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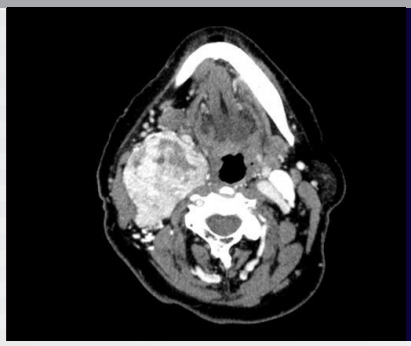

The Importance of Prostate-Specific Membrane Antigen Expression in Carotid Body Paragangliomas

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Original Investigations

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Owner and Responsible Manager on behalf of the Turkish Otorhinolaryngology Head and Neck Surgery Society / Türk Kulak Burun Boğaz ve Baş Boyun Cerrahisi Derneği adına Sahibi ve Sorumlu Yazı İşleri Müdürü: Özgür YİĞİT • Publication Type: Yerel süreli / Local periodical • Basım yeri / Printed at: Matsis Matbaa Hizmetleri San. ve Tic.Ltd.Şti, Tevfikbey Mah., Dr. Ali Demir Cad. No: 51, 34290 Sefaköy, Turkey (+90-212-624 21 11) • Basım tarihi / Printing Date: Eylül 2020 / September 2020 • Türk Kulak Burun Boğaz ve Baş Boyun Cerrahisi Derneği tarafından yayımlanmaktadır. Published by Turkish Otorhinolaryngology Head and Neck Surgery Society, Çobançeşme Sanayi Cad. No: 11 Nish İstanbul A Blok D: 8 Yenibosna, İstanbul, Turkey (+90 212 233 11 26)

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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. The journal is released at three-month intervals, in March, June, September and December, and one volume of the journal comprises four issues. 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 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readers and experts concerning the published studies, articles about general practice and subject of the journal with historical content, memories of scientific significance, educative and catechetical manuscripts about medical deontology and publication ethics.

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 (doaj.org/bestpractice).

Turkish Archives of Otorhinolaryngology is indexed in PubMed Central, PubMed, Web of Science-Emerging Sources Citation Index, TUBITAK ULAKBIM TR Index, DOAJ, EBSCO, CINAHL and ProQuest.

Processing and publication are free of charge with the journal. No fees are requested from the authors at any point throughout the evaluation and publication process. All manuscripts must be submitted via the online submission system, which is available at www.turkarchotolaryngol.net. The journal guidelines, technical information, and the required forms are available on the journal's web page.

All expenses of the journal are covered by the Turkish Otorhinolaryngology Head and Neck Surgery Society. Potential advertisers should contact the Editorial Office. Advertisement images are published only upon the Editor-in-Chief's approval.

All published content is available online, free of charge at www.turkarchotolaryngol.net. Printed copies of the journal are distributed to the members of the Turkish Otorhinolaryngology Head and Neck Surgery Society, free of charge.

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Turkish Archives of Otorhinolaryngology
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Instructions to Authors

CONTEXT

The Turkish Archives of Otorhinolaryngology (Turk Arch Otorhinolaryngol) is an international, 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 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 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 processes of the journal are shaped in accordance with the guidelines of the International Council of Medical Journal Editors (ICMJE), the World Association of Medical Editors (WAME), the Council of Science Editors (CSE), the Committee on Publication Ethics (COPE), the European Association of Science Editors (EASE), and National Information Standards Organization (NISO). The journal conforms to the Principles of Transparency and Best Practice in Scholarly Publishing (doaj.org/bestpractice).

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 that have been 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.

ETHICAL PROCEDURES

An approval of research protocols by the Ethics Committee in accordance with international agreements (World Medical Association Declaration of Helsinki "Ethical Principles for Medical Research Involving Human Subjects," amended in October 2013, www.wma.net) is required for experimental, clinical, and drug studies and for some case reports. If required, ethics committee reports or an equivalent official document will be requested from the authors. For manuscripts concerning experimental research on humans, a statement should be included that shows that written informed consent of patients and volunteers was obtained following a detailed explanation of the procedures that they may undergo. For studies carried out on animals, the measures taken to prevent pain and suffering of the animals should be stated clearly. Information on patient consent, the name of the ethics committee, and the ethics committee approval number should also be stated in the Materials and Methods section of the manuscript. It is the authors' responsibility to protect the patients' anonymity carefully.

For photographs that may reveal the identity of the patients, signed releases of the patient or their legal representative should be enclosed, and the publication approval must be provided in the Materials and Methods section.

PLAGIARISM

The Turkish Archives of Otorhinolaryngology is extremely sensitive about plagiarism. All submissions are screened by a similarity detection software (iThenticate by CrossCheck) at any point during the peer-review and/or production process. Even if you are the author of the phrases or sentences, the text should not have unacceptable similarity with the previously published data.

When you are discussing others' (or your own) previous work, please make sure that you cite the material correctly in every instance.

In the event of alleged or suspected research misconduct, e.g., plagiarism, citation manipulation, and data falsification/fabrication, the Editorial Board will follow and act following COPE guidelines.

AUTHORSHIP

Each person listed as an author should fulfill the authorship criteria recommended by the International Committee of Medical Journal Editors (ICMJE - www.icmje.org). The ICMJE recommends that authorship is based on the following four criteria:

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

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

3. Final approval of the version to be published; AND

4. 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, 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.

The Turkish Archives of Otorhinolaryngology requires corresponding authors to submit a signed and scanned version of the authorship contribution form (available for download through www.turkarchotolaryngol.net) 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.

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.

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. When needed, an ombudsperson may be assigned to resolve claims that cannot be resolved internally. The Editor in Chief is the final authority in the decision-making process for all appeals and complaints.

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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 (updated in December 2019 - http://www.icmje.org/icmje-recommendations.pdf). Authors are required to prepare manuscripts in accordance with the CONSORT guidelines for randomized research studies, STROBE guidelines for observational original research studies, STARD guidelines for studies on diagnostic accuracy, PRISMA guidelines for systematic reviews and meta-analysis, ARRIVE guidelines for experimental animal studies, and TREND guidelines for non-randomized public behavior.

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, and

Instructions to Authors

- 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 www.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 email address of the corresponding author,
- Acknowledgment of the individuals who contributed to the preparation of the manuscript but who do not fulfill 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 three to a maximum of six 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 (https://www.nlm.nih.gov/mesh/MBrowser.html).

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" targeting 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 important 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 Materials and 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).

Editorial Comments: Editorial comments aim to provide a brief critical commentary by reviewers with expertise or with 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, and Tables, Figures, Images, and other media are not included.

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,

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
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

Clinical and Research Consequences, and Conclusion sections. Please check Table 1 for the limitations for Review Articles.

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, and 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.

Clinical Images: This 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.

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 labeled (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 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 the shortcomings of original articles should be mentioned in the Discussion section before the conclusion paragraph.

References

Both in-text citations and the 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.

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 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.

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

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

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

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

Manuscripts Published in Electronic Format: Morse SS. Factors in the emergence of infectious diseases. Emerg Infect Dis (serial online) 1995 Jan-Mar (cited 1996 June 5): 1(1): (24 screens). Available from: URL: http:/ www.cdc.gov/ncidodlElD/cid.htm.

REVISIONS

When submitting a revised version of a paper, the author must submit a detailed "Response to the reviewers" that states point by point how each issue raised by the reviewers has been covered and where it can be found (each reviewer's comment, followed by the author's reply and line numbers where the changes have been made) as well as an annotated copy of the main document. Revised manuscripts must be submitted within 30 days from the date of the decision letter. If the revised version of the manuscript is not submitted within the allocated time, the revision option may be canceled. If the submitting author(s) believe that additional time is required, they should request this extension before the initial 30-day period is over.

Accepted manuscripts are copy-edited for grammar, punctuation, and format. Once the publication process of a manuscript is completed, it is published online on the journal's webpage as an ahead-of-print publication before it is included in its scheduled issue. A PDF proof of the accepted manuscript is sent to the corresponding author and their publication approval is requested within 2 days of their receipt of the proof.

WITHDRAWAL POLICY

Out of respect to the reviewers, journal staff and the Editorial Board, authors are asked to submit a withdrawal request only if the reasons are compelling and unavoidable.

Withdrawal requests should be submitted in written form, signed by all contributing authors of the manuscript. Reasons for withdrawal should be stated clearly. Each request will be subject to the Editorial Board's review and manuscripts will only be assumed withdrawn upon Editorial Board's approval.

PEER-REVIEW AND ETHICS

Peer-review

Editorial policies of the journal are conducted according to the rules advised by Council of Science Editors and reflected in the Uniform Requirements for Manuscripts Submitted to Biomedical Journals: Writing and Editing for Biomedical Publication (http://www.icmje.org/).

Submitted manuscripts are subjected to single-blind peer-review. The scientific board guiding the papers' selection to be published in the Journal consists of elected experts of the Journal and if necessary, selected from national and international experts in the relevant field of research. All manuscripts are reviewed by the editor, section associate editors and at least three internal and external expert referees. All research articles undergo review by a statistical editor as well.

The authors of the accepted manuscripts should be in consent that the editor and associate editors could make corrections without changing the main text of the paper. Manuscript format should be in accordance with Uniform Requirements for Manuscripts Submitted to Biomedical Journals: Writing and Editing for Biomedical Publication (http://www.icmje.org/).

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Instructions to Authors

Ethics

For the experimental, clinical and drug human studies, approval by ethical committee and statement on the adherence of the study protocol to the international agreements (Helsinki Declaration revised 2008 (www.wma.net/e/policy/b3.html) are required. In experimental animal studies, the authors should indicate that the procedures followed were in accordance with animal rights (Guide for the care and use of laboratory animals, www.nap.edu.catalog/5140.html) and they should obtain animal ethics committee approval. The Ethics Committee approval document should be submitted to the Turkish Archives of Otorhinolaryngology together with the manuscript.

The approval of the ethics committee, statement on the adherence to international guidelines mentioned above and that the patients` informed consent is obtained should be indicated in the `Material and Method` section and is required for case reports whenever data/media used could reveal the identity of the patient. The declaration of the conflict of interest between authors, institutions, acknowledgement of any financial or material support, aid is mandatory for authors submitting a manuscript and the statement should appear at the end of the manuscript. Reviewers are required to report if any potential conflict of interest exists between reviewer and authors, institutions.

Plagiarism: To re-publish whole or in part the contents of another author's publication as one's own without providing a reference.

Fabrication: To publish data and findings/results that do not exist. Duplication: Use of data from another publication, which includes re-publishing a manuscript in different languages.

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Turkish Archives of Otorhinolaryngology
Türk Otorinolarengoloji Arşivi

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Editorial

Call for Emergency Action to Limit Global Temperature Increases, Restore Biodiversity, and Protect Health

Wealthy nations must do much more, much faster

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The United Nations General Assembly in September 2021 will bring countries together at a critical time for marshalling collective action to tackle the global environmental crisis. They will meet again at the biodiversity summit in Kunming, China, and the climate conference (COP26) in Glasgow, UK. Ahead of these pivotal meetings, we—the editors of health journals worldwide—call for urgent action to keep average global temperature increases below 1.5°C, halt the destruction of nature, and protect health.

Health is already being harmed by global temperature increases and the destruction of the natural world, a state of affairs health professionals have been bringing attention to for decades (1). The science is unequivocal; a global increase of 1.5°C above the pre-industrial average and the continued

loss of biodiversity risk catastrophic harm to health that will be impossible to reverse (2, 3). Despite the world's necessary preoccupation with COVID-19, we cannot wait for the pandemic to pass to rapidly reduce emissions.

Reflecting the severity of the moment, this editorial appears in health journals across the world. We are united in recognising that only fundamental and equitable changes to societies will reverse our current trajectory.

The risks to health of increases above 1.5°C are now well established (2). Indeed, no temperature rise is "safe." In the past 20 years, heat related mortality among people aged over 65 has increased by more than 50% (4). Higher temperatures have brought increased dehydration and renal function loss, dermatological malignancies, tropical infections, adverse mental health outcomes, pregnancy complications, allergies, and cardiovascular and pulmonary morbidity and mortality (5, 6). Harms disproportionately affect the most vulnerable, including among children, older populations, ethnic minorities, poorer communities, and those with underlying health problems (2, 4).

Global heating is also contributing to the decline in global yield potential for major crops, falling by 1.8%–5.6% since 1981; this, together with the effects of extreme weather and soil depletion, is hampering efforts to reduce undernutrition (4). Thriving ecosystems are essential to human health, and the widespread destruction of nature, including habitats and species, is eroding water and food security and increasing the chance of pandemics (3, 7, 8).

The consequences of the environmental crisis fall disproportionately on those countries and communities that have contributed least to the problem and are least able to mitigate the harms. Yet no country, no matter how wealthy, can shield itself from these impacts. Allowing the consequences to fall disproportionately on the most vulnerable will breed more conflict, food insecurity, forced displacement, and zoonotic disease—with severe implications for all countries and communities. As with the COVID-19 pandemic, we are globally as strong as our weakest member.

Rises above 1.5°C increase the chance of reaching tipping points in natural systems that could lock the world into an acutely unstable state. This would critically impair our ability to mitigate harms and to prevent catastrophic, runaway environmental change (9, 10).

Global Targets Are Not Enough

Encouragingly, many governments, financial institutions, and businesses are setting targets to reach net-zero emissions, including targets for 2030. The cost of renewable energy is dropping rapidly. Many countries are aiming to protect at least 30% of the world's land and oceans by 2030 (11).

These promises are not enough. Targets are easy to set and hard to achieve. They are yet to be matched with credible short and longer term plans to accelerate cleaner technologies and transform societies. Emissions reduction plans do not adequately incorporate health considerations (12). Concern is growing that temperature rises above 1.5°C are beginning to be seen as inevitable, or even acceptable, to powerful members of the global community (13). Relatedly, current strategies for reducing emissions to net zero by the middle of the century implausibly assume that the world will acquire great capabilities to remove greenhouse gases from the atmosphere (14, 15).

This insufficient action means that temperature increases are likely to be well in excess of 2°C (16), a catastrophic outcome for health and environmental stability. Critically, the destruction of nature does not have parity of esteem with the climate element of the crisis, and every single global target to restore biodiversity loss by 2020 was missed (17). This is an overall environmental crisis (18).

Health professionals are united with environmental scientists, businesses, and many others in rejecting that this outcome is inevitable. More can and must be done now—in Glasgow and Kunming—and in the immediate years that follow. We join health professionals worldwide who have already supported calls for rapid action (1, 19).

Equity must be at the centre of the global response. Contributing a fair share to the global effort means that reduction commitments must account for the cumulative, historical contribution each country has made to emissions, as well as its current emissions and capacity to respond. Wealthier countries will have to cut emissions more quickly, making reductions by 2030 beyond those currently proposed (20, 21), and reaching net-zero emissions before 2050. Similar targets and emergency action are needed for biodiversity loss and the wider destruction of the natural world.

To achieve these targets, governments must make fundamental changes to how our societies and economies are organised and how we live. The current strategy of encouraging markets to swap dirty for cleaner technologies is not enough. Governments must intervene to support the redesign of transport systems, cities, production and distribution of food, markets for financial investments, health systems, and much more. Global coordination is needed to ensure that the rush for cleaner technologies does not come at the cost of more environmental destruction and human exploitation.

Many governments met the threat of the COVID-19 pandemic with unprecedented funding. The environmental crisis demands a similar emergency response. Huge investment will be needed, beyond what is being considered or delivered anywhere in the world. But such investments will

produce huge positive health and economic outcomes. These include high quality jobs, reduced air pollution, increased physical activity, and improved housing and diet. Better air quality alone would realise health benefits that easily offset the global costs of emissions reductions (22).

These measures will also improve the social and economic determinants of health, the poor state of which may have made populations more vulnerable to the COVID-19 pandemic (23). But the changes cannot be achieved through a return to damaging austerity policies or the continuation of the large inequalities of wealth and power within and between countries.

Cooperation Hinges on Wealthy Nations Doing More

In particular, countries that have disproportionately created the environmental crisis must do more to support low and middle income countries to build cleaner, healthier, and more resilient societies. High income countries must meet and go beyond their outstanding commitment to provide $100bn a year, making up for any shortfall in 2020 and increasing contributions to and beyond 2025. Funding must be equally split between mitigation and adaptation, including improving the resilience of health systems.

Financing should be through grants rather than loans, building local capabilities and truly empowering communities, and should come alongside forgiving large debts, which constrain the agency of so many low income countries. Additional funding must be marshalled to compensate for inevitable loss and damage caused by the consequences of the environmental crisis.

As health professionals, we must do all we can to aid the transition to a sustainable, fairer, resilient, and healthier world. Alongside acting to reduce the harm from the environmental crisis, we should proactively contribute to global prevention of further damage and action on the root causes of the crisis. We must hold global leaders to account and continue to educate others about the health risks of the crisis. We must join in the work to achieve environmentally sustainable health systems before 2040, recognising that this will mean changing clinical practice. Health institutions have already divested more than $42bn of assets from fossil fuels; others should join them (4).

The greatest threat to global public health is the continued failure of world leaders to keep the global temperature rise below 1.5°C and to restore nature. Urgent, society-wide changes must be made and will lead to a fairer and healthier world. We, as editors of health journals, call for governments and other leaders to act, marking 2021 as the year that the world finally changes course.

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From Past to Present: The Journey of Female Doctors in Medicine and Otorhinolaryngology in Turkey

Historical Article

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Introduction

Patience, diligence, being able to adapt to changing conditions, having good communication skills, being open to innovations, and coping with crises are the almost-innate skills of women. The same features are predictive for success in the field of medicine. However, throughout history, women have been kept away from the field of medicine by claiming that they do not possess the aforementioned features. Further, their prescribed roles were limited to midwives and assistants to male doctors. Although all these, female physicians/surgeons, who have made their mark since ancient times, have existed in Turkey as well as worldwide.

Ancient Times

The lands of the Republic of Turkey have been the cradle of civilizations for centuries, which pioneered the birth of philosophy, art, architecture, and medicine. In ancient times (10,000-5,500 B.C.), people believed that diseases were created by evil spirits. Therefore, clerics or shamans conducted the treatments. Although magic and herbs were often used for treatment, surgical procedures such as trepanation (opening a hole in the skull), possibly aimed at removing evil spirits from the body, were also used to cure diseases. The evidence obtained during excavations has not revealed how and by whom these operations were conducted. However, it is not difficult to guess that these were most likely done by male clerics.

Among Hittites, who ruled in Anatolia from 2,100-1,190 B.C., scribes documented and archived the events of their civilization on clay tablets very

carefully. These documents showed that women also affected the field of medicine. The clay tablets described female doctors with the names Makiya and Mammitum-um-mi. For Hittites, medical treatment was based on herbs, besides fortune-telling and magic. According to some authors, the women in medicine only took part in magic rituals (1). On one of the clay tablets, it was reported that a female doctor/healer probably treated a male with sexual dysfunction through religious ritual prayers (2). Additionally, women were thought to be superior in obstetrics and gynecology. They helped during births as midwives and cast spells to facilitate the birth process. Spells and prayers made for the baby to lead a healthy and good life after birth were also made by midwives. According to the inscriptions, the Hittites also documented abnormal births and interpreted abnormal findings as harbingers of future events. One tablet said: "in the absence of the right ear; the enemy will claim the King's land and in the absence of the left ear; the King will claim his enemy's land." That is interesting, as we know that microtia is more common in the right ear and in boys.

The records of the Eastern Roman period (395-1071 A.D.) stated that despite mainly male doctors working in hospitals established in Istanbul, a female doctor and a female surgeon were also present. Interestingly, 12 assistants and eight support staff were employed to assist male doctors, whereas this number was four assistants and two support staff for female doctors. This finding indicated discrimination against female doctors at that time, which continues today (2).

It is impossible to find any documents proving the practice of female doctors in Anatolia from the 11th to the 17th century. However, Al-Zahrawi, who worked in Andalusia in the 10th and 11th centuries and was one of the pioneers of modern surgery, stated that especially gynecological procedures in female patients could not be performed by male doctors and hence needed to be performed by a woman or midwife, who was instructed by a male doctor. Based on this information, it can be suggested that women were only trained based on practice and they were primarily expected to perform gynecological/obstetrical procedures.

Ottoman Period

The records show that after the death of Deniz bin Gazi, the famous inguinal hernia surgeon in the 17th century in the Ottoman Empire, his wife Saliha bint-i Küpeli performed hernia surgery. Saliha bint-i Küpeli continued the surgical procedure learned from her husband and could successfully treated not only patients from Istanbul but patients from all regions dominated by the Ottoman Empire such as Rumelia, Chios, Morea, Erzurum, and İnegöl (3). The records also show that janissaries Mehmed Beşe and Ali Beşe were among her patients. Female surgeon Küpeli Saliha Hatun had to obtain a consent form from all her patients in the Religious Court before performing the procedure like other physicians and surgeons who served in the Ottoman Empire, the consent deed was a document that would protect a physician performing the treatment or procedure from relatives filing blood feuds and blood money lawsuits in the case of any complication. The fee to be received by the surgeon was also recorded. The examination of the Ottoman archives showed clear differences between the fees charged by the same physician for similar procedures. This depended on the difficulty of the procedure or the patient's financial status. While Saliha bint-i Küpeli received between 300 and 3,000 coins for her surgical procedures, Deniz bin Gazi, from whom she learned the profession, received between 400 and 1,000 coins for hernia surgeries. This finding indicated that a surgeon who successfully performed the surgery was valued, regardless of whether it was a woman or a man (4).

Zeynep bint-i Mustafa, who could be considered a contemporary of Saliha bint-i Küpeli, removed a mass in a man's neck in the village Köyviran of the small town Şuhud in 1691 (5).

Apart from the aforementioned rare examples, the woman's place in social life in the Ottoman Empire was limited. No women could work, show herself in public, or own certain rights due to religious and traditional reasons. However, women's movements, which were initiated by educated women in the 19th century in cities such as Istanbul, Thessaloniki, and İzmir, gained momentum during the Tanzimat period. Although women gained certain rights with these efforts, their applications made for admission to medical faculties were evaluated and rejected by the State Council on their decision on May 23rd, 1898 claiming that women could not work as doctors due to the perception that their chastity would be impaired, they would not be able to examine male patients, they would not be eligible to participate in anatomy dissections, and they could not continue their profession after getting married and having children (5).

Women, who wanted to become doctors during this period, received medical education in the United States of America (Chicago University of Illinois), Switzerland (Geneva Medical School), Germany (Würzburg University Faculty of Medicine), and England (London Medical School), then returned to Turkey. However, according to the Ottoman rules, these women could not practice on their own. One of these students, Zaruhi Kavalcıyan, an Ottoman citizen, returned to her country in 1903 following her medical education in the United States and started working next to her father Dr. Serope Kavalcıyan, who was also a doctor. Dr. Kavalcıyan took over her father's patients after he suffered from a stroke, and attended educational activities (6).

The first Turkish female doctor, Hatice Safiye Ali, shared the same fate despite having excellent education. Safiye Ali, who lost her father at a young age and grew up in the mansion of her grandfather Hacı Emin Pasha, who was the shaykh

al-Islam of Mecca, did not give up. She went to Germany with the support and scholarship of the board of education. She was sad that she could not study in her own country, which also had a medical school. However, she stated in her correspondence with Mr. Ali Haydar, Official of the Ottoman Student Delegation, that she was aware she represented Turkish women in Germany and would strive to glorify Turkish women. With this motivation, Safiye Ali graduated from the Faculty of Medicine, Würzburg University, in 1921 and returned to her country. However, Dr. Safiye Ali was not satisfied with her education and returned to Germany six weeks later to specialize in women's and pediatric diseases. Meanwhile, the Republic was not announced yet, but the Grand National Assembly Government of Turkey had been founded. After her application was evaluated, she was granted permission in 1923 to become the first female doctor in Turkey. The permission was named "Mr. Safiye Ali," which perfectly summarized the situation (5).

After that, Safiye Ali opened a clinic in Istanbul but again faced various difficulties. First, patients did not trust a female doctor and did not want to go to her, after which they proposed only to pay half of the examination fee. She was also subjected to negative propaganda from her male colleagues. Safiye Ali continued her work and treated patients free of charge in the Hilal-i Ahmer Ladies' Center Little Children Practice, the Mothers Union, and the Turkish Women's Association Children's Practice. Also, she educated women in the Himaye-i Etfal Society Milk Drop Establishment, where she worked and later managed, in subjects such as the importance of breast milk, baby care, nutrition, and health rules.

Besides being the first woman to teach in a medical school in Turkey (she became the first female lecturer to teach at the first female medical school established within the American College), she was the first Turkish female doctor attending international congresses. The first congress she attended was the International Women Doctors Congress in London in 1924. She was one of 300 female doctors attending from 300 countries. In the same year, she attended a congress in Vienna, and in 1928, she attended two more international congresses in Bologna. In these international congresses, she mentioned the reforms and health services provided for women and children by the newly established republic and its founder Gazi Mustafa Kemal Atatürk (7).

Period of Republic of Turkey

During the years when Safiye Ali started to practice as a doctor, six female students admitted to the medical faculty for the first time, thanks to the efforts of Dr. Besim Ömer Pasha and Dr. Adnan Adıvar, graduated in 1928 and started to practice in a more relaxed atmosphere in the young Republic founded by Mustafa Kemal Atatürk. One of these doctors İffet Naim (Onur) (1906-1995) practiced obstetrics and gynecology received training from Dr. Akif Şakir at Turkey's first Orthopedic Surgery and Pediatric Surgery Clinic, and continued her career as a surgeon. She is one of the founding members of the Turkish Orthopedics and the Traumatology Surgery Association. Fatma Müfide Kazım (Küley) (1899-1985) became an expert in Internal Medicine and Gastroenterology; Hamdiye Abdürrahim (Rauf) Maral (1895-1975) in Dermatology, Physical Therapy, and Radiotherapy; Sabiha Süleyman (Sayın) (1903-1984) in Pediatrics; Suat Rasim (Giz) (1903-1980) in Surgery, especially Thoracic Surgery; and Fitnat Celal (Taygun) (1898-1985) in Surgery. Mrs. Sabiha Süleyman, one of these pioneer women, received a salaried staff position for the first time as a female doctor in 1929 (6). These pioneering women continued their struggle so that other women could get a medical education and set an example for future generations. Meanwhile, thanks to the implemented revolutions, the women in Turkey gained more rights.

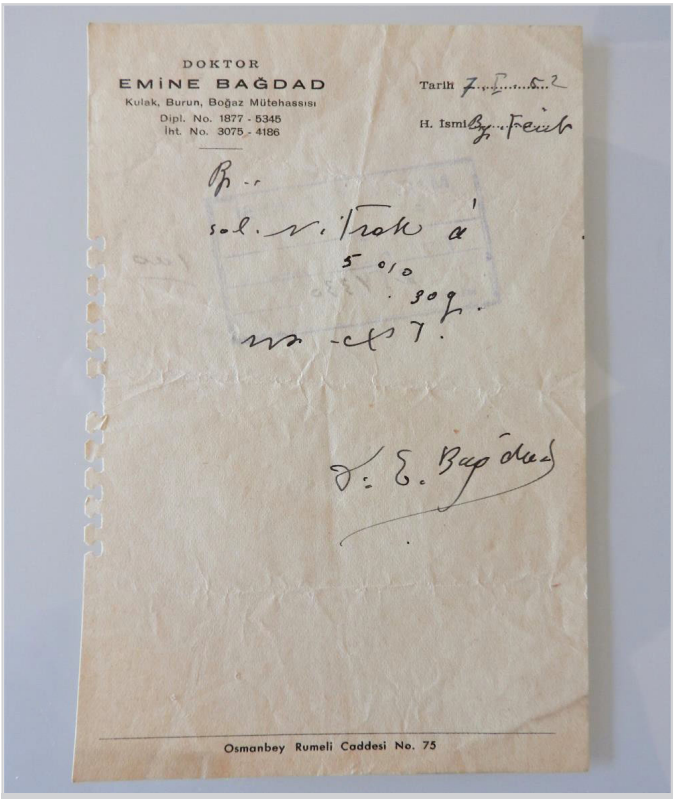
The first female Ear-Nose-Throat (ENT) specialist in Turkey was Dr. Hatice Bodur. Dr. Bodur was initially unaccepted by the medical school and started the Chemistry Department of Istanbul University Science Faculty. Dr. Bodur graduated as valedictorian, worked as a chemist for the next nine years and applied to medical school in 1931. After graduation, she worked as an honorary assistant at the Istanbul University Otorhinolaryngology Clinic between 1940 and 1943, besides her biochemistry education and received the title of otorhinolaryngologist after successfully passing the exam in 1943 (Figure 1) (8).

Dr. Emine Bağdad was the second female otolaryngologist in the young Republic of Turkey, although it was unknown

Figure 1. Hatice Bodur. The first female Ear-Nose-Throat (ENT) specialist in Turkey

when she has obtained her title as a medical doctor and when she received the degree of otorhinolaryngologist. We only have her prescription dated 1952 and her business card (Figures 2 and 3).

Prof. Dr. Nermin Başerer, who is the first female academic in Otorhinolaryngology, was born in 1943. In 1966, she graduated from medical school, which she attended at the insistence of her father, then started her residency in the Otorhinolaryngology Clinic under the guidance of her Physiology instructor. Prof. Dr. Başerer, who decided to leave the department of otorhinolaryngology after only a few months, was motivated by the words of Prof. Dr. Behbut Cevanşir who was head of the department: "… Your report card is full of success, if you work hard and show persistence you will become the first female academician in the field of otorhinolaryngology in Turkey." After this, she successfully finished her training (Figure 4). During 1973-1975, she worked as a foreign chief assistant in Lyon, France, performed successful surgeries, and investigated the effects of psychoactive drugs on the balance system. Prof. Dr. Başerer, who stated with pride that her teacher in France introduced her everywhere as "The daughter of Mustafa Kemal," successfully represented Turkey with her surgeries and work as a woman coming from Atatürk's Turkey.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EMİNE BAĞDAD
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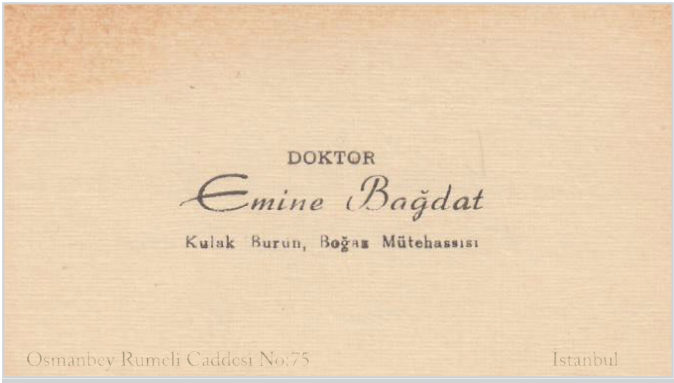
Dipl. No. 1877 - 5345
İht. No. 3075 - 4186
Tarih
H. İsmi
Osmanbey Rumeli Caddesi No. 75

Figure 2. Dr. Bağdad's prescription dated 1952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İstanbul

Figure 3. Dr. Emine Bağdat's business card

Prof. Dr. Nermin Başerer, who became an assistant professor in 1976, successfully served as the first female head of department in Turkey between 1988 and 1991 (Figure 5). Prof. Başerer, who served as the first woman president of the Turkish Otorhinolaryngology Head and Neck Surgery Association founded in 1932, between 2006 and 2008, said the following about her duty: "The fact that I became the first female president of the association was a duty given to me and I accepted that duty. This was not a vision. However, it was gratifying for me to show the whole world, as a Turkish woman, that the medicine in Turkey and that an academician in Turkey can reach this point in a world that is dominated by man. This is a heavy duty but also peaceful." Prof. Dr. Nermin Başerer, who served the field of otorhinolaryngology for 44 years, worked extensively on head and neck cancers and aural atresia and applied the "Başerer shunt" method. She successfully published many scientific articles and books, achieved immense academic success and practiced her profession until she passed away in 2020 (9).

The number of female otorhinolaryngology specialists and academicians, who are on the path opened by Professor Başerer, increases with each passing year; however, it is still relatively low. Although the second female president

Figure 4. Prof. Dr. Nermin Başerer. She was giving a lecture in National ENT Congress in 1969 in İstanbul. It was the first congress she attended.

Figure 5. Prof. Dr. Nermin Başerer. the first female head of Otorhinolaryngology department in Turkey between 1988 and 1991

was not nominated in the history of the Turkish Society of Otorhinolaryngology Head and Neck Surgery (TSO-HNS), sub-association presidencies were successfully carried out by Prof. Dr Nesil Keleş, Prof. Dr Sema Başak, and Prof. Dr Ülkü Tuncer. Moreover, female instructors contributed to our field by serving in several committees of the TSO-HNS.

Prof. Dr. Emine Elif Altuntaş, Prof. Dr. Sema Başak, Prof. Dr. Aslı Şahin Yılmaz, Prof. Dr. Sema Zer Toros, Prof. Dr. Berna Uslu Coşkun, Prof. Dr. Ceren Günel, Prof. Dr. Fatma Tülin Kayhan, Prof. Dr. Ülkü Tuncer, and Prof. Dr. Serap Köybaşı in Qualification Board of TSO- HNS Higher Education Commission; Prof. Dr. Güleser Saylam, Prof. Dr. Emel Çadallı Tatar, Prof. Dr. Müge Özcan, Prof. Dr. Adin Selçuk in Qualification Board of TSO-HNS Examination Board; Prof. Dr. Zeynep Alkan and Prof. Dr. Tuba Bayındır in Qualification Board of TSO-HNS Education Commission; Prof. Dr. Armağan İncesulu and Prof. Dr. Nesil Keleş in Qualification Board of TSO-HNS Executive Board enthusiastically acted in their term.

The Journey of Female Doctors in the World

While these developments occurred in Anatolia, the journey of female physicians in the world was not easy. The acceptance of female students by medical schools in England started only at the end of the 19th century. An interesting example is that of Dr. James (Miranda) Barry. Dr. Barry finished her education in 1812 and worked as a doctor for many years. She even attained the honor in the British army as the Inspector General of Hospitals. Dr. Barry was a woman who was understood only after her death in 1865 (10).

The first woman to be accepted by the "male" medical school in the United States of America was Dr. Elizabeth Blackwell (1821-1910). Blackwell, who graduated in 1949, was accepted by the school only with the approval of the male students. Dr. Blackwell, who was born in England and had to move to the USA for family reasons, was previously rejected by 20 medical schools. Returning to England after her graduation, Dr. Blackwell was the first female member accepted by the General Medical Council in 1858, which allowed doctors to work in England. The fact that it was difficult for women to be accepted at medical faculties has led to the necessity of opening *"female"* medical schools. The New England Female Medical College was opened in 1848, the Woman's Medical College of Pennsylvania was opened in 1850 in the USA, and the London School of Women was opened in England in 1874 (10,11). The Constantinople Women's College, which was opened in Turkey for the same reason in 1920 within the İstanbul American Girls' College, was closed due to the Unification of Education Law of March 3rd, 1924, which prohibited foreigners from establishing and employing higher education institutions (5).

Dolors Aleu i Riera was the first female doctor to receive her doctor's license in Spain in 1882, three years after graduating. The first female doctor in Germany was Dorothea Christiane Erxleben who got her diploma in 1754. Ogino Ginko gained the right to practice as a doctor in 1885 in Japan. The first female doctor in Italy was Dorotea Bucca, who had a chair at Bologna University in 1390 (12).

Margaret F. Butler was the first female doctor in the United States to become specialized in the field of Otorhinolaryngology; she became the department head and a professor in 1906. Besides the surgeries she performed, she also designed different instruments. The first female consultant in the field of Otorhinolaryngology in England was Mrs. Esme Hadfield. The Polish female neurologist Lucia Frey was not an otorhinolaryngologist, but she defined Frey's syndrome in 1923, which was an important breakthrough in related field (10).

Current Situation of Female Otolaryngologists

The journey of women in medicine in Turkey and across the world has not been easy. Although women's movements, which started in the 1800s, gained momentum, the journey was not always easy, especially in surgical branches and otorhinolaryngology. The first study in this regard published in 2019 reported that 53.2% of female ENT specialists were exposed to discrimination during their specialty training. The decision-making positions in universities and professional associations are still "male-dominated." Despite no significant difference in the remuneration of female and male otorhinolaryngologists in Turkey, the salary of women is less than that of men in some countries, including developed ones (13). Among the academic journals in the field of ENT

in Turkey, only the journal *Ear Nose Throat Applications* have a female editor (Prof. Dr. Fatma Tülin Kayhan). Also, none of the world's leading otorhinolaryngology journals employ female editors as the Editor-in-Chief (14).

Conclusion

The admission of women to medical schools or to the surgical field is no longer a fundamental problem, but this area still has some challenges. Sex discrimination still exists when trying to find a place in decision-making positions, in work environments and professional organizations, as well as in academic and senior leadership positions. Steps to be taken in this regard may help reveal the existing potential in women.

Authors Contributions

Conception: S.A.İ., Design: S.A.İ., Supervision: S.B., Data Collection and/or Processing: H.E., İ.H.C., Literature Review: S.A.İ., İ.H.C., Writing: S.A.İ., Critical Review: İ.H.C.

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Turkish Validity and Reliability Study of the Speech, Spatial and Qualities of Hearing Scale

Original Investigation

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Abstract

Objective: The Speech, Spatial and Qualities of Hearing Scale (SSQ) is a self-report scale that evaluates hearing in complex daily life situations in the areas of hearing quality, speech perception, and spatial perception. It is also frequently used in the follow-up of hearing-impaired people, hearing aid and cochlear implant users. It is aimed to translate and adapt SSQ into Turkish, and to investigate its test-retest reliability, and construct validity and reliability, and further to present associations of SSQ scores with the pure tone averages (PTA).

Methods: The Turkish SSQ (Tr-SSQ) scale was administered on 114 adults including those with and without hearing loss. Cronbach's alpha was used to assess its reliability. The reliability coefficient of the scale was calculated by test-retest method. Associations of SSQ scores with PTAs in better and worse hearing ears (BHE and WHE) were evaluated.

Results: Tr-SSQ presented high internal consistency (Cronbach's alpha = 0.984) and test-retest reliability (r=0.994). Tr-SSQ scores were lower in the subjects with hearing loss and correlated with PTAs. Age was found to be correlated with PTAs; regression analysis demonstrated that only WHE-PTA was extracted as explanatory variable for average Tr-SSQ, speech perception and spatial perception scores while both BHE-PTA and WHE-PTA were found to be predictors of hearing quality, but not age for any of Tr-SSQ scores.

Conclusion: Tr-SSQ is a convenient tool for assessing the hearing abilities of individuals with hearing impaired.

Keywords: Hearing loss, self-report, audiology, questionnaire, pure tone audiometry, reproducibility of results

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Introduction

Hearing is one of the most important senses that connect man to the outside world. "Normal" hearing is necessary to ensure adequate and effective communication and adaptation to the environment. Restoration of communication via hearing needs more refined auditory functions and this is the most difficult aspect of

the audiologic intervention and rehabilitation. Routine clinical audiological evaluation for hearing covers pure tone audiometry and speech audiometry including also speech in noise tests, which are the subjective tests; and the objective tests such as (immitancemetric measurements, otoacoustic emissions, and auditory brainstem response tests). By using these assessment methods, audiologists can manage to make diagnosis about the type, degree, and localization of hearing loss and intervention. However, these tests provide limited information about the impact of hearing loss on people and their daily lives (1-4); therefore, intervention strategies recommended to the subjects suffering from hearing impairment may not meet the exact needs of those subjects when only these tests are used.

The self-report scales, which are self-evaluated and graded by the patient about their own illnesses, handicaps and/or health problems have an important role in the evaluation of patients in the field of health (5, 6). These scales contain substances that are standardized in different areas and provide reliable and comprehensive information in the clinic. Furthermore, they ensure that the effectiveness of the therapy and/or treatment is concrete and measurable. Such scales that evaluate complaints about hearing are closely associated with the person's perception of his or her hearing disability and the healing process. Speech, Spatial and Qualities of Hearing Scale (SSQ) was developed by William Noble and Stuart Gatehouse in 2004 to evaluate the sub-components of hearing and quality of hearing in adults and to determine the level of disability perception of the current hearing problem. It is an assessment tool that allows self-evaluation of a wide range of hearing reality in everyday life (7).

The SSQ scale includes 3 sub-scales which are speech perception, spatial perception, and qualities of hearing (7). The developers declare that the first subscale, "Speech", measures the ability to understand, discriminate and follow the speech sounds. As stated by Gatehouse and Noble (7), the second subscale, "Spatial", presents the data about the ability to determine the direction, distance and mobility of the audible voice, and "Qualities" is the third subscale of SSQ which contains items about the identifiability of simultaneous sounds experienced in daily life and provides quantity for the clarity, naturalness, comprehensibility, and effort of hearing. Gatehouse and Noble (7) pointed out that the elements in Qualities subscale were to some extent driven by capacities in the speech and space domains, as well. In each item, complex listening conditions from daily life are described and the person is asked to evaluate his/her hearing by imagining this situation.

The translated versions of SSQ scale have been widely used in the Western languages [in Dutch by (8); in German by (9); in French by (10), in Portuguese by (11)], and in Columbian Spanish by (12), and in the Eastern languages [in Korean by (13); in Malay by (14) and in Persian by (15)]. By using the SSQ scale, the data about the efficacy of amplification by hearing aids, cochlear implants and bone-conducted hearing aids (16), advantages of bilateral hearing aids (17-19), effects of aging on hearing (20-21) have been documented.

Turkish version of SSQ scale has not been developed yet; therefore, the aim of this study is to translate and culturally adapt SSQ into Turkish, and to investigate its test/retest reliability, and construct validity and reliability.

Methods

This study was carried out at the audiology center of a university hospital between December 15th, 2015 and May 25th, 2017. The written informed consent was obtained from all the participants of the study. The study design and the consent form were approved by the Clinical Research Ethics Committee of Gazi University under protocols 77082166-604.01.02.

In this study, the original English version of SSQ scale was used (7). The first step of the process was translation and back translation. Then test validation and the test reliability studies were performed.

In the first step of the study, translation of SSQ into Turkish was done by the first translator, and then the second translator performed the back translation into English. Both translators were bilingual native Turkish speakers. A committee composed of two audiologists and an experienced bilingual translator reviewed the preliminary Turkish version of the SSQ. This version was applied to 20 participants who were randomly selected. Then their recommendations about the clarity, content and order of the questions were examined and Turkish version of the SSQ (Tr-SSQ) was completed by making minor changes in line with these recommendations. The Tr-SSQ, which was named as KUİK (Konuşma, Uzaysal Algı ve İşitme Kalitesi Ölçeği) (Appendix 1) in Turkish, comprised 49 items and 3 sub-scales which are speech perception, spatial perception and qualities of hearing, as in the original one (7). In each item, complex listening conditions from daily life are described and the person is asked to evaluate his or her hearing by imagining this situation. Each item in the scale is scored from "0" to "10"; "10" points indicate that the skill can be performed perfectly in the situation described, "0" point indicates that the described thing cannot be done.

The next step was the construct validity. The reliability coefficient of the scale was calculated by test-retest method. For these steps, Tr-SSQ scale was applied to volunteers aged between 18 and 55 years. The subjects were recruited from the patients suffering from hearing loss in the department, their relatives accompanying them and the staff or students of the university hospital. The literate subjects with normal

otoscopic examination were included. The exclusion criteria for the study were as follows: abnormal tympanogram, conductive and/or mixed type-hearing loss, inadequate cognitive ability to fill the scale, and inability to complete the survey.

At this stage, the participants were informed, a quiet environment was provided, and sufficient time was given for them to fill out the scale. Each participant completed the scale independently. The construct validity of the Tr-SSQ Scale was calculated via factor analysis, and the internal consistency reliability was calculated via Cronbach's alpha (α) coefficient. The reliability coefficient of the scale was calculated via test-retest method. The invariance of the scale according to time was evaluated by using the same method. At this stage, randomly selected 60 participants were re-tested four weeks after the first application of the scale. Pearson test was used for test-retest reliability analysis. In order to test the homogeneity of variances, Levene's test was used. According to the result of the Levene's tests, homogeneity or non-homogeneity of variances for the groups were determined and independent two sample t-test was conducted. By using analysis of variance test, the differences between groups were assessed. Average SSQ score is calculated by summing all item scores and dividing by 49. Speech, Spatial and Qualities scores in the Tr-SSQ are obtained by dividing the total score in each subscale by the number of items in the subscale, which results in14 items for Speech, 17 for Spatial and 18 for Qualities, respectively, as originally described by Gatehouse and Noble in 2004 (7).

The audiological evaluation was performed by using calibrated clinical audiometer (Interacoustic AC-40) with Telephonics TDH 49 headphones in a quiet soundproof room. Pure tone audiometry performed between frequencies 125-8,000 Hz, pure tone average (PTA) was calculated between frequencies of 500-4,000 Hz. While the subjects presenting PTA over 15 dB-HL in one or two ears without any gap higher than 10 dB-HL between air and bone thresholds were included in the sensorineural hearing loss (SNHL) subgroup. Those with PTA lower or equal to 15 dB-HL in both ears were included in the normal hearing (NH) subgroup. Then, PTA of the better hearing ears (BHE) and worse hearing ears (WHE) of each subject were calculated to test the relationship of SSQ score with hearing levels, and to find out the relationship between SSQ scores and audiological variables by using correlation and regression tests.

Statistical analyses were performed using the e Statistical Package for the Social Sciences 21 for Windows (SPSS Inc., Armonk, NY, USA), p-values ≤0.05 were considered significant. The evaluation of distribution of variables was investigated using Kolmogorov-Simirnov/Shapiro-Wilk's tests. As the A correlation analyses were performed between groups of subjects and SSQ score and subscale scores, age, and hearing loss with Spearman correlation test. Linear regression analyses were used to identify predictors of Tr-SSQ scores.

Results

Validity-Reliability

As a result of the factor analysis of the participants' responses to 49 items, it was seen that the responses to the items were expressed with a total of four factors. For each item, four factor values are given in the Table 1.

The questionnaire had a high level of internal consistency with a 0.984 Cronbach's alpha value. In the first column of Table 2, the effect on the scale score mean when the item is removed and in the last column the change of Cronbach's Alpha value when the item is removed are shown. As can be seen, the removal of any items from the questionnaire did not increase the number of Alpha higher than the initial value shown in Table 2.

The reliability coefficient of the scale was calculated by test-retest method. Test–retest interval was four weeks with 60 participants. The Pearson correlation coefficient between the first and second assessment for Tr-SSQ scale scores were r=0.994, p=0.00, for Speech scale r=0.987, p=0.00 for Spatial scale r=0.989, p=0.00 for Qualities scale r=0.982 p=0.00. They were very close to +1. In this case, it has been determined that our scale is a steady and consistent measure which is not based on time. These findings showed that Tr-SSQ scale demonstrated high test-retest reliability and the measurement accuracy of the scale did not change radically over time.

Of 114 participants (female: 62, mean age: 34.9±9.4; male: 52, mean age: 36.3±10.9) enrolled in this study, 53 (female: 27, mean age: 39.3±11.4; male: 26, mean age: 38.3±10.5) were diagnosed with SNHL (bilateral: 40, unilateral: 13); (Table 3), while 61 (female: 36, mean age: 32.5±7.8; male: 25, mean age: 33.0±9.5) had NH in both ears. Means of BHE-PTA and WHE-PTA are presented in Table 3. There was no difference in female/male ratio between the subjects in SNHL and NH subgroups (x^2 test, p=0.544) (Table 3). Age was significantly lower in NH subjects than those with SNHL (Mann-Whitney U test, p=0.002), and age was correlated with both BHE-PTA (r=0.373 p=0.00) and WHE-PTA (r=0.340 p=0.00) in the total group, but not in the subgroups (p> 0.05).

In Table 4, no difference in average SSQ score and subscale scores was detected between males and females in either total group or the subgroups (Student t-test, p>0.05). Mann-Whitney U tests showed that average Tr-SSQ and subscale scores of the NH and bilateral SNHL groups were different, p=0.00 for all pairs. It was observed that average Tr-SSQ, Speech, Spatial and Qualities scores were significantly

Table 1. Factor values of items

Items	Factor					
	1	2	3	4	5	6
Speech-1	0.550	0.455	0.314	0.323	0.173	0.112
Speech-2	0.754	0.286	0.138	0.143	0.083	0.156
Speech-3	0.597	0.456	0.310	0.100	0.102	0.133
Speech-4	0.434	0.686	0.287	0.049	0.189	0.039
Speech-5	0.466	0.639	0.334	0.142	0.186	-0.010
Speech-6	0.238	0.701	0.414	0.249	-0.031	0.123
Speech-7	0.444	0.724	0.263	0.271	0.039	0.052
Speech-8	0.316	0.684	0.227	0.017	0.323	0.040
Speech-9	0.393	0.708	0.245	0.200	0.159	0.121
Speech-10	0.190	0.751	0.267	0.284	0.247	-0.010
Speech-11	0.340	0.733	0.286	0.286	0.032	0.031
Speech-12	0.299	0.744	0.303	0.328	0.117	-0.068
Speech-13	0.758	0.242	0.154	0.242	0.115	0.106
Speech-13	0.207	0.647	0.439	0.334	0.157	-0.097
Spatial-1	0.491	0.336	0.533	0.026	0.378	0.059
Spatial-2	0.559	0.305	0.458	0.063	0.330	0.112
Spatial-3	0.764	0.151	0.362	0.135	0.155	0.124
Spatial-4	0.584	0.236	0.580	0.072	0.236	-0.014
Spatial-5	0.409	0.261	0.636	0.214	0.284	-0.039
Spatial-6	0.368	0.302	0.646	0.196	0.262	0.137
Spatial-7	0.404	0.305	0.508	0.321	0.100	-0.087
Spatial-8	0.248	0.289	0.708	0.291	0.024	-0.047
Spatial-9	0.306	0.288	0.717	0.302	0.051	0.005
Spatial-10	0.188	0.318	0.808	0.233	0.012	-0.064
Spatial-11	0.169	0.434	0.777	0.206	0.026	0.075
Spatial-12	0.466	0.234	0.671	0.121	0.169	0.172
Spatial-13	0.458	0.202	0.701	0.153	0.210	0.211
Spatial-14	0.128	0.153	0.227	0.300	0.746	0.114
Spatial-15	0.193	0.231	0.369	0.561	0.392	0.203
Spatial-16	0.153	0.126	0.279	0.628	0.308	0.339
Spatial-17	0.367	0.289	0.531	0.545	0.101	0.094
Qualities-1	0.796	0.203	0.321	0.123	0.200	0.048
Qualities-2	0.298	0.306	0.043	0.055	0.631	-0.260
Qualities-3	0.709	0.379	0.151	0.196	0.272	0.010
Qualities-4	0.776	0.265	0.309	0.200	0.133	0.156
Qualities-5	0.139	0.034	0.057	0.066	-0.028	0.836
Qualities-6	0.770	0.178	0.313	0.164	0.091	0.160
Qualities-7	0.615	0.420	0.380	0.111	0.107	-0.035
Qualities-8	0.647	0.356	0.372	0.384	-0.003	0.044
Qualities-9	0.685	0.330	0.254	0.454	-0.014	0.011

Table 1. contiuned

Qualities-10	0.650	0.350	0.253	0.520	-0.044	-0.126
Qualities-11	0.516	0.332	0.232	0.591	0.171	-0.059
Qualities-12	0.589	0.212	0.347	0.352	-0.042	-0.169
Qualities-13	0.672	0.448	0.171	0.048	0.257	-0.133
Qualities-14	0.312	0.421	0.270	0.612	0.106	0.054
Qualities-15	0.470	0.432	0.305	0.523	0.105	0.005
Qualities-16	0.692	0.340	0.248	0.446	0.054	-0.101
Qualities-17	0.689	0.307	0.261	0.350	0.120	-0.127
Qualities-18	0.353	0.441	0.277	0.475	0.123	-0.158

higher in NH subgroup than in the subjects with SNHL (p=0.00) for three comparisons. Qualities subscale showed highest score in all three group. Due to the small sample size of the unilateral NH group, no comparison was performed with this group.

The correlation analysis disclosed that age was correlated with SSQ scores in total group (Spearman's test; r=-0.258 (p=0.006) (Table 5), but not in the subgroups (Spearman's test, p>0.05). As presented in Table 5, the SSQ scores in all subjects and those with bilateral SNHL were significantly correlated with PTA values in a negative direction (Spearman's test). In the subjects with NH, WHE-PTA was correlated with total SSQ, Speech and Qualities scores while BHE-PTA was only correlated with Qualities score. Qualities score presented highest correlations in all groups.

Stepwise regression analysis including age, BHE-PTA and WHE-PTA revealed that only WHE-PTA was extracted as explanatory variable for Tr-SSQ (R^2: 0.464; B: -0.036; p<0.0001), Speech (R^2: 0.367, B: -0.4, p= 0.001) and Spatial: (R^2: 0.392, B: -0.34, p=0.002). For Qualities, both WHE-PTA and BHE-PTA were found to be its predictors (R^2: 0.499; for WHE-PTA B: -0.35, p=0.001; for BHE-PTA, B: -0.026; p=0.03). When age was taken out from the independent variable list, no explanatory variable change was seen. When WHE-PTA was taken out, BHE-PTA was appeared as the only explanatory variable for all SSQ scores.

Discussion

The data of this study clearly supports that SSQ can be conveniently used for the assessment of hearing handicap in everyday complex situations in Turkish, as in other languages (7-9, 11-15).

In the adaptation study conducted by Moulin et al. (10), it was determined that the scale had four factors and Cronbach's Alpha coefficient was found to be 0.91. In our study, the validity of the scale was determined by factor analysis in parallel with these studies and it was revealed that 49 items in the scale were expressed with four factors. The internal consistency of the scale was calculated as Cronbach's alpha coefficient and found to be 0.984 or 98.4%. Since this result is higher than the 70% threshold, it can be said that the internal consistency of the survey is quite high. Moreover, in this study, test- retest method was used to determine the reliability of the scale. Reliability is a concept associated with the test-retest sub-assessment and the stability of the tool used. The high value of reliability is one of the important assessment points for any measurement tools. In our study, Tr-SSQ scale was administered to the same participants twice with an interval of approximately four weeks. The correlation between the scores obtained from these two evaluations was analysed and r=0.813 was found (p<0.001), which was in accordance with the original study (7).

In our study, the subjects with NH were younger than those with SNHL, as in the previous studies (8-10). Demeester et al. (8) presented the data of young subjects with normal hearing, the older subjects with clinically normal hearing according to PTA, and the older subjects with hearing loss. Maulin et al. (10) compared difficulty SSQ scores between normal hearing subjects and those with hearing impairment and demonstrated that it is higher in the subjects with hearing loss (mean age: 54.2) then in normal hearing subjects (mean age: 20.8).

Mean average SSQ scores in our study, found in the subjects with bilateral normal hearing and unilateral and bilateral hearing loss (8.1, 7.1, and 6.0, respectively), were in accordance with the previous studies. In the study of Demeester et al. (8) mean average SSQ in young subjects (18–25 years of age) with normal hearing and clinically normal hearing subjects between 55 and 65 years of age were 8.8 and 8.1, respectively. Banh et al. (20) also compared normal hearing young and older adults and reported that younger adults with mean age of 19 years presented higher scores (8.8) than older adults (7.7). Mean of average SSQ in our NH subgroup composed of the subjects aged between 18 and 50 years (mean age: 32.7) was 8.1.

Table 2. Change of Cronbach's alpha value for 49 items in the scale

Item	Total mean when item is removed	Total variance when item is removed	Item total correlation coefficient	Coefficient of multiple determination (R2)	Cronbach's when item is removed
Speech-1	7.10	6703.14	0.851	0.861	0.983
Speech-2	7.13	6771.25	0.717	0.864	0.984
Speech-3	7.14	6730.26	0.780	0.891	0.984
Speech-4	7.15	6715.79	0.786	0.864	0.984
Speech-5	7.16	6679.58	0.834	0.884	0.983
Speech-6	7.17	6694.87	0.768	0.856	0.984
Speech-7	7.16	6672.77	0.850	0.912	0.983
Speech-8	7.15	6741.82	0.698	0.854	0.984
Speech-9	7.15	6708.59	0.803	0.890	0.984
Speech-10	7.17	6683.10	0.755	0.899	0.984
Speech-11	7.16	6677.33	0.806	0.898	0.984
Speech-12	7.16	6660.30	0.819	0.904	0.984
Speech-13	7.14	6741.77	0.746	0.823	0.984
Speech-13	7.17	6678.93	0.790	0.870	0.984
Spatial-1	7.15	6711.14	0.798	0.874	0.984
Spatial-2	7.15	6722.35	0.789	0.881	0.984
Spatial-3	7.14	6727.17	0.777	0.898	0.984
Spatial-4	7.15	6703.04	0.803	0.901	0.984
Spatial-5	7.16	6707.37	0.797	0.900	0.984
Spatial-6	7.15	6697.46	0.801	0.849	0.984
Spatial-7	7.16	6711.24	0.753	0.831	0.984
Spatial-8	7.16	6737.72	0.725	0.871	0.984
Spatial-9	7.16	6727.61	0.777	0.911	0.984
Spatial-10	7.16	6713.50	0.722	0.898	0.984
Spatial-11	7.16	6700.46	0.762	0.929	0.984
Spatial-12	7.15	6737.66	0.790	0.903	0.984
Spatial-13	7.15	6729.10	0.810	0.930	0.984
Spatial-14	7.15	6787.72	0.513	0.723	0.984
Spatial-15	7.16	6749.13	0.681	0.805	0.984
Spatial-16	7.15	6783.46	0.573	0.785	0.984
Spatial-17	7.15	6701.50	0.824	0.899	0.984
Qualities-1	7.13	6722.39	0.801	0.916	0.984
Qualities-2	7.15	6808.75	0.468	0.568	0.984
Qualities-3	7.14	6701.35	0.797	0.866	0.984
Qualities-4	7.13	6725.85	0.839	0.898	0.984
Qualities-5	7.13	6866.28	0.173	0.737	0.984
Qualities-6	7.14	6742.43	0.773	0.878	0.984
Qualities-7	7.14	6712.52	0.804	0.886	0.984
Qualities-8	7.14	6685.58	0.870	0.926	0.983
Qualities-9	7.14	6712.25	0.841	0.944	0.984
Qualities-10	7.14	6702.83	0.835	0.952	0.984
Qualities-11	7.14	6706.90	0.811	0.866	0.984
Qualities-12	7.14	6767.23	0.712	0.842	0.984
Qualities-13	7.14	6754.83	0.758	0.885	0.984
Qualities-14	7.16	6665.76	0.752	0.860	0.984
Qualities-15	7.15	6640.46	0.838	0.900	0.983
Qualities-16	7.14	6688.65	0.849	0.921	0.983
Qualities-17	7.14	6700.29	0.813	0.869	0.984
Qualities-18	7.16	6703.15	0.732	0.769	0.984

Table 3. Mean age and audiological values (BHE-PTA and WHE PTA) of the subjects

Groups		Males: Females	Age (years)	BHE-PTA (dB HL)	WHE-PTA (dB HL)
NH subgroup (n=61)		25:36	32.7±8.5 (18–50)	6.4±3.6 (0–14)	8.4±4.2 (0–15)
Subjects with SNHL	Bilateral (n=40)	19:21	39.1±11.0 (18–50)	39.9±18.2 (16–88)	46.4±19.9 (18–90)
	Unilateral (n=13)	08:05	37.8±10.9 (18–50)	36.9±19.9 (5–15)	8.4±4.2 (18–85)
Total (n=114)		52:62	35.5±10.1	18.5±19.3	25.3±22.9

BHE, better hearing ears, n: Number of the subjects, NH: Normal hearing, PTA: Pure tone averages at 5,000 to 4,000 Hz, SNHL: Sensorineural hearing loss, WHE: Worse hearing ears

Table 4. SSQ scores in the study group

		Speech	Spatial	Qualities	Tr-SSQ
Gender	Males	7.2±1.7	7.3±1.7	7.7±1.7	7.4±1.7
	Females	6.7±1.9	6.8±1.7	7.6±1.8	7.1±1.7
NH subgroup (n=61)		7.8±1.4	7.8±1.3	8.5±1.0	8.1±1.1
Subjects with Bilateral SNHL (n=40)		5.9±2.0	5.8±1.7	6.3±2.1	6.0±1.8
All subjects (n=114)		7.0±1.8	7.0±1.7	7.6±1.8	7.2±1.7

BHE: Better hearing ears, n: Number of the subjects, NH: Normal hearing, PTA: Pure tone averages at 5,000 to 4,000 Hz, SNHL: Sensorineural hearing loss, Tr-SSQ: Average score over all items of speech, spatial and quality of hearing scale, WHE: Worse hearing ears

Table 5. Spearman's rho correlations of SSQ scores with age and audiological values

All subjects (n=114)				The subjects with bilateral SNHL (n=40)			The subjects with bilateral NH (n=61)		
	Age	BHE-PTA	WHE- PTA	Age	BHE- PTA	WHE- PTA	Age	BHE- PTA	WHE- PTA
Tr-SSQ	r=-0.258 p=0.006	r=-0.550 p=0.000	r= -0.654 p=0.000	r=0.103 p=0.528	r=-0.453 p=0.003	r=-0.518 p=0.001	r=-0.194 p=0.13	r=-0.231 p=0.073	r=-0.364 p=0.004
Speech	r=-0.212 p =0.024	r=-0.470 p=0.000	r= -0.581 p=0.000	r=0.124 p=0.44	r=-0.470 p=0.002	r=-0.520 p=0.001	r=-0.187 p=0.150	r=-0.234 p=0.069,	r=-0.37 p=0.003
Spatial	r=-0.246 p=0.008	r=-0.514 p=0.000	r=-0.602 p=0.000	r=0.114 p=0.484	r=-0.357 p=0.024	r=-0.460 p=0.003	r=0.147 p=0.259	r=-0.109 p=0.401	r=-0.218 p=0.092
Qualities	r=-0.301 p=0.001	r=-0.605 p=0.000	r=-0,696 p=0,000	r=0.066 p=0.688	r=-0.537 p=0.000	r=-0.602 p=0.000	r=-0.216 p=0.094	r=-0.333 p=0.009	r=-0.438 p=0.000

BHE: Better hearing ears, n: Number of the subjects, NH: Normal hearing, PTA: Pure tone averages at 5,000 to 4,000 Hz, SNHL: Sensorineural hearing loss, Tr-SSQ: Average score over all items of speech, spatial and quality of hearing scale, WHE: Worse hearing ears

Ages of the subjects with SNHL in our study were between 18 and 50 years, and Demeester et al. (8) documented that mean average SSQ score was 7.7 for the subjects with hearing loss between the ages of 55 and 65 years. In the study of Gatehouse and Noble (7) average SSQ score was 5.5 in the subjects with mean age of 71 years. In Iran, average SSQ was found to be 5.1 in the hearing-impaired subjects with a mean age of 62 (15).

Maulin et al. (10) compared SSQ scales in Dutch, German and French and reported that regardless of the language version considered, the pattern of the items was remarkably similar. They pointed out that a question with a lower score in one SSQ language would also have a low score in another language. They reported Qualities subscale as the most difficult and the spatial subscale as the easiest, and the best reproducibility was found for Speech and the worst was seen in Qualities. In the study of Demeester at al. (8), the highest score was obtained from Qualities subscale in all groups, like our findings. Noble and Gatehouse (17) also documented that the highest score in the subjects with SNHL was found in Qualities subscale. Although Speech presented the lowest score in both the studies of Demeester et al. (8) and Noble and Gatehouse (17) values of Speech and Spatial were very close to each other in our study.

Moulin and Richard (22) reported that correlation between BHE-PTA and total SSQ score were r=-0.56, speech r=-0.57, spatial r=-0.47, qualities r=-0.49, WHE-PTA and total ssq r=-0.52, speech r=-0.43, spatial r=-0.56, qualities r=-0.44 in SNHL group. They claimed that BHE-PTA predictor for scale score after regression analysis. According to their results SSQ and subscale scores decreased with increased PTA values. In our study both WHE-PTA (-0.52) and BHE-PTA (-0.45) correlated negatively with SSQ and subscale scores but in contrast to Moulin and Richard (22) WHE-PTA showed higher correlation for hearing impaired groups. NH group also showed significant and negative correlations with WHE-PTA except Spatial subscale. However, compared to hearing-impaired group, the NH group showed the smallest correlation with WHE-PTA (Table 5). This may be due to small changes in NH participants' PTA values (between 0–15 dB). On the other hand, our analyses on NH group BHE-PTA did not show any significant correlation between Tr-SSQ, Speech and Spatial scores except Qualities score. These values support the results of validation study of French version of SSQ (10). They stated that there was no significant correlation between NH's PTA values and SSQ scale/subscale scores. Zahorik and Rothpletz (23) pointed out that even young normal-hearing listeners did not necessarily rate their listening abilities at the top of the ability scale. As pointed out above, Demeester et al. (8) and Banh et al. (20) reported worse SSQ scores in the older subjects with clinically normal hearing than younger adults with normal hearing. Previously, Banh et al. (20) looked for correlations of SSQ scores with bilateral PTA and Words-in-Noise test thresholds in the normal hearing groups composed of younger and older adults, and reported that the younger adults showed significant correlation only between Speech and Words-in-Noise test thresholds while a positive correlation between Spatial and bilateral PTA was seen. The correlations we observed between SSQ scores and PTA could be either due to our normal hearing subgroup composed of the subjects between the ages of 18 and 50 years or the use of WHE-PTA and BHE-PTA instead of mean PTA values of the subjects.

Since age was also different between NH and SNHL subgroups, step-wise regression analysis was performed and it was seen that WHE-PTA value were predictors for SSQ score. Only Qualities subscale revealed both BHE-PTA and WHE-PTA as the predictors. Age was not observed as a predictor of SSQ scores in any setting.

In our study regression analysis and correlations showed that WHE-PTA was the stronger predictor than BHE-PTA. As expected, we found that as WE-PTA increased, the scale scores decreased. Noble and Gatehouse (24) researched the interaural asymmetry of hearing loss and they demonstrated that average SSQ score was negatively correlated with WHE-PTA (-0.40) and BHE-PTA (-0.43) in the subjects with symmetric hearing loss (24) which was in accordance with our data. But, since our unilateral sample was small, our data in this study was not useful to evaluate the interaural asymmetry.

The major limitation of our study, since number of individuals in the group with unilateral hearing loss was small, the scale and subscale scores in the unilateral subjects were not compared with other groups. As known, unilateral hearing loss has an important negative effect on hearing perception of the subjects in everyday life. For revealing this aspect future studies are necessary. Furthermore, the average age of the normal hearing was lower than in SNHL groups, and there were apparent correlations between age and PTA values in the better and worse ears; correspondingly age was negatively correlated with SSQ scores. However, age was not found as an explanatory variable of Tr-SSQ. These data also support that Tr-SSQ is directly related with only WHE-PTA, not in direct variables. Therefore, although age difference between the study groups was a drawback of this study, Tr-SSQ is a capable scale presenting directly hearing reality in everyday life.

Conclusion

In line with the other versions of SSQ in English, Dutch, German, French, Korean, Portuguese, Persian, Malay and Columbian Spanish, our data supports that Turkish version of SSQ (Tr-SSQ) is a convenient and reliable scale to screen hearing impaired people within the society before inviting to them to the clinics for audiological evaluation and to further evaluate the benefits supplied by hearing aids or cochlear implants via speech, spatial and quality aspects of hearing, which are important in daily life. Since all screening tools including screening of hearing loss recently become more and more popular during COVID-19 pandemic, Tr-SSQ would provide a great opportunity not only to audiologists and otolaryngologists but also to all healthcare professionals who are in charge of following hearing disability of the

special groups, such as people who are regularly exposed to noise in work or the elder subjects. However, as pointed out by Maulin and Richard (22), filling out the whole scale that is composed of 49 questions requires substantial cognitive effort and takes time. Therefore, the use of short SSQ form appears to be more optimistic for easy screening.

Ethics Committee Approval: The study design and the consent form were approved by the clinical research ethics committee of Gazi University under protocols 77082166-604.01.02.

Informed Consent: The written informed consent was obtained from all the participants of the study.

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Authorship Contributions

Conception: N.K., G.İ.Ş.K., İ.B., Design: N.K., G.İ.Ş.K., B.G., Supervision: B.G., İ.B., Data Collection and/or Processing: N.K., Analysis and/or Interpretation: N.K., G.İ.Ş.K., Y.K.K., Literature Review: N.K., G.İ.Ş.K., B.G., İ.B., Y.K.K., Writing: G.İ.Ş.K., Y.K.K., Critical Review: G.İ.Ş.K., B.G., İ.B., Y.K.K.

Main Points

- The SSQ Scale is a self-report scale, which evaluates hearing abilities in complex daily life situations.
- The Tr-SSQ Scale is a valid and reliable tool, presenting high internal consistency and test-retest reliability.
- Since the Tr-SSQ is a convenient scale to assess hearing loss, it could also be used for evaluating effectiveness of the hearing aids, cochlear implants, etc.

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Appendix 1. Konuşma, uzaysal algı ve işitme kalitesi (KUİK) ölçeği

KONUŞMA, UZAYSAL ALGI VE İŞİTME KALİTESİ (KUİK) ÖLÇEĞİ

Aşağıdaki soruların amacı günlük işitme koşullarınızdaki farklı durumlarda işitme ve dinleme yeteneğinizi ve deneyiminizi ortaya koymaktır.

Her soru için, soruların karşısında gösterilen, "0" ila "10" aralığındaki ölçeğin herhangi bir noktasını çarpı (x) ile işaretleyin. "10" noktasına bir işaret koyulması, soruda tanımlanan şeyi kusursuz biçimde yapabilir durumda olduğunuz; "0" noktasına bir işaret koyulması ise tanımlanan şeyi yapamayacak durumda olduğunuz anlamına gelir.

Örneğin, 1. soruda televizyon açıkken aynı anda biriyle sohbet edilmesi ile ilgili bir soru yöneltilmektedir. Eğer bunu yapabilecek durumdaysanız, ölçeğin sağ ucuna yakın bir yere işaret koyun. Böyle bir ortamda sohbetin yarısını takip edebilecek durumdaysanız, ortadaki bir noktaya işaret koyun ve diğer durumlarda da aynı yöntemi kullanın.

Tüm soruların günlük deneyimlerinize uygun sorular olduğunu düşünüyoruz, ancak bir soru sizin için geçerli olmayan bir durumu tanımlıyorsa, "uygun değil" (UD) kutusuna çarpı işareti koyun.

Ad Soyad:

Tarih:

İşitme cihazı kullanıyor musunuz?

□ Evet

□ Hayır

Kullanıyorsanız

□ Sağ Kulak

□ Sol Kulak

□ Her iki kulak

Ne kadar zamandır kullanıyorsunuz?

_____ yıldır

_____ aydır

veya _____haftadır

(İki cihazınızı da farklı zamanlarda aldıysanız lütfen belirtiniz)

KONUŞMA ALGISI

Bir kişiyle konuşuyorsunuz ve aynı oda içinde açık bir televizyon var. Televizyonu kapatmadan konuştuğunuz kişinin ne söylediğini takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Sessiz bir salonda bir başka kişiyle konuşuyorsunuz. Karşınızdaki kişinin söylediklerini takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Bir masanın etrafında oturan beş kişilik bir grubun içindesiniz. Bulunduğunuz yer sessiz bir ortam. Gruptaki herkesi görebiliyorsunuz. Sohbeti takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Kalabalık bir restoranda beş kişilik bir grubun içindesiniz. Gruptaki herkesi görebiliyorsunuz. Sohbeti takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Bir kişiyle konuşuyorsunuz. Arka planda fan veya akan su sesi gibi sürekli bir gürültü var. Kişinin söylediklerini takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Kalabalık bir restoranda beş kişilik bir grubun içindesiniz. Gruptaki herkesi göremiyorsunuz. Sohbeti takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Cami ya da tren garı gibi çok yankı yapan bir yerde biriyle konuşuyorsunuz. Karşınızdaki kişinin söylediklerini takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Sesi sizin konuştuğunuz kişiyle aynı tonda olan başka bir kişi konuşurken, biriyle sohbet edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Sesi sizin konuştuğunuz kişiden farklı tonda olan başka bir kişi konuşurken, biriyle sohbet edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Sizinle konuşan birini dinliyorsunuz ve aynı anda televizyondaki spikeri takip etmeye çalışıyorsunuz. Her iki kişinin de ne dediğini anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Birçok kişinin konuşmakta olduğu bir odada bir kişiyle sohbet ediyorsunuz. Konuştuğunuz kişinin ne dediğini takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Bir grup ile birliktesiniz ve sohbet bir kişiden diğerine çok çabuk geçiyor. Her yeni konuşmacının ilk söylediklerini kaçırmadan sohbeti kolayca takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Telefonda kolaylıkla sohbet edebiliyor musunuz? [cihaz kullanmadan, bir ya da iki cihaz kullanarak]

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Telefonda birini dinliyorsunuz ve yanınızdaki kişi konuşmaya başlıyor. Her iki konuşmacının da ne dediğini takip edebilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

UZAYSAL ALGI

Bilmediğiniz bir dış mekanda bulunuyorsunuz. Birinin çim biçme makinesi kullandığını işitiyorsunuz. Nerede olduğunu göremiyorsunuz. Sesin nereden geldiğini anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Birkaç kişiyle bir masanın etrafında oturuyorsunuz veya toplantı yapıyorsunuz. Herkesi göremiyorsunuz. Bir kişi konuşmaya başlar başlamaz o kişinin nerede olduğunu anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

İki kişinin ortasında oturuyorsunuz. Biri konuşmaya başlıyor. Konuşan kişinin solunuzdaki kişi mi yoksa sağınızdaki kişi mi olduğunu bakmadan anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Bilmediğiniz bir evde bulunuyorsunuz. Ev sessiz. Bir kapının gürültüyle kapandığını işitiyorsunuz. Bu sesin nereden geldiğini anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Bir binanın altınızda ve üstünüzde katların olduğu merdiven boşluğundasınız. Başka bir kattan sesler duyuyorsunuz. Sesin nereden geldiğini kolayca anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Dışarıdasınız. Bir köpek yüksek sesle havlıyor. Köpeğin nerede olduğunu bakmadan anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

Kalabalık bir sokağın kaldırımında ayakta duruyorsunuz. Gelen aracın bir kamyon mu ya da otobüs mü olduğunu bakmadan anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

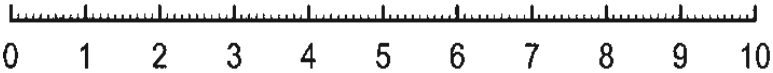
Sokaktayken, yürüyen bir kişinin kendi sesinden veya ayak sesinden o kişinin ne kadar uzakta olduğunu anlayabilir misiniz?

UD □

(Kesinlikle değil) 0 1 2 3 4 5 6 7 8 9 10 (Mükemmel bir şekilde)

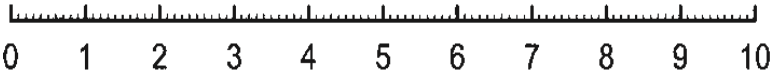
Bir otobüs ya da kamyonun ne kadar uzakta olduğunu sesinden anlayabilir misiniz?

UD □



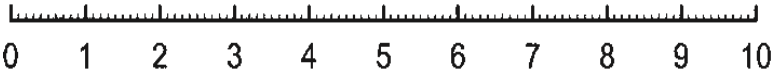
Bir otobüs ya da kamyonun hangi yönde hareket ettiğini sesinden anlayabilir misiniz, örneğin soldan sağa mı yoksa sağdan sola mı hareket ediyor?

UD □



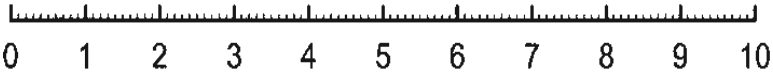
Bir kişinin hangi yönde hareket ettiğini sesinden veya ayak sesinden anlayabilir misiniz, örneğin soldan sağa mı yoksa sağdan sola mı hareket ediyor?

UD □



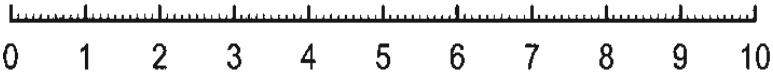
Bir kişinin size doğru mu geliyor yoksa uzaklaşıyor mu olduğunu sesinden ya da ayak sesinden anlayabilir misiniz?

UD □



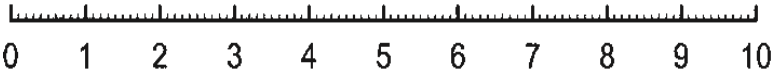
Bir otobüs veya kamyonun size doğru mu geliyor yoksa uzaklaşıyor mu olduğunu sesinden anlayabilir misiniz?

UD □



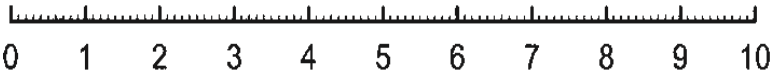
Duyduğunuz sesler size dış dünyadan değil de kafanızın içindeymiş gibi mi geliyor?

UD □



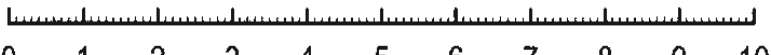
Sesini duyduğunuz ancak ilk başta göremediğiniz kişi veya nesnelere baktığınızda, tahmin ettiğinizden daha yakında olduğunu mu görüyorsunuz?

UD □



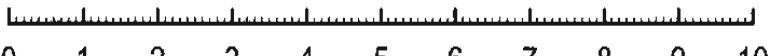
Sesini duyduğunuz ancak ilk başta göremediğiniz kişi veya nesnelere baktığınızda, seslerinin tahmin ettiğinizden daha uzakta olduğunu mu görüyorsunuz?

UD □



Seslerin tam olarak tahmin ettiğiniz yerden geldiğini mi düşünüyorsunuz?

UD □



İŞİTME KALİTESİ

İki sesi aynı anda duyduğunuzu hayal edin; örneğin, suyun lavaboya akışı ve bir radyonun çalışı. Bu seslerin birbirinden ayrı olduğunu fark edebilir misiniz?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Aynı anda birden fazla ses duyduğunuzda, bunlar size birbiriyle karışmış tek bir ses gibi mi geliyor?

UD □

(Karışmış) (Karışmamış)

Radyodan müzik sesinin geldiği bir odadasınız. Aynı odada başka biri de konuşuyor. Konuşan kişinin sesini müzikten ayrı olarak duyabilir misiniz?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Bildiğiniz farklı kişileri seslerinden kolayca tanıyabilir misiniz?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Aşina olduğunuz farklı müzik parçalarını birbirinden kolayca ayırt edebilir misiniz?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Farklı sesler arasındaki farkı anlayabiliyor musunuz; örneğin, bir otomobil ile otobüs; tencerede kaynayan su ile tavada pişen yiyecekler?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Müzik dinlerken, bildiğiniz kadarıyla hangi enstrümanların çalındığını anlayabiliyor musunuz?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Müzik dinlerken, sesler net ve doğal geliyor mu?

UD □

(Kesinlikle değil) (Mükemmel bir şekilde)

Günlük hayatta duyduğunuz sesler size net bir şekilde geliyor mu?

UD □

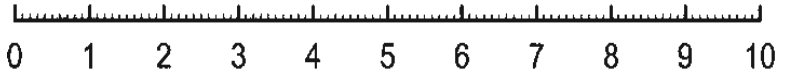
(Kesinlikle değil) (Mükemmel bir şekilde)

Diğer insanların konuşma sesleri size net ve doğal geliyor mu?

UD □

Günlük hayatta duyduğunuz sesler size yapay ve doğal olmayan bir şekilde mi geliyor?

UD □



Konuştuğunuzda, sesiniz kendinize doğal geliyor mu?

UD □

Başka bir kişinin ruh halini sesinden kolayca tahmin edebiliyor musunuz?

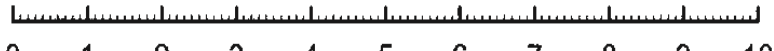
UD □

Bir kişiyi veya şeyi dinlerken çok fazla konsantre olmak zorunda kalıyor musunuz?

UD □

Başkalarıyla konuşurken ne dediklerini anlamak için çok fazla çaba sarf ediyor musunuz?

UD □



Bir arabada sürücü olarak bulunduğunuz sırada, yan koltuğunuzda oturan kişinin ne söylediğini kolayca işitebilir misiniz?

UD □

Yolcu olarak bulunduğunuzda, yan koltuğunuzda oturan sürücünün ne dediğini kolayca işitebilir misiniz?

UD □

Bir şeyi dinlemeye çalışırken diğer sesleri kolayca yok sayabiliyor musunuz?

UD □

Differentiated Thyroid Carcinoma: Distant Metastasis as an Unusual Sole Initial Manifestation

Original Investigation

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Abstract

Objective: The objective of this study was to identify the characteristic features of patients with distant metastasis as the only manifestation of well-differentiated thyroid cancers and to analyze the treatment outcomes.

Methods: A retrospective review of all patients with well-differentiated thyroid cancers and distant metastasis as the sole initial presentation was carried out. Data regarding age, gender, tumor histology, site, symptoms, and treatment outcomes were collected.

Results: There were 10 patients who presented with distant metastasis as the only presentation. The mean age was 56.1 years. Eight (80%) patients had osseous metastasis, one (10%) had pulmonary and one (10%) had both. Follicular thyroid carcinoma was more common and seen in six (60%) patients. Seven (77.8%) out of nine patients had demised within five years of initial presentation.

Conclusion: Distant metastases without a neck lump as the initial presentation of well-differentiated thyroid cancers are extremely rare. No specific guidelines are available to manage such patients due to lack of relevant data in the literature.

Keywords: Differentiated thyroid cancer, neoplasm metastasis, distant metastasis, prognosis, surgery, radioactive iodine, survival

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Introduction

Thyroid cancers account up to 1% to 5% of all cancers worldwide (1). The most common endocrine malignancy is thyroid carcinoma which presents as an enlarging lump in the neck (2). The bulk of thyroid malignancies, including papillary and follicular thyroid cancers, have a well-differentiated form, and usually are clinically indolent with good prognosis (3). The overall survival for 10 years in well-differentiated thyroid cancers (WDTC) ranges from 85% to 95% and drops to 50% in patients with distant metastasis (4).

One of the key factors for poor prognosis is distant metastasis; although patients who have WDTC with distant metastasis have comparatively better survival rates than other forms of thyroid cancers (2). Studies have reported 53% and 58% five-year survival rates, respectively, in patients with differentiated thyroid cancers with distant metastasis (5, 6). This is due to the use of multimodality treatment including surgery, radioactive iodine, radiotherapy and surgical removal of metastatic deposit depending on the type and site of the metastasis (5, 7). Most of the times, thyroid carcinoma metastases remain asymptomatic and are only discovered on surveillance or whole-body metastatic work up (2). However, symptomatic distant metastasis can rarely present as an unusual and sole initial presentation of WDTC without any swelling in the neck (2, 8).

Because of the rarity of such cases, there is limited data available on this subject in literature and the course of disease as well as the treatment outcomes in these patients are not well documented. Therefore, we decided to describe the treatment outcomes of such patients treated at our center; hence, the objective of our study was to report and identify the characteristic features of the patients who presented with distant metastasis as the only manifestation of WDTC and analyze the treatment outcomes.

Methods

A retrospective review of all patients treated for WDTC at a dedicated cancer center between 1995 to 2015 was carried out. Approval was obtained from the Institutional Review Board (IRB) of the Shaukat Khanum Memorial Cancer Hospital & Research Centre, Lahore, Pakistan with IRB number EX-15-07-20-01 and written informed consent was taken from all the patients. All patients with cytologically or histologically proven diagnosis of WDTC and presenting with distant metastasis as the sole initial presentation without any complaint of neck swelling or goiter were included. Patients with simultaneous distant metastasis and thyroid or neck swelling, or those with missing data were excluded. Cervical lymph node metastasis was not considered as distant metastasis. Data regarding age, gender, tumor histology, site, symptoms and treatment outcomes were collected. The data were analyzed using SPSS software v25.0.

Results

The total number of patients with WDTC managed between 1995 to 2015 were 886 (papillary thyroid cancer = 689, follicular thyroid cancer = 114 and medullary thyroid cancer = 83). Out of these, 76 (8.6%) patients presented with metastasis at presentation. Ten patients were identified to fulfill the inclusion criteria of presenting with distant metastasis as the only presentation without any prior history of thyroid or neck lump (Figures 1 and 2). The mean age at presentation was 56.1±10.6 years (range: 36–78 years). There were seven (70%) female and three (30%) male patients.

All patients were symptomatic at the time of presentation, examinations, including fine needle aspiration cytology (FNAC) or incisional biopsies of the metastatic lesion were performed, led to the final diagnosis of differentiated thyroid cancer with distant metastasis. The most common

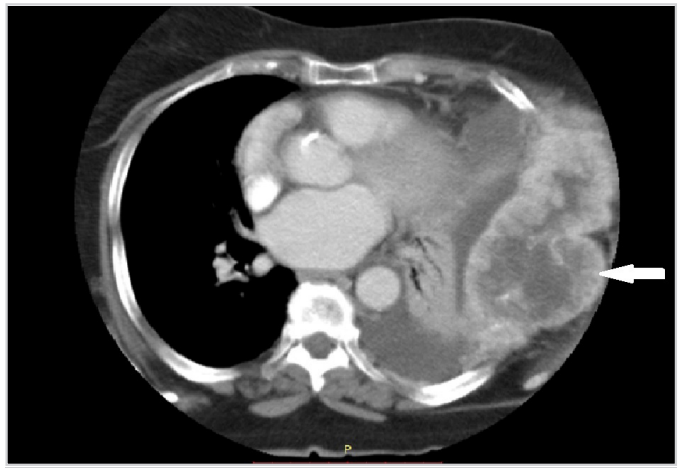

Figure 1. Papillary carcinoma of thyroid with follicular variant. CT of thorax showing the destructive lesion in left lower rib cage with large soft tissue component measuring 13.3 x 6.5cm (arrow)
CT: Computed tomography

Figure 2. Follicular neoplasm of thyroid. MRI of upper limb shows large lobulated soft tissue mass centered on left scapula associated with its erosion and destruction (arrow)
MRI: Magnetic resonance imaging

initial presentation was bone pain or fracture, seen in five (50%) patients, followed by mass or swelling seen in three (30%) patients. Two (20%) patients presented with more than one symptom, one of them presented with hoarseness and difficulty in breathing whereas the other presented with difficulty in breathing along with pain in sternal bone. Eight (80%) patients had osseous metastasis, one (10%) had pulmonary and one (10%) had both pulmonary and osseous metastasis. In nine patients with osseous involvement, the appendicular skeleton was more commonly involved and seen in six (66.7%) patients whereas osseous metastasis was in the axial skeleton in three (33.3%) patients.

Regarding histology, follicular thyroid carcinoma was seen in six (60%) patients of whom five had extracapsular extension and angioinvasion. The remaining four (40%) patients were diagnosed with papillary thyroid carcinoma, two of which were follicular variant. There were six (60%) patients who underwent surgery. Five (50%) patients underwent total thyroidectomy. Of these, one patient also underwent lateral neck dissection. There was one patient who underwent wide excision of sternal mass. Adjuvant radioactive iodine (RAI) was given to six (60%) patients (200 mci in three patients and 150 mci in the other three patients); out of these, four (66.7%) patients received multiple sessions of RAI (200 mci in two patients and 150 mci in two patients). Post therapy thyroglobulin and antithyroglobulin levels were raised in four patients. Eight (80%) patients received radiation therapy and majority of them (n=5, 62.5%) were given radiation with a palliative intent.

All patients were followed up regularly, except for one patient who was lost to follow-up. Mean follow-up period was 41.7 months (minimum one month and maximum 156 months). Seven (77.8%) out of nine patients died within the five years after initial presentation. Except for one case, all deaths were related to the disease process. The characteristics of all patients included in the study are provided in Table 1.

Discussion

WDTCs limited to thyroid gland have an excellent survival outcome, with a 10-year disease-specific survival outcome reported up to 90% (9). Even in the presence of distant metastasis, it has a better survival outcome compared to the other malignancies with distant metastasis (5, 6). WDTCs are more often limited to thyroid gland only, and distant metastasis is rare, with rates reported from 1% to 15% in the literature (10-12). The prevalence of distant metastasis in WDTC was reported up to 2.2% in a comprehensive Surveillance, Epidemiology, and End Results (SEER) database study (13). The most common sites of distant metastasis are the bones and the lungs, although involvement of other organs is also reported in the data, at a rate less than 5% (4, 8, 12). In our series, eight (80%) patients had osseous metastasis, one (10%) had pulmonary and one (10%)

Table 1. Characteristics of patients with distant metastasis as the sole initial presentation of differentiated thyroid cancers

Seq.	Patient	Presentation	Histology	Metastasis	Management	Outcome	Cause of death
1	68, F	Rib cage swelling	Papillary	Osseous	TT, Adjuvant RAI, RT	Demised at 64 months	Disease
2	36, F	Clavicular swelling	Follicular	Osseous	TT, Adjuvant RAI, RT	Demised at 48 months	Disease
3	48, M	Sternal swelling	Follicular	Osseous	Metastasectomy, Adjuvant RAI, RT	Demised at 6 months	Disease
4	42, F	Femur fracture	Follicular	Osseous	TT, Adjuvant RAI, RT	Alive	N/A
5	56, F	Iliac crest fracture	Follicular	Osseous	TT, Adjuvant RAI, RT	Demised at 156 months	Disease
6	65, F	Pain in scapular region	Follicular	Osseous	Declined treatment of primary tumor, Palliative RT	Demised at 19 months	Disease
7	61, M	Femur fracture	Follicular	Osseous	TT + Lateral ND, Adjuvant RAI	Demised at 1 month	Cardiogenic shock
8	60, M	Pain & lump in clavicular region	Papillary	Osseous	Declined treatment of primary tumor, Palliative RT	Demised at 12 months	Disease
9	61, F	Shortness of breath	Papillary	Pulmonary	RT	Demised at 10 months	Disease
10	64, F	Pain in sternal region and shortness of breath	Papillary	Osseous & pulmonary	Lost to follow-up		

F: Female, M: Male, TT: Total thyroidectomy, ND: Neck dissection, RAI: Radioactive iodine, RT: Radiation therapy, Seq: Sequence

had combination of pulmonary and osseous metastasis, these findings are similar to the data reported in the literature (2, 8).

Distant metastasis at the time of initial presentation has a significant prognostic implication as the 10-year survival rate falls significantly to 50%. Factors including age, gender and distant metastasis are associated with survival outcomes in patients with WDTC (12, 14, 15). Metastasis to organs other than the bones and the lungs is not well understood and usually missed in clinical settings because of the rarity of such cases. Shaha et al. (11) reported a case series of 44 patients, over a period of more than half a century, with distant metastases as the only initial presenting symptom. Since then, the literature regarding this topic has remained scarce.

Shaha et al. (11) reported that acceptable long-term survival outcomes were achieved after adequate treatment of primary tumor and aggressive RAI for metastatic disease. Similar management approach is reported in our series and in the literature (2, 8).

See et al. (2) found that follicular thyroid carcinoma, which has hematogenous spread, was most frequently associated with patients with distant metastasis followed by papillary thyroid carcinoma which has lymphatic spread. Also, they found that papillary thyroid carcinoma was more frequently associated with multiple foci of distant metastasis as compared to follicular thyroid carcinoma. In our series we had similar findings that were found comparable with data available in the literature (11, 16).

Mazzaferri (17) took 40 years of age as cut-off and found that patients younger than 40 had a significantly lower risk of developing distant metastasis compared to patients older than 40. Similar findings were noted in our study. The mean age of our patients at the time of their presentation was 56.1 years and only one patient was younger than 40. Comparable results are seen in studies reported in the literature (2, 8, 18).

Patients with swelling in the neck, an easily noticeable and palpable region of the body, would normally seek early medical advice. In our series, patients did not present to head and neck surgeons due to the atypical initial presentation and usually sought medical advice from other specialty departments for complaints such as bone pain, bone fracture and difficulty in breathing, leading to delays and providing the disease the opportunity to metastasize further and thereby adversely affecting the survival outcomes. Studies in the literature also reported similar trends of presentation, with patients being referred to head and neck surgeons by other specialty departments (2, 8).

The American Thyroid Association (ATA) guidelines are widely accepted and used in the management of non-metastatic WDTC (2). There is, however, lack of consensus as to which management protocols should be opted for patients with metastatic thyroid cancer because of the paucity of data on thyroid cancers with distant metastasis. The management plan for all the patients included in our study was discussed and decided by the Multidisciplinary Tumor Board and we advocate that a similar approach is adopted for all patients with rare and atypical presentation.

Our study has several limitations, namely the small sample size, single institution experience and retrospective study design. We were not able to identify the prognostic and risk factors because of our smaller sample size. Nonetheless, the findings of our study on the characteristic features of a rare entity can help further investigations on the risk and prognostic factors in patients diagnosed with WDTC, a condition which generally is regarded as a relatively favorable condition but may have a poor clinical outcome.

Conclusion

We conclude that distant metastases without a neck mass as the sole initial presentation of WDTC is extremely rare. While follicular thyroid carcinoma is the most common etiology, osseous metastases are seen more commonly. Its prognosis is low. Due to the paucity of data, there are still no guidelines available for the treatment of such patients. We recommend further studies in order to better understand the natural course of the disease and subsequently to formulate the guidelines that will help clinicians devise a correct management plan when dealing patients with thyroid cancers with distant metastases.

Ethics Committee Approval: Approval from Institutional Review Board (IRB) of Shaukat Khanum Memorial Cancer Hospital and Research Centre was taken. IRB number: EX-15-07-20-01.

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

Peer-review: Externally peer-reviewed.

Conflict of Interest: The authors declare no conflict of interest.

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Authorship Contributions

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

Main Points

- The incidence of thyroid cancers is on a rising trend, mainly as a result of the advancement in diagnostic modalities.
- Distant metastasis in well-differentiated thyroid cancers is rarely seen and its presentation without any neck mass is even rarer.
- Here, we report an interesting investigation on the characteristic features of the patients who had presented with distant metastasis as the only manifestation of well-differentiated thyroid cancers, and their treatment outcomes.
- These patients have low prognosis. Due to paucity of data, there are still no guidelines available to treat such patients.

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The Bibliometric Aspects of Case Report/Series in Science Citation Index Otorhinolaryngology Journals

Original Investigation

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Abstract

Objective: The rate of case report/series (CR/S) acceptance by the high impact journals is steadily declining mainly due to low citations. The aim of this study is primarily to investigate the bibliometric aspects of CR/S in the field of otorhinolaryngology (ORL) and secondarily to guide prospective authors as to which type of CR/S have better chances of acceptance and citation in the current publication climate.

Methods: Bibliometric and citation analysis of CR/S published in Science Citation Index (SCI) journals of ORL covering the years of 2012-2016 was conducted.

Results: There were 1332 (8.9%) CR/S among 14900 publications in 11 SCI ORL journals published between January 1st, 2012 and December 31st, 2016. The most common published field and subject were the 'pediatric ORL' (33.2%) and 'rare cases/conditions' (47.1%) respectively. 'General ORL' (5.13) and 'treatment' (4.93) categories had the highest citations. Only 10% of CR/S had ≥10 citations. The mean citation counts were positively correlated with impact factors of journals (r=0.131, p<0.001), mean number of authors (r=0.151, p<0.001), mean number of cases (r=0.192, p<0.001), mean number of references (r=0.315, p<0.001) and mean number of Web of Science visits (r=0.291, p<0.001).

Conclusion: Although CR/S provides low citation rates in ORL SCI journals, they may serve important topics in terms of diagnosis, treatment, or complications. The findings and the main discussions of this study may direct the rationale for the consistent publication of CR/S in the evidence-based medicine era.

Keywords: Bibliometrics, case report, case series, citation analysis, otorhinolaryngology, abstracting and indexing

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Introduction

In the last few decades, the place of case report or series (CR/S) in the era of evidence-based medicine (EBM) has been changing. However, in the past, physicians recognized and consequently treated rare conditions with the help of reports presented by their colleagues (1).

Furthermore, the emergence of CR/S dates back to ancient Egypt and it still has didactic value today likewise those times (2).

Because CR is often considered as being unscientific or of little scientific value, there is still not a consensus on the worth of publishing CR/S in the EBM age (3). The Science Citation Index (SCI), an online database of Web of Science (WoS) platform, calculates the impact factor (IF) of a journal annually according to the number of citations made to the publications of that journal (4). While meta-analysis and prospective randomized controlled studies come to the fore with this concept, CR/S, which are at the bottom of the hierarchy of evidence, have lost their importance to a great extent (1, 3). Since IF is determined by the number of citations the publications receive, editors tend to publish as few CR/S as possible in their journals (5-7).

In this study, we aimed to make quantitative and qualitative analysis of the articles in the category of CR/S published in 16 SCI journals of Otorhinolaryngology (ORL) field with respect to their bibliometric characteristics and citation data. As a secondary aim, we tried guiding authors about the current trend in the acceptance of CR/S in high impact ORL SCI journals.

Methods

The citation analysis with bibliometric characteristics of CR/S, which were published in ORL journals indexed in SCI category during the period January 1st, 2012 and December 31st, 2016 was conducted. Any study, conducted for citation analysis, is better designed to include the data of the second-third year prior to the date of the study as the most recently, to be able to collect citations. That was the reasonableness of the time period choice in this study. In order to design a non-biased journal sample, we preferred to select ORL journals listed in 'SCI', which was the actual journal listing for high-impact journals at the time of the study.

Total number of articles in each journal and the number of CR/S were manually collected by screening journal issues. We defined CR/S by looking at how the journal classified the paper on its official website. If that knowledge was not available, the article was defined as CR/S or not, according to the methodology of the paper. The following types of texts were excluded; e-report article only, proceedings article, early printing, conference notes, meeting/congress abstracts, comments, reflections, highlights, corrections, historical reports, specific focus sections, erratum, corrigendum, discussions, biographic items, retracted papers, clinical practical guideline supplements, meeting/conference programs, editorials, list of reviewers and invited editor talks. The clinical problem solving and video cases were excluded as well.

After listing the articles, the two authors reviewed data separately. They included the reports published under the journals' category of 'case report' or 'case report/case series', plus, when the journal was lack of this classification; they included the study if study design was stated as CR/S in the article.

Research Sample

There were 16 SCI indexed ORL journals between 2021-2016. Among the journals, 'Dysphagia', as it is more specific to pharyngo-esophageal disorders, and 'Journal of the Association for Research in Otolaryngology (JARO)', as it publishes particular type of scientific reports, were excluded. The journal of 'Otolaryngologic Clinics of North America' was also excluded because it only publishes review-type articles. There were 1,336 (8.4%) articles under CR/S category (among 16,207 articles) in 13 SCI ORL journals covering the period 2012-2016. Additionally, the journals of 'Hearing Research' (with only one multifamily study) and 'Ear and Hearing' (with only three case studies) were excluded as well. After exclusions, there remained 1,332 (8.9%) CR/S articles from 51 countries (among 14,900 papers in 11 journals).

Before conducting analysis regarding citations, 9 papers (three CR/S with 11 cases, one with 12 cases, two with 15 cases, one with 16 cases, one with 18 cases and one with 67 cases), which have case numbers above 10, were extracted to let the rest of the articles more likely be defined as case reports.

Categorization of Bibliometric Features

The subspecialties of ORL were categorized as otology, rhinology, general ORL (benign ORL disorders other than malignant diseases: infections, benign head and neck tumors, thyroid disease and obstructive sleep apnea), head and neck oncology, laryngology, pediatric ORL (reports of cases under the age of 18 years) and facial plastic surgery. There were only two CR/S in the field of facial plastic surgery; therefore, they were placed to the section of rhinology. According to their subjects, CR/S were observed in the categories of rare condition/disease/anatomy, etiology/symptomatology, foreign body/trauma, diagnostic tool/examination, medical or surgical treatment, complications and unexpected conditions, pathology, genetics and audiology. To provide a more accurate analysis, the types of study subjects were classified under four main categories particularly for this study; rare cases (rare condition/disease/anatomy, foreign body, trauma, etiology/symptomatology), diagnosis and examination (diagnostic tool/examination diagnosis, pathology, genetics and audiology), treatment, and finally complications.

Citation Analysis

The WoS database was evaluated in terms of citations between October 20th and 26th, 2019. Typing the whole title of the manuscript at the section of 'title' on the search page, the data provided by the database were collected. The count of citations and WoS visits were obtained from the citation report of WoS. The values of IFs for 2018/2019 periods were noted from both the journal websites and the http://www.bioxbio.com/if/ on 10th of December 2019 (8).

Statistical Analysis

The relationship of correlation between variables was evaluated by Spearman's correlation test. Continuous variables were also compared among different groups using Mann-Whitney U or Kruskal-Wallis tests. Cross-tabs with likelihood ratios were used to analyze categorical variables among the different groups. All statistical analyses were performed using the SPSS 20.0 software package (IBM® SPSS® Statistics 20.0, Armonk, N.Y., USA). The results were evaluated with 95% confidence intervals, and $p<0.05$ was considered as statistically significance level.

Ethics committee approval was not obtained for this study, as no human subject was included in this bibliometric research.

Results

A total of 14,900 articles were published in 11 SCI ORL journals within a five-year period covering 2012-2016, of which 1,332 were (8.94%) CR/S. The bibliometric characteristics of journals regarding CR/S are given in Table 1. The trend for publications of CR/S according to the years is shown in Figure 1.

The journals with CR/S publication rates of more than 10% were as follows; 'Head & Neck' (16.5%), 'International Journal of Pediatric Otorhinolaryngology' (14.8%), 'Annals of Otology, Rhinology & Laryngology' (13.6%) and 'Otology & Neurotology' (11%).

The most common subspecialty of ORL was the pediatric ORL (n=442, 33.18%), followed by otology (n=284, 21.32%),

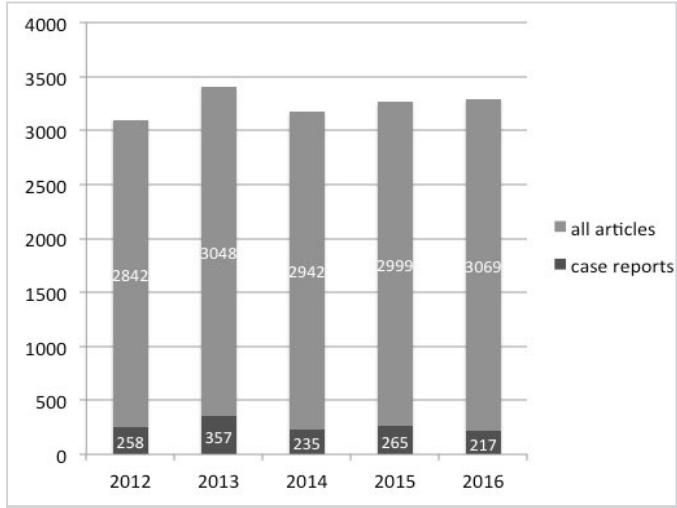

Figure 1. The number of case report/series among all articles and trends in years covering 2012-2016

Table 1. Frequency findings of the case report/series with respect to the numbers, percentage among all articles, year of publication, institution and department of the first author of the study

Journal name*	Total article number (n)	Case report n (%)	Year					Institution			Department	
			2012	2013	2014	2015	2016	University	General	Private	ORL	Other
Acta Otolaryngol.	1,022	8 (0.78%)	7	0	0	1	0	5	3	0	6	2
Am J Rhinol Allergy.	657	3 (0.46%)	0	0	0	2	1	3	0	0	3	0
Ann Otol Rhinol Laryngol.	765	104 (13.59%)	14	18	24	29	19	84	19	1	100	4
Audiol Neurootol.	209	12 (5.74%)	3	2	5	1	1	11	0	1	12	0
Clin Otolaryngol.	620	22 (3.55%)	6	7	3	3	3	21	0	1	22	0
Head Neck.	1,589	262 (16.49%)	19	96	36	51	60	241	13	8	147	115
Int J Pediatr Otorhinolaryngol.	2,275	337 (14.81%)	51	70	81	86	49	295	30	12	283	54
JAMA Otolaryngol Head Neck Surg.	1,205	66 (5.48%)	24	9	16	10	7	55	2	9	57	9
Laryngoscope.	3,028	201 (6.64%)	38	73	29	33	28	191	6	4	192	9
Otolaryngol Head Neck Surg.	1,782	126 (7.07%)	43	33	19	23	8	122	4	126	125	1
Otol Neurotol.	1,748	191 (10.98%)	53	49	22	26	41	181	10	0	182	9
Total	14,900	1332 (8.94%)	258	357	235	265	217	1209	87	162	1129	203

N: North, ORL: Otolaryngology, S South, n: Number

*Abbreviated name of the journals

head and neck oncology (n=250, 18.77%), general ORL (n=208, 16.62%), rhinology (n=83, 6.23%) and laryngology (n=65, 4.88%) respectively (Table 2). The category of rare cases (47.1%) and treatment (26.8%) were the most common main subjects (Table 3).

The only significant contingencies were between the subspecialty of the ORL and subject of the CR/S (p<0.001). Laryngology was out of the usual trend and the CR/S about the treatments and complications in this field were more common than the other subspecialties of ORL. Most (>50%) of CR/S were about rare cases in the fields of Otology, Rhinology, General ORL and Pediatric ORL.

Of 1332 CR/S, 258 (19.37%) failed to have citations. The percentage of CR/S, which had only one citation at the time of this study, was 21.02% (n=280). One hundred twenty-four (9.31%) CR/S had ≥10 citations. The highest mean citation counts were belonging to 'Head and Neck' and 'American Journal of Rhinology and Allergy', which were 5.69 and 5.67, respectively (Table 4). 'American Journal of Rhinology and Allergy' seems to be the most citation-effective journal with lowest number of CR/S but highest number of citations.

International journals tended to publish CR/S not only from the American or European countries, but also from worldwide. USA (48%) stood first with the highest number of CR/S publications in this study, followed by Japan (6.1%), Italy (4.7%) and Canada (4.1%).

Table 2. Frequency findings of the case report/series with respect to the fields of Otorhinolaryngology

	Subspecialties of ORL, n (%)					
Journal name*	**Otology**	**Rhinology**	**GORL**	**HNO**	**Laryngology**	**Ped. ORL**
Acta Otolaryngol.	7 (87.5%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	1 (12.5%)
Am J Rhinol Allergy.	0 (0.0%)	2 (66.7%)	0 (0.0%)	1 (33.3%)	0 (0.0%)	0 (0.0%)
Ann Otol Rhinol Laryngol.	33 (31.7%)	5 (4.8%)	22 (21.2%)	10 (9.6%)	13 (12.5%)	21 (20.2%)
Audiol Neurootol.	9 (75%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	3 (25%)
Clin Otolaryngol.	5 (22.7%)	6 (27.3%)	1 (4.5%)	5 (22.7%)	3 (13.6%)	2 (9.1%)
Head Neck.	5 (1.9%)	5 (1.9%)	71 (27.1%)	174 (66.4%)	0 (0.0%)	7 (2.7%)
Int J Pediatr Otorhinolaryngol.	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	337 (100%)
JAMA Otolaryngol Head Neck Surg.	13 (19.7%)	11 (16.7%)	14 (21.2%)	3 (4.5%)	10 (15.2%)	15 (22.7%)
Laryngoscope.	29 (14.4%)	31 (15.4%)	52 (25.9%)	28 (13.9%)	29 (14.4%)	32 (15.9%)
Otolaryngol Head Neck Surg.	20 (15.9%)	23 (18.3%)	42 (33.3%)	15 (11.9%)	10 (7.9%)	16 (12.7%)
Otol Neurotol.	163 (85.3%)	0 (0.0%)	6 (3.1%)	14 (7.3%)	0 (0.0%)	8 (4.2%)
Total	284 (21.3%)	83 (6.2%)	208 (16.6%)	250 (18.8%)	65 (4.9%)	442 (33.2%)

GORL: General Otorhinolaryngology, HNO: Head and Neck Oncology, ORL: Otorhinolaryngology, Ped. ORL: Pediatric Otorhinolaryngology, n: Number

*Abbreviated name of the journals

Table 3. Frequency findings of the case report/series with respect to the subject groups

	Subgroups of Subject, n (%)			
Journal name*	**Rare case**	**Examination**	**Treatment**	**Complication**
Acta Otolaryngol.	3 (37.5%)	1 (12.5%)	4 (50%)	0 (0.0%)
Am J Rhinol Allergy.	2 (66.6%)	0 (0.0%)	1 (33.3%)	0 (0.0%)
Ann Otol Rhinol Laryngol.	55 (52.9%)	11 (10.6%)	29 (27.9%)	9 (8.7%)
Audiol Neurootol.	0 (0.0%)	8 (66.7%)	4 (33.3%)	0 (0.0%)
Clin Otolaryngol.	0 (0.0%)	0 (0.0%)	21 (95.5%)	1 (4.5%)
Head Neck.	112 (42.7%)	34 (13%)	99 (37.8%)	17 (6.5%)
Int J Pediatr Otorhinolaryngol.	191 (56.7%)	28 (8.3%)	77 (22.8%)	41 (12.2%)
JAMA Otolaryngol Head Neck Surg.	38 (57.6%)	10 (15.2%)	7 (10.6%)	11 (16.7%)
Laryngoscope.	97 (48.3%)	8 (4.0%)	82 (40.8%)	14 (7.0%)
Otolaryngol Head Neck Surg.	82 (65.1%)	1 (0.8%)	28 (22.2%)	15 (11.9%)
Otol Neurotol.	163 (85.3%)	14 (7.3%)	5 (2.6%)	9 (4.7%)
Total	743 (55.8%)	115 (8.6%)	357 (26.8%)	117 (8.8%)

n: Number

* Abbreviated name of the journals

Table 4. Descriptive findings of the case report/series with respect to the number of cases, authors, references, citations and Web of Science visits; plus, first ten countries with highest number of report/series publications, citations and citation effectiveness

Journal name*	Impact Factor**	Case (n)		Author (n)		Reference (n)		Citation (n)		WoS visit (n)		The Countries with respect to number of publications	The Countries with respect to number of citations	The Countries with respect to citation effectiveness
		min-max	mean	min-max	mean	min-max	mean	min-max	mean	min-max	mean			
Acta Otolaryngol.	1.286	1-11	2.63	1-6	4.25	7-19	12.75	0-12	4.13	0-6	2.13	(n, MCN)		
Am J Rhinol Allergy.	2.015	7-18	11.33	5-8	6.33	16-19	17	2-11	5.67	1-5	3.33	USA (640, 3.64)	Sweden (16, 5.33)	Scotland (10, 3.5)
Ann Otol Rhinol Laryngol.	1.458	1-8	1.69	2-12	4.47	3-36	16.21	0-16	3.29	0-10	2.01	Japan (81, 3.94)	Germany (26, 5.27)	Sweden (16, 5.33)
Audiol Neurootol.	2.053	1-7	3.75	3-9	6.42	15-55	26.92	1-11	4.58	1-8	3.33	Italy (63, 4.73)	Turkey (24, 5)	Greece (11, 3.45)
Clin Otolaryngol.	2.377	1-9	4.82	1-7	3.82	0-12	7.05	0-6	2	0-6	1.14	Canada (54, 4.13)	Italy (63, 4.73)	Australia (14, 4.29)
Head Neck.	2.442	1-11	1.59	1-13	5.24	1-61	19.09	0-40	5.69	0-42	2.56	South Korea (51, 2.76)	Holland (28, 4.46)	Brazil (13, 3.92)
Int J Pediatr Otorhinolaryngol.	1.225	1-67	1.7	1-11	4.41	3-93	15.78	0-20	3.04	0-26	2.16	China (45, 4.27)	Australia (14, 4.29)	Switzerland (16, 4.06)
JAMA Otolaryngol Head Neck Surg.	3.502	1-16	1.98	1-8	3.83	3-45	10.62	0-13	3.21	0-10	1.29	England (38, 2.89)	China (45, 4.27)	Finland (13, 3.25)
Laryngoscope.	2.343	1-15	1.9	2-11	4.34	3-31	11.52	0-31	4.54	0-80	2.09	India (34, 2,59)	Canada (54, 4.13)	Turkey (24, 5)
Otolaryngol Head Neck Surg.	2.310	1-10	1.55	1-9	3.05	1-10	4.61	0-35	2.13	0-7	0.99	France (32, 3.81)	Switzerland (16, 4.06)	Germany (26, 5.27)
Otol Neurotol.	2.063	1-10	1.26	1-11	4.04	0-50	6.49	0-21	2.24	0-23	1.28	Taiwan (29, 2.52)	Japan (81, 3.94)	Spain (17, 3.41)

max: Maximum, MCN: Mean citation number, min; Minimum, WoS: Web of Science, n: Number

*Abbreviated name of the journals.

** The values are collected from the official websites of journals and http://www.bioxbio.com/if/ on 10th of December 2019

Table 4 shows the first ten countries with the highest number of CR/S publications, citations and citation effectiveness.

The mean citation numbers were statistically different between all subgroups of study subjects and ORL subspecialties ($p<0.05$). New treatments/surgical techniques (4.93) and new examinations/diagnostic tools (4.46) gathered the highest number of citations. The mean number of citations for rare cases and complication topics were 2.96 and 2.86, respectively. The subspecialty of General ORL diseases received the highest number of citations (5.13), followed by rhinology (4.07), head and neck oncology (3.85), pediatric ORL (3.13), laryngology (2.97) and otology (2.85). The first authors of other specialties showed a higher number of citations (5.27 vs. 3.31) than otolaryngologists ($p<0.05$).

There were positive correlations between the value of IFs of the journals with mean citation counts of CR/S ($r=0.131$, $p<0.001$) and mean author numbers ($r=0.061$, $p=0.027$). The mean citation counts were positively correlated with the mean number of authors ($r=0.151$, $p<0.001$), mean number of cases ($r=0.192$, $p<0.001$), mean number of references ($r=0.315$, $p<0.001$) and mean number of WoS visits ($r=0.291$, $p<0.001$) as well.

Finally, descriptive characteristics of the most cited ten CR/S published in the studied five years in SCI ORL journals are summarized in Table 5.

Discussion

There are many bibliometric studies, especially about the citations the articles receive, but this report is detailed for solely case reports in our field. Therefore, the findings of this study bring important perspective to the bibliometrics of the case reports in ORL. The striking point of the present study is the low citation rates that only 10% of CR/S articles in ORL journals of SCI category (published during the period January 1st, 2012 and December 31st, 2016) had ≥10 citations by October 2019.

The case presentations can be stimulating for the reader. When a physician confronts with a collection of history, symptoms and signs; it is human nature for him to consider the most common disorders at first. However, thanks to the similar CR/S reported in literature, one can notice the rarer conditions or figure out the new treatment modalities. Before the modern science age, reporting CR/S was the basic way to document rare conditions and to provide an environment for discussing difficult cases. But, in recent times, the existence of massive information and easy online access to resources has replaced their needs (1).

Nevertheless, there are still conflicts about the definition of case reports and case series or case studies in the literature (5-7). A case report is defined as a detailed description of the experience gathered from a single patient (1, 9). If more than one case is reported, it is called a case series or case series report. They usually consist of 3-10 cases (10). In 2008, Glossary of Epidemiology (11) defined 'case series' as follows; "A collection of patients with common characteristics used to describe some clinical, pathophysiological, or operational aspect of a disease, treatment, or diagnostic procedure." However, the term of 'case reports' was described as; "Detailed descriptions of a few patients or clinical cases (frequently, just one sick person) with an unusual disease or complication, uncommon combinations of diseases, and unusual or misleading SEMIOLOGY, CAUSE or OUTCOME (may be surprising recovery…)" (11). However, in 2001, the Glossary of Epidemiology (12) explained nothing for CR/S, indicating that an epidemiologist had tried defining these terms at the early 2000s (13).

To moderate the above discussions, we suggest the following definitions; case presentation or report is a descriptive article on a single case; whereas, case series is a descriptive article on multiple cases with common rare condition/disease/anatomy/pathology/genetic structure. Apart from them, a case study can be retrospective, cross-sectional or prospectively followed and/or may include further analysis regarding new cases/methodologies/experimental findings or novel treatment techniques. Therefore, if the article is original and analytic; it is not recommended to be identified as CR/S, accordingly any CR/S should not be accepted as an original article if it does not involve the research hypothesis (14). Consequently, the articles with research purposes or retrospective/prospective investigations on a respectable case sample would rather be accepted as case studies than CR/S (15, 16). In the same way, CR/S are preferably unaccepted as analytical studies, but if only the descriptive aspects are specified; then they are identified as CR/S (17). To adhere to the formal categorization of the journals, the articles listed as CR/S or stated as CR/S in their methods section were included in this study.

Even though CR/S have fallen out of favor in the era of EBM, their place in academic medical publication should be promoted as they have potential educational benefits (18). In fact, there was a fluctuating but finally declining trend in publication rates of CR/S in the period of 2012-2016 in this study. We found an incidence of 8.94% for CR/S among articles published in SCI ORL journals in the study period. This ratio corresponds to the recent CR/S publishing tendencies in the field of medicine (5, 6, 18, 19). However, the frequency of CR/S is 15.1% among ORL articles for the period between 1945-2016 (19), which supports the decline in our sample (Figure 1). Some journals, as a rule, do not intend to accept the submission of CR/S at the initial stage or some recommend early rejection or desk rejection. Considering the fast-growing volume of scientific literature and their concern regarding earning citations, it seems that CR/S have lost their value of evidence and become weaker in recent decades (3, 19).

Table 5. The descriptive characteristics of the most cited ten report/series published in SCI ORL journals in the period covering 2012-2016

CR/S Paper		Categorical Variables				Numerical Variables (n)				
No	'DOI' number	Country	Specialty	ORL subspecialty	Subject	Case	Author	Reference	Citation	WoS visit
1	Fully 3-dimensional digitally planned reconstruction of a mandible with a free vascularized fibula and immediate placement of an implant-supported prosthetic construction									
	10.1002/hed.21922	Holland	Oral Maxillofacial	Head & Neck Oncology	Treatment	1	8	16	40	42
2	Mammary analogue secretory carcinoma of the parotid gland in a pediatric patient									
	10.1177/0194599811419044	USA	ORL	Pediatric ORL	Rare Case	1	4	5	35	3
3	Combined transnasal endoscopic and transoral robotic resection of recurrent nasopharyngeal carcinoma									
	10.1002/hed.21731	China	ORL	Head & Neck Oncology	Treatment	1	3	17	31	5
4	Evaluation of adult Pott's puffy tumor: our five cases and 27 literature cases									
	10.1002/lary.23490	Japan	ORL	Rhinology	Rare Case	5	3	31	31	1
5	Combined transoral robotic surgery and endoscopic endonasal approach for the resection of extensive malignancies of the skull base									
	10.1002/hed.23238	USA	ORL	Head & Neck Oncology	Treatment	2	5	44	30	7
6	Definitive treatment of androgen receptor-positive salivary duct carcinoma with androgen deprivation therapy and external beam radiotherapy									
	10.1002/hed.23383	USA	Radiation Oncology	Head & Neck Oncology	Treatment	1	3	9	30	3
7	Human epidermal receptor 2-amplified salivary duct carcinoma:regression with dual human epidermal receptor 2 inhibition and anti-vascular endothelial growth factor combination treatment									
	10.1002/hed.23429	USA	Hemato/Oncology	Head & Neck Oncology	Treatment	1	4	32	30	0
8	Transoral robotic total laryngectomy: report of 3 cases									
	10.1002/hed.23226	Canada	ORL	Head & Neck Oncology	Treatment	3	8	12	28	1
9	Multimodal nonlinear microscopic investigations on head and neck squamous cell carcinoma: toward intraoperative imaging									
	10.1002/hed.23139	Germany	Cell Biology	Head & Neck Oncology	Examination	3	8	43	27	26
10	Detection of evolving injury to the brachial plexus during transaxillary robotic thyroidectomy									
	10.1002/lary.22429	USA	ORL	Head & Neck Oncology	Examination	1	5	10	26	2

CR/S: Case report/series, ORL: Otorhinolaryngology, WoS: Web of Science, SCI: Science Citation Index, n: Number

In this study, the field of General ORL diseases is in the first place for the highest mean citation number, followed by rhinology and head and neck oncology for CR/S. In the study by Coelho et al. (20) the fields, most frequently associated with citation classics, were otology and lateral skull base both in 1999 (48.75%) and 2009 (55.71%). Head and neck oncology, benign head and neck disorders and laryngology remained at similar citation rates in 1999 and 2009. Whereas, the number of rhinology and anterior skull base related citation classics declined in 2009 (10.50%), compared to 1999 (17.50%) for original articles (20). In the report of Lenzi et al. (21), the leading research field was otology/neurotology, followed by rhinology and head and neck surgery, for all article types. It seems that the ORL subspecialty with the most citation potential can vary according to the type of paper.

It has been shown that most of the CR/S in the ORL journals were under the rare cases class (7). On the other hand, only two of the most cited 10 CR/S were included in the category of rare cases. Indeed, 'new treatment' subject was the most frequent among these ten most cited CR/S (Table 5). In line with this finding, the highest citation rates were observed in the field of treatment (4.93+/-5.809) as well. It is an expected result that the case series, which have not yet had a large number of patients enough for reporting a clinical study, but where the results of an innovative surgical technique applied to a reasonable number of cases are reported, will receive more citations.

The most citation-effective publications were from Scotland, Sweden, Greece, Australia, Brazil, Switzerland, Finland, Turkey, Germany and Spain (Holland was the eleventh) in this study. In another study, the country of origin (for all type of publications) influenced the citation possibility and effectiveness as well (21). The journal 'Head & Neck' (16.5%) showed the highest percentage of publishing CR/S and the highest mean citation count of 5.69 in this study.

In the report of Fenton et al. (22) in 2004, the number of citations to CR/S, published from January to December 2000, ranged from 3 to 28 (mean: 12.4) in Auris Nasus Larynx and 2 to 28 (mean: 10.2) in The Journal of Laryngology and Otology. Edelmeyer et al. (19) reported only three CR/S with more than 100 citations and were considered as 'citation classics'. Whereas, 905 citation classics were found in 2014, compared with 80 in 2001, for original articles (19). In this study, the highest citation count was 40 (Table 5).

The study by Laccourreye et al. (23) analyzed the reasons for acceptance and rejection for CR/S in 'European Annals of Otorhinolaryngology Head and Neck Diseases' and reported the most common flaws; in 74.5% of cases, as the lack of originality and lack of new data contributing to the medical literature. The number of CR/S that SCI ORL journals have taken over the course of five years (2012-2016) has also declined a bit in this study. Similarly, as reported by Edelmayer et al. (19) six of the top 10 ORL journals (by IF) did not publish CR/S anymore.

The evaluation of the CR/S in terms of the citations they receive shows that the index of journal plays an important role in the citation gain of the study. The interest of readers to the articles published in the journals that listed in noteworthy indexes would be higher (24). Although it was claimed that the citations received by CR/S, published in an average ORL journal or that of higher IFs, are similarly not high (20), there were positive correlations between the value of IFs of the journals with mean citation counts of CR/S in this study.

In a recently released national paper, Susaman and Erdağ (25) underlined that while the highest number of case reports was in the field of head and neck surgery and in the category of rare diseases and unexpected conditions, citations they received were not noteworthy, which was also not different from the international literature.

Over the last several years, new peer-reviewed journals that exclusively publish CR/S have emerged. These are mostly open-access journals with considerably high acceptance rates (1). Because CR/S do not rank highly in the hierarchy of evidence and are not frequently cited, they are seldom published by high-impact medical journals. Laccourreye et al. (23) showed that 5% of the CR/S submissions was accepted for publication in the journal of 'European Annals of Otolaryngology Head & Neck Diseases', whereas, 7% were not resubmitted by the authors and 88% were rejected.

The development of modern experimental techniques may have lead clinical trials to supersede observations, CR/S in this context. The descriptions of new syndromes and ground-breaking new modalities are not being made lately unless they make novel discoveries that worth citing. However, CR/S type of papers advance medical knowledge and constitute data for EBM, especially when they were combined with follow-up reports. Case reports also provide information on new conditions, examination and imaging procedures, treatment modalities, complications, and side effects, with literature, have also induced the formation of hypotheses for more comprehensive and sophisticated studies (5, 13, 24). Moreover, the positive correlation between the number of cases presented and the number of citations is a critical finding of this study. Perhaps, as the number of cases increases, the more value a CR/S is taken due to the more knowledge and experience reported about these cases in EBM.

The findings of the present study, on the bibliometric and citation journey of CR/S in SCI ORL journals, can be beneficial for the academicians, who are interested in writing CR/S. In fact, case reporting is like writing an academic story of a particular medical condition and an entry pathway for young physicians into the medical publication world. Alternatively, the findings of this study will provide the

editorial boards of the journals with an evidence-based guide to evaluate their attitudes toward CR/S submissions.

Conclusion

In this study, the publication and citation aspects of CR/S in SCI ORL journals were served to literature. The decline in publication and citation rates of CR/S in SCI journals should not discourage the academicians submitting their works, instead they should become aware of the possibly accepted and cited types of CR/S.

Ethics Committee Approval: Ethics committee approval was not obtained for this study, as no human subject was included in this bibliometric research.

Informed Consent: Since this bibliometric research did not include any human or animal subject, informed consent was not obtained.

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Authorship Contributions

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Main points

- Only 10% of CR/S articles in SCI-indexed ORL journals (published during the period January 1st, 2012 and December 31st, 2016) had ≥10 citations by October 2019.
- The most published CR/S were in the field of 'pediatric ORL', whereas 'general ORL' field got the highest citations.
- The most published CR/S were in the category of 'rare cases', whereas 'treatment' category got the highest citations.
- Changes in publication and citation numbers may reflect changing interests in CR/S type of articles in the field of ORL.

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The Importance of Prostate-Specific Membrane Antigen Expression in Carotid Body Paragangliomas

Original Investigation

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Abstract

Objective: Prostate-specific membrane antigen (PSMA) is a transmembrane protein expressed in prostate cancer. It is, however, also expressed in the neovasculature of some non-prostatic solid tumors. Carotid body paragangliomas (CBPs) are highly vascular neoplasms. In this study, we aimed to investigate the possible role of PSMA expression in CBPs. There are no studies in the literature that report to have investigated the relationship between PSMA and CBPs.

Methods: This study is a retrospective analysis of cases diagnosed with CBP based on their demographic, clinical, radiological, surgical and immunohistochemical findings. Immunohistochemical examination results of Ki-67, S100, synaptophysin, chromogranin were retrieved from patient files. Then, the paraffin blocks of CBPs specimens, stained by PSMA-antibody by immunohistochemical methods were examined histopathologically.

Results: The number of patients operated on for CBP was 12 (four men and eight women). Ten out of 12 specimens were suitable for staining and histopathological examination. Capsular and/or vascular invasions of tumors were seen in complicated cases. Intratumoral vascular PSMA expression was seen in all specimens except one. Extratumoral vascular PSMA expression was not detected in any of the cases. Tumoral cell PSMA staining was seen in six of ten cases.

Conclusion: We found higher intratumoral vascular expressions of PSMA nearly in all CBPs, but we could not assess the statistical significance because of the small number of specimens. These data might be a guide for future studies that are planned for either diagnostic or therapeutic approaches to CBPs.

Keywords: Carotid body, carotid body tumor, immunohistochemistry, paraganglioma, prostate-specific membrane antigen, surgery, histopathology

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Introduction

While carotid body paragangliomas (CBPs) are uncommon tumors, they account for the majority of the paragangliomas (PGLs) in the head and neck (60-70%) (1). Patients with persistent hypoxia and complaints like chronic obstructive pulmonary disease, congenital cyanotic heart disease, and

those living in high-altitude areas are at risk (especially women) (2, 3). Immunohistochemistry shows that PGLs are positive for neuroendocrine markers such as chromogranin and synaptophysin (3).

Prostate-specific membrane antigen (PSMA) is a transmembrane protein that is found in the neovasculature of some non-prostatic solid tumors, as well as of prostate cancers. PSMA expression is not only found in prostate epithelial cells, but also in tumor-associated endothelial cells in a variety of solid tumors (4). If surgery is difficult or impossible to perform, *in vivo* PSMA expression in tumors can serve as a deputy biomarker for angiogenesis, and this rationale can be used for non-invasive targeted therapy in solid vascular tumors such as head and neck PGLs (5).

The target is labeled with a diagnostic radionuclide, and once expression is confirmed, the same target is labeled with a therapeutic radionuclide, and the procedure is carried out. Choudhury and Gupta (6) reported the treatment of neuroendocrine tumors with 177-lutetium (Lu-177) by this mechanism. Expressions of new molecules may give rise to new imaging and/or treatment modalities. To that end, PSMA may be a promising novel target in CBPs. We investigated the staining pattern and other findings of CBP cases that were operated on in our clinic. This study is the first to investigate the relationship between PSMA and CBPs.

Methods

The files of 12 patients (four men and eight women) who were diagnosed with CBP and operated on in Süleyman Demirel University Faculty of Medicine, Ear Nose and Throat Clinic from January 2000 to January 2021 were analyzed. The Clinical Research Ethics Committee of Süleyman Demirel University approved our study protocol. (approval date: 11/09/2020; approval number: 254). The study was designed as a retrospective study; therefore, informed consent was not obtained from the patients.

Files of all patients that were diagnosed with CBP were thoroughly examined. The findings retrieved from patient files were: clinical examination results (horizontally mobile but vertically fixed mass in the carotid triangle, brown skin patches, etc.), computed tomography (CT) images, magnetic resonance images (MRIs) of the neck, and angiography of the carotid system, operational findings, and histopathological records. CT, MRI, and angiographic images were re-evaluated from the recordings. Immunohistochemical investigation results of Ki-67, S100, synaptophysin, chromogranin were retrieved from the reports in patient files. All findings were compared and interpreted.

All patients were routinely followed up four times (every three months) in the first year and annually in the following years. The basic follow-up consisted of routine physical examination and MRI. After evaluating all files, paraffin-embedded blocks were taken from the archives of the pathology department. Immunohistochemical staining and examination were done using PSMA antibody.

PSMA Immunohistochemistry

PSMA antibody was applied to the specimen blocks via immunohistochemical method in the hematoxylin and eosin (H&E) stained sections. PSMA antibody (clone 3E6, isotype: IgG1, kappa, DAKO flex ready to use monoclonal mouse antibody provided in liquid form in a buffer containing stabilizing protein and 0.015 mol/L sodium azide; Dako, California, USA) was prepared according to the manufacturer's instructions as expressed in the data sheet. The tissue samples that were obtained from the formalin-fixed paraffin-embedded blocks and taken to four -µm thickness adhesive-coated slides were processed together with prostate tissue sample as antibody positive control and the sample sent alongside as negative control to the automated immunohistochemistry device.

Evaluation of Immunohistochemical Staining

Immunohistochemically stained slides were evaluated by the same experienced pathologist. PSMA immunohistochemically stained slides were examined under light microscope and accepted as positive cytoplasmic staining and/or positive cytoplasmic membrane staining.

The vascular structures in the epithelial and stromal foci of the tumor were identified from H&E-stained tumor sections and taken to the simultaneously stained PSMA. Vascular percentage score (expression score) was defined as the number of cells stained by PSMA throughout counted cells. The intensity score was defined as the staining intensity of the cells examined.

Vascular percentage score was defined as Score 0=0%–5%; Score 1=5%–50%, and Score 2=>50%. The intensity score was defined as Score 0=none-weak, Score 1=moderate, and Score 2=strong. PSMA expression was scored according to the intensity of the tumor epithelium, as Score 0=none-weak, Score 1=moderate, Score 2=strong.

Results

Findings of 12 patients who were diagnosed with CBP (four men and eight women) and operated on in our clinic from January 2000 to January 2021 were analyzed. All demographic, diagnostic, surgical and follow-up results were re-evaluated. Paraffin blocks of ten patients (out of 12) were suitable for PSMA staining and immunohistochemical examination. The age of the patients ranged from 25 to 81 years with a mean of 50 years. The most common initial complaint reported by patients was neck mass. Additionally, four patients had pain, one patient had odynophagia,

one patient had dysphagia. The time between the initial symptoms and patients' presentation to our clinic ranged from one to 60 months. One patient had bilateral, five had left-sided and six had right-sided CBP. The follow-up period ranged from eight months to ten years. There was no recurrence of the disease in the surgically treated patients (Table 1). Angiographically determined blood supply of the tumors are given in Table 2. Three patients had type I, six patients had type II and three patients had type III tumors according to the Shamblin classification (preoperative and postoperative CT imaging of Shamblin type III CBP are shown in Figures 1a and 1b). In terms of complications, one patient had cerebral emboli that needed embolectomy, and one patient had superior laryngeal nerve paralysis. Carotid artery resection and arterial grafting was needed in only one patient. There was one patient with incomplete resection. Data on complications are shown in Table 2.

PSMA Staining Results

Intratumoral vascular PSMA expression was strong in eight patients, moderate in one patient. One patient had no PSMA expression (Figures 2 and 3). There was no extratumoral vascular PSMA expression in any of the cases. Tumoral cell PSMA expressions were mild to moderate in six patients and absent in four patients. Comparisons of PSMA expression

Table 1. Demographic features and clinical presentations of patients with CBP

No	Gender	Age	Complaint	DC	Side of the tumor	Family history	Follow-up period	Recurrence
1	M	54	Mass	6 m	L	No	3 y	No
2	F	55	Mass	48 m	R	No	8 m	No
3	M	44	Mass, pain	6 m	L	No	4 y	No
4	M	52	Mass	4 m	B	No	2 y	No
5	F	42	Mass	6 m	R	No	10 y	No
6	F	53	Mass, pain, odynophagia	5 m	L	No	7 y	No
7	F	62	Mass	3 m	R	No	2 y	No
8	F	32	Mass	12 m	L	No	10 y	No
9	F	25	Mass	8 m	R	No	10 y	No
10	F*	81	Mass, pain, dysphagia	24 m	R	No	6 y	No
11	M	47	Mass, pain	60 m	R	No	5 y	No
12	F	53	Mass	1 m	L	No	7 y	No

M: Male, F: Female, m: Months, y: Years, DC: Duration of complaints, R: Right, L: Left, B: Bilateral.

*Patients with tumor embolization before surgery

Table 2. Radiologic and surgical features of patients with CBP

No	Size of tumor (mm)	Angiography (blood supply)	Shamblin classification	Complications	Arterial grafting	Incomplete surgery
1	28x25x20	ECA	I	No	No	No
2	31x20x19	Ncd	II	No	No	No
3	30x25x20	ICA	II	Cerebral emboli	No	No
4	53x36x35	Ncd	II	No	No	No
5	38x28x20	ECA	II	No	No	No
6	57x44x40	Vertebral - pharyngeal artery & ICA	III	No	No	Yes
7	45x36x32	Ncd	II	No	No	No
8	25x21x20	ECA, ICA	III	SLN paralysis	Yes	No
9	31x26x11	Ncd	I	No	No	No
10	55x52x44	ICA & ECA	III	No	No	No
11	30x25x17	Occipital artery	I	No	No	No
12	44x37x34	Occipital artery	II	No	No	No

ECA: External carotid artery, ICA: Internal carotid artery, SLN: Superior laryngeal nerve, Ncd: Not clearly determined

with the other immunohistochemical stainings (Ki-67, S100, synaptophysin, chromogranin) and capsular and/or vascular invasion are shown in Table 3.

Discussion

CBPs or carotid body tumors are also known as chemodectomas because of the carotid body's physiological function as a chemoreceptor (7). Despite the limited number of cases in our study, characteristics such as age, gender and side of tumor were found compatible with the literature (2, 8, 9). CBPs are common in patients that have been exposed to hypoxia for a long time, as well as in those who have chronic obstructive pulmonary disease, congenital cyanotic heart disease, or live at high altitudes (2). The city where our patients live is situated at a moderately high altitude of 1,000-1,500 meters.

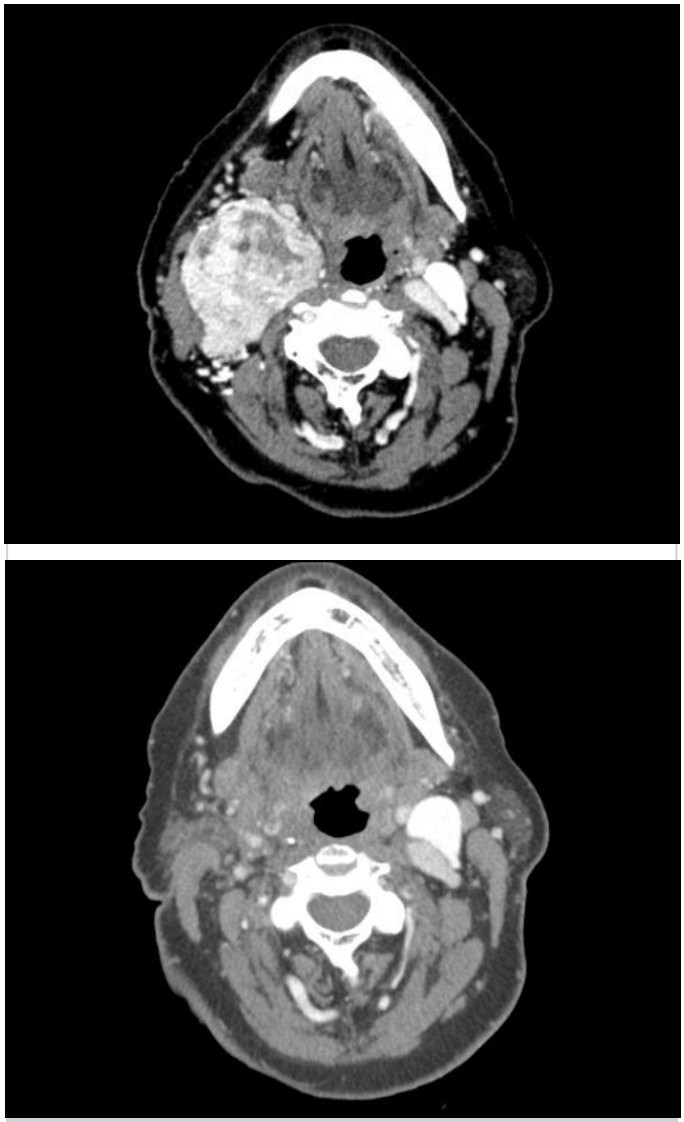

Figure 1. a. Preoperative contrast enhanced CT of the neck demonstrating right-sided Shamblin type III CBP of an 81-year-old female patient; b. Postoperative contrast enhanced CT of the neck

CT: Computed tomography, CBP: Carotid body paraganglioma

The most common complaint of CBP is neck mass. A painless observable pulsatile neck mass, laterally mobile, but vertically immobile, is found only anterior to the sternocleidomastoid muscle, at the plane of the hyoid joint, and under the angle of the mandible on physical inspection (10). The common complaint of all our patients was neck mass. Additional complaints were reported in the clinical course as the tumors grew. Odynophagia and dysphagia were other complaints reported by patients (by one patient each).

The bulk of CBPs are benign, but 6% are cancerous. Malignancy is defined by the dissemination of cancerous cells to the lymph nodes or distant metastases (7). One out of 12 patients had lymph node metastasis in our study group. Except for one patient, no incomplete surgery or tumor recurrence was observed during the eight-month follow-up period.

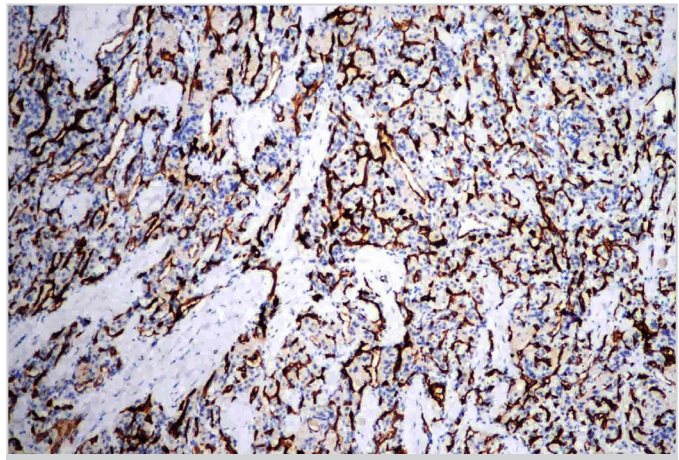

Figure 2. x100 PSMA expression with Score 2 staining in the vessels of the tumoral stroma

PSMA: Prostate-specific membrane antigen

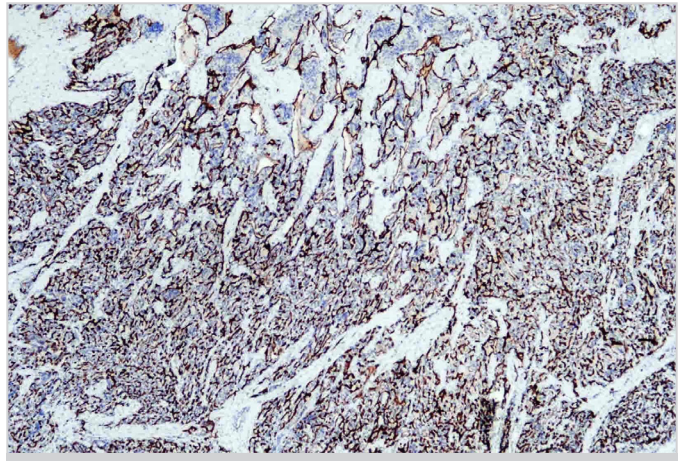

Figure 3. x400 PSMA expression with Score 2 staining in the vessels of the tumoral stroma

PSMA: Prostate-specific membrane antigen

The gold standard for assessing the vascular function of these tumors is conventional optical subtraction angiography. PGLs are hypervascular masses that splay the internal and external carotid arteries and have major feeding arteries, as well as rapid contrast washout on angiography (11). In our study, all 12 patients underwent angiography. Major feeding vessel(s) were determined immediately before the surgery (Figure 4 shows the angiographic imaging of a right sided CBP). However, only one patient's tumor was successfully embolized.

The Shamblin classification can inform us about the size of the tumor and its relationship to the carotid arteries. Localized tumors with splaying of the carotid bifurcation, but no attachment to the carotid arteries are known as Shamblin Class I CBPs. The carotid vessels are marginally surrounded by Shamblin Class II CBPs, but intimately surrounded by Shamblin Class III CBPs, which are very difficult to dissect completely. It is common for vascular repair to necessitate a temporary disruption in cerebral circulation. In comparison to Class I and Class II, the probability of chronic vascular and neurological deficits is considerably higher in Class III (9). Three of our patients had type I, six had type II and three had type III tumors according to the Shamblin Classification. Incomplete surgery, superior laryngeal nerve paralysis and necessity for arterial grafting were the challenges for these patients. Another aspect worth noting was capsular/vascular invasion seen in the histopathological examination of two out of three patients.

Surgery is the preferred treatment for CBPs, even though it can often be complicated by factors such as carotid artery involvement, tumor vascularity, proximity to cranial nerves, and expansion to the skull base (2). All our patients underwent surgery and only one resulted with incomplete surgery. All patients were followed-up regularly.

Many histopathologic stains, such as chromogranin, synaptophysin, serotonin, and neuron-specific enolase, may be used to differentiate these tumors based on their neuroendocrine nature. These histopathological stains, on the other hand, could be positive for other neural crest tumors,

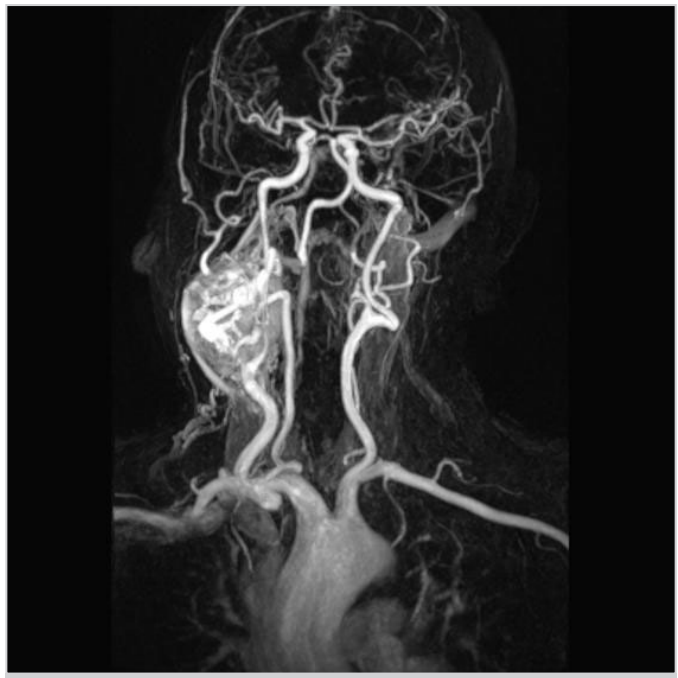

Figure 4. Angiographic appearance of right-sided CBP of 81-year-old female patient

CBP: Carotid body paraganglioma

Table 3. Histopathologic findings of resected CBPs

No	Intratumoral vascular PSMA expression	Extratumoral vascular PSMA expression	Tumoral cell PSMA staining	Ki-67	S100	Snp	Chr	C&V inv	Met
1	2	0	1	5% +	+	-	-	No	No
2	1	0	1	3% +	+	+	Uir	No	Yes
3	2	0	1	3% +	+	Uir	+	Yes	No
4	2	0	0	4% +	+	-	-	Yes	No
5	2	0	1	Uir	+	+	+	Uir	No
6	2	0	0	Uir	Uir	Uir	Uir	Yes	No
7	0	0	0	1% +	+	+	+	No	No
8	2	0	1	Uir	Uir	Uir	Uir	Yes	No
9	Upb	Upb	Upb	Uir	+	+	-	Uir	No
10	2	0	0	5% +	+	-	+	No	No
11	2	0	1	8% +	+	+	?	No	No
12	Upb	Upb	Upb	Uir	Uir	Uir	Uir	No	No

Snp: Synaptophysin, Chr: Chromogranin, C&V inv: Capsular and/or vascular invasion, Met: Metastasis, Uir: Unexplored in initial reports, Upb: Unsuitable paraffin blocks

which is a drawback that should be considered before making a differential diagnosis (7).

PSMA is a potential molecule for tumor-specific vascular targeting. PSMA's role in endothelial cell activity is not well understood. According to *in vitro* and *in vivo* studies PSMA can play a role in endothelial cell invasion through peptidase activity (4). In case of vascular tumors like CBPs, the investigations of PSMA can provide us new therapeutic and diagnostic ways. Our study has essentially showed the presence of intense PSMA in intratumoral vascular structures. Although tumoral cells do not show strong positivity, their vessels are also major targets for both diagnosis and treatment. Zade et al. (12) determined a neck mass in PSMA-PET/CT compatible with metastasis of prostate cancer. They excised the mass and diagnosed it as glomus jugulare (another PGL of the neck).

According to Foss et al. (13), tumor neovasculature and the neovasculature of certain normal proliferative tissues have higher PSMA expression on the apical and luminal surface of new blood endothelial cells (endometrium, heart valve injuries, pleural lesions, and keloid scars). All these findings show the importance of PSMA in the neovascularization and blood supply of the solid tumors.

In vivo PSMA expression in tumors can serve as a replacement molecule for neovascularization, according to Tripathy et al. (5), and this concept can be used for selective therapy in vascular benign tumors such as head and neck PGLs if resection is difficult or impossible. Our study revealed that strong expression of PSMA occurred in the intratumoral vascular structures of CBPs. PSMA targeted prospective studies (either diagnostic or therapeutic) may be planned according to intratumoral vascular expression of PSMA.

The limitations of our study were mainly its preliminary nature and limited numbers of cases: While the number of female patients was twice that of male patients, the small number of patients did not allow us to reach a conclusion about the effect of gender on PSMA expression. The same limitation exists for age groups. More comprehensive studies involving larger numbers of patients are required to overcome these limitations.

Conclusion

We detected PSMA expression in CBPs. The most important part of this expression is intratumoral vascular structures. We did not find PSMA expression in the extratumoral vascular structures. We recommend conducting prospective studies targeting intratumoral vascular PSMA expression. PSMA targeted modalities may be the future of diagnostic and therapeutic methods. This needs to be confirmed by prospective studies.

Ethics Committee Approval: The Clinical Research Ethics Committee of Süleyman Demirel University approved our study protocol. (approval date: 11/09/2020; approval number: 254).

Informed Consent: The study was designed as a retrospective study; therefore, informed consent was not obtained from the patients.

Peer-review: Externally peer-reviewed.

Conflict of Interest Statement: The authors declare no conflict of interest.

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Authorship Contributions

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

Main Points

- The most common paragangliomas in the head-neck region are CBPs, and they are primarily treated with surgery.
- For CBPs, new treatment strategies can be developed in cases which surgery cannot be performed.
- The presence of PSMA has been detected in the neovasculature of some non-prostatic solid tumors.
- This study has shown the presence of PSMA in CBPs, and PSMA is expected to shed light on new treatment strategies.

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Author Self-Citation in the Turkish Otorhinolaryngology Literature

Original Investigation

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Abstract

Objective: To evaluate the prevalence and other characteristics of author self-citations in six Turkey-originated general otorhinolaryngology (ORL) journals of Turkish ORL literature.

Methods: A total of 970 articles published in six Turkey-originated general ORL journals (ENT Updates, Journal of Ear Nose Throat and Head Neck Surgery, KBB-Forum, Praxis of Otorhinolaryngology, The Turkish Journal of Ear Nose and Throat, and Turkish Archives of Otorhinolaryngology) in 2016-2020 were analyzed for author self-citations. The association between author self-citations and journal types, study types, study topics, country of origin, and compatibility with the topic were also evaluated.

Results: There were 265 author self-citations (0.273 per article) which corresponded to 1.36% of all citations. There was no significant difference between the journal types, study topics, and origin of the studies in terms of mean self-citation values per study, whereas case reports had significantly lower self-citations than review and original investigations. There were three citations (1.1%) that were irrelevant to the study topic.

Conclusion: To the best of our knowledge, this is the first study that investigated the practice of author self-citation in Turkish ORL literature. Author self-citation rate in the Turkish-originated general ORL journals was found remarkably lower than the medical literature, whereas the self-citations were found compatible with the study topic to a very large extent. Members of the scientific community including authors, readers, and journal editors should be cautious regarding the unethical practices of self-citations.

Keywords: Bibliometric analysis, publications, self-citation, otorhinolaryngology, publication ethics.

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Introduction

One of the main goals of publishing a scientific work is to introduce its existence to scientific and public recipients as much as possible which can also be defined as "recognization". It is also anticipated from scientific work to provide a significant impact on medical literature which is also valid for researchers and journals. Hence, every scientific content requires

objective measurement methods to quantitatively present its impact and recognization. Several bibliographic indicators have been utilized to objectively assess the recognization or impact of research, author or journal such as citations, Hirsch index (H-index), and impact factor with the presence of their limitations (1-3). Although bibliometric significance and accuracy of citations are still a matter of debate, highly cited scientific works are theoretically considered to be more important and influential (4). Therefore, high citation numbers are assumed to be a constant parameter that warrants scientific achievements such as career promotion and academic reputation.

Self-citation is a type of citation that refers to citing one's own work (5). It comprises multiple subtypes that are ascribed for authors, journals, institutions, countries, and languages (6). Self-citations are likely to be an inevitable entity for productive researchers especially focus on a specific field since they have a higher probability to cite their own papers (7). Also, self-citations have multiple benefits since they enable authors to expand earlier hypotheses, refer to the established methodology and utilize them for further investigations (8). However, self-citations can also be manipulated by the researchers to affect their influence and recognization. Due to the potential role of author self-citations in citation manipulation, the accurate utilization of self-citations has become an ethical concern and there is ongoing effort to better delineate the self-citation-based ethical misconducts in the academic era (9-14).

Author self-citation is simply defined as any instance where a given author cites their own articles in subsequent scholarly works (11). The role of author self-citation in different medical specialties including otorhinolaryngology (ORL) has been investigated in the literature (6-8, 15-17). However, the prevalence and other characteristics of author self-citation in Turkish ORL literature are unidentified to date. In the present study, we aimed to evaluate the prevalence and other characteristics of author self-citations in six Turkey-originated ORL journals of Turkish ORL literature.

Methods

The articles from six Turkey-originated general ORL journals (ENT Updates, Journal of Ear Nose Throat and Head Neck Surgery, KBB-Forum, Praxis of Otorhinolaryngology, The Turkish Journal of Ear Nose and Throat, and Turkish Archives of Otorhinolaryngology) published between 2016 and 2020 were evaluated for author self-citations. The studies that belonged to the supplements were not included in the study. The articles were analyzed according to the authors, citations, study topics and types, country of origin, and language. Original investigations, case reports, and reviews were included among the article types. Study topics were categorized as otology, rhinology, head/neck, pediatric ORL, and general ORL. The total and the mean number of self-citations were calculated per study. Self-citations were evaluated according to the journals, type and topic of the articles, and country of origin. Study authors were categorized as the first author, last author, and other authors, and the mean number of self-citations calculated according to the author arrangement was compared. Additionally, self-citations were reviewed in terms of compatibility with the issue of the main text to reveal possible manipulations.

Statistical analysis was performed using Statistical Package for the Social Sciences (v. 21; SPSS Inc., IBM Corp., Armonk, NY). Data were tested for normal distribution using the Kolmogorov–Smirnov test. Mann-Whitney U and Kruskal-Wallis tests were used to compare the mean author self-citation values between the journal types, study topics, study types, and study origins. A p-value less than 0.05 was considered significant for all comparisons.

Results

Characteristics of the published articles and distribution of the self-citations are shown in Table 1. A total of 970 articles were found in 2016-2020 with the Turkish Archives of Otorhinolaryngology (21.1%) and The Turkish Journal of Ear Nose and Throat (19.5%) had the highest number of articles. Of the 970 articles, 287 (29.6%) were written in Turkish, whereas 683 (70.4%) were written in English. The most frequent study topics were otology (29.8%) and general ORL (21.7%), respectively, whereas original investigations had the highest rate among the study types (75.3%). There were 909 (93.7%) Turkey-originated studies, while 61 (6.3%) were published from foreign countries (Table 2). The mean number of authors and citations per study were 4.3 and 20.1, respectively.

One hundred sixty-eight (17.3%) articles involved author self-citations. The maximum and minimum numbers of self-citation for a single article were 11 and one, respectively. The total number of self-citations was 265 with an average number of 0.273 per article. The total number of self-citations constituted 1.36% of all citations. Turkish Archives of Otorhinolaryngology and The Turkish Journal of Ear Nose and Throat had the highest mean value of self-citations per study, whereas otology and reviews had the highest mean value of self-citations among the study topics and types, respectively. Case reports involved a significantly lower mean value of self-citations than original reports and reviews ($p<0.001$). Reviews had also significantly higher mean value of self-citations than original reports. There were no significant differences between the journal types, study topics, and origin of the articles in terms of mean self-citation values per study. There was no significant difference between the first (n=90, 0.119 per study), last (n=98, 0.155 per study),

Table 1. Characteristics of the published studies and distribution of the self-citations

		Number of study (%)	Number of self-citation (%)	Self-citation per study	p*
Journal	TAO	205 (21.1)	74 (27.9)	0.36	0.276
	Tr-ENT	189 (19.5)	54 (20.4)	0.285	
	KBB-Forum	167 (17.2)	45 (17)	0.269	
	ENT Updates	155 (16)	39 (14.7)	0.251	
	Praxis of ORL	137 (14.1)	23 (8.7)	0.167	
	ENT-HNS	117 (12.1)	30 (11.3)	0.256	
Study topic	Otology	289 (29.8)	101 (38.1)	0.349	0.083
	General ORL	210 (21.7)	52 (19.6)	0.247	
	Head and Neck	202 (20.8)	43 (16.2)	0.212	
	Rhinology	196 (20.2)	55 (20.8)	0.28	
	Pediatric ORL	73 (7.5)	14 (5.3)	0.191	
Study type	Original Investigation	730 (75.3)	207 (78.1)	0.283	<0.001
	Case Report	202 (20.8)	15 (5.7)	0.074	
	Review	38 (3.9)	43 (16.2)	0.883	
Country of origin	Turkey	909 (93.7)	239 (90.2)	0.262	0.561
	Foreign countries	61 (6.3)	26 (9.8)	0.426	
Total	Number of study	970	265	-	-
	Number of self-citation				

ENT: Ear Nose Throat, ENT-HNS: Journal of Ear Nose Throat and Head Neck Surgery, ORL: Otorhinolaryngology, Tr-ENT: The Turkish Journal of Ear Nose and Throat, TAO: Turkish Archives of Otorhinolaryngology.

*Statistical comparison for self-citation per study values among the parameters

Table 2. Distribution of the studies according to the country of origin

Country	n (%)	Country	n (%)
Turkey	909 (93.7)	Bosnia	1 (0.1)
India	19 (1.9)	Croatia	1 (0.1)
Malaysia	6 (0.6)	Greece	1 (0.1)
England	3 (0.3)	Iran	1 (0.1)
Pakistan	3 (0.3)	Japan	1 (0.1)
Portugal	3 (0.3)	Morocco	1 (0.1)
Saudi Arabia	3 (0.3)	Montenegro	1 (0.1)
Brazil	2 (0.2)	Nepal	1 (0.1)
Egypt	2 (0.2)	New Zealand	1 (0.1)
Ghana	2 (0.2)	Philippines	1 (0.1)
Italy	2 (0.2)	Russia	1 (0.1)
Serbia	2 (0.2)	USA	1 (0.1)
Spain	2 (0.2)		

n: Number of items

and other authors (n=107, 0.158 per study) in terms of the mean number of self-citations (p=0.396). There were three citations (1.1%) which were irrelevant with the study topic.

Discussion

In the competitive academic world, researchers necessarily endeavor to improve their productivity. Since higher productivity doesn't always mean higher quality, it is also anticipated from the researchers to confirm their productivity with higher impact. To quantitatively reveal their balance of productivity and impact, citations have become one of the major goals of the researchers that are tried to be obtained. As a consequence, a potential risk has arisen which is manipulating the number of citations as a career motivation and self-citations provide a potential microenvironment for the authors to artificially boost their citation-based metrics (18). Bartneck and Kokkelmans (19) demonstrated that H-index can be significantly inflated through self-citations by the authors. In the present study, we found that author self-citation rates were lower than the literature for the studies published in six Turkey-originated ORL journals between 2016 and 2020. Also, the author self-citations were found compatible with the study topic to a very large extent (98.9%) which may suggest that the author self-citations are not served for unethical purposes in Turkish ORL practice.

Self-citations were reported to constitute 6-20% of all citations in the medical literature (8, 15). In a study

investigating the author self-citations in the five highest-ranked journals of ORL, Tolisano et al (17) found that about two-thirds of all articles included at least one self-citation. The authors reported that self-citations represented 10% of all citations with an average of 2.6 self-citations per article. In the present study, nearly one of the five studies (17.3%) published in six Turkey-originated general ORL journals involved at least one self-citations, whereas self-citations constituted 1.36% of all 100 citations. The mean value of self-citations per study was 0.273 which was remarkably lower than the literature (17). Also, there was no significant difference between the author arrangements in terms of self-citations in the present study. However, Tolisano et al. (17) reported that lead authors and last authors were the frequent self-citers in the ORL field. As primary conductors, it is logical to expect that the first author appears as a leading self-citer among the authors of a scientific paper. On the other hand, since the last authors are usually composed of researchers with more senior academic positions, they may have a higher opportunity for self-citing due to their long track record of publications and expertise. These discrepancies between the present study and the literature can be associated with a number of facts. In Turkish ORL practice, Turkey-originated journals mostly constitute the second line for submitting a study and author preferences regarding the journal choice for submission may influence the self-citation characteristics. For instance, due to academic promotion and recognization concerns, more experienced and productive researchers may tend to publish their high-quality scientific works regarding their specified research fields in journals having high impact factor values rather than publishing in Turkey-originated journals. Mishra et al. (20) reported that self-citations are the hallmark of productive and experienced authors specialized in certain medical fields who may have a higher potential for self-citation. Also, recruit researchers may initially prefer Turkey-originated journals during their learning curve of scientific publishing processes and their low self-citation potentials due to the smaller number of previous works and low expertise may diminish the chance for author self-citation. However, these speculations should be verified with further investigations.

The studies investigating the association between author self-citations and the type and topic of the studies revealed conflicting results in the literature. Livas et al. (7) screened six orthodontic journals with the highest impact factor and found that there was no association between self-citation counts and the type and topic of the studies. However, Tolisano et al. (17) reported that original articles had a significantly higher author self-citation rate, whereas no difference was found among the otolaryngological study topics. The authors found that case reports had significantly lower number of self-citations per article when compared with reviews and original investigations. Similarly, in the present study, the author self-citation values were not significantly different among the study topics, whereas the case reports had significantly lower self-citation values than the other study types.

The present study has several limitations. First, we included the articles published in six Turkey-originated general ORL journals between 2016 and 2020 and the results of the present study cannot be generalized for the entire Turkish ORL literature. Secondly, our study primarily intended to offer a general view of the author self-citations in Turkish ORL practice, hence, it does not provide further data regarding the possible causes of the discrepancies with the literature. Nevertheless, we think that similar studies investigating the author self-citation trends in Turkish ORL practice may have vital importance to raise the awareness of the academic community concerning the author bias of self-citation.

Conclusion

To the best of our knowledge, this is the first study investigated the practice of author self-citation in Turkish ORL literature. In the present study, author self-citation rate in the Turkish-originated general ORL journals was found remarkably lower than the medical literature, whereas the self-citations were found compatible with the study topic to a very large extent. Although the results of the present study can be interpreted as the authors in Turkish ORL practice do not use self-citations at a level suggesting citation manipulation, members of the scientific community including authors, readers and journal editors should be cautious regarding the unethical practices of self-citations.

Ethics Committee Approval: Bibliometric study.

Informed Consent: Bibliometric study.

Peer-review: Externally peer-reviewed.

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Otolaryngology Residents' Attitudes, Experiences, and Barriers Regarding the Medical Research

Original Investigation

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Abstract

Objective: It was aimed to investigate the attitudes, experiences, and barriers towards scientific research among otolaryngology residents in Turkey.

Methods: Anonymous data were collected via an online survey. The demographic characteristics and experience in scientific research were investigated in the first section of the survey. The attitudes of participants towards scientific research and the barriers to the scientific research were examined in the second section of the survey using 17 items.

Results: The present study involved 119 otolaryngology residents continuing their education. It was determined that 68.1% of participants think that "*participating in scientific research*" is a part of otolaryngology training. In the present study, it was shown that the residents having journal club hours in clinics on regular basis participated in various steps of scientific research projects ($p<0.05$). Residents stated that they participated in the "*literature review*" stage of the preparation of a scientific publication (mean value of 2.58±1.88) most and in "*verbal presentation in a congress*" least (mean value of 0.74±1.44). It was determined that 80.7% of participants have not attended in any training on scientific research. It was found that the residents receiving structured scientific research training participated more in steps of scientific research projects which was statistically significantly ($p<0.05$).

Conclusion: In our country, otolaryngology residents are very enthusiastic about having research education and participating in researches. However, residency students frequently have time deficiency, lack of knowledge-skill, and lack of financial support. Dedicated time should be allocated for research training and practice in specialty programs. Journal club activities should be organized on regular basis and integrated with research education. On the other hand, the scholarly activities of residents should be supported by means of various countrywide educational activities on research training.

Keywords: Otorhinolaryngology, scientific research, resident, research education, community survey, educational activities

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Introduction

For a successful scientific research, having knowledge on writing a manuscript and on technical subjects such as clinical study design, bias, confounders, and statistics is a must. Besides the curriculum, the research education in residency education can be improved by implementing the theoretical knowledge in a research project and by sharing the experiences of a mentor (1). In the entire world, scholarly activity is an essential component of otolaryngology residency training programs. In our country, however, the involvement of residency students in researches during their residency period originates for the first time from "Regulation on Medical Residency" in 1974 (2). The research component has been included first time in the Turkish Otolaryngology-Head and Neck Surgery Residency Core Curriculum published in 2012 (3). In this program, the research activities were identified as a learning tool and it was recommended for every resident to participate in a minimum of one research (except for the thesis study) during the residency period. In the section of cognitive learning, it was requested for the resident to plan and conduct a research and make a scientific presentation. In the following years, the Curricula and Standard Development System of the Council of Specialty in Medicine (TUKMOS) developed the current otolaryngology specialty curriculum (4). In this program, research is defined as "the process of designing a research on a subject solely by a student or with a team and, by determining the learning needs in this period, meeting these needs from any educational source". Research activity has been employed in medical residency education since 1974 but there is no study examining how successfully the research component has been implemented.

In the present study, it was aimed to determine the ear nose throat (ENT) residents' attitudes, experiences, and barriers in scientific research in Turkey, as well as the troubles they have experienced.

Methods

In the present study, it was planned to collect the data via an anonymous web-based survey. In the first step, a comprehensive literature review was performed by researchers and an item pool was established (5-8). Then, two researchers working in different institutions conducted interviews with ENT residents and the item pool was prepared by enriching with the items including the conditions of ENT residency in our country. And then, considering the simplicity and understandability of items, the preliminary corrections were made and the opinions of experts about the item pool were collected. The preliminary implementation of the survey, on which the experts had a consensus, was performed with 10 ENT residents asked to answer the draft items and it was confirmed that the items were understandable.

The first part of the survey includes the items examining the demographical characteristics (gender, type of hospital where they receive residency training, and years in residency education) and experience in the cognitive research field. Using 17 items in the second section of the survey with 5-point Likert scale, the participants were asked to assess the attitudes towards research and the barriers they face. The surveys were conducted online using Google® forms. Announcements were regularly performed through "Kanal KBB" online communication platform, in which ENT residents are members, and via training clinic supervisors in order for ENT residents to fill in the surveys. The online survey was kept accessible for the participants for a month between August 15th and September 15th 2020. Ethics committee approval was obtained from the Clinical Research Ethics Committee of Pamukkale University (08.01.2020/456).

Statistical Analysis

All the analyses were performed using IBM SPSS 23.0 package program (IBM Corp., Armonk, NY). Descriptive statistics were expressed as frequency and percentage for categorical variables and mean ± standard deviation (SD) for continuous variables. The relationships between categorical variables were analyzed using Fisher's Exact Test or Pearson's chi-square test. The difference between measurements in two groups was analyzed using Student's t-test. A two-sided p-value <0.05 was interpreted as statistically significant.

Results

The target sample size was determined to be 544, the number of ENT residents actively continuing their education. In total, 119 (rate of participation is 21.8%) ENT residents (woman: 35, man: 84) responded. Demographic characteristics of residents are presented in Table 1. The mean age of volunteer participants was found to be 28.22±1.81 years (min: 25-max: 35). Seniorities of participants are 28 in the first year, 25 in the 2nd year, 38 in the 3rd year, 10 in the 4th year, and 18 in the 5th year. In order to compare the residents' participation in scientific researches and their attitudes with the core curriculum in ENT residency education, the seniorities of residents were examined by dividing in two (≥24 months and <24).

Comparison of research experience of residents by gender, type of hospital and residency training periods is presented in Table 2. From the aspect of gender, a statistically significant difference in favor of men was observed in preparing a method for scientific research (p=0.024). Regarding preparing ethical committee application, presenting a poster in a congress, submitting a manuscript to a journal, and published research manuscript, a statistically significant difference was observed between residents by the clinics, in which they received training (p=0.008, p<0.001, p=0.026, p=0.009, and p=0.003, respectively). Examining the data by seniority, it can be seen

that those having seniority of or higher than 24 months participated in most of the items related with playing a role in scientific activities (Table 2). Comparison of the attitudes of ENT residents to the scientific research by gender, type of hospital, and residency training periods is presented in Table 3. The effects of having a structured scientific research education in residency training and having a journal club on regular basis on the participation of residents in scientific activities are statistically presented in Table 4.

Table 1. Residents' characteristics

Variables	(n=119)
Age (years)	28.2±1.8
Gender	
Female	35 (29.4%)
Male	84 (70.6%)
Type of hospital	
University hospital	68 (57.1%)
Training and research hospital	51 (42.9%)
Residency training periods (years)	2.2±1.4

Discussion

The objectives of "research education" are to promote evidence-based medicine practice, to provide skills for life-long learning, and to support critical-thinking skills. These objectives are in corroboration with the "journal club" activity (9, 10). Besides keeping the knowledge on education activity updated, the other objectives of the journal clubs are to provide critical thinking skills and evidence-based practice skills, life-long learning, determining the subjects to research in literature, learning the research methods, to provide the skills of research planning and manuscript writing, and to learn bio-statistical analysis methods (11). Research

Table 2. Comparison of research experience of residents by gender, type of hospital and residency training periods

	Gender			Type of hospital			Residency training periods		
Steps of research that resident has participated in, n (%)	Female (n=35)	Male (n=84)	p-value	University hospital (n=68)	Training and research hospital (n=51)	p-value	<24 months (n=53)	≥24 months (n=66)	p-value
Literature review	28 (80%)	68 (81%)	0.905	51 (75%)	45 (88.2%)	0.070	35 (66%)	61 (92.4%)	<0.001
Preparing an application to the ethics committee	25 (71.4%)	55 (65.5%)	0.528	39 (57.4%)	41 (80.4%)	0.008	28 (52.8%)	52 (78.8%)	0.003
Preparing a method for research	16 (45.7%)	57 (67.9%)	0.024	37 (54.4%)	36 (70.6%)	0.073	27 (50.9%)	46 (69.7%)	0.037
Presenting a poster in congress	17 (48.6%)	48 (57.1%)	0.392	30 (44.1%)	35 (68.6%)	0.008	20 (37.7%)	45 (68.2%)	0.001
Writing a manuscript	16 (45.7%)	45 (53.6%)	0.435	33 (48.5%)	28 (54.9%)	0.491	21 (39.6%)	40 (60.6%)	0.023
Statistical analysis	15 (42.9%)	36 (42.9%)	0.999	31 (45.6%)	20 (39.2%)	0.487	19 (35.8%)	32 (48.5%)	0.166
Submission to a journal	9 (25.7%)	35 (41.7%)	0.100	16 (23.5%)	28 (54.9%)	<0.001	17 (32.1%)	27 (40.9%)	0.321
Verbal presentation in a congress	7 (20%)	29 (34.5%)	0.116	21 (30.9%)	15 (29.4%)	0.863	9 (17%)	27 (40.9%)	0.005
Published case presentation	12 (34.3%)	35 (41.7%)	0.453	22 (32.4%)	25 (49%)	0.066	16 (30.2%)	31 (47%)	0.063
Published research article	14 (40%)	33 (39.3%)	0.942	21 (30.9%)	26 (51%)	0.026	18 (34%)	29 (43.9%)	0.268
Having education on scientific research	5 (14.3%)	18 (21.4%)	0.369	12 (17.6%)	11 (21.6%)	0.592	4 (7.5%)	19 (28.8%)	0.004
Having a certificate for using experimental animals	4 (11.4%)	7 (8.3%)	0.729	2 (2.9%)	9 (17.6%)	0.009	1 (1.9%)	10 (15.2%)	0.022
Having regular "journal club"	27 (77.1%)	62 (73.8%)	0.703	44 (64.7%)	45 (88.2%)	0.003	32 (60.4%)	57 (86.4%)	0.001

Pearson chi-square test, Fisher's Exact test.

Significant p-values are shown as bold and italic.

Table 3. Comparison of the attitudes of ENT residents to the scientific research by gender, type of hospital and residency training periods

	Gender			Type of hospital			Residency training periods		
Residents' attitudes regarding research experience, mean ± SD	**Female (n=35)**	**Male (n=84)**	**p-value**	**University hospital (n=68)**	**Training and research hospital (n=51)**	**p-value**	**<24 months (n=53)**	**≥24 months (n=66)**	**p-value**
Every resident should be given an education on doing scientific research	4.7±0.6	4.6±0.8	0.430	4.6±0.7	4.6±0.7	0.581	4.6±0.8	4.6±0.7	0.986
The education on scientific research should be given only to the residents planning to have an academic career	2.2±1.4	2.6±1.7	0.237	2.6±1.6	2.3±1.6	0.227	2.6±1.7	2.4±1.5	0.446
I'd like to participate in scientific research activities during my residency time	4.7±0.6	4.5±0.8	0.115	4.4±0.8	4.7±0.6	0.012	4.5±0.7	4.6±0.8	0.538
In the clinic, in which I'm having education, residents are not given any task in researches	1.8±1.3	2.2±1.4	0.235	1.8±1.2	2.4±1.5	0.034	1.9±1.2	2.2±1.5	0.273
We do not have time for research	3.2±1.4	3.5±1.3	0.335	3.3±1.4	3.4±1.2	0.692	3.4±1.3	3.4±1.3	0.892
I have no knowledge about counseling to a statistician for statistical analysis	3.9±1.2	3.7±1.4	0.416	3.8±1.2	3.6±1.5	0.331	4.2±1.1	3.3±1.4	<0.001
My educational background on research methodology is not sufficient	4.1±1.3	4±1.1	0.573	3.9±1.2	4.2±1	0.232	4.2±1.2	4±1.1	0.395
I do not want to do a scientific research	1.5±0.7	1.9±1.3	0.053	2±1.3	1.5±0.9	0.024	1.7±1	1.8±1.2	0.805
It is very hard to collect data for a scientific research	2.9±1.1	2.6±1.2	0.166	2.8±1.2	2.4±1.2	0.048	2.7±0.9	2.6±1.3	0.487
Data entry for research is an annoying task	3.2±1.4	3.3±1.2	0.775	3.3±1.2	3.2±1.3	0.680	3.3±1.2	3.2±1.3	0.927
It's hard to find financial support for scientific researches	4±1.1	4.3±0.8	0.149	4.1±1	4.3±0.8	0.187	4.1±1	4.2±0.9	0.449
There is no incentive or reward for me to participate in researches	3.6±1.3	4.1±1.1	0.058	3.8±1.2	4.1±1	0.155	3.8±1.2	4±1	0.387
There is no guiding educators leading the way for participating in researches	2.7±1.3	2.9±1.5	0.492	2.9±1.4	2.9±1.5	0.999	2.9±1.5	2.9±1.4	0.976
It should be obligatory for residents to participate in researches	3.4±1.5	3.4±1.4	0.934	3.1±1.5	3.8±1.2	0.004	3.3±1.5	3.5±1.4	0.624
I do not have opportunity to access to full texts of research articles	2.5±1.5	2.1±1.3	0.193	2.4±1.3	2±1.4	0.100	2.2±1.2	2.3±1.5	0.838
"Even" the residency thesis is unnecessary	1.5±0.9	1.8±1.1	0.199	1.9±1.2	1.5±0.7	0.028	1.4±0.8	1.9±1.2	0.018
My foreign language is insufficient for reading and writing scientific articles	3.1±1.2	2.8±1.3	0.311	2.9±1.4	2.9±1.2	0.903	3.1±1.3	2.7±1.3	0.140

ENT: Ear nose throat, SD: Standard deviation. Student's t-test, n: Number

Significant p-values are shown as bold and italic.

Table 4. Comparison of research experience of residents by having education on scientific research and regular "journal club" activity

Steps of research that resident has participated in, n (%)	Having education on scientific research			Having a certificate for using experimental animals			Having regular "journal club"		
	Yes (n=23)	No (n=96)	p-value	Yes (n=11)	No (n=108)	p-value	Yes (n=89)	No (n=30)	p-value
Literature review	22 (95.7%)	74 (77.1%)	0.043	10 (90.9%)	86 (79.6%)	0.367	77 (86.5%)	19 (63.3%)	0.005
Preparing an application to the ethics committee	23 (100%)	57 (59.4%)	<0.001	11 (100%)	69 (63.9%)	0.015	65 (73%)	15 (50%)	0.020
Preparing a method for research	18 (78.3%)	55 (57.3%)	0.064	9 (81.8%)	64 (59.3%)	0.199	61 (68.5%)	12 (40%)	0.006
Presenting a poster in congress	17 (73.9%)	48 (50%)	0.039	11 (100%)	54 (50%)	0.001	54 (60.7%)	11 (36.7%)	0.022
Writing a manuscript	18 (78.3%)	43 (44.8%)	0.004	8 (72.7%)	53 (49.1%)	0.135	52 (58.4%)	9 (30%)	0.007
Statistical analysis	17 (73.9%)	34 (35.4%)	0.001	5 (45.5%)	46 (42.6%)	0.999	41 (46.1%)	10 (33.3%)	0.223
Submission to a journal	13 (56.5%)	31 (32.3%)	0.031	6 (54.5%)	38 (35.2%)	0.325	38 (42.7%)	6 (20%)	0.026
Verbal presentation in a congress	10 (43.5%)	26 (27.1%)	0.124	3 (27.3%)	33 (30.6%)	0.999	29 (32.6%)	7 (23.3%)	0.340
Published case presentation	15 (65.2%)	32 (33.3%)	0.005	7 (63.6%)	40 (37%)	0.110	39 (43.8%)	8 (26.7%)	0.096
Published research article	14 (60.9%)	33 (34.4%)	0.020	8 (72.7%)	39 (36.1%)	0.024	40 (44.9%)	7 (23.3%)	0.036

Pearson chi-square test, Fisher's Exact test.

Significant p-values are shown as bold and italic.

education in residency shares such common objectives with the journal club activity. Thus, the journal club is the residents' most important educational activity, in which their research education begins. Besides the common objectives, it was also reported that journal clubs also frequently incorporate the subjects to be investigated (12). The "journal club" is an activity, in which the theoretical background of residents' research education is constructed. It was determined that 76% of otolaryngology residents participating in the present study have a regular journal club in their clinic. Moreover, it was statistically presented that the residents with regular journal club hours participated in many steps of scientific research projects (Table 4). Thus, it can be stated that the regular journal club activity in residency training of ENT residents increases their participation in research activities.

As in all the surgical branches, also the ENT residency is a tiring and challenging process for the residents (13). Despite this fact, the results of the present study suggest that 89% of ENT residents wanted to participate in scientific research activities during their residency period. Given this ratio, it can be seen how otolaryngology residents in our country are enthusiastic about this subject. Canadian plastic surgery residents reported the rate of willingness to participate in research to be 70% (9). The reason for this willingness might be their plans for building an academic career after the residency (14, 15). In our country, 40% of residents plan to build an academic career (16). Although it was not questioned in the present study, the motivation of academic career after the residency might be a factor motivating them to conduct research.

In Turkey, master of science and doctorate programs offer research education, whereas there is no structured formal research education in residency. Otolaryngology residents have already stated that residency should incorporate a formal and structured research education (17). Similarly, the present study showed that 91.6% of the participants requested research education to be incorporated into the residency program. Moreover, as seen in Table 4, it was determined in the present study that residents having a scientific research training participated in steps of scientific researches more than those having no such training which is statistically significant. This finding corroborates that a training program to be added to ENT residency training would increase the efficiency of students' participation in researches. In Turkey, it is aimed to standardize the residency education by means of the Council of Specialty in Medicine. The best solution would be the integration of research education into the specialty program. However, it cannot be stated that, despite the efforts, no common standards could be achieved in terms of either education centers or educators. However, the scholarly activities of residents should be supported through countrywide activities on research education, in which both residents and educators can participate. Online education programs, in which many residents can participate, can be used in these education programs (18-20). These programs should be planned in the light of science and in guidance of

experienced specialists in accordance with the principles of educational sciences. Moreover, it should be noted that these educational activities should be financially supported.

In literature, it was reported that, although residents might want to participate in scientific research projects, they may sometimes face several obstacles such as lack of knowledge-skill, insufficient time, and lack of financial support (14, 21). The surgical residency educations incorporate an intense study program (13). For this reason, no time can be allocated to scientific research projects and educations while running routine healthcare services. In the present study, the ratio of those accepting the hypothesis *"We do not have time for researches"* was found to be 45.3%. In previous studies, the most important barrier towards residents' scientific research was reported to be the time factor (64%–78.9%) (14, 15, 21). The only solution for the time insufficiency is the research time devoted to scientific research activities in residency programs (22, 23). In literature, there is no common practice about how to plan the dedicated research time. There are samples such as research rotation between a year and several months or the activities planned for specific days of a month (5). This time can be determined and planned according to the working conditions of the training clinics and the expectations from the research training. This period can be defined as optional that will not be included in the residency training period.

Another barrier towards residents' scientific research was specified to be the lack of financial support (21, 24). In literature, it was reported that 44% of residents had no sponsors in their scientific studies (24). In the present study, 89% of participating residents stated that they couldn't access to financial supports. In Turkey, a research designing culture should be established and the financial supports for thesis work, which is one of the requirements of residency programs, should be increased.

Nowadays, because the most common science language is English, incompetence in a foreign language might be an important problem for the residents. In order to do a research and write a manuscript to be published, it is necessary to have a good command of both verbal and written English language. In a study carried out by Yılmaz et al. (16), it was reported that only 25% of residents were found to be capable of writing and reading in a foreign language. The results achieved in the present study were in parallel with those reported by Yılmaz et al. It was determined that 36.9% of participating residents stated that they had no problem in writing and reading in a foreign language, whereas 33.6% specified it as a problem. For countries, where English is not the native language, such as our country, inefficacy in a foreign language is still considered to be a problem in the education of residents.

Another one among the most important problems in the education of ENT residents was the lack of mentorship to be provided by the educators. During the residency period, mentor should transfer his/her experience in this field, provide guidance at every step, and help with establishing a professional network (25). Residency students should be assigned an active role in every step of research such as guiding to project, preparing a project, and time management, and this can be achieved via mentorship (26). Moreover, another problem is the insufficiency of the mentors (21, 25). It was determined that 43.6% of residents participating in the present study do not agree with the statement *"There is no educator guiding me to participate in researches"*, whereas 21.8% were neutral. The residents might be thinking so because they witnessed researches at any step in the clinics, in which they were having education. However, in our country, there is no structured research education, which is incorporated in doctorate programs, but not in in residency. Even the academicians giving residency education develop their knowledge on doing research with their own means. The qualifications required for being a researcher have been identified (8). At least, for the academicians participating in residency training, providing structured research and statistics education similar to those in doctorate programs might solve this deficiency.

In the present study, an interesting result on the timing of research training was achieved. In the ENT core curriculum, all the research skills are expected to be achieved in the first half (2). However, at the end of the study, it was determined that the participation of residents in researches increased after the first two years (Table 2). This finding should be considered while updating the ENT core curriculum.

In this study, the experience of residents in research was also questioned. It can be seen in their answers regarding their experiences on the subject that they have participated in almost every step of research process (Table 2). Making a verbal presentation in a congress (30.3%) was found to be the least frequently performed activity, whereas the most frequently performed one was literature review (80.7%). Moreover, even the rate of participating in the process of article publication among the participating residents was found to be 39.5%. These results indicate that, even though they haven't received any standard and structured research education, ENT residents in Turkey have somehow participated in research processes. In literature, the rate of lack of interest in research was reported to be 43% (15). In the present study, the ratio of lack of interest in research was found to be 9.3%. This result indicates how high the interest of ENT residents of our country in this subject is. Other interesting findings of the present study are that 19.3% of participants have received education in various fields of research, and that 9.2% of participants have "certificate for using experimental animals". These results show the success and motivations of inquiring students when given an opportunity.

In our country, residency education is given in university hospitals and research and training hospitals. As a result of this study, it was determined that residents having education in research and training hospitals (51%) participated in more published research which is statistically significant when compared to those having education in university hospitals (30.9%) (p=0.026). This might be because the participation of residents in researches is supported more in research and training hospitals or because more researches are conducted in training and research hospitals. It is recommended to examine this finding in further studies. From the aspect of gender, a statistically significant difference was found only in the step "Preparing a method for research" and comprehensive qualitative studies are needed to investigate the effects of individual factors.

The individual and mass causes of the factors preventing residents from participating in scientific activities should be investigated in detail. Providing formal scientific research training integrated into residency education would eliminate the lack of education. Allocating time for scientific research in study programs for both educators and residency students can prevent the lack of time and motivate them to participate in scientific research projects. In order to tackle the lack of financial support, which is stated as the most critical barrier in the literature, national measures are needed.

In the present study, the most important limitation is that, although it was aimed to reach 544 ENT residents having education in Turkey, only 119 (21.9%) residents participated in survey. However, considering the fact that ENT residents at different seniority levels have participated, it can be stated that the results reflect the general residency student population in Turkey. Another limitation is that the present study involves only the results of ENT residents. The attitudes and thoughts about and barriers towards research education among the educators, who are in charge in residency education, should be investigated. The educators should be supported in their inadequacies such as research education and mentorship, etc. For further studies, it is recommended to examine if the residents' scholarly activities allow them to gain critical thinking and evidence-based medical practice skills or, in other words, if the objectives of the educational programs are achieved.

Conclusion

The otolaryngology residents in Turkey are very eager to have research education and participate in researches. However, residency students may face obstacles such as lack of knowledge-skill, lack of time, or financial support deficiency. The scholarly activities of residents should be supported through countrywide activities on research education, in which both residents and educators playing a role in residency education can participate. Journal club activities should be performed on regular basis and combined with research training.

Ethics Committee Approval: Ethics committee approval was obtained from the Clinical Research Ethics Committee of Pamukkale University (08.01.2020/456).

Informed Consent: Anonymous data were collected via an online survey.

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Authorship Contributions

Concept: H.E., C.O.K., Design: H.E., C.O.K., Data Collection and/or Processing: H.E., C.O.K., Analysis and/or Interpretation: H.E., C.O.K., Writing: H.E., C.O.K., Critical Review: H.E., C.O.K.

Main Points

- The objectives of education are to promote evidence-based medicine practice, to provide skills for life-long learning, and to support critical-thinking skills.
- The results of the present study suggest that 89% of ENT residents wanted to participate in scientific research activities during their residency period.
- Journal club activities should be organized on regular basis and integrated with research education.
- The scholarly activities of residents should be supported through countrywide activities on research education, in which both residents and educators playing a role in residency education can participate.

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Topical Intranasal Fluorescein to Diagnose and Localize Cerebrospinal Fluid Leak: A Systematic Review

Systematic Review

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Abstract

Objective: This study evaluates the available evidence regarding using topical intranasal fluorescein (TINF) to diagnose and localize nasal cerebrospinal fluid (CSF) leak.

Methods: A literature search was conducted through PubMed, the Cochrane Database, Scopus, and Ovid to identify the articles providing insight into using TINF to diagnose CSF leak preoperatively or to localize the leak intraoperatively. The articles from the database were screened and filtered by two authors according to the selection criteria. A spreadsheet was created to collect the data including demographic characteristics, the sensitivity and specificity of TINF for diagnosing and localizing a CSF leak, the protocol of applying TINF, and the complications.

Results: After excluding duplicates and articles that did not meet our selection criteria, we included five reports in the final analysis. The average age of the 94 participants was 39.5, and there was an equal distribution of males and females. The sensitivity of TINF to make a preoperative diagnosis of CSF leak was 100%, and it was 97% to localize the site intraoperatively. Complications associated with TINF were not reported in any of the reports. This review showed a grade C recommendation based on five case series.

Conclusion: Based on the current evidence, TINF cannot be recommended for standard clinical practice. It can, however, be considered in situations where other gold standard tools are unavailable since it is feasible and easy to use. A standardized control trial should be conducted to yield additional unbiased evidence.

Keywords: Cerebrospinal fluid rhinorrhea, paranasal sinuses, skull base, spinal puncture, transferrin, systematic review

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Introduction

Nasal cerebrospinal fluid (CSF) leak has multiple known etiologies that could be traumatic, iatrogenic, or spontaneous. In general, traumatic CSF leaks, whether accidental or iatrogenic, account for 70%–80% of cases (1-3). The incidence of CSF leaks range from 0.17% in endoscopic sinus surgery to around 6.0% in transsphenoidal surgery (4, 5), which may result in complications in about 10%–37% of those cases (6). Such complications include meningitis, pneumocephalus, and meningo-encephalocele (7). Management of this pathology consists of three steps. Confirming the diagnosis is the first step, in which the fluid is analyzed for beta-2 transferrin as a primary screening test in a suspected symptomatic patient (6). The reported beta-2 transferrin sensitivity and specificity are approximately 97% and 99%, respectively, with positive and negative predictive values of 97% and 99% (8). Also, beta trace protein presents in high concentration in CSF. Studies showed that the sensitivity and specificity for a beta trace protein immunoassay were 100% and 86%, respectively (6). The next step is the localization of the leak site, which is necessary to repair the site in the third step. Imaging studies, including high resolution computed tomography (HRCT) of paranasal sinuses and magnetic resonance cisternography (MRC) play an essential role. Studies on HRCT in localizing a CSF leak showed sensitivity and specificity of 44% to 100% and 45% to 100%, respectively (9, 10), while reported positive and negative predictive values were 100% and 50% to 70% (6, 11, 12). On the other hand, studies reported MRC accuracy, sensitivity, and specificity to be 89%, 87%, and 100%, respectively (13, 14). If the site of a CSF leak cannot be localized radiologically, intrathecal fluorescein (IF) can be used intraoperatively. Although IF can help with localization, it is an invasive procedure requiring lumbar puncture to administer fluorescein, increasing the operating time. Moreover, IF is not approved by the FDA. Therefore, benefits and risks should be discussed with the patient. A consent form must be signed before using IF as it may lead to complications such as malaise, headache, numbness in the extremities, seizures, and chemical meningitis (15). Published reports of implementing IF demonstrated varying rates of detecting the CSF leak site, ranging from 46% to 100% (16-18). Therefore, topical intranasal fluorescein (TINF) has been tried (19, 20). This alternative technique is non-invasive, faster, and causes no complications.

We conducted a systematic review to analyze the efficacy of TINF. Using the available literature, we aimed to investigate the sensitivity, specificity, and safety of TINF. Our secondary objective was to assess the possibility of applying TINF in current practice.

Methods

Data Sources and Literature Search

We conducted an extensive literature review in different databases using specific keywords from January 2000 to December 2019:

PubMed: We used a PubMed Medical Subject Heading (MeSH) search according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines with the search terms: (topical[All Fields] OR local[All Fields]) AND (intranasal[All Fields] OR ("nose"[MeSH Terms] OR "nose"[All Fields] OR "nasal"[All Fields]) AND ("fluorescein"[MeSH Terms] OR "fluorescein"[All Fields]).

Cochrane Database: We used the search terms: (topical AND nasal OR intranasal AND fluorescein).

Scopus: We used the search terms: (topical AND nasal OR intranasal AND fluorescein).

Ovid: We used the search terms: (topical AND nasal OR intranasal AND fluorescein).

Other Resources for Data: We screened the reference lists of identified publications for additional trials and contacted some authors when necessary. We also ran a non-systematic search on Google Scholar to retrieve grey literature and other sources of potential trials.

Study Selection

Following the predetermined inclusion and exclusion criteria, the studies selection was made in two stages by two authors independently. In the first stage, titles, and abstracts of the most relevant studies from the electronic database search were reviewed. The full-text articles of these retrieved studies were collected and reviewed in the second stage. In case of a discrepancy between the two reviewers, a third reviewer intervened until a consensus was reached. In cases of duplicates, the most recent version was selected.

Inclusion and Exclusion Criteria

Studies were selected if TINF was used to diagnose the presence of a CSF leak preoperatively or to localize the leak intraoperatively. To demonstrate the efficacy of TINF, this review included clinical trials, cohort studies, and diagnostic case series. Cases were included if a preoperative diagnosis of a CSF leak was made by TINF and confirmed by a beta-2 transferrin test or another radiological test. If, however, a laboratory or radiological test was used to diagnose a CSF leak before applying TINF, the study was excluded. Another inclusion criterion was the identification of the leak site by

TINF intraoperatively. If IF was used to detect the location of the leak before TINF, the study was excluded. Only studies on CSF leaks were included. However, an iatrogenic CSF leak during endoscopic skull base surgery was not counted.

Data Synthesis and Data Extraction

To ensure that data extraction was accurate and reproducible, two authors independently reviewed the studies. We created review-specific data extraction forms to make the process consistent and to avoid missing data. If additional data was needed, we sent a request with all inquiries via email to the corresponding author of the article.

For each study, we extracted the following data:

- Year of publication and country of origin
- Demographic characteristics of the patients
- Accuracy of preoperative diagnosis of CSF leak with TINF
- Accuracy of intraoperative localization using TINF
- Protocol of applying TINF
- Complications

Assessment of Outcomes

To assess the outcomes, data were divided into two categories: preoperative diagnosis and intraoperative localization. For each type of diagnosis, the sensitivity, specificity, positive prediction rate, negative prediction rate, and cause of the CSF leak were examined.

Assessment of Risk of Bias in Included Studies

The methodologies of the included studies were evaluated for bias. Since all included articles were case series, we used the Joanna Briggs Institute (JBI) critical appraisal checklist tool for case series (21).

Registration

This systematic review protocol was registered in the Prospero database with ID: CRD42020186463.

Results

Results of the Search

We found 266 articles by primary research using PubMed, the Cochrane Database, Scopus, and Ovid. After removing duplicate articles, 109 remained. The titles and abstracts were then screened by two authors independently for relevant articles based on the inclusion and exclusion criteria, and we selected eight for full-text review. Out of these, three were excluded: two had no full-text English version, and one did not meet the inclusion criteria. The five remaining articles included were all reports of case series. A flow chart of the study selection process is shown in Figure 1.

Design

All articles included in this analysis are case series. Three of them describe preoperative diagnosis and intraoperative localization of a CSF leak using TINF, while the other two report only intraoperative localization.

Sample Sizes

In total, the included case series in this review had a small sample size of 94 patients for analysis.

Patient Characteristics

Ninety-four participants were included in this review and had equal gender distribution with 47 patients in each group. The age ranged from 14–68 years with an overall mean of 39.5 years. The most common etiology of a CSF leak among patients was a spontaneous leak (55.3%) followed by trauma (29.7%) and iatrogenic causes (15.9%). We counted 100 sites of CSF leaks, which exceeds the total number of cases as three patients had revision surgery, and three suffered from bilateral leaks. Patient characteristics and distribution of defect sites are shown in Table 1.

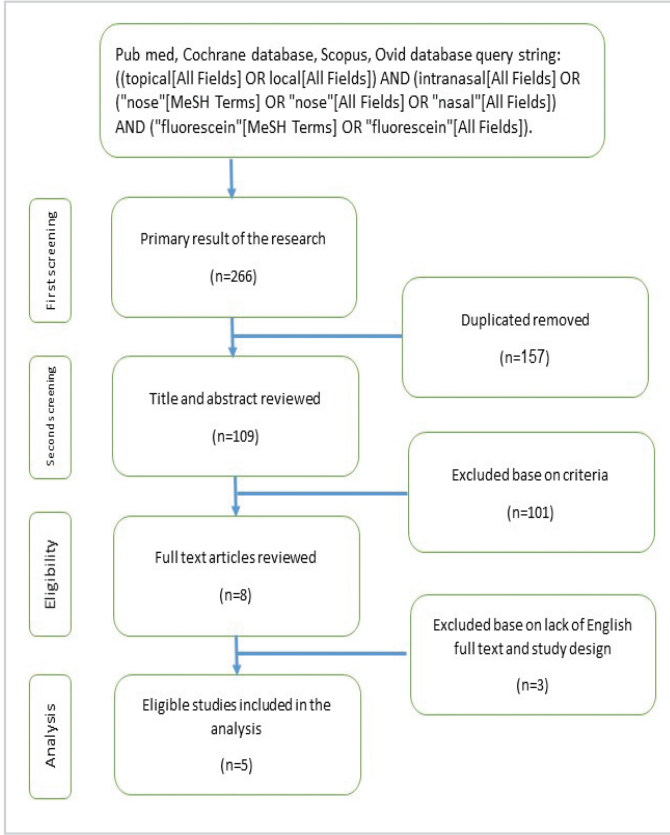

Figure 1. Flow chart of the process for search results and selecting studies for inclusion

Table 1. Clinical characteristics of patients included in the review

Number of patients	**(n=94)**
Sex (male)	47 (50%)
Age	(n=94)
Mean	39.5
Cause of CSF leak	
Spontaneous	49
Traumatic	28
Iatrogenic	15
Meningoencephalocele	2
Site of CSF leak	n=100
Ethmoidal roof	59
Cribriform plate	31
Sphenoid	9
Frontal	1

CSF: Cerebrospinal fluid

Intervention

There was no consistent regimen of using the fluorescein intranasally. However, all studies used plain fluorescein 5%–10% and applied directly into the nose. A test was considered positive for the presence of CSF when the fluorescein color changed from orange to green. When we excluded the study of Jones et al. (22), which does not specify the fluorescein concentration and includes only three cases, we found 40 participants who received 5% of fluorescein, and 51 who received 10% fluorescein. Using the chi-square test which yielded a p-value of 0.58, we observed no significant difference between the two groups concerning the detection of CSF leak sites.

Protocol for Preoperative Diagnosis

Most authors agreed on applying fluorescein intranasally with cotton pledgets after anesthetizing the nasal mucosa. There was no agreement on the local anesthetic or decongestant used. Cotton pledgets were placed with a rigid nasal endoscope at the middle meatus, the roof of the cribriform plate, and the sphenoethmoidal recess.

Protocol for Intraoperative Localization

Various endoscopic sinus approaches were used to access the CSF leak, according to the site. Once the endoscope reached the suspected site, TINF was applied using cotton pledgets. A color change from brown to green fluorescence and sometimes streaming of the fluorescein over the nasal mucosa and blood denoted the presence CSF. If CSF was suspected to be at a specific site, the area was irrigated with saline to clean the fluorescein applied.

All authors of the included reports used the regular light source without a blue light filter to detect the fluorescein color change.

Outcomes

Topical Fluorescein for Preoperative Diagnosis of CSF Leaks

Among the included studies, only three discussed preoperative diagnosis using TINF. Sixty-four cases were tested, and a diagnosis was successfully attained in all of them with 100% sensitivity. All the included articles were case series describing cases of confirmed CSF leaks. However, there were no cohort- or case-controls to assess the specificity of TINF in rolling out CSF leaks (Table 2).

Topical Fluorescein for Intraoperative Localization

TINF was used in all 94 cases to detect the CSF leak site. Except for three cases, the leak's location was successfully identified intraoperatively, achieving a sensitivity of 97%. It is unclear from the reports whether the leak sites were not visible without fluorescein supplementation and were found with fluorescein support intraoperatively. The exceptions were the three cases presented by Jones et al. (22), where the leak sites were identified after application of TINF. Specificity was not calculated since the case would not have been taken for intraoperative localization and repair without confirming the diagnosis at a prior stage (Table 2).

Complications

No complications were reported with TINF application. Thus, according to our review, using plain fluorescein 5%–10% intranasally to detect CSF leak was safe.

Quality Assessment and Level of Evidence

Using the JBI assessment tool for case series, we checked the included articles for bias. They all generally shared the same level of bias: high selection bias on including patients of high clinical suspicion for CSF rhinorrhea, low bias in measurement outcome, low bias of result reporting, and low bias in missing data.

According to this review based on five case series describing the use of TINF (Level 4 evidence), the aggregate grade of recommendation is C.

Meta-Analysis

We could not proceed to meta-analysis because of the lack of a standardized cohort and randomization.

Discussion

The available and commonly used methods for diagnosing a CSF leak have some limitations. Some tests, such as glucose testing, are unreliable (23, 24). Both beta trace protein

Table 2. Summary of the studies included in the review

Date	Author	Study type	Sample size	Causes of CSF leak	Number of Preoperative diagnoses	Number of intraoperative localizations	Formula used for topical fluorescein	Sensitivity for Diagnosis	Sensitivity for localization
2000	Jones et al. (22)	Case series	3	1 spontaneous 2 post-surgery	------	All	5%–10% intranasal fluorescein	-----	100%
2006	Saafan et al. (19)	Case series	25	11 traumatic 9 spontaneous 5 iatrogenic	All	All	Intranasal 5% fluorescein	100%	100%
2009	Liu et al. (29)	Case series	15	10 spontaneous 5 traumatic	All	All	Intranasal 5% fluorescein	100%	100%
2012	Ozturk et al. (30)	Case series	24	10 traumatic 12 spontaneous 2 meningoencephalocele	All	23	Two cotton pledgets soaked with 10% fluorescein	100%	95.8%
2019	Eren et al. (31)	Case series	27	17 Spontaneous 2 Head traumas. 8 Iatrogenic.	------	25	10% fluorescein	------	92.5%

CSF: Cerebrospinal fluid

and beta-2 transferrin tests need to be performed in a specialized laboratory, which may not be always feasible (6). Although these might be inexpensive (approximately $20 and $37.9 for beta trace protein and beta-2 transferrin, respectively), the samples need to be sent to tertiary labs for analysis and sometimes across borders. Days to weeks may pass before results come in. Unlike the proteins, radiological tools are expensive, ranging from $280 for HRCT to $807 for MRC (6). On the other hand, an ampule of 10% fluorescein costs only $1.7 (25), and is widely available, as it is used in many ophthalmic procedures and brain tumor surgeries (26).

In areas with limited resources, alternative methods to diagnose CSF leaks have been used. These methods, such as skull X-rays or Valsalva and squatting maneuvers, are not standardized and thus have unknown efficacy rates (27, 28). Our review shows that TINF offers a feasible method and can be applied quickly and easily. Moreover, TINF is the only suggestive tool available for both preoperative diagnosis and intraoperative localization of CSF leaks. Although it does not substitute the gold standard tools, it should be considered in situations where such tools are not available.

Intraoperative localization of CSF leak is not straightforward. The most important advantages of using TINF over IF for intraoperative localization are safety and rapid applicability. Nonetheless, IF is a more reliable and preferable method based on its high sensitivity and specificity rate. It remains however, an invasive technique as it requires lumbar puncture, and is time-consuming and less safe than TINF, and causes discomfort to the patient (18).

One of the limitations of our review is that only case series were included. Case series can increase the evidence level, strength, and credibility of a review. However, these advantages must be balanced against the risk of bias associated with the lack of a control group. Another drawback is that none of the case series studied the efficacy of TINF in ruling out a diagnosis of CSF. Hence, the specificity of the test was not calculated in this review.

Therefore, TINF has an insufficient grade of evidence for it to be recommended in current clinical practice. More control trials would help achieve a higher level of evidence for TINF implementation.

Conclusion

This review explores the application of TINF to detect CSF leaks. The available evidence showed a low level of recommendation. To increase the amount of

unbiased evidence, thereby provide more legitimate support, standardized control trials should be conducted.

Ethics Committee Approval: This systematic review protocol was registered in Prospero data base with ID: CRD42020186463.

Informed Consent: Not applicable since the study conducted through the databases PubMed, the Cochrane Database, Scopus, and Ovid.

Peer-review: Externally peer-reviewed.

Conflict of Interest: No conflict of interest was declared by the authors.

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Authorship Contributions

Conception: H.A., S.A., Mo.A., Sa.A., A.A., Design: H.A., M.A., Sa.A., A.A., Supervision: Sa.A., A.A., Data Collection and/or Processing: H.A., M.A., Analysis and/or Interpretation: H.A., M.A., S.A., Mo.A, Sa.A., A.A., Literature Review: H.A., M.A., Writing: H.A., M.A., Critical Review: S.A., Mo.A., Sa.A., A.A.

Main Points

- TINF is a test used to diagnose and localize CSF leak.
- Fluorescein 5%–10% is applied directly into the nose.
- The test result is considered positive when fluorescein color changes from orange to green.
- There is insufficient evidence to recommend using TINF in current clinical practice.
- TINF can be considered when other gold standard tools are not available.

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A Patient with Severe Cervicofacial Subcutaneous Emphysema Associated with Munchausen's Syndrome: A Case Report

Case Report

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Abstract

Subcutaneous cervicofacial emphysema is a rare and life-threatening condition that results from various causes. In this report, we documented a case of a patient with severe subcutaneous cervicofacial emphysema a condition that falls under the umbrella of Munchausen's syndrome and discussed the workup of this patient. Thorough diagnostic investigations seeking the etiology of the condition proved unsuccessful. When faced with cases of recurring subcutaneous cervicofacial emphysema, where the root cause remains ambiguous a diagnosis of Munchausen's Syndrome should be considered.

Keywords: Munchausen syndrome, subcutaneous emphysema, neck, pediatric otorhinolaryngology, case report

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Introduction

Munchausen's Syndrome (MS) was initially documented by Asher in 1951 (1). Sufferers of the syndrome are characterized as being wholly conscious persons who falsify physical or psychological signs in order to appear sick. Self-induced subcutaneous emphysema, a rare form of MS was first observed by Gershwin in 1971 (2). The following report outlines a case of an adolescent with MS who required aggressive management due to multiple subcutaneous cervicofacial emphysema.

Case presentation

The patient, a 16-year-old female complained of recurrent instances of swelling on her neck, chest wall, and left arm. Her medical history included two surgeries under general anesthesia with orotracheal intubation; septoplasty (one

year prior) and nasal septal perforation repair (two months prior). After septoplasty, she had recurrent epistaxis episodes for six months which were treated with nasal packings when needed. She eventually had a nasal septal perforation and was referred to another tertiary care hospital by her primary surgeon. There she underwent her second surgery under general anesthesia which involved septal perforation repairment. After this surgery she started having epistaxis episodes which were treated with cauterization and nasal packing.

One month following the septal perforation repairment surgery, the patient suffered her first episode of subcutaneous emphysema on her left arm, chest, and neck. The chest computed tomography (CT) revealed pneumothorax and subcutaneous emphysema. The pneumothorax was treated with chest tube insertion. The patient underwent an endoscopic examination of head and neck, as well as bronchoscopy to discover the etiology of pneumothorax and subcutaneous emphysema, which revealed normal findings. The subcutaneous emphysema regressed slowly and disappeared. Two months after the first episode of emphysema, she was referred to the pediatric surgery department at our hospital with swelling on her neck and on her upper part of the chest. She underwent a detailed investigation, involving several consultations with otolaryngology, dentistry, pediatric gastroenterology, infectious disease, allergy and dermatology departments, however, these provided little clarity into the patient's condition. All initial laboratory tests (erythrocyte sedimentation rate, blood count, urine tests, liver and kidney function tests) and cultures (blood, nasal cavities, nasopharynx, lung aspiration and throat for bacteria) were essentially normal. Oral examination was negative for perforated ulcers or dental abscess, and ear-throat-nose examination did not unearth any irregularities. CT scan of the chest excluded pneumomediastinum and other pulmonary pathologies that could result in subcutaneous emphysema. A subsequent endoscopy did not reveal any irregularities. The patient had spontaneous resolution of symptoms and was discharged from the hospital. Two months following her discharge the patient presented with bilateral massive periorbital edema and subcutaneous emphysema on her neck. She was hospitalized and transferred to our department. Panendoscopy of head and neck and bronchoscopy were performed for a second time and the examinations were inconclusive. CT of the chest and neck revealed subcutaneous emphysema on her neck and face (Figure 1). The patient was subjected to a second course of laboratory tests and with cultures being taken again and similarly to the first round the results did not unearth pathological signs.

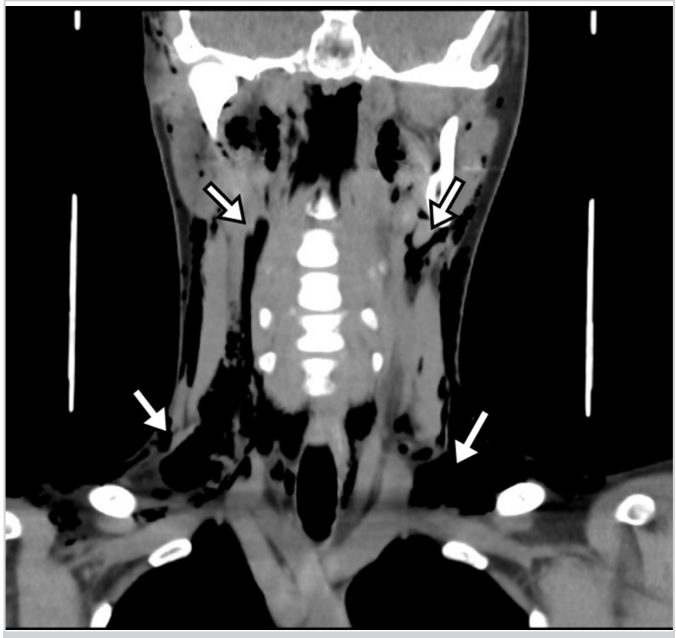

Figure 1. Computed tomography scan of the patient reveals the presence of extensive air (arrows) within the fascial planes of the head and neck

After a detailed inquiry into her previous surgeries, the parents consented our team to contact the surgeon who had previously operated her for nasal septal perforation. The previous surgeon mentioned that he was suspicious that the recurrent epistaxis episodes and the resulting nasal septal perforation, as well as the recurrent epistaxis episodes after the perforation repair, could be due to self-induced digital trauma since no underlying hematologic condition was detected.

With the etiology proving elusive, self-injected air was deliberated as a potential diagnosis and the psychiatry department was consulted. Mounting suspicion that the illness may be self-inflicted meant that a stricter watch of the patient was required. The risk posed by misdiagnosis and to prevent the administering of unnecessary surgical procedures hospital staff carried out a search of the patient's belongings. The search uncovered four syringes as well as blood-stained needles. The patient was referred to the department of psychiatry. Based on her recurrent negative clinical and laboratory evaluations and evidence found in the search, a diagnosis of MS was suspected which would indicate her symptoms resulted from self-abusive behavior and would explain her sustained exaggeration nature of said symptoms. When the patient was confronted with the evidence outlining the justification for her diagnosis, she became agitated and uncooperative, finally discharging herself from the hospital against her caregivers' wishes. The patient was discharged leaving behind a high medical bill consisting of 40 days of hospitalization, two surgeries under general anesthesia and eight CT scans of head, neck and chest region. Monthly follow-up appointments with psychiatry outpatient continued for a duration of six months and these confirmed the diagnosis of MS.

Discussion

There are few reports in literature that discuss subcutaneous emphysema arising from MS. Koufagued et al. (3) reported a case who presented with subcutaneous emphysema of the shoulder girdle and the right arm. The treatment included a wide surgical debridement, hyperbaric oxygen therapy, and parenteral antibiotic therapy. Tosun et al. (4) recorded an episode of subcutaneous cervicofacial emphysema that stemmed from MS. The patient underwent a tracheotomy to secure the airway. Yucel et al. (5) forwarded a case of an adolescent with orbital emphysema. The authors performed a tracheostomy, anteroposterior nasal packing. In total 41 consultants were involved in administering treatment to the patient across the eight months the patient was hospitalized. A case of a woman aged 36 who presented with swelling of left orbital, pain, vision loss, diplopia, fever, and prostration associated with MS was reported by Greene et al. (6). The common denominator in all these cases is recurrent emphysema, the cause of which could not be found, and the patients were diagnosed with MS. In the present case, the subcutaneous emphysema recurred three times, moreover, no etiological factors were found.

Studies into factitious disorder demonstrate the impact unnecessary investigations, treatments, and hospital admission have on the health care system as evidenced by our case (7).

The patient underwent several surgeries, consultations and radiological assessments that resulted in unnecessary exposure to general anesthesia and radiation, as well as a high medical bill.

The literature detailed cases involving children where the deception was discovered either because they were so obvious even after a brief consultation or because health care workers at the hospital carried out a search of the patient's belongings after considering all other diagnoses (8). In the present case, hospital staff carried out a search of the patient's belongings. The search uncovered four syringes as well as blood-stained needles giving rise to suspicions of MS.

A review on pediatric factitious illnesses confirmed the mean age of the children to be 13.9 years, with the range spanning years 8 to 18. The authors noted that the average length of deception was shorter amongst the younger cohort of children that being (12.8 months) compared with the older group (18.9 months). Furthermore, older children, as well as adolescences' medical deceptions, included ingesting steroids leading to an onset of factitious Cushing's Syndrome or by injecting air and thereby causing subcutaneous facial emphysema. Evidence also points to the introduction of eggs and other foreign materials into their bladders to induce proteinuria as well as used tourniquets and feculent urine to cause swelling and wrist pain. In addition, at least 45% of older children stubbornly maintained their denial even when confronted with concrete evidence (8). In the present case, the adolescent did not own up even when presented with the evidence that was full proof.

Conclusion

MS offers a diagnostic predicament that requires increased medical and social attention. Furthermore, it is a condition that demands sustained research, and more effort should be made to spread awareness not only amongst the public at large but also to health care providers. Moreover, early recognition of MS is essential in order to restrict wastage of healthcare resources and reduce patient suffering. When a thorough diagnostic investigation fails to ascertain the cause for a patient's recurring symptoms, a diagnosis of MS should be considered.

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Main Points

- If detailed diagnostic investigations fail to reveal the cause of subcutaneous emphysema, a diagnosis of MS should be considered.
- The correlation between anamnestic data and clinical and para-clinical examinations are essential for the diagnosis of MS.
- Early recognition of MS is paramount to spare the patient from needless and potentially harmful diagnostic tests as well as saving hospitals from unnecessary financial outlays.

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An Incidental Chorda Tympani Schwannoma Identified During Middle Ear Surgery

Case Report

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Abstract

Chorda tympani schwannomas are rare benign tumors of the middle ear cleft. This is a case of incidental chorda tympani schwannoma identified intraoperatively. The patient was a 50-year-old male with chronic active otitis media complicated by left-sided facial paralysis. During closed mastoidectomy and tympanoplasty approach, a well-demarcated swelling on the left chorda tympani nerve was identified and sectioned for pathologic evaluation. The histopathologic evaluation established the diagnosis of schwannoma. Although the tumor was unlikely the cause of the disease process in this patient, his facial paralysis and middle ear disease were resolved after surgery.

Keywords: Chorda tympani, schwannoma, facial nerve, middle ear tumor, chronic otitis media, case report

This study was presented as a poster at 39th Turkish National Otorhinolaryngology, Head and Neck Surgery Congress on November 8th-12th, 2017, Antalya, Turkey.

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Introduction

Schwannomas are the most common tumors of the facial nerve in the middle ear cleft. They can arise from any segment of the facial nerve or its branches (1). Although isolated chorda tympani schwannomas (CTS) are extremely rare, there are several reported cases in the English literature (2-4). The presented case is an incidental CTS identified during middle ear surgery for chronic otitis media. Clinical and histopathological characteristics of CTS are reviewed in this report.

Case Presentation

A fifty-year-old male presented to our clinic with a 12-day history of left facial asymmetry. He had a three-month history of foul-smelling ear discharge. He used oral ciprofloxacin two times a day for 10 days, topical 0.3% ciprofloxacin and topical 0.1% dexamethasone three times a day for three weeks, but his ear discharge persisted. He had no vertiginous symptoms, taste disturbances, or any previous history of facial asymmetry.

On physical examination, he had left-sided House-Brackmann grade 5 peripheral

facial paralysis. Otomicroscopic examination revealed purulent ear discharge, a bulging mass beneath the posterior quadrant of the tympanic membrane, and a millimetric central perforation just anterior to it (Figures 1a and b). A pure-tone audiogram showed a left-sided conductive hearing loss with an average bone and air conduction thresholds of 15 dB and 29 dB at 500, 1,000, 2,000, and 4,000 Hz frequencies. Electroneurography demonstrated more than 90% degeneration of the left facial nerve. Temporal bone computed tomography (CT) without contrast showed soft tissue density in the middle ear and mastoid cavity without an identifiable mass lesion. The fallopian canal was intact. Scutum erosion was not present. A closed mastoidectomy was planned to relieve mastoid infection and its burden on the facial nerve.

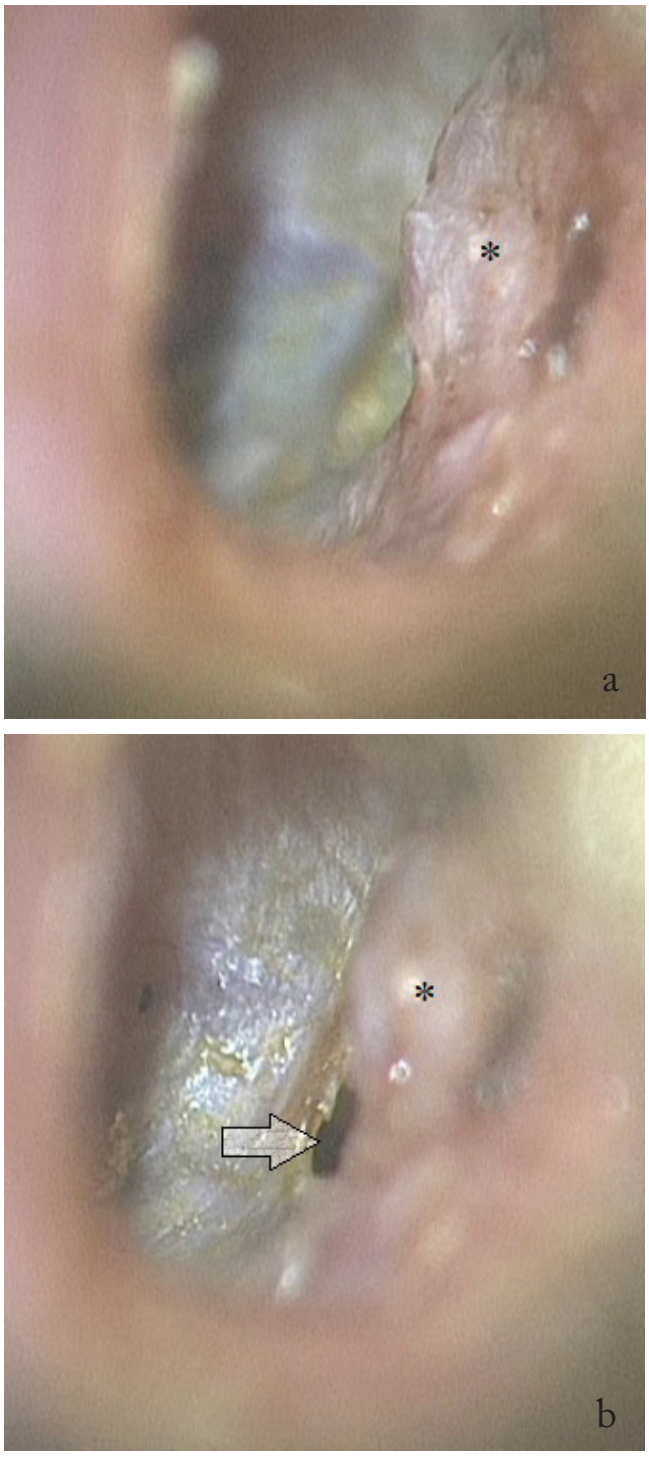

Figure 1. Otomicroscopic view of the patient is showing: (a) a posterosuperior bulge on the left tympanic membrane (asterisk), (b) and a millimetric central perforation (arrow)

After elevating the tympanomeatal flap, the surgeon detected that the 1-2 mm segment of the chorda tympani nerve was thickened, which was corresponding to the bulge on the tympanic membrane (Figure 2). The remaining segments of the nerve were normal in size. The fallopian canal was intact. The mastoid cavity was full of inflamed mucosa and exudate, thus mastoidectomy with an intact canal wall was performed. After obtaining verbal consent from the next of kin of the patient, thickened segment of the chorda tympani nerve was excised and sent for histopathologic examination.

Histopathologic evaluation of the specimen demonstrated proliferating spindle cells with a round nucleus, dense chromatin, delicate eosinophilic cytoplasm, and indistinct cell borders in the sections (Figures 3 and 4). There was no cellular atypia, nuclear pleomorphism, or necrotic areas on the specimen. Tumor cells showed diffuse and strong S-100 positivity in the immunohistochemical staining (Figure 5). These histopathological findings confirmed the diagnosis of schwannoma.

During follow-up, the patient's facial paralysis completely resolved two weeks after surgery. Tympanic membrane

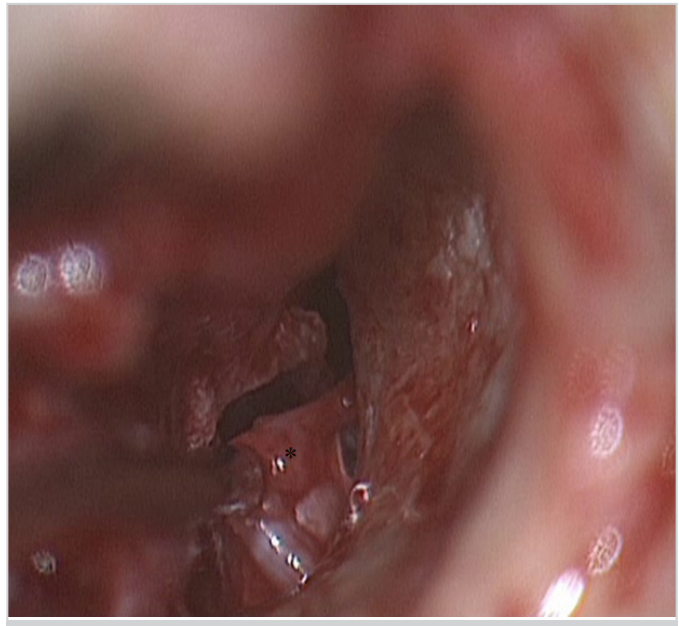

Figure 2. Intraoperative view of the thickened segment of the chorda tympani nerve at the tip of the suction tube (black asterisk)

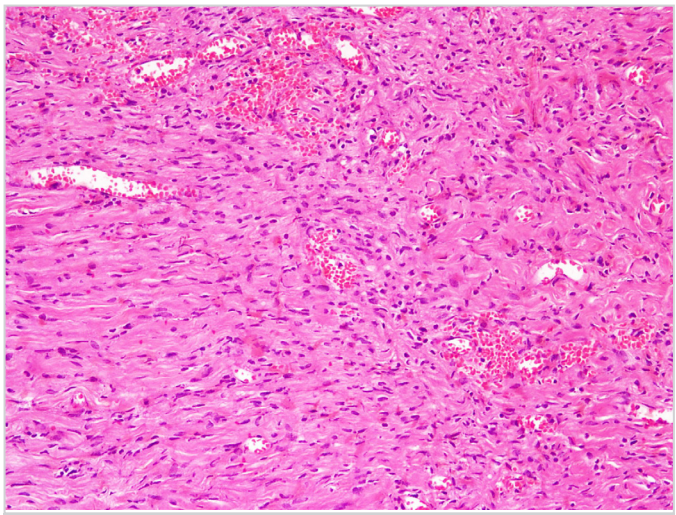

Figure 3. A histopathological section of the mesenchymal tumor is shown. The tumor is composed of uniform spindle cells accompanied by irregularly spaced vessels (x200, H&E)

H&E: Hematoxylin and eosin

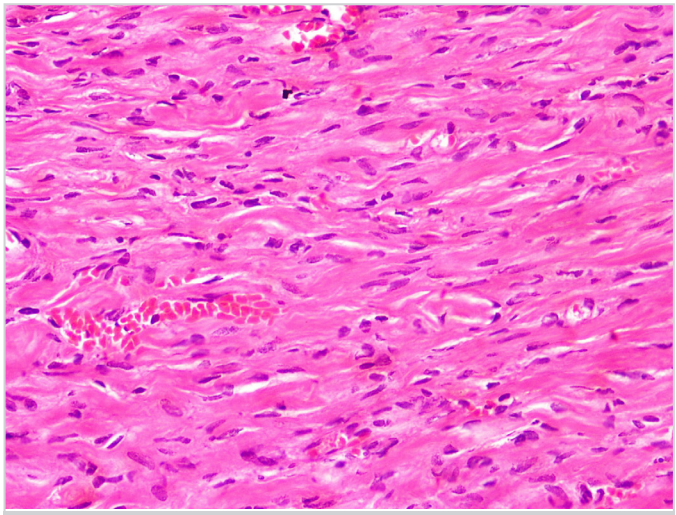

Figure 4. Narrow, elongated, and wavy cells with tapered ends are interspersed with collagen fibers (x400, H&E)

H&E: Hematoxylin and eosin

perforation was closed, and there were no signs of ear discharge. The patient did not report any dysgeusia or hearing loss.

Discussion

Facial nerve schwannomas are benign, slow-growing tumors that destruct the surrounding structures. Expanding tumoral mass fills up the middle ear and the mastoid cavity and causes symptoms. They may be confused with cholesteatoma, paraganglioma, adenoma, meningioma, or other middle ear tumoral masses (1, 5). As the tumor size was very small in this case, our initial diagnosis was also chronic otitis media,

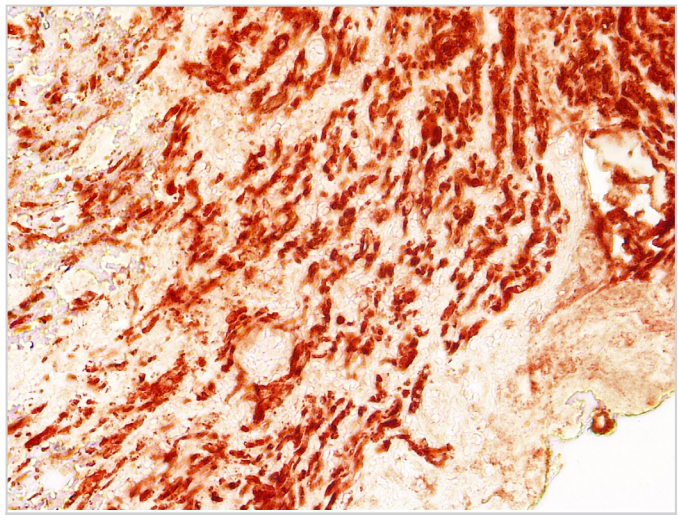

Figure 5. The tumor cells are stained positive for S100 (immunohistochemistry, x400)

and we were not expecting to encounter any type of tumor in the middle ear.

Schwannomas may arise throughout the facial nerve or from the branches, such as chorda tympani and nerve to the stapedius muscles. CTS is quite rare. Patients present with slowly worsening conductive type hearing loss, tinnitus, and rarely facial weakness, or paralysis. Since facial paralysis is rarely appreciated in initial presentation and is mainly seen due to the mass pressure effect on the facial nerve itself; some authors advocate CTS as a distinct subgroup of facial nerve schwannomas (6, 7). Accordingly, asymptomatic CTS cases have been reported in the literature (2). Our patient had facial paralysis and purulent ear discharge in presentation, however, we believe such a small tumor was unlikely responsible for the facial paralysis. Due to the slow-growing nature of CTS and compensatory mechanisms, gustatory dysfunction has not been reported (2, 3, 5). As in our case, gustatory dysfunction is rarely appreciated even after complete resection of the nerve (2, 5).

On otomicroscopic examination, a bulge on the posterosuperior quadrant of the tympanic membrane may be seen in CTS (5, 8). In our case, the presence of chronic otitis media concealed the diagnosis of middle ear mass, and there was no identifiable mass lesion on CT. Normally, osteolytic erosion of the ossicles and temporal bone with space-occupying soft tissue density along the course of the chorda tympani is observed in the CT scan (6, 7). Gadolinium-enhanced magnetic resonance imaging helps to better delineate the extent of the tumor and its relation with the facial nerve (8). Furukawa et al. (9) developed a new technique that combined CT and magnetic resonance imaging (MRI) images based on non-rigid registration and showed the benefits of this imaging in tumor localization

and surgical planning. Both of the imaging techniques are complementary with each other and help the surgeon with preoperative planning (6, 10). As our initial diagnosis was chronic otitis media, we did not request MRI imaging.

Middle ear adenoma, paraganglioma, and meningioma are in the differential diagnosis of schwannoma during the histopathologic examination (8). Middle ear adenomas show variable patterns of glands/tubules composed of a uniform single layer of columnar cells with round hyperchromatic nuclei and eosinophilic cytoplasm. Immunohistochemically, these lesions are stained positively for pan-cytokeratin, and sometimes neuroendocrine differentiation can be seen. Paragangliomas have central, oval chief cells with a nested pattern, containing uniform nuclei with scattered chromatin and abundant eosinophilic granular cytoplasm. Chief cells are stained positively with chromogranin and synaptophysin. Sustentacular cells are stained positively with S100, which is difficult to see with hematoxylin-eosin staining. They settle at the periphery of the nests. Meningiomas show a whorls growth pattern, and they have oval or spindle cells with pale cytoplasm, intranuclear cytoplasmic inclusions, and indistinct cell borders. They may contain psammoma bodies. These tumors are EMA positive and S100 negative (3). In our case, the tumor cells were diffuse and strongly positive with S100; but negative for pan-cytokeratin, chromogranin, and synaptophysin.

Although there is no consensus about the best treatment strategy, restoration of hearing and maintaining normal facial motor functions are the mainstays of treatment for facial nerve schwannomas. In cases of facial paralysis due to the tumor itself, complete tumor excision with cable grafting is recommended (1). There is no need to restore the integrity of the chorda tympani nerve due to compensatory mechanisms. So, the tumor can be excised, or small asymptomatic tumors may be followed.

In the presented case, the facial paralysis was due to chronic otitis media, since CTS was very small in size, and not in the fallopian canal. Simple biopsying of the lesion was not possible. The small size of the tumor and the presence of chronic otitis media might complicate follow-up for this patient. Secondary surgeries would be required. So, we chose to excise the tumor completely.

Conclusion

Surgeons should be cautious about the underlying tumoral mass in cases of chronic otitis media and request imaging with contrast if suspected. Patients should be informed about the possibility of a middle ear tumor, therefore the decision to observe or excise can be made preoperatively.

Informed Consent: Informed consent was obtained

Peer-review: Externally peer-reviewed.

Authorship Contributions

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Main Points

- Chorda tympani schwannomas are rare benign tumors of the middle ear cleft that may present with a bulge on the posterosuperior quadrant of the tympanic membrane.
- The otologic surgeons should be prepared for the incidental middle ear tumors which may also cause facial paralysis and chronic ear discharge.
- MRI with contrast and CT may be used together to better delineate the tumoral mass.

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Immunocompetent Young Patient Presenting with Unilateral Lip Abscess Due to Peeling Exfoliated Lip Skin

Case Report

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Abstract

Lip abscess is a rare condition encountered in clinical practice. Generally, it may be due to an infective agent, such as virus, bacteria, entering through a skin wound, or it can be seen through hematogenous spread when there is a serious underlying condition such as a general condition disorder or immunodeficiency. It requires rapid diagnosis and treatment as it may cause significant complications in terms of localization and lymphovascular drainage. In this case report, an 18-year-old male patient with unilateral lip abscess that regressed rapidly with external drainage and antibiotic therapy is presented with imaging and clinical-laboratory findings.

Keywords: Lip, abscess, edema, lip abscess, immunocompetence, deep neck infection, case report

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Introduction

Sudden painful lip swelling is a rare condition that we see in clinical practice. Lip swelling usually develops secondary to traumatic or allergic conditions. Infectious agents such as bacteria and viruses are the second most common cause of swollen lips (1). Herpes labialis or Herpes zoster should be considered especially in unilateral swelling accompanied by pain (2). In such viral infective cases, apart from systemic examination, the oral mucosa should also be examined, and the patient should be investigated for the presence of vesicles-ulcers (3, 4).-Tumors of vascular origin such as hemangioma and lymphangioma or malignant tumors may also be the cause of swelling or asymmetric discoloration of the lips. In this case report, we aimed to provide clinicians a perspective in terms of lip swelling and lip infections by presenting the case of a young patient with unilateral lip abscess which regressed rapidly in a very short time with antibiotherapy and drainage.

Case Presentation

An 18-year-old male patient was admitted to our dermatology outpatient clinic with

complaints of swelling, pain, redness in the right half of his lip. The patient who had widespread pustular acne on his face was questioned about his history of medication use, such as isoretinoic acid, that could cause infection by mucosal integrity loss. The patient stated that he did not use any medication. The patient, in whom empirical antibiotherapy and antiviral treatment (amoxicillin, valasiclovir) was initiated by the dermatology doctor, was admitted to the Ear-Nose-Throat outpatient clinic three days after the increase in lip swelling and the onset of systemic complaints such as fever and weakness.

Physical examination revealed extremely edematous erythematous lips, more prominent in the right half of the lower lip, and multiple palpable lymphadenopathies (LAPs) in the right cervical chain (Figure 1a). In the evaluation of the oral and mucosal structures, there were no findings other than erythema and swelling on the lips. Initially, laboratory tests were requested to investigate infective causes and the patient was referred for ultrasonography. In ultrasonographic examination, diffuse thickening, more prominent in the right half of the lip, and marked increase in vascularity, as well as a heterogeneous non-locular collection with a dense content compatible with abscess was observed (Figures 2a-b). Multiple LAPs with thickened cortex were detected in the right cervical chain at levels 1-2-3-4. Linear effusions consistent with edema were observed in the right half of the face and around the lips under the skin. Allergic causes were not expected since the patient's complaints had onset only three days ago, but immunological tests were also requested. However, when findings in favor of an abscess were described in ultrasonographic examination, the tests were canceled to focus on infection. Culture antibiogram was not taken since empirical antibiotherapy had already been started and the patient was using antibiotics for three days. In laboratory tests, leukocytosis (15.8x10 µL) was detected in neutrophil dominance (80%) with erythrocyte sedimentation rate (ESR: 41 mm/h) and high CRP (7.5 mg/dL). While HSV 1, HSV 2, VZV IgG antibodies were positive, IgM antibodies were negative. The patient stated that he had been picking and peeling his dried lips for a long time. The condition of the patient—who had no known chronic disease—was then evaluated as a primary lip abscess due to exfoliating his lips. Drainage was performed with a syringe, and about 10 cc of pus was evacuated and intravenous antibiotic treatment (sultamicillin), oral antibiotics (amoxicillin) and antiviral therapy (valasiclovir) were continued for two days. Complaints of the patient, who also had spontaneous drainage during follow-up, regressed almost completely within three days and the lip returned to its normal state (Figure 1b). The patient provided his signed informed consent for publishing his data and images.

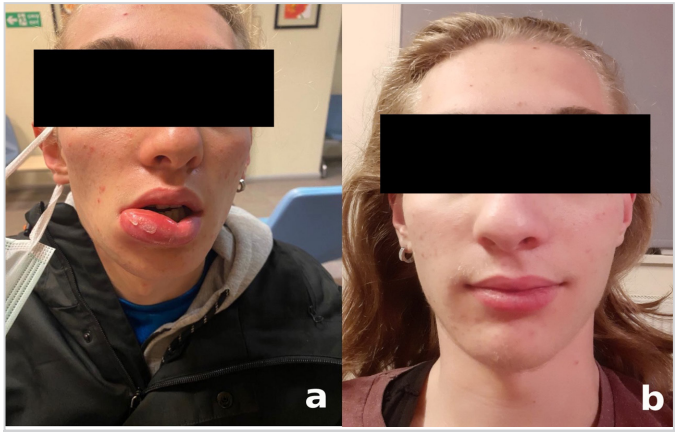

Figure 1a. Extremely edematous and erythematous appearance in the right lateral of the lower lip. Note the desquamation and peeling of the lip skin. b. View of lip that quickly returned to normal on the 3rd day of treatment

Discussion

In this case report we present a very rare cause of lip swelling in the form of a lip abscess that was detected in a young patient with localized involvement, together with clinical, laboratory, imaging findings. We emphasize the path to follow in differential diagnosis, follow-up and treatment. Viral agents such as Herpes and Varicella, which frequently affect the mouth and the face, are among the most common infective causes of lip swelling (2). Pain is more prominent than swelling and redness, especially in Herpes zoster. Cases can even be confused with trigeminal neuralgia (5). In these

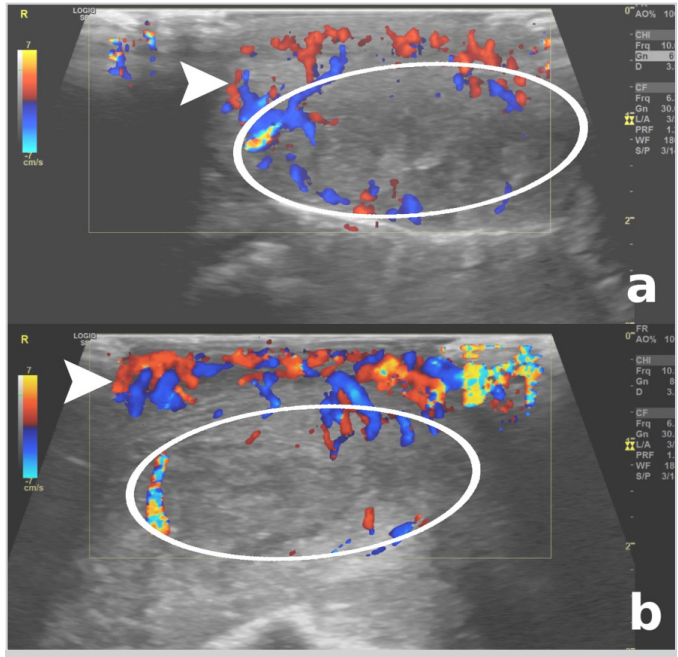

Figure 2a. In the ultrasonographic examination of the lip, a marked increase in lip thickness, a heterogeneous collection with a dense content compatible with abscess in the central part (circles), and b. increased vascularity in the periphery with Doppler sonography are observed (arrowheads)

cases, vesicles-ulcers should be sought in the oral mucosa or the face during physical examination.

Bacterial agents such as *Staphylococcus aureus* may also cause lip swelling with or without lip abscess (1, 6). Bacterial infections should be kept in mind especially in immunocompromised patients who also have systemic complaints and rapidly progressive bacterial lip swelling. Primary-isolated lip abscess is an extremely rare condition and usually develops in an acute-rapidly progress secondary to bacterial agents. With a very rapid increase in lip sizes, it may lead to necrosis due to vascularization insufficiency (1). The lip becomes very swollen and erythematous, which can be confused with angioedema (7). Angioedema is important in differential diagnosis as it is an immunological entity that causes diffuse edema, progresses very rapidly and can lead to vital consequences, therefore requiring urgent intervention. Skin defects on the lips or face cause infection in soft tissues by opening the entrance door for these agents. In immunocompromised cases, it may spread to the face and neck region and cause deep neck infection (7). Therefore, lip abscess, like other bacterial infections in the head-neck-face region, is a serious problem in terms of complications and mortality, and requires timely and accurate medical intervention.

In the literature, there are reports about cases presenting in the adolescent age group with lip abscesses caused by isotretinoin which is frequently used in the treatment of acne (8). Isotretinoin creates a predisposition for bacterial entry by creating skin dryness and cracks, especially around the lips. Likewise, a rapidly developing lip abscess secondary to an insect bite was reported in a young patient (9). Our case differs from other reported cases in that there was no risk factor.

To conclude, lip infections should be considered in patients with acute lip swelling, and bacterial-suppurative abscesses should be ruled out in rapidly developing conditions. In such acute and rapidly progressing situations, it is crucial to drain the abscess detected in imaging or examination without delay, initiate intravenous antibiotherapy, and obtain culture before antibiotherapy for rapid and specific treatment. With the timely and correct medical approach, it is possible to prevent complications that may even lead to mortality such as deep neck infections.

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Main Points

- Unilateral lip abscess is a rare clinical condition.
- The clinical situation regresses rapidly with early drainage and antibiotic therapy.
- Serious progresses up to deep neck infection may occur in case of delay in treatment.

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Where is my ear? – Cervical Chondrocutaneous Branchial Remnant

Clinical Image

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A 6-year-old boy presented with the absence of pinna on the left side since birth. A cutaneous appendage emerging from the neck on the left side was discovered during the examination (Figure 1). Deep palpation revealed a firm texture like cartilage. On the left side, a primitive firm cartilage was palpated in the helix region (Figure 2). The rest of the cartilaginous framework and external auditory canal (EAC) opening was absent on the left side. On the right side, the pinna was smaller in size and loped. High resolution computed tomography (HRCT) of temporal bone showed an atretic and stenosed EAC on the left and ride side, respectively. Ossicular lump was noted on both sides on HRCT, but all inner ear structures appeared normal in both ears. Pure tone audiometry revealed moderate and mild conductive hearing loss on the left and right sides, respectively. Magnetic resonance imaging of the neck was done to rule out any branchial abnormality. It did not reveal any cyst/sinus/fistula. A provisional diagnosis of cervical chondrocutaneous branchial remnant (CCBR) with left-sided microtia and EAC atresia was made on clinico-radiological basis. After a thorough discussion with the father, the patient was planned for the excision of the cervical mass and staged pinnaplasty and canaloplasty on the left side at a later stage. Histopathological examination after excision showed elastic cartilage rests covered by normal skin consisting of epidermis, dermis, adnexal structures, and subcutaneous fat compatible with CCBR.

The development of six cartilaginous tubercles known as "Hillocks of His" initiates the development of pinna in the sixth week of pregnancy. The first three hillocks are formed by the first branchial arch, while the remaining three are formed by the second branchial arch (1). The lobule is the first external ear component to develop. The growing pinna is first found in the neck, but by the 20th week of pregnancy, it has ascended to its usual adult location and structure (2).

An accessory tragus is formed from the first branchial arch and is usually found along an imaginary line drawn from the tragus to the angle of the mouth, whereas CCBR

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Figure 1. Frontal view showing cervical chondrocutaneous branchial remnant arising from neck on the left side (red arrow)

is derived from the second branchial arch and is found over the lower one-third of the sternocleidomastoid (SCM) (3, 4). However, they may be of auricular origin (due to failure of migration and contain elastic cartilage) or may represent primordial laryngeal remnants of the second and third branchial arches containing hyaline cartilage. They present as painless neck masses resembling skin tags anterior to SCM, with no overlying skin changes and more common in males. Imaging of the neck is done to see the extent of the lesion and to rule out other branchial abnormalities. CCBRs usually do not involve underlying neck structures. However, they may be associated with other congenital malformations involving the genitourinary tract and may warrant ultrasound screening of the kidney and urinary tract. In our case, the patient did not have any other systemic symptoms and his abdominal, renal, and urinary system ultrasound scanning was normal. The differential diagnosis may include fibroepithelial polyp, epidermoid cyst, and squamous papilloma. The treatment is usually surgical excision until the superficial muscular plane. Recurrence and malignant transformation have not been reported in the literature (4, 5).

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Figure 2. Lateral view showing cervical chondrocutaneous branchial remnant (red circle) and a rudimentary cartilaginous structure (accessory tragus?) buried under the skin in region of helix on the left side (blue arrow)

Authorship Contributions

Concept: S.K., Design: S.K., Data Collection and/or Processing: A.A., A.G., Analysis and/or Interpretation: S.K., A.A., A.G., Literature Search: S.K., A.A., A.G., Writing: S.K., A.A.

Main Points

- Pinna develops from six cartilaginous tubercles known as "Hillocks of His".
- Cervical chondrocutaneous branchial remnant usually forms due to defect in migration of these tubercles during embryonic development and can be confused with accessory tragus.
- Treatment is surgical excision till superficial muscular plane.

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