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A comparison of endoscopic and microscopic techniques for the repair of tympanic membrane perforations

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Abstract

Objective: This study aims to compare endoscopic and microscopic tympanoplasty in terms of graft success rate and hearing gain.

Methods: Medical records of 236 patients (99 females and 137 males) aged 18 to 74 years with chronic otitis media who underwent myringoplasty, between January 2014 and June 2017 were retrospectively compared. Patients were divided into two groups; microscopic myringoplasty (140 patients), endoscopic myringoplasty (96 patients). Demographic data, pure tone audiometric results preoperatively and 6 months postoperatively, operation time, hearing gain and graft success rate were evaluated.

Results: The mean operation time was 57.8±9.6 minutes for the endoscopic group and 78.6±17.7 minutes for the microscopic group (p<0.001). The mean preoperative air-bone gap (ABG) was 24.1±11.5 dB for the microscopic group and 22.7±9.2 dB for the endoscopic group, whereas the mean postoperative ABG was 11.6±9.9 and 9.8±9.3 dB respectively. Graft success rate was 90.3% (213 patients) for entire group, 89.7% (131 patients) for the microscopic group, and 91.1% (82 patients) for the endoscopic group (p=0.727). The functional success rate (ABG≤10 dB) was 72.5% (171 patients) in the entire group. The mean hearing gain was 12.4±10.8 and 12.8±9 dB in the microscopic and endoscopic groups respectively.

Conclusion: Endoscopic technique offers similar graft success and hearing outcomes to microscopic technique along with a shorter operation time.

Keywords: Conductive, hearing loss, otitis media, tympanic membrane perforation.

Myringoplasty, which has been used for the closure of tympanic membrane (TM) perforations for about seventy years [1-5], has been traditionally performed with the assistance of an operating microscope through postauricular, transcanal and endaural approaches. While microscopic myringoplasty via a postauricular approach enhances the visibility of the operation field, especially in patients with anterior and large TM perforations or those with a narrow ear canal, this procedure results in incision scars, esthetic problems, and considerable pain for the patient and requires hair shaving and general anesthesia in most cases.[3-6] Furthermore, the straight-line view offered by a microscope limits the visualization and exploration of the hidden regions of the middle ear cavity. Hence, recently, the use of endoscopic instead of microscopic ear surgery has gradually increased. Middle ear endoscopy was first introduced in 1967 by Mer et al [7] for the diagnosis of TM perforation and middle ear disease. In 1975, Willemot [8], for the first time, recorded the middle ear cavity using an endoscope. Since the early 1990s, endoscopic ear surgery procedures have been increasingly per-

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formed in various middle ear surgery. A wide field of view can be obtained by a high-resolution endoscopic system. In contrast to the microscope, the endoscopes enable the surgeon to "look around the corner" with better visualization and access of the hidden areas of the tympanic cavity, such as the anterior epitympanum, attic region, sinus tympani, facial recess, and hypotympanum. Endoscopic surgery is also utilized for educational purposes. The surgeon and the resident viewing the monitor at the same time facilitates the understanding of the surgical anatomy and procedure.[9] In addition, the risk of residual disease and recurrence can be reduced by the use of an endoscope in cholesteatoma surgery.[5,9-11] Furthermore, the endoscopic technique does not require canaloplasty or skin incisions, and thus it is less invasive and painful than the microscopic technique. Therefore, using endoscopes may shorten the operating time as a result of the decrease in the time required for gaining access to the tympanic cavity and closure at the end of the surgery.[12] However, endoscopic ear surgery is a single-handed surgical technique, and it may be difficult to proceed the operation in case of a massive bleeding. It can also damage some middle ear components due to the thermal effect produced by the light source.[13,14]

In this study, we aimed to compare the graft success, hearing outcome, and operating times for patients who underwent endoscopic and microscopic myringoplasty. We then evaluated whether the endoscopic technique offered an advantage over microscopic surgery.

Materials and Methods

The study was carried out during the period from January 2014 to June 2017 and the study protocol was approved by the Ankara Numune Training and Research Hospital ethical committee (No: E-18-2069). Medical records of 236 chronic otitis media (COM) patients (99 females and 137 males) aged between 18 and 74 years, who had undergone tympanic membrane repair in our hospital were reviewed. The data consisted of the patients' age, gender, perforation side, location of perforation (central-marginal, anterior-posterior-inferior), pre- and postoperative audiometry and air-bone gaps (ABGs), operative technique (microscopic, endoscopic), graft uptake, and operative duration. The exclusion criteria were discharging ear, revision cases, COM with cholesteatoma, and cases that underwent ossiculoplasty or mastoidectomy. The minimum follow-up period was 12 months for all patients.

The local ethics committee of our hospital approved the study (no: E-18-2069). The study was performed in accordance with the ethical standards of the institution and the 1964 Helsinki declaration and its later amendments or comparable ethical standards. All patients provided written informed consent.

All surgeries were performed under general anesthesia. The patients were classified into two groups according to the surgical procedure they received: The microscopic group (Möller-Wedel Optical®; Hamburg, Germany) underwent microscopic myringoplasty (146 patients) and the endoscopic group (Karl Storz, Tuttlingen, Germany) underwent endoscopic myringoplasty (90 patients). A postauricular incision was used in the microscopic group. The edges of the perforation were desepithelized in a circular fashion. Then, the canal skin flap was elevated, and the middle ear was visualized. In cases where the anterior canal wall bulging was present, a canalplasty was performed using a diamond drill. Ossicular movement was checked by the observation of the round window reflex. The tragal cartilage was removed and prepared as a graft, leaving the perichondrium intact on both sides.

In the endoscopic group, an endoscopic system and a 0-degree rigid endoscope was used. The edges of the perforation were desepithelized in a circular fashion, and a vertical incision was made 5 mm laterally from the annulus. This incision was integrated with radial incisions at 6 and 12 o'clock. Next, the tympanomeatal skin flap was elevated, and the middle ear was visualized. Ossicular movement was checked by the observation of the round window reflex. The graft harvesting and placement procedures were similar to Group 1.

Pure-tone average audiometry was performed before and after surgery (6 months) using a clinical audiometer (AC 40, Interacoustic, Denmark, Headphone: TDH39). Audiometric data were analyzed for average, standard deviation (SD), and ABG.

Follow-up controls were performed at postoperative months 1, 2, 3, 6 and 12, postoperatively. The anatomic success was defined as the complete closure of TM perforation without medialization, lateralization, or perforation. An ABG of ≤10 dB was accepted as functional success.

SPSS software (version 21.0 for Windows, IBM Corp., Armonk, NY, USA) was used for all data analyses. The descriptive data were given as average±SD. The results of the categorical data were given as percentages (%). Student's

t-test and paired-sample t-test were used to compare the quantitative data with a normal distribution. The comparison of the qualitative data was made by Fisher's exact test. P values less than 0.05 were considered statistically significant.

Results

This retrospective study included 236 COM patients (average age, 36.2±13.2 years) with 236 ears (134 left, 102 right) treated surgically. The average follow-up was 28.9±14.6 months ranging between 12-80 months.

No statistically significant differences were found between the characteristics of the microscopic and endoscopic groups in terms of age, gender, operated side, and location of TM perforation (Table 1).

Table 1: Demographic date of all cases.

Characteristics	Endoscopic	Microscopic	p-value
Age (years)	35.1±13.2	36.8±12.2	0.271
Gender (F/M)	39/51	60/86	0.735
Side operated (L/R)	43/47	59/87	0.267
Location of perforation			
Anterior	43 (47.8%)	57 (39.0%)	0.082
Posterior	32 (35.6%)	73 (50.0%)	
Inferior	15 (16.4%)	16 (11.0%)	0.304
Type of perforation			
Central	69 (76.7%)	103 (70.5%)	
Marginal	21 (23.3%)	43 (29.5%)	

F: female, M:male, L:left, R:right

No statistically significant differences were found between the groups in terms of the graft take rate, which was 90.3%, 89.7% and 91.1% for the entire, microscopic and endoscopic groups, respectively (p=0.727). On the other hand, the duration of operation was significantly shorter in the endoscopic group (57.8 ± 9.6 minutes) than in the microscopic group (78.6±17.7 minutes) (p<0.001) (Table 2).

Table 2. Comparision of Endoscopic and Microscopic groups by graft success and operation time.

Parameters	Endoscopic	Microscopic	p-value
Graft success	82/90 (91.1%)	131/140 (89.7%)	0.727
Operation time (min)	57.8±9.6	78.6±17.7	<0.001

Min: minute

The average preoperative ABG was 23.5 ± 10.7 dB for the entire group, 24.1±11.5 dB for the microscopic group, and 22.7±9.2 dB for the endoscopic group, while the average postoperative ABG was 10.9±9.7, 11.6±9.9, and 9.8±9.3 dB, respectively. There was a statistically significant difference between the groups in terms of the pre- and postoperative ABGs (p<0.001).

The average hearing gain was 12.4±10.8 in the microscopic and 12.8±9 dB in the endoscopic groups. There was no statistically significant difference between groups in terms of hearing gain (p=0.789). The functional success was 72.5% (171 patients), 70.5% (103 patients), 75.5% (68 patients) in the entire, microscopic and endoscopic groups, respectively, and this difference was not statistically significant (p=0.403) (Table 3).

Discussion

The character of ear surgery changed after the introduction of operating microscopes. Today, many otological procedures are performed with both hands under an operating microscope, which provides the surgeon with stereoscopic vision and bimanual handling. However, microscopes may not be sufficient to view the hidden areas of the middle ear cavity or to confirm the circumference of the perforation, especially in the presence of a protruding anterior canal wall.[15] For this reason, recently, there has been an increase in the number of studies on endoscopic ear surgery that compare the two techniques and indicate that otological procedures conventionally performed under a microscope, such as TM reconstruction, repair of ossicular chain defects, cholesteatoma, and stapes surgery could be performed using an endoscope.[9,15-20] Nevertheless, the question of whether endoscopic surgery is entirely preferable over microscopic procedures is yet to be answered.

We compared the data gained from our study with those reported by other authors in order to better analyze the advantages and disadvantages of the endoscopic and

Table 3: Pure-tone audiometry of endoscopic and microscopic group.

Variables	Endoscopic	Microscopic	p-value
Mean preoperative ABG (dB)	22.7±9.2	24.1±11.5	0.319
Mean postoperative ABG (dB)	9.8±9.3	11.6±9.9	0.185
Mean hearing gain (dB)	12.8±9.0	12.4±10.8	0.789
Functional success (%)	68/90 (75.5%)	103/146 (70.5%)	0.403

ABG:air-bone gap, dB:decibel

microscopic techniques. The results demonstrated that although the outcome and success of both techniques were similar, endoscopic surgery involved a significantly less operation time and offered better visualization.

Since zero-degree endoscopes, coupled with the different angled endoscopes, provide a panoramic, wide-angled and clear view of the operating field, the surgeon can precisely visualize the entire TM, ossicular chain, epitympanum, hypotympanum, and retrotympanum compared to the limited view of operating microscopes. With a thin and rigid endoscope, the surgeon can perform minimally invasive and conservative operations by protecting the physiological anatomy, which provides functional reconstruction during surgery.[21]

The decreased operation time is the main advantage of endoscopic ear surgery, which reduces the duration of anesthesia and anesthesia-associated side effects, and improves surgeon's concentration. In the present study, the average duration of operation was 57.8 minutes for the patients that underwent endoscopic myringoplasty while it was 78.6 minutes for the patients that underwent microscopic tympanoplasty, revealing a statistically significant difference between the two groups (p<0.001). Similarly, in their studies, Huang et al [22] reported the main operative time as 50.4 minutes for endoscopic tympanoplasty and 75.5 minutes for microscopic tympanoplasty, whereas Dündar et al [23] calculated it as 51.3 and 67 minutes, respectively. This difference can be attributed to the endoscopic technique requiring no additional time to perform or suture an incision, elevate the vascular flap, or control bleeding.

Another advantage of endoscopic tympanoplasty is that it lessens the requirement of canalplasty or the curettage of the external auditory canal. Karhuketo et al [24] stated that they had to perform canalplasty or the curettage of the external auditory canal for some of their patients undergoing microscopic tympanoplasty while none of their patients who underwent endoscopic tympanoplasty required such interventions. Having compared 60 patients undergoing either endoscopic or microscopic myringoplasty, Lade et al [25] reported that out of the 30 patients in the microscopic group, five required canalplasty. In our study, no patient required canalplasty in the endoscopic group, which is in accordance with the findings of Ayache et al [26] and Harugop et al.[27]

Consistent with previous studies, no statistically significant difference between the two techniques was revealed in our study in terms of the graft success rate, which was 91.1% and 89.7% for the endoscopic and microscopic groups, respectively (p=0.727). Similarly, Dündar et al [23] and Kuo and Wu [28] reported that the graft success rates in their studies were 87.5% and 97.3%, respectively for endoscopic myringoplasty, and 93.1% and 98.2%, respectively for microscopic myringoplasty.

In terms of postoperative hearing gain, numerous studies have reported successful results with both techniques. In our study, the hearing gain was 12.8 dB and 12.4 dB in the endoscopic and microscopic groups, respectively, while functional success was observed in 75.6% and 70.5% of the cases in the endoscopic and microscopic groups, respectively.

Despite all the advantages, the endoscopic technique lacks three-dimensionality, adequate magnification, and focus provided by operation microscopes. Endoscopes offer a two-dimensional view of the surgical field, which leads to the loss of depth perception. However, variable magnification and the insufficiencies of two-dimensional images can easily be compensated by endoscopic surgeons. Another disadvantage of endoscopes is the need to perform one-handed surgery, which makes it difficult to proceed the operation in case of a massive bleeding. Frequent contamination of the operation field, bleeding, and fogging of en-

doscopes may occur during endoscopic surgery, which are not encountered in the microscopic technique. Heat generation caused by the light source of the endoscope is also considered to negatively affect some of the vital structures of the middle ear, for which Kozin et al [29] recommended the use of submaximal light intensity and frequent repositioning of the endoscope. However, in a study by Ozturan et al [30], the authors indicated that the heat generated by all types of endoscopes fell within the safe limits of the ear in clinical settings.

Conclusion

The data obtained from the current study indicated that the anatomic and functional success rates were similar for both endoscopic and microscopic techniques while the endoscopic technique offered a shorter operation time compared to the microscopic method. There was no need for an external incision or auditory canal drilling in the endoscopic technique, which can also lead to higher patient satisfaction. The lack of a three-dimensional view, loss of depth perception, and one-handed surgery are the main challenges in the endoscopic technique that can be overcome by practice. However, further prospective studies with experimental designs are required to suggest that the endoscopic technique is entirely preferable over microscopic surgery.

Ethics Committee Approval: The local ethics committee of our hospital approved the study (no: E-18-2069).

Informed Consent: All patients provided written informed consent.

Author Contributions: Designing the study – i.G, M.Ö.; Collecting the data – i.G, M.Ö.; Analyzing the data – i.G; Writing the manuscript – i.G, M.Ö.; Confirming the accuracy of the data and the analyses – i.G, M.Ö.

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

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Balance disorders and hypothyroidism: A rare cause worth remembering

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Abstract

Objective: This study aimed to evaluate balance disorders and potential risk factors for falling in patients with hypothyroidism, as well as postural stability and the risk of falling.

Methods: The participants' sex, age, body mass index (BMI), falls history in the past one year, and Falls Efficacy Scale - International (FES-I), Neuropathic Pain Diagnostic questionnaire (DN4) and clinical symptoms were recorded. The stability index (SI), weight distribution index (WDI) and falls risk analysis were performed for postural stability by objective computerised dynamic posturography.

Results: 50 patients were matched for age and sex (mean age:41.7±11 years; age range:19-61 years) with 47 healthy (euthyroid) controls (mean age:39±9.6 years; age range:23-60 years). All SI, HL-WDI and falls risk scores, except FES-I, DN4 and NO-SI, were significantly higher in the patient group. In addition, other symptoms were present at a higher rate, with the exception of morning stiffness ($p<0.05$). There was no significant association between sex, age, BMI, FES-I, DN4, serum FT4 and TSH levels, clinical symptoms and WDI values and the risk of falls ($r<0.3$ and $p>0.05$). However, there was a slight positive correlation between the existence of paraesthetic symptoms, a history of falling, and the overall SI and HLWDI values ($r>0.3$ and $p<0.05$).

Conclusions: It was determined that, as postural stability is affected in cases of hypothyroidism, balance becomes disordered and the risk of falls increases. This increase in the risk of falls was associated with being hypothyroid, but not with FT4 or TSH levels or the other factors evaluated.

Keywords: Hypothyroidism, postural balance, falling.

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Hypothyroidism is one of the most prevalent endocrine disorders and has a frequency in the general population of between 0.5 and 1%. Hypothyroidism can affect multiple physiological systems, but especially affects the central nervous system, the peripheral nerves and muscles and may cause various symptoms and signs within the neuromuscular system. These signs range from various myopathies and entrapment mononeuropathy causing neuropathic pain in a glove-stocking distribution to a sensorimotor polyneuropathy, and can give a varying clinical picture consisting of symptoms like muscle and joint pain, cramp, weakness and fatigue.[1] Besides its strong association with the risk of falls, hypothyroidism is known to be a major cause of cerebellar and gait ataxias.[2,3] It is known that the frequency of neuropathy is between 10 and 70%, and that of myopathy between 20 and 80%, in cases of hypothyroidism.[4] Nonetheless, what is less recognised as a consequence of hypothyroidism, is the fact that, depending on the severity and duration of thyroid deficiency, various parts of the vestibular system, which play a key role in maintaining balance and postural stability, may be affected, resulting in vertigo, dizziness and other similar problems.[5]

Polyneuropathies and other neuromuscular symptoms found in patients with hypothyroidism include insufficiency of motor and sensory function secondary to profound loss of superficial, vibrational and proprioceptive sensation, and may cause disordering of static and dynamic postural stability and hence increase the risk of falls.[6,7] Despite the fact that a great many studies have been undertaken assessing postural stability and balance in patients with diabetes mellitus, one of the key causes of neuropathy, as far as the authors are aware, this kind of study has yet to be undertaken in patients with hypothyroidism.[6] Such a study, by clarifying postural stability, falls risk and associated risk factors in adults with hypothyroid illness would contribute to their recognition, treatment and management.

In our study, we aim to assess postural stability and the risk of falls in adult patients with known hypothyroidism by means of an objective computerised system.

Materials and Methods

This cross-sectional study was undertaken between November 2018 and April 2019 by the Physical Medicine and Rehabilitation (PMR), Neurology and Endocrinology divisions of the Atatürk University School of Medicine. The study protocol received ethical approval from the Medical Faculty of Atatürk University Ethics Committee (29.11.2018/7-1). Informed consent was obtained in writing from each participant. The study was conducted in accordance with the principles of the Declaration of Helsinki. The patients included in the study had a diagnosis of either subclinical or overt hypothyroidism according to endocrinological assessment, and were not confirmed to have any previous pathology in terms of postural stability or being at risk of falls. They received detailed clinical investigation by neurology specialists and neurological examination. Electromyography (EMG) did not indicate the existence of a polyneuropathy. The exclusion criteria for the study were: being unable to tolerate posturographic assessment or being unable to co-operate with it; having a previous history of orthopaedic or spinal surgery; suffering from a muscle disorder, movement disability, motor insufficiency, rheumatological disorders, degenerative osteoarthritis or diabetes mellitus; auditory or visual disorders; and any other type of pathology that could otherwise account for having a polyneuropathy. The control group consisted of individuals with normal thyroid function, and with no disorder that might cause balance problems.

To evaluate thyroid functions, free T4 (FT4; normal range:0.61-1.12 ng/dl) and thyroid stimulating hormone (TSH; normal range:0.34-5.6 mIU/ml) levels were measured by the chemiluminescent enzyme immunoassay method. A level of TSH>5.6 uIU/ml or FT4<0.6 ng/dl was considered overt hypothyroidism, TSH>5.6 uIU/ml with FT4 in the normal range was considered subclinical hypothyroidism.

The sex, age, body mass index (BMI), any history of falls in the last year, the score on the Falls Efficacy Scale - International version (FES-I), score on the neuropathic pain questionnaire (DN4) and clinical symptoms (weakness, tiredness, morning stiffness, exercise intolerance, muscle and joint pain, diffuse pain and paraesthesia etc.) was recorded for all participants in the study. None of the patients stated they were using any medication which may cause balance problems. In addition, 25 of the patients were receiving thyroid hormone replacement therapy at a dosage of between 50-150 µg.

The Falls Efficacy Scale - International version (FES-I) is a questionnaire based on self-reporting that allows quantification of the level of concern about falls whilst undertaking daily activities. The reliability and validity of the Turkish version of FES-I has previously been evaluated by Ulus et al.[8] The DN4 scale consists of ten questions, each worth one point. Seven questions concern symptoms,

whilst three are scored on physical examination. A total score of 4 or above is considered indicative of neuropathic pain. The Turkish language adaptation of the scale has been assessed for validity and reliability by Çevik et al.[9]

The Tetrax Interactive Balance System (Sunlight Medical Ltd, Tel Aviv, Israel) was used to assess both postural stability and to perform the falls risk analysis, in both cases and controls. The technique used was that outlined in the user guide for the device.[10] In the Tetrax system, posturographic evaluation consists of a detailed investigation that allows calculation of the balance and falls risk indices. There are two configurations. Changes made to the centre of gravity are assessed in the stability index (SI). Fourier transformations are applied to the four independent wave signals gathered from the device's four platforms to separate out 8 frequency bands. The weight distribution index (WDI) is calculated from the footplate data, and compares the way the weight is distributed between left and right feet and between the heels and toes. By using the four measurements described above, and comparing them in eight separate positions, the postural performance analysis can be carried out. These 8 different positions are as follows: eyes open and closed, with head held upright (NO, NC); eyes open and closed, head down, whilst standing on the foot pads (PO, PC); head turning to right and left whilst the eyes are closed (HR, HL); and eyes closed whilst extending and flexing the neck to the full extent (HB, HF). Using all these postural measurements, the various parameters were then calculated. The parameters output from the Tetrax device may be used to calculate a falls index, which reflects the patient's risk of having a fall. This index takes into account the various mechanisms which normally should prevent falling. The value of the index is between zero and 100, allowing risk stratification into low (score of 35 or below), moderate (score between 36 and 57) and high (between 58 and 100) risk.[11]

Statistical Analysis

The G* Power application was used to claculate the minimal sample size needed. This indicated that a minimum of 31 were needed as participants in the patient and control groups for the 95% confidence interval. A sample size of 50 for both the cases and controls was chosen in view of probable drop-outs from the study. Three volunteers from the control group left the study at the beginning because they were unwilling to provide blood samples. At the end of the study, 50 healthy volunteers and 47 cases completed the protocol.

The SPSS v. 23 statistical software package (SPSS Inc, Chicago IL, USA) was used for all statistical analyses. The Kolmogorov-Smirnov test was used to assess the normality of the data. The descriptive statistics generated were the mean±standard deviation for normally distrubuted data, the median (minimum-maximum) for non-normally distributed data and the number (as a percentage) for categorical data.

The independent samples t test and the Mann-Whitney U test were employed in group comparisons. The Chi-square test was used for 2x2 comparisons of variables of categorical type.

Results

In the cases group, 50 individuals participated, with a mean age of 41.7±11 years (range 19-61 years), of which 5 were males and 45 females. In the control group, matched for age and sex, 47 euthyroid individuals took part, with a mean age of 39±9.6 years (range 23-60 years), of which 11 were males and 36 females. See Table 1.

The demographic and clinical characteristics, postural stability parameters and values for the risk of falling for both the cases and healthy controls are given in Table 1. There was no significant difference between cases and controls, either in terms of age or gender ($p>0.05$), but the BMI of individuals in the cases group was higher at the level of statistical significance ($p<0.05$). The history of falls in the last 1 year was similar in both groups, with six hypothyroid individuals having a history of falling 14 times within the previous one year ($p>0.05$). FES-I scores were higher in the patient group, and this was a statistically significant difference between the groups ($p<0.05$). Mean FT4 and TSH levels in the patient group were 0.8±0.2 ng/dl and 9.3±12.2 mIU/ml, respectively. The control group's values for FT4 and TSH were 0.9±0.2 ng/dl and 1.8±1 mIU/ml, respectively. The FT4 level in the cases group was significantly lower, and the TSH significantly higher ($p<0.05$). Coming to the evaluation of the groups in terms of neuropathic pain and clinical symptoms, it was seen that the values obtained on the DN4 scale were significantly higher ($p<0.05$), as were all other symptoms except morning stiffness in the group with hypothyroidism ($p<0.05$). The overall SI and WDI scores obtained for postural stability in the groups are shown for the eight positions. There was a significant difference between the groups ($p<0.05$) in the other SI values obtained, apart from NO-SI, but for the WDI values obtained, only HL-WDI was significantly dif-

Table 1. Demografic and clinical characteristics, postural stability parameters and fall risk values in patients with hypothyroidism and healty controls

	Patients (n=50)					Controls (n=47)					
	n	%	Mean±SD	Median	Min-Max	n	%	Mean±SD	Median	Min-Max	p
Gender (M/F)	5/45					11/36					0.077
Age (year)			41.7±11					39±9.6			0.196
BMI (kg/m2			29.1±5					26.5±3.6			0.04*
Falls history		12					4				0.068
FES-I			25.4±8.8					20.3±6			0.001**
Serum FT4 level (ng/dl)			0.8±0.2					0.9±0.2			0.04*
Serum TSH level (mIU/ml)			9.3±12.2					1.8±1			0.001**
DN4			3,5±2,2					1.1±1.2			<0.001**
Clinical Symptoms (% frequency)											
Weakness		82					45				<0.001**
Fatigue		78					45				0.001**
Morning Stiffness		4					10				0.211
Intolerance of exercise		46					0				<0.001**
Muscle pain		78					2				<0.001**
Arthralgia		72					30				<0.001**
Widespread pain		60					25				<0.001**
Paresthesia		68					13				<0.001**
Postural stability parameters											
SI values											
NO-SI				15.9	9.3-29.5				14.9	10-78.7	0.262
NC-SI				23.3	14.2-50.1				19.4	13.7-32.1	<0.001**
PO-SI				18.1	11.4-53.9				16.6	10-38.1	0.022*
PC-SI				29.4	13.1-65.9				25.1	17.9-44.3	<0.001**
HR-SI				23	12.6-49.6				19.3	11.5-42.5	0.007**
HL-SI				25.5	12.1-56.6				21.3	14.5-48.1	<0.001**
HB-SI				27.4	12.6-74.6				20.9	14.8-39.3	<0.001**
HF-SI				28.1	16-56.8				19.9	13.6-36.1	<0.001**
WDI values											
NO-WDI				6.2	1.7-16				5.3	1.1-12.6	0.431
NC-WDI				6.4	1.2-15.5				4.9	1.9-11.5	0.073
PO-WDI				8.2	1.4-17.5				7	2.1-21.4	0.321
PC-WDI				6.8	0.8-14.2				6.6	1.4-17.1	0.718
HR-WDI				6	0.5-19				5.2	1.8-14.1	0.083
HL-WDI				6.6	2.2-17.4				4.9	1.8-13.6	0.01**
HB-WDI				6.2	1.4-17.8				6.1	1.2-13.5	0.127
HF-WDI				6	1.6-21.5				5.3	1.4-13	0.330
Fall risk assessment (0-100)				53	0-100				30	4-92	<0.001**
Fall risk category											
Low	14/50	28				31/47	66				
Moderate	16/50	32				11/47	23				
High	20/50	40				5/47	11				

SD: Standard deviation; Min: Minimum; Max: Maximum; M: Male, F: Female, BMI: Body mass index; FES-I: Falls Efficacy Scale International, SI: Stability Index, WDI: Weight Distribution Index, NO: open eyes- head neutral, NC: closed eyes- head neutral, PO: open eyes–sponge pad under the feet, PC: closed eyes–sponge pad under the feet, HR: closed eyes – head rotated to the right, HL: closed eyes – head rotated to the left, HB: closed eyes – head fully extended, HF: closed eyes – head fully flexed, *. p<0.05 was considered as statistically significant between patient and control groups, **. p<0.01 was considered as statistically significant between patient and control groups.

ferent ($p<0.05$). In the falls risk analysis performed by the computerised system, the fall risk category was found to be moderate (risk assessment: 53) in the cases group and was higher than the controls group at the level of statistical significance ($p<0.05$) (Table 1).

In the cases group, there were no significant correlations between sex, age, BMI, FES-I, DN4, FT4 or TSH level, clinical symptoms and the risk of falling ($r<0.3$; $p<0.05$). For the risk of falling, there was a weak positive correlation only amongst symptoms of paraesthesia and previous history of falls ($r>0.3$ with $p<0.05$ and $r=0.281$ with $p<0.05$, respectively). However, for postural stability and all values of SI, there was a significant positive association ($r>0.03$ with $p<0.05$). There was no demonstrable association between the values obtained for WDI and the risk of falling, except in the case of HL-WDI ($r<0.3$ with $p>0.05$). In addition, the risk of falling was discovered to be at a low level in the control group, and a significant positive correlation was found between the risk of falling and all SI values ($p<0.05$), (Table 2).

Discussion

By using the objective computerised technique, postural stability was shown to be disordered and the risk of falling increased in individuals with hypothyroidism. This increase in the risk of falling had an association with hypothyroidism, but not with the levels of FT4 or TSH, or the other factors under investigation.

Balance and postural stability is a complex physiological function, encompassing various neuromuscular processes. Sensory input, sensory evaluation and the neuromuscular response need to be controlled. Sensory input involves the vestibular, visual and proprioceptive systems. For an effective motor response, an intact neuromuscular system and sufficient muscular strength are needed.[12] Hypothyroidism causes slowing of metabolic pathways, reduction in ATP synthesis, the inability to break down glycosaminoglycans, malfunction of the Na-K pumps and thus damage to axonal transport. It therefore leads to the neuromuscular and neurological problems that can be observed in the disorder. Hence hypothyroidism may be a cause for impairment of postural stability and an increase in the risk of falls occurring.[13]

Factors that increase the risk of falling include advanced age (>65 years), being female, past falls and a fear of falling, living alone, having known medical problems, physical condition, impaired cognitive function, insufficient physical activity, and environment, amongst multiple others.[14] For this study, the age and sex distribution of the groups were similar, the cases were aged 41 years on average, and there were no additional medical conditions which might impair balance or increase the risk of falling. Thus no correlation was discovered amongst them with the risk of falling. Although the mean BMI values of both groups were close and in both cases indicated the subjects were overweight, there was a significant difference between them. However, the authors do not consider this situation to be clinically significant since it is not amongst the risk factors associated with falling. Although the history of falls was higher in the patient group, the result lacked statistical significance. FES-I values were, however, found to be significantly higher. Despite this, a relationship with the risk of falling was not established. This situation may be attributed to clinical symptoms insufficient for neuropathy, the absence of a polyneuropathy according to EMG results, and the fact that participants were not of advanced age.

Although the pathogenesis of polyneuropathy in hypothyroidism has not yet been completely elucidated, it has been linked to the accumulation of mucopolysaccharides in the endo- and perineurium, segmental demyelination, aggregation of glycogen, axonal degeneration and similar mechanisms of injury.[15-16] Neuropathy eventually results in superficial and deep sensory losses such as proprioceptive and vibration, and this sensory loss may cause the impairment of balance and postural stability, with a consequently raised risk of falling. As far as the authors are aware, there has been no study undertaken on postural stability and the risk of falling utilising an objective methodology in hypothyroid patients whose electrophysiological test results were normal and who lacked polyneuropathy. Diabetes mellitus is one of the leading causes of polyneuropathy and multiple studies concerning postural stability, balance problems and the risk of falls exist in the literature. In diabetes mellitus, disorders affecting the somatosensory system have been reported to produce postural problems, loss of balance and a subsequent increase in the risk of falls.[13,17-18] In addition, electrophysiological studies indicate that diabetic patients, even if they do not have a polyneuropathy, may lose their balance.[19] From this study, postural stability and the risk of falling can be seen to be affected in hypothyroid patients, even where polyneuropathy is not present.

Our research agrees with the literature in showing that, in hypothyroidism, the incidence of cramp rises [1,10-

Table 2. The correlation between demographic, clinical, stability index and weight distribution index values with fall risk.

	B12 patients with deficiency (fall risk)		Healthy controls (fall risk)	
	r	p	r	p
Age (year)	0.206	0.152	0.037	0.807
BMI (kg/m2)	0.198	0.168	0.321	0.028*
Falls history	0.281	0.048*	0.248	0.93
FES-I	0.170	0.238	0.243	0.099
Serum FT4 level (ng/dl)	-0.086	0.551	-0.348	0.017
Serum TSH level (mIU/ml)	-0.099	0.495	-0.156	0.295
DN4	0.155	0.281	-0.214	0.149
SI values				
NO-SI	0.411	0.003*	0.378	0,009*
NC-SI	0.566	<0.001**	0.720	<0.001**
PO-SI	0.403	0.004*	0.658	<0.001**
PC-SI	0.656	<0.001**	0.518	<0.001**
HR-SI	0.720	<0.001**	0.701	<0.001**
HL-SI	0.761	<0.001**	0.707	<0.001**
HB-SI	0.800	<0.001**	0.689	<0.001**
HF-SI	0.792	<0.001**	0.751	<0.001**
WDI values				
NO-WDI	0.139	0.334	0.174	0.242
NC-WDI	0.157	0.275	0.197	0.185
PO-WDI	-0.073	0.616	-0.054	0.717
PC-WDI	0.080	0.579	-0.065	0.663
HR-WDI	0.146	0.313	0.205	0.168
HL-WDI	0.291	0.041*	0.028	0.854
HB-WDI	0.187	0.193	0.014	0.928
HF-WDI	-0.076	0.601	0.016	0.913

SD: Standard deviation, BMI: Body mass index; FES-I: Falls Efficacy Scale International, SI: Stability Index, WDI: Weight Distribution Index, NO: open eyes- head neutral, NC: closed eyes- head neutral, PO: open eyes–sponge pad under the feet, PC: closed eyes–sponge pad under the feet, HR: closed eyes – head rotated to the right, HL: closed eyes – head rotated to the left, HB: closed eyes – head fully extended, HF: closed eyes – head fully flexed, *. Correlation is significant at the 0.05 level, **. Correlation is significant at the 0.01 level.

[12], complaints of fatigue are at a rate of 40-70% [1,4,13], and diffuse pain [4,11,14-15] and paraesthesia increase in frequency. [6,15] Even if, in theory, subclinical hypothyroidism is considered an asymptomatic disorder, our study agrees with the literature in revealing a clear increase in neuromuscular symptoms in such patients.

This study confirms the literature on hypothyroidism in showing a statistically significant rise in weakness, fatigue, exercise intolerance, muscle and joint aches, diffuse pain and symptoms of paraesthesia, in those with hypothyroidism, as compared to healthy controls. This was not shown for morning stiffness, however. However, with the exception of symptoms of paraesthesia, no correlation could be established between the risk of falling and these symptoms. There are studies available in the literature reporting that pain, weakness and fatigue have an adverse effect on balance [21-25]. The absence of a significant correlation in our study may arise from the fact that the cases group had no additional problems, were not advanced in age, possessed full muscular strength and may have exaggerated the symptoms to an extent, or for other similar reasons. It has been demonstrated in this study that complaints indicating paraesthesia,

such as burning, numbness and tingling may increase the risk of falls through impairment of postural stability.[26]

In our study, there was a significant difference in all the values of SI apart from NO-SI, but only the HL-WDI scores were significantly different amongst the WDI values. In the study, SI values were higher in the cases group. This indicates in a numerical form the changes in the parameters affecting balance, i.e. stance, muscle control and compensation that cannot be seen clinically. Higher values of the SI indicate a higher degree of imbalance in an individual, whereas a low SI indicates better balance and stability. Furthermore, the positive correlation found between all the parameters measured in the SI and the risk of falling indicate that balance plays a key role in determining an individual's risk of falling. In particular, where the PO-SI is more impaired than the NO-SI, which is a more straightforward position and reference point, this may show us that the patients cannot utilise their somatosensory systems (touch, proprioception and vibration sense etc) and are affected by the illness. In terms of the second of our parameters, the WDI, when there is an asymmetric distribution between the right and left feet, we can observe interactions between the heels and toe regions and between the two sides. High WDI is indicative of pathology, whilst values approaching zero indicate maximal postural stability, and are often produced by mechanisms responsible for attaining compensatory equilibrium. Although there was no significant difference between the groups, higher WDI values in the patient group could indicate that the compensatory mechanisms (e.g. somatosensory, visual and vestibular) in patients work less well than in the control group. Potentially, had the patients had greater symptoms or comprised more individuals, the difference between them might have reached statistical significance. The raised HL-WDI value (which was statistically significant) found in the patient group may indicate an undetected orthopaedic problem affecting the feet. There was also a subset of individuals within the control group, who fell into the low risk category for falling. In this group, there was a positive correlation between values on the stability index and the risk of falling. This correlation has no clinical significance, however, since there is no need to take precautions in a group with such a low risk of falls.

The elevated risk of falling observed in this study in patients with hypothyroidism may, we believe, arise from defects in the visual, vestibular or proprioceptive systems causing an impulse disorder. Due to the moderate risk of falls shown to exist in patients with hypothyroidism, patients should do regular exercise and be cautious when walking or running. Furthermore, when they are in contact with or walking on slippery surfaces, they need to take the necessary precautions to avoid falling. Advancing age, as well as various vitamin deficiencies, a rise in symptoms of neuropathy, the increase in degenerative disorder and, specifically in women, osteoporosis, can lead to fractures and their complications linked to falls. Our study may contribute to awareness of these problems and how to prevent them and thereby raise the quality of life.

One of the limitations of the study may be accepted as arising from the fact that some of the hypothyroid patients were already taking thyroid replacement therapy, and the group was not divided into two groups, subclinical or overt hypothyroidism, which may have permitted a better analysis.

Conclusion

In this study, it was found that postural stability was impaired and the risk of falling increased in hypothyroid patients. This increase in the risk of falling correlated with hypothyroidism but not with the level of FT4 or TSH, nor the other factors investigated. Accordingly, this increase in risk of falls may have occurred as a result of neuronal degeneration due to disease, even though the existence of a polyneuropathy is not reflected in electrophysiological tests. In addition, hypothyroidism should be considered as a possible cause of loss of balance, postural instability and increased risk of falls in patients without evidence of vestibular pathology.

Ethics Committee Approval: The study protocol received ethical approval from the Medical Faculty of Atatürk University Ethics Committee (29.11.2018/7-1).

Informed Consent: All patients provided written informed consent.

Author Contributions: Designing the study – A.K., A.B., K.S., H.U.; Collecting the data – A.K., A.B., N.B., K.S., H.U., R.D.; Analyzing the data – A.K; Writing the manuscript – A.K., N.B., F.B.; Confirming the accuracy of the data and the analyses – A.K., K.S., F.B.

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An Approach to Orbital Complications in Rhinosinusitis

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Abstract

Objective: Whilst orbital complications arising from acute rhinosinusitis are infrequent in contemporary practice, the spread of infection orbitally or intracranially can result in severe complications and a high mortality. This study aims to review cases of orbital complications arising from rhinosinusitis in the light of the published literature on the topic.

Methods: Eight cases of individuals presenting to a particular clinic with the development of orbital complications following sinusitis within the previous four years were included in the study. The characteristic features of these cases in terms of diagnosis, treatment and follow-up were investigated.

Results: The mean age of all cases was 29.5 years. In three cases, orbital abscess, and in five cases, preseptal cellulitis, were observed as complications. Both surgical and medical treatment was administered in three cases, while medical treatment alone was used for five cases.

Conclusion: The region where the complications of rhinosinusitis are most commonly observed is the orbit, due to its proximity to the paranasal sinuses, especially the ethmoid cells. Where limitation of eye movements, proptosis, and a decrease in vision exists alongside an orbital abscess which itself has developed as a complication of sinusitis, surgical treatment is the most important option to consider to prevent complications.

Keywords: Orbit, sinusitis, orbital cellulitis, abscess.

Introduction

Orbital infections can result from multiple different causes such as initial sinusitis, trauma, dental abscess, peribulbar operations, closed fractures, dacryocystitis, immunosuppression, and so forth.[1] Nowadays, although antibiotics are frequently used in the treatment of sinusitis, complications may still occur.[2] There are a number of complications that can occur if sinusitis leads to the spread of infection intracranially or intraorbitally. Orbital complications are rarely observed, but occur more frequently in children than adults. The orbital septum acts as a barrier preventing the spread of infection from the preseptal region to the orbital region. Infection in the sinuses can spread via lymphatic, venous, or perineural routes.[3] Since the ethmoid cells are in close proximity to the orbit, this is the region most commonly affected when complications do occur. Preseptal cellulitis, orbital cellulitis, subperiosteal abscess, orbital abscess, and cavernous sinus thrombosis are types of orbital complication found in sinusitis.[2,3] As infection takes hold, limitation of movement in the bulbus oculi, diplopia, chemosis, proptosis, a decrease in the pupillary reflex, a decrease in visual acuity or even permanent vision loss may be observed.[1-3]

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The objective of this study was to review those cases presenting to our clinic over the previous four years where orbital complications had developed due to rhinosinusitis, and to compare them with the current state of knowledge, as reflected in the literature on this topic.

Materials and Methods

The study was undertaken in accordance with the principles contained in the Declaration of Helsinki, and involved a retrospective file review. In this study, eight patients were reviewed retrospectively. These individuals had been hospitalised, diagnosed or had undergone treatment and/or follow-up in our clinic between January 2014 and November 2018 because of orbital complications from rhinosinusitis. Details of the history were extracted from the patients' case records. The cases had been evaluated by routine otorhinolaryngological examination and nasal endoscopy. In the endoscopic examination, Karl-Storz 4mm rigid 0°, 30°, and 70° Hopkins lens rigid endoscopes (Storz, St Louis, MI, USA) had been used. In the detailed eye examination performed by an ophthalmologist, the pathological appearance of the eyelids and surrounding tissues (particularly the presence of oedema or hyperaemia) were evaluated on the basis of eye movements, visual field examination, and functional integrity. Radiological diagnosis and follow-up of the patients was achieved by taking 4mm coronal and axial sections in the axial and coronal planes so as to include the paranasal sinus and orbital sections, by means of a Toshiba Aquilion 640-slice CT (Computed tomography) (Toshiba Medical Systems, Otawara, Japan, 2012). The patients' ear, nose, and throat disease examination results, demographic data, radiological imaging results, details of medical and surgical treatment, and follow-up results were analysed.

In all cases, intravenous ceftriaxone (adult dose 1-2 g/day, paediatric dose 50-75 mg/kg/day) + ornidazole 2x500 mg (adult dose 2x500 mg/day, 25 mg/kg in children, divided into 2 doses per day) + saline irrigation + local decongestant treatment were started as medical treatment. Surgical intervention was undertaken in cases featuring limitation of eye movements, a decrease in vision or no response to medical treatment within a 24 hour period.

Results

A total of 8 patients, five males (63%) and three females (37%), with a mean age of 29.5 (range: 11-70) years, were included in the study. The diagnosis in five cases was preseptal cellulitis (63%) (Figure 1). Three patients were diagnosed with an orbital abscess (37%) (Figure 2). The main complaints in the cases with orbital complications were fever, nasal congestion, postnasal drip, headache, swelling of the eyes, and decreased visual acuity. The frequency of the various symptoms and signs of orbital complications is presented in Table 1. The mean hospital stay was nine days. Endoscopic sinus surgery (ESC) + orbital decompression + external abscess drainage were performed in two patients, and ESC + orbital decompression were performed in one patient, while medical treatment alone was undertaken in five patients. After treatment, no functional or aesthetic problems were encountered in the patients. The clinical characteristics, complications, and details of treatment of the patients are presented in Table 2.

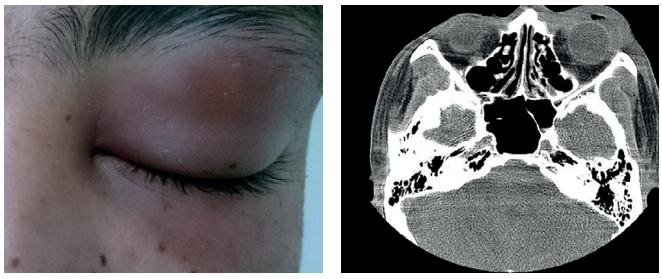

Figure 1. Left preseptal cellulitis.

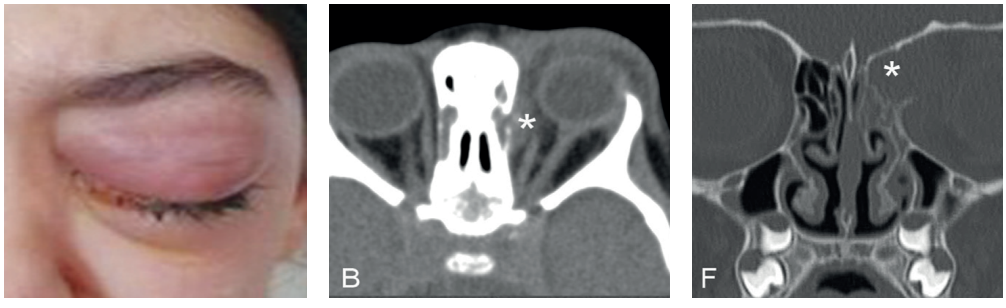

Figure 2. Left orbita abscess (white star)

Table 1. Symptoms and signs of orbital complications.

Sign	n	(%)
Nasal congestion/discharge	6	(75%)
Fever	4	(50%)
Headache	5	(62%)
Eyelid Edema	8	(100%)
Eye pain	5	(62%)
Blurred vision	2	(25%)
Loss of vision	2	(25%)

Discussion

The frequency of orbital complications observed in acute rhinosinusitis significantly decreased following endoscopic nasal examination, radiological imaging, and appropriate antibiotic therapy.[4] Complications are frequently observed in patients who do not receive adequate treatment for acute suppurative sinusitis or who suffer acute exacerbations in chronic sinusitis. These complications are grouped under three main headings: orbital, intracranial and osseous. The most common are orbital complications. *Haemophilus influenzae*, *Streptococcus pneumoniae* and *Staphylococcus aureus* are the principal pathogens in orbital complications. In acute rhinosinusitis infections, the route of spread to the surrounding tissues may be direct, venous, lymphatic or perineural. Direct regional spread occurs via osteitis in compact bones and osteomyelitis in diploic bones.[4,5] When the orbital tissues become infected, clinical signs appear over a broad spectrum, ranging from mild to severe. The most important factor determining the presentation of symptoms and signs in infection is which anatomical region of the orbit is involved, and the severity of the involvement. Orbital complications are associated with ethmoid, maxillary, frontal and, rarely, sphenoid sinusitis, with ethmoid sinusitis the most common and sphenoid sinusitis the least. Since these complications can result in serious morbidity and high mortality, it is critical to diagnose and treat them quickly.

Paranasal sinus tomography (PNS-CT) is a radiological imaging method used both to confirm the diagnosis of acute rhinosinusitis and to plan the surgical treatment of the complications of paranasal sinusitis or rhinosinusitis. PNS-CT has a high degree of effectiveness and is frequently a preferred modality in the evaluation of paranasal sinus infections and complications. PNS-CT offers benefit at each stage in the diagnosis and treatment of complica-

Table 2: Clinical features and treatment modalities of the patients.

Age-Gender	Complication	Eye signs	Treatment
14-M	Orbital abscess	Reduced vision + limited movement	ESC + orbital decompression + external drainage
70-F	Preseptal cellulitis	Normal	Medical
19-F	Preseptal cellulitis	Normal	Medical
11-M	Orbital abscess	Reduced vision + limited movement	ESC + orbital decompression + external drainage
66-M	Preseptal cellulitis	Normal	Medical
28-F	Preseptal cellulitis	Normal	Medical
15-M	Orbital abscess	Normal vision, limited movement	ESC + orbital decompression
13-M	Preseptal cellulitis	Normal	Medical

tions, including abscess formation. MRI imaging is the modality of choice for the evaluation of soft tissues in cases of cavernous sinus thrombosis and cerebral abscess. MRI may be preferable to CT for paediatric patients who will be monitored at close intervals and thus are at risk from cumulative radiation damage.[6]

Since the retina and optic nerve can only tolerate ischaemia for a very short period, typically 1-2 hours, vision and eye movements should be evaluated as soon as possible.[7] Patients with orbital complications require hospitalisation, and the antibiotic selected for treatment needs to be effective against streptococci, staphylococci, aerobes and anaerobes, which may be responsible for sinusitis. Good results have been reported in the literature with 3rd-generation cephalosporins, clindamycin and metronidazole in combination, and sulbactam-ampicillin, and researchers stress the need for intravenous antibiotic therapy to last for 7-14 days.[8,9]

In the literature, there are different approaches described to achieve surgical drainage in cases where a subperiosteal or orbital abscess develops. Garcia et al [10] monitored abscesses that were small, medial and not secondary to a tooth infection, in patients under nine years of age on medical therapy. They reported obtaining a response to medical treatment in 93% of cases. Yang et al [11] argued that, since the ostium is wider and the sinus receives superior ventilation at earlier ages, the proliferation of anaerobic flora is prevented, and thus spontaneous drainage is easier. Siedek et al [12] report draining surgically 90% of cases where abscesses occurred.

Patients in stages 1 and 2 of Chandler's classification [5] are often treated with a conservative approach, whilst patients in stages 4 and 5 are usually treated surgically. Treatment methods in stage 3 patients are controversial.[13] Teinzer et al [14] stated that in stage 3, 4 and 5 patients, urgent surgery should be performed within 24 hours. There are currently some authors who suggest that medical treatment can still be used in patients with a subperiosteal abscess in whom vision and intraocular pressure are normal, proptosis is smaller than 5 mm, eye movement is not limited, and abscess width is smaller than 4 mm.[14,15] In another evaluation, it was reported that the best treatment modality was surgery in cases where vision is impaired and eye movements limited, proptosis is greater than or equal to 5mm, intraocular pressure is greater than 20 mmHg, and the size of the abscess exceeds 1 cm.[16] While in the case of a medially located subperiosteal abscess, the endoscopic approach is widely used in surgical treatment, external drainage is needed if the abscess is superiorly located.[17,18] Currently, some authors suggest the combined approach (external drainage + endoscopic approach) in large-sized abscesses with medial, superior or medial/superior co-location.[19] Additionally, some authors have stressed the necessity of surgical drainage in subperiosteal orbital abscesses greater than 500 mm^3 in volume or 1cm in diameter.[20]

In this study, patients with acute rhinosinusitis, who were suspected, on the basis of a suitable history, physical examination and detailed endoscopy, of having orbital complications, were hospitalised, and intravenous antibiotherapy (ceftriaxone+ornidazole) + local decongestant + saline irrigation therapy were initiated. A specialist ophthalmic consultation was sought in order to evaluate vision in all patients, and PNS-CT was requested. Surgical treatment was undertaken within 24 hours in cases where an orbital abscess was accompanied by proptosis, ophthalmoplegia, and loss of vision. Consistently with the recommendations in the literature, the combined surgical approach was applied in 2 patients with a superiorly located abscess, and ESC + drainage were performed in 1 patient with a medially located abscess. Kim et al [21] have reported that navigation-aided endoscopic surgery in a superiorly located orbital abscess is safe. Roithmann et al [22] report the successful application of endoscopic drainage to a patient with a superiorly located abscess. Another author states that, in the presence of large collections of pus, medical treatment is usually insufficient as sole therapy, and should be combined with surgical treatment in older children and in cases featuring visual impairment.[23] While the conservative approach is strongly recommended in the literature for abscesses which are medially located and smaller than 0.5cm^3, Gavriel et al [23] argue that the conservative treatment approach is also safe for superiorly located abscesses smaller than 0.5 cm^3.

Conclusion

In patients diagnosed with sinusitis with swelling and redness of the eyelids, a detailed ophthalmologic examination is essential, including globe movements and visual acuity. In cases where there is limitation in eye movements and a decrease in visual acuity, surgical drainage should be performed as an emergency. Conservative treatment can be applied in small-sized subperiosteal abscesses when vision is normal and there is no preseptal cellulitis, orbital cellulitis, or limitation of eye movements.

Ethics Committee Approval: The local ethics committee approved the study with the project number of E-771.

Informed Consent: Informed consent was not received due to the retrospective nature of the study.

Author Contributions: Designing the study – S.Ç.; Collecting the data – S.K.; Analyzing the data – S.Ç., S.K.; Writing the manuscript – S.Ç.; Confirming the accuracy of the data and the analyses – S.Ç., S.K.

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

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An Analysis of Sociodemographic and Clinical Characteristics in Children and Adolescents Diagnosed with Childhood Onset Speech Fluency Disorder

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Abstract

Objective: Childhood onset speech fluency disorder (stuttering) is a communication disorder beginning in childhood, and is characterised by interruption to speech flow, sound prolongations and pauses produced by repetition of a particular sound or word. The aim of this study was to investigate the sociodemographic characteristics, comorbid psychiatric diagnoses and other variables that are associated with stuttering.

Methods: Sixty-four children and adolescents aged under 18 years and attending the child and adolescent psychiatry or otorhinolaryngology (ENT) outpatient clinic between November 2017 and June 2019 were enrolled in the study. ENT examination was carried out. A sociodemographic questionnaire was also administered to the participants. The psychiatric evaluation of the patients was performed according to DSM-5 criteria.

Results: The mean age was 7.36±3.76 years. 70.3% of the cases were male. The mean age at onset of stuttering was 4.85±2.34 years and the average stuttering duration was 2.31 years. In 31.3% of cases a life stressor had preceded stuttering, 21.9% had a positive family history, and 40.6% had comorbid psychiatric morbidity. The most common comorbid psychiatric diagnoses were specific learning disorder, speech sound disorder and attention deficit hyperactivity disorder.

Conclusion: According to the results obtained, stuttering was 2.3 times more common in males, and 68.7% of all cases were below the age of 5 years. The most common psychiatric diagnoses found in cases of stuttering are within the neurodevelopmental disorders grouping. Since psychiatric diagnoses occur at such a high frequency, it is important to address this comorbidity when treating stuttering.

Keywords: Child, language, learning disorder, speech, stuttering.

Childhood onset speech fluency disorder (stuttering) is a communication disorder beginning in childhood, and characterised by interruptions to speech flow, sound prolongations and pauses produced by repetition of a particular sound or word. Overall, 95% of stuttering cases begin before the age of 5 years.[1] Although it is estimated that the lifetime prevalence of stuttering is only around 0.3-1%, it has been ascertained that 5 out of 100 people do actually have a stuttering problem at some period in their lives.[2,3] When undiagnosed cases, or those which have already re-

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mitted spontaneously are taken into account, it is thought that the true rate will be higher than that previously reported.[4] Some investigators have noted that the frequency of stuttering in children varies according to race or ethnicity.[5] In addition, it is more frequent to observe stuttering in children with genetic syndromes, such as Down Syndrome or Fragile X.[6] Stuttering can appear in different guises: it can be in the form of repeating a particular sound or word, prolonging a sound, interjecting, pausing within words, filled or unfilled pauses in speech, circumlocutions, excessive word stress and repetition of words consisting of a single syllable.[7] These clinical findings and symptoms are listed amongst the diagnostic criteria for "childhood onset speech fluency disorder", within the category of neuro-developmental disorders in the Diagnostic and Statistical Manual of Mental Disorders-5 (DSM 5).[7] The intensity and frequency of stuttering may vary in different contexts, or depending on whom a child is talking to. Additionally, secondary behaviours may also be observed, such as clearing the throat, repeating the "um" sound, blinking the eyes, or shaking the head when initiating speech. These behaviours may also be linked to the avoidance of speaking. Stuttering may be classified into developmental, neurogenic, and psychogenic types.[8] Although DSM 5 does not include such a system of classification, it does acknowledge that neurological factors should be ruled out as a cause when making a diagnosis of stuttering, and stuttering should be coded "adult-onset" when it starts in the post-adolescence period.[7] Childhood stuttering is a multi-factorial diagnosis, encompassing genetic, psychological, neurological and behavioural characteristics, and each of the factors causing stuttering should be focused on in turn.[9-12] Stuttering in adults is usually related to an underlying neurogenic or psychogenic condition.[13] In studies which have investigated the family history of stuttering children, the rate of a positive family history ranges between 20 and 74%.[14] Although the high frequency of a positive family history in sufferers suggests a genetic basis, no chromosomal anomaly or model for how the disorder may be inherited has yet been identified.[14] Amongst some of the neurogenic factors implicated in the aetiology of the disorder, differences in the relative volumes of grey and white matter in the brain, differences in neuronal pathways, atypical lateralisation of functions to the opposite hemisphere and increased white matter connections within the right hemisphere have been described.[15,16] In the psychiatric evaluation of children who stutter, behavioural problems, various fears or other psychosocial stressors preceding the onset of stuttering have been observed.[17] In addition, it has been reported that a lack of self-confidence, introversion and obsessional character traits are encountered more frequently in subjects who stutter. Shyness, being ridiculed, inhibitions and worries can adversely impact the lives of children who stutter.[18]

In this study, the aim was to evaluate the sociodemographic characteristics and comorbid psychiatric diagnoses of children and adolescents diagnosed with stuttering.

Materials and Methods

The study was granted ethical approval by the Human Research Ethics Committee of Mugla Sıtkı Koçman University. The design was a retrospective chart review study. The participants in the study consisted of 64 children and adolescents under the age of 18 diagnosed with stuttering at the Child and Adolescent Psychiatry and Otorhinolaryngology outpatient clinics in Mugla Sıtkı Koçman University Training and Research Hospital. All the cases underwent examination by a child psychiatrist. The age of onset of stuttering, its duration, the existence of triggering factors, any family history, developmental characteristics, comorbid psychiatric diagnoses and whether the subjects received speech therapy or special training were investigated, in addition to their sociodemographic characteristics. The diagnosis of stuttering was made in accordance with DSM-5 diagnostic criteria, following detailed clinical examination. Cognitive function and level of intelligence were assessed by means of age-appropriate tests (i.e. the Ankara Development Test Inventory, Denver Developmental Screening Test [Denver II], Wechsler Intelligence Scale for Children, [WISC-IV]) alongside psychiatric examination. All of the subjects underwent otorhinolaryngological examination. Depending on examination findings, the patient's history and family history, patients at risk of hearing loss also underwent audiometric examination. Children under 5 years of age (n=2) underwent play audiometry and children over 5 years (n=11) underwent pure tone audiometry.

The data obtained were evaluated using the SPSS 17.0 (SPSS Inc. Chicago, Illinois, USA) software application running on Windows. Descriptive data were obtained as minimum, maximum, mean, standard deviation, number and percentage. In the evaluation of normal distribution, the Kolmogorov-Smirnov test, and in the comparison of independent groups, the Student t-test, were used. A p value <0.05 was accepted as indicating statistical significance.

Results

Of the 64 subjects included in the study, 19 were female and 45 male. The mean age was 7.36±3.76 years. The mean age of their mothers was 34.39±6.12 years and the mean age of the fathers was 40.05±7.51 years. Prenatal complications had occurred in 17.7% of cases, perinatal complications in 3.2% and postnatal complications in 25% (i.e. history of incubator use or phototherapy). The mean number of siblings was 1.98±1.49. 60.9% of the subjects were the first child, and 23.4% the second child, of their families. The mean time interval elapsed before starting walking was 15.06±6.72 months and the mean age of utterance of the first meaningful words was 17.48±9.26 months. The sociodemographic characteristics of the subjects are given in Table 1.

The mean age at which stuttering began was 4.85±2.34 (min 2, max 16) years. The mean duration of stuttering was 2.31 years. A statistically significant difference was not found between the sexes in terms of the age at which stuttering began (p=0.12, independent groups t-test). In 31.3% of the subjects, a life stressor was identified which led to stuttering (Table 1).

There was a comorbid psychiatric diagnosis in 40.6% of cases. The most common comorbid psychiatric diagnoses were specific learning disorder (SLD), speech sound disorder and attention deficit hyperactivity disorder (ADHD) (Table 2). Eleven out of the 35 children who were at school age had SLD (31.4%). 11 subjects were taking psychiatric medication. The most frequently used medications were psychostimulants and selective serotonin reuptake inhibitors. The age of onset of stuttering for subjects with a psychiatric diagnosis was 6.09±2.94 years, whereas it was 4.13±1.55 years for those with no psychiatric diagnosis. A statistically significant difference was determined between the two groups (p=0.001, independent groups t- test).

Discussion

The mean age when stuttering began was determined as 4.85±2.34 years in this study. It was detected that stuttering began before 5 years of age in 68.7% of cases, at school age in 14 cases, and during adolescence in 2 cases. Developmental stuttering usually starts between the ages of 2-5, which is the period during which the most rapid development in speech occurs, and spontaneous remission occurs within 2 years in 75% of cases.[19] Similarly to the results of our study, Morley [20] states that the point at which stuttering begins in 85% of cases is prior to age 8.

Stuttering is seen twice as often in males compared to females.[21] Similarly to some other studies found in the literature, 70.3% of cases in our study were male, and the proportion of males to females was 2.3. However, it has been stated that in cases of stuttering occurring at an early age, the male-female ratio is similar, but as age increases, this ratio increases towards a male predominance.[4] In our study, it was observed that the male/female ratio (n=5/6) is close to unity in cases under the age of 4. Different theories have been put forward to explain the sex ratios observed. The greater effect on males than females of life stressors, the earlier maturation of females, the possibility of hereditary transmission and the varying profiles of androgenic hormones have been cited in such theories. In a study by Selçuk et al [22], it was determined that the testosterone level in males who stutter was higher than in a control group. The authors therefore suggest there might be a relationship between stuttering and testosterone levels.

The effect of birth complications on stuttering has also been investigated. According to the results of one review, children who suffer cerebral injury either at birth or at other times stutter more frequently than control subjects of the same age.[23] In a large sample study, the risk factors for stuttering were ascertained as the following: perinatal complications, incubator occupancy (due to prematurity), likely attention deficit hyperactive disorder, parental alcohol misuse, a family history of obsessive compulsive disorder, maternal disability and having a foreign-born parent.[24] In our study, only 3 cases of premature birth were found. In addition, a history of incubator occupancy and phototherapy were present in 25% of cases.

In a study comparing cases of stuttering with healthy subjects, 30-60% of the cases with stuttering had a positive family history, whereas this occurred in less than 10% of the healthy control group.[4] In our study we found a family history of stuttering in 21.9% of cases. Avcı et al [17] likewise determined that 18.6% of cases had first degree relatives with a history of stuttering. The high rate of positive family history in stuttering cases has meant aetiological studies have generally focused on genetic factors, but a definite model of heredity has not yet been proposed.

Acquired stuttering almost always emerges due to neurological or psychological factors.[25] In our study, in virtually all of the school age or adolescent cases of stuttering where there were no pre-existing problems with the fluency of speech, there was an identifiable triggering event. More specifically, starting primary school was the most frequently observed triggering factor for stuttering to occur in the school age period. Avcı et al [17] reported that 33.5% of the children diagnosed with stuttering had suffered a life

Table 1. Sociodemographic characteristics of the subjects diagnosed with stuttering and variables related to stuttering

Variable	n	%
Gender		
Female	19	29.7
Male	45	70.3
Educational status		
Not school age	18	28.1
Pre-school	11	17.2
Primary school	24	37.5
Middle-school	3	4.7
High-school	8	12.5
Marital status of parents		
Married	55	86
Divorced/Single parent	9	14
Mother's occupation		
Employed	22	34.4
Unemployed*	42	65.6
Father's occupation		
Employed	62	96.9
Unemployed*	2	3.1
Prenatal complication**		
Yes	11	17.7
No	51	82.3
Natal complication**		
Yes	2	3.2
No	60	96.8
Type of birth		
Vaginal birth	21	32.8
C-Section	43	67.2
Time of birth		
Term	60	93.7
Premature	3	4.7
Postmature	1	1.6
Incubator history		
Yes	8	12.5
No	56	87.5
Neonatal jaundice		
Yes	12	18.8
No	52	81.2
Phototherapy history		
Yes	8	12.5
No	56	87.5
Convulsion history		
Yes	6	9.4
No	58	90.6
Surgery history		
Yes	9	14
No	55	86
Chronic disease		
Yes	8	12.5
No	56	87.5
Accompanying body movement		
Yes	7	10.9
No	57	89.1
Special education		
Received	20	31.2
Not received	44	68.8
Speech therapy		
Received	14	21.9
Not received	50	78.1
Family history of stuttering		
Yes	14	21.9
No	50	78.1
Life stressor related to the stuttering***		
Yes	20	31.3
No	44	68.7
Intelligence level		
Normal	52	81.3
Border	8	12.5
Mild	4	6.2
Use of psychotropic drug		
Yes	11	17.2
No	53	82.8

* The unemployed category includes housewives, unemployed or retired people.
** Missing values: prenatal (2) and natal (2) complications. Percentages calculated among cases with non-missing values
*** Starting school, death of the mother, moving, birth of a sibling, being chased by an animal, being afraid of his/her own shadow, falling, skin prick test, starting to sleep separately, father's beginning his military service, amusement parks, horror movies, change of teachers

stressor at no more than one week prior to stuttering and the prognosis of stuttering which emerged after a stressor factor was positive. Similarly, we found that the rate of occurrence of a life stressor was 31.3%. Life stressors can be listed as starting school, various fears, death of the mother, birth of a sibling and moving. Since our study was not a follow-up study, it was not possible to compare and evaluate the rate of spontaneous remission in these cases. It is generally accepted that stuttering cannot be explained by any single-factor theory, and the pathogenesis is thus multifactorial.[9]

In 67.1% of cases, stuttering had been present for more than 6 months. The following are risk factors for persistent stuttering: male, family history of stuttering, stuttering beginning at 3.5 years of age or older, having a comorbid language or speech disorder, stuttering severely at the age

Table 2. Comorbid psychiatric diagnosis.

	n	%
Specific learning disorder	12	18.7
Attention deficit hyperactivity disorder	6	9.3
Speech sound disorder	5	7.8
Post-traumatic stress disorder	2	3.1
Generalized anxiety disorder	2	3.1
Social anxiety disorder	2	3.1
Enuresis nocturna	1	1.5
Autism spectrum disorder	1	1.5
No diagnosis	38	59.4

of 4 to 5 years and early delays in the acquisition of the fundamental motor processes involved in speech.[1,4,26,27] In our study, 74.4% of the cases whose stuttering lasted more than 6 months were male, 23.3% had a family history of stuttering, and 7% had an accompanying speech sound disorder. The age at the onset of stuttering in 76.7% of cases was 3.5 years or older. According to the normal developmental stages for speech, it is expected that a child will utter her first word at 12 months, will have 1-50 words at 18 months and be capable of 2-word sentences at the age of 2.[28,29] It was noted that the mean age of saying a meaningful first word for all participants diagnosed with stuttering was 17.48 months and they therefore had speech delay. Speech delay is a risk factor for the development of language-speech disorders in general.[30]

Blood et al [31] report that articulation disorder is comorbid with stuttering in 33.5% of cases, phonological disorder in 12.7%, learning difficulty in 11.4%, and ADHD in 5.9%. Arndt and Healey [32] determined that there was a phonological and/or language disorder in 44% of children who stutter. Avcı et al [17] stated that in 28% of children who stuttered, a comorbid psychiatric diagnosis existed, the most frequently seen disorder being enuresis nocturna. In our study, 40.6% of cases had a comorbid psychiatric diagnosis. The most frequently encountered diagnoses were SLD, speech sound disorder and ADHD. It is noteworthy that the age at which stuttering began was higher in cases where a psychiatric diagnosis was also appropriate than in those where no such diagnosis applied. The rate of SLD was 18.7% overall, whilst in school age cases, this rate increased to 31.4%. Comparing these rates with the overall prevalence of SLD in the general population, 5-15%, it is evident that SLD is considerably more prevalent in individuals who stutter.[7] In one study evaluating school age subjects with stuttering, it was ascertained that the scores for attention deficit and impulsivity were raised compared to normal controls.[33] Some of the comorbidity with disorders in the neurodevelopmental group seen in stuttering subjects can be explained by shared aetiological or risk factors. Stuttering is, in any case, a neurodevelopmental disorder in its own right. In addition, it was observed that psychotropic drugs had been prescribed for comorbid psychiatric diagnoses.

This study has certain limitations, amongst which may be mentioned the fact that the intensity of stuttering was not determined using the measurement tools available. Not being able to comment on the duration of recovery and stuttering of the study participants is also a limitation, and was due to the study design not allowing for follow-up.

Conclusion

In conclusion, psychiatric diagnoses frequently accompany a diagnosis of stuttering. Children admitted to otorhinolaryngology and paediatric units with speech disorders should be evaluated by a child and adolescent psychiatrist to allow for recognition of any accompanying psychiatric disorder, and thus to allow the treatment process to be conducted more efficiently. Since SLD is frequently comorbid with childhood onset speech fluency disorder, particularly in school age children, assessment for SLD in these patients to facilitate improvement in academic performance is crucial. There is also a need to undertake preventive measures to improve the mental health of patients with stuttering.

Ethics Committee Approval: The study was received ethical approval by the Human Research Ethics Committee of Mugla Sıtkı Koçman University.

Informed Consent: Informed consent was not received due to the retrospective nature of the study

Author Contributions: Designing the study – B.G.Ö., E.Ö.; Collecting the data – B.G.Ö., E.Ö.; Analyzing the data – B.G.Ö., E.Ö.; Writing the manuscript – B.G.Ö., E.Ö.; Confirming the accuracy of the data and the analyses – B.G.Ö.

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Benefits on language development and auditory perception performance of using a contralateral hearing aid in cochlear implanted children

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Abstract

Objective: To evaluate the benefits of a contralateral hearing aid on expressive and receptive language development and auditory perception performance over a 36-month post-surgical period in children implanted on a single side with a cochlear device for bilateral pre-lingual profound sensorineural hearing loss.

Methods: Seventy-five patients with idiopathic profound sensorineural hearing loss were included. The cases were divided into two groups: cochlear implant users (50 patients, "CI group") and cochlear implant plus hearing aid users (25 patients, "CI+HA group"). Language development and auditory performance were compared in the two groups during the first 3 years following cochlear implant surgery. The Pre-school Language Scale-4 was used to assess language development and the LittlEars® Auditory Survey, Meaningful Auditory Integration Scale and Meaningful Use of Speech Scale were employed to assess auditory perception performance.

Results: Language development in the CI+HA group was superior to that in the CI group at 6 months post-surgery, in terms of receptive and expressive language development; auditory perception performance was also superior in the CI+HA group, compared to the CI group.

Conclusion: The use of a contralateral hearing aid in cochlear implanted children with prelingual sensorineural hearing loss positively contributes to language devel-opment and auditory perception performance.

Keywords: Auditory perception, cochlear implants, hearing aids, language development.

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Hearing loss is one of the most common disabilities encountered in children. If not corrected, its negative effects may last a lifetime. Sensorineural hearing loss occurs in approximately 1-2 infants in every 1,000 births.[1,2] Hearing loss may be congenital or acquired after birth. Whatever the cause, hearing loss in the pre-lingual period should be diagnosed as soon as possible, and hearing amplification provided.

The first years of life represent the most critical period for speech and language development. If hearing loss in a child goes unrecognised during this period, speech and language development at the desired level cannot be achieved. Children who do not have auditory perception skills have an inadequate ability to recognise, distinguish, and understand voices. Unrecognised hearing loss during infancy and early childhood adversely affects the development of the central nervous system, and social, emotional, cognitive and academic development lags behind compared to the child's healthy peers.[3] Cochlear implants facilitate a significant increase in hearing and speech skills in patients with severe or profound pre-lingual hearing loss when compared to the period preceding implantation of the device.[4] However, unilaterally implanted patients have difficulty locating sounds, such that all sounds appear to be coming either directly from their ear, or to be inside their head. Additionally, their ability to understand speech in every day, noisy or reverberant environments is relatively poor.[5-7]

Currently, bilateral cochlear implantation is a common procedure. However, in some patients, cochlear devices cannot be implanted bilaterally due to medical complications, insurance restrictions or personal preference. Combining electrical hearing (i.e. a cochlear device) with an acoustic hearing aid in the other ear may be an option for such patients. This combination of electrical and acoustic stimulation is termed "bimodal stimulation". Binaural hearing via bimodal stimulation can improve speech perception in both quiet and noisy environments, and also improve the ability to localise sounds, determine sound quality, and perceive melodies and other aspects of music.[8,9] Looking at the benefits obtainable, one would be tempted to suppose the presence of residual hearing in the opposite ear were essential. Surprisingly, though, it appears that the benefits of bimodal stimulation are obtainable even where profound hearing loss is present.[10]

In our study, we evaluated the effectiveness of providing bimodal hearing (by supplying a contralateral hearing aid alongside a unilateral cochlear implant) on the development of receptive and expressive language, as well as auditory perception performance, in patients with bilateral pre-lingual profound sensorineural hearing loss.

Materials and Methods

A retrospective case-control study was undertaken at Gazi University Hospital. The study was conducted in accordance with the principles of the Helsinki Declaration. Ethical permission number 469 was granted on 13/10/2014 by the Corporate Ethics Committee of the Gazi University Hospital.

A retrospective review was undertaken of patients from the clinic who had undergone cochlear implantation between 2002 and 2014 for profound pre-lingual sensorineural hearing loss. A total of seventy-five patients (50 single CI users, 25 CI and contralateral HA users) were included in the study. Table 1 presents the demographic data for the study participants. The inclusion criteria were pure-tone averages (PTAs) for hearing thresholds at 0.5, 1, 2 and 4 kHz worse than 90 dB hearing loss in both ears, diagnosed in the pre-lingual period. The exclusion criteria were: patients who did not regularly use the cochlear implant or hearing aids, who suffered from hearing loss secondary to meningitis or a specific syndrome, had mental retardation, had not attended the rehabilitation programme regularly or had not attended audiological check-ups on a regular basis. The patients enrolled in the CI+HA group continued to use the hearing aids that they were using before the implant surgery.

In the present study, for the evaluation of language development, a ratio was calculated in which the average score obtained for each language term was compared to the expected score at the patient's chronological age, rather than recording the change over the period in which the patient was assessed. If the value of the ratio obtained is equal to or greater than 1, the patient's language age is consistent with the chronological age; if it is less than 1, the language age is lower than the chronological age. When the level of language development between groups was compared in our study, this measure was used to compare outcomes.

For each of the participants selected from the audiology service follow-up documentation, the preoperative and postoperative 36-month language development test results and auditory perception test results were entered into a data format suitable for use with Microsoft Excel (Excel Version 12.0, Microsoft, Redmond, WA, USA). Other data,

Table 1. Demographic data of the patients

	CI	CI+HA	p
Male	25 (50%)	9 (36%)	
Female	25 (50%)	16 (64%)	
Diagnosis age* (month)	9.1±4.8	10.7±4.7	p>0.05
Operation age* (month)	19.4±6.6	20±5.9	p>0.05
Duration of hearing aid before surgery* (month)	5.8±4.6	6±4.3	p>0.05
PTAs (.5, 1, 2, 4 khz) implanted side* (before surgery) (dB)	104.9±9.6	104.2±9.8	p>0.05
PTAs (.5, 1, 2, 4 khz) non-operated side* (dB)	103.5±9.6	100.2±9.3	p>0.05

CI: Cochlear implant group, CI+HA: Cochlear implant+hearing aid group, PTAs: pure tone averages, dB: decibel, khz: kilohertz, *: mean±SD

including the patients' sex, age of diagnosis, age of operation and pre-operative device usage times were also recorded. We used the Turkish-validated version of the Preschool Language Scale-4 for the assessment of language development. To determine the performance of auditory perception, we used the LittlEARS® Auditory Questionnaire, the Meaningful Auditory Integration Scale (MAIS) and the Meaningful Use of Speech Scale (MUSS).

Tests Used

Pre-school Language Scale-4 (PLS-4)

The PLS-4 test, which plays an important role in studies on language development, is a psychometric test to evaluate both the receptive and expressive language skills of children aged up to 6 years and 11 months of age. There are 62 test items for receptive language, 68 test items for expressive language and 104 pages in the illustrated test books. Testing begins at a level at least one year below the chronological age of the patient. For each question, the child's answer format is marked with 'D', 'S', or 'M' (Directly, Spontaneously, Mother or nanny). Each question has a passing criterion stated under the question. A child who meets the passing criterion is awarded 1 point and considered successful for that question. Any child who passes more than half of the questions in each language level then progresses on to the higher level questions. As a result of the test, the development period which the child has reached in terms of receptive or expressive language is determined (9-11 months, 18-23 months, etc).[11]

LittlEARS® Auditory Questionnaire

The LittlEARS® Auditory Questionnaire is a questionnaire evaluation that shows the auditory development, speech development and pre-verbal speech phase in normal or hearing-impaired children. It evaluates auditory development up to two years of age in hearing children, or in the first two years after a cochlear implant or hearing aid is supplied. It consists of 35 questions marked 'Yes' or 'No'. The 'Yes' option scores 1 point and the 'no' option 0 points. The test has a maximum total score of 35 points.[12]

Meaningful Auditory Integration Scale (MAIS)

This test is a questionnaire consisting of 10 items that can be applied to children at all ages, both before and after implantation. The items evaluate the child's ability to listen with the hearing aid or implant, recognise sounds and combine them with their meaning. The survey is divided into three sections. The first two questions include the initiation of listening; questions three, four, five and six include recognition of voices; the seventh, eighth, ninth and tenth questions concern the ability to ascribe meaning to voices. There are five different response options for each question (0=never, 1=rarely, 2=sometimes, 3=frequently, 4=always), giving a minimum item score of zero points and a maximum of four points. The test is evaluated over 40 points in total.[13]

The Meaningful Use of Speech Scale (MUSS)

MUSS is used to assess the child's communication strategies, the ability to produce the sounds used in the mother

tongue, and the ability to control those sounds. The test may be used with children of all ages. The questionnaire may be filled in by parents or teachers. The first three items on the questionnaire assess the patient's voice control, the next five questions examine the speaking voice, and the last two questions evaluate speaking strategies. There are five different response options for each question (0=never, 1=rarely, 2=sometimes, 3=frequently, 4=always), giving a minimum item score of zero points and a maximum of four points. The test is evaluated over 40 points in total.[13]

Statistical Analysis

All data were analysed by means of the Statistical Package for Social Sciences application (SPSS for Windows version 17.0; SPSS Inc., Chicago, IL, USA). The parametric Student's t-test was used to compare language development levels between the groups. Comparisons of within-group language development level over time were made using the paired sample Student's t-test. The non-parametric Mann-Whitney U test was used to compare auditory perception level development between groups, at different monthly intervals. The level set as indicating statistical significance was $p<0.05$, for all tests.

Results

There were no statistically significant differences between the CI and CI+HA groups ($p>0.05$) in mean age at diagnosis, mean time of use of the device before surgery, the age at which surgery was performed, or PTAs before surgery of both implanted ears and non-operated ears.

There was no significant difference in receptive or expressive language development between groups until the 6th month. However, after the sixth month, both the receptive and expressive language development of the CI+HA group was significantly greater ($p<0.05$). Table 2 and Figure 1 show the mean language development rates of each group. At the end of thirty-six months, the language development of patients in neither group matched chronological age, but the most significant approximation to this goal was in the CI+HA group (Figure 2).

With reference to the results of the MAIS test, there was no difference between the groups up to the 6th month, while the mean test results of the CI+HA were significantly greater for all months from 6 months to 36 months (36th month, $p=0.01$). Similarly, the mean MUSS test scores in the CI+HA group were significantly higher after six months, and this significant elevation continued until the 36th month (36th month $p<0.01$). When the LittlEARS® test results were compared, it was observed that the CI+HA group received higher scores from the postoperative period up to 12 months, but this result failed to reach the level of statistical significance. The mean scores in the CI+HA group were significantly higher from the twelfth to the thirty-six months inclusive. At the end of the thirty-six months, the mean test results of the CI+HA group were also significantly higher ($p=0.01$). Table 3 and Figure 3 indicate the mean LittlEARS®, MAIS and MUSS test results for the groups.

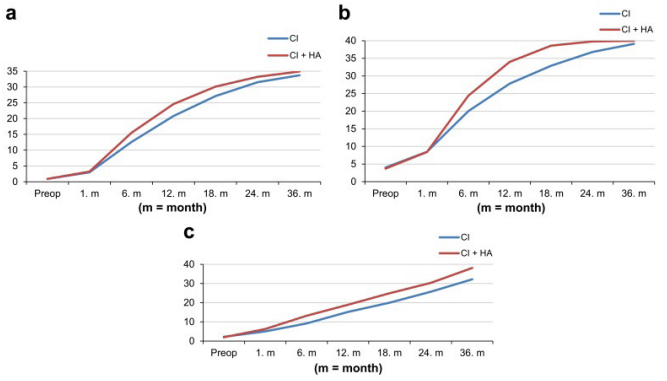

Figure 1. Expressive (a) and Receptive (b) language development of the groups

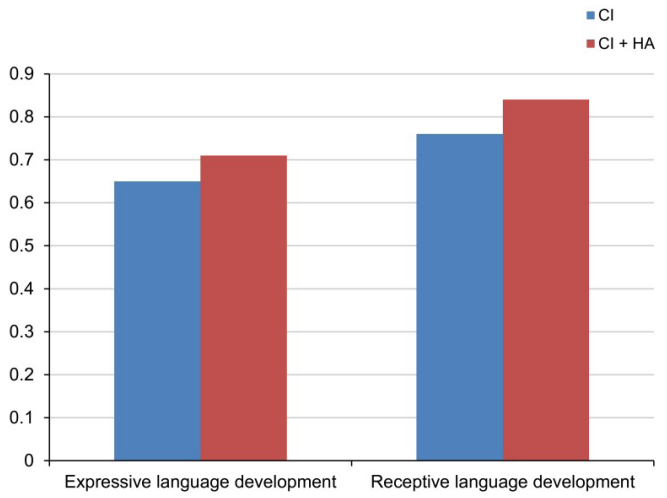

Figure 2. Expressive and receptive language development of the groups at 36th month.

Discussion

The results of this study suggest that both receptive and

Table 2. Expressive and receptive language development of CI and CI+HA comparison by month

	CI		CI+HA			
	ELD	RLD	ELD	RLD	P1	P2
0. m*	0.44±0.13	0.44± 0.12	0.47±0.12	0.48±0.13	0.44	0.209
1. m*	0.43±0.11	0.44±0.10	0.47±0.11	0.50±0.14	0.11	0.032
6. m*	0.52±0.15	0.53±0.16	0.71±0.21	0.69±0.22	0.001	0.003
12. m*	0.59±0.20	0.64±0.19	0.76±0.17	0.76±0.16	<0.001	0.011
18. m*	0.65±0.19	0.70±0.18	0.76±0.15	0.80±0.16	0.014	0.027
24. m*	0.69±0.20	0.75±0.18	0.79±0.16	0.86±0.16	0.041	0.013
36. m*	0.65±0.17	0.71±0.17	0.76±0.14	0.84±0.13	0.009	0.002

CI: Cochlear implant group, CI+HA: Cochlear implant+Hearing aid group, ELD: expressive language development, m: month, RLD: receptive language development, * mean±SD, P1: ELD CI vs CI+HA, P2: RLD CI vs CI+HA

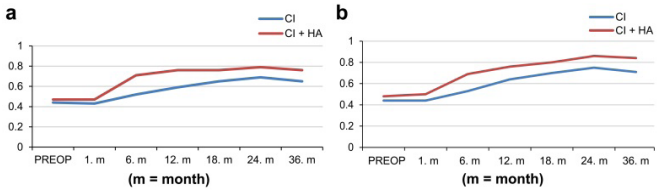

Figure 3. Auditory perception performance of the groups. LittlEARS® (a), MAIS (b), MUSS (c)

expressive language development and auditory perception performance are significantly better in children using bimodal stimulation hearing aids than in those using a cochlear implant alone. Permanent hearing loss in childhood entails a lifetime of negative impacts on affected individuals and their families. Early diagnosis and treatment of these children are essential for correct speech, language

Table 3. LittlEARS®, MAIS and MUSS test results of the groups' comparison by month

	CI			CI+HA					
	LittlEARS	MAIS	MUSS	LittlEARS	MAIS	MUSS	P1	P2	P3
0. m*	0.92±1.3	4±3.5	2.2±1.9	0.92±1.3	3.7±2.3	2±1.1	>0.05	>0.05	>0.05
1. m*	3±2.5	8.5±4.6	5±2.9	3.2±2.1	8.4 ± 3	6.3± 2.9	>0.05	>0.05	>0.05
6. m*	12.6±6.2	20±6.9	9.2±4.1	15.5±8.6	24.4±7.9	13.2±4.1	0.16	0.02	<0.01
12. m*	20.8±6.6	27.8±6.7	15.2±5.4	24.6±6.5	34±5.1	18.9±5.1	0.02	<0.01	<0.01
18. m*	27.1±6.1	32.9±6	20±6.6	30.1±4.7	38.6±2.2	24.9±4.7	0.03	<0.01	<0.01
24. m*	31.5±4.5	36.8±1	25.7±6.7	33.2±3	39.8±1	30.3±3.7	0.02	<0.01	<0.01
36. m*	33.7±3	39.1±1.9	32.2±6.5	34.9±0.2	40±0	38.1±3	0.01	0.01	<0.01

CI: Cochlear implant group, CI+HA: Cochlear implant+hearing aid group, MAIS: Meaningful auditory integration scale, m: month, MUSS: Meaningful use of speech scale, *mean ± sd P1: LittlEARS CI vs CI+HA P2: MAIS CI vs CI+HA P3: MUSS CI vs CI+HA

and cognitive development. Hearing and language learning processes begin at birth and are complete to a greater or lesser extent within approximately four to five years of development in normal children. Neuroplastic activity at this time is at a maximum level, and the various stimuli activate neuronal encoding of language in the fastest and most accurate way. Integration begins within the learning and language centres of the brain. The activity of the auditory cortex begins increasing at birth.[14] This activity of the cortex does not occur in children with bilateral pre-lingual hearing loss of a severe to profound degree. Although unilateral cochlear implants do confer many benefits, including improved speech perceptual ability in implanted children, in daily life, children with cochlear implants continue to experience problems under challenging listening conditions such as noisy classrooms, playgrounds or within the house, due to the auditory stimuli being unilateral.[15] These difficulties can be obviated by bilateral cochlear implants or, where this is not possible, by providing bimodal hearing via the use of a hearing aid.

The benefits of bimodal hearing are reported throughout the literature.[16-18] Speech perception skills when both a cochlear implant and hearing aid are used simultaneously may be better than when hearing is unimodal, using a cochlear implant alone. In patients with bimodal hearing, single-word recognition may undergo a 15%–20% improvement, and a 20%–30% improvement in sentence recognition when exposed to background noise has been observed. Such patients are also better able to locate the origin of sounds.[19-21]

There is little doubt that cochlear implants are responsible for improving speech and language skills in children with hearing loss, but their language skills overall continue to be weaker than their hearing peers. In children with hearing loss, bimodal stimulation provides early access to vocal pitch contrasts that are important for early speech perception.[22] Nittrouer and Chapman investigated bilateral cochlear implanted infants with bimodal stimulation experience and found that they manifested improved language development compared to bilaterally implanted infants lacking such experience.[23]

The native language of children with CI may exert an effect on language development. Using an identical test battery may introduce a handicap when used to evaluate the development of different languages spoken in different countries. For this reason, different countries should use an appropriately adapted test when assessing development of the particular local language. Iwasaki et al [24] employed a package for the assessment of language development in Japanese hearing-impaired children (ALADJIN) to evaluate the language development in children using cochlear implants and hearing aids together. They report that children who used both a cochlear implant and hearing aid had superior language development to those using a cochlear implant alone. In our study we used the Turkish-language validated version of the Pre-school Language Scale-4. We observed that both expressive and receptive language development were significantly higher in the CI+HA group than in the CI group, from the sixth month following implant surgery.

Perception refers to the ability of our different senses to process the information received from the environment. Auditory perception can be defined as the ability to receive and interpret information reaching our ears through the air or other media in the form of audible frequency waves. Since auditory perception is involved in virtually every task we undertake, naturally it plays a vital role in our daily life, giving us the ability to interact adequately with the environment, communicate fluently, be alerted to potential dangers around us, and even to enjoy music. Numerous studies have proven that unilateral cochlear implantation (i.e. unimodal hearing) enhances auditory perception in children with severe to profound hearing loss.[25,26] Bimodal stimulation is a less well-researched area. Bimodal stimulation promotes central integration of auditory stimuli and supports the acquisition of auditory perceptual skills. Thus, whilst stimulating the contralateral auditory pathway is not crucial for the development of meaningful hearing, it does provide additional perceptual benefits compared to not using the hearing aid.[27] There is evidence suggesting that bimodal stimulation augments the performance of auditory perception by the use of a cochlear implant alone.[28,29] Similarly, in all the three test batteries we used (LittlEars®, MAIS and MUSS), we found that auditory perceptual performance was significantly higher in the CI+HA group than in the CI group.

Looking at the benefits obtainable, one would be tempted to suppose the presence of residual hearing in the opposite ear were essential. While electrical stimulation has a greater effectiveness at higher frequencies, acoustic amplification is more effective in boosting the lower frequencies. Bimodal stimulation may be more advantageous than bilateral cochlear implantation in cases where meaningful residual hearing is present, since perception of pitch information,

including voice pitch contrasts in speech, is directly influenced by the level of hearing at low frequencies.[22,30] Against this explanation, Morera et al [8] state that the amount of residual hearing level cannot be used to predict the benefits of bimodal stimulation. Similarly, Beijen et al [31] hypothesise that the benefits of bimodal stimulation are seen, regardless of whether residual hearing is present or not in the opposite ears of children using unilateral cochlear implants. The results of the present investigation support that hypothesis. Although the PTAs of the patients in the CI+HA group were worse than 100 dB, the results confirmed benefit on language development and auditory perception performance from bimodal stimulation in these patients.

Conclusion

The key finding in this study is that bimodal stimulation should be recommended to appropriate patients, regardless of the presence of residual hearing, to allow them to experience a social and academic life similar to their hearing peers. Contralateral hearing aid use contributes to language development and auditory perception performance in unilaterally implanted children with profound hearing loss. The retrospective methodology used here imposes certain limitations and, thus, we can expect stronger evidence to be obtainable in future studies using a prospective methodology and enrolling greater numbers of participants.

Ethics Committee Approval: The study protocol was approved on 13/10/2014 and permission number 469 was granted by the Gazi University Hospital Corporate Ethics Committee.

Informed Consent: All participants provided written informed consent to the study.

Author Contributions: Designing the study – H.T.; Collecting the data – M.Ç., Ş.A.; Analyzing the data – M.Ç.; Writing the manuscript – M.Ç.; Confirming the accuracy of the data and the analyses – İ.B.

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Computed Tomographic Findings in the Nasal and Paranasal Sinuses of Patients Scheduled for Rhinoplasty at Mostafa Khomeini Hospital between 2011-13

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Abstract

Objective: Rhinoplasty is done to improve the morphology of the nose whilst also restoring its physiological function. Currently it is possible to perform rhinoplasty and endoscopic sinus surgery simultaneously. The need has therefore arisen to evaluate anatomical variation within the nose and paranasal sinuses in symptom-free patients who are scheduled for rhinoplasty. Coronal computed tomography (CT) is the standard method used to evaluate the nose and paranasal sinuses. We aimed to evaluate the frequency of anatomical variation noted within the nose and paranasal sinuses on coronal CT in patients scheduled for rhinoplasty.

Methods: This is a descriptive, cross-sectional, retrospective study of CT findings in 84 patients who underwent rhinoplasty at Mostafa Khomeini Hospital, Shahed University, Tehran between 2011-13. The CT findings were evaluated in three different predefined categories: nasal septal, nasal turbinate, and paranasal sinuses abnormalities.

Results: The study involved 84 individuals, consisting of 26 men and 58 women. The most frequent findings in the nasal septum category were nasal septal deviation (69.04%) and nasal septal spur (33.33%), whereas concha bullosa (45.23%) and inferior turbinate hypertrophy were the most frequent abnormality in the nasal turbinates category. Mucosal thickening of the sinuses (45.23%) and partial opacification of the sinuses (21.42%) were the most common findings in the paranasal sinuses.

Conclusion: The high frequency of anatomical variation, inflammatory and congenital abnormal findings in CT images of the paranasal sinuses in patients scheduled for rhinoplasty indicates that preoperative CT examination may be useful in avoiding multiple surgical operations and in reducing costs.

Keywords: Rhinoplasty, computed tomography, nasal septum, nasal concha, paranasal sinuses.

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Introduction

Rhinoplasty is a surgical procedure which modifies the appearance and function of the nose by alteration of the nasal skin, cartilages, and bones. It is either corrective or cosmetic. Corrective rhinoplasty is performed in cases of nasal tumour, congenital defect, trauma or surgical injury. The aim of cosmetic rhinoplasty is to enhance the appearance of the nose while maintaining nasal function.[1] Nasal cosmetic surgery is one of the most common and yet interesting and challenging types of cosmetic procedure. The final result of the operation cannot be predicted with certainty since the dynamicity of the nasal tissues can influence the outcome. In addition, there are both normal and abnormal variations within the nasal and paranasal sinus structures that can affect the final results.[2]

Given that the majority of patients seeking rhinoplasty have nasal obstruction or sinus problems, such as inflammatory disorders, anatomical deformity, or osteomeatal complex obstruction and that simultaneous rhinoplasty and functional endoscopic sinus surgery (FESS) has become feasible, careful evaluation of such problems prior to surgery is clearly crucial.[3,4] Computed tomography (CT) is the method of choice and the most reliable modality for evaluating the extent of sinonasal disease and displaying the bony and soft tissues and anatomical variations as well as congenital and inflammatory problems of the nose and paranasal sinuses.[5-7] The aim of the present study was to evaluate the CT findings in three different areas, i.e. nasal septum, nasal turbinates, and paranasal sinuses, in patients scheduled for rhinoplasty. By identifying anatomical variations and pathological problems, more suitable preoperative surgical plans can be drawn up, which can save time and money as well as prevent multiple operations and related complications.

Materials and Methods

This was a descriptive, cross-sectional, retrospective study of CT findings in 84 patients who underwent rhinoplasty at Mostafa Khomeini Hospital, Shahed University, Tehran during 2011-13. Patients with a previous history of rhinoplasty, septoplasty or FESS, as well as those with acute sinusitis, were excluded from the study.

The CT findings on the patients were gathered and entered into a specially designed data collection form. The patients were informed about the aims and protocol of the study and informed consent was obtained verbally from them. The data were sorted into three separate categories: nasal septum (spur, tubercle, deviation, etc.); nasal turbinate (concha bullosa, hypertrophy, paradoxical middle turbinate, etc.) and paranasal sinus disorders (total or partial opacification, mucosal thickening, cyst retention, etc.). All CT examinations were performed in coronal section with 3-mm cuts using a Shimadzu Model 8500 scanner (Shimadzu Corporation, Kyoto, Japan). The data from the patients' archived files as well as the CT findings were assembled, then analysed using the Statistical Package for the Social Sciences application (v. 18; SPSS Inc., Chicago, IL, USA). Descriptive statistics were reported.

Results

The age range of the 84 cases scheduled for rhinoplasty was 19-44 years (mean=25.3 years), of which, 58 (68.29%) were female and 26 (31.71%) male. Amongst nasal septal disorders, deviation was the most common (58 patients, 69.04%), whereas concha bullosa was the most common pathology to affect the nasal turbinates (38 patients, 45.23%) (Table 1). Increased mucosal thickening was the most prevalent pathology involving the paranasal sinuses. Increased mucosal thickening was most common at ages 25-29 years. There was no correlation with the sex of the patient. The other paranasal sinus findings, including anatomical variations, are presented in Tables 2 and 3.

Discussion

Many patients with cosmetic nasal concerns also have functional complaints such as sinus problems and/or nasal septum or turbinate abnormalities. The surgeon operating on the nose needs to have the full picture about these functional problems before surgery commences. In addition, many patients with sinonasal complaints also seek cosmetic improvement. Clear and complete information about the sinonasal anatomy and its variants in individual cases allows for a better surgical approach. Preoperative paranasal sinus CT scanning is the modality of choice for diagnosis and assessment of the nasal cavity and paranasal sinuses. In this study, we assessed paranasal sinus CT scans of patients who went on to have rhinoplasty.

Evaluating the nasal septum constitutes a major part of the assessment of the nose and paranasal sinuses. Paranasal sinus CT scanning is very accurate in the evaluation of the nasal septum prior to septorhinoplasty.[8] Addressing any issues with the septal cartilage is a key task, since the septum is the most important source of grafts used in rhinoplasty. According to Bolger's theory, non-traumatic nasal

Table 1. Computed tomographic findings of nasal septum and turbinates in patients underwent rhinoplasty.

Nasal septum and turbinate pathologies	Right (n)	Left (n)	Bilateral (n)	Total (n)
Septal deviations	28 (33.33%)	30 (35.71%)	-	58 (69.04%)
Septal spur	10 (11.9%)	18 (21.42%)	-	28 (33.33%)
Concha bullosa	14 (16.66%)	6 (7.14%)	18 (21.42%)	38 (45.23%)
Inferior turbinate hypertrophy	-	4 (4.76%)	8 (9.52%)	12 (14.28%)
Paradoxical middle turbinate	-	-	10 (11.9%)	10 (11.9%)

Table 2. Computed tomographic findings of paranasal sinuses in patients underwent rhinoplasty.

Paranasal sinus	Side	Increased mucosal thickening (n)	Partial Opacification (n)	Total Opacification (n)	Retention cyst (n)	Osteomeatal obstruction (n)
	Right	2 (2.38%)	-	-	-	-
Ethmoid sinus	Left	2 (2.38%)	-	-	-	-
	Bilateral	8 (9.52%)	-	-	-	-
	Right	-	-	-	-	-
Frontal sinus	Left	2 (2.38%)	-	-	-	-
	Bilateral	-	4 (4.76%)	-	-	-
	Right	6 (7.14%)	4 (4.76%)	-	4 (4.76%)	-
Maxillary sinus	Left	8 (9.52%)	4 (4.76%)	-	-	2 (2.38%)
	Bilateral	10 (11.9%)	6 (7.14%)	-	-	-
Total		38 (45.23%)	18 (21.42%)		4 (4.76%)	2 (2.38%)

Table 3. Computed tomographic findings of paranasal cells in patients underwent rhinoplasty

Paranasal cells	Right (n)	Left (n)	Bilateral (n)	Total (n)
Haller cell	-	4 (4.76%)	-	4 (4.76%)
Agger nasi cell	2 (2.38%)	2 (2.38%)	6 (7.14%)	10 (11.9%)
Onodi cell	2 (2.38%)	2 (2.38%)	4 (4.76%)	8 (9.52%)
Frontal cell	4 (4.76%)	4 (4.76%)	4 (4.76%)	12 (14.28%)

septal deviation, especially where the cartilage and vomer join, can be seen in 10% of the normal population.[9] This prevalence has been reported as between 21% and 75% in different studies.[7,10-15] In our study, the prevalence of a nasal spur was 33.33% and nasal septal deviation was found in 69.04% of cases. The prevalence of nasal septal deviation in our study was very similar to that found in reports from other parts of Iran. The difference in prevalence seen when compared with the reports from other countries may be attributable to racial differences, larger sample sizes, or different study populations (for example, studies including patients with sinusitis).

Concha bullosa (i.e. pneumatisation of the middle turbinate) ranges in size from very large to very small. It can

lead to mucosal contact within the osteomeatal complex, middle meatus obstruction, ethmoiditis, maxillary sinusitis, and even mucocoele. The prevalence of concha bullosa in our study was 45.23%, while it has been reported to be between 9.09% and 61% in the literature.[7,10-12,15,16] The reason for the apparently varying prevalence is that different definitions of the concha bullosa are in use. In studies reporting a higher prevalence, any air in the middle turbinate was considered concha bullosa, whilst in our study, only air in the bullous part of the middle turbinate was counted as concha bullosa.

Another anatomical variation in middle meatus is paradoxical middle turbinate, a condition in which the middle turbinate deviates laterally. This condition can lead to stricture of the middle meatus and sinusitis, either per se or when accompanied by other anatomical variations. The prevalence of paradoxical middle turbinate in our study was 11.9%, while the prevalence has been reported as 3.03% to 26.1% in other studies.[9,15,17-19] In all the studies mentioned above the prevalence of a paradoxical middle turbinate of sufficient degree to affect the middle meatus was evaluated, whilst in our study we evaluated the prevalence of paradoxical middle turbinate, whether it could affect the middle meatus or not.

More than 20% of patients referred with nasal obstruction have turbinate hypertrophy. Inferior turbinate hypertrophy can be an important factor in nasal obstruction, especially in those patients with maxillary crest or nasal septal deviation. Severe nasal septal deviation can lead to compensatory hypertrophy of the inferior turbinate on the contralateral side.[20] In our study the rate of inferior turbinate hypertrophy was 14.28%.

Agger nasi cells are any of the anterior ethmoid cells, which are located in the anterior of the frontal recess. They normally lead to no complications. However, if they extend posteriorly, they may constrict the frontal recess, leading to frontal sinus problems. An agger nasi cell, due to its vicinity to the lacrimal sac, may lead to inflammation of the lacrimal sac, producing tearing. The prevalence of such conditions varies between studies, ranging from 2% to 80%.[7,9-11,21,22] The prevalence of such cells in our study was 11.9%. The reason for such differences in reported prevalence is that in some studies only large agger nasi cells were considered, whilst in our study, in line with the work of Zinreich [23], any air cell below the frontal sinus and bordered superiorly by the frontal recess, located laterally to the lacrimal sac, and anteriorly to the lacrimal bone was considered an agger nasi cell.

Haller cells are defined as ethmoidal air cells that may protrude into the orbital floor and narrow the maxillary ostium. The frequency of these cells has been reported as 4% to 39%, in various studies.[7,24] Significant pneumatisation within such a cell can lead to obstruction of the maxillary sinus orifice, resulting in ethmoiditis and maxillary sinusitis, but the presence of such a cell is not necessarily pathological. In our study the prevalence of Haller cells was 4.76%. Our study found a prevalence in line with other studies conducted in Iran. The prevalence is low compared to other anatomical variations in the paranasal sinuses. Although there is an apparent difference in the reported prevalence in the literature, this can be explained by use of differing definitions of the Haller cell, or because of differences in the CT technique used.

The Onodi cell is the most posterior cell of the ethmoid. It is very important because of its vicinity to the carotid canal and the optic nerve. Its position should be considered during endoscopic sinus surgery. The prevalence of an Onodi cell in Talaiepour's study was 7% [11], in Stammberger's 8% [25], and in our study 9.52%. The results are similar in each study.

Anterior ethmoidal air cells are referred to as frontal recess cells if they are positioned towards the frontal area. The prevalence of this type of cell has been reported as between 20%-41% in various studies.[26-28] In our study, the rate of a frontal cell was 14.28%. Such a difference in prevalence may be due to the fact that different populations were involved in the study. In Karzki's study [26], patients with chronic rhinosinusitis were investigated, whilst in Han's research [28], Chinese patients were studied. Furthermore, unlike in our study, Han's study involved axial CT as well as coronal CT to diagnose all types of frontal cells.[28] In the study by Abdi et al [16] involving patients referred with symptoms of chronic sinusitis, osteomeatal complex obstruction was seen in 45.6% and pansinusitis in 17.3% of the patients. In another study, a significant relationship between some anatomical variations and the extent of sinus mucosal disease has been reported following evaluation of CT scans.[7] As these studies were done on patients with chronic sinusitis, their findings cannot be directly compared with our results. We could not identify any other study done on the prevalence of sinus diseases in individuals without sinus symptoms.

Conclusion

We have discovered that there are anatomical variations and particular findings in the CT findings of patients scheduled for rhinoplasty. The most frequent findings were septal deviation and concha bullosa. The high frequency of anatomical variation, inflammatory and congenital abnor-mal findings in CT images of the paranasal sinuses in pa-tients scheduled for rhinoplasty indicates that preoperative CT examination may be useful in avoiding multiple surgi-cal operations and in reducing costs, as well as managing such conditions.

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

Author Contributions: Designing the study – M.A., M.E.Y, P.I.; Collecting the data – M.A., M.E.Y, P.I.; Ana-lysing the data – M.A., M.E.Y, P.I.; Writing the manuscript – M.A., M.E.Y, P.I.; Confirming the accuracy of the data and the analyses – M.A., M.E.Y, P.I.

Conflict of Interest: The authors have no conflict of in-terest to declare.

Financial Disclosure: The authors declare that no finan-cial support was received for this paper.

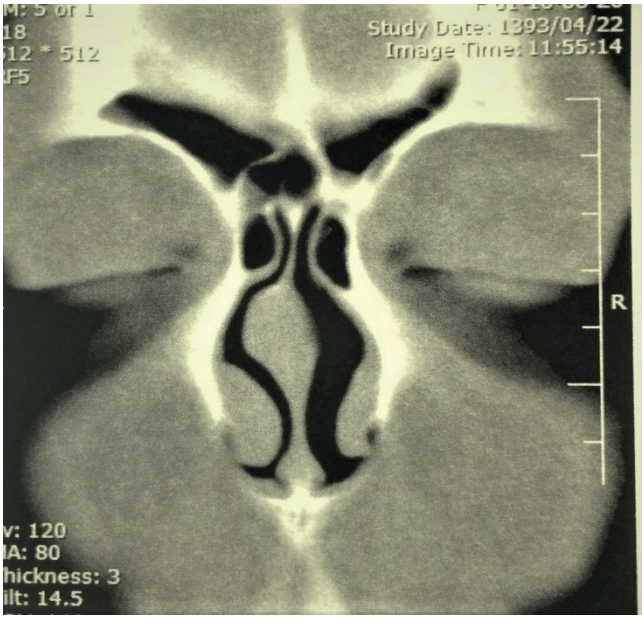

Figure 1. A coronal CT image of an agger nasi cell.

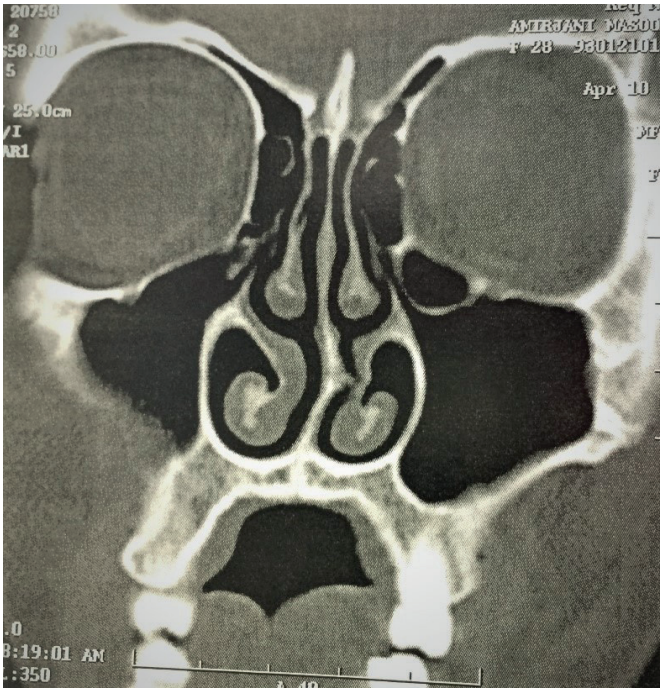

Figure 2. A coronal CT image of a Haller cell

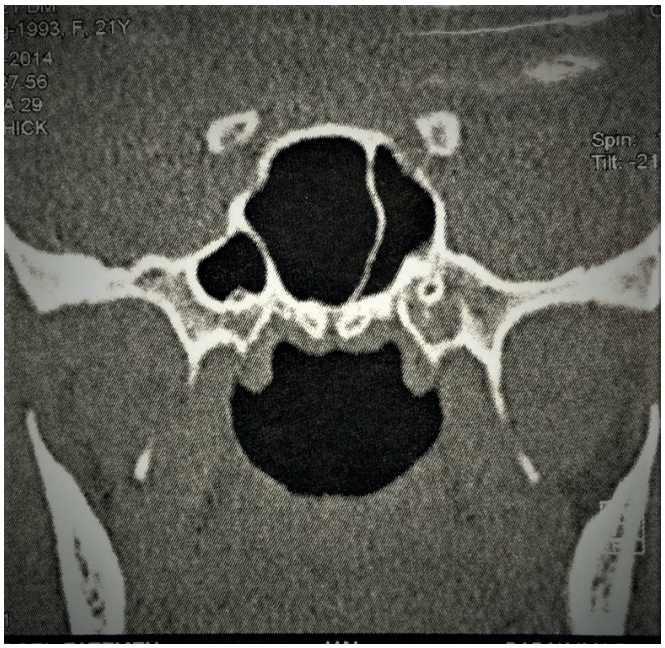

Figure 3. A coronal CT image of an Onodi cell

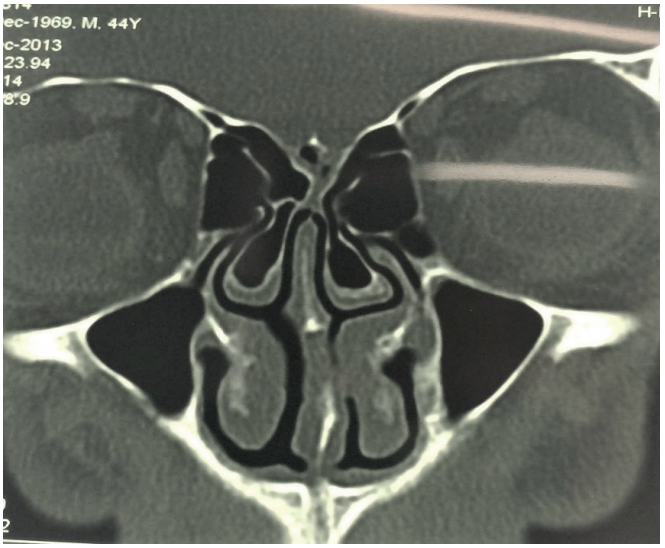

Figure 4. A coronal CT image of a concha bullosa.

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Head and Neck Schwannomas: A Tertiary Referral Single-Centre Experience

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Abstract

Objective: The aim is to share our experience of the clinical features and important issues encountered in diagnosing and treating multiple head and neck schwannoma cases seen at our centre, all of which went on to have surgical treatment.

Methods: This was a retrospective review of the medical records of cases over an 11-year period (2007–2018) diagnosed with schwannoma by post-surgical histopathology. The demographic characteristics of these cases, along with their clinical characteristics, namely, tumour location, preoperative diagnostic tests undertaken, surgical approach used, and any postoperative complications, including the management of such complications, were reviewed.

Results: A total of 31 patients (18 male, and 13 female) were included in the study. Contrast-enhanced magnetic resonance imaging was the most commonly used pre-operative diagnostic method (77%). 58% of the cases were extra- and 42% intra-cranial. Extracranial schwannomas were noted to arise from several different areas of the head and neck region. The most common neurological deficit post-operatively was facial paralysis.

Conclusion: Since head and neck schwannomas can develop from any area where the nerve sheath is present, they may present with a wide variety of non-specific symptoms. The treatment plan should be made with the anticipated preoperative and postoperative neurological deficit firmly in mind.

Keywords: Schwannoma, head and neck neoplasms, neurilemmoma.

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Introduction

Schwannomas, also known as neurilemmomas, are encapsulated, solitary, benign and slow-growing tumours originating from the sheaths of peripheral, cranial or autonomic nerves. Approximately half of these rare tumours occur in the head and neck region, but they can occur anywhere in the body.[1] They are generally classified according to their anatomical location and the nerve from which they originate.[2] Schwannomas can be extra-, intra- or transcranial. In addition, extracranial schwannomas of the neck are also classified into lateral or medial. While tumours arising from the brachial and cervical plexuses are considered to be lateral, tumours originating from the glossopharyngeal, vagus, accessory, and hypoglossal nerves are considered of medial type.[3]

The symptoms and signs of schwannomas vary since they can be found in many different locations within the head and neck region. They typically present with rather non-specific symptoms and therefore are easily overlooked in the differential diagnosis. Malignant transformation is rare but should be suspected if pain or neurological deficits are present. Preoperative diagnostic modalities can provide valuable information and even diagnostic evidence, but a definitive diagnosis requires histopathological examination.[2] However, whilst surgical excision does permit a definitive diagnosis and treatment to occur, there is also the potential for postoperative morbidity. The rarity of tumour occurrence, the uncertain presentation and difficulty in diagnosis, and the potential morbidity associated with surgical treatment mean managing a schwannoma is clinically challenging. Therefore, in this study, we aim to share the expertise gathered in our centre, by outlining the clinical features and the key issues encountered in diagnosis and treatment of head and neck schwannomas that proceeded to surgical treatment.

Materials and Methods

A retrospective study was undertaken following approval by the Kocaeli University Non-Interventional Ethics Committee (KU/GOKAEK 2018/157). A retrospective review was carried out on cases where schwannoma had been diagnosed histopathologically following surgical excision at the Kocaeli University Otorhinolaryngology and Head & Neck Surgery Clinic between 2007 and 2018, and who had been followed up regularly in the postoperative period. Cases where non-surgical treatment was employed or follow-up did not occur were excluded from the study. Demographic information on all the cases was recorded, as was the presenting complaint, the clinical features of the schwannoma, the location and nerve of origin, any preoperative diagnostic tests undertaken, the surgical approach followed, postoperative complications and their management, and the duration of follow-up. The gathered data were analysed using the Statistical Package for Social Sciences software version v16.0 (SPSS Inc., Chicago, IL, USA). Categorical variables were expressed as numbers (percentage) and analysed using Pearson's chi-square test. A p value of <0.05 was considered statistically significant.

Results

Thirty-one cases of head and neck schwannoma, who underwent surgical treatment in our tertiary referral clinic between 2007 and 2018 were included in the study.

Demographic Information

Of the 31 cases, 18 were male and 13 female. The average age was 43.6 years, with an age range of 9 to 65 years. The peak age of tumour occurrence was the 4th decade. There were no significant differences between male and female cases in terms of demographic variables. Although male cases predominated in the extracranial group, the result was not statistically significant (p=0.439). The age and sex distribution of the cases of extracranial and intracranial locations are given in Table 1.

Table 1. Demographic Characteristics of Head and Neck Schwannoma Patients

Head and Neck Schwannomas	Number of patient n (%)	Age (years) Mean (range)	Sex M/F
Extracranial	18 (58.1)	43.4 (9-65)	12/6
Intracranial	13 (41.9)	44 (22-61)	6/7
Total	31 (100)	43.6 (9-65)	18/13

Location

Eighteen (58%) cases were extra- and 13 (42%) cases intracranial. All the intracranial tumours were vestibular schwannomas. Seven (54%) tumours were located entirely within the internal acoustic canal, whilst 6 (46%) tumours extended to the cerebello-pontine angle. Extracranial head and neck schwannomas were for the most part located in the neck region (n=8, 44%). The other extracranial locations in order of frequency were; middle ear (n=3), tongue (n=2), larynx (n=2), pterygopalatine fossa (n=1), parotid (n=1) and nasolabial region (n=1) (Table 2).

Table 2. Location, clinical characteristics, distribution of schwannomas according to origin nerve and surgical approach.

Location	Complaint	Surgical Approach	Origin Nerve
Extracranial, Neck (n=8, 44%)			
Lateral (n=5, 28%)	Neck mass (n=4) Numbness of left hand (n=1)	Transcervical (n=8)	Cervical sympathic chain (n=3) Brachial plexus (n=1) Unknown (n=1)
Medial (n=3, 17%)	Neck mass (n=2) Headache (n=1)		Unknown (n=3)
Extracranial, Other (n=10, 56%)			
Middle Ear (n=3, 16%)	Facial paralysis (n=3)	Transmastoid (n=3)	Facial Nerve (n=2) Chorda tympani (n=1)
Larinx (n=2, 11%)	Hoarseness (n=2)	Transcervical (n=2)	Unknown (n=2)
Tongue (n=2, 11%)	Tongue Mass (n=2)	Transoral (n=3)	Hypoglossus (n=2)
Nasolabial area (n=1, 5%)	Swelling over upper lip (n=1)		Unknown (n=1)
Parotis (n=1, 5%)	Swelling below the ear (n=1)	Transcervical (n=1)	Facial Nerve (n=1)
Pterygopalatine fossa (n=1, 5%)	Numbness of the face (n=1)	Transnasal (n=1)	Trigueminal nerve (n=1)
Intracranial Internal acustic canal (n=7, 54%) Cerebellopontine angle (n=6, 46%)	Dizziness (n=7, 54%) Hearing loss (n=6, 46%) Tinnitus (n=6, 46%)	Translabyrinthine (n=12) Retrosigmoid (n=1)	Superior Vestibular (n=7) Inferior Vestibular (n=6)

Clinical Features

The presenting complaints in the patients with intracranial tumours was dizziness in 7 (54%) and hearing loss in 6 (46%) patients. Amongst the patients with extracranial tumours, the presenting complaints were: neck mass without pain (6 cases, 33%); facial paralysis (3 cases, 16%); hoarseness (2 cases, 11%); and tongue mass (2 cases, 11%). Numbness in the arm, headache, swelling below the ear, numbness on the face and an over-lip mass, were noted less commonly: these symptoms each occurred only once, i.e. in 5% of cases (Table 2).

Preoperative diagnostic tests

Contrast-enhanced magnetic resonance imaging (MRI) was the most commonly used diagnostic imaging method (n=24, 77%) (Figure 1). All of the 13 cases with intracranial

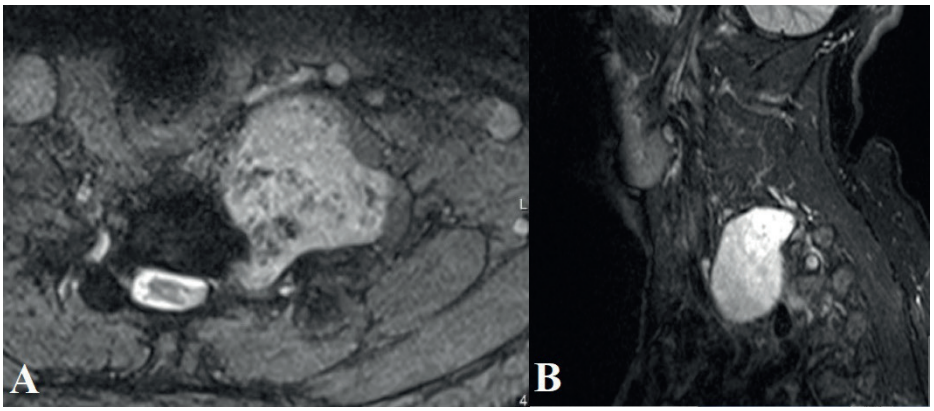

Figure 1. Magnetic resonance imaging of a mass. A. Axial section B. Sagittal section

head and neck schwannoma were assessed by MRI. In 10 cases, schwannoma was reported as the diagnosis, whilst in a further 3 cases schwannoma was mentioned as a differential diagnosis. Of the 18 cases where the tumour was extracranial, eleven patients were imaged using MRI, three patients were assessed by contrast enhanced computed tomography (CT), and two patients were assessed by Doppler ultrasonography (USG). In two cases, where the tumour was located within the tongue, there was no preoperative imaging. These individuals presented with a smoothly circumscribed polypoid mass visible on the tongue surface. The excised material was reported histopathologically as a schwannoma following a diagnostic total excision performed via a transoral approach under local anaesthesia. Only 3 of the extracranial head and neck schwannomas were directly diagnosed on MRI. One case was reported as a possible pleomorphic adenoma, and in one case the lesion was reported as indistinguishable from a paraganglioma, which was included in the differential diagnosis. For the rest of the cases, 6 patients underwent MRI, three patients CT, and two patients ultrasonography. All (11 cases) were reported as a non-specific mass. Fine needle aspiration cytology (FNAC) was performed in 8 of these cases where the diagnosis was indeterminate. Following FNAC, 2 cases were diagnosed with schwannoma.

Surgical Approach

In 26 (84%) of cases, the tumour was able to be totally resected, but in 5 (16%) cases where the tumour could not be totally excised, debulking was instead performed.

A trans-labyrinthine approach (n=12, 92%) was the preferred method in the treatment of the cases of intracranial vestibular schwannoma, with a retrosigmoid approach only used in one case (Table 2). Due to incomplete data, the 3 kHz hearing threshold was not considered when assessing pre- and postoperative hearing. The pure-tone average (PTA) was calculated as the mean of the hearing thresholds at 0.5, 1, and 2 kHz. The average PTA of the 12 patients who underwent surgery via a trans-labyrinthine approach was 72 dB (range:30-118 dB) preoperatively and hearing was unable to be preserved because of the particular surgical technique chosen. The preoperative and postoperative 3rd month PTAs of the single case who underwent surgery via a retrosigmoid approach were 22 dB and 34 dB, respectively.

For the extracranial head and neck schwannomas, total excision was performed by a transcervical approach in 11 (61%) patients, transmastoid in 3 (17%), transoral in 3 (17%) and transnasal approach in one (5%) patient, the route being chosen according to the location of the lesion (Table 2, Figure 2).

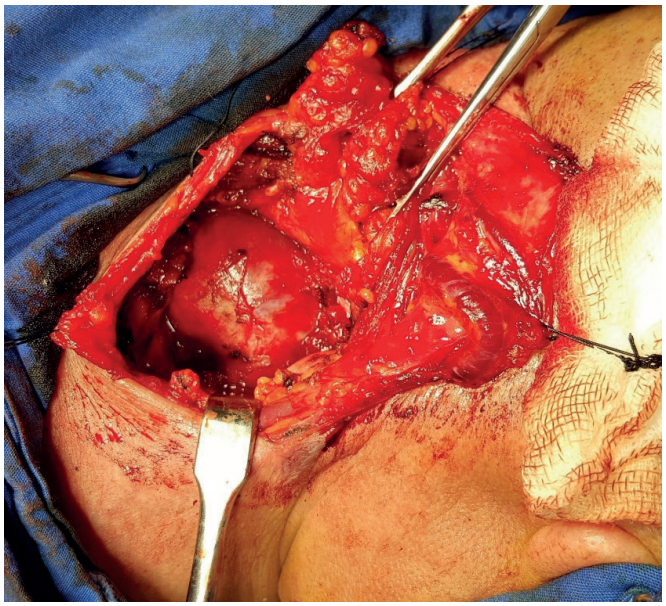

Figure 2. Intraoperative image of cervical tumor.

Nerve origin

For the intracranial schwannomas, the inferior vestibular nerve was the most common nerve of origin, being found in 6 (46%) such cases. For the extracranial schwannomas, the cervical sympathetic chain in the neck (n=3, 37%) as well as the facial nerve in other areas (n=3, 30%) were the most common nerves of origin. In 7 (22%) patients, the neural origin of the tumour could not be determined with precision (Table 2).

Postoperative complications and management

Postoperative complications and complaints were reported in 7 (23%) of the vestibular schwannomas treated via a trans-labyrinthine approach. The complications consisted of: dizziness, rhinorrhoea, cerebrospinal fluid (CSF) leak, facial paralysis and Horner syndrome. Facial paralysis was the most common (n=4, 13%) complication. The management of complications was as follows: pharmacotherapy for vertigo was supplied for one week postoperatively. Rhinorrhoea was noted in one case with facial paralysis, and

lumbar drainage was performed following neurosurgical advice. Due to ongoing CSF leakage, a revision exploration was performed, and facial nerve decompression with hypoglossal nerve anastomosis were performed. One of the cases with facial paralysis was treated intraoperatively with a gold implant to the upper eyelid. Other patients with facial paralysis received medical treatment and were followed up.

The individual in whom the retrosigmoid approach was used had CSF leakage at the wound site. Following referral to neurosurgery, lumbar drainage was performed and the leakage ceased with conservative treatment. Speech impairment was observed postoperatively in one case, with a CT/MRI evaluation revealing an epidural haematoma. This case was treated by neurosurgery. In another case, where the schwannoma had arisen from the brachial plexus, numbness of the hand developed postoperatively. Physiotherapy to the affected hand resulted in acceptable improvement.

Follow up

The mean follow-up period for the intracranial cases was 39 months, with a minimum of 5 months and maximum of 67 months follow-up. No recurrence or malignant transformation was detected during the follow-up period. The mean follow-up period of the extracranial cases was 21 months with a minimum one month and maximum 96 months follow-up. No recurrence or malignant transformation was detected during the follow-up period.

Postoperative histopathological appearances were of benign cells with typical spindle cells and hypercellular (Antoni A) and hypocellular (Antoni B) areas, in all cases. Immunohistochemical studies showed S100 positivity in all cases.

Discussion

Schwann cells are neural crest-derived glial cells that provide myelin isolation to axons of the peripheral nervous system. Schwannomas are benign tumors originating from Schwann cells, first described by Verocay in 1908.[4] Although such tumours may occur at any age, it has been reported that they are most commonly seen between the ages of 20 and 50, with no significant differences in terms of ethnicity or sex.[5-7] In the present study, the majority of patients were between the ages of 20-50 years (n=19, 61%) and the peak occurrence in this sample was in the patients' thirties. Therefore, schwannomas should always be included in the differential diagnosis of head and neck masses in both sexes in both middle and old age.

Since these tumours do not often impair the function of the nerve from which they originate, non-specific symptoms may occur, depending on the location. This results in frequent misdiagnosis of schwannomas. Vagal or glossopharyngeal schwannoma may commonly present with complaints such as dysphonia, dyspnoea, dysphagia, or even cough. Although rare, schwannoma originating from the facial nerve or its surroundings may be the underlying cause of acute peripheral facial paralysis. In our study, three cases presented with facial paralysis, one with facial numbness and another with numbness of the arm. The most common symptom in cases where there was an intracranial vestibular schwannoma was dizziness, and the second most common symptom was hearing loss. In our study, 72% of cases with an extracranial schwannoma presented with a painless neck mass, which is a very common symptom. Since nerve dysfunction is generally not expected in benign cases, the development of neurological deficits should raise the suspicion of malignancy, which must then be ruled out. This situation also strengthens the surgical indication for these patients. Likewise, a careful radiological screening will be required for local and distant metastasis or other concomitant schwannomas.

Although schwannomas of the head and neck most commonly originate in the cervical sympathetic plexus, they may arise from any of the cranial nerves. Leu and Chang [8] reported on 52 cases of extracranial schwannoma, of which 18 were located in the neck. The origin was reported as the cervical plexus in 9 cases, brachial plexus in 3 cases, cervical sympathetic chain in 2 cases, vagus nerve in 2 cases, hypoglossal nerve in 1 case, and the greater auricular nerve in 1 case.[8] Biswas et al [5] reported that in 17 cases out of their series of 31 extracranial schwannomas, the nerve from which the tumour originated could not be identified. The identified origins were reported as the brachial plexus in 5 cases, facial nerve in 3 cases, vagus nerve in 2 cases, hypoglossal nerve in 1 case, cervical sympathetic chain in 1 case, glossopharyngeal nerve in 1 case, and recurrent laryngeal nerve/vagus in 1 case.[5] Hoing et al [9] reported that they were able to find the nerve from which the tumour originated in only 11 of their 20 extracranial schwannoma cases (facial nerve in 4 cases, vagus in 4 cases, and cervical sympathetic chain in 3 cases). In our study, most of the extracranial cases (n=8, 44%) were located in

the neck and only three of them had arisen from the cervical sympathetic chain. In addition, we encountered tumours of facial nerve origin in the middle ear as well as tumours of the chorda tympani. The presence of neurological deficits, especially prior to surgery, may be useful in finding the nerve of origin. However, given that most patients are asymptomatic, detection of the nerve of origin of the tumour may depend on high-quality and well-evaluated preoperative imaging methods.

Various imaging methods are in use to evaluate the tumour before intervention. MRI is undoubtedly superior to other imaging modalities in terms of determining the location of the lesion and its relationship with the surrounding soft tissue, as well as for determining the origin of the lesion. Yasumatsu et al [10] reported that the diagnostic sensitivity of MRI was 80%. In our case series, the diagnosis of schwannoma was reported in 13 (54%) of 24 patients evaluated by MRI. The rate of radiological diagnosis undoubtedly increases with greater experience and interest in this area. We have observed that radiologists, particularly those who have not encountered a schwannoma previously, may overlook schwannoma as a diagnosis, even when typical features are present on imaging. Multidisciplinary evaluation of such cases may improve the differential diagnosis and increase successful recognition of the lesion. FNAC provides benefit in cases where the diagnosis cannot be made by radiological imaging methods alone, but the preoperative diagnostic sensitivity of FNAC alone is inadequate.[10] FNAC was used in 8 cases in which the diagnosis was unclear and vascular pathology had been excluded by imaging. Two such cases were reported as schwannoma. FNAC was inadequate to diagnose the disease sufficiently in our case series and the method did not contribute to preoperative diagnosis. Nevertheless, FNAC retains some usefulness as a way to rule out other malignancies.

The management of schwannomas includes both surgical and non-surgical treatment options, such as watchful waiting, complete tumour excision or intracapsular enucleation. Due to the possibility of malignant degeneration and recurrence, it is important to remove the entire mass wherever possible. Therefore, the recommended treatment is usually surgical excision. On the other hand, since tumours arise from the nerve sheath, even with highly delicate and careful surgery, it is very likely that patients who have no neurological deficit prior to surgery may develop neurological deficits postsurgically. Çakır et al [6] reported a failure rate of 57% in protecting the nerve of origin in their extracranial head and neck schwannoma cases. Therefore, the surgical decision should be informed by an evaluation of the nerve of origin and its likely impact on the patient's quality of life if a postoperative deficit occurs. While nerve dysfunction is more acceptable in cases where the tumour arises from a small sensory nerve, deficits in major nerves and the cranial nerves will be more apparent. In our study, 19% of the cases reviewed had significant neural deficits after surgery. Facial paralysis developed in 4 such patients, although the deficit improved partially in the late postoperative period. In one patient with a brachial plexus-derived tumour, postoperative numbness in the hand improved acceptably with physiotherapy. Giving detailed information before surgery and planning rehabilitation strategy in the postoperative period facilitates the management of the complications and increases success. For all these reasons, the surgical decision should be made considering the balance between risks and benefits, i.e. by comparing the severity of preoperative symptomatology and the anticipated postoperative neurological deficit. In current treatment management algorithms [11], asymptomatic benign lesions have been reported to be good candidates for observation as long as they remain stable Our general approach in clinical practice is to observe asymptomatic extracranial masses, especially those located medially, in patients who can be relied on to attend follow-up.

Even some cases treated surgically required careful postoperative follow-up, with annual MRI. It is very important to inform the patient in detail about what symptoms and signs may develop due either to the disease or to treatment, and to advise the individual to attend hospital without waiting for a follow-up appointment if any additional symptoms occur. The follow-up period should also include the evaluation, follow-up and rehabilitation of treatment-related deficits and complications. The effects of neurological deficits may change or worsen with age, plus further disease is a possibility in young individuals with otherwise full life expectancy. Long-term follow-up should be planned around these issues.

Conclusion

Head and neck schwannomas are rare tumours which may present with multiple various and non-specific symptoms, since they can develop from any area where nerves are present. The treatment decision should be made on the basis of the potential neurological deficits that exist preoperatively, and those anticipated postoperatively. Therefore, accurate

preoperative diagnosis with secure identification of the nerve from which the tumour originates allows patients to be informed about possible nerve damage and make an informed decision about accepting surgery or follow-up. In preoperative diagnostic imaging, MRI is superior to other imaging modalities in determining the relationship of the lesion with the surrounding tissue, its location and especially in identifying the nerve of origin. FNAC may provide additional benefit for the differential diagnosis and may allow the recognition of other pathologies. If surgical excision is planned, total excision of the tumour should be performed and a multidisciplinary team should then work together to address potential postoperative morbidity.

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Ethics Committee Approval: Ethical approval was obtained from the Institutional Review Board of the Kocaeli University, School of Medicine (KU/GOKAEK 2018/157).

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

Author Contributions: Designing the study – M.Ö., S.Ş., F.R.A.; Collecting the data – M.Ö., S.Ş., F.R.A.; Analyzing the data – M.Ö., S.Ş., F.R.A., F.M., M.İ.; Writing the manu-script – M.Ö., S.Ş., F.R.A., F.M., M.İ.; Confirming the accura-cy of the data and the analyses – M.Ö., S.Ş., F.R.A., F.M., M.İ.

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Possible Effects of Chronic Otitis Media with and without Cholesteatoma on Bone Conduction Thresholds: An Evaluation of 112 Cases

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Abstract

Objective: The aim of this study is to evaluate any possible effects of chronic otitis media (COM), with or without accompanying cholesteatoma, on bone conduction thresholds (BCT).

Methods: A total of 112 cases who underwent surgery for unilateral COM between 2006 and 2014 were enrolled in the study. Eighty cases had no cholesteatoma (Group 1). Thirty-two cases did have cholesteatoma (Group 2). Prior to surgery, the temporal bone was evaluated radiologically by use of high resolution computed tomography. The presence of a clinically and radiologically normal contralateral ear was the principal selection criterion for the cases. BCT at 0.5, 1, 2, 4 kHz and their averages were evaluated with pure tone audiogram (PTA) and the normal and diseased ears in each group were compared. This comparison was also made between the diseased ears in Groups 1 and 2.

Results: There was a statistically significant difference observed between the mean BCT scores obtained by PTA for the normal/diseased ears in Group 1 (9.78±0.98/17.34±1.71 dB) and in Group 2 (9.10±0.99/17.58±2.59 dB). This statistically significant difference was observed for each of the four different frequencies ($p<0.0001$). However, there was no statistically significant difference observed between the mean BCT scores obtained by PTA for the diseased ears in Groups 1 and 2 (17.34±1.71/17.58±2.59 dB). Similarly, in the comparison between the diseased ears, the differences of BCT at all four different frequencies (0.5, 1, 2, 4 kHz) lacked statistical significance ($p>0.05$).

Conclusion: As a result of this study, we can say that COM may lead to sensorineural hearing loss. However, we observed that the presence of cholesteatoma does not exert an additional negative effect on cochlear function.

Keywords: Chronic otitis media, cholesteatoma, sensorineural hearing loss.

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Introduction

Chronic otitis media (COM) is a highly prevalent disease worldwide. Its prevalence in developing countries is as high as 72 cases per 1000 inhabitants. Although hearing loss is the main health issue in COM, the complications of the disease may lead to death.[1]

Whilst COM is usually associated with conductive hearing loss, it may also cause sensorineural hearing loss (SNHL).[2-5] Conductive hearing loss due to COM is extensively discussed in the literature, but the relationship between SNHL and COM remains controversial.[1] Inflammatory mediators and lytic enzymes produced by the reactive middle ear epithelium are thought to be the main reason for SNHL in cases of COM. These molecules pass through the round window membrane and cause destruction of hair cells.[6-8] COM with accompanying cholesteatoma is a more destructive process than COM on its own. Thus, it is hypothesized that cochlear damage might be more evident in cases involving cholesteatoma.[9,10]

The aim of this study, then, is to evaluate the relationship between COM and the bone-conduction threshold. We also investigate whether the presence of cholesteatoma may have an additional negative effect on cochlear function.

Material and Methods

The files of patients undergoing surgery between 2006 and 2014 for either unilateral COM without cholesteatoma (Group 1) (n=80), or unilateral COM with cholesteatoma (Group 2) (n=32), were retrospectively reviewed. Ethical approval was first obtained from the hospital ethics committee (Necmettin Erbakan University Meram Medical Faculty, Ethics Committee Decision - 017/777). In all cases, the temporal bone had been evaluated radiologically by high resolution computed tomography prior to surgery. The key selection criterion for inclusion in the study was that patients should have a clinically and radiologically normal contralateral ear (Figure 1). Previous ear surgery, a history of severe head or acoustic trauma, the presence of an otic capsule erosion revealed either radiologically or intraoperatively, or perilymphatic fistula resulted in exclusion from the study. Age, sex, otoscopic findings and audiograms were examined within each patient's file. Bone conduction thresholds (BCT) obtained on pure tone audiogram (PTA) at four different frequencies (0.5, 1, 2, 4 kHz), and their averages, were evaluated and compared, between the normal and diseased ear for the individuals in each group. This comparison was also made between the diseased ears in Groups 1 and 2. The data were analysed statistically using the Independent-Samples Kruskal-Wallis Test contained within the software (SPSS FW, SPSS Inc. Chicago, IL, USA).

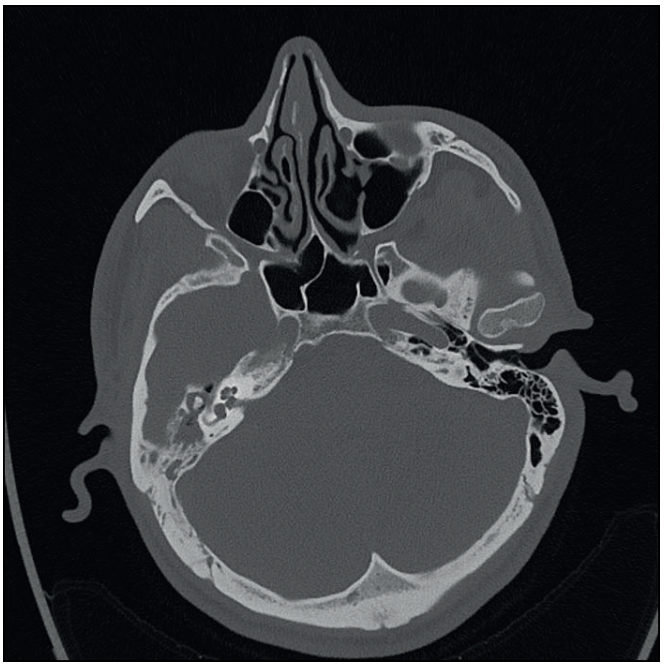

Figure 1. Axial temporal CT, unilateral COM (right).

Results

Group 1 (n=80) consisted of 33 men and 47 women, whereas there were 19 men and 13 women in Group 2 (n=32). The mean age of individuals in Group 1 was 27.60±11.77 (range:11-60) years and in Group 2 was 26.50±13.03 (range:10-60) years.

There was a statistically significant difference between the mean BCT obtained on PTA of the normal/diseased ears in Group 1 (9.78±0.98/17.34±1.71 dB) and Group 2 (9.10± 0.99/17.58±2.59 dB). This statistically significant difference was observed for each of the four different frequencies ($p<0.0001$) (Table 1, Table 2). However, there was no statistically significant difference observed between the mean BCT scores obtained by PTA for the diseased ears in Groups 1 and 2 (17.34±1.71 vs 17.58±2.59 dB). Similarly, in the comparison between the diseased ears, the differences of BCT at all four different frequencies (0.5, 1, 2, 4 kHz) lacked statistical significance ($p>0.05$) using the Independent-Samples Kruskal – Wallis Test (Table 3). These findings are summarized in Figure 2.

Table 1. BCT-PTA at 4 frequencies for normal and diseased ears in Group 1.

Group 1 (n=80)	0.5 kHz	1 kHz	2 kHz	4 kHz	Mean kHz
BCT-PTA Normal ears	10.50±3.61 dB	8.94±3.62 dB	8.94±4.48 dB	10.75±7.68 dB	9.78±0.98 dB
BCT-PTA Diseased ears	19.10±9.18 dB	15.88±8.99dB	18.56±7.92 dB	15.88±9.44 dB	17.34±1.71 dB
p	<0.0001	<0.0001	<0.0001	<0.0001	<0.0001

BCT: bone conduction thresholds, kHz: kiloHertz, PTA: Pure tone audiogram

Table 2. BCT-PTA at 4 frequencies for normal and diseased ears in Group 2.

Group 2 (n=32)	0.5 kHz	1 kHz	2 kHz	4 kHz	Mean kHz
BCT-PTA Normal ears	8.75±4.92 dB	8.13±4.71 dB	9.06±4.30 dB	10.47±7.11 dB	9.10±0.99 dB
BCT-PTA Diseased ears	19.69±8.51dB	14.38±6.32 dB	19.69±9.58 dB	16.56±8.18 dB	17.58±2.59 dB
p	<0.0001	<0.0001	<0.0001	<0.0001	<0.0001

BCT: bone conduction thresholds, kHz: kiloHertz, PTA: Pure tone audiogram

Table 3. BCT-PTA at 4 frequencies for diseased ears in Group 1 and Group 2.

Diseased Ears in Group 1 and Group 2	0.5 kHz	1 kHz	2 kHz	4 kHz	Mean kHz
BCT-diseased ears Group 1 (n=80)	19.10±9.18 dB	15.87±8.99 dB	18.56±7.92 dB	15.88±9.44 dB	17.34±1.71 dB
BCT-diseased ears Group 2 (n=32)	19.69±8.51 dB	14.38±6.32 dB	19.69±9.58 dB	16.56±8.18 dB	17.58±2.59 dB
p	>0.05	>0.05	>0.05	>0.05	>0.05

BCT: bone conduction thresholds, kHz: kiloHertz, PTA: Pure tone audiogram

Discussion

Chronic otitis media may cause a clinically significant SNHL without eroding the otic capsule.[10,11] Although the explanation for this phenomenon remains controversial, it has been suggested that inflammatory mediators, lytic enzymes and toxins may penetrate the inner ear via the round-window membrane and cause destruction of the hair cells, especially in the basal turn of the cochlea where the higher frequencies are represented.[5,7,9,12,13] The permeability of the round window membrane increases due to chronic inflammation.[11,14] This pathogenic mechanism may explain the early appearance of an interaural difference in BCT obtained on PTA at higher frequencies (4 kHz) in patients with unilateral COM.[9,13] Cureoglu et al [7] found a statistically significant loss of both inner and outer hair cells in the basal turn of the cochlea in diseased temporal bones. They also demonstrated the destruction of the stria vascularis and spiral ligament.

Blunt or penetrating otologic trauma, an occult perilymphatic fistula, drilling and ossicular manipulation from previous otologic surgery may also produce negative effects on cochlear function.[2,8,10] In this study, in order to

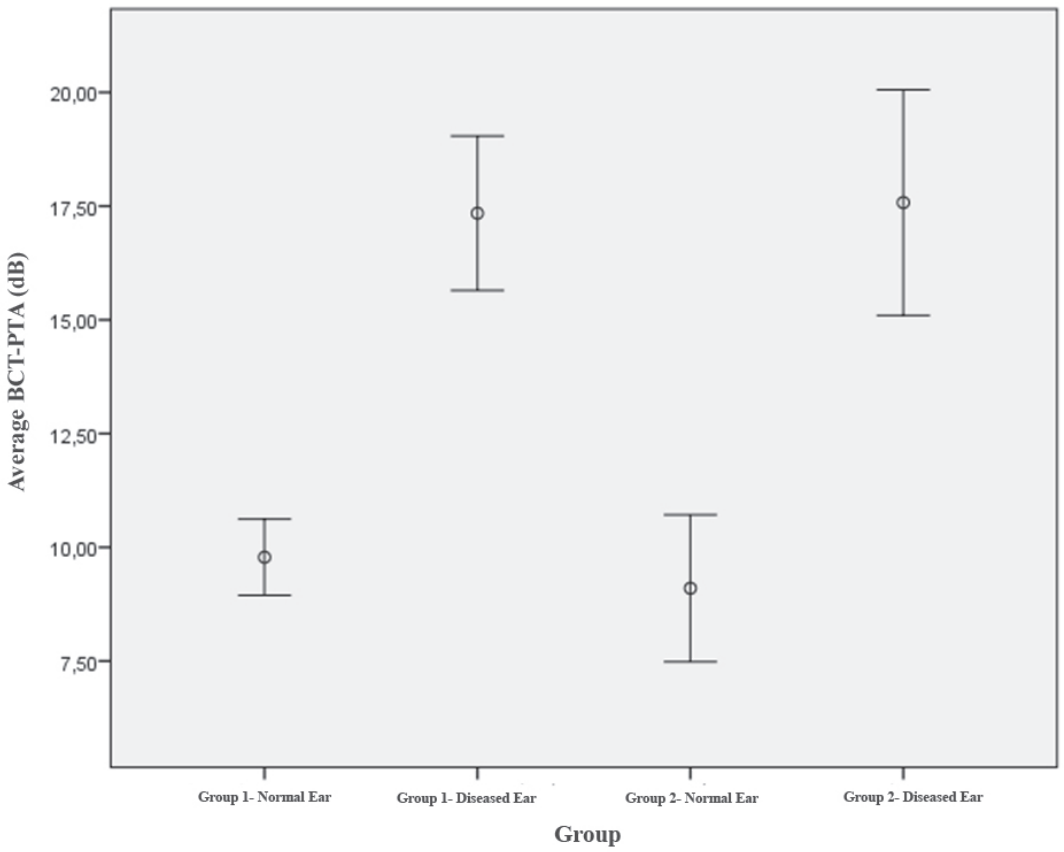

Figure 2. Mean BCT obtained on PTA for normal and diseased ears in Group 1 and Group 2.

focus on the effects of COM alone, individuals with potential SNHL from other causes, as mentioned above, were excluded.

The duration of disease and the existence of accompanying cholesteatoma may be risk factors for the development of SNHL in adult patients with COM. Cochlear damage and SNHL may be more apparent when cholesteatoma co-exists, because of the destructive nature of cholesteatoma.[8,9] Walby et al [12] reviewed case notes relating to the temporal bone of 87 patients with unilateral COM, to ascertain the effects of the disease on the cochlea. They report a decrease in BCT obtained on PTA in diseased ears. However, they failed to find any evidence for inflammatory penetration through the round-window membrane. There was no difference in BCT obtained on PTA between cases of COM with cholesteatoma and those without, in their study. Paparella et al [6] noted an elevation in BCT obtained on PTA at 1, 2 and 4 kHz in 279 COM cases. According to the authors of that study, penetration by inflammatory cells and mediators via the round-window membrane of the inner ear was the principal cause of cochlear dysfunction. The presence or absence of cholesteatoma did not feature among the variables considered by the study.

Levine et al [8] reported that SNHL was greater in the diseased ear at all frequencies than in the contralateral ear in patients with unilateral COM. The difference in mean BCT obtained on PTA in 161 cases was reported to be 9.1 dB (range:5.6-12.8 dB). In that study, the frequencies where the greatest effect was seen were 2 and 3 kHz. The authors also investigated the potential effects of varying middle ear disease severity and patient age on SNHL. They reported that 64% of the 161 cases enrolled in the study consisted of COM with cholesteatoma, and the presence of choleste-

atoma had additional adverse effects on cochlear function. However, this observed difference was only statistically significant at a frequency of 0.5 kHz. It was also reported that there was a small, but statistically significant, effect of age on increasing the BCT.

Papp et al [13] notes a strong correlation between patient age and BCT in COM cases. SNHL has been reported as more pronounced at higher frequencies. According to the authors, due to the proximity of the round window to the basal turn of the cochlea, inflammatory mediators, lytic enzymes and toxins which arise from the middle ear may be present at higher concentrations. Thus SNHL will occur at high frequencies first.

Azevedo et al [15] retrospectively reviewed case notes from 115 patients with COM. Neither the presence of cholesteatoma nor the duration of disease were found to affect SNHL, however, the patient's age was identified as a significant factor. Kolo et al [16] observed a significant degree of SNHL in diseased ears compared to normal ears. However, they did not identify any correlation between the patient's age or duration of disease and the degree of SNHL. Amali et al [4] report that a statistically significant difference in mean BCT was obtained on PTA between normal and diseased ears at all frequencies. The authors found that there was a significant correlation between age and SNHL, but that the duration of the disease did not have a significant effect on SNHL. Furthermore, they report that there is no relationship between the presence of cholesteatoma and the degree of SNHL.

In the present study, a statistically significant difference in BCT obtained on PTA between the normal and the diseased ears in both groups was found for all 4 frequencies tested ($p<0.0001$). However, in the comparison between the diseased ears, the differences of BCT at all four different frequencies (0.5, 1, 2, 4 kHz) lacked statistical significance ($p>0.05$). Ninety percent of our patient sample was under 50 years of age, and the duration of COM was similar, therefore the data were unsuitable for investigating any potential effect of patients' age and duration of disease on BCT.

Our study suffers from certain limitations. The possible effects of age and disease duration on BCT were not evaluated. Moreover, the number of patients included in the study was relatively small.

Conclusion

According to the results obtained in this study, it can be stated that COM may cause SNHL. However, the presence of cholesteatoma does not result in a statistically significant additional negative impact on cochlear function. Surgical management of COM should be routinely recommended to patients in order to circumvent serious complications of COM, such as SNHL.

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Ethics Committee Approval: All procedures involving human participants were in accordance with the ethical standards of the Necmettin Erbakan University Meram Medical Faculty (Ethics Committee Decision – 017/777)

Informed Consent: Informed consent was not required due to the retrospective study design.

Author Contributions: Designing the study – C.H.U.; Collecting the data – C.H.U., D.A., A.Y., F.A.; Analys-ing the data – D.A., A.Y., F.A.; Writing the manuscript – C.H.U., D.A.; Confirming the accuracy of the data and the analyses – C.H.U., D.A., A.Y., F.A.

Conflict of Interest: The authors have no conflicts of in-terest to declare.

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The association between subjective and objective masculine vocal quality in hormone-naïve trans-male individuals

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Abstract

Objective: The aim of this study is to investigate the association between subjective and objective masculine vocal quality in hormone-naïve trans-male individuals.

Methods: Twenty-seven hormone-naïve trans-male individuals were recruited for the study. All the study participants had applied to undergo the gender transition process and been referred to the Voice Clinic. The Self-Perception of Voice Masculinity (SPVM) scale and the mean fundamental frequency (F0) were used to assess any association between subjective and objective vocal masculinity.

Results: The mean age of study participants was 25.3 years. The median F0 lay within the normal limits for cisgender females. However, in 12 out of 27 cases, F0 lay within the gender-ambiguous frequency range. SPVM scores were higher in this ambiguous group, a result with a strong tendency toward statistical significance (p=0.053). A moderate negative correlation between SPVM and F0 scores was observed (r=-0.484, p=0.027). The smoking frequency was high, with 77.8% of individuals in the group as a whole being smokers. Not only were F0 values of smokers lower than those of non-smokers, but their SPVM scores were also higher.

Conclusion: The findings from this study indicate that diversity in objective and subjective voice parameters exists even within the hormone naïve period. There appear to be several factors which influence F0 to a significant extent. Accordingly, a more comprehensive approach is called for when assessing transgender voice at all stages of the gender-affirming medical treatment process. The high frequency of smoking amongst the trans-male population should also be noted as it constitutes a serious health hazard.

Keywords: Transgenders, voice, masculinity, smoking behaviour, voice quality.

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Introduction

The term "transgender" is an umbrella term used to refer to individuals whose gender self-identity mismatches their sex as assigned at birth.[1] The term "cisgender" is used to denote persons whose gender identity corresponds to their sex as assigned at birth.[1] Discomfort arising from a discrepancy between the sex assigned at birth and gender self-identity results in an impairment of psychosocial and mental functioning, known as "gender dysphoria".[2,3] In this article, we will use the term "voice-related gender dysphoria" to denote the discomfort arising due to vocal features that are incongruent, in terms of masculinity and femininity, between the sex assigned at birth and gender self-identity.[4]

Cross-sex hormone therapy is the dominant treatment in the gender transition process as well as voice-related gender dysphoria in trans-male individuals. For the most part, individuals desire and expect their vocal pitch to be congruent with their lifestyle and physical appearance, particularly in terms of their gender identity. Therefore, the pitch-lowering effect of testosterone treatment allows trans-male individuals seeking treatment to achieve a voice that is more compatible with their gender identity and thus hormonal therapy is usually considered to be an effective method to alter vocal quality in the direction desired.[5] However, in spite of hormonal therapy remaining the most effective treatment to achieve masculinisation of the voice, recent studies have reported diversity in treatment response, which indicates that hormonal treatment may not always be satisfactory.[6-8] In a recent meta-analysis, an overall rate of 16% was reported for incomplete satisfaction with vocal quality in spite of hormonal treatment, and in particular subgroups, this dissatisfaction reached a rate approaching 30%.[9] Factors thought to affect treatment outcome include differing sizes of the laryngeal framework and androgen insensitivity, as well as marked variations in the testosterone regime followed.[9] The treatment regimens in use include various administrations and dosages and are not standardised.[10] In any case, it is still unclear to what extent treatment should be maintained and whether or not it should be administered to every individual in an identical manner.[11] To make the matter yet more complicated, even if the pitch-lowering effect is successfully achieved, it may not always be satisfactory from the point of view of how individuals perceive the masculine quality of their own voice.[12]

Trans individuals cannot be considered a homogeneous group in terms of voice perception and expectation, a point we highlighted in a recent study, which highlighted the existence of variation among individuals from the very beginning of the process, even before treatment begins.[4]

Thus, considering the dearth of literature in this area, the aim of our study was to investigate the association between subjective and objective vocal masculinity in the pre-treatment period.

Materials and Methods

Sample

Since its inception in 2004, Kocaeli University's Gender Identity Clinic (KoUGIC) has provided services for trans individuals from an interdisciplinary perspective (a psychiatrist, endocrinologist, gynaecologist, urologist, plastic surgeon, and voice specialist) aiming to assess, protect and manage both the physical and mental health of patients during the gender transition process. The objective is to provide an equitable delivery of high-quality care reflecting the recommendations of the World Professional Association for Transgender Health.[3,4] The phoniatric evaluation protocol includes perceptual assessment of the voice, initial voice recordings, and the use of standardised subjective rating scales as well as videolaryngostroboscopic examination.

The medical records were evaluated from hormone naïve trans-male individuals referred to the Voice Clinic who had completed the perceptual assessment measures as well as having their voice recorded postmenstrually during the same visit. The exclusion criteria were as follows: aged under 18 years, incomplete scales and/or voice samples, individuals who were already under hormonal treatment, and individuals who had received previous voice therapy. The study protocol was approved by the Institutional Review Board of the University of Kocaeli Medical School (KU/GOAEK 2019/272). Written informed consent was obtained from all the participants included in the study.

Measures

Sociodemographic Questionnaire

A sociodemographic questionnaire covering age, job, educational status, socio-economic status, employment status, relationship status, alcohol and/or smoking history, family and/or social support, outward appearance, and current so-

cial gender identity was administered to the participants.

Self-Perception of Voice Masculinity (SPVM)

The self-perception of voice masculinity scale (SPVM) is in use as a subjective voice measure to assess the perception of voice gender. Participants rated their SPVM on a five-point Likert scale. Each item offered the following possible choices, ranging from 1 to 5: 'very female', 'somewhat female', 'gender neutral', 'somewhat male' and 'very male'. The rating scale was based on equal interval scales with very female/feminine at one end and very male/masculine at the other.

Acoustic Analysis

The Computerised Speech Lab software, Multi-Dimensional Voice Program (MDVP) model 5105 (Kay Elemetrics Corporation, Lincoln Park, New Jersey, USA) was used for the analysis of vocal samples. The average of two recorded voice samples of a sustained /a/ vowel was used, following a demonstration by the examiner in a quiet room with less than 50 dB of background noise and with a microphone placed at a distance of 10 cm from the speaker.

The mean fundamental frequency (F0) is a frequently employed major objective cross-gender measure of acoustic difference. [13] Therefore, we chose this measure as the acoustic analysis parameter of interest. Although there is no definitive absolute distinction between masculine and feminine F0, in a recent meta-analysis the following values were used to evaluate the response to testosterone in trans males: cisgender male normative frequencies are at or below 131 Hz, cisgender female normative frequencies are 185 Hz or above, and the gender ambiguous frequency range is 185 Hz or less.[9,13,14]

Statistical analysis was performed using the SPSS v22 (IBM Corp., Armonk, NY, USA) application. Descriptive statistics were generated for all the variables. The Shapiro-Wilk test and graphical examinations were used to test the normality of the data. Non-parametric tests were applied for non-parametric data or for when the sample size was small. Descriptive data are expressed as the mean (plus standard deviation) and the median (with corresponding range). The correlation coefficients analyses between the SPVM measure scores and F0 were performed by means of Spearman's correlation test. The Mann–Whitney U-test was used to compare independent groups. All differences where the p value was 0.05 or less were considered statistically significant.

Results

Twenty-seven hormone naive individuals were enrolled in the study. The mean age was 25.3 years (range:18-43 years). Five of the twenty-seven individuals' social gender identity and outward appearance were still female due to working in a very conservative environment. The sociodemographic and gender transition-related characteristics of all participants are given in Table 1.

Table 1. Sociodemographic and gender transition characteristic data of the participants

Sociodemographic characteristics	**n***	**%**
Educational Status		
College	13	48.1
High School	13	48.1
Middle School	1	3.8
Employment Status		
Student	7	25.9
Employed	17	63.0
Unemployed	3	11.1
Socioeconomic Status		
Low	-	
Moderate	27	100
High		
Relationship Status		
Partnered	14	51.9
Single	13	48.1
Married	-	
Smoking habit	21	77.8
Gender transition-related characteristics		
Presence of family support	14	51.9
Presence of social support	22	81.5
Outward appearance and		
Male	22	81.5
Female	5	18.5
Social gender identity		
Male	22	81.5
Female	5	18.5

* Total Number of patients

The median SPVM score was 3 (range: 1-4), indicating gender neutrality. The median F0 was 190 Hz (range:154-267 Hz), thus within the normal limits for a cisgender female (Table 2). However, 12 out of 27 individuals had an F0 lying within the gender ambiguous frequency range, i.e. less than 185 Hz. Median SPVM scores were higher in this group, a result showing a strong tendency toward statistical significance (p=0.053) when compared with individuals within the cisgender female normative range for F0. The median SPVM and F0 scores of the participants compared to normative F0 values are given in Table 3.

Table 2. Mean (SD) and median (range) SPVM and f0 scores of the participants

	Mean (SD)		Median (range)	
SPVM	2.67	(0.92)	3	(1-4)
f0 (Hz)	196.11	(31.15)	190	(154-267)

f0: Mean Fundamental Frequency, Hz: Hertz, SD: standart deviation, SPVM: Self-perception of voice masculinity

The strength and direction of the correlation between the SPVM scores and F0 was examined using Spearman's correlation test. A moderate negative correlation between the SPVM and the F0 scores was observed (r= -0.484, p =0.027).

The median F0 scores were evaluated in detail depending on the SPVM scores of individuals, and these results are presented in Table 4. None of the individuals perceived their own voice to be very male. As the median F0 decreased, the SPVM scale scores moved towards to a greater perception of masculinity.

The subjective and objective vocal masculinity parameters were also investigated in relation to smoking habit.

Table 4. Median (range) f0 scores according to SPVM

	n (27)	f0 (Hz) Median(range)
SPVM		
1 (very female)	3	223.00 (190.00-262.00)
2 (somewhat female)	8	202.00 (162.00-248.00)
3 (gender neutral)	11	191.00 (167.00-267.00)
4 (somewhat male)	5	168.00 (154.00-190.00)
5 (very male)	-	

f0: Mean Fundamental Frequency,
Hz: Hertz, SPVM: Self-perception of voice masculinity

Although the result was not statistically significant, the F0 values of smokers were lower than those of non-smokers. Additionally, their median SPVM scores indicated a neutral gender perception, while for non-smokers the SPVM scores corresponded to somewhat female (p=0.195) (Table 5). Smoking habit was also compared with the normative F0 values for individuals. Ten out of 12 individuals whose F0 fell within the gender ambiguous frequency range plus

Table 5. Median (range) SPVM and f0 scores of the participants according to smoking habit

	Non-smokers n (6)	Smokers n (21)	P-value*
SPVM Median (range)	2 (1-4)	3 (1-4)	0.195
f0 (Hz) Median (range)	218.50 (168.00-262.00)	190.00 (154-267)	0.195

f0: Mean Fundamental Frequency,
Hz: Hertz, SPVM: Self-perception of voice masculinity
*Mann–Whitney *U*-test

Table 3. Median (range) SPVM and f0 scores of the participants according to normative f0 values

	Gender ambiguous f0 range (≤ 185 Hz) n (12)	Female f0 range n (15)	P-value*
SPVM Median (range)	3 (2-4)	2 (1-4)	0.053
f0 (Hz) Median (range)	168.50 (154.00-184.00)	215.00 (190.00-267.00)	<0.001

f0: Mean Fundamental Frequency, Hz: Hertz, SPVM: Self-perception of voice masculinity
*Mann–Whitney *U*-test

11 of 15 individuals who were within the normal limits for cisgender females were current smokers.

With regard to outward appearance, no significant correlation was observed with either SPVM score or F0.

Discussion

As the overall prevalence of trans individuals seeking treatment has increased globally, research has increased into the psychosocial and physical conditions and the specific expectations and needs of trans-gendered individuals.[15]

The human voice, like the face, contains important social clues about a person's mood, personality, age, and gender, and has even been described as an "auditory face" that facilitates social perception.[16] Therefore, voice is of particular importance for trans individuals. The literature investigating voice in transgender individuals has mostly so far concerned trans-females, and there is a paucity of research focusing on the voice in trans-males.[8] Furthermore, most of the studies involving trans-male vocal characteristics have been conducted with relatively small sample sizes, lack information about pre-treatment vocal status and have often focused on the outcomes of testosterone treatment.[8,9] Although androgen therapy is generally considered adequate to alter the voice in the desired direction for trans-males, recent studies have reported significant diversity in response to treatment.[9] A recent meta-analysis including the results of individuals who had been receiving treatment for at least 1 year, investigated the efficacy of testosterone therapy in masculinizing the voice in transgender individuals.[9] A failure ratio of 21% to achieve the cisgender male normative frequency range (i.e. ≤131 Hz) has been reported. The individuals concerned (21% of cases) were reported to have voices which fell in the gender ambiguous frequency range (i.e. ≤185 Hz).[9]

The findings of this study indicate that diversity in F0 is already present even in the pre-treatment period. This diversity in F0 is also associated with SPVM and, therefore, contributes to the variation in self-perception of voice gender among hormone naïve trans-male individuals. In our study sample, 44.4% of the participants had an F0 already within the gender ambiguous frequency range. This pre-treatment diversity implies that F0 has already been significantly influenced by other factors and thus it is likely that these same confounding factors would have an impact on testosterone treatment outcomes. When evaluating hormone treatment efficacy, therefore, these confounders need to be borne in mind.

As the main therapeutic target is to achieve the vocal quality that the trans individual desires and will be satisfied with, how they perceive their own voice should also be evaluated at the beginning of the gender affirming treatment process, together with F0. Watt et al [17] reported that a perception of their own voice as masculine in character lead to trans-males gaining a greater sense of psychosocial well-being. A decrease in F0 lead to a perception of the voice as more masculine in character, and, for most trans-males, it is important to be recognized as male by the community.[12] In a study by Sandmann et al [18] in both cisgender female and male subjects, the relation of F0 to how masculine or feminine-sounding individuals regarded their own voice was evaluated. While a lower F0 correlates significantly with self-assessment of a voice as masculine in cisgender males, no correlation was reported in cisgender females.[18] Nygren et al [6] reported a moderate to strong negative correlation between F0 and the self-perception of vocal masculinity in trans-male individuals who had undergone hormone therapy for at least 3 months. In our study group of hormone naïve trans-males, a moderately powerful negative correlation was observed between the SPVM and the F0 score. As F0 decreases, the SPVM scale moves towards to a greater perception of masculinity. This might well be related to the high number of individuals in the study sample whose F0 fell within a gender ambiguous range in the pretreatment period.

Vocal pitch and its acoustic correlate, F0, is known to be the most important indicator of voice gender.[13] Various factors other than biological sex can affect F0. Specifically, the ageing process, linguistic differences, and smoking are known to play a role in influencing the F0 of normal healthy individuals.[19]

In the two largest longitudinal series presenting voice data before and after treatment in trans-males, Bultynck et al [20] reported a mean age of 25.6 years (range:17-47) for 80 individuals and Nygren et al [6] reported a mean age of 27 years (range:18-64) for 50 individuals. Along with an increase in prevalence, the age at presentation is also reported to have become lower.[15] The demographic data in our study is comparable with previous studies, consisting as it did of 27 hormone naïve trans-males with a mean age of 25.3 years (range:18-43) seeking gender-affirming medical treatment.

Few studies have reported acoustic parameters across different language contexts before the beginning of hormonal treatment.[6,7,21-24] In the largest study evaluating

the effects of testosterone treatment on vocal F0, which was conducted in Sweden by Nygren et al [6], the mean F0 was reported to be 192 Hz, with a median value of 190 Hz (range:147-242 Hz) at baseline for 50 trans-male individuals. These values fit with the reported reference F0 of 188 Hz for cisgender vocally healthy Swedish females.[25] Deuster et al [7] reported a median speaking F0 of 192 Hz (range: 164-255Hz) at baseline for 11 trans-male individuals, which also fits in with the German cisfemale normative speaking F0 of 163.7 Hz (SD:27.5).[19] In our study, the median and mean F0 were 190 Hz (range:154-267) and 196.11 Hz (SD:31.15), respectively. As expected, at baseline, none of the reported F0 have been within the cisgender male normative limit, and this was the case with our series, too. However, in our series, it was observed that the median F0 was at, or even slightly below, the lower limits of the reported normative F0 value of 223.9 Hz (SD:23.4) for cisgender Turkish females of a similar age range during the postmenstrual interval.[26] In addition, 12 out of 27 individuals were already within a gender ambiguous frequency range (range:154-185 Hz). In fact, there have also been cases within the gender ambiguous frequency range at baseline in other studies.[6,7] In the study by Deuster et al [7], 6 out of 11 cases had an F0 within this ambiguous range, and Nygren et al [6] reported that in 10 out of their 50 cases, F0 was below 175 Hz at baseline, but they failed to comment further on this.

Smoking is another major factor affecting acoustic voice parameters. Studies in healthy cisgender female individuals have reported a significant decrease of 15-45 Hz in F0 due to smoking.[19,27] The effect of lowering F0 may cause cisgender female voices to be perceived as male by others. Indeed, a smoking cisgender woman being perceived as a man on the telephone is a classical clinical presentation of Reinke's oedema, a common laryngeal pathology. Thus, there are several studies reporting more frequent smoking in trans-male individuals seeking to benefit from this effect.[8,21] In a cross-sectional study by Cosyns et al [28] reporting long-term androgen treatment outcomes, only 34 % of 38 trans-males had never smoked. T'Sjoen et al [29], however, reported a non-smoker rate of 80% for 20 individuals, whereas Van Borsel et al [21] reported this rate as 50% based on a sample of 16 cases.[21,29]

An elevated smoking rate of 77.8% was observed in our study group. Although the majority of the sample were aware that smoking has a pitch lowering effect, no individual reported taking up smoking with this aim in mind. While age may not have significantly influenced F0 in our study, smoking frequency likely had a significant effect on lowering the overall median F0. Given that 10 out of 12 individuals who were within the gender ambiguous frequency range were current smokers, this might well be the reason why almost half of our study group had an F0 within the gender ambiguous frequency range even before hormone treatment could begin.

F0 is the principal acoustic difference between feminine and masculine-sounding voices. However, it should be noted that F0 is not the only acoustic parameter influencing the perception of voice gender. Other acoustic parameters such as voice range, sound pressure level and formant frequencies may also play a role.[25,30] Furthermore, the voice quality, resonance, and other aspects such as speech and communication properties differ between females and males.[21] Therefore, the femininity or masculinity of a voice is a very complex issue, about which relatively little is yet understood and this issue should also be taken into consideration when studying trans-voices.

Although these evaluations were performed in conjunction with a psychiatrist and voice specialist and these individuals were also participants in group psychotherapy, leading to a good therapeutic rapport, the relatively small sample size is the main limitation of the study. Second, the results reported may be subject to confounding. An age and smoking matched cisgender female control group was not used for our study, and other objective parameters such as vocal range, sound pressure level, formant frequencies, and speaking F0 were not investigated due to some data being missing from the records. These limitations might cause a degree of uncertainty about the conclusions. Lastly, being a single-institution study and including only individuals actively seeking treatment prevents immediate generalisation of our results to other institutions.

Conclusion

On the basis of our findings, it could be argued that diversity in objective and subjective voice parameters exists not only in cross-sex hormone treatment response, but also within the pre-treatment period. The main aim of androgen treatment is to lower the vocal pitch in order to sound more masculine. However, there are other factors influencing F0 from an early stage, which may bear on treatment efficacy and patient satisfaction. Therefore, a more comprehensive approach should be implemented when assessing transgender voices throughout the gender af-

firming treatment process. Furthermore, smoking, a major confounder in the results, is yet another serious health hazard in an already neglected sexual minority group in terms of general medical health. The high frequency of smokers among the trans-male population should also be taken into account when considering public health.

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

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Successful partial cochlear implantation in a patient with relapsing polychondritis

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Abstract

The author reports on a case of relapsing polychondritis in which partial insertion of a cochlear implant has been successful. A woman with known relapsing polychondritis gave a history of increasingly severe auditory loss of sensorineural type occurring bilaterally. Previous repeated treatments with corticosteroids and immunosuppressive medications had been attempted. A right-sided cochlear implantation with partial electrode array insertion was carried out due to the patient's cochlea being obliterated. The outcome was excellent and the improvement in hearing was maintained. Relapsing polychondritis (RP) is a rarely-seen disorder, the aetiology of which is obscure, but which results in inflammation within cartilage throughout the entire body. The cochlea may become obliterated by this process. Cochlear implantation may be needed in such circumstances. The successful outcome in this case shows a potential role for cochlear implantation in individuals with inner ear disorders produced by immune disorders or relapsing polychondritis.

Keywords: Polychondritis, relapsing, sensorineural hearing loss, cochlear implantation.

Jaksch-Wartenhorst may have been the first person to describe the condition currently termed relapsing polychondritis (RP), writing in 1923.[1] RP is a rare condition which features recurrent exacerbations of inflammation affecting cartilage. The pathogenic basis and aetiology of RP is uncertain. Some 40-50% of individuals with RP develop conditions affecting the inner ear at some point in the disorder.[2,3] This report is of a case of RP in which severe auditory loss had occurred, affecting both ears, and of sensorineural type. Previous treatment with corticosteroids and immunosuppressive medications had been to no avail. A cochlear device was implanted successfully, with partial electrode placement.

Case report

A woman aged 53 years was referred to an ENT clinic in 2004 with a history of profound auditory loss of sensorineural type occurring bilaterally, secondary to RP. She also had vertigo. Total deafness had been present in the left ear for 15 years, and in the right for two months. A diagnosis of RP was reached due to chondritis of the nose, a non-erosive polyarthropathy with negative serology, abnormal vestibulo-cochlear function and appropriate histopathological appearances. Pure-tone audiometric analysis indicated bilateral total deafness. Neither corticosteroid nor immunosuppressant treatment had restored hearing. As expected,

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there was no response on either auditory brain stem response or distortion product otoacoustic emission testing. The temporal bone had been imaged using computed tomography (CT) in 2004, as shown in Figure 1. The appearances were interpreted as indicating an intact cochlea on the right, whilst also showing that the left cochlea was partially obstructed.

It was proposed that a cochlear device be implanted in the right ear. An aortic aneurysm required surgery before the device could be implanted, entailing a delay until 2006. The temporal bone was re-scanned (Figure 2), the appearances at that point showing partial obstruction of the basal turn of the cochlea on the right side, and a more significant obstruction on the left. Right-sided cochlear implantation was performed in 2006 using a Digisonic SP cochlear implant from the Neurelec Company. As we found an obliterated basal turn in the cochlea, the device was partially introduced starting at the second turn of the cochlea. Seven electrodes out of 20 were introduced, and their placement confirmed by transorbital-ocular view.

One month later, the Digisonic SP processor was fitted. Six electrodes were found to be active. Pure tone audiometry showed good thresholds of around 20 dB between frequencies of 250 - 4000 Hz, with the cochlear implant operating in a free field. Intelligibility was around 30% with the cochlear implant alone. The patient reported a high level of satisfaction with the device, which she was using throughout the day. In 2011, the patient benefited from an upgrade to a Saphyr SP processor from the Neurelec Company. There was a definite improvement in the result obtained. Intelligibility with Lafon lists of monosyllabic words was around 50% at 65dB with the cochlear implant alone, and reached 94 % with the cochlear implant used in conjunction with lip reading. The score on the MMBA battery sentences was 41% at 65dB with the cochlear implant alone, and 90% with the cochlear implant used in conjunction with lip reading. The patient scored 205 on the Nijmegen cochlear implant questionnaire.

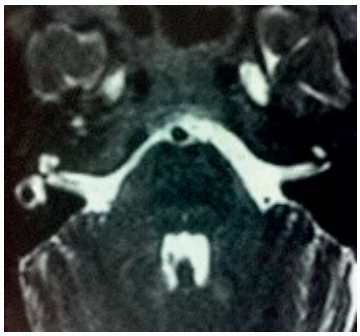

Figure 1. The CT image shows a normal cochlea on the right side and a partial obstruction on the left side.

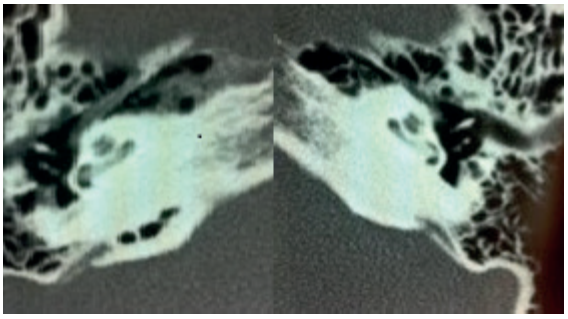

Figure 2. The CT image shows a partial obstruction of the basal turn of cochlea.

Discussion

RP is a disease affecting multiple body systems and has an association with autoimmune disorders such as Sjögren's syndrome, systemic lupus erythematosus and rheumatoid arthritis. The condition was first recognized by Jaksch-Wartenhorst in 1923, being described as a polychondropathy.[1] RP presents with chondritis affecting the pinna, joint inflammation, symptoms affecting the larynx and trachea, chondritis of the nose, eye inflammation, symptoms arising from the audiovestibular apparatus and cardiovascular problems.[4] The rate of inner ear complications in RP sufferers is thought to be 40-50%. Auditory loss of sensorineural type may affect one or both sides and the loss may worsen slowly or very rapidly, with resulting cophosis.[5,6] Exactly how the inner ear complications unfold is not currently known. Given that the inner ear does not contain cartilaginous tissue, it is theorised that a different mechanism must be responsible, such as vasculitis of the internal auditory artery, affecting the cochlear or vestibular branches.[2] Hoshino et al [7] examined temporal bone taken from a case of RP in which deafness had rapidly occurred in both ears. They suggest that viral labyrinthitis occurring alongside RP led to cophosis, as the histological appearances showed encapsulation of the tectorial membrane, and severe degeneration of the organ of Corti, as well as of sensory neurons. Miyazawa et al [8] used immunohistochemical techniques to highlight type 2 collagen in

histological sections obtained from three cases of RP in which RP had led to sensorineural type auditory loss. They discovered that degeneration particularly affected type 2 collagen in the tectorial membrane, where it is abundant, and thus, they suggest, degeneration can be linked to auditory dysfunction. Type 2 collagen is found in various structures of the external and middle ear: the pinna, the cartilage supporting the external auditory meatus, the ear drum and the anulus of the ear drum.[9]

Issinget et al [10] performed serology for anti-labyrinthine antibodies, finding these were positive in a case of RP where there was impairment of auditory and vestibular function. Circulating immunoglobulins or T lymphocytes targeting the self may produce an inflammatory response and apoptosis in the inner ear. Schuhknecht [11] observed that the inner ear lacks cartilaginous structures and suggested that a destructive vasculitis of the labyrinthine artery, or branches thereof, was the pathological mechanism. Such an explanation fits with the observation that systemic necrotizing vasculitis can occur in RP.[12] Whilst auditory loss of sensorineural type does affect 40-50% of RP sufferers, the majority of such cases do not develop severe auditory impairment.[13] Usually, the therapeutic interventions used for RP, i.e. nonsteroidal anti-inflammatory drugs, corticosteroid and immunosuppressive agent administration, do partially rectify or protect auditory function in RP cases.[14] But rare cases also occur in which auditory loss secondary to RP is not responsive to the usual therapeutic measures. Implantation of a cochlear device has been found beneficial previously in cases of auto-immune ear disorders, namely systemic lupus erythematosus, Cogan's syndrome, Buerger's disease and related disorders.[15] Cochlear implants work by stimulating the para-cochleal spiral ganglion directly. They are potentially highly effective in cases where cochlear function is impaired. One such scenario would be RP resulting in cophosis. Nevertheless, as may happen when there is ossification of the cochlea for other reasons, such as meningitis, cochlear obliteration may render the implantation of a cochlear device highly challenging. Thus, a high level of clinical vigilance is needed in any case where cochlear obliteration is deemed a risk, so that the device may be implanted prior to the point where cochlear obliteration prevents the siting of electrodes. In the case reported here, good results were still obtainable, despite only partial implementation (five electrodes out of 20 were possible). This outcome is highly encouraging, as the results are superior to other possible options, including an auditory brainstem implant. In clinical practice, whenever possible a cochlear implantation is preferable, even if only partial, to an auditory brainstem implant.

Conclusion

Relapsing polychondritis is a rarely seen disorder, the ae-tiology of which is obscure, but which results in inflam-mation within cartilage throughout the entire body. The cochlea may be obliterated by this pathological process. Cochlear implantation may be needed in such circum-stances. The successful outcome in this case shows a poten-tial role for cochlear implantation in individuals with inner ear disorders produced by immune disorders or relapsing polychondritis.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

Author Contributions: Designing the study – B.A.; Collecting the data – B.A.; Analyzing the data – B.A.; Writing the manuscript – B.A.; Confirming the accuracy of the data and the analyses – B.A.

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