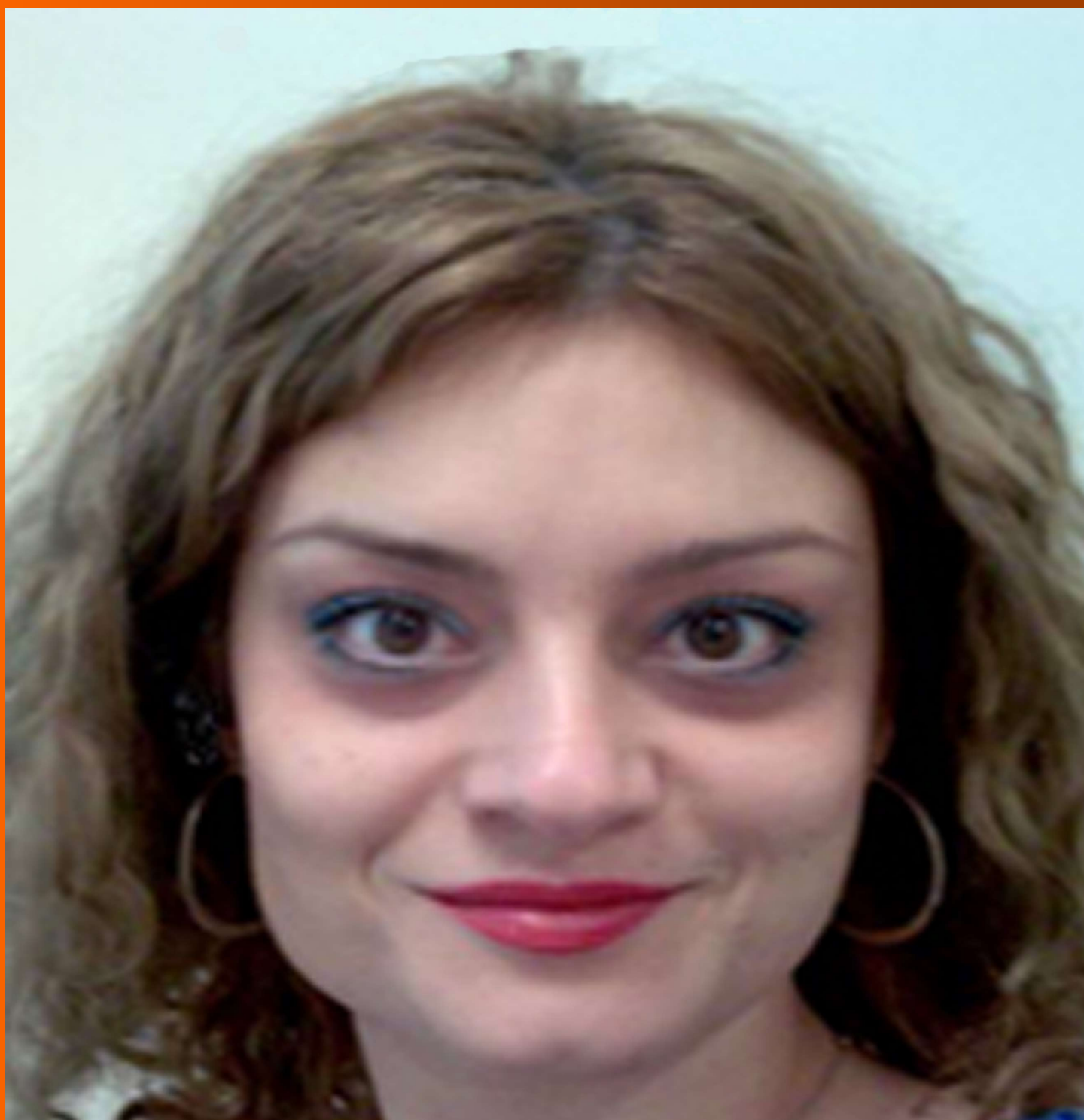


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Room 903, Building D, Ocean International Center,
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Obstructive sleep apnea: An interdisciplinary challenge for otorhinolaryngologists

Haralampos Gouveris

Haralampos Gouveris, Department of Otorhinolaryngology, Medical Centre of the University of Mainz, 55131 Mainz, Germany

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Correspondence to: Haralampos Gouveris, MD, PhD, Department of Otorhinolaryngology, Medical Centre of the University of Mainz, Langenbeckstr.1, 55131 Mainz, Germany. hagouve@yahoo.de

Telephone: +49-6131-177361

Fax: +49-6131-176637

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diagnosis and treatment of SRBD requires a team approach and hence building interdisciplinary teams with other involved relevant specialties is necessary from the patients' perspective.

Key words: Apnea; Surgery; Obstructive; Continuous positive airway pressure; Sleep

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Core tip: Otolaryngologists are quite often the first medical specialists to be contacted by patients with such complaints as snoring, episodic sleep apnea observed by the bed partners with or without reported excessive daytime sleepiness and may therefore emerge as important gatekeepers of the general health of an individual by means of an active preventive, and in many cases therapeutic, role.

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Abstract

Otolaryngologists play a pivotal role in the diagnosis and management of sleep-related breathing disorders (SRBD) in both adults and children. Otolaryngologists are often the first medical specialists to be contacted by patients with complaints as snoring, episodic sleep apnea observed by the bed partners with or without reported excessive daytime sleepiness and therefore emerge as important gatekeepers of the general health of an individual by means of an active preventive, and in many cases therapeutic, role. Comprehensive

INTRODUCTION

Otolaryngologists may play a pivotal role in the diagnosis and management of sleep-related breathing disorders (SRBD) in both adults and children. The importance of such a role can be appreciated by the fact that otolaryngologists are often the first medical specialists to be contacted by patients with complaints as snoring, episodic sleep apnea observed by the bed partners with or without reported excessive daytime sleepiness.

The overall prevalence of severe obstructive sleep

apnea (OSA) may be as high as 38% in patients aged 2 to 18 years who undergo tonsillectomy or adenotonsillectomy^[1]. Obstructive sleep apnea syndrome (OSAS) is a common disease, affecting approximately 2% of women and 4% of men residing in Western countries. A similar prevalence has been found in the general population in some eastern countries^[2,3]. Patients with SRBD require a holistic approach to management involving both diagnosis and therapy.

DIAGNOSTICS

A complete medical history including in particular information on arterial hypertension (with emphasis on hypertensive syndromes refractory to medical therapy)^[4], cardiovascular and cerebrovascular disease, pulmonary disease, diabetes mellitus^[5] and the metabolic syndrome^[6] should be taken. In patients with the metabolic syndrome, the prevalence of moderate to severe OSA is 60%^[6]. Hypertension, age, and obesity have been associated with OSA severity in males, whereas only age and obesity are associated with OSA severity in females^[4]. Nondiabetic patients with OSA are at increased risk of developing insulin resistance and diabetes. Insulin resistance and diabetes are factors independently contributing to increased cardiovascular and cerebrovascular morbidity and mortality^[5]. The prevalence of atherosclerosis, as assessed by coronary artery calcification, carotid intima-media thickness, brachial artery flow-mediated dilation and pulse wave velocity was found higher in OSA patients and correlated with the severity and duration of OSA. Obstructive sleep apnea is therefore an independent predictor of subclinical cardiovascular disease^[7]. Moderate-to-severe OSA is independently associated with an increased risk of all-cause mortality, incident stroke, and cancer incidence and mortality^[8]. On the contrary, in a large cohort, the severity of obstructive sleep apnea was not independently associated with either prevalent or incident cancer. More studies are needed to elucidate whether there is an independent association with specific types of cancer^[9].

Additionally, a thorough history involving social and psychological status is always relevant. Especially the frequent coexistence of insomnia and depression (or related disorders) with SRBD^[10,11] necessitates an evaluation of the respective patients during the first contact visit by means of historical information and clinical assessment as well as by means of the respective standardized specific questionnaires (e.g., the Regensburg Insomnia Scale)^[12]. Such an association is more common in case of obese obstructive sleep apnea syndrome (OSAS) patients with posttraumatic stress disorder or major depressive disorder^[13]. Social and psychological factors are confounders of nutrition in these patients.

Polysomnography (PSG) and structured stand-

ardized questionnaires (such as the Epworth Sleepiness Scale) improve diagnostic accuracy and guide treatment in SRBD^[14]. The impact of the first-night effect on PSG-findings, especially in patients with nose and throat pathology should always be considered^[15]. Drug-induced sleep endoscopy emerges as an important diagnostic method, especially for the selection of candidate patients for implantation with an hypoglossal nerve stimulator^[16]. Notably, in recent reports, OSAS has been definitely associated with increased cancer incidence, especially cancer of the lung^[17,18]. Chronic or intermittent hypoxia (such as the one associated with OSAS) may lead to tumour growth and resistance to radiotherapy. Obesity-associated OSAS may provide a possible mechanism by means of which obesity may promote cancer development. Obesity and intermittent hypoxia increased tumor growth in a mouse model of sleep apnea, but did not exhibit any synergistic effects^[19]. Moreover, increased hypoxia during sleep in OSAS patients has been associated with increased cancer incidence in males and in patients younger than 65 years of age^[20].

THERAPEUTICS

Oral appliance therapy should be seriously considered as an effective treatment alternative to continuous positive airway pressure (CPAP) in patients with mild to moderate OSAS. In patients with severe OSAS, continuous positive airway pressure (CPAP) remains the treatment of first choice^[21]. Definitive treatment of severe and moderate OSAS is CPAP treatment^[22]. Moreover, in patients with cardiovascular disease or multiple cardiovascular risk factors, the treatment of OSAS with CPAP, results in a significant reduction in blood pressure in addition to treatment of OSAS^[23]. Additionally, treatment of OSA with CPAP may lead to improvement in insulin sensitivity, hemoglobin A1c levels, systemic hypertension, and other components of the metabolic syndrome^[5].

Isolated nasal surgery in OSAS patients experiencing daytime nasal obstruction reduces therapeutic CPAP device pressures and increases CPAP use and compliance in select patients^[24,25]. Reduced compliance with CPAP therapy is an important factor that limits CPAP efficacy in both the adult and pediatric OSAS populations. Hypoglossal nerve stimulation emerges as a possible treatment option in OSAS patients who do not tolerate CPAP treatment^[26]. By means of a surgical procedure the stimulation electrode is placed on the hypoglossal nerve, the sensing lead is placed between the internal and external intercostal muscles to detect ventilatory effort and the neurostimulator is implanted in the right ipsilateral mid-infraclavicular region^[26]. Patients with pronounced anatomical abnormalities (such as tonsils visible beyond the pillars or extending to midline) or with complete concentric collapse at

the retropalatal airway during drug-induced sleep endoscopy are not suitable candidates for hypoglossal nerve stimulation^[16,26]. Surgery of the soft palate^[27] and tongue base as well as hyoid suspension^[28], minimally invasive surgery of the inferior turbinates, such as radiofrequency tissue ablation^[29] and surgery of the nasal septum provide satisfactory and definitive treatment in patients with mild or even moderate OSAS^[30].

Adenotonsillectomy remains the treatment of first choice in pediatric OSAS. Nonetheless, many children, especially the obese or those with other underlying medical conditions, have residual OSAS after adenotonsillectomy. CPAP could be an effective treatment modality in these children. Nonetheless, poor adherence and compliance appears to be a significant frequent limitation of CPAP in this pediatric group. Therefore, new treatment modalities for the pediatric OSAS are needed, such as anti-inflammatory substances^[31], treatment by means of an oral appliance, high-flow nasal cannula, and measures to promote weight loss. To date there are few randomized controlled trials assessing the effectiveness of these therapies^[32].

CONCLUSION

Given the enormous systemic impact of SRBD, otolaryngologists emerge as important gatekeepers of the general health status of an individual by means of an active preventive, and in many cases therapeutic, role. Other disciplines involved in diagnosis and therapy of SRBD are internal medicine, neurology, pediatrics and even sleep medicine as own discipline in some countries. Consequently, sleep medicine can be associated with the ENT department and the internal medicine department. Providing comprehensive diagnosis and treatment of SRBD requires a team approach and hence building interdisciplinary teams with other involved relevant specialties is necessary from the patients' perspective.

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Vessel selection and free flap monitoring in head and neck microvascular reconstruction

Ryan M Smith, Vicky Kang, Samer Al-Khudari

Ryan M Smith, Vicky Kang, Samer Al-Khudari, Department of Otorhinolaryngology-Head and Neck Surgery, Rush University Medical Center, Chicago, IL 60612, United States

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Correspondence to: Ryan M Smith, MD, Department of Otorhinolaryngology-Head and Neck Surgery, Rush University Medical Center, 1611 W. Harrison St., Suite 550, Chicago, IL 60612, United States. ryan_m_smith@rush.edu

Telephone: +1-312-9426100

Fax: +1-312-9426653

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Abstract

Microvascular free flap surgery has become a successful and reliable method of reconstruction following head and neck cancer resection. The effectiveness of free flap reconstruction has increased with improved surgical technique as well as technological refinement in vessel selection and flap monitoring. Few papers have studied the factors that influence success or failure rates of free flap reconstructions, particularly with an eye towards the technologic advancements that have refined the procedure in the last several decades. Here we present a comprehensive review of perioperative

and intraoperative considerations that influence free flap outcomes as well methods of vessel selection and flap monitoring important during microvascular reconstruction of the head and neck.

Key words: Neck dissection; Free flap; Microvascular anastomosis; Mandibulectomy; Head and neck cancer

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Core tip: Microvascular free flap reconstruction of the head and neck has become a successful and reliable procedure important in the treatment of head and neck cancer. Careful consideration of the donor site, strategies for vessel selection, and intra- and post-operative flap monitoring protocols and procedures are crucial for successful free tissue transfer. In this review, we present an overview of the latest techniques and technologies proven useful during free flap surgery including strategies for vessel selection, the use of computer aided modeling programs, indocyanine green near-infrared angiography, and ultrasonic transit-time flowmetry.

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INTRODUCTION

The management of tumors of the head and neck pose a significant challenge to the head and neck surgeon owing to the complex anatomy of the region as well as the dynamic physiology of breathing, speaking and swallowing. Reconstruction after

oncological resection with the goal of restoring both form and function must be carefully considered prior to proceeding with surgery. Traditionally, the concept of the reconstructive ladder has provided a framework helpful in determining which reconstructive procedure will best accomplish this goal. As surgical techniques have been perfected, we have seen a shift away from the regional pedicled flaps popular during the 1980's towards more advanced microvascular free tissue transfer procedures that currently exist at the pinnacle of the reconstructive ladder. Free flap surgery has become a reliable method of reconstruction enabling aggressive resection and the use of custom designed vascularized tissue to repair the post surgical defect. The effectiveness of free flap reconstruction has increased with improved surgical technique as well as technological refinement in vessel selection and flap monitoring. Studies have shown that patients who received free flap reconstructions had shorter hospital stays and intensive care unit admissions than those who received pedicled flap reconstructions. The benefits of reduced hospital stays are reflected in both reduced cost and complication rates^[1,2]. Free flap success rates exceed 90% in many large clinical series, with complication rates reported at around 5%^[1,3-5]. Few papers have studied the factors that influence success or failure rates of free flap reconstructions, particularly with an eye towards the technologic advancements that have refined the procedure in the last several decades. Here we present a comprehensive review of perioperative and intraoperative considerations that influence free flap outcomes as well methods of vessel selection and flap monitoring important during microvascular reconstruction of the head and neck.

PRE-OPERATIVE EVALUATION

Free tissue transfers are time-consuming and technically challenging, but innovative preoperative planning modalities have decreased both operative time and costs, while maintaining the reliability and quality of the postoperative outcomes. In cases of radial forearm free flaps, the Allen's test is always performed preoperatively to assess whether the radial artery is a suitable donor vessel; however, additional examination is recommended to ensure the successful perfusion of the anastomoses.

Imaging and modeling

Preoperative CT and MRI are essential for the diagnosis of head and neck cancer and indispensable while planning surgical extirpation of tumor. Computer modeling programs are now being used in combination with traditional imaging modalities to plan reconstruction. The clinical utility of combining preoperative CT with computer-guided planning programs and the use of osteotomy guides, stereoli-

thographic modeling and pre-fabricated reconstruction plates has been demonstrated in mandibulectomy with free fibula flaps. Preoperative computerized planning has eliminated the need to estimate the shape of the fibula to create the neo-mandible as this technology enables more precise and accurate bony alignment during fibula free flap reconstruction following segmental mandibulectomy. Surgeons can better define tumor resection margins and plate mandibles with exophytic lesions that make estimation difficult^[6]. A systematic review of virtual surgical planning (VSP) with computer aided design (CAD) and computer aided modeling (CAM) for oncologic reconstructions also reported a number of qualitative benefits, including reduced operative time, flap ischemic time, complication rate, and allowed for more precise resection margins and improved accuracy of reconstruction as well as predictability of outcomes and patient satisfaction scores^[7]. These new technologies eliminate the need to spend valuable ischemic time designing and modifying the neomandible prior to inset. Additionally, VSP simulates the maxillo-mandibular relationship preoperatively to achieve better occlusion, alignment and orthognathic relationship of the reconstructed mandible. The reduced costs associated with decreased operative time and complications offset the additional technological costs for the preoperative planning^[7].

Patient characteristics

Many studies have assessed the impact of intrinsic patient factors on postoperative outcomes like flap survival. Factors including American Society of Anesthesiologists (ASA) grade ≥ 2 (95%CI: 15.3-18.1, $P < 0.009$), smoking history (95%CI: 5.5-7.2, $P < 0.049$), body mass index ≥ 25 kg/m² (95%CI: 20.8-22.1, $P < 0.003$) and peripheral vascular disease (95%CI: 5.9-7.5, $P < 0.036$) were independently predictive of free flap complications^[1]. Several investigators have studied the impact of patient characteristics such as age and BMI^[8], tobacco and/or alcohol consumption, the use of preoperative radiotherapy^[9], type of anticoagulation therapy^[4] and the incidence of free flap failures and have found no significant associations. It is well known that a history of diabetes mellitus significantly increases the postoperative complication rate, yet patients with diabetes are not contraindicated for free flap reconstructions. They do however require a more thorough preoperative assessment and more vigilant postoperative monitoring for flap failure^[10]. The microangiopathic consequences of diabetes on the skin and muscle leads to disrupted intimal repair and an increased risk of flap compromise^[11]. Female patients were found to have higher rates of flap failure and compromise than male patients in a few studies, but this finding was not replicated in other studies and the mechanism behind this

finding remains unclear^[9,12,13]. Thrombophilia and a preoperative platelet count of greater than 300 were found to be associated with lower rates of successful salvage when free flap exploration became necessary^[13].

An important consideration must be made in patients who have undergone previous neck dissection or have received previous radiation to the head or neck due to a potentially reduced availability of recipient vessels for free flap reconstruction^[14,15]. Pedicled flap reconstructions should be considered for high-risk patients, including those who suffer from severe co-morbidities or elderly patients. Using alternative pedicled flaps instead of free flaps in these patients reduces the risk of flap failure and other post-operative complications^[16]. A meticulous review of previous operative reports is necessary to determine the status of preserved vessels of the internal and external jugular venous systems. The external carotid artery and its medial branches are commonly preserved and available to use, however the facial artery and superior thyroid artery are frequently ligated during neck dissection and therefore may be unavailable. Contralateral recipient vessels may be needed in 50% of patients who have undergone previous neck dissection. Selection of a flap that provides a long vascular pedicle would be desirable in this case and the need for vein grafts can be avoided if careful preoperative consideration is given to the post-dissection anatomy of the patient^[17,18].

Irradiated tissue is associated with endothelial dysfunction, including degeneration and necrosis of the endothelium, a thickened tunica media that narrows the vessel lumen, and prothrombotic processes^[19]. Previous studies have found a higher rate of flap loss in patients with previous radiation and/or neck dissection; however, the difference was not statistically significant^[16]. Hanasono *et al.*^[20] reported significantly higher complication rates in patients with a history of both neck dissection and radiation than patients who did not have a history of either. Unfortunately, due to the often aggressive behavior of head and neck cancer, a single patient may require multimodality treatment including free flap reconstruction in less than the ideal setting of a non-irradiated and un-dissected field. Therefore, these pre-operative patient factors should be considered prior to free flap reconstruction but should not hinder plans for definitive treatment.

INTRA-OPERATIVE CONSIDERATIONS

Intraoperative technique is one of the most important factors influencing free flap survival. In particular, careful planning during vessel selection and intraoperative monitoring of vascular perfusion can optimize the post-operative outcome and minimize surgical complications.

Vessel selection

Classic considerations for recipient vessel selection include the vessel's viability, length and location. The vessel must be able to be exposed atraumatically and allow a sufficient length for the positioning of the anastomosis. Strong pulsatile flow before and after the division indicates good vessel quality and perfusion. In patients with previous head and neck dissection or radiation therapy, suitable recipient vessels should be sought in locations outside the zone of radiation^[19].

The most frequently used and readily available recipient arteries include the facial, lingual, superficial temporal, superior thyroid, and transverse cervical arteries^[18]. Arterial anatomy is more consistent than venous anatomy; however, there are variations in the branching patterns of the facial, lingual and superior thyroid arteries. The most common pattern has the three vessels anteriorly directed and branching individually from the external carotid artery, but sometimes the facial and lingual arteries may have a lower or higher take-off from the common linguofacial trunk off the external carotid artery. The high take-off form of the linguofacial trunk is more susceptible to ligation during the neck dissection and therefore may not be amenable to use^[21].

Past reviews have created algorithms for the selection of recipient vessels, taking into consideration the defect location, operative history, type of anastomosis, and characterizations of frequently used arterial and venous systems^[18,22]. The superficial temporal artery is the first choice for a defect in the upper third of the head. This vessel has little anatomic variation and is usually the smallest of all the external carotid artery branches. It is located within 5 cm of the oral cavity but can also be useful for lower head and neck defects in patients with previous neck dissection and/or irradiation compromising more inferiorly located vessels^[23].

The facial artery, located 1 cm below the border of the mandible, is the first choice vessel for defects in the mid or lower third of the head in patients without a history of neck dissection or radiation; however, care must be taken to protect the marginal mandibular nerve branch as it crosses superficially over the facial artery. The facial artery is particularly useful when mandibulectomy is required because the osteotomy enables easier access for the microvascular anastomosis.

The superior thyroid artery is the first choice vessel for lower face defects with a concurrent neck dissection. The superior thyroid artery runs in the carotid sheath caudally towards the thyroid gland, affording it protection from the resection. The superior thyroid artery should be mobilized to a sufficient extent to allow its reach without kinking or excessive stretching.

The algorithm for recipient vein selection is slightly different from that of the artery as the venous

anatomy is more variable with unpredictable interconnections of adjacent vessels. The first choice recipient vein is the corresponding vein of the selected artery. Vessels are chosen from the internal or external jugular venous system and selecting a recipient vein closest to the recipient artery can decrease kinking and stretching of the vessels.

The internal jugular vein (IJV) and its branches are more accessible than the external jugular vein (EJV). Additionally, the negative intrathoracic pressure generated with respiration increases the blood flow of the IJV. However, the IJV has a rate of thrombosis between 14%-33% and may be ligated during the neck dissection. Some reports have suggested large branches of the IJV in close proximity to the selected artery are the better and more frequently used recipient veins. The EJV is considered when all branches of the IJV are unavailable or unsuitable because the external jugular vein (EJV) is more superficial and vulnerable to compression and ligation^[23]. The cephalic, thoracodorsal, and internal mammary veins can be considered as alternative options if both the internal and external jugular venous systems are unavailable. Vein grafts should be avoided and only considered as a last resort because their use requires an extra anastomosis, which adds to the technical difficulty of the case.

Anastomosis considerations

The configuration of the anastomosis of the vascular pedicle linking the recipient to the donor vessel is another important factor to consider during free flap reconstruction. Some reviews recommend an end-to-end (ETE) over the end-to-side (ETS) technique for the arterial anastomosis. It is argued that ETE anastomoses are safer and more reliable because they avoid directly manipulating the carotid artery, a vessel that houses more turbulent flow and is vulnerable to plaque deposition and atherosclerosis. Manipulation during ETS anastomosis risks intimal separation of the atherosclerotic plaques^[22]. Additionally, ETS anastomosis is associated with a higher inflow into the flap, increasing the risk of venous congestion, and ETS positioning also predisposes the anastomosis to kinking and vessel occlusion. ETE anastomosis leaves the vessels in a relatively mobile and flexible position, decreasing the risk of compression and kinking with intra- and post-operative changes. ETS technique is used if there is a significant difference in the diameter of the vessel lumens, indicated as a size discrepancy greater than a 2:1 ratio^[18]. If they are to be used, ETS anastomoses have been suggested as a more suitable technique for the venous anastomosis because veins are less likely to be atherosclerotic. However, a large cohort study comparing ETE and ETS anastomosis found no significant differences in the rates of flap failure or need for re-exploration^[24]. Advocates of ETS

anastomoses claim benefits such as using vessels of different sizes and the ability to create multiple anastomoses using a single recipient vessel. There is still a lack of consensus on which technique is superior; it is suggested that reconstructive surgeons should select which anastomotic configuration to use based on the characteristics of each case, including patient anatomy, type of defect and selected donor site, and the preference and experience of the surgeon.

The geometry and positioning of the vascular pedicle must also be considered in free tissue transfers. Kinks caused by poor positioning or redundancy of the vessel can cause arterial occlusion, insufficiency and flap loss. Vessel positioning is a more significant factor in head and neck free flaps than in extremity free flaps, because it is not possible to completely immobilize the head and neck with the need for tracheostomy care. Aligning the vascular pedicle on a longitudinal axis can reduce complications associated with vessel positioning, because side to side movements of the head and neck will less likely kink or create tension on the pedicle than if the vessels were aligned on a horizontal or oblique axis^[25].

The gold standard for achieving anastomosis in microvascular free tissue transfers is by hand-sewn techniques using nylon sutures; however, mechanical anastomotic coupler devices (MACDs) have recently become an effective alternative to the hand-sewn anastomosis. Hand-sewn anastomosis is both time-consuming and technically challenging, including poor vessel edge eversion that exposes thrombotic material in the lumen and poor suture placement resulting in anastomotic leaks^[26]. The mechanical microvascular coupler can decrease flap anastomosis time^[27], flap ischemia time and reduce complications and risk of free flap loss. Coupler sizes range from 1.5 to 4.0 millimeters and the size of the couplers do not affect tissue transfer outcomes. Limitations of couplers include the inability to achieve anastomosis in vessels that are not pliable or thickened and the potential risk of late exposure^[28]. Mechanical venous anastomosis has proven to be a reliable method of anastomosis in head and neck reconstructions; however, the effectiveness of microvascular couplers for arterial anastomosis is still under debate and some studies have shown poor flap outcomes.

Vessel monitoring

Intraoperative monitoring of candidate vessels and the vascular anastomosis is crucial for successful flap perfusion. The diameter of the vessel is directly related to the flow of the vessel and inversely related to vessel resistance, thus well-perfused flaps use the vessels with the largest diameter and the highest flow^[29]. Vessels with a diameter of less than 1 mm are associated with a higher risk of free flap failure^[30]. Surgeons have traditionally evaluated

recipient vessels as healthy and viable when there is a brisk and pulsatile flow on release of the vascular clamp after vessel transection, but recent technologies have enabled more objective and reproducible intraoperative assessments of vascular perfusion.

The implantable Doppler is an invasive monitoring technique that collects qualitative data on blood velocity to detect vascular compromise both intra- and post-operatively. A silicone cuff attached to an ultrasonic probe is secured around the vessel with a suture or clip and can be kept in the neck for postoperative monitoring. Literature reports that the probe should be placed on the venous pedicle, as arterial compromise can be detected through venous changes within minutes. A perfect fit of the cuff around the vessel is important for an accurate measurement, as the vessel may slip out of the cuff resulting in a false positive signal loss and an unnecessary intervention. Conversely, if the cuff is too tight, it may obstruct flow through the vessel^[31]. The implantable Doppler is effective in detecting intraoperative vascular compromise, and in recent studies, patients with intraoperative Doppler monitoring had a significantly higher success rate (95%) compared to patients who were evaluated by clinical signs of compromise alone (40%)^[32].

Postoperative monitoring of vascular perfusion using the implantable Doppler is more unreliable than intraoperative monitoring. Postoperative positioning and maneuvering of the patient's head and neck may result in a lost Doppler signal leading to false positives and unwarranted re-explorations that increase both costs and risks of morbidity^[9]. The inaccuracy of the Doppler technology has also been reported, and measurements can be skewed by the insonation angle and vessel wall thickness and diameter^[9,33,34]. Literature suggests that the implantable Doppler should be used as an adjunct method to monitor vascular perfusion. For instance, the combined use of the clinical exam and the implantable Doppler is a safer and more reliable method^[32,35].

Indocyanine green near-infrared angiography (ICGA), with a sensitivity of 100% and specificity of 86%^[36], is another validated method for intraoperative and postoperative assessment of vessel and anastomosis quality. Intravenous administration of the ICG dye can visualize the arterial or venous network and reveal any stenosis of the vasculature. It is especially effective for detecting microthrombosis that may cause partial insufficiency of the circulation. ICGA also provides detailed angiographic information that enables a more targeted approach to resolve a vascular compromise. For instance, re-exploration surgery that would require taking down the anastomosis is avoided when ICGA reveals an intact inflow and outflow, allowing the surgeon to expedite and avoid a misdiagnosis^[36]. Due to the invasive

nature of the ICGA, its application is only suggested for high-risk and complicated cases like patients with diabetes or prior irradiation^[37,38].

Color Doppler Sonography (CDS) is used both intraoperatively and postoperatively to monitor for vascular compromise. The non-invasive probe is applied to the vascular pedicle and changes in hemodynamics are displayed through color Doppler waveforms, enabling calculations of flow velocity and a pulsatility index (PI). The flow velocity is unreliable and highly dependent on the angle between the probe and the vessel, whereas the PI, calculated by subtracting the minimum flow rate from the maximum flow rate divided by the mean flow rate is a more reliable measurement of the vascular resistance^[39]. Although CDS is unable to record continuously and its use in the operative or exam room may be limited by the large size of the system, it signals any increase in vascular resistance resulting from vessel compression or thrombus occlusion before changes are apparent through clinical signs. Studies have also reported its ability to facilitate preoperative planning of the reconstruction by assessing the vascular patency and flow of the donor site^[40].

Ultrasonic transit-time flow meters record precise and reproducible intraoperative transit-time flow measurements (TTFM) using a non-invasive probe that transmits ultrasonic beams^[41]. It is easy and safe to use^[42] and accurately quantifies blood flow of recipient arteries and flap pedicles. Transit time flow measurements are not confounded by the angle of the ultrasonic beam to the vessel area, as is the Doppler. It can accurately diagnose graft occlusions^[41,43-45], and Lundell *et al*^[46] found that patients with vascular occlusions had significantly lower transit-time flow measurements than patients with patent vascular pedicles; this finding has been replicated in recent diagnostic angiography studies^[47]. A study evaluating postoperative graft patency in coronary surgery found that intraoperative TTFM successfully predicted grafts found to have a higher incidence of failure within the first postoperative year^[43].

POST OPERATIVE EVALUATION

Total flap loss rates are low, reported at a range of 0.6% to 6%^[10], but rates of surgical complications after free flap reconstruction of the head and neck are significantly higher. The impact of complications on the overall outcome of free tissue transfers cannot be underestimated; events such as venous and arterial thrombosis, wound infection, leakage, flap dehiscence, fistula formation and partial flap necrosis commonly require surgical re-exploration and can ultimately lead to partial or total flap failure. Studies report that vascular compromise is the most common reason for flap re-exploration,

representing 76.8% of compromised flaps^[48]. Common causes of vascular compromise include arterial and venous insufficiency, anastomotic bleeding and hematoma. Venous insufficiency was the most common reason for flap re-exploration; the low pressure of the venous system is susceptible to compression from improper positioning of artery over vein, pedicle tension, hemodynamic compromise, and neck flexion^[8,48]. Arterial insufficiency, frequently from small thrombi that occlude perforators, was associated with the highest percentage (49.3%) of unsalvageable flaps^[48]. Currently, the only evidence-based strategy for optimizing free flap salvage is an early intervention. The rate of successful flap salvage is inversely related to the time duration between the onset of flap ischemia and recognition of the failing free flap, and the rate of salvage declines as duration of this time period increases^[9,14,49]. An unrecognized venous thrombosis can progress to an arterial thrombosis and ultimately result in a flap failure. Additionally, if re-exploration is delayed and circulation is not restored within 8 to 12 h, the no-reflow phenomenon^[50] and reperfusion injury^[51] will develop when flow is finally restored, leading to a flap that is impossible to salvage. Due to the importance of early recognition, post-operative flap monitoring protocols are extremely important for successful free tissue transfer.

Post operative monitoring

Vigilant postoperative monitoring, including both clinical and instrumental observation, is vital for ensuring success after free flap reconstruction. The most common cause of late flap failure, indicated as occurring 7 d postoperatively, is an unrecognized failure of a buried flap^[9].

Post-operative vascular perfusion monitoring is classically assessed with clinical observation, including the color, temperature, swelling and capillary refill of the flap. The pinprick test or placement of surface temperature probes may also be used to monitor flaps with a skin paddle^[4,9,24,52]. A needle used to prick the flap and cause bleeding will indicate a viable and perfused flap if the flow appears bright red but may suggest a failing flap if dark red blood is observed^[52]. Clinical assessments are highly subjective and dependent on the observer's experience and skill; on the other hand, innovative monitoring systems can deliver reliable and objective measurements of flap perfusion.

The hand-held Doppler is commonly used for postoperative flap monitoring. The skin site corresponding to the area of the anastomosis can be marked with a suture in order to provide an accessible and reliable point at which the Doppler probe may be placed. Compression of a vein will result in an augmented venous signal, while compression of an artery will lead to a loss of arterial signal^[24], making use of the Doppler operator dependent.

Tissue oxygenation measurements (TOx) using non-invasive near-infrared spectrophotometry (NIRS) is used postoperatively to detect circulatory compromise before it is noticeable through clinical assessment. Although, it is most commonly used for postoperative monitoring for flap compromise, studies have acknowledged the potential of TOx to be used intraoperatively for characterizing recipient vessels and measuring pre- and post-anastomosis flap perfusion. TOx is calculated by measuring the reflected near-infrared light emitted by the probe that is attached and secured intraoperatively over the skin paddle. The computer uses spectrophotometric principles to calculate the percentage of oxygenated hemoglobin to evaluate tissue oxygen saturation (StO₂), which is closely correlated with vascular perfusion. Keller reported diagnostic algorithms to predict vascular complications; a drop rate indicator ($\Delta\text{StO}_2/\text{time}$) equal to or greater than 20% per hour sustained for 30 min or longer is indicative of vascular complications and a StO₂ of less than or equal to 30% is indicative of occluded blood flow by a venous or arterial thrombosis.

Studies have examined both systemic and surgical factors that may confound the TOx measurements and concluded that only the patient's hemodynamic status, determined by his or her blood SO₂ (pulse oximetry), had a significant effect on the TOx measurements. A 1% increase in SO₂ correlated with an average 0.36% increase in TOx^[53]; however, only a truly significant drop in SO₂ would have a significant skewing effect on the TOx. Use of tissue oximetry significantly improves rates of flap salvage (57.7% to 93.75%), reduces rates of total flap loss (2.9% to 0.43%), and provides continuous monitoring of flap perfusion with numeric data^[49]. Limitations of TOx include its requirement for a cutaneous skin paddle, rendering it ineffective for buried or wet flaps, and the high costs associated with the probes and monitors.

Computed tomography angiography (CTA) is another non-invasive method to monitor for post-operative vascular stenosis. CTA is a rapid exam using iodinated contrast and ionizing radiation to visualize the course of the flap vessels with high spatial resolution; it is able to detect a failing flap and grade varying degrees of vascular stenosis at the site of the anastomosis^[54]. Preoperative CTA imaging of flap vasculature have also been demonstrated to optimize the surgical planning process and allow surgeons to create precise skin paddle placements and avoid potentially poor vascular perfusion. Preoperative CTA visualizes hypoplastic or diseased arteries, allowing surgeons to modify operative plans, utilize alternative sites with verified vascular sufficiency, and take more direct courses for vessel exposure^[55].

Contrast enhanced ultrasound (CEUS) is an emerging technology that may be useful in the post-operative monitoring of free flap perfusion. This technology involves the application of ultrasound contrast medium to traditional medical sonography

and has been used for the detection of focal malignant liver lesions and the measure of cardiac blood flow during echocardiography^[56]. More recent literature suggests an application in free flap reconstructive surgery as well. Prantl *et al.*^[57] investigated 15 patients with history of lower limb orthopedic trauma who underwent parascapular free flap reconstruction and used CEUS to assess the flow and patency of the small vessels of the flap. The authors report CEUS to be a sensitive technique allowing detection of the flow and patency of the microvascular anastomoses as well as the perfusion within the microcirculation of the free flap itself. Lamby *et al.*^[58] used CEUS in the immediate post-operative period and were able to detect partial flap necrosis as well as post-operative hematoma in 2 of the 10 patients studied, concluding that this technology can provide an optimal assessment of perfusion in multiple tissue layers of the free flap and may help prevent flap loss.

Microdialysis is another technique that can be used for post-operative monitoring after free flap reconstruction. This technology involves an analysis of the metabolic by-products present within free flap tissue as a means of detecting ischemia and allowing early surgical intervention to prevent flap loss. A microdialysis probe, with a dialysis membrane at the end, is introduced into the free flap tissue. Perfusion fluid is then instilled into the catheter and allowed to equilibrate across the membrane with the extracellular fluid of the free flap tissue. Dialysate is then collected and analyzed and the concentrations of glucose, glycerol and lactate are compared to those of a control dialysate collected from a similar microdialysis probe inserted at a distal site into native tissue. During flap ischemia, the concentration of lactate and glycerol will increase and that of glucose will decrease. Udesen *et al.*^[59] used microdialysis for post-operative monitoring of 14 female patients with free TRAM flaps. In one patient with arterial anastomotic thrombosis, the glucose level fell to an immeasurable level while the glycerol and lactate concentrations increased. The difference in concentrations between the flap and control was statistically significant and after reperfusion, the concentrations returned to normal levels. Although microdialysis is an invasive procedure with the inherent risk of damage to the vasculature of the free flap, it represents a potentially useful method of detecting early tissue ischemia.

CONCLUSION

Microvascular reconstruction of the head and neck using free tissue transfer has undergone considerable technical refinement in the recent decades, making it a reliable and successful method of reconstruction following head and neck cancer surgery. Due to the complex nature of the procedure, numerous factors combine to determine the success rate following

free flap surgery. In this review article, we have presented a comprehensive review of perioperative and intraoperative considerations that influence free flap outcomes as well methods of vessel selection and flap monitoring important during microvascular reconstruction of the head and neck.

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Exostoses of the external auditory canal

David R Lobo

David R Lobo, Department of Otolaryngology, Hospital El Escorial, San Lorenzo de El Escorial, 28200 Madrid, Spain
David R Lobo, Instituto Antoli-Candela, 28043 Madrid, Spain
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Correspondence to: David R Lobo, MD, PhD, Department of Otolaryngology, Hospital El Escorial, San Lorenzo de El Escorial, Ctra. M-600 de Guadarrama a San Lorenzo de El Escorial, Km. 6,255, 28200 Madrid, Spain. dlobo28@gmail.com
Telephone: +34-91-8973021

Fax: +34-91-8973031

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review offers an overview of the recent advances in the understanding of this condition, with a special focus on the etiology and physiopathology of this condition, the different surgical procedures and their outcomes, the risk factors for recurrence and the results of preventive measures. Finally, this review suggests the need for the otological surgeon to acquire a great deal of experience before undertaking surgical treatment of exostoses as it is a challenging operation and, besides expertise, demands great patience and extreme care in order to achieve good results.

Key words: Exostoses; Surfer's ear; Ear canal; Surgical procedures; Recurrence; Prevention

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Core tip: Readers interested in aquatic sports medicine, otological procedures or bone lesions will find in this review a thorough summary of the most relevant aspects of ear canal exostoses, with a special focus on the major advances achieved in the knowledge of its etiology and pathophysiology, prevention and surgical therapy, and the challenges that remain and may guide research in the next few years and beyond, so that complications are minimized and the best outcomes are achieved.

Abstract

Ear canal exostoses are bilateral, usually symmetric multiple bony growths occurring in the medial portion of the external auditory canal. Also known as surfer's ear, exostosis is thought to be a reactive process from repeated stimulation by cold water and is much more common than external auditory osteoma. Exostoses are usually asymptomatic and discovered on routine otoscopy. Indications for surgical treatment are recurrent otitis externa, hearing loss, otalgia and other conditions in which access medial to the exostoses is required. Surgery is not risk-free and postoperative complications are the most important factor for negative impact on the patient's health-related quality of life. This

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INTRODUCTION

Ear canal exostoses, also known as surfer's ear, are bilateral, usually symmetric multiple bony growths occurring in the medial portion of the external auditory canal, just lateral to the tympanic membrane

and medial to the sutures of the tympanic bone. Clinically, they are easily differentiated from the similar but less common external auditory osteoma, which is a unilateral, pedunculated lesion occurring mostly in the outer half of the ear canal, arising from the tympanosquamous or tympanomastoid suture lines. Histopathologically, however, there is no consensus regarding differentiation of exostoses from osteomas^[1,2]. Histologic examination of exostoses reveals remodeled densely lamellar bone tissue superficially, running parallel to the periosteum, suggesting continued periodic apposition and deep to this, loosely compact and more vascular bone, less organized around fibrovascular channels^[3].

Exostoses are usually asymptomatic and discovered on routine otoscopy. They have been described as a functional physiological organ in aquatic mammals like hooded seals^[4]. Their prevalence in humans has been well studied among surfing populations as well as among people involved in other water activities such as kayaking, diving, swimming or sailing^[5-12]. The prevalence of exostoses in coastal inhabitants is much higher than in valley or mountain inhabitants, with men much more affected than women^[8]. This susceptibility among coastal populations or those engaged in water activities has also been studied in different ancient or prehistoric populations^[13-17]. Although most cases do not present to medical care, when the stenosis caused by exostosis exceeds a certain limit, defined as greater than 80%, the patient may develop conductive hearing loss due to occlusion and other symptoms, usually recurrent wax retention, recurrent otitis externa with otalgia or tinnitus^[18]. When these symptoms become bothersome and cannot be managed with medical treatment, surgical excision of exostosis is warranted. Surgery may also be necessary as an access path for treating other middle ear conditions such as otosclerosis or chronic suppurative otitis media.

ETIOLOGY

Exostoses build up in various locations in the body as new subperiosteal bone is formed as a response to increased tension on the periosteum that induces osteoblastic activity. Prolonged reflex vasodilatation occurring in the bony meatus following exposure to cold water or cold winds offers such an increase in tension, for there is no insulating layer of subcutaneous tissue between epithelium and underlying periosteum in the deep meatus. Moreover, venous congestion promotes lamellated new bone production as seen microscopically in exostoses^[19].

Early theories proposed some form of prolonged irritation to explain the origin of exostoses. For instance, chronic otitis externa, gout, syphilis, subperiosteal abscess or in individuals who routinely use stethoscopes^[19-22].

However, growing evidence suggests that prolonged

exposure to cold water, as seen most frequently in aquatic sportsmen, is the most important etiopathogenic factor^[23-26]. Cold air may also be a contributing factor in the formation of exostoses. This is supported by a series of findings. First, the skeletal remains of coastal populations from areas with mild atmospheric temperatures and wind chill factor do not show high frequencies of ear canal exostoses^[14]. In addition, the most affected water-sport athletes reported in a study practiced sailing, which exposes the subjects to continuous cold jets, and according to another study the prevalence of exostoses was higher among the group of patients who practiced head-above-surface aquatic sports such as surfing and sailing (and therefore were more exposed to wind chill factor) than among the group of patients who practiced head-immersed aquatic sports such as swimming or diving^[12,27]. Interestingly, the prevalence and severity of exostoses in breath-hold divers, who stay longer above surface, is more similar to previously published results for surfing populations than to those for scuba-diving populations^[28]. Finally, the severity of exostoses seems to correspond to the ear that is more exposed to the predominant coastal winds^[29].

Regular exposure to low air temperatures and/or cold winds could explain the development of exostoses in non-aquatic sport enthusiasts or people not engaged in long-term cold water activities.

Genetic factors may also explain a higher predisposition to aural exostoses as these have not been reported in black people^[30]. There might be other factors as well involved in the origin of exostoses. Half of the population did not report exposure to cold water in one study^[31]. Nevertheless, most studies have focused on populations engaged in water activities for recreational, occupational or military reasons.

DIAGNOSIS

Exostoses are discovered on routine otoscopy. Computed tomography scans are rarely performed, usually before the surgery to evaluate the extension of exostoses, but generally they are not believed to be as helpful as direct microscopic transcanal visualization of ear canal anatomy.

For ear canals that are less than obliterative, the degree of stenosis, the percentage of closure, can be estimated. This can be achieved either by outlining the approximate lumen of the ear canals in photographs taken at the start and conclusion of surgery and then measuring the respective areas using a computer program, or by taking photographs using an endoscope connected to a camera, with each print being assessed by three independent observers^[19,27]. Unfortunately, in most publications authors do not report how the percentage of closure is evaluated.

Audiologic testing is mandatory, as conductive

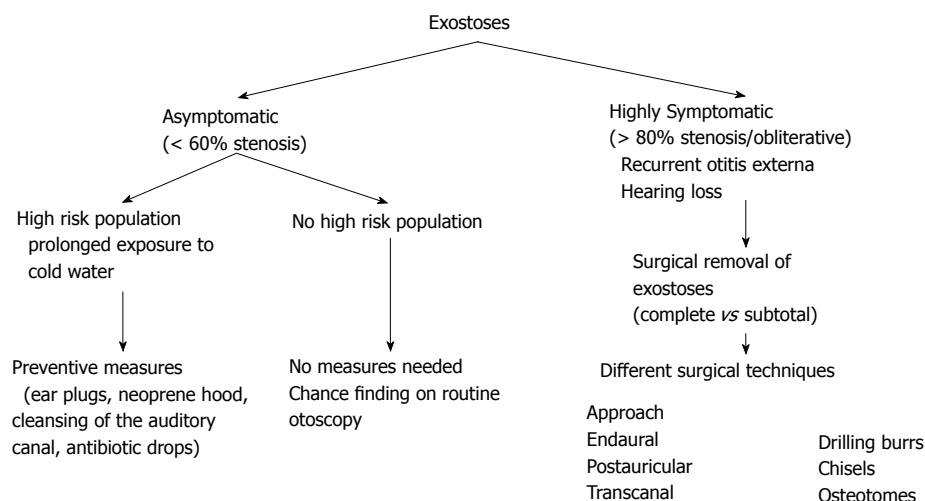


Figure 1 Proposed algorithm scheme for the management of exostoses and surgical options. Some studies report surgical removal of exostoses in asymptomatic patients, although this approach remains controversial and is not supported by most authors.

hearing loss is the second most frequent indication for surgery after repetitive otitis externa.

TREATMENT

Most patients with exostoses are asymptomatic. When symptoms make their first appearance, these are due to wax accumulation in the external auditory canal, and therefore the most frequent symptom is ear fullness, which can lead to otitis externa and ultimately to conductive hearing loss if all these conditions do not improve. In these cases, the initial treatment is the clearance of the ear canal to prevent wax retention and hearing loss. Antibiotic and anti-inflammatory drops should be prescribed in the case of otitis externa. Surgical intervention should be considered only in the most severe cases with obliterative exostosis, when cleansing of the auditory canal is no longer possible, when otitis externa becomes very recurrent or when there is conductive hearing loss. This usually occurs when the degree of stenosis is more than 80%, and hardly ever when it is less than 60%^[18].

Although most authors agree that exostoses should be operated on only when the patient is symptomatic or when the exostoses prevent medial access in the ear canal for other procedures, some studies report surgical removal of exostoses in asymptomatic patients, when the degree of stenosis was assessed as minimal (< 30%) or moderate (30%-60%) or to facilitate the fitting of a hearing aid^[27,32]. However, surgery has a considerable complication rate and cannot achieve excellent symptom control in all cases. This surgical procedure demands considerable patience and a variety of precautions, is difficult and tedious, and the outcome is uncertain due to a number of factors such as the proximity of the exostoses to the tympanic membrane, temporomandibular joint and facial nerve, the narrow or non-existent space between the exostoses, the inability to visualize medial ear canal

landmarks, the thin skin covering the exostoses, and the ease with which modest bleeding can obscure the surgeon's view^[19]. Moreover, the patient's desire to return to aquatic sports as rapidly as possible after surgery may jeopardize the results of surgery and lead to complications or early recurrence. Thus, correct patient selection and indication for surgery is paramount. An algorithm for the surgical options and management of exostoses is provided in Figure 1.

The most frequently employed surgical techniques involve the postauricular or endaural approaches and the use of different drilling systems and drilling burrs. Transcanal approaches and the use of osteotomes, chisels, curettes and gouges have also been employed^[18,19,29]. The beaver knife is used to incise the posterior meatal skin, forming a flap which is held anteriorly. This flap is usually protected by a small piece of aluminium harvested from the packing of surgical threads^[32,33].

Most authors recommend complete removal of exostoses to prevent early recurrence. However, several authors have advocated subtotal removal of exostoses so that patients are not placed at risk of injury to the ossicles, facial nerve or chorda tympani nerve^[34,35]. Although facial nerve monitoring is not generally used in these procedures, the surgeon must be well aware of the course of facial nerve in the posterior inferior medial bony canal wall. In an anatomical study of the temporal bone, the facial nerve was located in a plane lateral to the tympanic membrane in the posterior-inferior quadrant of the medial bony ear canal in 70% of specimens^[36]. In cases of severe narrowing, facial nerve protection becomes a priority. This is achieved by restricting blind drilling of the posterior canal wall while the tympanic membrane is not visible^[32].

When the stenosis is severe the procedure can be very demanding, especially given the absence of adequate landmarks. This is why some authors prefer the retroauricular approach for all but the mildest narrowing^[32,33]. The retroauricular approach

Table 1 Different approaches and surgical techniques

Ref.	Year	Endaural (operations)	Postauricular (operations)	Transcanal (operations)	Drilling (operations)	Osteotomes, chisels ¹
Altuna Mariezkurrena <i>et al</i> ^[33]	2004		× (52)		×	
DiBartolomeo <i>et al</i> ^[30]	1979			× (21)	× (21)	
Bordure <i>et al</i> ^[37]	1994	×	×		× (64)	
Frese <i>et al</i> ^[38]	1999	× (56)	× (3)		× (59)	
Hempel <i>et al</i> ^[31]	2012	× (33)	× (2)		NA	
Hetzler ^[19]	2007			× (221)	× (42) ²	× (179)
Hurst <i>et al</i> ^[39]	2001	× (58)	× (6)		×	
King <i>et al</i> ^[29]	2010		× (80)		× (71) ³	× (4)
Oostvogel <i>et al</i> ^[40]	1992	×	×		×	
Portmann <i>et al</i> ^[41]	1991		× (25)		× (25)	
Sanna <i>et al</i> ^[32]	2004		× (65)		× (65)	
Stougaard <i>et al</i> ^[35]	1999	× (7)	× (1)	× (16)	× (24)	
Timofeev <i>et al</i> ^[27]	2004	NA	NA	NA	× (46)	
Whitaker <i>et al</i> ^[18]	1998			× (27)		× (27)

¹Hetzler^[19] used osteotomes whereas King *et al*^[29] and Whitaker *et al*^[18] employed chisels; ²Both osteotomes and drilling burrs were used in these 42 patients;

³Hand-held surgical drill and chisel were used in 61 patients whereas drill only was employed in 10 patients. NA: Not applicable.

also gives access to the temporalis fascia, which can be harvested for grafting bare areas of the external auditory canal^[32].

Sensorineural hearing loss has been reported after the removal of exostoses by drilling techniques, presumably as a result of prolonged noise exposure from drilling. The osteotome technique has the potential advantage of less risk of cochlear damage compared to the drilling technique. Moreover, osteotomes pose little risk of avulsing skin, which is really important to prevent postoperative canal stenosis^[19]. Nevertheless, most surgeons are more familiar with drilling techniques than with the use of osteotomes, which has the risk of mobilizing an entire segment of bone, of traumatizing the tympanic membrane, or of exposing periosteum anterior to the anterior bony wall and thus damaging the temporomandibular joint. To minimize complications with osteotomes, incremental removal of bone is advised^[19]. The different approaches and techniques employed are shown in Table 1^[18,19,27,29-33,35,37-41].

There is no consensus regarding which canal exostoses should be removed first. Some authors prefer removing the anterior exostoses first (or these are even the only exostoses removed)^[34] whereas other surgeons address the posterior exostoses first^[33]. Finally, other authors recommend removal the superior, inferior and anterior exostoses first, and only when the tympanic membrane position is verified do they remove the posterior wall protrusion^[32].

At the end of the procedure, the skin does not usually have the circumference necessary to cover the new canal, and several vertical incisions are, therefore, performed in the skin with small scissors in order to ensure intimate contact of the skin with the bone. If any significant amount of bone remains bare, it is covered with fascia temporalis or a split thickness skin graft, as this allows for faster healing and re-epithelialization. Finally, pieces of gelatin

sponge soaked with antibiotic and anti-inflammatory drops are inserted in the ear canal along the various skin incisions and a cotton ball is placed in the ear meatus as a dressing.

COMPLICATIONS

Surgical removal of exostosis is not a procedure without risks and in fact the number of complications is not low when we revise the literature.

Postoperative canal stenosis is one of the most frequent complications (Table 2)^[18,19,27,29-33,35,37-45]. Integrity of healthy meatal skin is a key factor in the success of the operation. Loss of skin can lead to formation of granulation tissue, fibrous stenosis and membranous atresia. This fibrous stenosis could require surgical revision with excision of the scar and relining of the external auditory meatus with a split thickness skin graft. Preoperative severe persistent external otitis should be addressed before the operation as it can be another risk factor for developing this complication^[38].

Tympanic membrane perforation is another of the most frequent complications. These tympanic membrane tears may be pinpoint and require no additional treatment, healing by the time the ear canal has healed^[19]. But in other cases, repair of the defect requires an underlay tympanoplasty, which is usually performed in conjunction with removal of the exostoses.

Other less frequent but more feared complications reported in the literature are damage to the temporo-mandibular joint or the facial nerve, sensorineural hearing loss, cervical subcutaneous emphysema and petrositis^[43,44,46]. Not all cases of anterior canal wall dehiscence, protrusion, prolapse, herniation of the temporomandibular joint or even exposure of the temporomandibular joint capsule are symptomatic but they could lead to chronic

Table 2 Complications of exostoses removal

Ref.	Year	No. of patients	No. of ears	No. of operations	TM tears or perforations ¹	Stenosis or scarring ¹	SNHL	Tinnitus	TMJ entry
Altuna	2004	45	52	52					
Mariezcurrera <i>et al</i> ^[33]									
Di Bartolomeo <i>et al</i> ^[30]	1979	16	21						1 (4.7%)
Bordure <i>et al</i> ^[37]	1994	36	64		8 (12.5%)		3 (4.7%)	2 (3.1%)	2 (3.1%)
Fisher <i>et al</i> ^[42]	1994	102		127	11 (8.6%)	2 (1.5%)	1 (0.8%)		1 (0.8%)
Frese <i>et al</i> ^[38]	1999	48		59	3 (5%)	4 (6.8%)	4 (6.8%)	1 (1.7%)	
Hempel <i>et al</i> ^[31]	2012	30	35			1 (2.8%)	2 (5.7%)	1 (2.8%)	
Hetzler ^[19]	2007	140	221	221	1 ²		3 (1.3%)	3 (1.3%)	3 (1.3%)
Hurst <i>et al</i> ^[39]	2001	49		64	1 (1.5%)	2 ³		1 (1.5%)	2 (1.9%)
King <i>et al</i> ^[29]	2010	58	83	83		4 (4.8%)		7 (8.4%)	
Oostvogel <i>et al</i> ^[40]	1992	51			1 (2%)		3 (5.9%)		
Portmann <i>et al</i> ^[41]	1991		25		1 (4%)		2 (8%)		1 (4%)
Reber <i>et al</i> ^[43]	2000	20		22	2 (9%)	2 (9%)	4 (18.2%)		2 (9.1%)
Sanna <i>et al</i> ^[32]	2004	57	65	65		2 (3%)	1 (1.5%)		
Seehy <i>et al</i> ^[44]	1982			79	8 (10.1%)				
Stougaard <i>et al</i> ^[35]	1999	19	24	24	1 (4.1%)		1 (4.1%)		1 (4.1%)
Timofeev <i>et al</i> ^[27]	2004	31	46	46		4 (8.7%)	1 (2.2%)	1 (2.2%)	
Vasama <i>et al</i> ^[45]	2003	136	182	182	5 (2.7%)	2 (1.1%)	4 (2.2%)		
Whitaker <i>et al</i> ^[18]	1998	18	27						7 (25.9%)

¹TM tears or perforations requiring underlay tympanoplasty, or stenosis or scarring requiring revision surgery; ²Although only one case required tympanoplasty, there were 29 tympanic membrane pinpoint tears that required no additional treatment and were healed by the time the ear canal skin had healed; ³One tympanic membrane perforation and 2 ears requiring revision surgery for removal of granulations and sequestered bone. SNHL: Sensorineural hearing loss; TM: Tympanic membrane; TMJ: Temporomandibular joint.

symptoms that do not resolve with anti-inflammatory medication, such as temporomandibular joint pain, trismus and masticatory problems, and unnecessary movement in the canal which disrupts the healing process and facilitates postoperative stenosis.

Dizziness, vertigo, worsening of the preoperative air-bone gap, tinnitus and diminished sense of taste have also been reported^[19,27,29,31,32,38-40]. Interestingly, dizziness and vertigo were reported in two studies in which osteotomes were used, and were probably due to the concussive forces applied to the temporal bone^[19,29]. This was resolved with canalith repositioning.

SURGICAL FOLLOW-UP AND PROGNOSIS

Careful observation of the ear canal should be emphasized during follow-up in order to recognize and treat early infection or granuloma, and prevent the risk of restenosis. Audiometric follow-up is also important to assess post-operative air-bone gap and detect any deterioration of bone conduction and sensorineural hearing loss.

The healing rate should also be considered, as this can be a major concern for patients who are mostly water sports enthusiasts and wish to resume their normal activity as soon as possible. Postoperative healing ranges from 2 to 16 wk (average 8 wk)^[18,19,30,42-44]. The best results have been achieved with the osteotome technique by maximizing ear canal skin preservation and minimizing skin disruption^[19].

Surgery for ear canal exostoses improve patients'

health-related quality of life (HRQOL)^[31,45]. Ninety percent of the patients were satisfied with the result of the operation, and would decide in favor of the operation in retrospect, according to one study^[31]. Complications were the most important factor for the lack of patient benefit and had a negative impact on the patient HRQOL.

With regard to prognosis it should be pointed out again that exostoses are typically a benign condition that does not usually require surgical therapy. Here our interest and comments will therefore focus on the risk factors for severe exostoses in high risk populations and for restenosis after surgery. The relation between exposure to cold water and development of exostoses has been dealt with in the section on etiology. The risk of developing exostoses and the degree of ear canal obstruction increase proportionally to the frequency of exposure according to many different studies^[5-8,11,23,25]. Indeed, a surfing index has been proposed to better predict the risk of the formation of external auditory exostoses and is expressed as the product of the period (years as an active surfer) and the frequency (number of surfing days per week)^[5]. Thus, subjects who have participated in water sports for longer than 10 years show some evidence of exostoses. This may be preventable since those who use earplugs are less likely to develop exostoses^[9]. Similarly, individuals who postoperatively participate in aquatic sports experience recurrence of the stenosis more rapidly, and protecting the ear canals increases the recurrence-free interval^[27]. Nevertheless, very few participants in water activities use these precautions

(ear plugs, neoprene hood), even among those aware of the preventability of surfer's ear^[47]. One reason for this could be that wearing earplugs impairs hearing. In this respect, soft prefabricated elastomer earplugs have been shown to cause less hearing impairment than custom-fitted silicone and custom-fitted acrylic ear plugs and are therefore preferable^[48].

However, the risk of recurrence does not only depend on new exposure to cold water. Although ear plugs could protect against the development of new exostoses, recurrence has been observed even in patients who stopped water sport activity completely after surgery^[27]. The age of the patient at the time of operation is also a factor associated with the recurrence rate of stenosis. According to Timofeev et al^[27] the older the patient, the faster the recurrent disease develops.

CONCLUSION

Exostosis of the auditory canal is a prevalent condition in water sports enthusiasts and in those who engage in an aquatic activity professionally or out of necessity. Most cases are asymptomatic and are chance discoveries at otoscopy. However, when they cause recurrent infections or hearing loss, surgical intervention becomes necessary. Many different surgical approaches and techniques are employed, and the outcomes seem to depend more on the experience and expertise of the surgical team than on technical aspects, although the latter should be borne in mind in order to avoid complications. The possibility of even serious complications should not be underestimated since they have a negative impact on patient quality of life. Finally, many patients wish to resume their aquatic activities as soon as possible so that it becomes even more important to maximize precautions and to avoid the bare areas of skin and prevent infections and restenosis in the postoperative period.

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Revision surgery for otosclerosis: An overview

Sertac Yetiser

Sertac Yetiser, Department of ORL and HNS, Anadolu Medical Center, Gebze, 41400 Kocaeli, Turkey

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Correspondence to: Sertac Yetiser, MD, Professor, Department of ORL and HNS, Anadolu Medical Center, Cumhuriyet mahallesi, 2255 sokak No:3, Gebze, 41400 Kocaeli, Turkey. syetiser@yahoo.com

Telephone: +90-532-3248433

Fax: +90-262-6540529

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Abstract

Stapes surgery for otosclerosis has been proved to be a very satisfying procedure. However, the condition is difficult for the patients with no or little hearing gain after surgery and for those who had sudden or gradual hearing loss after a successful air-bone gap closure in the follow-up period. The issue of re-exploring the middle ear is challenging. A general review of this subject from several points of view remains lacking. In this study, articles related with the revision surgery for otosclerosis have been reviewed after a PubMed research and common and/or contradictory points were documented. The aim of this study is to give an insight to diagnostic and therapeutic approaches for

the clinicians in patients who need a revision surgery. In conclusion, prosthesis problems, loose prosthesis in stapedotomy and migrated prosthesis in stapedectomy are the most common causes for revision surgery. Most important indicators which effect better hearing outcome following revision surgery are those ears with the presence of incus, with no obliteration of oval window, with small fenestra stapedotomy and the experience of surgeon. The risk of neurosensorial hearing loss in revision cases is not high but the hearing gain is limited as compared to primary cases. The rate of 10 dB air-bone gap closure is around 60%-70% at most and even less promising results have been reported. Patient's demands and expectations have to be clarified in a realistic way.

Key words: Otosclerosis; Stapedotomy; Stapedectomy; Revision

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Core tip: It is very difficult for the patients with otosclerosis having no or little hearing gain after surgery and for those who had sudden or gradual hearing loss after a successful air-bone gap closure in the follow-up period. The issue of re-exploring the middle ear is challenging. A general review of this subject from several points of view remains lacking. In this study, articles related with revision surgery for otosclerosis have been reviewed after a PubMed research and common and/or contradictory points were documented.

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INTRODUCTION

Revision surgery for otosclerosis is always difficult

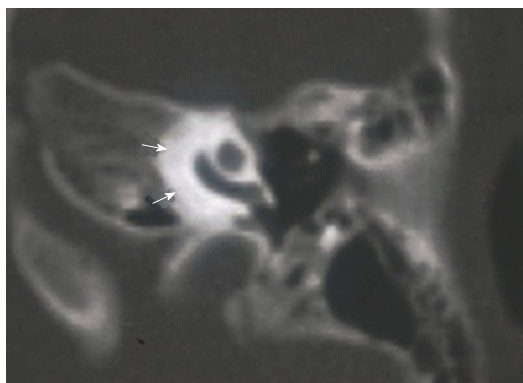


Figure 1 CT appearance of cochlear otosclerosis. Note for perilyabyrinthine decalcification (marked with arrows).

to decide and also challenging for both the surgeon and the patient. Difficulties of re-operation are higher and the success rate is less predictable. Before going back to the operating room, all related issues about the primary intervention (type of otosclerotic focus, facial nerve dehiscence, the length, diameter and the type of prosthesis, type of anesthesia, balance problem, graft for oval window, operation time, bleeding, the technique, type of laser, etc.) and about the patient (his age, occupation, his opinion about the secondary surgery, his current psychological status, hearing level, any possible cause for hearing loss, etc.) have to be looked at. The details which have to be reviewed before surgical planning are numerous. What type of anesthesia will be used, what if the revision side is the better hearing one, what if the patient has an associated balance problem. An option of hearing aid has to be frankly discussed with the patient. The aim of this review study is to enlighten this subject from its all aspects.

EPIDEMIOLOGY, INCIDENCE AND ETIOLOGY

The incidence of revision surgery for otosclerosis has been declined over the years. Major indications for re-exploring the middle ear are usually for patients who had no hearing recovery after stapes surgery at the early period or those who had sudden or gradual conductive or neurosensorial hearing loss in the long run. Revision surgery is also needed for patients with intractable or chronic and recurrent balance problems after surgery and no relief with medical treatment whether they have hearing loss or not. For those with stable and long term hearing loss, chance of hearing restoration is quite low. There seems to be no common understanding in the literature deciding the surgery in terms of hearing loss. Most surgeons are not willing to operate the patients with less than 20 dB air-bone gap and discrimination score less than 60%. Some of the air-bone gaps are very mild

and could be resolved easily by medical measures. On the other hand, cochlear otosclerosis could be the main cause for those with slowly progressive sensorineural hearing loss and there is no indication for revision surgery. Cochlear otosclerosis causes progressive damage to the organ of Corti and stria vascularis. Venous congestion and abnormal blood circulation leads to hyalinisation of spiral ligament^[1]. Ear drum looks like more reddish than it was before. Increased blood flow of the promontrium vessels gives a typical finding to the tympanic membrane, so-called "Schwartz sign"^[2]. Linear perilyabyrinthine decalcification as seen on the temporal bone CT scanning is diagnostic finding (Figure 1)^[3].

Prosthesis dislocation from oval window, incus erosion and incus-prosthesis detachment, short prosthesis, postoperative fibrosis in the middle ear and re-ankylosis, perilymphatic fistula, insufficient fenestra and too tight prosthesis, footplate resclerosis, incus subluxation, facial nerve dehiscence and prosthesis friction, reparative granuloma, vestibular symptoms due to long prosthesis, malleus-incus fixation, neurosensorial hearing loss are some of the main causes of revision surgery. However, the incidence of the causes for revision surgery has greatly changed over the years mostly due to the technique and the materials used for hearing restoration. One of frequent causes of revision surgery during the era of wire-gel foam or wire-adipose tissue prosthesis was perilymphatic fistula presenting with neurosensorial hearing loss and prolonged unsteadiness^[4,5]. However, common causes of re-operation, more recently are due to re-fixation of prosthesis, prosthesis coming off the oval window for some reason, and incus necrosis presenting with gradual or sudden conductive hearing loss^[6-10].

Fisch *et al*^[11] have reported that in almost 80%-85% of cases, revision surgery is related with either prosthesis (too tight or fixed, too loose, too long, too short, bended, etc.) or oval window problems (fibrosis, narrowing, granulation tissue, new bone formation, fistula). In addition to problems like inadequate crimping of prosthesis to incus or prosthesis detachment or overlooked incus mobility problems, one of the most common causes of revision surgery is substantially incudo-malleal ankylosis or malleal-epitympanic fixation especially for patients who have limited hearing gain after primary surgery. For this reason, some clinicians claim that a malleo-stapedotomy or disconnection of malleus head after drilling attical bone could be necessary and superior canaloplasty incision should always be included during classic end-aural approach to inspect the anterior malleal ligament and incudo-malleal articulation, as well^[11-13]. However, 46% incidence of incus-malleus ankylosis claimed by Fisch *et al*^[11] had not been supported by others^[14-17]. The incidence is about 2% for Lippy *et al*^[18] and 10% for Causse *et al*^[19]. On the other hand, histopathological investigations on fixed malleus head revealed normal

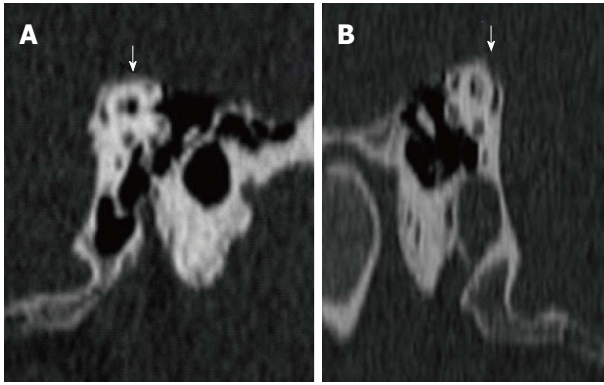


Figure 2 Bilateral superior semicircular canal dehiscence of A and B (marked with arrows).

bone or tympanosclerotic focus^[20]. But, of course it has primary importance to control the mobility of incus and malleus during primary surgery. Some authors proposed Laser-Doppler-Interferometer to objectively distinguish otosclerosis and malleus fixation before the surgery^[21]. A constant minimum 10 dB air-bone gap could always be present even after a perfect stapes surgery if such malleus problem is overlooked.

Malleus fixation can be an acquired problem due to tympanosclerotic process of childhood otitis media or it can be congenital. It can be found as an isolated problem in 80% of incidence. However, it may be associated with incudo-malleal fixation in 15% of cases^[22]. However, its co-existence with otosclerosis is an interesting subject. Malleus fixation associated with otosclerosis was first reported by Guild^[23]. Nandapalan *et al*^[24] have found a kind of hyalinization process in anterior and superior malleal ligaments in 30% of otosclerotic temporal bones. This high incidence was not supported by other studies. Subotic *et al*^[25] have reviewed 1108 normal temporal bones and reported 14 congenital malleus fixations. Oktay *et al*^[26] have found no relation between hyalinization of anterior malleal ligament and otosclerosis. Vincent *et al*^[22] have reported that 30% of patients with malleus fixation have a history of otitis media in the past. If it is a congenital abnormality, it should be related with an anomaly of Meckel cartilage during 7th month of fetal life probably due to cessation of resorption of some embryonic mesenchymal tissue^[27]. Genetic aspect and the type of transmission is not clear although familial cases have recently been reported^[28].

Probabilities for the presence of air-bone gap after primary surgery are listed below: (1) Middle ear stiffness: clots, too much gel foam in the middle ear, adipose tissue placed around the prosthesis could be the reason. But, they have a minor effect; (2) Eustachian dysfunction and associated ventilation problems: this is a temporary condition; (3) Problems of tympanic membrane: tympanic membrane which is not flexible yet, tiny perforations or restored ear

drum with underlay fascia or perichondrium may affect hearing; (4) Edema of the external auditory canal: restoration of hearing is expected after resolution of the edema; (5) Immobile prosthesis: hearing gain is very little if the prosthesis is too tight in the oval window fenestra; (6) Loose prosthesis: the attachment of prosthesis to incus is too floppy; (7) Prosthesis out of oval window or too short prosthesis: there is no hearing gain. It is even worse; (8) Prosthesis with small caliber: little air-bone gap can be found; (9) Incudo-malleal ankyloses: sometimes, this is overlooked and again there is no hearing gain; (10) Partial dislocation (subluxation) of incus; incus can be dislocated, if it is forced too much during insertion of prosthesis. Air-bone gap is mild; (11) Oval and round window abnormalities: hearing gain could be very limited in case of obliterative otosclerosis; (12) Dehiscence of superior semicircular canal: one of the interesting clinical entity that may mimic otosclerosis is superior semicircular canal dehiscence^[29-31]. Those patients have low frequency conductive hearing loss because of absorption of sound energy through the bony defect which is defined as "third window effect". Stapes surgery will not restore the hearing in those patients. Therefore, patients with conductive hearing loss should be evaluated with temporal bone CT (Figure 2); (13) Otosclerosis with cavitations: hearing gain could be very limited due to "third window effect"^[32]; (14) Large vestibular aqueduct: there is no enough air-bone gap closure because of "third window effect"^[33]; (15) Pressure of dehiscent facial nerve: bulging facial nerve can press the prosthesis and can hamper its mobility; (16) Inner ear pressure: inner ear pressure can affect the mobility of stapes in cases with stapes gusher; (17) Meniere and otosclerosis: an attack of Meniere can lead to temporary hearing loss, even though it is very rare^[34]; (18) Pneumolabyrinth: air in the labyrinth can cause limited gain, if too much perilymph is aspirated; and (19) Audiometric inaccuracy: technical problems (inadequate testing conditions, masking problems, etc.) or patients' faulty guidance (too much tinnitus, medico-legal conditions, etc.) may lead to pre or postoperative faulty audiograms.

PATIENT COUNSELLING AND SURGICAL PLANNING

One of the important subjects before deciding a revision surgery is the patient's demand and his expectations which have to be truly clarified in a realistic way. Hearing loss after primary surgery is very frustrating for both the patient and the surgeon. Surgeons experience has utmost importance. But, it is a mutual understanding that the risks are higher in revision cases. One of the first things to do is to calm down the patient, explain the situation and wait for a couple of months for a more reliable audiogram.

The patient should be evaluated with temporal bone CT scanning and MRI during this waiting period. Age seems to be unimportant for decision making^[35]. A detailed history of the patient is necessary. Every pieces of information should be explored before the surgery. Each steps of the primary surgery from the beginning or even the diagnosis of otosclerosis should be checked again, especially if there is no hearing gain or the gain is very little. Those are the main questions which have to be reviewed thoroughly: (1) What are the hearing level and word recognition scores before and after the primary surgery; (2) What is the operation time, the technique, (stapedectomy/stapedotomy) and type of anesthesia? (If it is local, what are intraoperative hearing and balance findings?); (3) What is the age of patient, gender, occupation, associated health problems and date of diagnosis of otosclerosis; (4) What is the type of prosthesis, length and diameter; (5) What are the otosclerotic and the footplate findings; (6) How did the hearing loss happen (sudden, progressive, etc.)? What are the possible causes and associate symptoms? What is the duration from primary surgery and hearing loss; (7) How is the facial nerve, the graft over the footplate, any associated problem (bleeding, perilymph leakage, etc.); (8) How is the intra or postoperative balance; (9) What is the method used to open the footplate (laser, pick, drilling)? What are the particular findings (obliteration, floating footplate, fragile/thin footplate, etc.); and (10) What are the patient's thoughts about the primary and revision surgery.

PROSTHESIS PROBLEMS (FIXATION-DISLOCATION)

Prosthesis problems are generally related with lateralization of the prosthesis or prosthesis re-fixation at the oval window which are presented with conductive type hearing loss being more evident at higher frequencies^[36]. Fibrosis around the oval window and re-stenosis may intervene with the mobility of the prosthesis. Fibrosis is mostly related with mucosal injury and foreign body reaction (silicone blocks, gel foam, wire prosthesis, etc.). A kind of soft tissue quickly coats the prosthesis and envelops all around it. However, its effect on hearing is questionable. Sim *et al*^[37] have investigated the influence of postoperative tissue formation on sound transmission with laser Doppler and electron microscopy and have found that this is negligible. Hearing loss is pretty much related with re-stenosis and can be seen as early as a year after primary surgery. Nadol points out that new lesions can be related with drilling around the otosclerotic lesion. He recommends less possible drilling^[8]. The incidence of extensive and obliterative otosclerosis is about 7%-11%^[38-40]. Re-stenosis of oval window is one

of the major problems. Sheehy reported that 10% of his revisions were because of oval window re-stenosis and 75% of them were primary obliterative cases^[41].

The incidence of primary round window otosclerosis is less than 1%^[8]. However, round window narrowing and obliteration due to otosclerotic focus extending from oval window have been found in 23% of cases. Therefore, it is an important precaution to always inspect the round window during primary surgery. On the other hand, round window abnormalities may mimic otosclerosis. Borrmann *et al*^[42] have reported non-syndromal round window atresia with otosomal dominant penetrance in 2 members of the family. Identification of severe obliteration of round window will prevent unnecessary stapes surgery. However, what is best to do if the otosclerotic lesion is extending to the round window. Studies indicate an increased risk of neurosensorial hearing loss during cleaning process of round window^[43,44].

It was commonly seen in revision cases of the earlier period that prosthesis displacement was mostly related with wire-gel foam and wire-adipose prosthesis. Prosthesis-incus attachment was somehow functional and often times, prosthesis sliding out of the intact incus was not case. On the contrary, dislocation was more common at the inferior side of oval window in which the stapedectomy was the common technique^[14,15]. Prosthesis coming off incus while it is still in place at oval window in cases of stapedotomy is usually associated with incus necrosis or oval window re-stenosis. This is called as "lateralized piston" and is seen in 18.5% of stapes surgery revision^[45].

INCUS PROBLEMS

Incus problems can be seen with almost every types of prosthesis. Incus necrosis is not the result of reduction of vascular supply or mucosal disruption due to very tight holding of prosthesis as believed once but it is because of vibratory movements at the prosthesis-incus contact site due to flaccid prosthesis or re-stenosis of the oval window constricting the prosthesis^[7]. Fibrosis or adhesions can also disturb synchronous movement of incus and the prosthesis and give rise to increase of friction and a kind of different phase of the piston movement^[8,46]. This is known as "Loose-wire syndrome". The characteristic sign is the short term improvement of patient's hearing following Valsalva maneuver.

The impact of incus necrosis on hearing after revision stapes surgery is not predictable. All techniques of ossicular reconstruction should be considered in case of incus necrosis. One option is to lengthen the long arm of incus with bone cement and secure the prosthesis if its movement in the fenestra is fine (Figure 3)^[47]. However, if the long arm of incus is completely gone, if incus is luxated or



Figure 3 Revision stapes surgery due to incus necrosis. Bone cement was used to reconstruct the incus. Titanium prosthesis was placed over the fixed incus.

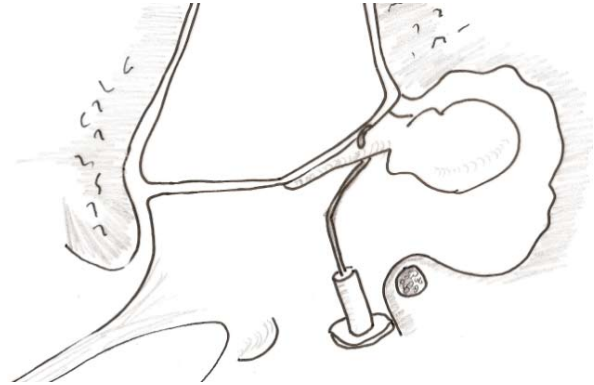


Figure 4 Ossicular reconstructions between the mobile malleus and stapes footplate fenestra if incus is not available due dislocation or extensive necrosis.

head of malleus is fixed at epitympanium, surgeon must consider to by-pass the incus with or without removal of head of malleus (Figure 4)^[48]. Placement of the prosthesis between the mobile malleus handle and stapes footplate or a TORP between the ear drum and oval window may be required^[13,49]. If it is not possible to use any ossicles then TORP is the only option. Sheehy has reported that he had to use TORP in 20% of revision cases. He had 64% of 10 dB air-bone gap closure and 95% of 20 dB air-bone gap closure^[49]. Prosthesis can be inserted to malleus anterior or posterior to the processus brevis without stripping the ear drum from malleus handle. Attachment of the prosthesis to the neck of malleus at the back of the processus is preferred. But, if fixed malleus head is resected then prosthesis is attached in front of the processus. Mangham *et al*^[50] have compared the hearing results with incus vs malleus reconstruction and have reported that results of reconstructive procedures with malleus (with incus by-pass) are much better. Ghonim *et al*^[51] have reported 10 dB air-bone gap closure in 58.3% of cases and 20 dB air-bone gap closure in 83.3% of cases in which malleus was re-located and incus was transferred between the malleus and footplate.

Malleus diameter is about 3.5 mm and fine attachment is very important. This will prevent prosthesis dislocation and excessive movement in the vestibule synchronous with the movement of the ear drum. Titanium clips and wire pistons are used for this purpose^[52,53]. In case of narrow oval window, the wire prosthesis is angulated which has to be little bit far away from the upper part of the oval window where utriculus is located. Malleus-oval window pistons can provide 10 dB air-bone gap closure^[22]. Hausler *et al*^[54] proposed titanium malleus prosthesis when ideal angulation and placement of wire prosthesis is not possible. Seidman and Babu reported considerable hearing gain in patients with incus-malleus fixation by epitympanic liberation of the malleus with laser and incus transposition

without insertion of prosthesis and removal of malleus head^[55].

OSSICULAR RECONSTRUCTION IN REVISION SURGERY

When the middle ear explored, one should pay much attention to long arm of incus, prosthesis-oval window relation, mobility of the ossicles and prosthesis and finally, should inspect the site of leakage in patients with vertigo. What the surgeon must do if there is nothing particular in the middle ear. Should he change the prosthesis, anyway or should he replace with the new one although the old prosthesis still looks good after restoration of the problem? Exchanging the prosthesis probably has least importance for additional hearing. However, Jahnke *et al*^[56] have reported better hearing when they replaced the old one with titanium as compared those in which the old one was left (69.4% vs 76.2%).

Laser is very effective in revision cases especially in those with extensive granulation tissue or fibrosis^[57,58]. Laser helps for less bleeding during surgery. However, it is important to remove the old prosthesis before using laser and not to shoot directly the prosthesis. Teflon prosthesis, silicone blocks could melt and get sticky, granulation tissue around the metal piston may increase the heat, hydroxyl-appetite could break. It has been reported that KTP laser is not suitable for patients with middle ear implant in the presence of blood and granulation tissue^[59,60]. Haberkamp *et al*^[57] have reviewed revision cases with or without CO₂ laser and have reported that laser allows positive identification of the oval window and assures placement of prosthesis. However, its role on better hearing outcome is not clear. Silverstein *et al*^[61] have found no difference in comparison of hearing outcome of patients following revision stapes surgery with and without laser. On the other hand, Wiet *et al*^[62] have found better hearing outcome in those with

Table 1 Studies regarding the hearing after revision stapes surgery

Ref.	No. of cases	10 dB AC-BC gap (%)	20 dB AC-BC gap (%)
Crabtree <i>et al</i> ^[67]	35	46	-
Sheehy <i>et al</i> ^[41]	258	44	71
Pearman <i>et al</i> ^[68]	95	58	73
Derlacki ^[14]	217	65	72
Glasscock <i>et al</i> ^[15]	82	39	64
Bhardwaj <i>et al</i> ^[69]	120	46.5	-
Lesinski ^[7]	57	66	89
Farrior <i>et al</i> ^[6]	102	58	85
Langman <i>et al</i> ^[17]	66	61	84
Somers <i>et al</i> ^[70]	332	40	64
De La Cruz <i>et al</i> ^[71]	356	59.8	77.5
Lippy <i>et al</i> ^[72]	483	71	-
Gros <i>et al</i> ^[73]	63	52.4	-
Babighian <i>et al</i> ^[74]	78	54	-
Bakhos <i>et al</i> ^[10]	89	52	-

laser surgery.

HEARING RESULTS FOLLOWING REVISION SURGERY

It should be kept in mind that hearing restoration after revision stapes surgery is not as successful as primary surgery. The rate of 10 dB air-bone gap closure is around 60%-70% at most and even less promising results have been reported^[7,63]. A realistic approach is to tell the patient that the chance of better hearing is about 60%, but also the hearing may not change or even may get worse. Pedersen have reported 17% worsening in a series of 186 revision cases^[9]. Palva *et al*^[64] have reported 23% worsening in 76 revision cases. Richards *et al*^[65] have reported hearing loss in the counter lateral ear in some of the revision cases which was termed as "sympathic cochleolabyrinthitis". The role of age in revision cases has been investigated, but no difference in hearing gain have been found between elder and young patients^[26,66]. Glasscock *et al*^[15] have reported that those cases with better hearing results following primary surgery also have better results following revision surgery. On the other hand, long term follow-up studies demonstrate that early hearing gains are prone to decline over the years. Lippy have reported that 72% air-bone gap closure rate drops to 50% in 10 years. Besides, those with multiple interventions have even worse results. Table 1 shows hearing results of several studies following revision surgery. The rate of 10 dB air-bone gap closure after revision surgery ranges between 39%-71%^[3,4,7,11,12,14,38,67-74].

REVISION SURGERY FOR POST-OPERATIVE BALANCE PROBLEM

Besides its audiological gain, stapes surgery also

means an intervention to the closed labyrinth system. Patients usually have abnormal caloric responses lasting for a long time^[75]. Increased utricular activity and dysfunction of sensorial organization have been documented by subjective visual horizontal test and posturography^[76-78]. Decrease in perilymphatic amount, mechanical effects of aspiration, dryness, heat, prosthesis irritation, air or blood infiltrating to the vestibule and probable enzymatic reactions could possibly alter the micromechanics of the labyrinth. An irritative nystagmus beating toward the counter lateral ear is seen for 3-4 d following surgery which disappears in normal condition^[79,80]. There is usually no relation with the transient vertigo and hearing gain after surgery. However, persistent nystagmus is indicative of chronic vestibular irritation. Vestibular exercises and medical therapy can provide relief of symptoms in some patients or symptoms may disappear with no obvious reason. Some patients get used to it. However, symptoms sometimes could be unbearable. One of the challenging problems is to decide for re-operation in patients with post-operative vertigo resistive to medical therapy and normal hearing.

The most definite cause for prolonged unsteadiness is the presence perilymphatic fistula. The incidence is about 10% and is due to inadequate sealing around the prosthesis in the oval window^[12]. It is less seen in patients with oval window grafting^[81]. Nakashima *et al*^[2] have reported the incidence of perilymph fistula was 22% in patients with gel-foam sealing and 4% in patients with tissue sealing. Sheehy and Perkins have reported 3.5%, 1.9% and 0.6% of fistula rate with gel-foam, adipose tissue and fascia, respectively^[38]. Tinnitus, aural fullness, neurosensorial hearing loss and vertigo which is more evident when the patients lie on the non-operated side or with Valsalva maneuver is found. Nystagmus as seen on tympanogram and pneumolabyrinth on MRI support the perilymphatic fistula^[82]. Another possibility for post-operative balance problem is the irritation of long prosthesis which is presented with similar symptoms (Figure 5). Symptoms are worse with head movement and after Valsalva maneuver. However, hearing loss may not always accompany the vestibular problem. In less than 1% of cases, progressive hearing loss and vertigo could be related with reparative granuloma^[83,84]. If medical treatment including anti-vertiginous drugs, prophylactic antibiotics, diuretics, steroids fails, the middle ear re-exploration is indicated.

CONCLUSION

Prosthesis problems, loose prosthesis in stapedotomy and migrated prosthesis in stapedectomy are the most common causes for revision surgery. Most important indicators which effect better hearing outcome following revision surgery are those ears with the presence of incus, with no obliteration of oval window, with small fenestra stapedotomy and

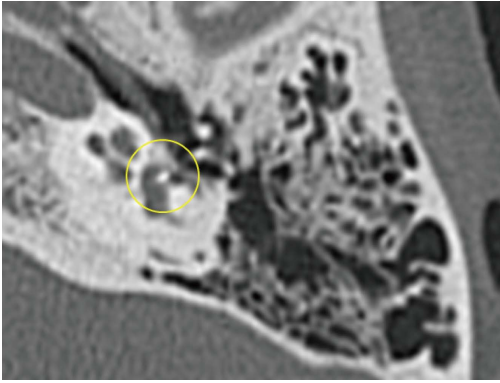


Figure 5 Temporal bone tomography with long prosthesis inside the vestibule (marked with yellow circle).

the experience of surgeon^[9]. Obliterative cases have the worst outcome. Finally, the risk of neurosensorial hearing loss in revision cases is not high but the hearing gain is limited as compared to primary cases.

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Retrospective Study

Complicated sinusitis in children: 18 cases report

Ali Mardassi, Nabil Mathlouthi, Hajer Mbarek, Chiraz Halouani, Sameh Mezri, Cyrine Zgolli, Ghassen Chebbi, Rania Ben Mhamed, Khemaies Akkari, Sonia Benzarti

Ali Mardassi, Nabil Mathlouthi, Hajer Mbarek, Chiraz Halouani, Sameh Mezri, Cyrine Zgolli, Ghassen Chebbi, Rania Ben Mhamed, Khemaies Akkari, Sonia Benzarti, ENT Department, Military Hospital, Tunis 1008, Tunisia

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Correspondence to: Ali Mardassi, MD, ENT Department, Military Hospital, Montfleury, Tunis 1008, Tunisia. alimardassi@gmail.com

Telephone: +216-22-552252

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itted to the Ear, Nose and Throat Department of the Military Hospital of Tunis, Tunisia for a complicated rhinosinusitis between 2006 and 2013 were evaluated. Data related to patients and the disease were collected and analyzed: past medical history, complaints, clinical examination, radiologic findings, therapeutic management and evolution.

RESULTS: Eighteen cases were identified with a mean age of 5.1 years (5 mo to 13 years) (SD \pm 3.1). A male preponderance was noted in 72% of the cases. Rhinorrhea and fever were the most common presenting symptoms. Radiological explorations (computed tomography-scan \pm magnetic resonance imaging) have been practiced for all of our patients. Orbital involvement was found in 77% of the cases associated with meningitis in 2 cases. Antibiotherapy was prescribed to all our patients. Surgical procedures were performed in 8 cases: endoscopic sinus surgery and/or external drainage of orbital abscess. After an average follow-up period of 2.5 years, 3 of our patients were lost. The ophthalmic sequelae noted in 3 cases (16%) were permanent and caused important functional and social problems. A favourable outcome has been noted in the rest of our patients.

CONCLUSION: Rhinosinusitis can be extremely severe in children requiring urgent radiological imaging and aggressive treatment to avoid orbital and intracranial complications.

Key words: Rhinosinusitis; Children; Imaging; Orbit; Meningitis; Antibiotherapy; Surgery

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Abstract

AIM: To precise the clinical characteristics of rhinosinusitis in pediatric population, their complications and therapeutic approaches.

METHODS: All infants younger than 15 years adm-

Core tip: Rhinosinusitis is a common condition in childhood. However, complicated cases occur less frequently and are potentially life-threatening. The clinical presentation is often modified by a prior antibiotic

prescription. In this paper we report our experience in the management of complicated sinusitis in infants and compare it with literature data. Orbito-cranial extension must be suspected in presence of proptosis, Swelling and/or redness of the eye or persistent headache. Urgent contrast-enhanced computed tomography-scan is the recommended initial imaging. Once the diagnosis confirmed, intravenous antibiotherapy should be started. Surgery is indicated in selected cases. A regular follow-up is mandatory.

Mardassi A, Mathlouthi N, Mbarek H, Halouani C, Mezri S, Zgolli C, Chebbi G, Ben Mhamed R, Akkari K, Benzarti S. Complicated sinusitis in children: 18 cases report. *World J Otorhinolaryngol* 2015; 5(1): 30-36 Available from: URL: <http://www.wjgnet.com/2218-6247/full/v5/i1/30.htm> DOI: <http://dx.doi.org/10.5319/wjo.v5.i1.30>

INTRODUCTION

Acute rhinosinusitis is a relatively common disease affecting both children and adults. Its prevalence rate is between 6% and 12% with viral origin in most cases and a bacterial one in only 0.5%-2%^[1]. Fungi have been described as causative agents especially in immunocompromised patients^[2,3]. Diagnostic criteria and therapeutic management are still a controversial issue. Complications of acute bacterial rhinosinusitis are uncommon in children, but they can be extremely severe and potentially life threatening^[1,4,5]. Mortality rate has decreased significantly from 42% in the 1950s to 12% in the 1990s thanks to medical progress^[5].

MATERIALS AND METHODS

We retrospectively reviewed the medical records of complicated rhinosinusitis in children admitted to the Ear-Nose-Throat Department of the Military Hospital of Tunis Tunisia between 2006 and 2013. The children treated for an acute and non complicated rhinosinusitis by oral therapy and without the need for an overnight stay in hospital were excluded from the study. Parameters recorded were epidemiologic data, clinical presentation, biological results, radiological investigations, localization of affected sinuses, types and sites of complications, therapeutic management and outcomes.

Statistical analysis

In this retrospective and descriptive study, 18 patients have been reported. For quantitative data, the mean and the standard deviation have been obtained. For qualitative data, the percentage of each subgroup has been calculated. The statistical method is adequate because there was no comparison between

the variables. *P*-value was not calculated since no hypotheses are tested in the study.

RESULTS

Eighteen children with complicated rhinosinusitis were identified over 8 years (2006 to 2013). Ages ranged from 5 mo to 13 years with an average of 5.1 years (SD \pm 3.1). A male preponderance was noted in 72% of the cases. No medical history of allergic rhinitis or sinusitis has been found. Besides, our patients weren't suffering from any immunodeficiency disorders. The main complaints reported were: rhinorrhea (16/18; 88.9%), fever (10/18; 55.6%) headache (2/18; 11.1%) and ophthalmic signs (14/18; 77.8%). The duration of symptoms prior to presentation was inferior to 7 d in 71.4% of the cases (2-10 d). Prior antibiotic treatment has been prescribed for 6 patients (33%). The results of the clinical examination are resumed in Table 1.

Brain and sinus CT-scan have been practiced for all our patients and brain magnetic resonance imaging (MRI) in 2 cases. The most frequent localization of sinusitis was ethmoid (77%), ethmoid + maxillary (33%) and the sphenoid sinus (11%). According to Chandler's classification, orbital complications were divided into: class II (7 cases; 38%), class III (4 cases; 22%), class IV (2 cases; 11%) and class V (1 case; 5%) (Table 2) (Figures 1-4). In case of sphenoiditis, different degrees of bony lysis have been noted (Figures 5 and 6).

Laboratory tests found leukocytosis in 9 cases and elevation of the C-reactive-protein in 6 cases (33%).

Medical treatment was based on intravenous antibiotherapy for all the patients. Intravenous heparin at curative dose has been administrated in the case of the cavernous sinus thrombosis for 3 wk. A complementary surgical intervention was indicated in 8 cases. All medical and surgical treatments are detailed in Table 3.

The outcomes were almost good in cases of class II and III ethmoiditis after medical or medicosurgical treatment. The clinical signs (fever, pain, swelling) disappeared in a period of 5 to 10 d. No ophthalmic complications have been noted among this group of patients.

For class IV "Chandler" complications (orbital abscess = 2 patients) and for the cavernous sinus thrombosis (1 case), severe ophthalmic sequelae have been diagnosed: ophthalmoplegia, loss of vision and 3rd, V1 and V2 cranial nerve palsies.

The 2 cases of sphenoiditis have benefited from endoscopic sphenoidotomy with intravenous antibiotherapy (Figure 7). The computed tomography (CT)-scan showed during the follow-up, a notable regression of the inflammatory lesions (Figures 5 and 6). The regression of the lytic bone lesions have taken several months but without notable clinical signs.

Table 1 Data of the clinical examination

	Findings	No. of cases	%
Nasal endoscopy	Congestion and pus at the middle meatus	5	27.78
	Purulent discharge at the nasopharynx	2	11.12
	Polypoid lesion	1	5.56
Ophthalmic exam	Congestion of the upper eyelid	8	44.45
	Swelling and/or redness at the medial angle of the eye	6	33.34
	limitation in the medial ocular movement	5	27.78
Signs of meningeal irritation		2	11.12

Table 2 Findings of the imaging exams

		No. of cases	%
Sites of the lesions	Ethmoid	14	77.78
	Ethmoid + maxillary	6	33.34
	Sphenoid	2	11.12
Lytic bone lesions		2	11.12
Orbital complications	Class II: orbital cellulitis	7	38.89
	Class III: periosteal abscess	4	22.23
	Class IV: orbital abscess	2	11.12
	Class V: cavernous sinus thrombosis	1	5.56
Meningeal reaction		2	11.12

Table 3 Therapeutic procedures

		No. of cases	%
Medical treatment	Cefotaxime (100 mg/kg/j) + fosfomycin (100 mg/kg/j)	14	77.78
	Cefotaxime + fosfomycin + amikacin (15 mg/kg/j)	2	11.12
	Cefotaxime (200 mg/kg/j) + vancomycin (60 mg/kg/j)	2	11.12
	Intravenous heparin (Bolus 50 units/kg, 2 units/kg per hour)	1	5.56
Surgery	External medial orbitotomy	4	22.23
	Endonasal orbitotomy	2	11.12
	Sphenoidotomy	2	11.12

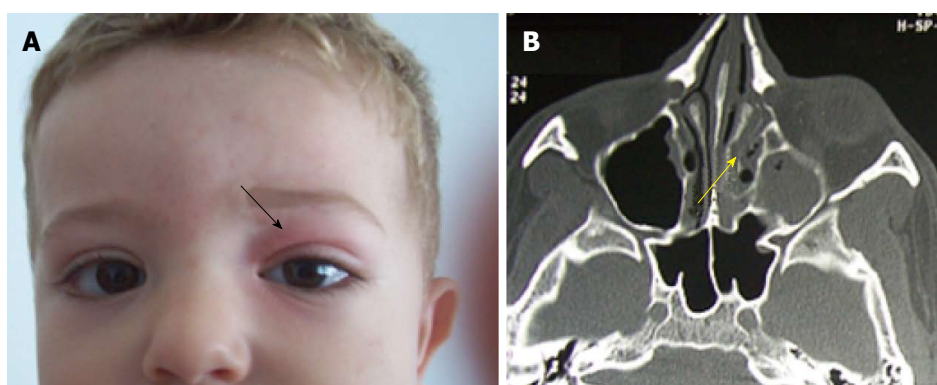


Figure 1 Swelling and redness at the level of the left upper eyelid (A); the computed tomography-scan shows a fulfilling of the ethmoid cells and the left maxillary sinus (B).

After an average follow-up period of 2.5 years, 3 of our patients were lost. The ophthalmic sequelae noted in 3 cases (16%) were permanent and caused important functional and social problems. A favourable outcome has been noted in the rest of our patients.

DISCUSSION

Rhinosinusitis is one of the most common diseases

during childhood, accounting for nearly a quarter of all pediatric antibiotic prescriptions^[6]. It is most of the time efficiently treated with nasal decongestants with or without antibiotics. Recovery is obtained in most cases of acute rhinosinusitis, however, between 5% and 10% will go on to develop complications^[7]. Complicated rhinosinusitis has been described as the adverse progression of bacterial infection beyond the paranasal sinuses. Complications are divided into orbital, intracranial or mixed manifestations^[1,8].

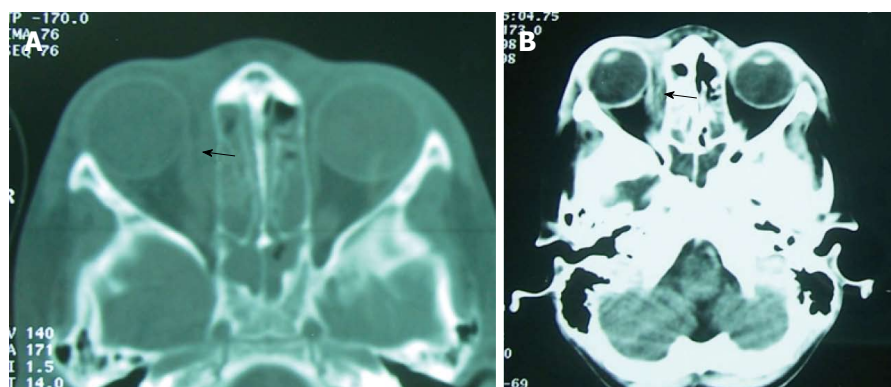


Figure 2 Bilateral ethmoiditis (A and B) complicated by a subperiosteal abscess (arrows).

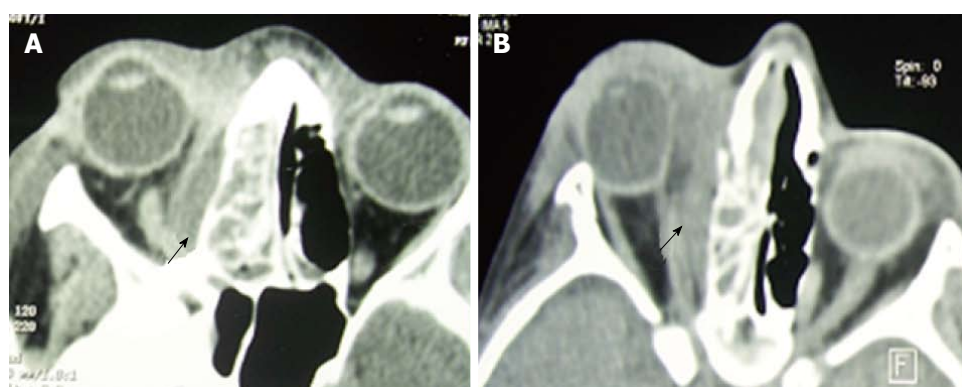


Figure 3 Right ethmoiditis complicated by intraorbital abscess (A and B).

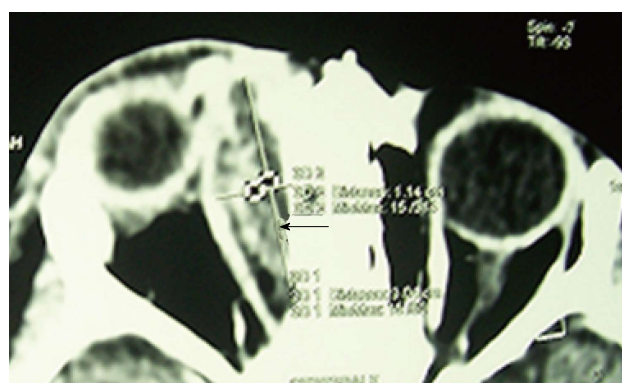


Figure 4 Intraorbital abscess displacing the ocular globe anteriorly and laterally (arrow).

Sinus infection can spread through either direct, hematogenous, or by retrograde extension, along the valve-less diploic veins^[4,5]. Orbital complications are more common than intracranial ones and are often classified using the criteria of Chandler^[1,4,5]. Briefly, class I is "preseptal cellulitis", class II is "orbital cellulitis", class III is "subperiosteal abscess", class IV is "orbital abscess" and class V is "cavernous sinus thrombosis"^[9]. Intracranial complications of rhinosinusitis consist of meningitis, subdural empyema, epidural and cerebral abscess, and venous sinus thrombosis^[5]. The most common of this type of complications is different from one series to another. It was subdural empyema in Yew Kwang Ong^[5], Skelton^[10] and Jones's series^[11], however for Clayman^[12] and Giannoni^[13] it was

cerebral abscess. Orbital complications occur most commonly in younger children^[4]. However, the extension to intracranial structures is rare in children under the age of 7 and occurs typically in young adolescents^[5,12]. Our finding of male preponderance in orbito-cranial complications of sinusitis is in keeping with literature^[1,14,15]. The clinical presentation can be fever, headache, vomiting, chemosis, eye pain, proptosis, cranial nerve palsy, seizures and focal neurological deficit^[4]. The symptoms are non-specific initially and the diagnosis may be overlooked for several reasons^[1,5]. In most cases of complicated rhinosinusitis in children, previous sinus disease or history of allergic rhinitis is often not found. For Jones^[11] this kind of medical history was present in only 10% of his patients and Yew Kwang Ong noted this in 28% of cases^[5]. Many patients consult a family physician and take prior antibiotic treatment. This prescription modifies the disease presentation and masks the diagnosis. However behind a swollen eye, a proptosis, or an impaired function of the extraocular muscles, an orbital extension must be suspected^[4]. A persistent headache, especially beyond one week, is the most consistent symptom of intracranial involvement^[16,17].

The average duration of illness from presentation is 3-5 d^[14]. In our series it was inferior to 7 d in 71.4% of cases.

Orbital complications are most commonly secondary to ethmoid sinus involvement and intracranial injury associated with frontal sinusitis^[1].

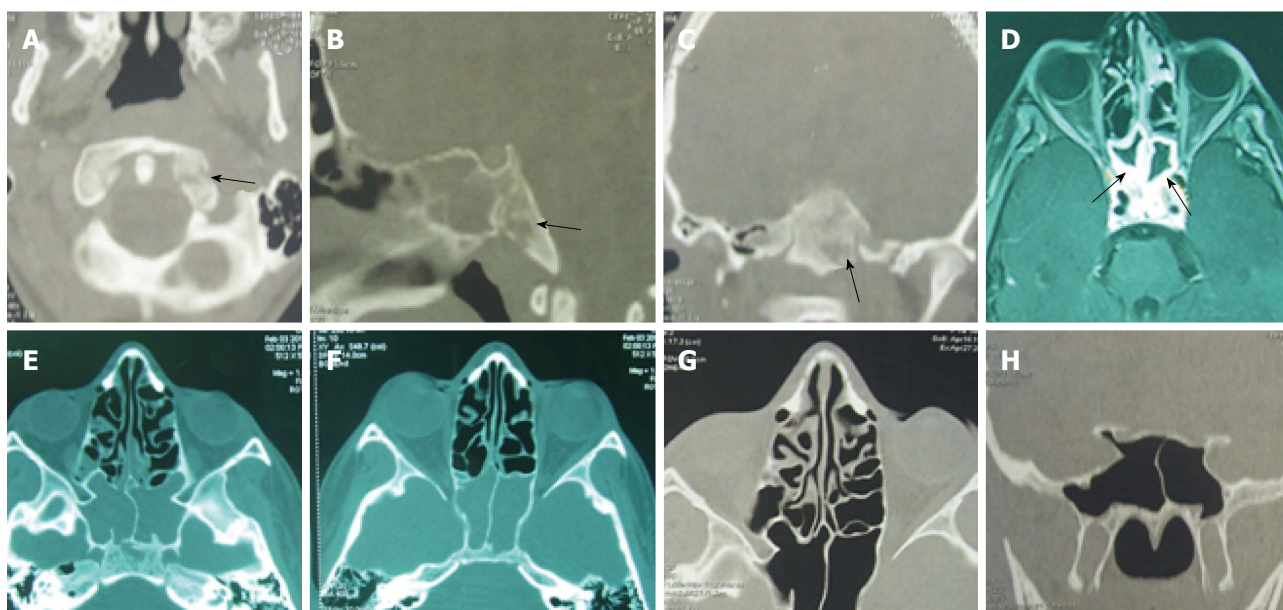


Figure 5 Sphenoiditis with bony lysis and a notable regression of the inflammatory lesions in a 9-year-old child. Sphenoiditis in a 9-year-old child with bony lysis in computed tomography-scan (A, B and C) and magnetic resonance imaging (D). The regression of the inflammatory lesions after sphenoidotomy and antibiotherapy was partial after 3 wk (E and F) and complete after 5 wk (G and H).

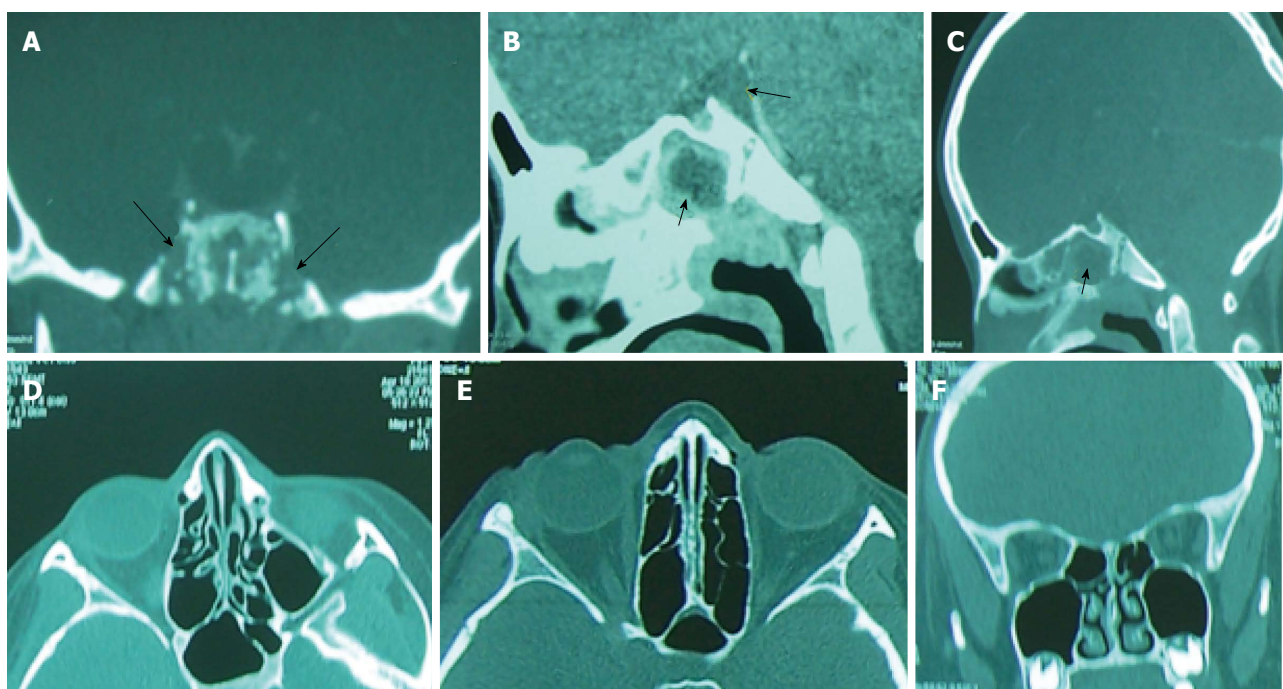


Figure 6 Sphenoiditis with meningitis in an 11-year-old child treated by sphenoidotomy and intravenous antibiotherapy. Note the lytic lesions of the sphenoid bone (A-C). The computed tomography-scan, performed after 1 mo shows a complete regression of the inflammatory signs (D-F).

The diagnosis of rhinosinusitis is clinical^[18]. However, when orbital or intracranial complications are suspected, urgent radiological examination is recommended. Contrast-enhanced CT with coronal and axial slices is the initial imaging modality of choice^[5,19]. It should be performed, in pediatric patients, with the lowest radiation dose possible allowing a good diagnostic study^[19]. Coronal cuts are useful for studying the ostiomeatal unit and the relations between orbit and brain^[20]. The contribution

of CT is also considerable in delineating bony anatomy for a potential endoscopic sinus surgery^[4,20]. MRI is more efficient in soft tissue analysis but its high cost and limited accessibility hinder its routine use^[5,20]. In our series it was practiced in only 2 cases. Yew Kwang Ong recommends to practice simultaneously sinus and brain CT when intracranial complications are suspected, and to repeat it if a complication is strongly suspected^[5]. In Skelton's series 50% of children with subdural empyemas had

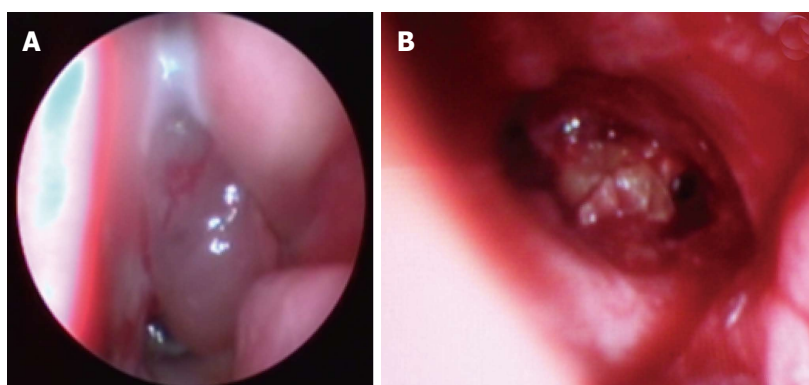


Figure 7 Sphenoiditis have benefited from endoscopic sphenoidotomy with intravenous antibiotherapy. Nasal endoscopy showing an inflammatory polyp with purulent secretions coming from the right sphenoid sinus (A). The sphenoidotomy revealed an inflammatory granuloma into the sphenoid cavity (B).

normal scans initially^[10].

Bacteriological analysis is required to adjust antibiotic treatment. Polymicrobial or culture negative specimens are not so rare, sterile samples are obtained in 25%-30% of cases^[1,4]. In recent years, there is an increase in reports of fungal invasion accompanying rhinosinusitis. These forms were connected with several promoting factors as diabetes, steroidotherapy and immunosuppression^[21,22].

A complicated rhinosinusitis is a therapeutic emergency. Once the diagnosis evoked, an adequate treatment should be started. The therapeutic management is divided into medical and surgical treatment. Intravenous broad-spectrum antibiotics are recommended until definitive culture and sensitivity results are obtained^[4,5,20]. The most common germs are *Streptococcus* species, *Anaerobes* and *Staphylococcus Aureus*^[4,5,20]. When intracranial complication is associated, used antibiotics must have a good blood brain barrier penetration and should be continued for 4 to 8 wk^[5]. The surgical treatment comprises endoscopic sinus surgery, orbital decompression and neurosurgical intervention^[4]. It is most commonly indicated when medical therapeutic fails to control the infection^[20]. However in cases with intracranial complications, it is recommended to initially operate^[1,4,5]. In Yew Kwang Ong's series^[5], repeated drainage was necessary to eradicate the intracranial infection. Kurt Denton Schlemmer^[1] prefers external approach to the endoscopic one and indicates surgery in the following cases: proptosis with orbital complications, intracranial complications, sub-periosteal and pre-septal collections, and cellulitis that does not respond to medical management within 24-48 h. In our series, surgery was indicated in 8 cases: external medial orbitotomy (4 cases), endonasal orbitotomy (2 cases) and sphenoidotomy (2 cases) because of a non response to medical treatment, for orbital collections or for associated meningitis.

In complicated acute rhinosinusitis, a follow-up for up to one year is recommended^[7]. Thanks to progress in diagnostic tools and therapeutic management, mortality has decreased significantly in developing countries to about 4%-6%^[1,4]. However sequelae

such as visual deficits or loss, cranial nerve palsies, epilepsy, hemiparesis, seizure, speech and cognitive impairment can unfortunately persist^[4,5].

In conclusion, rhinosinusitis is one of the most common infections in the pediatric population but complicated cases are rare. Orbito-cranial involvement carries high morbidity and changes radically the prognosis. It requires a management by a multi-speciality team including pediatrician, radiologist, otolaryngologist, neurosurgeon and ophthalmologist. Good outcomes can be reached given an early diagnosis, based on complete clinical examination and urgent radiological imaging, and aggressive medical and surgical treatments.

COMMENTS

Background

Contrary to adults, acute rhinosinusitis are more frequent in children, but with a high risk of severe and potentially life threatening complications.

Research frontiers

The study describes the clinical, paraclinical and therapeutic management of rhinosinusitis in pediatric population.

Innovations and breakthroughs

The treatment of rhinosinusitis in children must be very appropriate to the locoregional extension of the disease and guided by endoscopic and radiological findings.

Applications

The authors must retain that pediatric rhinosinusitis is a serious disease requiring a precise evaluation of the locoregional extension through radiological exams, intravenous antibiotherapy and, if necessary, a complementary surgical procedures.

Terminology

SD: Standard deviation; CT: Computed tomography; MRI: Magnetic resonance imaging.

Peer-review

It was a well written study.

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