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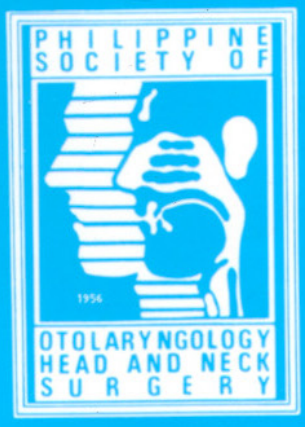
The Philippine Journal of Oto Rhino Laryngology Head & Neck Surgery



Editorial Policy

The Philippine Journal of Otorhinolaryngology-Head and Neck Surgery is committed to the publication of scientific work on the specialty. It seeks to disseminate timely and relevant information to improve practice and to inform health policy. It provides a forum for the continuous exchange of views among health professionals concerned with the provision of quality otolaryngologic care.

The Journal is peer-reviewed, published quarterly and distributed to Society members as well as to other relevant consumers.

Instructions to Authors

The Journal follows the "Uniform Requirements for Manuscripts Submitted to Biomedical Journals" as agreed upon by the International Committee of Medical Journal Editors.

Manuscripts should not have been published or being considered for publication elsewhere. Exceptional articles are accepted for secondary publication if authors can provide proof of having received permission from the editor of the primary version.

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4. Include an abstract of not more than 150 words with the following parts: objectives, setting, design, participants, outcome measures, methods (interventions, if any), results, conclusions.
5. Use Systeme International (SI) units throughout the manuscript, provide metric equivalents within parentheses as needed.
6. Use generic names of drugs, unless the trade name is relevant to the study.
7. Do not use abbreviations in the title and abstract and limit their use in the text.

Tables/Graphs

Type each table on a separate standard-sized white bond paper. Title each and number them in order of their citation in the text. Write all legends and footnotes completely.

Illustrations

Submit good-quality unmounted glossy prints. Number illustrations in order of citation. Indicate views for radiographs, magnification and stain for photomicrographs.

Acknowledge fully all illustrations and tables taken from other publications and submit written permission to reprint from the original publishers.

References

Number references in the order of citation in the text; do not alphabetize. In text, tables and legends, identify a reference with superscript Arabic numerals. In listing references, follow the examples given by the "Uniform Requirements".

Example: Garland C, Barrett-Connor E, Suarez L, et al. Isolated systolic hypertension and mortality after age 60 years: A prospective population-based study. Am J Epidemio 1983; 118:365-376.

Style

Write in simple and clear sentences. Express ideas as logically and succinctly as possible. Refer to Strunk and White's "The Elements of Style" (3rd ed.) for guidance.

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The Philippine Journal of OtoRhinoLaryngology - Head and Neck Surgery

Editorial

The Guidelines: What do we do with them now?

In 1998, our Society developed a set of clinical practice guidelines for thediagnosis and management of otitis media with effusion (OME) in children, chronic suppurative otitis media (CSOM) in adults, chronic rhinitis in adults, acute and chronic sinusitis in adults, acute and chronic tonsillitis and chronic obstructive adenoiditis. Not much has happened since then by way of active dissemination. No one knows how many ENT specialists are still aware of the recommendations contained in the guidelines. More uncertain still is the extent to which the guidelines have been incorporated into routine clinical practice or the training of new ENTs. We have only ourselves to blame.

The guidelines are in a varying state of finish. OME seems the most polished while CSOM needs to systematically incorporate clinical opinion. The tonsillitis guidelines definitely can be improved. The rhinitis and sinusitis guidelines need a more systematic collection and synthesis of evidence. What is the best and most efficient way of improving their quality?

The best way to update the guidelines would be to sytematically search and appraise the published and unpublished medical literature using explicit criteria for including and evaluating studies, synthesize their results and use them in revising and augmenting the evidence-based recommendations from which the guidelines were developed. For example, while the 1996 recommendation for managing OME is either to treat with antibiotics immediately or to wait for 6 weeks for the effusion to resolve on its own, new trials and economic analyses might suggest the superiority of immediate antibiotic therapy considering the prevalence of OME in our setting and the costs of persistent effusion.

For those recommendations which are based on poor quality evidence, expert opinion, patients' preferences and socio-ethical values must be incorporated in a more explicit and systematic fashion.

Guidelines never implement themselves. If we are serious in developing them, we should be doubly serious in making sure they get implemented.

But before we disseminate the guidelines, we must be clear about the target users so that we can plan how best to reach them. ENT specialists may be our initial targets but if we hope to benefit the majority of patients with ENT complaints who will never get to reach us, we should be setting our sights to family physicians, pediatricians and internists. This is just the beginning because guidelines typically have multiple users and uses. They can be used by nurses and other health professionals, provider organizations, such as hospitals and clinics, purchasers of health care, such as health maintenance organizations and the, government, consumers of health care, regulators of health care, accrediting and certifying agencies, researchers and health policy analysts, and interested parties, such as attorneys, journalists, politicians, and other stakeholders.

The rich literature on guideline dissemination has proven that publication alone does not work because clinicians rarely behave like "rational actors". Nonclinical factors are at work. Some of these factors act as filters that determine how much of the guidelines are disseminated and to what extent are they followed. Safeguarding the ENT turf, competition with fellow ENTs and with non-ENTs, the need to retain patients who insist on their own "guidelines", the need to encourage referrals from non-ENTs, HMOs and

their policies of reimbursements, the threat of malpractice, drug company influence are just some issues to which guidelines are vulnerable.

What seems important in guideline dissemination is the multiplicity of methods that would predispose, enable and encourage clinicians to behave in ways that would support evidence, expediency and ethics. Written reminders by themselves do not work. Lectures, unless given interactively, rapidly decay in clinicians' memories. Clinical reminders, when presented to clinicians at the moment of decision making may be effective. Individual detailing encourages compliance by persuasion. Focused group discussions help to engender ownership of the guidelines. Audit and feedback are effective in some settings but tend to be expensive. Computerized decision support systems can influence clinical behavior but are resource-intensive. Patient specific reminders attached to records, prescription pads and hospital forms may be cheaper alterantives.

While attempting to combine these dissemination methods optimally, we should also ask ourselves what is the PSOHNS willing to contribute? Support from the Society may come in the form of sanctions for non-compliers, support for the costs of dissemination and the subsequent regular monitoring of its effectiveness, incorporation of guidelines in the evaluation of residency training programs and of its fellows and diplomates?

Guidelines are supposed to promote evidence-based decision making but whether they do lead to good health outcomes need to be demonstrated and not just taken for granted. We will need to know if our guidelines are read, discussed, tested and complied with. Guideline evaluation tools would include surveys, questionnaires, record reviews, criterion-based audit, and routine monitoring of sentinel indicators.

Are we then up to these tasks? Perhaps, the key issues to examine are: How essential are the guidelines in the mission of the Society such that their existence would be worth all the trouble? And what are our alternatives if we choose not to develop them? The first question can only be answered by ourselves. The second, however, is already being answered as this editorial is being read. Guidelines are being developed by stakeholders outside our Society, and these guidelines will inevitably impact on the way we serve our patients, with our without our explicit involvement. As consumers awaken and third party payors insist on outcomes-based clinical performance of its health care providers, judgments on the appropriateness of our management could one day be made by those who may know very little of the realities of ENT practice. If we choose not to act.

Jose Acuin, MD, MSc

Original study

A Randomized Controlled Trial of Kersch-Wolff Versus Standard Posterior Nasal Packing in Preventing Alar Pressure Necrosis

Enriquez, Jose Erwin L, MD*

Objective: To determine if the Kersch-Wolff posterior nasal packing (KW-PNP) technique is better than the standard (S-PNP) technique in protecting the nasal ala from pressure necrosis.
Design: Randomized, blinded, controlled trial
Setting: Tertiary hospital, both pay and charity patients from January to July 1998.
Patients: 33 patients with severe posterior epistaxis from various causes were packed posteriorly using either the standard technique or the KW-PNP on one nostril and the other technique on the other nostril as determined randomly.
Results: There is a strong association between pressure alar necrosis and standard posterior nasal packing technique (RR= 6).
Conclusion: The use of the KW-PNP technique eliminates trauma to the external naris effectively preventing nasal alar pressure necrosis.

INTRODUCTION

Severe posterior epistaxis is a problem which the Otorhinolaryngologist – Head and Neck Surgeon must always be prepared to confront . While using a Foley catheter to control epistaxis is not a new technique, the problem of protecting the fragile external naris from pressure necrosis during fixation of the catheter still persists.

Many ways to anchor the catheter and distribute pressure evenly over the external naris have been described. These have included positioning cotton, felt and foam rubber padding, even eyepads over the naris before placing an anchoring umbilical clamp on the catheter (Johnson, 1956; Barton, 1970). Nahum and in a few centers placed a short segment of the plastic suction tubing over the Foley catheter before inserting it into the bleeding nostril (1972). After the catheter is placed and the balloon inflated, the plastic tube was secured against the anterior pack. This technique works well for large flaring nostrils, but those with less commodious nostrils are still at great risk of alar injury.

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In our center, the standard method of securing traction is rolling a small piece of soft operating sponge around the catheter at the alar rim and pressing against the naris with umbilical clamp. It has been observed that approximately 20% of our patients develop some form of pressure necrosis manifested as mild deformity and hyperemia of the alae, abrasion and scab formation around the rim, or obvious edema, oozing and necrosis of the entire vestibule resulting in perichondritis and consequently, stenosis later. This limited form of "ecthyma gangrenosum" has not only been very difficult to treat in as much as trying to save the cartilage and nasal contour but frustrating for both the otolaryngologist and the patient.

In December 1990 Kersch and Wolff in a "How I Do It" segment of the journal "THE LARYNGOSCOPE" presented a simple and effective technique in eliminating trauma to the external naris. They used a cut short segment of the drainage port of Foley catheters and slipped it over the catheter with the tapered end toward the balloon and the patient before insertion into the bleeding nostril. After the balloon is

inflated and a formal anterior packing placed, the previously threaded portion of catheter tube is then slid tightly against the anterior pack with the tapered end fitting safely into the naris. An umbilical clamp is then applied behind the tubing, securing adequate tension.

This study therefore aims to determine if the KW-PNP technique is better than the Std PNP technique in protecting the nasal ala from pressure necrosis by evaluating the degree, comparing and computing the relative risk of alar pressure necrosis from the two techniques. This study hopes to provide an alternative/ modified technique to totally eliminate this dreaded and disfiguring complication among our patients with severe posterior epistaxis.

SUBJECTS AND METHODS

33 patients (or 66 nostrils) were enrolled in the study from January to July 1998 who were all having profuse posterior epistaxis from various reasons and required bilateral posterior nasal packing. 23 were males, 10 were females with ages ranging from 23 to 58 years old. The following patients were excluded from the study: maxillofacial trauma patients and with multiple facial lacerations, patients with asymmetric nostrils (congenital deformities, previous rhinoplastic surgeries) prior to posterior nasal packing and moribund patients.

Upon confirmation of the diagnosis of active posterior epistaxis, bilateral posterior and anterior nasal packing was done by a resident. Simple randomization was employed by tossing a coin: heads, pack the Right nostril with KW-PNP, tails, pack the Left Nostril with KW-PNP.

While admitted, all patients were given the standard prophylactic oral or IV antibiotics, decongestants and analgesics. Packing remained in the nose for 48 hours and were removed on the third post - packing day. Guided by a questionaire, the nostrils were checked and examined for signs and symptoms of pressure necrosis by a designated resident who was blind to the nasal packing technique used in each nostril.

Consequently the left nostril was compared with the right nostril of each subject. This was done immediately after the removal of the packing and again 24 hours later prior to discharging the patients.

RESULTS

18% of patients with S-PNP and 3% of patients with KW-PNP developed signs of pressure alar necrosis (see Table).

Table. Alar necrosis by packing technique

Signs of alar necrosis	Nostrils with KW-PNP (n=33)	Nostrils with Std. PNP (n=33)
Deformity and redness resolving in 24 hours	1	1
Deformity and redness persisting after 24 hours	0	2
Epidermal abrasion of alar rim	0	1
Scab formation at alar rim	0	1
Edema of nasal ala	0	1
TOTAL	1	6

Using the McNemar test (test for matched and related data) the difference between these two proportions was significant at 90% confidence level.

The relative risk for developing pressure alar necrosis with KW-PNP relative to S-PNP was 0.17 (95% confidence limits = 0.02,1.31). This means that using KW-PNP protects patients from pressure alar necrosis and reduces its risk from 18% to 3%.

DISCUSSION

The correct and effective way of doing posterior nasal packing is a vital skill otolaryngologists should be adept with in doing. It is not enough to control the hemorrhage but one should also be aware of the complications inherent to this demanding procedure.

The danger of pressure alar necrosis is always present especially if packing is done either hastily or incorrectly. This is especially true if bleeding is massive and profuse such that traction or tension exerted by the balloon in the nasopharynx is greater. This is translated to an increase in countertraction pressure by the rubber catheter exerted on the ala. The standard PNP technique showed a higher incidence of pressure alar necrosis due to the OS pressing on the external naris. The KW-PNP technique on the other hand eliminates and obviates nasal alar necrosis by transmitting the countertension directly to the anterior packing.

Because previous studies have demonstrated that inadvertent pressure on the nasal ala causes pressure necrosis, and that there is a strong dose–response, time, cause and effect relationship and sound biological plausibility, the removal of this external pressure as proposed in this technique in study greatly reduces the risk of developing pressure alar necrosis.

This paper was based on textbook and consultants' personal definitions of pressure alar necrosis. A formal and more accurate description of pressure alar necrosis may be devised in future studies.

CONCLUSION

The Kersch-Wolff Posterior Nasal Packing technique is simple, effective and requires only commonly found equipment. It eliminates trauma to the external naris thus preventing and reducing the risk nasal alar pressure necrosis.

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Original study

Mandibular Fractures: A 2-Year Retrospective Study

Agbayani-De Jesus, Lei Edith P, MD*

A retrospective survey of mandibular fracture cases seen at Quirino Memorial Medical Center during a two-year period was made. By determining age and sex incidence, nature of incidence, place of occurrence, and most common site of fracture on local setting, the study aims to delineate areas where preventive measures might decrease such trauma. A total of 29 patients were included in the study and results were analyzed.

The majority of the patients were male, belonging to the 3rd decade of life. Most of the injuries occurred in a vehicle with blunt trauma as the most common type of injury. The angle of the mandible is the most frequent site fractured. The highest number was attributed to the vehicular accidents followed by assault / mauling. These injuries due to violence are difficult to prevent and improvement of socio-economic conditions of these patients may probably decrease the incidence of mandibular fracture.

INTRODUCTION

Fractures of the mandible are common maxillofacial injuries. Even the most minor injuries cause pain and discomfort, significant deformity, lost wages and health care expenses. It is important to characterize the causes and extent of the fracture to identify preventable factors. Classifying mandibular fracture helps to determine appropriate therapy.

Classification systems traditionally depend on location, fracture line, severity of fracture and relationship to favorable or unfavorable muscular forces.

This study aims to describe the characteristics and clinical features of patients with mandibular fractures and thus promote awareness of this type of injury. We aim to describe the patients' age, sex, type, nature and site of mandibular fractures and to identify predisposing and preventable factors in their occurrence.

SUBJECTS AND METHODS

A retrospective study was done at Quirino Memorial Medical Center, a government hospital that provides basic medical care to indigent patients in Quezon City and adjacent areas. Emergency records, log sheets of all patients seen on emergency basis at the ENT Department and hospital records of in- and outpatients treated for mandibular fractures from January 1996 to December 1997 were included. The type (blunt, blast injury), nature (assault, mauling, vehicular accident, fall, sports-related), place of occurrence (pedestrian, home, transportation, work, etc.) of the injury as well as the site of mandibular fracture (angle, body, ramus, coronoid, condyle, parasymphysis) were noted.

Excluded were patients with incomplete or unavailable records, congenital anomalies or underlying diseases of the mandible.

RESULTS

Twenty nine (29) patients were included in the study, 25 (86%) males and 4 (13%) females. Most patients were between 21-40 years (82%).

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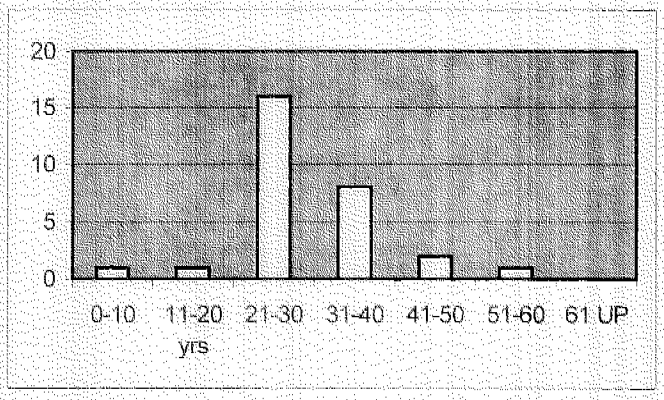

Figure 1. Age distribution of patients with mandibular fractures

Majority of the injuries occurred inside vehicles (motor vehicle, motorcycle, bicycle) or out in the streets while walking (Table 1).

Table 1 Place of Occurrence

Location	Number (%)
Street	7 (24.1)
Home	7 (24.1)
Inside Vehicle	12 (41.4)
Work	1(3.5)
Others	2 (7)

In 89% of patients the injuries were due to blunt trauma, the rest from blasts. Nearly half of the injuries were secondary to vehicular accidents, representing the most common cause. (Table 2)

Table 2: Causes of injuries

Nature Of Trauma	Number (%)
Vehicular Accident	13 (44.8)
Assault/ Mauling	7 (24.1)
Fall	4 (13.8)
Sports-Related	2 (6.9)
Others	3 (10.3)

It was also observed that vehicular accidents were predominant in the 3rd to the 4th decade of life and there was a noticeable increased incidence of mauling and assault cases in the 21 - 30 years age group.

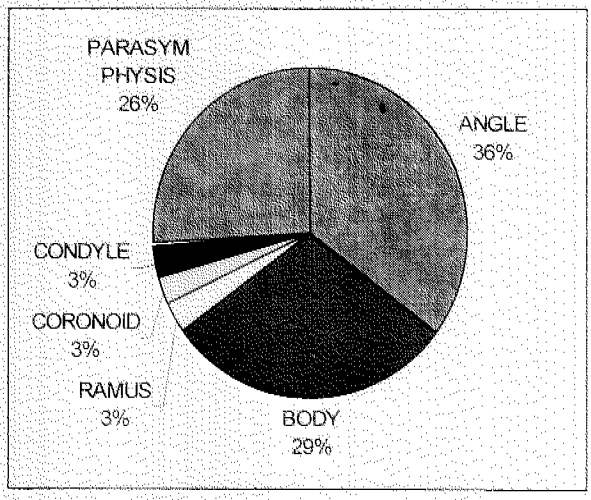

Figure 2 Sites of mandibular fracture

The most common site was the angle, 12 (35.29%) followed by body and parasymphysis (Fig.2).

Of the 29 patients only 19 (65.52%) were admitted. 17 (89.47%) underwent open reduction-internal fixation while 2 underwent closed reduction. Of those who were not admitted 8 were transferred to other hospitals and 2 absconded.

DISCUSSION

Our findings clearly indicate that more men, especially at the 20-40 years age group, are more at risk for mandibular fractures, either from vehicular accidents or from violence.

Johnson et. al. in a survey of maxillofacial trauma showed a higher incidence of facial fracture in nonhelmeted motorcyclist compared to those wearing helmets.[1] Vehicular accidents could also be vastly reduced by product modifications such as use of air bags and other passive restraints like seatbelts, anti-lacerative windshield and instrument panels without knobs and hard surfaces in likely impact areas. Their use should be encouraged by health education programs arranged in collaboration with trade unions. Government should also conduct publicity campaigns aimed at increasing the use of these automotive safety devices and compliance by motor vehicle occupants.

Ranking second as the most likely cause of mandibular fracture is assault/mauling which clearly illustrates a significant association of mandibular fracture with violent behavior. That most of our patients came from the low socioeconomic bracket might have predisposed them to violence.

Noteworthy too is the fact that most mauling incidents occurred at night and the patients were under the influence of alcohol at the time of injury. Falling while drunk or getting mugged could have caused the injury.

In this study, 3% of the patients were children below five years old. Mandibular fractures are relatively rare in children [3] and Pynn B.R. et. al., reported a facial fracture incidence of 1% in the 0-5 year old age group .[4] The most common cause was a fall. Since this could be due to inattentive caregivers, educating parents and caregivers on proper avoidance of hazards to children may prevent injuries.

CONCLUSION

In summary, this study observed that mandibular fractures are most common among males at the 2nd and 3rd decades of life who sustained injuries from vehicluar accidents or from mauling. The angle of the mandible was the most common fracture site and open reduction with internal fixation was the most common form of management.

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Original study

Laryngeal Papillomatosis Treated With CO_2 Laser Surgery

Gutierrez, Edelwissa I, MD*, Villafuerte, Jr,Cesar V, MD, Dimacali, Bernardo D, MD**, Yap, Eduardo C, MD****

Objectives: To describe six cases of laryngeal papillomatosis who were treated with CO_2 laser from April 1996 to June 1997 and the advantages and disadvantages of this procedure.
Design: Case series
Setting: Private tertiary hospital
Patients: All patients diagnosed to have laryngeal papillomatosis from April 1996 to June 1997 who were treated with carbon dioxide laser.
Results: Six patients were identified. There was an equal number of pediatric and adult cases who were treated with carbon dioxide laser using the Sharplan model 1055 surgical laser coupled to the Zeiss OPM 111 operating microscope with a 400-mm operating lens. The most common presentation was hoarseness. Endoscopic findings revealed lesions involving single or multiple sites. General anesthesia was given through the tracheostomy orendotracheal tubes. There was minimal bleedings and postoperative edema noted in all cases. They were discharged on the first post-operative day with monthly follow-up.

Flexible endoscopic findings on follow-up revealed no significant scarring on laryngeal sites. The laser procedures did not affect the rate of recurrences in pediatric patients which occurred at 4 to 8 months range of intervals. Two adult cases remained asymptomatic within 15 to 22 months with good voice quality at 4 to 6 months. One adult patient developed recurrence at same site after 14 months.
Conclusion: In our clinical experience, the use of CO2 laser did not affect the rate of recurrence of laryngeal papillomatosis due to the aggressive behavior of this disease. Tracheostomies can be avoided, no significant fibrosis is observed with minimal postoperative edema and discomfort. However, the cost of this treatment modality can be discouraging to those patients who may require periodic laser removal of papilloma.

INTRODUCTION

Laryngeal papilloma is the most common benign neoplasm seen by otolaryngologists. This was first described in the 17th century but it was only in 1871 that "papilloma" was coined to differentiate this disease entity from other laryngeal masses.[1] Laryngeal papillomatosis is not only confined to the larynx but can be seen in any portion of the upper aerodigestive tract thus, the term "recurrent respiratory papillomatosis" was proposed.[2]

The actual incidence of laryngeal papillomatosisis unknown, but the estimate of 1500 new cases diagnosed annually has been often quoted in most studies since 1964. Between 60% and 80% of cases are thought to be of childhood onset.[3]

Laryngeal papillomatosis is caused by human papillomavirus (HPV) types 6 and 11 and is characterized by recurrent proliferations of benign squamous papillomas within the respiratory tract. Although histologically benign, they may behave aggressively precipitating sudden airway obstruction and posing a potentially serious threat to life. The clinical course in terms of growth or spontaneous regression of papillomatosis seems unpredictable.

Several medical and surgical treatment modalities have been tried but none of them offers complete cure.

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However, an increasingly acceptable form of treatment for papillomas today is removal with the CO2 laser.

This paper aims to describe six cases of laryngeal papillomatosis who were treated with CO2 laser from April 1996 to June 1997. It also aims to elaborate on the advantages and disadvantages of this procedure.

CLINICAL DATA

Case 1:

K.L., 9 year old female is a diagnosed case of recurrent laryngeal papillomatosis and was referred to our institution for laser surgery last April 26, 1996. Between 1991 to 1996 she has undergone multiple endolaryngeal excisions of papillomas including 14 conventional and 6 laser procedures in Canada. She also underwent tracheostomy at some time during her previous admissions.

On admission, flexible endoscopic examination revealed fleshy mass at the anterior portion of the subglottic area. There was scarring from the supra-to subglottic areas.

General anesthesia was introduced through a pediatric endotracheal tube size 3. Excision of the mass was done using cup forceps wherein bleeding was immediately noted. The rest of the lesion was ablated with CO2 laser (5 watts single pulse mode). She was discharged on the third post-operative day and was advised monthly follow-up. She remained asymptomatic until after 6 months when she developed difficulty of breathing. She underwent tracheostomy at a government hospital and was subsequently referred to our institution for laser surgery.

Flexible endoscopy revealied a fleshy mass at the supra-to subglottic areas. She underwent CO2 laser excision (3 watts single pulse mode) of the mass under general anesthesia introduced through a tracheostomy tube. She was given oxygen at 35% FIO_2 through a T-piece and was decannulated on the fourth postoperative day. She was discharged improved on the fifth postoperative day and was advised monthly follow-up. She has remained asymptomatic thereafter.

However, on subsequent follow-up after eight months, patient complained of hoarseness, cough and difficulty of breathing. Flexible endoscopy revealed recurrence of the subglottic mass. Parents refused repeat laser surgery due to financial constraints.

Case 2:

A.C., is a 4 year old, male who presented with stridor at 20 months of age. He was diagnosed to have recurrent laryngeal papillomatosis for which three convention excisions were done at four to six months intervals from 1994 to 1995 at a government hospital. Tracheostomy was done on first admission in May 1994 because of upper airway obstruction. He was then referred to our institution for laser surgery of recurrent papillomatosis in April 1996.

On admission, flexible endoscopy revealed fleshy masses from the supra- to subglottic areas. Intraoperatively, there were areas of scarring from the supraglottic areas extending up to the anterior commissure. Excision of laryngeal papillomatosis was done using carbon dioxide laser (3-5 watts single pulse mode). General anesthesia was introduced through pediatric endotracheal tube size 3. Extubation was delayed and intravenous steroids and antibiotics were given. He was discharged on the fourth post-operative day and remained asymptomatic for 4 months.

However, on subsequent follow-up, he had on and off cough. Flexible endoscopy revealed recurrence from the supra-to subglottic areas. Parents refused repeat laser surgery due to financial constraints.

Case3:

K.P.H., is a 3 year old, female who presented with hoarseness 2 months prior to admission. Flexible endoscopy revealed fleshy masses at the supraglottic extending unto the subglottic areas. Excision was done using carbon dioxide laser (3-5 watts at single pulse mode). General anesthesia was introduced through a pediatric endotracheal tube size 3. Extubation was delayed and she was discharged on the second postoperative day. On subsequent monthly follow-up, patient developed on

and off cough and hoarseness. Flexible endoscopy revealed recurrence of the mass at the supraglottic area extending to the anterior commissure. Parents refused further intervention due to financial constraints.

Case 4:

J.P., a 24 year old female is a diagnosed case of laryngeal papillomas with a chief complaint of hoarseness for 6 months. She was referred to our institution for laser surgery in October 1996. Clinical examination revealed papillomatous lesions occupying supraglottic extending up to the glottic areas. Debulking of the mass was done using cup forceps; the rest of the lesion was later ablated using CO2 laser (3-5 watts setting, single pulse mode). General anesthesia was introduced through a size 6 endotracheal tube. Extubation was delayed and patient could be discharged on the first postoperative day. At present, patient has a good voice quality and there has been no evidence of recurrence for 22 months.

Case 5:

A 58 year old male was admitted because of hoarseness for 7 months. Clinical examination revealed a fungating mass at the left true vocal cord. He underwent CO2 laser excision (3-5 watts single pulse mode). General anesthesia given through size 6 adult endotracheal tube. Patient was discharged in stable condition on the first post-operative day with monthly follow-ups. At present, patient has a good voice quality and there has been no evidence of recurrence for 14 months.

Case 6:

A 48 year old male is a diagnosed case of laryngeal papillomas and was referred to our institution for laser excision. He was admitted because of hoarseness for 8 months. Clinical examination revealed a sessile, fleshy mass at the anterior commissure extending up to the subglottic area. He underwent CO2 laser excision (5 watts single mode). General anesthesia was given through size 6 adult endotracheal tube. Patient was discharged on the fifth post-operative day due to medical problems. On subsequent consult at the OPD, after 14 months, patient complained of hoarseness and difficulty of breathing. Indirect laryngoscopy revealed recurrence of the mass occupying the anterior commissure. He refused further intervention due to financial constraints.

From April 1996 to June 1997 six patients diagnosed to have larngeal papillomatosis in our institution treated with CO2 laser were reviewed. There were equal number of pediatric and adult patients. The average age of presentation of pediatric patients was 34 months. Intervals of recurrences ranged from 2 to 6 months. Papillomas involved multiple sites. In cases 1 and 2 areas of supra- to subglottic scarring were seen. This can be due to the multiple endolaryngeal surgeries which they went through.

There were three adult patients with ages ranging from 24 to 58 years old at onset of symptoms and diagnosis. All presented with horseness and lesions were seen involving multiple sites. All cases need not undergo tracheostomy prior to the operation.

All procedures were performed using the Sharplan model 1055 surgical laser coupled to the Zeiss OPM 111 operating microscope with a 400-mm operating lens. General anesthesia was given through the tracheostomy tube or endotracheal tubes. Debulking of papillomatous masses was done to cases 1 and 4 using cup forceps. Massive bleeding was immediately noted. Remaining lesions were excised using CO2 laser setting was at 3 to 5 watts single mode. There was minimal bleeding and postoperative edema noted in all cases and discharge at the first post-operative day was possible in all.

Postoperative flexible endoscopy revealed no significant scarring on larygeal sites. All pediatric cases had recurrences occurring at 4 to 8 months range of intervals. The laser procedures did not affect the rate of recurrences in all pediatric cases (Table 1). Cases 4 and 5 remained asymtomatic for 15 and 22 months respectively. Both cases have good voice quality at 4 and 6 months respectively. However, Case 6 had recurrence of the lesion at the anterior commissure after 14 months (Table 2).

Table 1. Pediatric Cases Treated with Carbon Dioxide Laser

Cases	Age at onset	Number of Conventional Procedures	Interval of Recurrences	Sites of Involvement	# of CO2 Laser Procedure	Results after treatment with Carbon Dioxide Laser
1 - K.L., 9/F	3	22	2-6 mos	Supra- to subglottic areas	8	Recurrence noted at subglottic area after 8 months No significant scarring
2 - A.C., 4/M	26 mos	3	4-6 mos	Supra- to subglottic areas	1	Recurrence noted at supra- to subglottic areas after 4 months No significant scarring
3 - K.L., 3/F	3 years 6 mos	-	-	Supra- to Subglottic areas	1	Recurrence noted at supraglottic to Glottic areas after 4 months No significant scarring

Table 2. Adult Cases Treated with Carbon Dioxide Laser

Cases	Age at onset (years)	Site of Involvement	Result after treatment with Carbon Dioxide Laser
4 - J.P., 24/F	24	Supra- to glottic areas	No evidence of recurrence for 22 months Good voice 4 months post-op No significant scarring
5	58	Left true vocal cord	No evidence of recurrence for 15 months Good voice 6 months post-op No significant scarring
6	48	Anterior commissure to subglottic areas	Recurrence noted at the anterior commissure after 14 months Hoarseness No significant scarring

DISCUSSION

Laryngeal papillomas are the most common benign lesion frequently encountered in children. These are considered an abnormal tissue response to mucosal infection by a viral agent, the Human Papilloma Virus type 6 and 11. The virus induces the formation of papillary like-projections that interfere with phonatory and respiratory function.[4] Likewise, HPV infection frequently persists in adjacent, clinically, normal site.and that the extent of nondiseased site involvement may predict both the extent of the disease and the likelihood of recurrence. [5,6]

Laryngeal papillomatosis is classified into juvenile- and adult- onset groups. There is however no distinctive histologic difference between these forms. In the juvenile type, infants and children are affected whose growth are always multiple and recurrence is common in spite of therapy. This form is often extremely aggressive and resistant to treatment, requiring frequent laryngoscopies. [7]

Intraoperatively, these lesions appear as white to pinkish-red grape-like

masses. They are friable and bleed easily on manipulation.

There is epidemiologic evidence linking maternal condyloma with juvenile onset laryngeal papillomatosis [1] although no history of genital warts during pregnancy were elicited among the mothers of our pediatric patients. However, patients delivered by Caesarean section are not immune to develop this, and so infection may be acquired transpalcentally or postnatally.

Multiple recurrences in our pediatric patients can be explained by the tendency of this lesion to seed into previously uninvolved areas that sustain epithelial injury, such as that which follows traheostomy or other surgical instrumentation. [8] Tracheostomy should be avoided if possible since it predisposes to distal papillomas seeding by activating latent HPV infection. According to Kashima et al, papillomas tend to grow at squamociliary junctions that can be induced in the trachea by traumatic metaplasia from a tracheotomy tubes. However, if this procedure is unavoidable, patients must be decannulated as soon as it is feasible.[9]

Three of our six cases were classified as adult-onset or senile types. Lesions were noted to be solitary and smaller than those seen in children. A slower, progressive though less aggressive behavior is also observed. The source of infection in adults is still unknown. Erisen et al reported cases of recurrent tumors in adult patients but the intervals between occurrences are long.

The otolaryngologist is faced with four problems in dealing with the management of laryngeal papillomatosis: (1) obstructive location of the lesions; (2) their multiplicity; (3) high recurrence rate; and (4) the ability of the papillomas to seed previously uninvolved mucosa.[10]

Different forms of treatment have been employed over the years. These can be grouped into physical, surgical, medical, and immunologic categories. Medical modalities have included the use of hormones, broad spectrum antibiotics, chemotherapeutic agents, retinoic acid and steroids which have shown to be of any significant benefit.

Ultrasound should be used cautiously with long term follow-up to evaluate the side effects on neural tissue and growth centers of the larynx. The use of liquid nitrogen began in the 1960's. Repeated cauterization with corrosives and electrocautery can lead to scarring and stricture. The sclerosing effect of nitric acid, trichloroacetic acid and formaldehyde can lead to laryngeal stenosis. The effectiveness of the use of Podophyllum in treating laryngeal papillomas has been questioned.[1] Radiation therapy should not be used due to its potential for injuring the laryngeal skeleton and for inducing malignant transformation.[6]

The nonsurgical treatment which appears to hold great promise is the use of interferon. Ongoing studies of the combination of CO2 laser and alfa recombinant interferon in the treatment of recurrent papillomatosis has been reported by Mattot et al wherein three of the five children are currently disease free for periods ranging from 22 to 68 months. According to Avidano, juvenile onset disease had a slightly higher response to interferon combined with standard laser excision than adult onset disease.

At present, periodic microsuspension laryngoscopy and carbon dioxide laser vaporization offers good control. The strategy of treatment with CO2 laser is to remove all exophytic papillomas with as little damage to underlying and adjacent tissues as possible.[8] Carbon dioxide laser has proven to be a superior method in treating laryngeal papillomatosis because of laser energy-soft tissue interaction: (1) an inherent hemostatic effect; (2) minimal production of edema in tissue postoperatively, (3) rapid healing due to minimal peripheral tissue damage; and (4) minimal resultant scarring.[4]

According to Mihashi et al, the rapid thermal drop of laser energy in the tissue surrounding the incision results in shallow and predictable tissue penetration thus limiting spread of thermal energy thereby decreasing postoperative edema. The heat generated by the laser seals small vascular channels as well as the lymphatic channels and sensory nerve endings which leads to better visibility for the surgeon in an almost bloodless surgical field with decreased

postoperative pain.[4,11] In addition to this, proper selection of appropriate CO_2 laser emission parameters and the use of the microspot micromanipulator, help to minimize lateral and/or deep thermal damage at the site of laser impact.[12]

Despite the advantage over other endoscopic modalities, laser vaporization is certainly not without its risks. The disadvantages are all related to thermal injury; thus, the key to laser safety is proper precaution to direct and indirect hazards to both patient and operating room personnel. Risks involved in CO2 laser include airway fire, pneumothorax, laryngeal and tracheal stenosis and tracheocutaneous fistula. To avoid airborne transmission of plume containing laryngeal papillomas viral-infected cells and infectious viral particles, Mahnke et al recommended that carbon dioxide laser parameters should be in a continuous mode with the power density equal to, or more than, 1667 W/cm^2.[13]

Additional precautions are concerned with the choice of anethetic technique, the choice and the protection of the endotracheal tubes, and the selection of proper instruments.[11] Another disadvantage is that the procedure is very expensive which can be discouraging to patients such as ours who may require multiple laser procedures.

CONCLUSION

Laryngeal papillomatosis is a serious and a potentially life-threatening disease. These lesions show an unpredictable pattern of active disease and remissions. The result of our clinical experience with the use of CO2 laser is similar to that of other investigators. Its use did not inhibit recurrence of laryngeal papillomatosis due to the aggressive behavior of this disease.

We noted no significant fibrosis and minimal postoperative edema and discomfort. However, the cost of this treatment modality can be one of its limitations. Likewise, laser safety should be properly observed to avoid any complications. A close follow-up in monthly basis with laryngeal examination is recommended to monitor recurrence. Removing the disease on a periodic basis before extensive growth occurs or obstructive symptoms develop is essential to minimizing the complications. Further experience with the use of adjuvant therapies in combination with CO2 laser may be helpful in decreasing the recurrence rate of juvenile onset laryngeal papillomatosis in our country.

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A Simple Fixation Device for the Anatomic Reconstruction of Segmental Mandibular Defects

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The challenge in reconstruction of segmental mandibular defects lies in the difficulty of re-establishing the three-dimensional relationship of the oral cavity ensuring restoration of form and function. An important prerequisite of a successful reconstruction is the maintenance and restoration of the original, preresection anatomic position of the mandibular condyles. Several methods employed to stabilize and align resected mandibular segments are discussed. A technique utilizing a locally manufactured three-dimensional mandibular fixation device for intraoperative stabilization and alignment of proximal mandibular segments is introduced. Utilization of such a device offers an easy, expedient and cost-efficient technique for accurate restoration of preoperative mandibular relations.

INTRODUCTION

Reconstruction of mandibular defects after trauma or tumor resection is one of the most challenging problems facing otolaryngologists and head and neck reconstructive surgeons. When undertaking mandibular reconstruction, the restoration of bony continuity and facial contour alone should not be considered the measure of success. The ultimate goal of mandibular reconstruction is to return the patient to their previous state of function.

Segmental defects are usually bridged with a reconstruction plate with the intent of restoring the continuity of the mandible in terms of its function, form and strength. Successful bridging is dependent on stable retention of the original anatomic position of the proximal mandibular segments which achieves adequate mandibular movement and occlusion. Optimal function in turn is critically dependent on the maintenance of physiologic condylar position. The condyle should be anatomically aligned in three planes of space. Slight torque of the condyle out of the glenoid fossa can have significant negative effects on mandibular Movement and dental occlusion and can cause pain.[4]

Herein lies the challenge of mandibular reconstruction---the difficulty of recreating the intricate three-dimensional relationship of the oral cavity ensuring occlusal relationships, oral competence and facial contour. Realizing the importance of maintaining mandibular position, a variety of methods have been employed to stabilize and align resected mandibles and restore the articular segment to its presurgical position. [1,3,4]

Our objective is to propose a simple technique for stabilizing proximal mandibular segments intraoperatively. The technique involves the use of a locally manufactured, modified adjustable mandibular fixation device originally conceptualized by Raveh et al (1983)[1] and recently described by Steinberg and Collins[4] in 1988 (manufactured by Leibinger, Howmedica-Leibinger Inc., Dallas, TX).

The use of the device is of clinical relevance as the technique is simple, easy, expedient, cost-effective and can be utilized in the majority of segmental mandibulectomy procedures. More importantly, its use assures the restoration of the original anatomic position of the condyles which is

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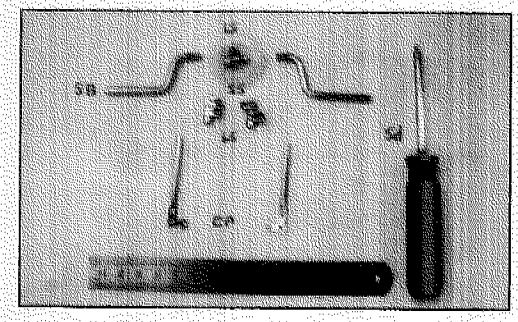
a prerequisite for successful mandibular reconstruction. The components of the device are illustrated in figure 1. The entire device has been manufactured locally.



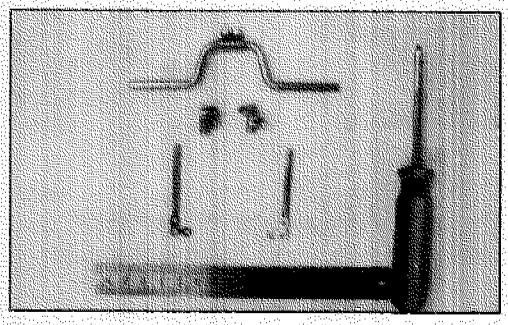


Fig. 1A and 1B. Components of the three-dimensional adjusdtable mandibular fixation device.

SB=stabilization bar: U-shaped curve designed to fit into the palatal arch. Our modified stabilization bar contains a midline swiveling joint with locking screws which allows each limb to move independent of each other. This modification allows another dimension of movement/adjustment to the device enabling fixation of the device to the most accessible proximal mandibular segments. This allows one stabilization bar to accommodate different mandibular widths thus obviating the need for different size bars.
SS=Swiveling clamps with locking screws for alignment of fixation plate bar
LS=locking screws on adjustable joints. All heads of screws oriented anteriorly for ease of adjustment. Joint lock simultaneously
CP=Carrier plates used to secure the device onto the mandible.
S=Screwdriver; The hexagonal drive-in screw head provide good force transmission from the screwdriver to the screw. In contrast to slotted or Phillips, the hexagonal drive does not require an axial force to apply torque to the screw. Thus the danger of slipping and damaging the screw head is reduced, and screw removal is not complicated.

TECHNIQUE (Figure 2)

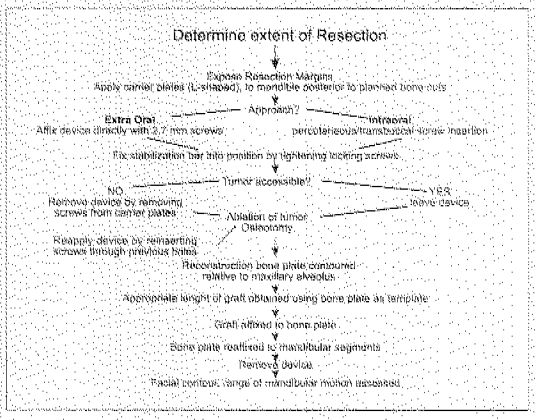


Figure 2 Steps Involved in Use of Mandibular Fixation Device

The surgical margins for the mandibular resection is determined. The lateral surface of the mandible posterior to the resection margin is exposed. The carrier/fixation plates (L-shaped) of the device are applied to the superior aspect of the mandible proximal to the planned osteotomy site. The plates are then fixed to the mandible with 2.7 mm screws. This device can be utilized when resecting and reconstructing mandibular defects from either an intraoral or an extraoral approach depending upon the clinical situation.

In the extraoral approach the screws can be applied directly to the mandible. Alternatively if an intraoral approach has been chosen, the screws can be applied percutaneously/transbuccally.[1,2] The stabilization is then applied to connect the two carrier plates.

The bar is U-shaped to fit over the tongue inside the curvature of the hard palate. The swivel joint of the bar allows greater maneuverability within the limited confines of the oral cavity. The stabilization bar is locked into position by tightening all screws using the hexagonal drive-in screw head manufactured with the device.

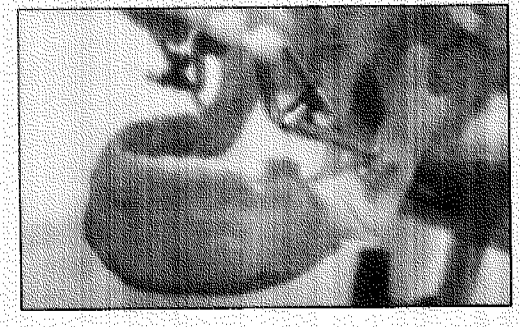
If the device does not interfere with tumor resection, it can be left in place, otherwise, the locked connecting bar and fixation carrier plates can be taken off the mandible in one piece by removing the 2.7 mm screws from the carrier plates. The locked stabilization device and the position of the holes drilled into the mandible record the preresection position of the mandible.

The mandible is then osteotomized in the previously determined locations. The

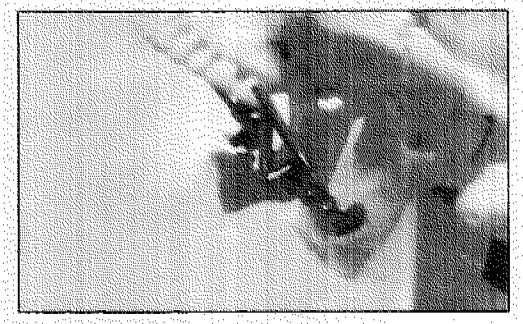
mandibular segments can be manipulated/retracted providing adequate surgical access for tumor excision. After the tumor is resected, the apparatus is reapplied with screws through the previously drilled holes, effectively repositioning the mandible in its original anatomical preresection position.

At this point a reconstruction bone plate can be contoured to the inferior border of the remaining mandibular segments. The continuity of the mandible is restored by closely contouring the reconstruction plate to the curvature of the maxillary alveolus. A fairly constant distance should be maintained between the maxillary alveolus and the reconstruction plate.[3] A suitable bone graft can then be shaped, secured to the reconstruction plate and attached to the mandibular segments. Once the reconstruction plate is secured, the fixation device is removed and the reconstruction bone plate and will maintain the preresection anatomic mandibular position.

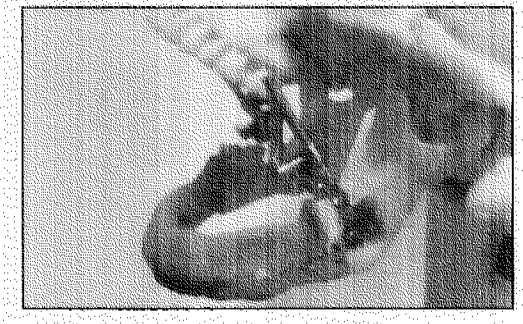
Figure 3 and Figure 4 illustrate the application of the device in hypothetical segmental mandibulectomy procedures using a skull-mandible model and a cadaver specimen respectively.



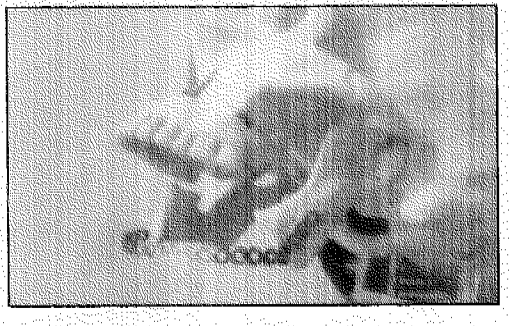
3A Model of a skull and mandible. The darker area represents an anterior-extending tumor that precludes prebending a reconstruction plate in the region. The adjustable fixation device has been applied to the ascending ramus bilaterally.



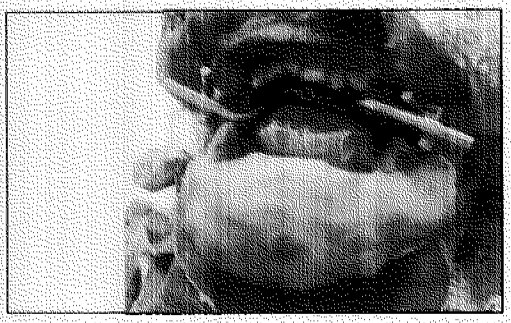
3B The involved segment of the mandible has been resected. The proximal mandibular segments are lined up to the carrier plate using the previously made drill holes. The anatomical position of the resulting fragments is retained by the fixation device. Note that the condylar position has been maintained by the device.



3C A bone graft secured to the reconstruction plate has been applied to the inferior border of the mandible. The continuity of the mandible is restored with a reconstruction plate closely contoured to the curvature of the maxillary alveolus. A fairly constant distance should be maintained between the maxillary alveolus and the reconstruction plate.



3D The adjustable fixation device is removed after the reconstruction bone plate is secured. The anatomical position of the mandible is now maintained by the bone plate. (no bone graft in this illustration)



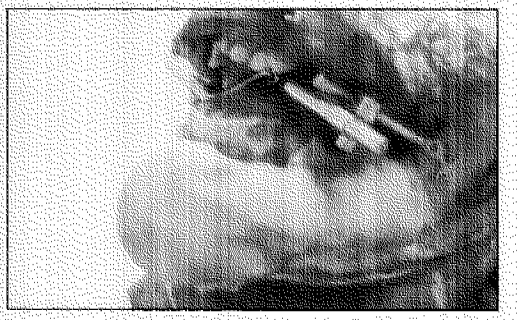


Figure 4 A & B Cadaver specimen. The darker area represents a significant tumor involving anterolateral portion of the mandible up to the left angle. The involvement of the lateral surface of the mandible precludes prebending a reconstruction plate in the region. The adjustable fixation device has been applied to the proximal mandibular segments

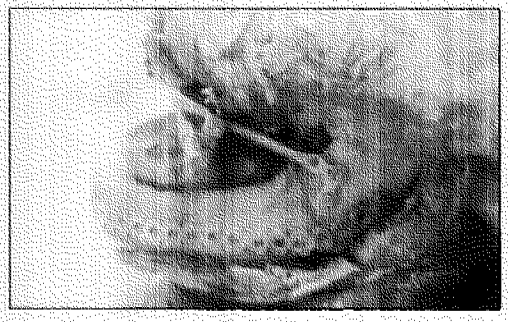


Figure 4C The involvement segment of the mandible has been resected. A bone graft secured to the reconstruction plate has been applied to the mandible.

Figure 4 D The adjustable fixation device is removed after the reconstruction bone plate is secured. The anatomical position of the mandible is now maintained by the bone plate.

DISCUSSION

In order to achieve successful mandibular reconstruction emphasis must be placed on optimizing mandibular position when segmental resection is required by appropriate intraoperative stabilization of proximal mandibular segments. Several methods to stabilize and align resected mandibles have been reported in the literature. For a variety of reasons no single technique can be used.

Perhaps the simplest method of fixation is what oral and maxillofacial surgeons' term maxillomandibular fixation (MMF) or what most people know as "wiring the jaw shut". With an intact dentition and a stable upper jaw, maxillomandibular fixation allows the upper jaw to act as a "cast" for the lower jaw while it heals.

Another well-described and reliable method to restore the basic shape if the mandible is attaching an appropriately contoured reconstruction plate to the intact mandible before resecting it. The plate is fixed distal to the proposed lines of resection with two screws on each side and then removed. The drill holes and the corresponding plate holes will provide the reference points for bridging the defect and for restoring the basic contour of the jaw.

Both techniques of adapting a reconstruction plate to the mandible prior to resection as well as of maintaining the spatial relationships of the mandible with intermaxillary fixation (IMF) are common.[1,3,4] However bone plates cannot be utilized when the tumor goes beyond

and deforms the lateral surface of the mandible and MMF is difficult in edentulous patients.

The use of dental splints, is another method used to maintain mandibular position. However, both MMF and splints will not hold the mandible together in three planes of space and will allow rotation of the free mandibular segments once the mandibular segment is removed.

The splint-plate technique, wherein temporary bone plates are attached to splints (Reece et al.), eliminates this rotation but requires bending and discarding additional bone plates, and consultation with prosthodontists for dental impressions and splint fabrication..

External pin fixation is reliable but can be time consuming to apply and can interferes with surgical access to the tumor.[1,3,4] Other methods utilize the maxilla as a stable reference from which the distance to the ascending ramus is determined in centric occlusion. Following the osteotomy the proximal segment is repositioned in accordance to the distance previously determined.

Spiessl and Tschopp (1974) developed a forklike device with a scale that attaches to the maxillary arch splint and retains the divided ramus in its original position. Leonard et al (1985) described a "Proximal Segment Orienting Device" (PSOD) with a paddle extension which offers a simple and reliable means if establishing the position of the proximal segment. Seto's technique of segmental repositioning (Seto & Matsuura 1984) is even simpler, as it used a detachable miniplate to reproduce the normal distance between the fixed reference (maxilla) and the ascending ramus.[1] Recently, Li et al (1996) reported temporary fixation of the mandible to the zygoma with plates.[3] Although the last two techniques are able to maintain condylar position. The techniques are complex, requiring additional exposure of the zygomaticomaxillary region, which requires additional bone plates and may open up new tissue planes to possible tumor cell implantation.[3,4]

In place of the simple miniplate, Raveh et al (1983) used a specifically designed holding device with two anchoring elements and an adjustable midpiece to restore the normal distance between the mandibular segments.[1] Recently Steinberg and Collins (March, 1998) described the use of such a mandibular fixation device (Leibinger, Howmedica-Leibinger Inc., Dallas, TX).[3] The stability of this device allows even more accurate repositioning of mandibular segments. The above device described by Steinberg and Collins necessitated use of two different size stabilization bars to accommodate different widths of the mandible.

Our modification involves the use of a single stabilization bar that contains a midline locking swiveling joint. The added joint allows each limb to move independent of each other. This modification added another dimension of movement/ adjustment to the device effectively increasing its maneuverability within the limited confines of the oral cavity. It also allows the device to accommodate to varying mandibular widths and dimensions without the need for different size bars.

CONCLUSION

We present a simple method of mandibular stabilization using a three-dimensional, adjustable fixation device that can be used regardless of the surgical approach and/or method of reconstruction (free flap, plate and soft-tissue flap or plate and bone graft) chosen. It is easily applied through the same exposure needed for bone plating, and does not require exposing other tissue planes to the risk of tumor cell implantation. Its maneuverability enables it to be accommodated within the limited confines of the oral cavity and can be applied regardless of mandibular width. It can be removed and later reapplied if needed to gain access to the tumor or it can be left in place if exposure is not hampered, eliminating steps in the procedure thus saving time.

The use of the device may be economically justified. It eliminates visits to prosthodontist with its additional laboratory time and costs. It is also reusable obviating the need to bend and discard additional bone plates. More significantly the use of this device and this technique allows accurate three dimensional restoration of the original

anatomic position of the mandible which would improve the functional and aesthetic outcome of the reconstruction.

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Endoscopic Guided-Laser Assisted Management of Glabellar Frown Lines

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Objectives: To present the management of glabellar frown lines in a Filipina patient using CO_2 laser through endoscopic technique, discuss its advantages and enumerate alternative methods in managing glabellar frown lines.
Design: Case Report
Setting: A private hospital in Metro Manila equipped with a CO_2 laser machine.
Patient: Sixty-four year old female with prominent frown lines in the glabellar area.
Results: Glabellar frown lines were satisfactorily removed under local anesthesia. No complications were noted.
Conclusion: The technique was successfully carried out in this patient. While the eventual results of surgery with or without the laser are the same, CO_2 laser diminishes the degree and length of postoperative morbidity. The desired effect was achieved endoscopically with minimal scarring and preservation of sensate flaps.

INTRODUCTION

As we age, our faces begin to show the effects of gravity, sun exposure and years of facial muscle movement, such as smiling, chewing and squinting. The underlying tissue that keep our skin looking youthful begin to breakdown, often leaving laugh lines and facial creases over the areas where muscle movement occurs. The appearance of glabellar frown lines in the forehead is directly related to the muscular dynamics of the area, namely, the pull on the skin of the underlying corrugator and procerus muscles.[1] Glabellar furrows alter facial expression so that people appear older, angrier or more tired than they are.

The management of glabellar frown lines varies in approach and principles. It includes injectable fillers such as collagen and fat to obliterate furrows and creases; botulinum toxin injection to weaken selected muscles; the standard forehead lifts approached by coronal incisions; and minimal incision endoscopic browlift.[2]

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Paramount to achieving a satisfied patient and optimum surgical effect in removing glabellar frown lines is minimal trauma to subcutaneous tissues. Three factors are required: small, well-hidden incisions, minimal disturbance of neural structures and absolute control of hemorrhage. By combining the use of CO_2 laser system with endoscopic visualization and direction, it is possible to readily satisfy these requirements. The CO_2 laser is used to cut and ablate the corrugator and procerus muscles to address skin folding in the glabellar region. Thus, a new technique for eliminating glabellar frown lines is presented.

CLINICAL DATA

E.B., a 64-year-old female consulted because of unwanted deep vertical lines in the glabellar region (Fig. 1) which had been present for several years. Physical examination showed two deep vertical creases in the glabellar area measuring 2 cm each. The rest of the ENT-head and neck findings were unremarkable.

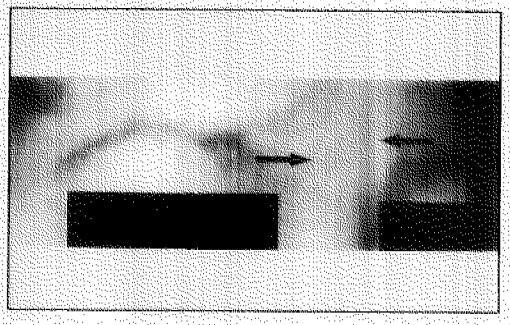

Figure 1. Preoperative facial features

I. Anesthesia

Preoperative medications consisted of promethazine HCl (Phenergan) 25 mg and pethidine HCl (Demerol) 25 mg IM. Lidocaine 1% with epinephrine at 1:100,000 dilution was injected over the glabellar and midline forehead areas to anterior hairline.

II. Operative technique

A 1.5-cm incision just behind the hairline was made using blade no. 15 and carried down to the galea. Subaponeurotic undermining of the mid-forehead area was done extending to the glabella over the root of the nasal dorsum, using curved Metzembaum scissors. A 4 mm rigid 30° nasal endoscope was inserted into the incision and to the undermined area. A Sharplan 1055 CO_2 laser machine with a Flexilase nasal fiber attached to a F-125 mm handpiece was used. The laser power was set at 5 watts superpulse and exposure mode was set to single at 1 sec on time. The Flexilase extension tip was inserted into the undermuned mid-forehead tunnel (Fig.2).

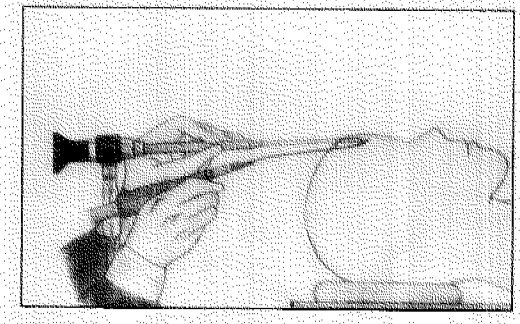

Figure 2. Insertion of Flexilase tip into undermined forehead skin.

Under endoscopic visualization, vaporization-myotomy of the corrugator and procerus muscles were done in a criss-cross pattern to partly erase the frownlines. Repeat vaporization of the muscle fibers directly underneath the vertical furrows totally removed the residual creases. A Sharplan smoke evacuator connected to a Frazer suction tip to suck out laser plumes. Both the endoscope and the Flexilase fiber were removed cautiously. The small hairline incision was sutured using nylon 5-0 interrupted sutures. Moderate pressure gauze dressing was applied over the mid-forehead and glabellar areas. The patient tolerated the procedure well. No postoperative complications were noted.

DISCUSSION

As aesthetic facial rejuvenation has become increasingly popular, surgeons and patients alike have searched for techniques that provide longer lasting natural-appearing results as well as decreased morbidity and recovery time. In the head and neck area, the endoscopic forehead and brow lift was the first clinical application of aesthetic endoscopy and is by far the most common endoscopic procedure performed today.[4] Because an excellent cavity can be created between the frontal bone and the soft tissues of the forehead and brow region, this area is ideally suited to the endoscopic technique.[4]

Endoscopy minimizes the dysesthesias associated with the coronal approach by making incisions shorter and radially oriented and by more precise muscle resection.[5] Using small incisions under endoscopic guidance, it is imperative that the field be dry at all times and the muscles be precisely cut. To achieve the driest filed possible and absolute control of hemorrhage, a 55-watt CO_2 laser machine was employed.

We used the CO_2 laser machine equipped with the Flexilase fiber to incise and ablate the corrugator and procerus muscles which were primary responsible for the vertical furrows seen in our patient. The CO_2 laser released the tissues from their attachments and the muscles ablated, allowing redraping of the glabellar skin.

Thus, wrinkles over the glabellar area were refined and subsequently removed. Intra- as well as postoperative bleeding and edema were not noted since the CO_2 laser can generate heat sufficient to seal off small vascular channels affording excellent hemostasis.[6] Because the CO_2 laser can be delivered precisely to the target tissues, there is minimal damage to surrounding structures and less edema formation[6] as demonstrated in our patient. The laser can also seal sensory nerve endings leading to decreased postoperative pain. The entire procedure was completed in an hour with no complications noted.

Alternative methods of treating glabellar frown lines include the use of botulinum toxin. This agent when injected into the corrugator or procerus muscles, blocks the nerve impulse from reaching that area and as a result, the muscles weaken. The skin overlying the muscles relaxes, thus diminishing glabellar kinetic folds.[8] Although there are no serious side effects associated with its use, improvement with the use of botulinum toxin is dose-dependent and temporary, which may last an average of 3 to 4 months.[9] Repeated injections may be needed, entailing extra costs for the patient.

Soft –tissue fillers, most commonly injectable collagen or fat (microlipoinjection), can also be used to fill in these glabellar frown lines, in conjunction with facial surgery procedures. When injected beneath the glabellar skin, these fillers can plump up the creases and furrows. However, there is generally some swelling, soreness and bruising after injections.[10] Just like botulinum toxin injection, correction is also temporary lasting only for 3 to 6 months,[11] since the body eventually metabolizes these injected fillers. Additional injections may be needed to achieve a longer lasting result. The outcome of treatment is never completely predictable.[11]

The traditional approach by which glabellar frown lines are removed is through a forehead or browlift wherein an ear-to-ear incision across the scalp is made.[12] The corrugator and procerus muscles are removed and the tissue is released from its attachments to the forehead, allowing the skin to be redraped, tightened and the excess removed. The classical browlift is primarily indicated for severe transverse wrinkling of the forehead, which was fortunately not seen in our patient. The main disadvantage of the open approach is that it is more extensive than other operations, and therefore carries a greater risk of complications.[13] The common sequelae include scar alopecia, and persistent forehead and scalp numbness.[4] The procedure requires more operating time, thereby increasing cost relative to the other procedures.[13]

The endoscopic surgical technique coupled with the use of a CO_2 laser machine has largely replaced the classic technique. It has revolutionized the removal of glabellar frown lines allowing similar results with minimal complications. Improved magnification and visualization of the structures allow precise dissection in a compact area.[4] The concomitant use of the CO_2 laser enabled the surgeon to cut precisely and produce a nearly bloodless field. Surgical time is shortened because of minimal incision and dissection, thus avoiding the secondary sequelae associated with the open technique such as bleeding, swelling and bruising. The traditional complications of postoperative cutaneous anesthesia and telogenic hair loss are almost nonexistent.[7] In addition, the endoscopic technique does not cause disfiguring scarring.

CONCLUSION

We have just presented a new method of managing glabellar frown lines, utilizing both the technologies of endoscopic and laser surgery. The endoscopic guided laser technique presents clear advantages over the other methods. It has gained popularity among plastic surgeons and patients because of the small incisions, preservation of senate flaps, reduction in operative time and bloodloss, less ecchymosis and decreased risk for incisional alopecia.[14] Thus, satisfactory and more acceptable results were achieved.

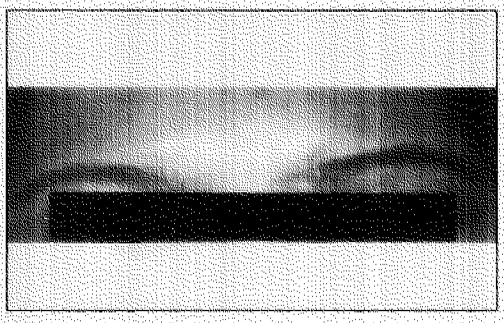

Figure 3. Four days post-op showing minimal edema and absence of ecchymosis.

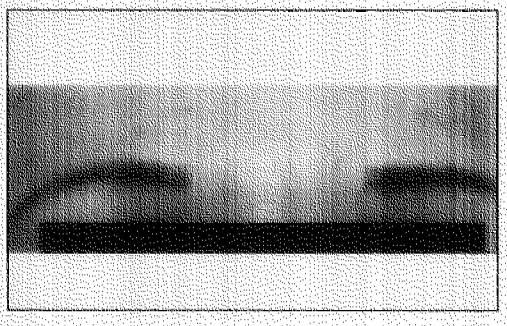

Figure 4 Eighteen months post-op showing the absence of glabellar frown lines

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Case Report

Anaplastic Cancer of the Temporal Bone presenting as Chronic Otitis Media

Penaflor, Neil Aldrine I, MD*, Jugo, Jose Carlos Z, MD*, Abes, Generoso T, MD**

INTRODUCTION

Temporal bone carcinoma is rare, occurring in only one in twenty thousand in the general population, as reported in foreign literature. To our knowledge, no case of anaplastic cancer of the middle ear or temporal bone has ever been reported either locally or internationally. Diagnosis of temporal bone carcinoma is not always straightforward, as in the case presented here, wherein the history, physical examination, clinical behaviour, and hence, management geared towards refractory chronic otitis media with mastoiditis. Awareness, and a high index of suspicion regarding this rare tumor, however, combined with routine histopathologic examinations for all tissues removed in 'chronic ears' refractory to usual medical and surgical therapy should allow us to detect temporal bone cancers early, and hopefully alter the clinical course of the disease.

CLINICAL DATA

DB, a 43 year old male, consulted for left ear pain that started approximately 9 years ago accompanied by ear fullness after a bout of coryza. An otoscopic examination showed left middle ear effusion while the rest of the ENT examination was normal. He was given unrecalled oral antibiotics and an oral decongestant. Pure tone audiometry showed mild-moderate hearing loss, bilateral. The patient underwent ventilation tube insertion for the left ear and remained assymptomatic for 6 years.

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However, 2 years prior to consult, he again experienced 2 months of intermittent left ear pain, associated with hearing loss. The same ENT specialist noted granulation tissue on the postero-superior quadrant of the left tympanic membrane. He was prescribed Garamycin otic drops and Hydrospor. No follow up was made.

1 year prior to consult, the patient consulted for the same symptoms. The left tympanic membrane had beginning retraction pocket on the postero-superior quadrant. A mastoidectomy with tympanoplasty, left, showed retraction pocket with cholesteatoma behind the pars flaccida, the cholesteatoma extending to the Prussak's space and epitympanum, additus and superior portion of the mastoid antrum. The posterior canal wall was taken down and the cholesteatoma was removed piecemeal. No specimen was sent for histopathology. The patient was discharged after two days, experienced relief of pain and was able to go back to his normal activities.

4 months prior to consult, while cleaning his left ear with a cotton pledget, he noted scant bleeding coming out of his left ear, accompanied by otalgia, ear fullness and sudden decrease in hearing. Otoscopic examination revealed a fleshy mass measuring approximately 0.5 X 0.5 cms. with smooth surface at the outer 1/3 of the inferior canal wall. At this point, a CT scan of both mastoids showed moderately enhancing soft tissue density mass within the left mastoid antrum. (Figures 1-3) Surgery revealed a tumor originating from the postero-superior canal. The mass was then excised which, on histologic examination was diagnosed as angiosarcoma. (Figures 4-5).

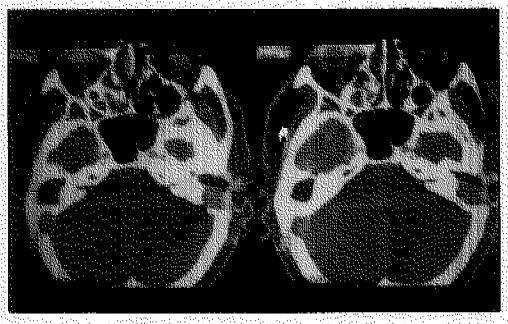

Figure 1 Plain CT scan of the mastoid:

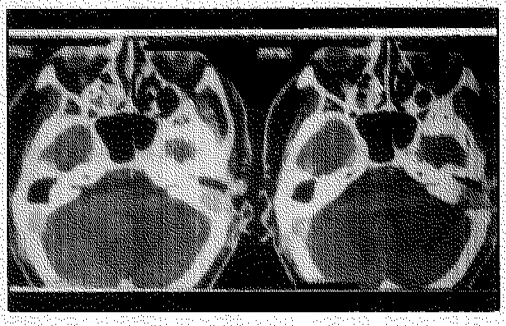

Figure 2 CT scan of the mastoid: with contrast showing) moderately enhancing soft tissue density mass within the left mastoid antrum

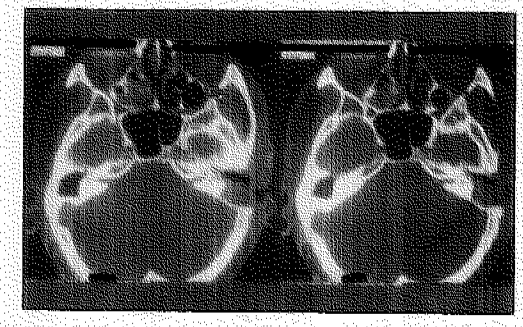

Figure 3 Bone Window. Thinning of the posterior portion of the petrous bone on the left.

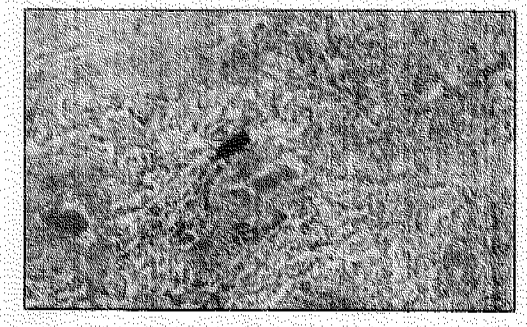

Figure 4. Photomicrograph:of angiosarcoma of the temporal bone, Low Power view) Slit like as well as elongated vascular spaces lined by atypical cells and numerous red blood cells are seen indicating that the mass is highly vascularized.

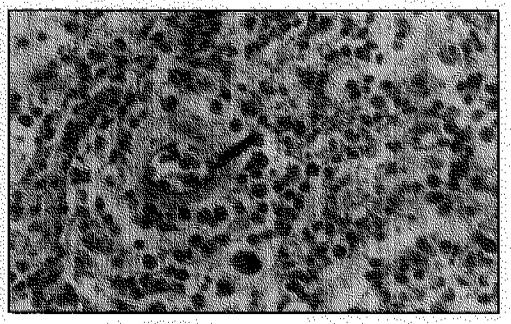

Figure 5. High Power view of angiosarcoma. Enlarged oval nuclei containing prominent nucleoli surrounding a blood vessel can be seen.

Post operative recuperation was uneventful, and the patient was discharged after two days. No medical or oncologic follow-up was made.

2 weeks prior to consult, he again experienced left ear fullness and severe otalgia radiating to the temporal area. Otoscopic examination showed a mass filling the ear canal. MRI showed a hypervascular mass in the left mastoid cavity with thinned out adjacent petrous bone, and with extension into the diploe. (Figure 6) Revision mastoidectomy showed granulation tissue filling the left ear canal and mastoid cavity. The tegmen mastoideum was dehisccent but the dura of the middle fossas was intact. Fibrin glue was then used to seal the defect on the tegmen.

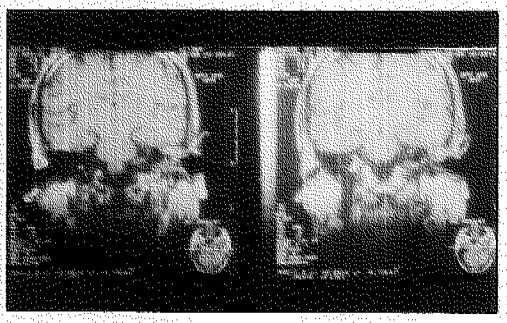

Figure 6. MRI Temporal bone. Hypervascular mass left antrum,with thinned out adjacent petrous bone and mild extension into the diploe.

Histopathologic examination of the ear mass revealed anaplastic carcinoma with chronic inflammation, left mastoid. (Figures 7-8)

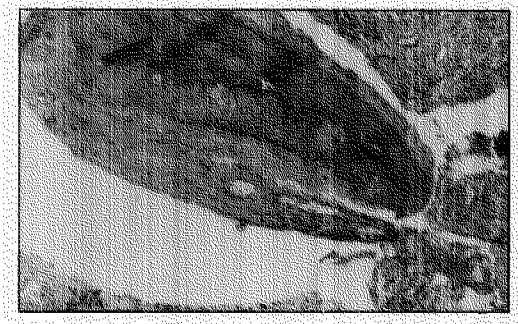

Figure 7. Anaplastic carcinoma of the temporal bone, low power view Presence of numerous chronic inflammatory cells surrounding the anaplastic tumor.

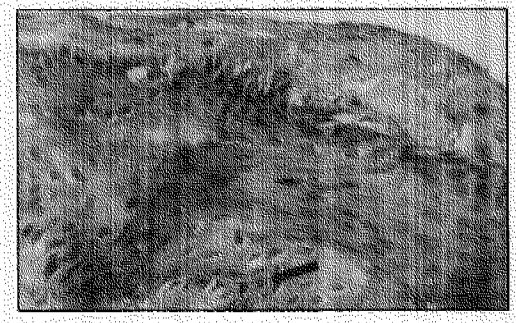

Figure 8. High power view: Anaplastic carcinoma, temporal bone. Abnormal pseudo-stratified enlarged columnar cells with malignant nucleoli. Hemorrhagic areas of necrosis are also seen.

14 years prior to consult he underwent mastoidectomy and tympanoplasty on his right ear for otalgia, hearing loss and chronic mucopurulent discharge. Hearing improved and his right ear remained dry since the above-mentioned surgery.

He was operated twice twenty years prior to consult, at a government hospital for nasal polyposis. Aside from occasional attacks of asthma, there were no other pertinent illnesses noted.

His mother died of breast cancer and his older brother died of complications from lung cancer, however these were not documented histologically.

Right after he was diagnosed to have anaplastic cancer of the temporal bone, he was referred to a medical oncologist and underwent radiotherapy of the left temporal bone for 37 treatment sessions receiving 180cGy daily for a total of 6,600cGy.

He experienced relief of symptoms while undergoing radiotherapy and was again able to resume work.

6 months after histopathologic confirmation, he experienced gradual abdominal enlargement accompanied by intermittent epigastric pain. Bone scan results were normal. (Figure 9) However, CT scan of the abdomen revealed ascites. Nodules and thick strands within the omentum were also noted, together with a mass lesion in the medial segment of the left hepatic lobe, measuring approximately 7 X 4 X 2 cms. all consistent with metastatic seeding. (Figures 10-11)

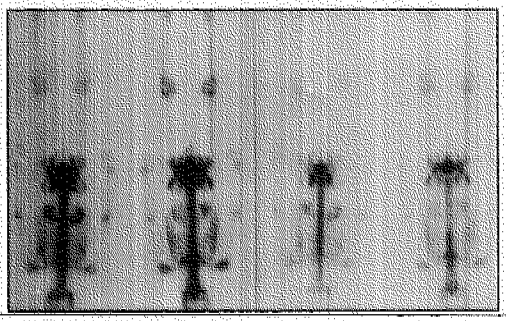

Figure 9. Bone scan. Normal except for arthritic changes.

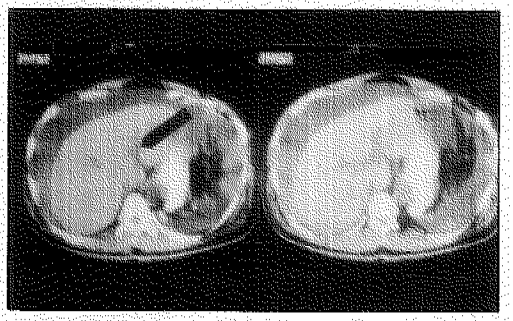

Figure 10. CT scan abdomen. Hepatic mass, medial segment of the left lobe, measuring approximately 7 X 4 X 2 cms.. Massive ascites is also seen surrounding the abdominal structures.

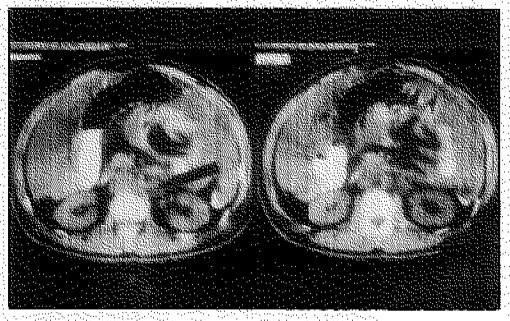

Figure 11. CT scan abdomen. Nodules and thick strands are noted within the omentum. Retroperitoneal lymphadenopathies beside the abdominal aorta is also seen, all consistent with metastases.

No biopsy of the liver mass was done. He was subsequently admitted and intravenous carboplatin was started. There was improvement after the first dose however the second dose was not started due to persistently low platelets count.

7 months after diagnosis, he was admitted for severe periumbilical pain. A CT scan showed massive ascites with bilateral pleural effusion. The liver mass, peritoneal infiltration and abdominal lymphadenopathies remained unchanged from the previous scan. Ultrasound guided paracentesis and Tenchoff catheter insertion drew almost 2 liters of bloody fluid. Cell cytology showed atypical cells.

He was transferred after six days to another hospital for radiotherapy of the abdomen in lieu of chemotherapy. After 2 sessions of radiotherapy, he became anuric. Repeat abdominal ultrasound showed septated fluid over the whole abdomen with irregularly thickened peritoneum, consistent with peritoneal metastases.

8 months from the time he was diagnosed to have anaplastic cancer of the temporal bone; he succumbed to multi-organ failure secondary to abdominal metastases.

DISCUSSION

Histopathologic studies for chronic middle ear surgery is not part of the recommendation from the Philippine Society of Otorhinolaryngology Head and Neck Surgery Clinical Practice Guidelines [4]. The incidence of temporal bone cancer arising as a complication of middle ear disease however is not well documented in local as well as international literature.[5] Nevertheless, the detection of such a cancer depends perhaps as much on the clinical acuity of the surgeon as well as a proper communication to the attending pathologist. The experience and high index of suspicion of both specialists cannot also be over-emphasized.

A patient with a known primary cancer who develops auditory symptoms should be suspected of having metastatic spread to the temporal bone until proven otherwise.[5] Ptients have been reported who have no history of prior ear disease, with onset symptoms resembling acute otitis media and mastoiditis, and during surgery are discovered to have metastatic carcinoma from an occult primary tumor often from the lungs, breast, kidney and prostate[5]. Saldanha et.al. in 1989 also documented a case of a bladder tumor that metastasized to the temporal bone long after the bladder cancer was treated by radiotherapy [6]. However, in the case presented here, no sign or symptom, or laboratory evidence related to the possibility of a distant primary site was present.

All primary middle ear neoplasms are rare with an incidence varying from one per 6,000 to one per 20,000 in patients with chronic ear disease. In Figis's review of middle ear malignancies at Mayo clinic the incidence was 0.003% among clinic patients.[7] The most common malignancies of the ear and temporal bone are still squamous and basal cell cancers.[8] In Conley's series of 100 cancers of the external auditory canal, middle ear and mastoid, 67% were squamous cell cancer. 69% of these tumors originated in the pinna, 22% in the external auditory canal and only 9% in the middle ear and mastoid. [9] Squamous cell carcinoma predominantly arises during the fifth decade of life.[10] Symptoms commonly found on presentation include aural fullness, pruritus and otorrhea. Serosanguinous drainage, deep seated otalgia and cranial neuropathies are ominous signs of a more advanced lesions. [10] Squamous cell carcinomas of the EAC and middle ear are associated with chronic otorrhea, chronic external otitis and cholesteatoma. Proper diagnosis at the earliest stages have been documented to make a lesion most amenable to surgical resection and maximize survival .[10]

Less commonly found tumors of the middle ear are glomus jugulare tumors, sarcomas, benign and malignant Glandular neoplasms (ceruminomas), basal cell carcinoma, and angiosarcoma .[11]

Angiosarcomas are tumors of mesenchymal origin, extremely rare in the head and neck and usually arise in adulthood. The M.D. Anderson Hospital (Houston) recorded only seven cases of angiosarcoma in the head and neck in the span between 1945 and 1963.[12] In a thirty-nine year period, the Memorial Hospital

(New York City) treated only ten cases of this tumor type occurring in the head and neck.[12] They found out that the most common site of origin is the scalp followed by the neck, mouth and antrum. In general, angiosarcomas are rapidly growing neoplasms with an insidous onset and with minimal related symptoms. Regional lymph node involvement occurs during the course of the disease in only thirty to fifty percent of the cases. There is a rapid clinical course; the patient being cured by the primary surgical excision or succumbing to the disease within three years of diagnosis.[12]

Our patient presented with chronic unilateral otorrhea, hearing loss and otalgia, three most common presentations among patients with chronic otitis media, followed by the appearance of a polypoid mass on the left superoposterior external auditory canal, again a frequent and well-documented sequelae of middle ear disease. However, on CT scan there was a mass noted in the left mastoid antrum which enhanced with contrast, accompanied by thinning of the posterior portion of the petrous bone. Hence, an external ear hemangioma was entertained preoperatively. Upon histopathologic examination, however, this seemingly innocent-looking lesion was read by the pathologist as angiosarcoma.

Anaplastic degeneration of a head and neck angiosarcoma has not been reported in our literature review of both local and foreign journals, rendering this case truly reportable if indeed it is such. However, there has been one locally reported case of a squamous cell carcinoma of the temporal bone by Garcia et.al in 1995 at UERM.[13]

Undifferentiated carcinomas are on the other hand composed of obviously malignant cells that by definition grow in sheaths and lack glandular mucinous, papillary, clear cell or squamous differentiation (see Figures 7,8). Distinction from lymphoma and sarcoma is based on the presence of a cohesive growth pattern and reticulin around large cell groups. Additional evidence of epithelial differentiation may be gained by ultrastructural demonstration of numerous primitive junctions between cells and immunohistochemical substantiation of keratin or epithelial membrane antigen expression.[14]

Bony erosion within the temporal bone on CT scan is highly suggestive of a malignancy but is not pathognomonic [5]; and correlates with a poor clinical prognosis. Chronic otitis media, in sharp contrast presents usually with occasional sclerosis of the mastoid antrum; not erosion of the petrous bone.

Nevertheless, biopsy is the single most important laboratory workup that would dictate the difference between chronic otitis media and a malignant neoplasm of the temporal bone presenting with signs and symptoms of a chronic and surgically refractory infection of the middle ear. Hence, it is our recommendation that a sample of the tissues removed pre-operatively or intra-operatively from chronic ears be sent for histopathologic examination, most especially those cases that remain refractory to usual medical and previous surgical therapy.

Microscopically, angiosarcoma is characterized by diffusely infiltrative masses of thin walled vascular channels lined by spindle to oval, swollen cells showing variable degrees of anaplasia.[14] (see Figures 4,5) Histopathologic report of our patient's tissue samples revealed fragments of the same malignant vascular neoplasm. The fragments are composed of slit-like as well as elongated vascular spaces lined by atypical cells with enlarged oval nuclei containing prominent nucleoli which is suggestive of a vascular tumor consistent with angiosarcoma.

Management of temporal bone tumor is still very controversial. Some authors advocate temporal bone resection alone, while others advise radiotherapy and chemotherapy combined, or radiotherapy alone Although there was some early enthusiasm for treatment of temporal bone cancers with radiation therapy alone, most recent series have demonstrated the superiority of combined therapy (surgery and radiotherapy) for epithelial cancers.[15] Sinha and Aziz reported a series of 31 patients treated with a combination of surgery and radiation had a 40 percent 5-year survival rate.[16] This was compared to a 14 per cent survival rate for those treated

with radiation alone. Lewis presented in 1983 a cure rate of 28% for en bloc resection of the temporal bone plus postoperative irradiation out of 132 cases.[17] Regardless of the mode of therapy, diagnosis must be early as management must be aggressive.

The clinical course of temporal bone cancer is a relatively rapid one. These patients may be treated with primary surgical excision of the tumor, followed by post-operative oncologic care, or may succumb to the complications of the disease within 3 years after diagnosis.[12] Demise is associated with lymph node and distant metastases and/or painful and ulcerohemorrhagic local recurrences into fascial planes, bone or cartilage[12] as what happened to our patient. He developed ascites secondary to liver and peritoneal lymph node metastases 6 months after he was diagnosed to have anaplastic cancer (see Figures 10,11). Prognosis is universally poor. In 1975 Lewis reported on 100 patients treated with surgery and post-operative irradiation. The 5 year survival rate was 27 per cent.[17] Wagenfeld reported a 32% four year survival rate in 25 cases of middle ear malignancies.[18]

The Temporal bone is composed of four distinct elements. The petrous portion, the squamous, mastoid and tympanic portion.[10] The foramen of Huschke, a developmental defect in the tympanic ring that normally closes, may remain patent throughout adulthood, which allows malignant processes access out of the bony ear canal anteriorly into the parotid gland and infratemporal fossa, then to lymphatic and venous channels of the head and neck to distant sites.[10] Violation of the tympanic membrane medially by a neoplastic process permits an unimpeded spread to all areas of the petrous bone via the aircell system. The external ear canal drains into the preauricular, parotid, upper cervical and deep internal jugular lymph nodes. The mucosa of the middle ear and mastoid drain into the deep upper jugular and retropharyngeal lymph nodes. Interestingly, the inner ear has no known lymphatic drainage.[10] Malignancies of the temporal bone metastasize via lymphatic and hematogenous spread.

Nevertheless, aggressive surgery and post-operative radiotherapy provides a lifesaving attempt at management, provided no distant metastases are present. While the risks are serious, the patient often has little choice, because untreated carcinoma of the temporal bone is a painful and very noxious way to die.[10]

There are no recorded predisposing factors to the development of anaplastic carcinoma of the head and neck, and temporal bone. However, studies have shown that chronic suppuration of the middle ear may be a significant factor in the development of middle ear carcinoma. It is also believed that continued purulent discharge produces a chemical - toxic irritation of the epithelium of the middle ear which results in the development of carcinoma.[13]

CONCLUSION

Any patient presenting with recurrent otorrhea, granulation and ulcerations of the external auditory canal, mastoid tenderness, deep ear pain, ear fullness and decrease auditory acuity that all remain peculiarly refractory to usual medical and surgical therapy requires a thorough otolaryngologic work-up geared at ruling out the possibility of a malignant neoplasm. The clinician, most especially the otolaryngologist must be aware and respectful of the common and not so common conditions affecting the temporal bone which may mimic chronic inflammatory disease of the middle ear. Early suspicion obviously may spell the difference between life and death for the patient, and not to mention, legal implications for the surgeon.

Hence, temporal bone cancer although rare should be considered among the differential diagnoses in patients presenting with the triad of recurrent otorrhea, deep ear pain and decrease auditory acuity, refractory to the usual medical and surgical therapy.

CT scan of the temporal bone with contrast studies ought to be requested for those cases presenting with recurrent granulation tissues or polyp in the external ear canal and middle ear after adequate surgical extirpation. The presence of bony erosions in the mastoid and external ear

canal should raise suspicion as to the presence of a malignancy.

Routine biopsy (pre-operative or intra-operative) of all granulomatous and neoplastic diseases of the ear is essential for early detection, and hence management of temporal bone cancers. A good working communication between the surgeon, the clinical pathologist and the radiation oncologist is essential for the long-term survival of the patient.

Temporal bone resection followed by radiotherapy remains to be the treatment of choice for middle ear malignancies. Radiotherapy alone has not been documented to give promising results for middle ear tumors.

Lastly, retrospective studies ought to be encouraged locally, and a diligent search for predisposing factors to the development of temporal bone malignancies should be made so as to minimize the incidence of this highly aggressive tumor, although already rare.

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Case Report

Recurrent Meningitis and Cerebrospinal Otorhinorrhea In A Pediatric Patient With Inner Ear Dysplasia

Santos, Regie Lyn P, MD* , Chiong, Charlotte M, MD**

Objective: To present the first documented case of congenital perilymphatic fistula with Mondini dysplasia, presenting with recurrent meningitis, cerebrospinal fluid (CSF) otorhinorrhea and unilateral profound sensorineural hearing loss. The report also aims to explain the anatomicopathophysiologic mechanisms behind the disease. An algorithm of management is likewise proposed.
Design: Case report
Setting: Tertiary care center
Results: A pediatric patient with a history of recurrent meningitis, hearing loss and CSF otorhinorrhea on CT scan of the temporal bone showed an enlarged vestibule and vestibular aqueduct on the left, with apparent communication with the internal auditory canal. The cochlea also appeared to have decreased basal turns consistent with Mondini dysplasia. Postauricular transtympanic repair of the perilymphatic fistula was done. Intraoperative findings include a congenitally absent footplate, an abnormal round window, and a CSF gusher at both oval and round windows, therefore confirming the presence of congenital perilymphatic fistula.
Conclusion: The presence of a congenital perilymphatic fistula with an inner ear abnormality should be highly considered when presented with a case of recurrent meningitis with sensorineural hearing loss. If the index of suspicion is high, high-resolution CT of the temporal bone and exploratory surgery may be enough to clinch the diagnosis. Early detection can result in a more timely repair thus preventing recurrence of life-threatening CNS infection and also increasing the likelihood of abating progression of the hearing loss.

INTRODUCTION

Congenital perilymphatic fistulas are rarely encountered clinical entities. Upon review of the foreign literature, only case reports or case series of the said condition can be found, with no real statistics as to its incidence. Usually these patients present with recurrent meningitis, and isolation of the inner ear as the focus may be chanced upon only after repeated bouts of life-threatening infection. To get to that focus, the battery of tests that the patient undergoes may be both psychologically and economically draining. Though diagnosis remains difficult, an awareness of the condition can highly increase the likelihood of early recognition of the disease and its timely management.

But the journey does not stop here. Should the inner ear be pinpointed as the culprit, there is a wide array of congenital inner ear anomalies to choose from which may present in multiple combinations. The documentation for the exact anatomic and physiologic pathway is not a purely academic exercise because a clear understanding of the pathway of the fistula guides the surgeon towards more effective management.

In this report we describe a case of congenital perilymphatic fistula presenting with recurrent meningitis, CSF otorhinorrhea and unilateral profound sensorineural hearing loss. Upon workup, the patient is proven to have a unilateral Mondini dysplasia which, to our knowledge, is the first reported case in the local literature. So that deeper understanding of the mechanisms can be achieved, the relevant temporal bone anatomy is also reviewed. Lastly a treatment algorithm is proposed.

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CLINICAL DATA

We present the case of R.V., a 7-year old female from Negros Occidental, who was admitted on March 24, 1998 for the chief complaint of watery discharge from the left ear. Her history started three years prior to admission when she experienced high-grade fever (undocumented), vomiting, generalized headache and increased sleeping time. She was brought to a local hospital where a lumbar tap was done. The assessment was bacterial meningitis and thus she was given IV ceftriaxone for 10 days. She was discharged asymptomatic with INH and rifampicin as home medications for nine months.

One year prior to admission, there was recurrence of the fever, vomiting, and headache and increased sleeping time. She was admitted at another hospital, where again the lumbar tap showed bacterial meningitis. A CT scan was read as otitis media. She received IV ceftriaxone for ten days and was discharged asymptomatic.

Three months prior to admission there was recurrence of the same symptoms, this time, however, with clear, watery non-foul smelling discharge from both nostrils and the left ear. She was then confined at the previous hospital with a diagnosis of CSF leak. A ventilation tube was inserted into the left tympanic membrane which relieved the rhinorrhea but not the left-sided otorrhea. She was initially maintained on IV chloramphenicol, and then later shifted to IV cefuroxime. She was then referred to Manila for further management.

On review of systems, the patient has had hearing loss on the left ear since early childhood. The patient has never had any symptoms of vertigo, imbalance nor tinnitus. She has no previous history of head trauma, surgery, measles or mumps. She has not had any purulent ear or nasal discharge. No complaints were localized to the nose or throat. The patient also has no previous history of blood dyscrasia nor syndrome complex.

The patient has no history of deafness or similar illness in the family. She was born full term via spontaneous vaginal delivery to a 35-year old G5P2 (7-0-2-1) at a local hospital with no perinatal complications. There was no maternal illness nor drug intake during pregnancy. The patient's immunization is complete for her age which includes vaccination for measles. The child's developmental history is at par with her age, presently being in Grade I with high marks.

Physical examination of the ear revealed a dull thickened tympanic membrane on the left with a ventilation tube inserted on the anteroinferior quadrant. There was also pooling of clear watery discharge at the external auditory canal. Aside from Grade I tonsillar hypertrophy on the left, the rest of the head and neck findings were unremarkable. Systemic and neurologic examinations were likewise normal.

Pure tone audiometry (Fig. 1) revealed normal hearing acuity on the right while the left ear had profound sensorineural hearing loss.

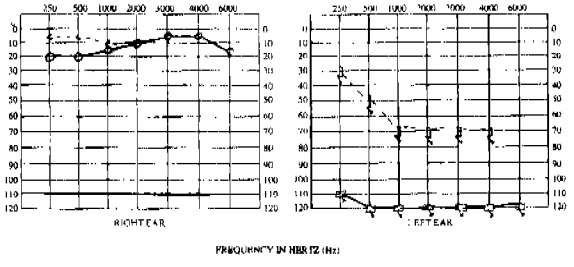


Figure 1 Pure tone audiometry curves, right and left ears

CT scan of the head with 4 mm cuts showed a mildly enlarged left vestibule and vestibular aqueduct. There is also an apparent communication between the vestibule and internal auditory canal. The cochlea on the left seems to have less septations compared to the right, with the left lacking one basal turn. The tympanic membrane was also noted to be thickened. (Fig. 2 a-c). Assessment upon admission was recurrent meningitis secondary to perilymphatic fistula with Mondini (inner ear) dysplasia, left S/P ventilation tube insertion, left. The patient was then admitted for surgery.

On the third hospital day, the patient underwent postauricular transtympanic repair of perilymphatic fistula, AS. On operation, the round window was confirmed to have CSF leak. The RW membrane was well in view and not hidden in a niche. The oval window was also confirmed to have a CSF leak. Repeated aspirations were followed by reaccumulation of clear fluid in both windows.

Repair of the perilymphatic fistula was performed by packing the oval and round windows with gelfoam and areolar tissue. The eustachian tube was likewise obliterated with muscle and areolar tissue. The ventilation tube was also removed with the TM perforation covered with a medial underlay graft. Post-operatively, the patient's course was unremarkable.

Figure 2a. Coronal cut showing the left cochlea with less basal turns compared to the right.

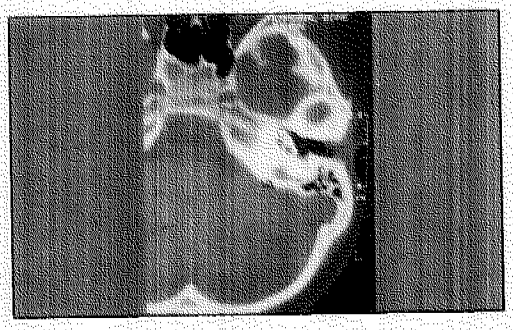


Figure 2b. Axial view showing cochlea (C) and enlarged vestibular aqueduct (VA). The tympanic membrane is shown to have the ventilation tube.

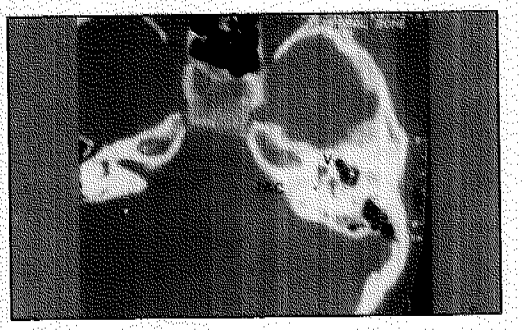


Figure 2c. The internal auditory canal (IAC) seems to have a communication (*) with the vestibule (V).

DISCUSSION

In 1976 Parisier and Birken reviewed 15 cases of recurrent meningitis in children also manifesting with otorrhea. The mean age of the patients was 6.4 years, with males and females equally affected. The otorrhea was unilateral in 2/3, bilateral in the rest. In most of the patients, vestibular symptoms were absent. Congenital ear anomalies were found in 11 out of 15 with Mondini dysplasia in at least four. The infecting organism was pneumococcus in most, which Fee explains is abundant in the middle ear mucosa.

Another case series by Quiney et al (1994) had 7 children with recurrent meningitis, later proven to have inner ear abnormalities by CT scan. Weber, Perez and Bluestone (1993) also reported that 16 out of 94 patients (17%) who underwent exploratory tympanotomy for perilymphatic fistula had inner ear anomalies. Reilly in 1989 and Fisher and Curtin in 1994 showed that 20-23% of patients presenting with progressive sensorineural hearing loss had inner ear malformations by CT scan.

Thus in a case of recurrent meningitis with hearing loss, when all other etiologic factors (eg. trauma, anatomical meningeal defects, parameningeal infection such as from the ear or sinuses, immunodeficiency states) have been ruled out, the likelihood of an inner ear abnormality with a perilymphatic fistula increases.

Quiney et al (1989) cites several reasons why inner ear abnormalities in children with CSF fistula are usually misdiagnosed. One reason is that the CSF

rhinorrhea may be mistaken for normal nasal discharge. Likewise CSF otorrhea or middle ear effusion may be diagnosed as otitis media and treated as such. CSF otorhinorrhea is rare as a presenting symptom in CSF fistulas since the drainage is usually down through the Eustachian tube. Also if the patient has only one ear with hearing loss, the child may compensate and thus may not complain of any deafness. As in this case, the deafness would not have been worked up had the patient not presented with other symptoms. Lastly, during documentation, the inner ear abnormality may not be demonstrated due to inadequate cuts with the CT.

A perilymphatic fistula is defined as an abnormal communication between the middle ear and the inner ear space. Studies show that the oval window is the most common pathway for this leak (Parisier and Birken, 1976; Quiney et al, 1994). However leaks from the round window can also occur, although they are less common (Bauer, 1962). Weber, Perez and Bluestone in 1993 further described the anatomic anomalies in the middle ear associated with perilymphatic fistulas. They found that stapes malformations were the most common congenital anomalies in the middle ear for patients with perilymphatic fistulas. It must be remembered that these malformations can occur even without identified conductive hearing loss. In terms of fistula formation, the authors postulate that "thinning out" around the anterior portion of the footplate may correspond to weak areas that are prone to fistula formation. This occurs in conjunction with transmission of increased pressure from the CSF compartment onto the inner ear surface of the footplate, gradually causing erosion or displacement. In this case the footplate was absent with only a membrane remaining. Therefore it is not difficult to imagine that constant pressure of CSF may result in ectasia and spontaneous rupture of this membrane.

Round window abnormalities can also occur in perilymphatic fistulas. The round window may lie in a more lateral rather than posterior orientation or a more oblong than round shape. In this case the round window was indeed noted to face laterally with no niche or bony shelf.

It is important to ascertain if the leak is confined just in the perilymphatic space or if it extends to the CSF compartment. Since the patient presented with recurrent meningitis and otorhinorrhea, one would suspect CSF leak; on the other hand, if the patient had mainly hearing loss and vestibular symptoms, the anomaly may be limited to the perilymphatic space.

Furthermore, Parisier and Birken (1976) described the occurrence of a "slow leak" when the patient simply had a recurrent meningitis. On the other hand the occurrence of CSF otorhinorrhea on top of the meningitis would mean a "fast leak". To understand this consider that there are four pathways through which the CSF and perilymph communicate: (1) cochlear aqueduct (2) lamina cribrosa and cochlear modiolus (3) vestibular aqueduct (4) internal auditory canal. If the leak were "slow", a smaller structure, such as the cochlear aqueduct, would probably be the source of the leak. However a fast leak would be caused by a more extensive violation of partitions, such as when the fundus of the internal auditory canal opens into the vestibule and / or the cochlear modiolus. In this case the presence of otorhinorrhea and the apparent communication between the vestibule and internal auditory canal by CT supports the suspicion of a large or "fast" CSF leak.

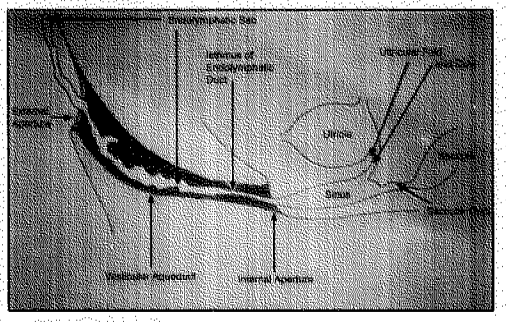


Fig. 3. Diagram of CSF pathways

The occurrence of hearing loss in a perilymphatic fistula is not explained simply by an abnormal communication between the perilymph compartment and the middle ear. Several theories have been put forth to explain this. Some authors have stated that decompression of the

perilymphatic space results in a secondary endolymphatic hydrops (Allen 1983, Walsted et al 1991, Syms et al, 1990) but tests to confirm perilymphatic fistula formation as a cause of hearing loss have been inconclusive (Meyerhoff and Marple, 1994). Simmons (1979) proposed that rapid shifting of pressures across Reissner's membrane could result in its rupture, altering the normal fluid balance within the cochlea with mixing of the perilymph and endolymph. In the present case, hearing loss might have occurred following recurrent bouts of meningitis where inflammatory cells find their way to the perilymphatic space via a patent cochlear aqueduct or lamina cribrosa. This may result in the onset of new bone formation that can slowly obliterate the cochlea,

In a prospective study involving patients undergoing exploratory tympanotomy for suspected perilymphatic fistula, Podoshin et al (1994) used clinical history, past medical history, hearing function and vestibular function as variables to test which among these four had the highest positive predictive value for perilymphatic fistula. They concluded that there was no single test that could be considered diagnostic for perilymphatic fistula.

Regarding the value of diagnostic myringotomy, some clinicians advocate insertion of a ventilation tube to facilitate specimen collection for CSF studies (Parisier and Birken 1976). This may be true if the focus for the meningitis is still at large, and the symptoms do not point clearly towards a CSF leak. However if the case is as clear-cut as this, the myringotomy may not be necessary, especially with the availability of CT scans to demonstrate any anatomic defect within the inner ear.

When the index of suspicion for a perilymphatic fistula and/or an inner ear abnormality is high, CT scan of the temporal bone with 1.0-1.5 mm cuts, axial and coronal, and with bone windows is used as a diagnostic test. In this case the CT scan showed a dilated and enlarged vestibule and vestibular aqueduct on the left side. Valvasorri in 1983 studied 160 cases of large vestibular aqueducts, 24 of which had associated anomalies of the vestibule, semicircular canals and cochlea, corresponding to the Mondini defect. It is now generally accepted that vestibular enlargement and Mondini dysplasia are part of a spectrum of inner ear anomalies.

The classical finding in Mondini dysplasia is the lack of septations within the cochlea by CT since the normal 2 ½ turns is decreased to 1 ½. It may also appear as a cystlike cavity without partitions, which may also represent abnormal communication with the vestibule and lateral semicircular canal. (Fig, 7 a-c) As a rule, the appearance of the inner ear by CT scan correlates with embryonic development. Thus the Mondini defect occurs at the time when the cochlear duct lengthens and begins to coil, which is at week 7-8. The patient's CT scan shows only 1 ½ basal turns on the left.

As previously mentioned, the CSF leak in this case most probably occurred from the internal auditory canal into the vestibule. With the presence of a Mondini defect, the pathway of the leak becomes clearer when we imagine the modiolus and the central bony spiral lamina to be absent or deficient. These parts of the cochlea should come in contact with the fundus of the internal auditory canal, therefore establishing the leak.

Ultimately it is direct visualization of the leak by exposing the oval and round windows via an exploratory tympanotomy, which clinches the diagnosis of a perilymphatic fistula. There are two main clinical observations that may indicate the possible pathogenetic mechanism. A perilymph "oozer" implies small defects in the fundus of the cochlear aqueduct. In the temporal bone collection of the Massachusetts Eye and Ear Infirmary, the widest diameter with the narrowest portion was noted to be 0.2 mm. (Fig. 8) On the other hand a perilymph or CSF "gusher" would mean larger defects in the fundus of the internal auditory canal (Shuknecht and Reisser, 1988). In this case the perilymphatic fistula was clearly demonstrated during surgery by clear fluid "gushing" out of the round and oval windows. This leak was controlled by packing the leak with subcutaneous areolar tissue and gelfoam. Though surgery is the gold standard for diagnosis, it is not

entirely foolproof since transudate, irrigation or anesthetic that pooled within the window niche can be mistaken for CSF or perilymph (Meyerhoff and Marple, 1994).

Several methods have been proposed to solve this problem. Intravenous flourescein has been used; however it was later proven to be an unreliable marker (Poe et al, 1993; Bojrab and Bhansali, 1993). Free amino acid assay on the other hand has been considered too complex (Levenson et al, 1996). Beta-2 transferrin has been shown to be highly sensitive and specific for CSF, and is being suggested for use intraoperatively. However the qualitative dipstick method used by Silverstein is very prone to cross-contamination (Thalmann et al 1994). The time required to perform the test is also considered a hindrance (Meyerhoff and Marple, 1994).

The choice of surgical approach in this case was guided by the tenet that local anesthetic, which is used with the exploratory tympanotomy approach, can be avoided with the postauricular transcanal approach. The latter approach precludes the use of local anesthesia which may falsely present as pooling in the windows. In addition a mastoidectomy can be easily done when other areas of CSF leak are found. Gacek and Leipzig (1979) noted that, other than the windows, other areas from which CSF otorrhea can occur are the facial canal, tympanomeningeal fissure and petromastoid canal.

Repair materials include muscle, perichondrium, autogenous blood or areolar tissue. The critical issue is meticulous placement of these materials and assuring enough flexibility so as to effectively seat these materials and close the leak.

Post-operatively the patient is advised to have careful gradual ambulation with the head elevated at all times. Stool, Leeds and Shulman (1967) reported an 8-42% incidence of improvement in hearing among patients who underwent surgical repair for perilymphatic fistula. However this may not apply if the patient is profoundly deaf. In this case it was stressed to the patient's family that the goal of surgery is not to correct the hearing loss which is irreversible. Rather it is to control the leak which leads to the life-threatening condition of recurrent meningitis. Since Mondini patients usually have compensated audiologic and vestibular functions, their quality of life is high.

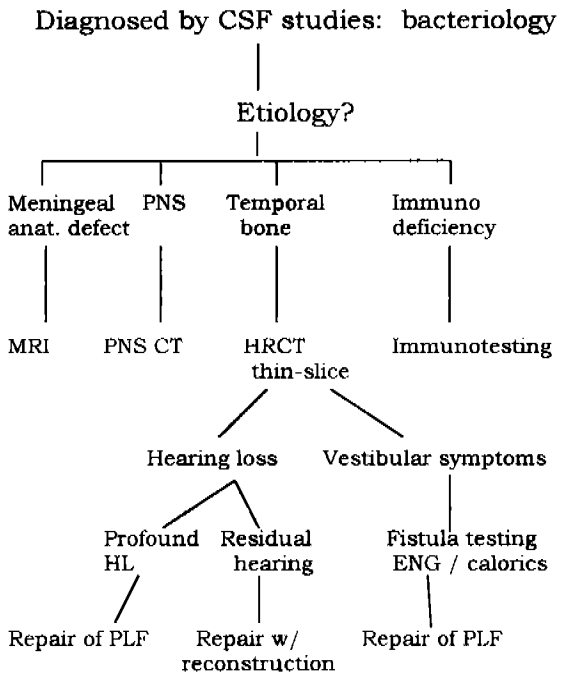
CONCLUSION

This is the first reported case of congenital perilymphatic fistula with Mondini dysplasia in the Philippines. In this child with recurrent meningitis and CSF otorhinorrhea, audiometric documentation of unilateral profound sensorineural hearing loss and high-resolution CT scan that demonstrated cochlear dysplasia aided in the diagnosis. The intraoperative finding of CSF "gusher" virtually confirmed the presence of the fistula with clear identification of leaks from the oval and round windows. A combined postauricular and transcanal surgical approach was guided by the preoperative evaluation and provided an efficacious manner of repair of the perilymphatic fistula.

This case has several implications. Since the identification of the focus of infection in recurrent meningitis may be taxing at times, an awareness of the clinical picture of this case would open many possibilities to the surgeon. A high index of suspicion for congenital inner ear anomalies and perilymphatic fistulas should be aroused when presented with recurrent meningitis combined with unilateral sensorineural hearing loss with no antecedent cause. In such case a high-resolution CT of the temporal bone becomes invaluable and exploratory surgery may be indicated.

Based on this experience, we propose the following treatment algorithm:

RECURRENT MENINGITIS



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