading cause of accidental death in children younger than three years of age and the third leading cause of death in children aged younger than one year (3). Impaired oxygenation and ventilation due to obstruction of the airway can lead to morbidity (4). The primary treatment approach is RB as it provides airway patency. RB has long been the method of choice for foreign body removal but is being replaced by FB which offers reduced levels of associated trauma and the ability to access distal bronchial regions. However, practice is limited in the literature, especially in children (5). In the presented study we assessed the patients who underwent FB for foreign body removal in our clinic.

Methods

We retrospectively reviewed the records of 20 patients who presented to our clinic in 2016 to 2020 with a history, physical examination, clinical symptom or finding, or a chronic cough that raised the suspicion of FBA and underwent FB for foreign body removal.

Sociodemographic data, the type of the foreign body, method of foreign body removal, foreign body location, time to diagnosis, and chest X-ray findings were analyzed. Diagnostic and therapeutic bronchoscopy were performed using a flexible fiberoptic bronchoscope (BF-3C160, BF-H1100, Olympus, Japan) with an outer diameter of 3.8 mm and a thickness of 4.9 mm. The procedure was performed in the operating room with an anesthesiologist, a bronchoscopy nurse and two pediatric pulmonologists present. Given the possibility of failure with flexible bronchoscopy, our hospital's thoracic surgery department was informed prior to the procedure about a possible need for RB.

All patients gave their consent before bronchoscopy. The procedure was initiated by inserting a laryngeal mask airway (LMA) while the patient was under sedo-anesthesia. In patients whom a foreign body was identified, anesthesia was deepened, and the procedure was continued under general anesthesia.

Forceps and multi prong snare were used in the first attempt to remove the foreign body, and those that could not be grasped or removed using forceps and snare were removed using a blunt-end urological basket and N-Gage.

For removals using forceps, the forceps were first advanced through the suction cannula, the bronchoscope was pulled upwards slightly to make the tip of the forceps visible, and the foreign body was grasped and pulled upward together with the bronchoscope. The same procedure was performed when using snare.

For removals using a basket, the basket was advanced through the suction cannula and the bronchoscope was withdrawn after it reached the level of the foreign body. Upon reaching at the edge of the foreign body, the basket was opened and rotated around its own axis to capture the foreign body in the basket. After capturing the foreign body, the basket was locked and pulled slowly upward to stabilize the foreign body. If the removal failed, the procedure was repeated in regard of the patient's oxygenation.

Following the removal procedure, a FB was performed again to ensure that no residue remained in the airways. After the procedure, the patients were followed up closely and observed for at least one day as inpatients.

Approval was given by the Ethics Committee of Necmettin Erbakan University (decision no: 2021/3231, date: 07.05.2021).

Statistical Analysis

Statistical analysis was performed using SPSS (Statistical Package for Science Studies) version 22.0 for Windows. Variables with non-normal distribution were analyzed with a Mann-Whitney U test and a Kruskal-Wallis test. The results of the statistical analyses were expressed at a 95% confidence interval and $p<0.05$ was accepted as statistically significant.

Results

Foreign bodies were removed using FB in 20 of our patients, of whom nine (45%) were female and 11 (55%) were male. Their mean age was 21.8 months, their median age was 20 months (minimum: 8, maximum: 44 months), and 13 patients were aged 12–18 months.

Regarding the time taken until diagnosis, the shortest duration was one day and the longest was six months. The mean duration of complaint was 32.5 days, and the median was 20.5 days.

The presenting complaint was cough in 18 (90%) patients, of whom 10 (50%) had a complaint of chronic cough (21 days–6 months). In addition, eight (40%) patients presented with wheezing, three (15%) with bruising, and one (5%) with fever in addition to cough and wheezing, while four (20%) had a history of antibiotic therapy after a diagnosis of pneumonia. Of the total, eight patients had undergone RB (twice in one patient) at another center due to FBA. In one patient, the foreign body was removed, but the complaint did not resolve. The patient who underwent RB twice could not be assessed for the presence of a foreign body due to edema of the airway. The duration of complaints of seven patients who previously had undergone RB ranged from

three weeks to three months, and the duration of admission to our department due to unresolved complaints following RB ranged from one month to two months. One of our patients had a one-day history.

Respiratory system examination of nine (45%) patients was normal. There were rales in two (10%) patients, rhonchi in three (15%) and decreased breath sounds on the side of the foreign body in six (30%) patients. Stridor was present in two (10%) patients, of whom one had hoarseness and dyspnea, and the other had a barking cough along with stridor.

Increased aeration was detected on the chest X-ray of 11 (55%) patients, pneumonic infiltrates were observed on the radiographs of three (15%) patients, and the chest X-ray was normal in nine (45%) patients.

During the procedure, sedo-anesthesia was administered to all of our patients, and LMAs sized 1.5–2.5 mm were selected based on the weight of the patient (6). The procedure was performed using a bronchoscope of 3.8 mm in 10 (50%) patients and 4.9 mm in 10 (50%) patients. The instruments used in our clinic are shown in Figure 1.

The removed foreign bodies were hard organic food such as hazelnuts, peanuts, seeds, almonds and raw corn kernel in 12 patients, pieces of fishbone in one patient and gelatin in one patient (Figure 2). The pieces of apple in one patient, egg in one patient and boiled corn kernel in one patient were removed by suction. Unidentifiable organic materials were removed from four of our patients (Table 1).

Initially, forceps and snare were used in all patients for removal. The foreign body was removed using forceps in nine patients, while a basket was used successfully in seven of the patients when foreign bodies could not be removed using forceps. One patient presented with two pieces of fish bones, of which one could be removed using forceps. The second, however, lodged under the epiglottis and could not be removed with the forceps and so it was removed using N-Gage. In one patient, two foreign bodies were removed from two different locations using forceps. Pieces of egg, apple, and boiled corn kernel were removed by suction in three of the patients. The locations of the foreign bodies are given in Table 2.

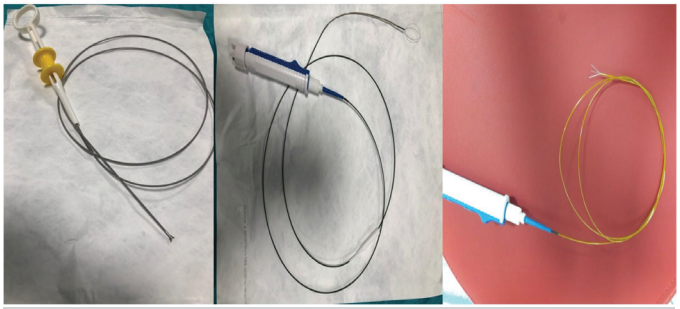

Figure 1. Tools used in our clinic (forceps, basket, snare)

None of the patients developed pneumothorax or hemorrhage. Edema of the airway developed in seven patients, while one patient who had a foreign body in the epiglottic tract developed laryngeal spasm, hypoxia, and edema during the removal procedure. In another patient, the foreign body was stuck in the vocal cords during the procedure, and hypoxia (SpO_2 decreased to 60% and was transient) and edema developed during removal with basket. None of the patients needed mechanical ventilation.

Of the total, five patients identified with granulation tissue and abundant secretion during the procedure were hospitalized for a week and placed on antibiotic therapy. The time of onset of complaint was ≥20 days in the patients with prolonged hospital stay. There was a statistically significant difference between the duration of complaints and the presence of granulation tissue, and between the length of hospital stay and the presence of granulation tissue ($p<0.004$ and $p<0.036$).

Table 1. Extracted foreign bodies and their numbers

Seed	4 (20%)
Hazelnut	2 (10%)
Almond	2 (10%)
Corn	2 (10%)
Egg mash	1 (5%)
Apple mash	1 (5%)
Peanut	1 (5%)
Chickpeas	1 (5%)
Fishbone	1 (5%)
Unidentified organic food	4 (20%)
Gelatin	1 (5%)

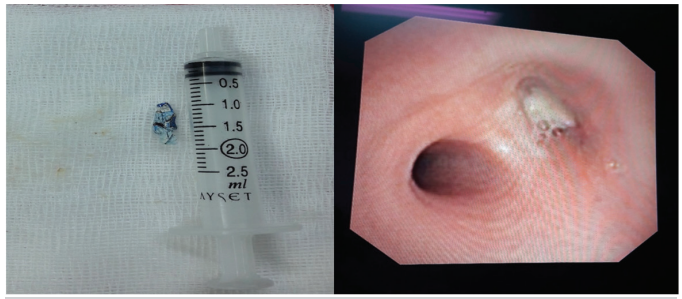

Figure 2. Foreign body specimens removed

Table 2. Anatomical localization of foreign bodies

Left main bronchus	6 (30%)
Left lower lobe	2 (10%)
Right main bronchus	5 (25%)
Right middle lobe	3 (15%)
Right upper lobe	1 (5%)
Trachea	1 (5%)
Epiglottis	2 (10%)

Growth in the cultured bronchoalveolar lavage sample collected during bronchoscopy was identified in 10 of the patients, namely *Klebsiella pneumoniae* in four patients, *Pseudomonas aeruginosa* in one patient, *Serratia marcescens* in one patient, *Moraxella catarrhalis* in one patient, *Enterobacteriaceae* spp. in two patients and coagulase-negative *Staphylococcus* in one patient. No such growth was identified in the remaining 10 patients (Table 3).

Discussion

FBA is the leading cause for concern in the field of pulmonology and requires immediate intervention. Among the reasons for the prevalence of FBA in young children are the tendency to place various objects in the mouth, the absence of posterior teeth, frequent and strong breathing while laughing and crying, immature swallowing and chewing functions, and habits of moving while eating (4). The risk of FBA is especially important in patients with intellectual disabilities due to impaired swallowing function. FBA is more common in males, which may be due to their more irregular behavior (7). In the presented study, males accounted for 55% of the sample.

For diagnoses of FBA, a physical examination should be supported by imaging methods, the most appropriate being posteroanterior (PA) chest radiography due to its ability to reveal findings to support FBA, such as mediastinal shift, difference in aeration between the lungs, obstructive emphysema, pneumonia, and atelectasis when a PA chest X-ray is completely normal. Previous studies in literature have reported normal chest radiography at rates of 10% to 46% (4, 8, 9). In the presented study, chest X-ray was normal in 45% of the patients. Chest X-ray alone is not sensitive or specific enough for the diagnosis of foreign bodies. If the history and physical examination findings support aspiration, a diagnostic bronchoscopy is required even if the chest X-ray is normal.

If the patient has dyspnea, persistent cough, asymmetry in auscultation and a localized increase in aeration on PA chest X-ray, therapeutic bronchoscopy should be performed. As the foreign body usually progresses to the bronchioles, the patient is clinically stable, and in such cases a recommended fasting period and appropriate equipment can be expected to remove the foreign body. However, if the patient develops severe respiratory failure, mediastinal shift, massive atelectasis,

Table 3. Demographic and clinical data of patients

Patient	Age (month)	Sex	Body weight (kg)	Bronchoscope outer diameter (mm)	Tool	Complication	Hospitalization (day/s)	Culture results
1	13	Male	12	3.8	Forceps	None	3	*Klebsiella, E.coli*
2	22	Male	14	3.8	Forceps	Edema	3	*Strep. salivarius*
3	22	Male	12.3	3.8	Forceps	None	4	*Strep. salivarius*
4	21	Male	10	3.8	Forceps	Edema	7	*Pseud. aeruginosa*
5	21	Female	11	3.8	Forceps	Nona	7	*Strep. salivarius*
6	19	Female	8.5	3.8	Forceps	None	7	*Streptococcus*
7	35	Female	12	3.8	Forceps	Edema	5	*Strep. mitis/Strep. oralis*
8	17	Female	9	3.8	Basket	Edema	8	*Klebsiella pneumoniae*
9	8	Female	8	3.8	Aspiration	None	1	*Moraxella catarrhalis*
10	25	Male	13	4.9	Basket	None	7	CNS
11	16	Male	10	4.9	Forceps	None	3	*Klebsiella*
12	42	Female	19	4.9	Forceps	None	0	*Enterobacter*
13	18	Male	15	4.9	Aspiration	Spasm, hypoxia, edema	1	Respiratory flora bacteria
14	17	Male	12	4.9	Basket	None	1	Respiratory flora bacteria
15	18	Male	8	3.8	Basket	None	3	*Serratia marcescens*
16	16	Female	9.8	4.9	Basket	None	1	Respiratory flora bacteria
17	44	Female	14	4.9	Aspiration	None	2	Respiratory flora bacteria
18	24	Male	11	4.9	Basket	None	3	*Klebsiella* spp.
19	18	Male	12	4.9	Basket	Edema	2	*Enterobacter*
20	21	Female	10	4.9	N-Gage	Spasm, hypoxia, edema	4	Respiratory flora bacteria

CNS: Coagulase-negative *Staphylococcus, E.coli: Escherichia coli, Strep. salivarius: Streptococcus salivarius, Pseud. aeruginosa: Pseudomonas aeruginosa*

or pneumothorax, or if there is a suspected foreign body such as a battery that can cause tissue necrosis, removal should be performed immediately.

Such procedures should be performed using FB if there are fewer specific symptoms of FBA, if the PA chest X-ray is normal, or if the patient has chronic or recurrent radiological abnormalities. RB is the standard treatment approach in FBA and is still used in many centers.

RB is mandatory when the foreign body is located in areas such as the larynx, subglottic region or trachea that may cause asphyxia, and when the foreign body has an irregular surface. In such cases, RB allows for good airway management and the use of more appropriate instruments for the removal of the object.

That said, peripheral bronchioles are difficult to reach using RB, and sometimes the field of view may be limited, and so there has been an increasing number of recent studies supporting the use of FB for foreign body removal (10). It should be noted, however, that the use of FB for foreign body removal in children younger than two years of age has been associated with several challenges, including the narrowness of the tracheal and bronchial areas, and the development of asphyxia and respiratory failure due to central airway stenosis caused by the use of an inappropriate bronchoscope. Although thin bronchoscopes can be selected for such cases, the procedure may fail due to the use of forceps that are less flexible and difficult to use. Tamate et al. (11) described a procedure in which a Fogarty balloon catheter was placed on the outer wall of the bronchoscope but concluded that the procedure was difficult. In our center, RB is still performed by the thoracic surgery department in patients who present with suspected FBA. The series in the presented study consisted of children in whom either a foreign body was detected by FB upon chronic cough, or no foreign body was detected by a previously performed RB, or who had chronic cough of which the etiology was being investigated or had unresolved pneumonia and wheezing.

In our experience, foreign bodies can be removed using flexible bronchoscopy more comfortably by experienced hands and with the appropriate equipment, although the removal of FBs using flexible bronchoscopy is still performed in very few centers and especially in pediatric cases.

Furthermore, foreign bodies, especially those in a peripheral location, may not be seen with RB, as was the case in eight of our patients, and the child's complaints may not be resolved despite RB.

After assessing this case series, our routine practice plan was to use flexible bronchoscopy to remove foreign bodies in the more distal bronchioles. In cases in which flexible bronchoscopy failed, we planned for RB to be performed by the thoracic surgery department.

In their adult study, based on the lack of any procedure-related complications in patients in whom foreign bodies were removed using FB, Sancho-Chust et al. (12) concluded that FB were more reliable than RB. The same study reported that procedures were successfully resolved using traditional biopsy forceps. The devices which are reported in the literature to be used to remove foreign bodies include standard biopsy forceps, forceps designed for the removal of foreign bodies (crocodile/alligator forceps), magnetic forceps (for metal objects), and various metal hooks, baskets, balloon catheters and cryoprobes (12).

There is always a risk of object displacement and backward movement when removing a foreign body, and factors such as size, shape irregularity, hardness, and consistency of the foreign body can affect this risk. If a basket is used to prevent backward movement, it is necessary to try to completely surround the foreign body. If forceps are used, it is necessary to grasp the object effectively and apply pressure. Foreign bodies are often lodged in the glottis as it is the narrowest part of the airway. If the bronchoscope is inserted nasally, the nasal fossae should be considered for stenosis (12). In the presented study, the procedure could not be completed in single attempt in some patients. When the object fell from the forceps or basket, the procedure was repeated from the point at which the foreign body fell. When foreign bodies remain in the airway for more than seven days (especially in dried fruit aspirations), edema and inflammation are more common and so procedure-related morbidity may increase and treatment may be challenging. A foreign body located in granulation tissue may restrict the field of view during bronchoscopy, and the mucosa may bleed easily. In such cases it is recommended that systemic steroids and antibiotics be administered for a few days before a second removal procedure, as this may facilitate the removal of the foreign body (13, 14).

The study by Varshney et al. (15) involving children reported that the foreign bodies were easily removed without damage to the tracheobronchial mucosa using flexible bronchoscopy in cases with severe laryngeal edema due to repeated RB, and that a postprocedural check was performed to be sure of the absence of residual foreign body. The patients in the presented study were also subjected to a control bronchoscopy to check the patency of the entire airway after the removal of the foreign body.

In all of the studies related to this issue, general anesthesia was administered to the patients. The most important factor in anesthesia management is the prevention of hypoxemia while removing the foreign body. In children, the inner diameter of the trachea is less than one cm, even at five years of age, while at the age of one year it is less than five mm (16). Matsushima et al. (17) suggested that bronchoscopy without intubation may be more advantageous due to the further narrowing of

the airway lumen and the increased pressure on the airway during procedures requiring intratracheal intubation. The authors also commented on the use of a laryngeal mask as a more appropriate approach to airway management in an increasing number of studies. In the present study, anesthesia was administered via a laryngeal mask in all patients, and high-frequency jet ventilation (HFJV) was reported to be effective in maintaining oxygenation during the insertion of the bronchoscope. In the presented study, none of the patients needed HFJV.

FB is a method that can be preferred when the foreign body advances to the distal bronchioles, and when it becomes necessary to reach the upper lobules. The consistency, the size and the shape of the foreign body should always be evaluated to prevent fragmentation or sticking in the laryngeal region during removal. If FB fails, there should be an option to continue with RB.

Conclusion

Foreign body removal using flexible bronchoscopy is not a commonly used approach in children, and although RB is still the general practice in many centers, FB seems to be a more comfortable and less invasive procedure for children as the airway can be assessed better by FB if oxygenation can be provided. We believe that this practice will become more common in pediatric pulmonology clinics in the years to come.

In the presented study we also observed a decline in the difficulties encountered with our first patients after hand skills were improved by appropriate training in our clinic. In our experience, foreign body removal using flexible bronchoscopy can be considered an alternative to RB in skilled hands due to its minimal complication risk and its reduced invasiveness and will be used more as the practitioners gain experience.

Ethics Committee Approval: Approval was given by the Ethics Committee of Necmettin Erbakan University (decision no: 2021/3231, date: 07.05.2021).

Informed Consent: Parents of the patients gave consent.

Peer-review: Internally peer-reviewed.

Authorship Contributions

Concept: G.Ü., S.P., Design: G.Ü., S.P., Data Collection and/or Processing: A.İ.Y., T.T., Analysis and/or Interpretation: G.Ü., Literature Search: G.Ü., A.İ.Y., T.T., Writing: G.Ü., S.P.

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Main Points

- Foreign body aspiration is the leading cause for concern in the field of pulmonology and requires immediate intervention.
- Flexible bronchoscopy is a method that can be preferred when the foreign body advances to the distal bronchioles, and when it becomes necessary to reach the upper lobules. If the flexible bronchoscopy fails, there should be an option to continue with rigid bronchoscopy.
- Foreign body removal using flexible bronchoscopy can be considered an alternative to rigid bronchoscopy in skilled hands due to its minimal complication risk and its reduced invasiveness and will be used more as the practitioners gain experience.

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Do the Age of Implantation, the Widths of Internal Acoustic Canal and Bony Cochlear Nerve Canal Affect the Auditory Performance of Primary School Children with Bilateral Cochlear Implants?

Original Investigation

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Abstract

Objective: To reveal the correlation between implantation age, the internal acoustic canal (IAC) width, bony cochlear nerve canal (BCNC) width, and auditory performance in primary school children with bilateral cochlear implantation (CI).

Methods: Preoperative IAC and BCNC widths of 57 pre-lingually deaf children aged 7–11 years who had previously undergone bilateral CI in our institution were reviewed and cut-off values were calculated. Twenty-four patients who had additional problems and could not attend school and those who refused to participate in the study were excluded. The remaining 33 were invited to the hospital, and their speech perception tests, and language development scores were analyzed (16 of 33 patients had been operated on before the age of 24 months).

Results: The cut-off values calculated from the 114 ears of 57 patients were 3.86 mm for IAC width and 1.56 mm for BCNC width. The auditory performances of the 33 patients after CI were not significantly different in the narrow and normal width groups. However, speech perception test results, and language development scores of patients implanted before the age of 24 months were significantly higher.

Conclusion: There are some studies showing that children with bilateral sensorineural hearing loss have narrower IAC and BCNC widths. However, we concluded that the widths of the IAC and the bone cochlear nerve canal did not affect auditory performance. We found that implantation age is the single most important determinant of speech-language development after CI.

Keywords: Cochlear implantation, age factor, bilateral hearing loss, temporal bone, surgery, cochlear nerve, pediatric, audiology, speech development

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Introduction

Cochlear implantation (CI) is an accepted treatment option for patients with severe to profound sensorineural hearing loss (SNHL) who do not benefit from hearing aids. Cochlear implants are useful in the perception of sound, speech understanding, and intellectual development in such patients (1, 2). Cochlear implants differ

from hearing aids in that they convert sound into electrical impulses. Implants also stimulate the cochlear nerve directly, bypassing the hair cells in the organ of Corti (2).

Retrocochlear SNHL occurs when the lesion is localized between the cochlea and the central auditory nervous system. While many malformations of the inner ear structure (Michel's deformity, cochlear aplasia, and Mondini's deformity) are associated with SNHL in pediatric patients, CI is a well-defined surgical procedure allowing hearing rehabilitation in such cases (3).

High-resolution computed tomography (CT) and magnetic resonance (MR) imaging are used in the preoperative surgical planning and the evaluation of congenital anomalies (4). In radiological findings, internal acoustic canal (IAC) width of less than 3 mm is considered as stenosis, and this has been reported as one of the causes of SNHL (5, 6). Recently, it has also been suggested that bony cochlear nerve canal (BCNC) stenosis could be associated with cochlear nerve deficiency (7). Currently, however, the number of reported studies is insufficient to determine the effect of BCNC and IAC dimensions on auditory performance after bilateral CI.

The main aim of this study is to reveal the correlation between the age of implantation, the widths of IAC and BCNC, and post-implant auditory performance in primary school patients with bilateral CI.

Methods

Ethical Approval

The study was approved by the University of Health Sciences Turkey, İstanbul Training and Research Hospital Clinical Research Ethics Committee (decision no: 2575, date: 13/11/2020). All procedures were performed in accordance with the ethical standards set forth by the World Medical Association Declaration of Helsinki (Scotland 2000). Informed consent forms were obtained from all parents before their participation.

Patients and Study Design

The study included 57 pre-lingually deaf primary school-aged children who underwent bilateral CI in our clinic. Children with peri- or post-lingually deafness, except 7-11 years old or those who underwent unilateral CI were excluded from the study. Severe to profound hearing loss was confirmed by the auditory brainstem response test in all 57 prelingually deaf children. Preoperative high-resolution CT and inner ear MR images were examined to detect abnormalities in the cochlea and the vestibulocochlear nerve.

Clinical evaluation of the patients and the CI decisions were made by a council of otolaryngologist, radiologists, speech-language therapists, and psychologists. We followed our patients with free field audiometry at the 2nd and 6th months after implantation. Routine post-operative speech and language rehabilitation sessions were held for at least two hours per week. Some patients, however, needed sessions up to 10 hours per week.

Preoperative CT images of children were reviewed by a single head and neck radiologist to determine the dimensions of the IACs and the widths of the BCNCs in the 114 ears of the 57 patients, and the means were used to determine their cut-off points which were found as 1.56 mm for BCNC and 3.86 mm for IAC.

To evaluate auditory performance, parents were asked to bring in their children, and patients were evaluated with speech perception test and verbal language development scale by a single experienced speech-language therapist. Of the 57 patients, 24 who had additional problems and could not attend school and those who refused to participate in the study were excluded. In the evaluation of speech perception, the Monosyllabic Trochee Polysyllabic Word Test (MTP) was used as a closed-set test and the Glendonald Auditory Screening Procedure (GASP) was used as an open-set test. The Turkish version of the Test of Early Language Development (TELD-3) was used to assess verbal language development. The auditory performances of the groups with narrow and normal canal widths were compared.

Measurement of IAC and BCNC Width

On transverse temporal CT image, the width of IAC was measured as the distance between the anterior and posterior bony margins at the middle of the canal (Figure 1a, 2a), and the width of the BCNC was measured as the distance between bony margins at the midportion between the base of modiolus and the fundus (Figure 1b, 2b). On coronal temporal CT image, the length of the IAC was measured as the distance from the falciform crest to the midpoint between the inferior and superior lips of porus acusticus internus and the height of the IAC was measured as the distance between superior and inferior bony margins at the middle of the canal (Figure 1c, 2c). All measurements were taken by the same head and neck radiologist.

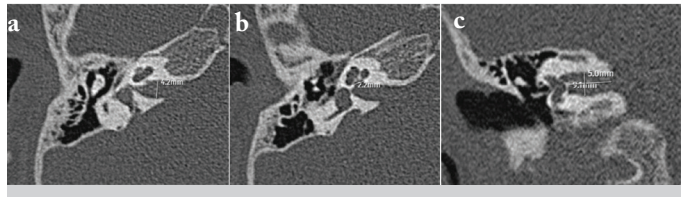

Figure 1. Temporal bone computerized tomography images showing the measurements of a) the width of internal auditory canal on transverse plane; b) the width of bony cochlear nerve canal on transvers plane; c) the length and height of internal auditory canal on coronal plane

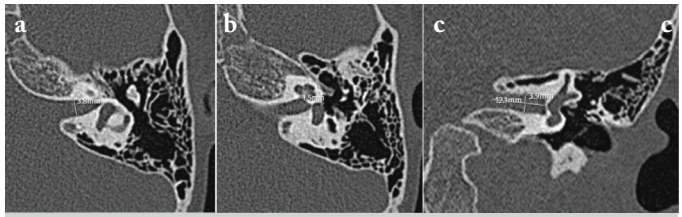

Figure 2. Temporal bone CT images showing the measurements of a) the width of internal auditory canal on transverse plane; b) the width of bony cochlear nerve canal on transvers plane; c) the length and height of internal auditory canal on coronal plane

CT: Computed tomography

Speech Perception Tests

Closed-Set Test: MTP, devised by Erber and Alencewicz (8), consists of monosyllabic, trochee, spondee, and polysyllabic words. The test can be applied to children aged two years and older. In the test, children are shown 12 images and asked to point to/repeat the word they heard. The correctly recognized words are scored. The test was administered only aurally, without the aid of lip reading. Each word was given twice in a 12-word test and the test was evaluated over 24 points.

Open-Set Test: Erber (9) developed the GASP. The test uses only question sentences. This is helpful as children can get confused when questions and commands are used together in the same test. Children can repeat or answer the question.

The Turkish Version of the Test of Early Language Development (TEDIL)

TEDIL is the Turkish adaptation of the TELD-3. The Turkish version, which was developed in 2005 and finalized in 2009, measures early verbal language development and covers three of the five basic components of language (semantics, morphology, and syntax). It measures the receptive and expressive verbal language skills of children. TEDIL consists of parallel forms, A and B and each form includes two subtests. There are a total of 76 items in each form. Some of these items address the skills to identify and describe images, and others address the ability to follow verbal instructions and verbally answering questions. TEDIL has five objectives: (a) to identify children who are significantly behind their peers in terms of early language development and to ensure that they receive early intervention; (b) to identify individual verbal strengths and weaknesses; (c) to set the program and process in language therapy; (d) to serve as a scale for examining language skills in early childhood; and (e) to support other assessment techniques (10). TEDIL is a useful tool for identifying children with language development problems.

Statistical Analysis

Statistical analysis was performed using the IBM SPSS Statistics 17.0 package program (SPSS Inc.; Chicago, IL, USA). The Kolmogorov–Smirnov test was used to determine whether or not the variables were normally distributed. Categorical variables of the data were presented with n (%). While presenting descriptive analyses, mean ± standard deviation (SD), median (minimum-maximum), Q1 and Q3 values were used. The Mann–Whitney U test was used for unpaired group comparisons and the Kruskal–Wallis test for comparisons between more than two groups. To determine the cut-off value for the 114 ears, a cut-off determination graph with normal distribution was used. Measurements were compared between the IAC AP and BCNC groups with the Kruskal–Wallis test. The Mann–Whitney U test was used for the comparison between groups separated by age of implantation. Spearman's correlation coefficient was used to examine the relationships between measurement data. The relationship between IAC and BCNC values and their relationship with the other measured parameters were analyzed using the Spearman's correlation coefficient. A p-value less than 0.05 was considered statistically significant.

Results

The mean age of the 57 primary school children participating in our study was 8.28 years (range 7 to11 years). The cut-off values calculated from 114 ears of 57 patients were 3.86 mm for IAC width and 1.56 mm for BCNC width.

The ears of 33 patients who could respond to language development tests were evaluated under two groups. The first group consisted of 17 patients (51.52%) who were implanted at the age of 24 months or older (mean age was 45 months), and the second group consisted of 16 patients (48.48%) who were implanted younger than 24 months (mean age was 17 months).

According to the cut-off values, there were three patients (9.09%) with unilateral stenosis in the width of IAC and three patients (9.09%) with bilateral stenosis. There were three patients (9.09%) with unilateral stenosis in BCNC width and five patients (15.15%) with bilateral stenosis (Table 1). In addition, when these 33 patients were examined, nine patients (27.27%) with radiological anomalies were detected. Of the nine patients, four had right high jugular bulb (12.12%), three had left high jugular bulb (9.09%), one patient (3.03%) had right chronic otitis media and one patient (3.03%) had incomplete partition type 2 (Table 2).

Auditory performance was tested in language development and speech perception. In the evaluation of speech perception, the MTP and the two-syllable test were used as closed-set test, and the GASP and the two-syllable test were used as open-set test. TEDIL was used to assess verbal language development.

Patients with and without IAC stenosis were compared, and no significant differences were found between the three groups

Table 1. Canal widths and test scores of patients with stenosis

	R IAC (mm)	R BCNC (mm)	L IAC (mm)	L BCNC (mm)	MTP	GASP	2 syllable Open-Set Test	TEDIL receptive language
1. R IAC stenosis	3.7	-	-	-	24	9	9	32
2. R IAC stenosis	3.7	-	-	-	24	10	9	37
3. b-BCNC stenosis	-	1.5	-	1.3	24	3	5	13
4. b-BCNC stenosis	-	1.3	-	1.5	24	10	8	32
5. b-BCNC stenosis	-	1.2	-	1.4	24	7	5	26
6. b-BCNC stenosis	-	1.3	-	1.4	24	8	7	28
7. b-BCNC + L IAC stenosis	-	1.3	3.7	1.3	24	10	10	33
8. R BCNC stenosis	-	1.4	-	-	24	10	7	29
9. L BCNC stenosis	-	-	-	1.5	24	10	8	34
10. b-IAC stenosis	3.7	-	3.7	-	24	10	9	28
11. R BCNC stenosis	-	1.5	-	-	24	10	8	36
12. b-IAC stenosis	3.5	-	3.4	-	24	8	7	22
13. b-IAC stenosis	3.4	-	3.4	-	23	7	5	12

R: Right, L: Left, b-: Bilateral, IAC: Internal acoustic canal, BCNC: Bony cochlear nerve canal, MTP: Monosyllabic Trochee Polysyllabic Word Test, GASP: Glendonald Auditory, Screening Procedure, TEDIL: Test of Early Language Development, (-): No stenosis

Table 2. Radiological characteristics of patients

	n	%
IAC width		
Normal (>3.86 mm)	27	(81.82)
Unilateral stenosis	3	(9.09)
Bilateral stenosis	3	(9.09)
BCNC width		
Normal (>1.56 mm)	25	(75.76)
Unilateral stenosis	3	(9.09)
Bilateral stenosis	5	(15.15)
Facial dehiscence		
Right	1	(3.03)
Left	2	(6.06)
Radiologic anomalies		
Normal	24	(72.73)
High jugular bulb	7	(21.21)
Chronic otitis media	1	(3.03)
IP type 2	1	(3.03)

IAC: Internal acoustic canal, BCNC: Bony cochlear nerve canal, IP: Incomplete partition

in terms of speech perception and language development (MTP: p=0.576, GASP: p=0.461, TEDIL: p=0.108) (Table 3). Similarly, no significant differences were found between the three groups (MTP: p=0.403, GASP: p=0.175, TEDIL: p=0.233) in terms of BCNC width (Table 4).

Thirty-three patients were divided into two groups according to implantation age. The open-set test and TEDIL language development test scores of the early implanted (<24 months of age) group were found to be significantly higher than late implanted (≥24 months of age) group (Table 5: early implanted open-set test score mean ± SD 8.06±1.88, late implanted mean ± SD 6.24±2.84; p=0.037; early implanted TEDIL score mean ± SD 28.75±7.57, late implanted mean ± SD 22.76±8.85; p=0.036). Neither were there any significant differences between the results of the closed-set tests (MTP scores: early implanted, mean ± SD 23.94±0.25; late implanted, mean ± SD 22.76±3.85; p=0.081).

The correlation between canal measurements and auditory performance tests were examined. Accordingly, positive correlations were observed between right IAC antero-posterior distance and the two-syllable open-set test, and between left AC height and GASP (Table 6).

Discussion

The most important goal of CI is to enable children born with severe hearing loss to attend mainstream education. Most children with cochlear implants attend mainstream schools and this is a measure of CI outcome success. The results of our study show that about 33 out of 57 implantees were enrolled in mainstream education.

CI is considered the best treatment option for severe to profound bilateral SNHL. Radiological dimensions of BCNC and IAC can be easily measured using temporal bone CT; however, the recommended cutting values for stenosis vary in different studies (11).

In our study, we compared the auditory performance of children who underwent bilateral cochlear implant surgery in terms of their implantation age, IAC and BCNC widths. Since

Table 3. IAC width and auditory performance

	IAC width					
	Normal		Unilateral stenosis		Bilateral stenosis	
	n=27		n=3		n=3	
	Mean ± SD	Median	Mean ± SD	Median	Mean ± SD	Median
MTP	23.56±1.58	24.00	24.00±0.00	24.00	23.67±0.58	24.00
2 syllable closed-set test	23.22±3.08	24.00	24.00±0.00	24.00	23.67±0.58	24.00
GASP	7.63±2.87	9.00	9.67±0.58	10.00	8.33±1.53	8.00
2 syllable open-set test	6.89±2.67	8.00	9.33±0.58	9.00	7.00±2.00	7.00
TEDIL receptive language performance	25.30±8.72	28.00	34.00±2.65	33.00	20.67±8.08	22.00
TEDIL expressive language performance	28.07±7.50	30.00	35.67±4.04	38.00	18.33±16.26	24.00

SD: Standard deviation, IAC: Internal acoustic canal, MTP: Monosyllabic Trochee Polysyllabic Word Test, GASP: Glendonald Auditory Screening Procedure, TEDIL: Test of Early Language Development

Table 4. BCNC width and auditory performance

	BCNC width					
	Normal		Unilateral stenosis		Bilateral stenosis	
	n=25		n=3		n=5	
	Mean ± SD	Median	Mean ± SD	Median	Mean ± SD	Median
MTP	23.48±1.64	24.00	24.00±0.00	24.00	24.00±0.00	24.00
2 syllable closed-set test	23.12±3.19	24.00	24.00±0.00	24.00	24.00±0.00	24.00
GASP	7.68±2.76	9.00	10.00±0.00	10.00	7.60±2.88	8.00
2 syllable open-set test	7.08±2.81	8.00	7.67±0.58	8.00	7.00±2.12	7.00
TEDIL receptive language performance	24.64±8.98	24.00	33.00±3.61	34.00	26.40±8.02	28.00
TEDIL expressive language performance	27.16±9.25	28.00	33.67±3.21	35.00	28.00±8.60	31.00

SD: Standard deviation, BCNC: Bony cochlear nerve canal, MTP: Monosyllabic Trochee Polysyllabic Word Test, GASP: Glendonald Auditory Screening Procedure, TEDIL: Test of Early Language Development

Table 5. Implantation age and auditory performance

	Implantation Age												
	Early implanted (<24 months)						Late implanted (≥24 months)						p
	n=16						n=17						
	Mean ± SD	min	Q1	median	Q3	max	Mean ± SD	min	Q1	median	Q3	max	
MTP	23.94±0.25	23.00	24.00	24.00	24.00	24.00	23.29±1.96	16.00	24.00	24.00	24.00	24.00	0.157
2 syllable closed-set test	23.94±0.25	23.00	24.00	24.00	24.00	24.00	22.76±3.85	8.00	23.00	24.00	24.00	24.00	0.081
GASP	8.88±1.67	4.00	4.00	9.50	10.00	10.00	6.94±3.15	0.00	4.00	7.00	10.00	10.00	0.080
2 syllable open-set test	8.06±1.88	3.00	3.00	8.50	9.00	10.00	6.24±2.84	0.00	5.00	7.00	8.00	10.00	0.037
TEDIL receptive LP	28.75±7.57	12.00	22.50	32.00	34.00	37.00	22.76±8.85	7.00	16.00	24.00	31.00	34.00	0.036
TEDIL expressive LP	29.94±9.41	0.00	27.00	31.00	36.50	39.00	25.94±7.99	11.00	21.00	26.00	31.00	38.00	0.079

MTP: Monosyllabic Trochee Polysyllabic Word Test, min: Minimum, max: Maximum, GASP: Glendonald Auditory Screening Procedure, TEDIL: Test of Early Language Development, LP: Language performance, Q1: First quartile, Q3: Third quartile

the development of the nervous system is completed at an early age, there is no age-related difference in IAC and BCNC dimensions. Our study included patients of similar ages.

Although BCNC and IAC widths are considered risk factors for severe SNHL, most patients in this study had normal BCNC and IAC width. According to our data, the cut-off values were 1.56 mm for BCNC and 3.86 mm for IAC. We found that BCNC or IAC stenosis did not significantly affect auditory performance after bilateral CI. Kim et al. (11) noted with a similar result that a narrow BCNC did not significantly affect auditory outcome after CI. However, there

Table 6. Correlation between canal measurements and auditory performance

		R IAC ap (mm)	R IAC length (mm)	R IAC height (mm)	R BCNC (mm)	L IAC ap (mm)	L IAC length (mm)	L IAC height (mm)	L BCNC (mm)
MTP	r	0.296	0.095	0.101	-0.110	0.151	0.074	0.196	-0.131
	p	0.094	0.598	0.575	0.543	0.402	0.680	0.276	0.467
2 syllable closed-set test	r	0.269	0.222	0.096	-0.198	0.125	0.202	0.227	-0.216
	p	0.130	0.214	0.595	0.270	0.489	0.260	0.203	0.227
GASP	r	0.225	0.144	0.294	-0.008	0.202	0.224	0.390	0.029
	p	0.207	0.423	0.097	0.964	0.259	0.211	0.025	0.871
2 syllable open-set test	r	0.371	-0.020	0.212	0.071	0.339	0.129	0.234	-0.140
	p	0.034	0.912	0.235	0.696	0.054	0.473	0.191	0.438
TEDIL receptive language performance	r	0.287	0.160	0.295	-0.023	0.231	0.277	0.317	-0.042
	p	0.105	0.374	0.095	0.899	0.195	0.118	0.072	0.818
TEDIL receptive language performance	r	0.314	0.103	0.316	0.071	0.299	0.226	0.312	0.006
	p	0.075	0.570	0.073	0.694	0.091	0.206	0.077	0.975

R: Right, L: Left, b-: Bilateral, IAC: Internal acoustic canal, BCNC: Bony cochlear nerve canal, ap: Anteroposterior distance, MTP: Monosyllabic Trochee Polysyllabic Word Test, GASP: Glendonald Auditory Screening Procedure, TEDIL: Test of Early Language Development

are some studies showing an association between BCNC or IAC width and auditory performance after bilateral CI (12, 13). The real reason for the low scores in these studies may not be IAC or BCNC stenosis, but concomitant inner ear anomalies.

In our study, the age of implantation was the single most important determinant of speech-language development after implantation. We found that children in the early implanted group had better performance, as also observed in the literature (14).

Speech perception may be impaired in patients with long-term hearing deprivation. Meister et al. (15) asserted that prolonged auditory deprivation adversely affected daily auditory performances. This situation is best avoided by early CI (16). Several studies have concluded that prelingually deaf children had improved speech perception following CI (17). In addition, Jain et al. (18) showed that early implant users developed more vocabulary than late implant users. Our findings were similar to the research findings mentioned above.

Our study has several limitations. First of all, a limited number of patient populations has been included in the study. Further studies with larger case numbers are needed. Secondly, auditory performance was tested in language development and speech perception in our study. The test battery can be expanded by applying daily communication skills and auditory reasoning performance tests. Thirdly, it would be useful to follow-up more closely on their routines, such as their postoperative speech and language rehabilitation schedules, whether or not they adhere to these schedules and how many hours per week they attend.

Conclusion

There are some studies showing that children with bilateral SNHL have narrower IAC and BCNC widths. However, we concluded that the width of the IAC and the bone cochlear nerve canal did not affect auditory performance. In our study, we found implantation age to be the single most important determinant of speech-language development after CI. Children with narrow BCNC are also candidates for early CI and rehabilitation.

Ethics Committee Approval: The study was approved by the University of Health Sciences Turkey, İstanbul Training and Research Hospital Clinical Research Ethics Committee (decision no: 2575, date: 13/11/2020).

Informed Consent: Informed consent forms were obtained from all parents before their participation.

Peer-review: Externally peer-reviewed.

Authorship Contributions

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

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Main Points

- In our study, unlike other studies, we concluded that the width of the internal acoustic canal and bony cochlear nerve canal did not affect auditory performance.
- We found that implantation age is the single most important determinant of speech-language development after cochlear implantation (CI).
- Children with narrow internal acoustic canal and bony cochlear nerve canal are also candidates for early CI and hearing rehabilitation.

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3D Model to Understand the Diagnosis and Treatment of Horizontal Canal BPPV

Video Article

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Abstract

Objective: Our primary objective was to develop a three-dimensional (3D) model of the vestibular labyrinth to understand the pathophysiological mechanisms of benign paroxysmal positional vertigo (BPPV) observed during common diagnostic positional tests. We secondarily aimed to monitor the effects of the repositioning maneuvers and use this tool in teaching.

Methods: A 3D model of a human semicircular canals (SSCs) system was created by 3D printing the core and assembling it with silicone tubing filled with lubricant oil containing colored small stones in the lumen mimicking otoconia. We used the model in horizontal canal BPPV diagnostic tests and therapeutic maneuvers. The working mechanism of the model we designed was recorded with video.

Results: The model allowed for a clear display of the anatomy and the respective orientations of the SSCs. Otolith movement in the horizontal canals could be imitated during diagnostic positional tests (Dix-Hallpike and Pagnini-McClure) and therapeutic maneuvers (Epley, Semont, Lempert and Gufoni).

Conclusion: As well as helping to understand the anatomy and physiology of the SSCs, this simple 3D model also provides a teaching tool for the diagnosis and treatment of BPPV. The mechanism of horizontal canal canalithiasis and the effect of therapeutic repositioning maneuvers could be clearly observed by watching the markers in the lumen demonstrating the progress of otolith movements with changes in head position relative to gravity.

Keywords: Benign paroxysmal positional vertigo, vestibular diseases, vestibular labyrinth, otoliths, three-dimensional printing, anatomic model

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Introduction

Benign paroxysmal positional vertigo (BPPV) is the most common cause of vertigo. The semicircular canals (SSCs) can be involved with the underlying mechanisms of canalithiasis or cupulolithiasis. Since there are many BPPV variants depending on the location of the otoliths in the canal sand the cupula, a clear understanding of the

pathophysiological mechanisms are crucial for the correct diagnosis and a successful treatment (1, 2).

The anatomy and biomechanics of the SSCs was previously studied using different techniques including radiology, histology, and computer-based modelling (3, 4). Most of them, however, were unclear and not simple enough, and we noticed that a practical understanding of the diagnostic and therapeutic aspects of the disease was necessary, especially for physicians practicing in medical fields different from neurotology.

Repositioning maneuvers (RMs) are performed to send the otoliths back into the utricle; most commonly, Epley and Semont maneuvers are used to treat posterior canal BPPV, while Lempert (Barbecue) and Gufoni maneuvers are performed for the horizontal canal involvement. These RMs are generally poorly understood by examining illustrations, photographs, or animations. Also, conventional three-dimensional (3D) anatomical models of the inner ear are not specifically designed for BPPV and are rather expensive. A 3D anatomical model is necessary to provide a clear understanding of the anatomy and physiology of the vestibular labyrinth, as well as the diagnostic tests and RM for those interested in BPPV.

BBPV most commonly involves the posterior SSCs, followed by the horizontal and anterior canals. There are more variants in the horizontal canal and their mechanisms of emergence are harder to understand. In our report, we present a horizontal canal canalithiasis model which we believe will help to understand these mechanisms.

Methods

Ultimaker 2+ extended (Build volume 223x220x205 mm, 8.77x8.66x8.07 ins), (Ultimaker, Netherlands), Porima 1.75 mm Pla filament (Porima, Turkey), Johnsons baby oil (Johnson & Johnson Medical, USA), silis astroturf sand (Tuna Silis Kum, Turkey), tennis bandanna, transparent silicone 3D printing model (Link: https://vestibularfirst.com/how-to-print-3d-vestibular-apparatus/), tubes (ID: 10 mm OD: 13mm), (Önder silikon, Turkey) were used in the study. This project took about six-ten hours to print (depending on the 3D printer and its settings). First, one end of the tube was inserted into the canal, and oil and sand were fed in. Then, the other end of the tube was inserted into the other canal. When the model was ready, it was mounted on the head of a voluntary subject using a tennis bandanna. The horizontal SSC was ensured to standard a 30-degree angle with the horizontal plane. This model was constructed in order to understand the anatomy and physiology of the SSCs, and also serve as a teaching tool for the diagnosis and treatment maneuvers of BPPV.

Results

The Dix-Hallpike test for the posterior canal and the Pagnini-McClure (Roll) test for the horizontal canal BPPV variants were recorded during head movements. Video recordings were also made during Epley, Lempert and Gufoni maneuvers. Close-up recordings were also shot to observe the migration of the marker otoliths in the horizontal SSCs, focus on heir behavior and the duration of movements until they reach their final destination.

Position changes appear much faster in the videos compared to the real-life implementation where a significant waiting period is needed to allow the otoliths to move by gravity. Also, close-up view of the mounted model is not provided for each video for simple demonstration.

Discussion

As the underlying mechanisms of BPPV are becoming more and more clearly understood, new variants, tests and treatment maneuvers are continuously being described. It is not easy to develop a simple concept model to understand and teach the pearls of BPPV. Illustrations, photographs, videos, and animations were somewhat successful to achieve these goals; but there are certain limitations for these media such as access to the sources, the clarity of the production and the contradictions between mathematical modeling and clinical experience.

A free 3D model readily obtained by printing might be expected to allow for a clearer understanding of the various possibilities of otolithic movement in the endolymph during certain head movements. By using 3D modeling, confusing and contradictory clinical observations may be explained by simulating the hypothetical mechanisms. In this report we presented a 3D horizontal canal canalithiasis model as the first part of our study; however, a single model is not enough to understand the different vestibular stimulation patterns or the therapeutic head positions relative to gravity.

In this model, the presentation of the SSCs may not be so perfect and the flow of induced movements of the marker otoliths may not reflect the temporal patterns of real situations. We believe that the inability to add a moving cupula is the most important limitation of the 3D model presented here, but we are currently working to add a bendable cupula like a swing door inside the lumen of the tubing to study the mechanics of cupulolithiasis. Also, bilateral 3D modeling, using liquids of different densities inside the canals and changing the size sand the shapes of the canals to study the effects of anatomical variations are under consideration.

In the study by Chien et al. (3) a 3D model of the temporal bone was built to better understand the anatomy. The aVOR application developed in Sydney was developed as a teaching, training, and testing tool for the vestibulo-ocular reflex, vestibular system and its disorders, including BPPV.

It demonstrates eye movements, including those caused by canalithiasis. The application has been downloaded more than 50,000 times and its usefulness in teaching BPPV to medical students has been demonstrated (4).

Santos et al. (5) used a 3D numerical approach to show the biomechanical behavior of the vestibular system. They worked on computational simulations and numerical models. They stated that such models would also contribute to rehabilitation processes. Similarly, Traboulsi and Teixido (6) and Teixido et al. (7) developed a biomechanical model showing that it could be useful in multicanal involvement and emphasized that a 3D software were useful for the diagnosis and treatment of BPPV.

Bhandari et al. (8, 9) stated in two separate studies that simulators are useful for visualizing otolith movements during maneuvers for treating BPPV. However, they stated that 2D visualizations were limited while 3D sand dynamic simulations were more effective in understanding BPPV.

Yılmazer and Topçuoğlu (10) showed the anatomy of the SSCs with a two-handed model and stated that it would be effective for understanding the diseases of the SSCs.

In our study, the movements of otoliths in the horizontal SSC were visualized in a 3D model for the first time. We believe that the model could facilitate the understanding of otolith movements during head positions. However, it does not evaluate the eye movements that occur in BPPV. Also in our study, only the horizontal canal was studied, and new studies are certainly needed for other SSCs involvements.

Conclusion

We believe that a simple and free 3D model as presented here is a useful clinical tool to understand and treat BPPV. With the 3D model, a detailed and accurate conceptualization of BPPV can be obtained.

Video 1. 3D Model of BPPV

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Video Link: https://cms.galenos.com.tr/SolvePark/Uploads/Files/3D-Model-of-BPPV.mp4

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

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Main Points

- Understanding of the anatomy of the semicircular canals and their position relative to various head movements is crucial for the diagnosis and treatment of BPPV.
- The 3D model allowed for a clear display of the anatomy and respective orientations of the semicircular canals.
- It can also be used as a teaching tool for the diagnosis and treatment of BPPV.

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Posterior Cervical Intramuscular Schwannoma Within the Trapezius Muscle: A Case Report

Case Report

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Abstract

Schwannomas are benign soft tissue tumors derived from the Schwann cells of the peripheral nerves. An intramuscular schwannoma arising within the trapezius muscle in the posterior neck is rare. We report a case of a 31-year-old woman with an intramuscular schwannoma in the trapezius muscle. A painless and smooth-surfaced mass from 10 years ago was evident on palpation in the right posterior neck. Ultrasonography revealed an oval mass with clear borders and slight internal blood flow. No continuous hypoechoic lesions were noted at the tip of the mass. Magnetic resonance imaging of the neck revealed a mass in the right posterior cervical trapezius muscle with isointensity on T1-weighted imaging and heterointensity on T2-weighted imaging. Based on these findings, a schwannoma was suspected. Ultrasonography guided fine needle aspiration cytology revealed no significant findings. During surgery, a white-colored, encapsulated-tumor mass was found in the trapezius muscle. Histopathologically, hypocellular and hypercellular areas of fusiform cells were conspicuous, and nuclear palisading was observed in a part of the hypercellular region, confirming the diagnosis of schwannoma. To our knowledge, this is an extremely rare report of an intramuscular schwannoma within the trapezius muscle; herein, we report its clinical, radiological, and pathological features.

Keywords: Neck neoplasm, benign soft tissue tumor, intramuscular, schwannoma, trapezius muscle, case report

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Introduction

Schwannomas are benign soft tissue tumors derived from the Schwann cells of the peripheral nerves, with the head and neck region being the most common site of their origin. The main clinical manifestation of schwannoma is a slow-growing, painless, soft swelling. Although it can be treated by surgical removal, postoperative neurologic deficits present a problem (1).

Intramuscular schwannomas, though rare, mostly occur in the thigh and trunk muscles (2, 3); they infrequently occur in the head and neck region. Moreover, there are scarcely any reports on intramuscular

schwannomas occurring in the upper fibers of the trapezius muscle in the posterior cervical region. Therefore, ours is a rare case of a posterior cervical intramuscular schwannoma within the trapezius muscle. In this report, we describe its clinical, radiological, and pathological features.

Case Presentation

A 31-year-old woman presented with a mass in the right posterior neck that had been growing for 10 years. On palpation, a soft, mobile mass (30×40 mm) with elastic-consistency was noted. Neurological symptoms such as pain, Tinel's sign, sensory disturbance, and motor paralysis were absent. Ultrasonography revealed an oval mass measuring 21×38×36 mm with a clear border and slight internal blood flow in the muscles of the posterior cervical region. No continuous hypoechoic lesion was noted at the tip of the mass. Ultrasonography guided fine needle aspiration cytology (FNAC) revealed no significant findings; additionally, FNAC did not elicit an electrical shock-like sensation. On magnetic resonance imaging (MRI), the mass in the right trapezius muscle was isointense on T1-weighted images and heterogeneous on T2-weighted images (Figure 1). Therefore, we considered a provisional diagnosis of schwannoma, neurofibroma, paraganglioma, or malignant peripheral nerve sheath tumor (MPNST). During surgery, a partial transverse incision into the trapezius muscle revealed a white encapsulated tumor (Figure 2). The nerve related to the origin of the tumor could not be identified. The tumor, measuring 25×30×25 mm, was excised with the capsule. On histopathological examination, the Antoni A region was characterized by dense, coarse, and long fusiform tumor cells in a swirling arrangement with wavy cell poles; a palisade arrangement was also observed in regions of hypercellularity. In the edematous area (Antoni B), floating spindle cells were observed (Figure 3). The tumor cells were positive for S-100 and vimentin, but negative for neurofilaments, desmin, and CD 34; the Ki-67 Index was <10%. Presently, i.e., 2 years after the operation, there is no evidence of recurrence.

Discussion

Schwannomas mainly occur in the fourth decade of life, with 80% of the patients aged between 30–69 years (4). The incidence rate of this lesion in the head and neck region is 25%–45% (1). In this region, schwannomas mostly occur in the lateral neck, but other sites include the parotid glands, the cheeks, the scalp, the tongue, and the pharynx. Majority of intramuscular schwannomas develop in the trunk and the extremities, and rarely occur in the head and neck region (4). Among the intramuscular schwannomas in the head and neck region, most cases have been reported in the masseter muscle (5).

Intramuscular tumors in the trapezius muscle have been rarely reported. Of these, hemangiomas were the most frequently reported; additionally, desmoid tumors and lipomas have been reported (6). Hemangiomas have different ultrasonographic and MRI features compared with schwannomas, making it possible to differentiate between the two. A search in the English-written literature revealed that only one case of intramuscular schwannoma within the trapezius muscle with false positive positron emission tomography/computed tomography finding, had been reported to date (7). Therefore, more cases are needed to obtain a comprehensive description of its clinical, radiological, and pathological features.

The clinical, imaging, and pathological features of the intramuscular schwannoma in this case were similar to the schwannomas found in other muscles. Intramuscular schwannomas typically present as swellings, and they differ clinically from non-intramuscular schwannomas in that they are not associated with neurologic symptoms, such as pain, Tinel's sign, sensory disturbances, and motor paralysis (8). These symptoms were also absent in the presented case. Secondly, ultrasonography, MRI, and pathological findings showed that this intramuscular schwannoma was nearly identical to schwannomas arising in the other parts of the body (5).

The differential diagnosis for schwannomas based on imaging includes neurofibromas, paraganglioma, and MPNST (9). As

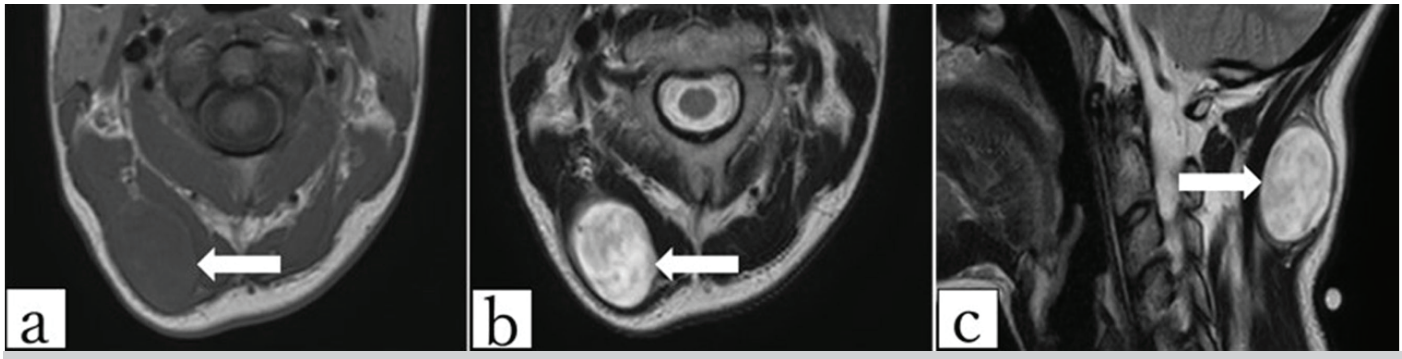

Figure 1. Magnetic resonance imaging of the neck
a) Horizontal section on T1-weighted image shows isointensity (arrow); b) and c) Horizontal section and sagittal section on T2-weighted image show heterointensity (arrow)

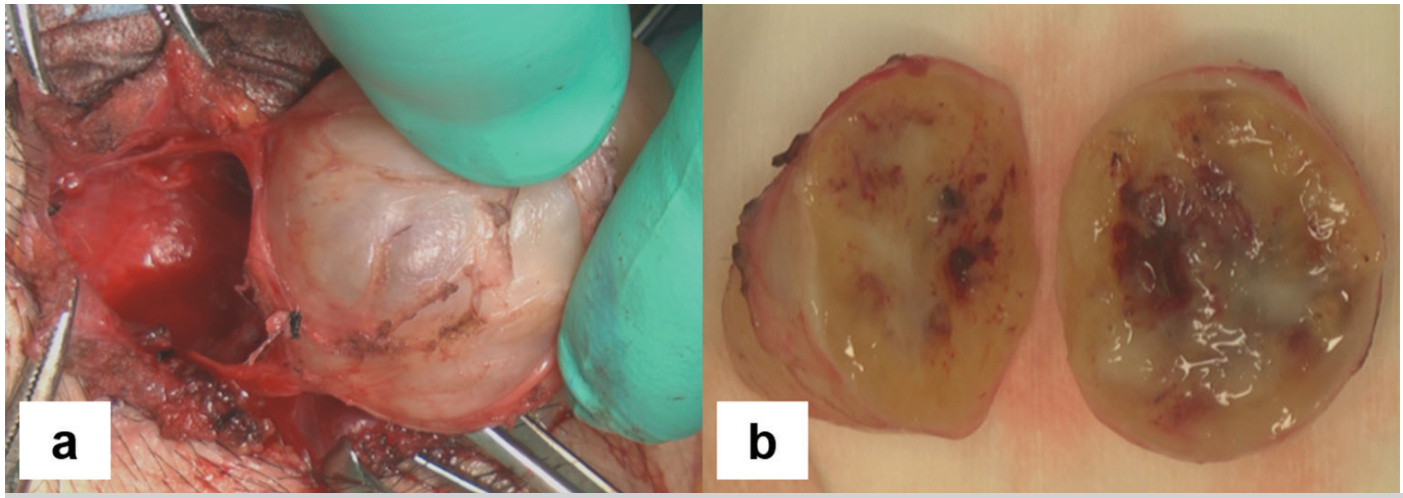

Figure 2. Surgical findings and specimens
a) Intraoperative findings. The tumor was excised; b) Specimen. The cut surface of the tumor appears yellowish white and is surrounded by a capsule

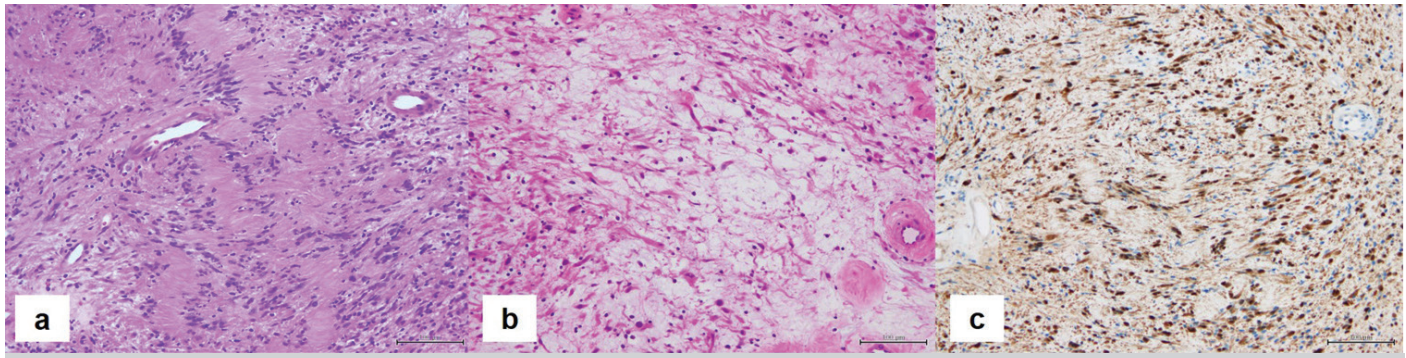

Figure 3. Pathological findings
a) and b) Coarse and spindle-shaped cells are prominent within the tumor. Hematoxylin & eosin, ×200; c) Immunostaining is positive for S-100 (arrow). S-100 immunostaining, ×400

for the echogenic findings on ultrasound of each differential disease, neurofibromas are characterized by internal chordal hyperechoic areas reflecting the inclusion of fatty tissue. Paragangliomas develop expansively and are hyperechoic. MPNSTs present as hypoechoic masses with some internal hyperechoic areas. In any case, the presence of a nerve sheath connecting the tumor to the nerve cord helps to differentiate it from other diseases. However, the presented case had no nerve sheath or continuous hypoechoic lesion making differential diagnosis for the intramuscular schwannomas challenging on ultrasonography.

In general, MRI (T2-weighted) findings of neurogenic tumors show that schwannomas show an irregular internal signal, as much as a non-solid mass. It is thought that the central part shows a high signal (10). Neurofibromas show a low signal in the center due to dense nerve fibers. Paragangliomas show an equal signal with T1-weighted image and a high signal with T2-weighted image, and when the tumors grow, a salt and pepper appearance is observed. MPNSTs show non-uniform signal intensity on T1-weighted images and have a contrast effect on the tumor margin. This case had no specific features on MRI.

Intramuscular schwannomas typically present as swellings, and they differ clinically from non-intramuscular schwannomas in that they are not associated with neurologic symptoms, such as pain, Tinel's sign, sensory disturbances, and motor paralysis (4, 8). We suggest that intramuscular schwannomas, on account of their deep location within the muscle, may not present with Tinel's sign. Altogether, it was difficult to determine a preoperative diagnosis, because an intramuscular schwannoma developing in the trapezius muscle in the posterior cervical region, as in this case, is very rare.

Conclusion

This is an extremely rare case report of a posterior cervical intramuscular schwannoma within the trapezius muscle, described with clinical, radiological, and pathological features.

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Main Points

- Intramuscular schwannoma arising within the trapezius muscle in the posterior neck has rarely been reported.
- The clinical, radiological, and pathological features of intramuscular schwannoma in this location are similar to other intramuscular schwannomas.
- They are not associated with neurologic deficits.

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Case Report

Promising Outcome of Radiation Therapy for Chondroblastoma of Temporal Bone in Childhood: A Case Report

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Abstract

Chondroblastoma is a rare cartilaginous benign bone tumor. Chondroblastoma in the temporal bone is also quite rare. Total excision is the main treatment. Data regarding tumor response to radiation therapy (RT) is insufficient. We describe a case of chondroblastoma that was treated with RT following subtotal tumor resection. In this case, the patient was a 14-year-old male who presented with a three-month history of ear fullness and hearing loss in his right ear. Magnetic resonance imaging revealed a mass partly filling the right external auditory canal and the inferior part of the middle ear. Histopathological findings indicated chondroblastoma. Subtotal tumor resection was performed due to risk of complications. RT was planned upon the growth of the tumor during follow-up. Treatment with subtotal resection and postoperative RT has been successful and the patient had no recurrence in the course of the 12-year follow-up. In chondroblastoma, complete surgical resection is still the gold standard. But the success of subtotal resection followed by adjuvant RT should also be kept in mind for cases where the total excision would pose high morbidity.

Keywords: Chondroblastoma, hearing loss, temporal bone, radiation therapy, pediatric otorhinolaryngology, case report

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Introduction

Chondroblastoma is a tumor rarely seen in temporal bone in childhood. It accounts for less than 1% of primary bone tumors (1). It is a benign cartilaginous tumor that often affects the epiphysis of long bones and is seen in the second to third decades of life (2). Chondroblastoma usually originates from the squamous part of the temporal bone (3). Clinical picture shows a painful mass with hearing loss and tinnitus. While total excision is the mainstay of the treatment, there is insufficient data

regarding tumor response to radiation therapy (RT). In this case report, we present our experience with the long-term follow-up of a teenager with chondroblastoma who had undergone RT following subtotal tumor resection.

Case Presentation

A fourteen-year-old male was admitted with ear fullness and hearing loss for three months in his right ear in 2008. A fragile and hemorrhagic mass filling the right external auditory canal (EAC) was observed in examination. The tympanic membrane could not be evaluated. No pathology was observed in the left ear. Pure tone audiometry (PTA) revealed slight conductive hearing loss with 3-frequency average of air and bone conduction thresholds of 30 and 5 dBHL, respectively, in the right ear. Air and bone conduction thresholds were 5 for each dBHL on the left. Non-contrast computerized tomography (CT) of the temporal bone showed an expansile and destructive lesion in right temporal bone (Figure 1a). Magnetic resonance imaging (MRI) of the temporal bone showed a mass partly filling the right EAC and the inferior part of the middle ear. Longest axis in the superior to inferior direction was 18 mm. The mass was irregularly shaped. It was hypointense in T1W, isointense in fat-saturated T2W images and showed contrast enhancement after post-contrast fat-saturated T1W images (Figure 1b). Following a biopsy, intact canal wall mastoidectomy was performed with posterior tympanotomy through the facial recess. It was noted that the reddish, fragile and bloody mass in the inferior part of the middle ear had remodeled the inferior part of the posterior wall of the EAC and reached the mastoid cavity. The fallopian canal was eroded in the middle part of the mastoid segment of the facial nerve. The ossicles were tumor-free. Upon obtaining the frozen section result as a benign lesion with no further detail, we tended to act based on the permanent histopathology report. Hearing levels could be preserved after the biopsy. The fact that permanent histopathologic examination confirmed the diagnosis of chondroblastoma led us to aim at intended-total excision of the lesion. During the main operation, the EAC was lowered in order to improve tumor exposure. The tumor was removed via canal wall down (CWD) mastoidectomy to the maximum extent possible and some residue was left behind the temporomandibular joint (TMJ) and on the jugular bulb in order to prevent inevitable injury. Meatoplasty was performed. The patient was discharged without any complications. The hearing level remained almost at the same level. Histopathologic examination of the main operation specimen revealed the same diagnosis (Figure 2). A wait-and-see policy was followed in order to observe the fate of the residual tumor. In a follow-up examination two years after the operation, it was noted that the entrance

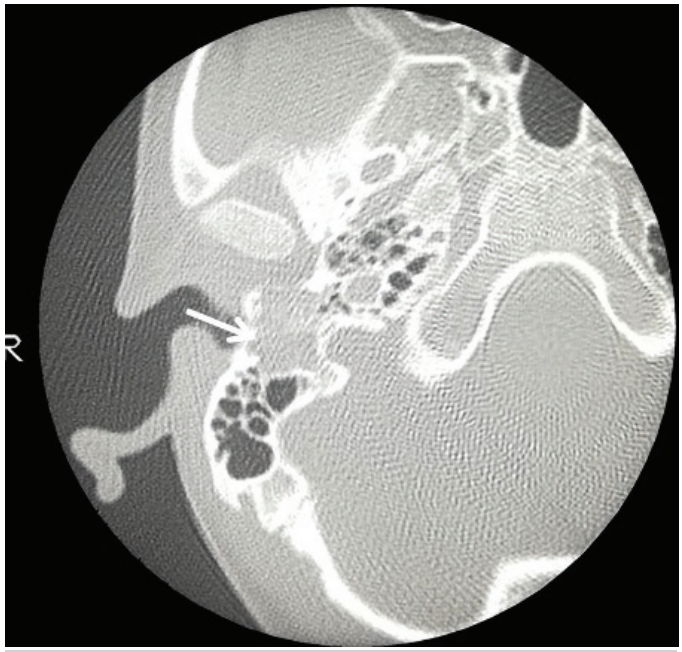

Figure 1a. Axial thin-section non-contrast CT image of temporal bone showing destructive and expansile lesion in right temporal bone (arrow)
CT: Computed tomography

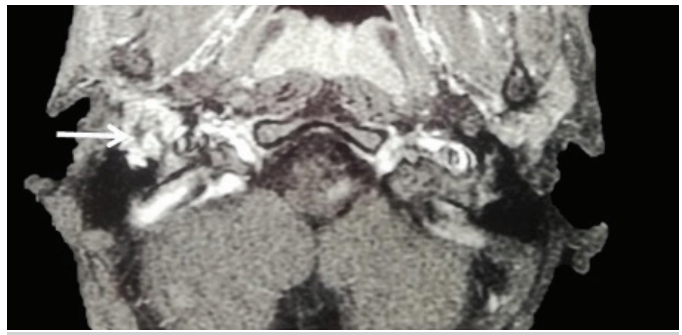

Figure 1b. Axial post contrast fat-saturated T1W image shows tumor in the right temporal bone (arrow)

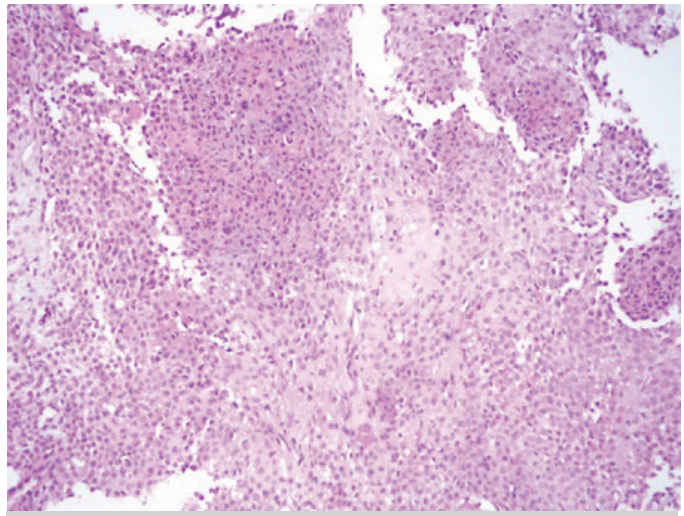

Figure 2. Sheet-like proliferation of chondroblasts with extracellular matrix. Scattered osteoclast-like giant cells are present and focal mineralization of matrix surrounding single cells can be seen (H&E 200X)

of the CWD mastoidectomy cavity was narrowed inferiorly by the tumor. PTA revealed increased conductive hearing loss in the right ear (Right: 45/10 dBHL, Left: 5/5 dBHL). Follow-up MRI showed increased dimensions of the residual tumor. It extended medially towards the vertical segment of the internal carotid artery and anteriorly towards the TMJ (Figure 3). Considering the morbidity of the required surgery led us direct the child to RT. Sixty Gy intensity modulated image guided RT was delivered in 30 fractions with 2 Gy fraction dose for six weeks (Figure 4). Grade 2 peripheral facial paresis and synkinesis developed on the right side a few months later which healed spontaneously in the next months. He developed total hearing loss in the right ear due to RT (PTA: Right air/bone: 110+/67+dBHL, Left air/bone: 10/5 dBHL).

The patient has been on follow-up for 12 years and does not have any complaints. A very recent MRI showed no evident contrast enhancement in the temporal bone (Figure 5). He is still on our yearly follow-up program.

Discussion

Chondroblastoma is a histologically benign but clinically aggressive tumor (4). Its etiology is unknown. It is caused by immature chondroblasts in the epiphysis of long bones (5). While the disease usually involves long bones and especially the femur and humerus, the involvement of flat bones such as the skull is extremely rare (<2%) (6). The temporal bone is the most frequently involved cranial bone. The squamous part is usually involved due to its cartilaginous origin (3).

In a systematic review of 100 cases, the mean age at diagnosis of skull and facial bone chondroblastoma was 42.3 years (2). In the same study, 4 out of 100 cases were diagnosed under the age of 18. There also was a slight male predominance (ratio, 1.3:1). Likewise, our patient is a 14-year-old male.

Hearing loss (especially the conductive type), tinnitus, cranial nerve involvement, facial swelling, and otalgia are the most common symptoms (4, 6, 7). TMJ dysfunction can also be seen. Diagnosing can be difficult. Chondroblastoma can be mistaken for the granulation tissue, giant cell reparative granulomas, giant cell tumors and osteosarcoma (3).

Radiological imaging is important for diagnosis. CT imaging typically shows internal calcification associated with intra-tumor calcium and hemosiderin deposition and rarely expansile intraosseous soft tissue mass with increased intravenous contrast enhancement (6).

The amount and morphologic pattern of calcification are variable on CT (8).

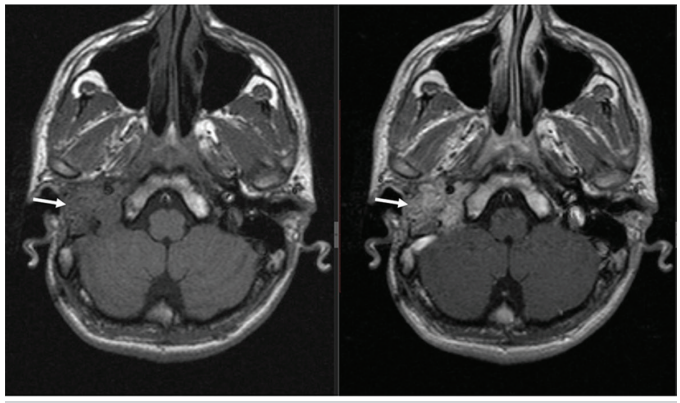

Figure 3. Axial pre- and post-contrast T1W images show increased dimension of the residual tumor (arrow)

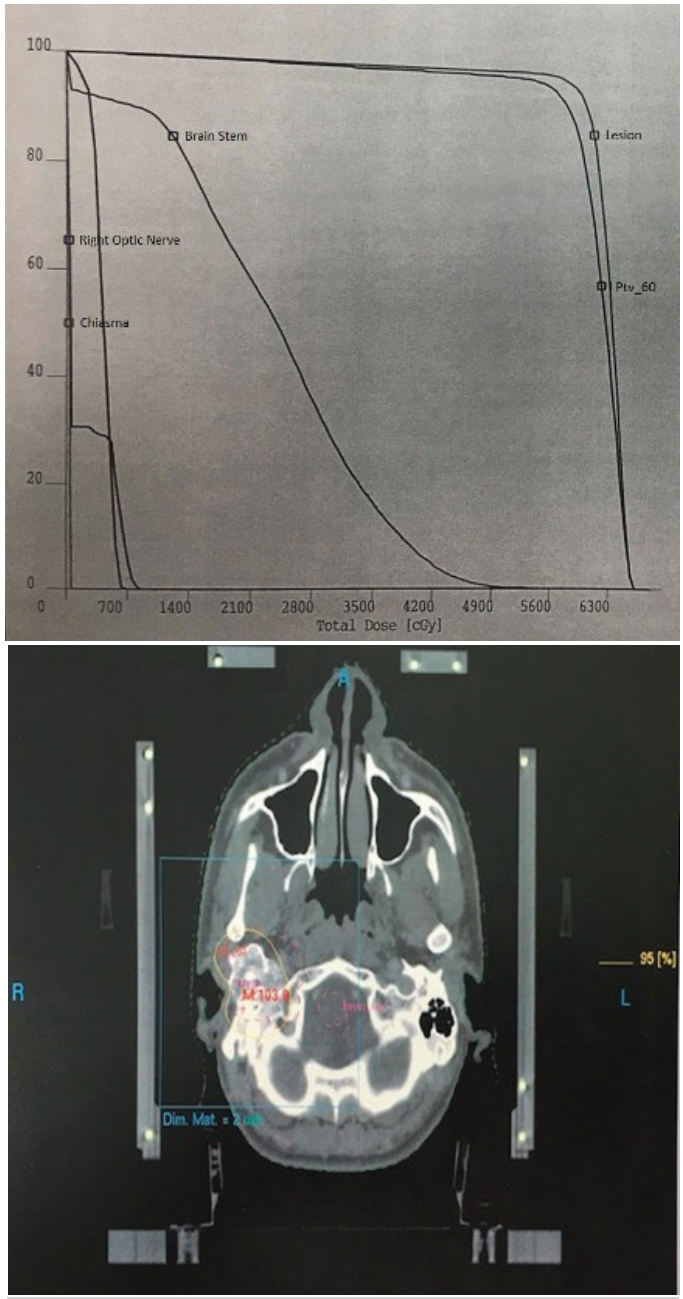

Figure 4. Dose volume histogram and digitally reconstructed radiography image of the planning target volume of the lesion

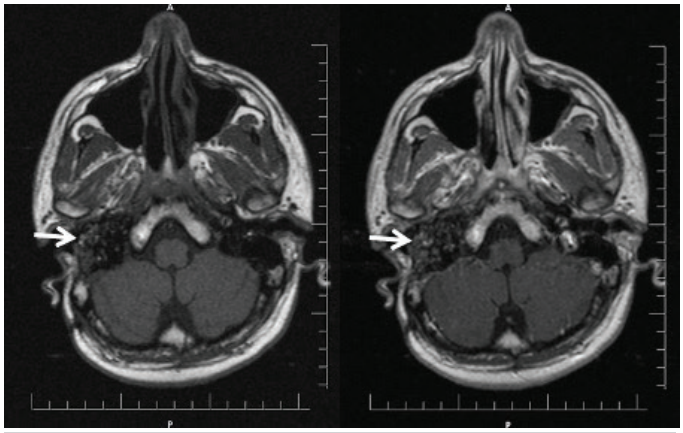

Figure 5. Axial pre- and post-contrast T1W images show no significant tumor enhancement (arrow)

MRI typically shows hypo-intermediate signal in T1 imaging and high signal in T2 due to bleeding into the mass. The cause of heterogeneity in T2 images is intense cellularity and dense vascular fibrous tissue. Post-gadolinium increase, which is a prominent hyperintensity component, shows heterogeneity in T1W imaging. In addition, MRI better describes intracranial/other soft tissue involvement such as dura and brain (6). The exact diagnosis is made pathologically. It is very difficult to diagnose with frozen section due to its similarity to other benign and malignant giant-cell neoplasms (2, 3). Diagnostic histopathological findings are the presence of chondroblast, osteoclastic-like giant cells, and chondromyxoid stroma around neoplastic cells. Chicken wire calcification is typical. These tumors secrete s-100 and vimentin (6).

The standard procedure for chondroblastoma is complete or wide/en bloc surgical resection. Complete removal of the tumor is recommended because of high local recurrence rate after incomplete surgery which is around 25% (4). However, complete tumor removal may not be possible in some circumstances, i.e., tumor proximity to vital structures or involvement of these structures. Also, less aggressive surgeries can be considered to preserve the quality of life in benign tumors like chondroblastoma. In these cases, adjuvant RT can be given. There is no clear information in the literature, possibly due to small sample size, about the use of adjuvant RT in case of residual disease. In fact, it is not recommended to be given due to malignant transformation risk (4). In a review conducted in 2020, only 11 of the 100 cases had a history of RT. Proton therapy combination with photon therapy were given in one patient and targeted biologic therapy using denosumab was given in one other patient (2). In a series of three cases in 2020, chondroblastoma were treated with a functional surgical approach. One case had recurrence after 125 months of follow-up and was treated with RT successfully (gamma knife radiosurgery; maximum dose of 23 Gy). No recurrence occurred in the following 100-month period (total follow-up period was 225 months) (7). However, in our case, total removal was not possible and control MRI showed residual tumor growth thus warranting RT as the best alternative treatment modality with many reports showing its effectiveness in tumor growth control (9).

A wait-and-see policy was adopted because of the possible side effects of RT, but because the patient came from countryside, close follow-up was recommended, but could not be done. This case is also important in terms of showing the importance of close follow-up in patients with residual tumors.

In this case report, we presented the long-term outcome of a young male with temporal bone chondroblastoma who received RT after the regrowth of the subtotally resected tumor. Clinical results favor RT, although this does not come to mean that RT can replace the surgical mode of treatment. Surgical treatment is still favorable wherever total excision is feasible.

Conclusion

Chondroblastoma is a rare tumoral formation seen in the mastoid bone in the first few decades. Although complete surgical resection is still the gold standard today, the success of subtotal resection followed by adjuvant RT should also be kept in mind for cases where total excision would pose high morbidity. It should be considered that hearing was sacrificed to RT and RT carries a potential risk of malignancy although tumor control was achieved for a long time in our case.

Informed Consent: Written informed consent for the publication of this report was obtained from the patient.

Peer-review: Externally peer-reviewed.

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Surgical and Medical Practices: B.S., E.Ç., Concept: B.S., Design: B.S., Data Collection and/or Processing: F.C.A.Ö., E.Ç., U.B., M.B., Analysis and/or Interpretation: F.C.A.Ö., U.B., M.B., Literature Search: F.C.A.Ö., Writing: F.C.A.Ö., B.S.

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Main Points

- Chondroblastoma is a tumor rarely seen in temporal bone in childhood.
- Total surgical resection is the main treatment.
- Although complete surgical resection is still the gold standard today, the success of subtotal resection followed by adjuvant radiation therapy should also be kept in mind for cases where the total excision would pose high morbidity.

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Glass Particles in the Nasal Cavity for 30 Years and Squamous Cell Carcinoma: Is There a Relationship?

Case Report

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Abstract

Nasal cavity tumors constitute a very small part of head and neck malignancies. Although paranasal sinus tumors due to the presence of backward foreign bodies, neoplasms of nasal cavity associated with a foreign body are extremely rare. In this article, we presented a rare case of carcinoma in the right nasal cavity which includes glass particles inside it, and the role of glass particles in carcinogenesis was discussed. The patient was a 55-year-old male with history of a car accident 30 years ago. During right medial maxillectomy via a right lateral rhinotomy approach, three pieces of glass beads, approximately 0.5 cm in size, were removed from the inside of the mass. The patient had also under gone postoperative radiotherapy. No complication emerged during the postoperative recovery period. The patient had been followed up with no finding of local recurrence for 12 months.

Keywords: Carcinogen, foreign body, glass particles, nasal cavity, neoplasm

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Introduction

The nasal cavity and paranasal sinus tumors constitute 0.3% of all malignant tumors and 3% of all head and neck cancers. The most common localizations of paranasal sinus tumors are maxillary sinus and nasal cavity respectively (1). The most common pathological diagnosis is squamous cell carcinoma (SCC). The most important factors responsible for the development of SCC are infections such as papilloma virus and exposure to occupational carcinogens like nickel, leather dust, wood shavings, and chromium. Besides these factors, chronic inflammation is also blamed although there is no consensus (2-5). Foreign bodies may play a role in the carcinogenesis depending on chronic inflammation (6, 7). Thanks to microinvasive technics like endoscopy, the rate of the comorbidities related to surgical interventions and surgical stres was declined and they enabled early diagnosis and treatment of foreign bodies. Foreign bodies in the nasal cavities are infrequent. The aim of this article is to present a rare case of SCC in the right

nasal cavity which includes glass particles inside it and to discuss the role of glass particles in carcinogenesis.

Case Presentation

A 55-year-old man had referred to our clinic with symptoms of nasal obstruction, foul-smelling nasal secretion, and right-sided facial pain. He described a long time history of nasal obstruction and foul-sniffing discharge. His medical history was insignificant except for the use of antihypertensive drugs for his cardiovascular problems, a car accident 30 years ago that caused the loss of his left eye ball, and a scar tissue formation over his nasal dorsum.

Endoscopic examination revealed a fragile mass easily bleeding in bilateral nasal cavities, which completely obstructed the right nasal cavity and partially obstructed the left nasal cavity.

Sampling of the lesion in the right nasal cavity was performed in the outpatient clinic and the histopathologic result was SCC. The preoperative computed tomography displayed a solid enlarged mass occupying the whole right nasal cavity and extending to the contralateral side at the medial nasal region. The medial wall of the maxillary sinus and inferior and medial walls of the orbit were intact. Signs related to sinusitis were observed in the right maxillary, frontal and, ethmoid sinuses (Figure 1 and 2). Magnetic resonance imaging (MRI) of the paranasal sinuses and positron emission tomography with computed tomography of the whole body were performed to assess metastases. The mass was extending to the left nasal cavity without any relevance of the regional skin, lymph nodes, intracranial invasion, and distant metastases.

Operation was carried out under general anesthesia with orotracheal intubation. Complete resection of the mass with right medial maxillectomy was performed via a right lateral rhinotomy approach. During the operation, we observed that the mass, which originated from right lateral nasal wall, infiltrated the nasal septum and extended into both nasal cavities. The right maxillary sinus, right inferior and medial orbital walls and mucosa of the left nasal cavity were intact. During the operation, 3 pieces of glass beads, approximately 0.5 cm in size, were removed from the inside of the mass (Figure 3 and 4). The intraoperative frozen section examination showed that the surgical margins were tumor-free. The mass was diagnosed as a poorly differentiated SCC with a pathological stage T2 N0 M0 according to the tumor node metastasis system published by the American Joint Committee on Cancer (2012). Our patient had also under gone postoperative radiotherapy. No complication emerged

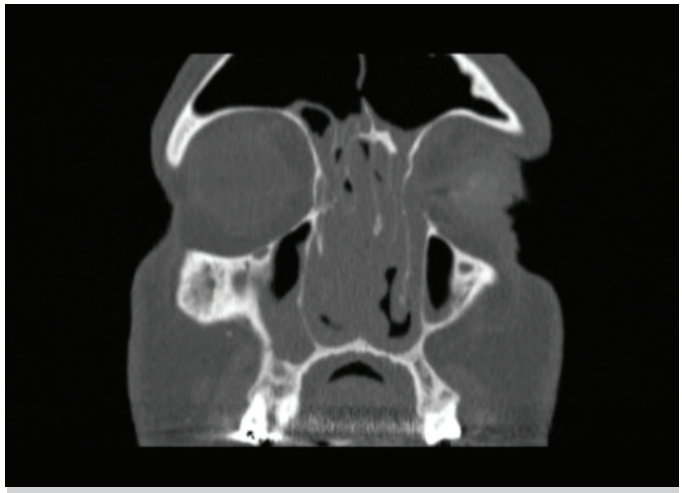

Figure 1. Preoperative computed tomography scan: soft tissue fills the nasal cavity in coronal section

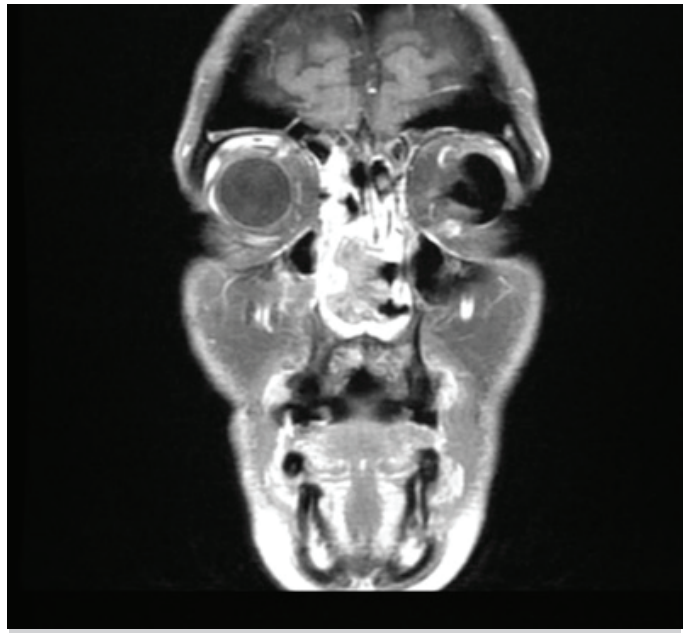

Figure 2. Preoperative magnetic resonance imaging: soft tissue seen in the nasal cavity on T1 weighted coronal section

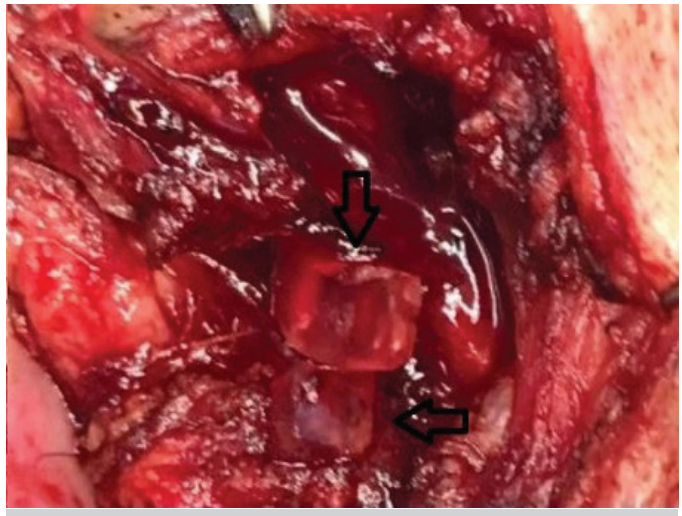

Figure 3. Photo taken during surgery: glass particles located in the tumor, marked with arrows

during the postoperative recovery period. The patient had been followed up with no finding of local recurrence for 12 months. On the MRI finding, six months after the surgery, a soft tissue mass without contrast enhancement which fills the cavity of the right maxillary sinus interpreted as mucosal thickness due to postoperative and post-radiotherapy changes (Figure 5). T2 weighted MRI, 12 months after the surgery revealed bilateral mucosal thickening in maxillary sinuses (Figure 6).

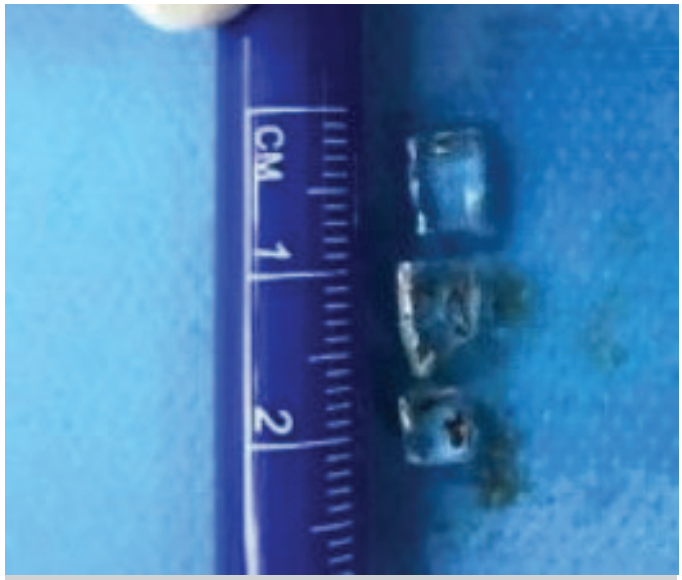

Figure 4. Photo taken during surgery: glass particles after resection

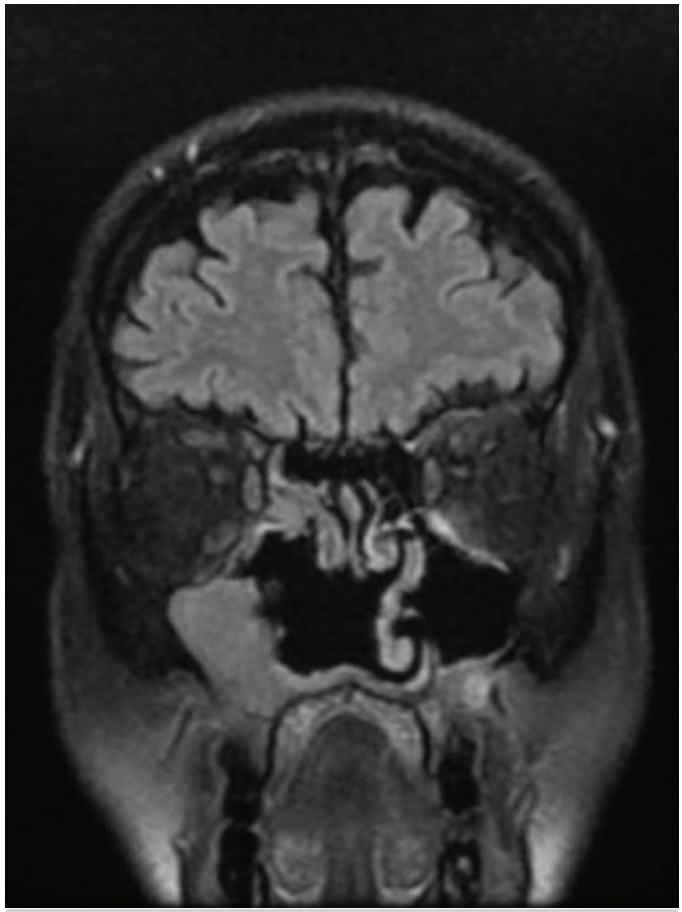

Figure 5. Magnetic resonance imaging at postoperative 6th month: mucosal thickness in the cavity of the right maxillary sinus seen on T1 weighted coronal section

Discussion

Chronic inflammation is one of the most important causes of the development and progress of tumors. Inflammation is a certain malignency-causing ingredient as revealed by accumulated basic, clinical and epidemiological works. It is frequently induced by infectious agents (5). It develops due to the infiltration of the active phagocytes/lymphocytes followed by the stimulation of the stroma reaction, which is induced by the cells related to angiogenes particularly fibroblasts and causes DNA damage and genetic mutation in the normal cells. The underlying pathogenesis in common is to induce and/or maintain inflammation (5). This process leads to uncontrolled cell proliferation, which explains the inflammation-dependent carcinogenesis (5).

Prosthesis, foreign bodies penetrating into the body during accidents and wars may cause chronic inflammation and lead to inflammation-related carcinogenesis (8). It was reported that tumor development may emerge even 20 years after the penetration of the foreign body in the organism (8). A patient with breast malignancy following breast augmentation with liquid silicone injection after sixteen years was reported in a recent paper (9). The carcinogenic effect may change depending on the shape, dimension of the implanted foreign body, roughness of the surface, electrostatic load and inflammatory properties of the host (10). In their animal experiment, Boone (6) and Boone and Jacobs (7) used glass particles and plastic plaques to investigate the carcinogenic effect of foreign bodies. In one of these animal studies, each of nine subjects were subcutaneously inoculated with

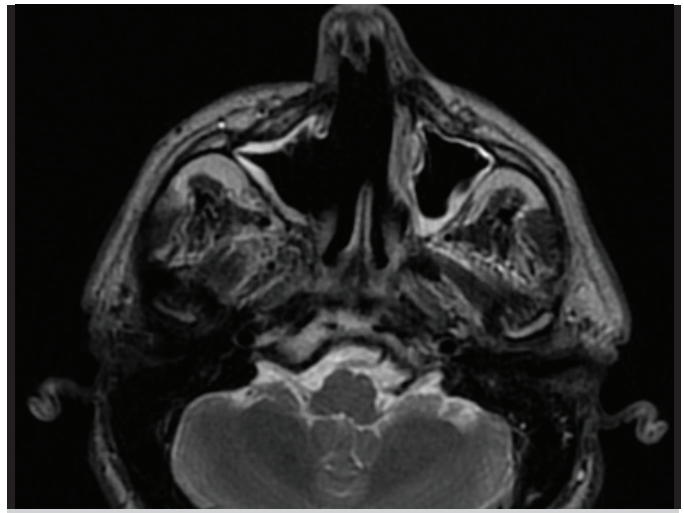

Figure 6. Magnetic resonance imaging at postoperative 12th month: bilateral maxillary sinusitis seen on T2 weighted axial section

an average of 15,400 Balb/3T3 cells attached to two glass particles. After eight weeks, all the subjects had developed large bloody masses that microscopically diagnosed to be hemangioendotheliomas. The inoculation of Balb/3T3 cells alone or particles alone produced no masses (6). Except for this animal study, we did not find any other report about malignancy cases related glass grains in the English literature. In this case report, we described the case of a right nasal cavity tumor which may have been caused by glass particles that had penetrated the nasal cavity in a car crash 30 years ago.

Conclusion

Although there are foreign body-related tumor cases and nasal cavity/paranasal sinus tumor cases in the literature, the presented case is interesting because of the nature of the foreign body and its location. A literature search showed that even biocompatible materials may cause inflammation-dependent carcinogenesis. In addition, foreign bodies may cause additional comorbidities like an infection. In such cases, detailed examination for the presence of foreign body and removal immediately when detected may be useful for the prevention of carcinogenesis and infections.

Informed Consent: The informed consent was obtained from the patient.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: S.Y., P.S.Y., A.V., L.Ş., Concept: S.Y., P.S.Y., A.V., L.Ş., Design: S.Y., P.S.Y., A.V., L.Ş., Data Collection and/or Processing: S.Y., P.S.Y., A.V., L.Ş., Analysis and/or Interpretation: S.Y., P.S.Y., A.V., L.Ş., Literature Search: S.Y., P.S.Y., A.V., L.Ş., Writing: S.Y., P.S.Y., A.V., L.Ş.

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

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Main Points

- Glass particles are among the rarest of foreign bodies reported in English literature to cause carcinogenesis.
- An experimental study about the development of malignancy due to glass particles have been reported.
- Detailed examination for the presence of foreign body and removal immediately when detected may be useful for the prevention of carcinogenesis and infections.

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