The osseous external auditory canal
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General discussion and conclusions
INTRODUCTION

This thesis focused on three themes:

1. Surgical treatment of acquired diseases of the OEAC. In order to define the optimal treatment we deemed it necessary to have a better understanding to which extent surgery is effective in alleviating the disease. Furthermore we investigated which aspects are relevant to address during surgery in order to be successful.

2. Shape of the OEAC. Although many authors have suggested that the shape of the OEAC can be a contributing factor in many cases of refractory inflammatory disease and in troublesome cavities we tried to supply clinical evidence for this prior unsubstantiated statement. We investigated the pretymppanic recess area as we hypothesize this region to be an important factor for chronicity.

3. Perceived sound quality after surgery addressing the OEAC. When altering the shape of the OEAC it stands to reason that the resonance function will change. Although this phenomenon has been described in the literature, it remains unknown to which extent this change in resonance function is perceived and whether it is clinically relevant. We investigated this phenomenon with more detail.

SURGICAL CONSIDERATIONS.

When extensive conservative therapy of acquired inflammatory disease of the OEAC is exhausted and a state of refractory pathology has arisen surgical intervention is indicated (1). Although Proud (2) recommended to resort to surgery after failure of three weeks of conservative treatment for external otitis, personal experience and experiences described in literature (3) show that many cases have a duration of many years prior to surgical intervention. The optimal moment of surgical intervention is unknown. Perhaps the lack of a clear consensus how to define when a refractory state is present is responsible for such delay? Or could it be that conservative treatment is continued for a longer period due to patient and doctor related reluctance towards surgical intervention?

Varying intervals have been described ranging from 72 hours (4), 2 weeks (5), 3 months (6) to as much as at least one year (7) before one states a chronic or refractory state is present. The Dutch national guideline (8) classifies otitis externa to be persistent after 3 weeks but chronic after 3 months. It states that for chronic otitis externa surgery is the primary treatment. This definition does not cover recurrent otitis externa which could be regarded as a type of refractory state as well.

Although for some diseases (for instance otitis externa) various definitions have been proposed to determine chronicity (see above) it has proven to be very difficult to implement them. This is due to the multi-aetiological origin and varying clinical presentation of these diagnoses. For instance, if a temporal resolution of the inflammation is achieved when does one regard a recurrence or exacerbation to be therapy resistant or chronic. If the middle ear is affected as well or a tympanic membrane perforation is present one cannot define the disease as
being solely located in the OEAC and a different strategy must be followed and therefore should not be seen as a sole otitis externa.

We would suggest to define acute otitis externa to have an acute start and a resolution within three weeks (with adequate treatment) and without a recurrence within one year. No concomitant ear disease such as tympanic membrane perforation, granular myringitis or middle ear effusion or infection should be present. Chronic otitis externa could therefore be defined to have a duration longer than three weeks or exacerbations and remissions within a one year period. Such a definition would mirror the definition of acute and chronic rhinosinusitis as was described in the European Position paper on Sinusitis and Nasal Polyps (EPOS document) (9).

Beside the abovementioned problems of disease definition and disease complexity one could argue that an increased reluctance towards a surgical therapy is present as one can assume it to be accompanied by more risks than the conservative therapy. We meticulously described adverse events and or complications when analyzing our results (Chapter 2.2, 2.3) in order to give insight into which sequelae are related to surgery of the OEAC (safety-aspect). The literature review (Chapter 2.1) and our own series (Chapter 2.2) show that complication rates vary but are a definite factor to consider when proposing surgery and that transient problems occur in a relatively high percentage of patients. This shows that surgery indeed is accompanied with a significant burden to the patient (10). Yet, when considering long term complications, unwanted and adverse effects, this percentage decreases significantly. (Chapter 2.1, 2.3) and the balance shifts in favour of surgical intervention if one assumes that no single form of conservative therapy actually will resolve the refractory OEAC disease (11). As the large majority does not suffer major and/or persistent sequelae of surgical intervention one could postulate that the risks of surgical interventions does not weigh up to its potential benefit.

Still some other considerations against surgical treatment can be taken into consideration. First surgical intervention has shown to vary in efficacy between different diseases of the OEAC (Chapter 2.1). For instance we showed (Chapter 2.2) that AMCF has less favorable surgical results compared to COE, exostosis and troublesome cavity groups. This group has a higher recurrence rate and therefore a surgical treatment of AMCF should be deemed less effective. Moreover, if a dry symptom free blind sac is present and only hearing is impaired one could argue that if hearing rehabilitation is successful (either with bone conduction devises or regular hearing aids) no indication remains for surgery at all.

An important aspect is the perceived burden of the disease by the patient. If a patient subjectively experiences limited burden of the disease and if he/she is satisfied with a conservative therapy one could argue that continuation of that treatment is preferred.

In conclusion, the literature review (in Chapter 2.1) and our own data (Chapters 2.2 and 2.3) show that surgery of the OEAC would lead to very high percentages of disease resolution in a wide variety of ear canal related diseases (including non-inflammatory diseases). Considering all above mentioned arguments and contemplations it seems that with current knowledge one could conclude surgical treatment to be the primary treatment for acquired chronic symptomatic OEAC disease and should be propagated as such.
We are not aware of studies in the literature comparing surgical and conservative treatment in patients with OEAC disease. We are in need of multicenter randomized clinical trials including sufficiently high patient numbers to enable multivariate analysis. Because of the patient number limitations, a careful consideration of the relevant determinants seems mandatory.

As second best a well described case series including all potentially relevant determinants could be used to strengthen recommendations regarding the indications of surgical intervention in acquired OEAC diseases.

When one has decided to perform surgery some practical considerations arise regarding the procedure to perform. The literature review (Chapter 2.1) showed that a wide variety of surgical techniques and underlying philosophies are currently propagated.

Regarding the epithelial lining, two possible contradictory philosophies can be distinguished. One advocating the use of skin grafts and another using secondary healing of the epithelial lining. Although evidence is lacking regarding both positions those in favour of grafting state that faster healing, the need for complete removal of diseased skin and lesser risk of restenosis are acquired when grafts are used. Those in favour of secondary healing state that the unique properties of the skin lining of the OAEC (migratory epithelium, wax production, the absence of subcutaneous tissue and direct adherence to the periosteum) are essential for a normal ear environment and that grafting introduces the need for life-long clinical cleansing and a higher recurrence rate. Further randomized prospective studies could be conducted comparing healing and functional results between a graft and secondary healing group.

Our techniques of canalplasty minimizes skin loss and showed that skin grafting is not obligatory which is in concordance with other observations (1, 12) (Chapter 2.2 and 2.3). Still we have used grafts in revision surgery and in cases of extensive shortage of epithelial lining. We found that these ears healed satisfactory but indeed needed life-long clinical cleansing as migratory properties were no longer present. We therefore think that secondary healing should be preferred and grafting only used in selected cases. We have been very reluctant to use the epithelial lining of the non-affected ear as a possible graft as this could potentially lead to bilateral problems if healing fails.

A promising development has been described regarding in vitro ear canal tissue growth (13). Although no clinical results have been described for the usage of such grafts it could potentially have the benefits of both philosophies. Functional epithelium could be used as a ‘transplant’ in order to prevent denuded bone surfaces. This would hypothetically lead to faster healing, less intensive post-operative care and possibly minimize ‘blunting’ of the OEAC while not compromising the unique properties of the skin of the OEAC.

Also, when considering which area to address and amount of bone to remove during surgery no consensus is currently present. The reviewed techniques (Chapter 2.1) showed that some advocate minimal bone removal of the posterior canal wall while others advocate extensive circular removal of bone especially of the anterior wall. No rationale is given for such decision. We will describe the role of the shape of the OEAC more extensively in the following paragraph but our work has shown that certain areas should be addressed in certain diseases. How much bone has to be removed to acquire a patent and disease free ear canal is completely unknown.
at the moment. Such knowledge will make surgical intervention even more successful and safe as minimal removal of bone limits potential complications and healing time. This knowledge should be expanded with future research.

A canalplasty only addresses the OEAC and therefore is insufficient when the cartilaginous EAC is involved as well. Some of the techniques described in Chapter 2.1 do not enable a simultaneous meatoplasty as the skinflaps do not leave any space for suturing in a ‘pessimistic’ flap management strategy (14) or because the lack of exposition of the subcutaneous layers due to the approach (15) or the need for post-operative use of stents (16,17). This results in a forced staging of surgery, even when one could foresee a meatoplasty to be needed. One can combine our described canalplasty technique with a myringoplasty, meatoplasty, mastoidectomy, and reconstruction of the posterior canal wall (Chapters 2.2 and 2.3) making the technique very versatile. At the moment personal preference or experience is used to decide if a simultaneous meatoplasty is necessary as no evidence based recommendations are present. Future research regarding the interaction between OEAC and the cartilaginous EAC is needed to base the choice of surgical intervention and the type of surgical intervention on clearly defined evidence based parameters such as described below.

THE IMPLICATIONS OF THE SHAPE OF THE OEAC.

We showed that the shape of the OEAC, especially the DPTR plays a role in chronic inflammatory OEAC disease (chapter 3.1 and 3.2). We mainly evaluated the bony aspects of the ear canal but we do realize that the cartilaginous part may play and important role that remains under exposed in this work. Although we know that a meatoplasty can influence chronic inflammatory OEAC disease significantly (18), we are in need of better methods to quantify the cartilaginous part of the EAC (CEAC) as it remains unknown which aspects of the shape of the CEAC contribute to the effect of a meatoplasty. Evaluation of our surgical cases suggests that the amount of occlusion due to the isthmus of this CEAC might be a crucial factor. Analysis of several dimensions, for instance the distance between conchal cartilage and tragal cartilage or the percentual difference in the cartilaginous circumference in regard to the OEAC circumference, could be used as parameters to investigate the relationship and to formulate a hypothesis that can be tested.

Although we showed that the pretympanic recess plays a role, in COE and draining cavities, we cannot disregard the fact that not all dimensions of this three dimensional complex anatomical shape have been investigated. We had no need to evaluate these parameters in order to investigate our primary hypothesis (being that the PTR was an important part in the chronic inflammatory EAC) and therefore did not include them. Furthermore the factor volume (a better quantifiable factor through tympanometry) is very interesting but unfortunately this information was lacking in our retrospective cohort. So further research could include aspects like the length, width, volume and their mutual relationships as contributing factors. Furthermore a classification should be based on evidence supporting its definition of subsets. With so little evidence presently available (i.e. only the DPTR for COE and draining cavities
at the moment) and so many unknown determinants it is crucial that further research defines the relevant factors before constructing a classification for inflammatory ear canal disease.

Based on theoretical grounds, we argued that the DPTR plays a role in the chronicity of inflammatory disease due to the inability of adequate (self) cleansing of the ear canal. Furthermore we postulated that a possible intertriginous eczema would occur due to a smaller angle in case of deeper PTR, allowing easier skin-to-skin contact (Chapter 3.1). This assumption provides a logical explanation for the fact that (at least a part of) the shape of the OEAC is a contributing factor to chronic inflammatory ear canal disease. Perhaps the OEAC shape influences other aspects like humidity and pH values in the external ear canal as well. Martinez Devesa (19) showed that also the pH is correlated with possible chronicity but immediately stated that more research was needed for conclusive statements. Gray (20) found similar results regarding humidity. A ‘smaller’ ear canal volume could give rise to an increased humidity and disruption of ear wax homeostasis due to accumulation, which could alter pH values significantly. A multifactorial analysis including shape, pH, humidity, volume and disease occurrence could elucidate the pathogenesis of chronic inflammatory disease of the ear canal. Such further knowledge will facilitate the development of precision medicine in the treatment of OEAC.

We have demonstrated that the shape plays a role in inflammatory disease (Chapters 3.1 and 3.2). Some other acquired diseases of the OEAC (such as AMCF, Canal stenosis, troublesome cavities and to some extent exostosis) could be regarded as closely related to inflammatory processes. Yet other acquired diseases (such as malignancies, benign tumors, cholesteatoma, exostosis and osteomata) are not associated with inflammation. Although it is more difficult to hypothesize how the shape would be a part of the pathogenesis of such diseases perhaps the shape would influence treatment outcome or affect symptoms. Further research regarding the non-inflammatory diseases and their relation to the shape could be interesting and useful.

Although not within the scope of this thesis the shape of the OEAC would logically play a role in treatment of other ear diseases as well. As the ear canal is an easy approach to the tympanic membrane, the middle ear and the cochlea it is reasonable to assume that surgical treatment outcomes of diseases of the middle ear and tympanic membrane will be influenced by the shape due to the amount of surgical exposure. Currently an interesting field of endoscopic ear surgery is developed in which the ear canal is used in treating a wide variety of middle ear disease (21). One could easily postulate that outcome of such surgery is related to ear canal shape.

Unpublished clinical observations made by our group suggest that the shape of the OEAC influences the width of the facial recess making a pre-operative assessment of the surgical approach possible, for instance in case of a cochlear implantation. One could decide whether a transmastoid posterior tympanotomy or drilling of the cochleotomy (or drilling of the crista antefenestrum in a round window implantation) is preferred or that a transcanal drilling would be easier. As results have shown to be similar (22) one could choose for an approach that minimizes risks for chorda tympani and facial nerve and maximizes exposure of the target organ.

We also suspect that some radical cavities are the result of lowering the posterior border of the ear canal in order to optimize exposure of the anterior part of the middle ear space. This
would not be necessary if a canalplasty is performed or if an endoscope is used. One could pre-operatively assess the problem of limited visibility as it is directly related to a larger ACPTTR. Our findings (Chapter 3.2) show that this abovementioned suspicion is definitely a possible factor in choosing for a radical cavity. An approach to further research this hypothesis is to prospectively assess the ACPTTR and investigate whether a conversion to a conservative radical mastoidectomy was needed / deemed necessary (with the assumption that a CWU approach is indicated beforehand). Or if an endoscope was used to assess the anterior middle ear space.

We feel that the success rate of tympanic membrane closure (especially in anterior and (sub) total perforations) increases if more exposure is present. It is commonly accepted that the residual disease rate of cholesteatoma (especially tympanic cholesteatoma) is lower when the ear canal shape allows more exposure (23). The drop in residual cholesteatoma when an endoscope is used to inspect the difficult regions of the middle ear cavity also strengthens this statement (24).

Another aspect in the treatment decision process of patients with acquired OEAC disease, but also in the treatment of hearing loss in general is hearing aid rehabilitation. We know that some patients do not tolerate the occlusion of the OEAC due to wearing acoustical hearing aids. Some even develop OE which in turn can become chronic. Although sound presentation via other modalities as vibration through Bone-Conduction Devices (BCD) has shown to be very effective in hearing rehabilitation in such cases (25,26), it has been generally accepted that acoustical hearing aids have distinct advantages like a higher maximum output and more effective gain than BCD’s. Consequently, acoustical hearing aids are the preferred option when tolerated.

Some of our study cohort patients had COE due to occlusion and the results of canalplasty are partly comprised with patients wearing conventional hearing aids (Chapter 2.2). It would be very interesting to perform sub analysis whether a canalplasty was successful in altering the shape in such a manner that hearing aids could be tolerated better post-operatively. Special attention is required in case of troublesome cavities. As hearing results are generally poorer in cavities (27) and mixed hearing loss is often present with a contralateral better hearing ear, conventional hearing aids are often a better choice than the abovementioned alternative of BCD’s. Unfortunately most cavities do not respond well to occlusion and often hearing aids are not tolerated (28). Clearly further research, preferably in a prospective randomized manner, is warranted to determine whether and to which extent a canalplasty can increase the tolerance for acoustic hearing aids in complicated cases. Multifactorial analysis in individuals that had unsuccessful hearing aid rehabilitation due to radical cavity, excessive sweating, aural fullness, sound distortion and inflammatory response could elucidate to which extent anatomical dimensions, and the humidity and pH play a role.

An anatomical model that allows us to alter the shape factor independently while leaving other factors unchanged could supply even more insight how altering the shape influences the final outcome. The predictions of such a model should be tested in clinical studies to evaluate them. These results could lead to a better prognosis of the post-operative results and to an improved evaluation of operative techniques.
PERCEIVED SOUND QUALITY CHANGES AND ITS IMPLICATIONS

We have shown that we can measure and simulate at least some of the alterations that are the result of the shape of the OEAC (Chapter 4.1). Significant effects can be achieved in perceived sound quality with surgery altering the shape of the OEAC (Chapter 4.2). Further studies are necessary to implement this knowledge in daily clinical practice and in pre-operative counseling when considering a canalplasty or canal reconstruction. We still face an extensive lack of knowledge, so these studies should at least include whether habituation/acclimatization takes place. Certain aspects of the sound perceived as more or less distorted and hearing rehabilitation can be used to compensate this perceived effect.

Acclimatization can be investigated by using patients as their own controls. Do patients still regard the simulated cavity condition presented to their normal ear to have a poorer sound quality? And can we reach better perception if we present ‘normal’ resonance conditions to the cavity ear? If acclimatization does not occur one can imagine that the altered resonance state can be compensated using hearing aids using filtering that takes into account the acoustical effects of the surgical interventions. If this is a viable alternative remains to be investigated. If similar results in perceived sound could be attained with hearing aids this could reduce the need for surgical intervention.

We showed how the real ear unaided response curves differed from the normal reference curve for several conditions (Chapter 4.1 and 4.2). Not very surprisingly we observed a less ‘natural’ sound or poorer ‘quality’ when these curves were more deviant from “normal”. This raises the question whether this difference could be used as an objective measure for subjectively perceived sound quality. Another question would be which aspect of the real ear unaided response is the most critical factor for the subjective sound quality? Is the gain loss more important when present in certain frequencies? Does an increase in gain lead to more or lesser distortion when evaluated subjectively? Further research should be performed to answer these questions. Such knowledge can be used in research regarding surgical changes made to the OEAC eliminating the need for normal hearing cohort analysis of outcomes. Perhaps mathematical models can be used to predict which adjustment of real ear gain would lead to positive of negative subjective effects. And the usage of the real ear unaided response and gain could be used to predict pre-operatively which effect can be achieved when certain types of surgical interventions are performed.

For the field of hearing rehabilitation, it will be important to understand how the resonance function leads to a perceptible effect. In children hearing aid rehabilitation often takes such effects into account (29) as the OEAC is not yet full grown. Yet when cavities or post-operative changes are present such effects are often ignored when applying a hearing aid. Whether hearing rehabilitation will improve when taking in account the effects of the alterations in shape of the OEAC after surgery can easily be explored in a randomized prospective trail using subjective outcome tools as primary outcome measurements.
Apart from our primary interest in OEAC we appreciate that other factors are important in patients undergoing ear surgery. Ideally, acoustic modeling including middle ear function (or dysfunction) and cochlear function could pre-operatively evaluate the acoustical effects for a broad scope of otological intervention. In addition to modeling we are in need of further developed patient reported outcome measures that can be used to personalize treatment counseling.

THE CONTRIBUTION OF THIS WORK.

This thesis indicated that an evidence-based approach was lacking in OEAC surgery. The work in this thesis tried to supply evidence and specify the rationale used regarding the indication and technique for OEAC surgery. Also we indicated areas where additional research is needed. Our proposal to standardize reporting of surgical outcome will hopefully lead to better comparisons between the techniques and to the identification of the key elements of surgery that influence outcome. We supplied evidence for the benefit of the canalplasty techniques we used in acquired OEAC disease and radical cavities. Because enlarging or widening the OAEC leads to a deterioration of perceived sound quality, we advocate a more individually tailored approach to OEAC surgery based on a systematic analysis of the contributing components. We showed the reduction of the depth of the pre-tympanic recess is an important component in OAEC surgery while the anterior curvature can be left untouched. Less extensive surgery can also lead to faster healing, less pain and to better care. We also suggest to perform earlier surgery in persistent otitis externa (duration of more than 3 weeks) especially when the DPTR is larger than 2.7 mm and other diagnoses have been considered. In conclusion this thesis has increased our understanding and knowledge regarding the surgical approach and the effects of shape and sound of the OEAC. The ultimate aim is to enable evidence based surgery. Further research as indicated in this discussion will help us to reach that goal.
REFERENCES


