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WIDENING SCOPE OF OTORHINOLARYNGOLOGY PRACTICE IN EAST AFRICA

I thank the Editor-in-Chief of the *East and Central African Journal of Otolaryngology, Head and Neck Surgery* Dr. Sophie Gitonga for the honour and privilege she has bestowed on me by requesting I be the guest editor for the journal's second edition.

The purpose of a journal can never be understated especially in the scientific realm. Apart from show casing the intellectual prowess of the researchers, journals also gather, store and disseminate knowledge. Consequently medical research and publication leads to new discoveries and gives birth to diagnostic and therapeutic innovations that ultimately lead to improvement in patient care and interventional outcomes. It's a well-known fact that there are very few scientific publications from Africa. *The East and Central African Journal of Otolaryngology Head and Neck Surgery* will serve as a spring board from where local and regional otolaryngologists can showcase their intellectual might to the rest of the world.

This second edition has 6 original research articles that are very exciting and relevant to the otolaryngologist and indeed the physician from the developing world. Two articles explore the relationship between hypertension and ear disorders. Maina *et al*[1] have looked at otologic manifestations in patients with autoimmune disorders. It should be noted that whereas this relationship between a systemic condition and ear disorders has been documented in numerous studies worldwide, this is the first of its type in this part of Africa.

Similarly the next two articles by Mwangi *et al*[2] and Muthoka *et al*[3] represent areas that have been studied widely in other parts of the world. However, data from sub-Saharan Africa is sparse and therefore these two studies also bring forth new knowledge as they look at patients from a different and distinct demographic group.

Currently there has been a shift towards use of quality of life questionnaires in most of the chronic illnesses in order to gauge how the diseases impact on the patient's day to day life. Whereas these questionnaires arise from different parts of the world thus in the language from that part of the world, this study by Warui *et al*[4] has translated into Kiswahili and validated the OSA-18 questionnaire.

Xerostomia is a common complaint in patients with oral cavity cancer presenting in the ENT outpatient clinics following surgery and / or radiotherapy. This article by Mutiso and Hwang[5] shows that submandibular gland invasion in oral cancer including advanced oral cancer is rare, therefore sparing of this gland during surgery or radiotherapy is advocated to reduce xerostomia. Finally the article by Magabe *et al*[6] is a case report of severe post tonsilectomy haemorrhage requiring coil embolization. Tonsillectomy is one of the most common surgical procedures performed by ENT surgeons worldwide. Post tonsillectomy bleeding is an expected complication that can and does occur either immediate post-operative period or 1-2 weeks after surgery. Therefore, possibility of encountering severe post tonsillectomy haemorrhage should never escape the mind of every ENT surgeon while performing this common surgery.

I finally note that all these studies have been done in one tertiary hospital in Kenya and I therefore take this opportunity to challenge colleagues from other hospitals and institutions to actively participate in scientific research and publishing.

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RESEARCH ARTICLE

OTOLOGIC MANIFESTATIONS AMONG ADULT PATIENTS WITH AUTOIMMUNE DISEASES AT A TERTIARY HOSPITAL IN KENYA: A CASE CONTROL STUDY

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ABSTRACT

Background: Autoimmune diseases are a consequence of an altered function of the immune system and can involve a single or multiple organs. Aetiology is unknown with genetic factors being a major component usually triggered by an infectious agent or environmental exposure. Otologic manifestations of autoimmune diseases represent a diagnostic challenge and are often ignored. They range from hearing disorders to balance disturbances. This study attempted to determine the otologic manifestations of autoimmune diseases amongst patients attending the Rheumatology clinic at a tertiary health facility in Kenya.

Methods: This was a case control study matched by sex and age. Cumulative sampling was carried out with estimated sample size of 130 adults (65 cases and 65 controls). Cases were patients with confirmed rheumatoid arthritis and systemic lupus erythematosus and controls were recruited from the dental clinic. For continuous data mean standard deviation was calculated. Frequency distribution was used to summarize the categorical variables. Bivariate analysis was conducted to identify the manifestation of otologic features according to case and control status. All otologic manifestations that were significantly associated with auto immune disease status in the bivariate analysis by Pearson's Chi-square were included in multivariable logistic regression models.

Results: Hearing loss was found to be 10 times more likely in the autoimmune group (13.8%) versus the control group (1.5%) ($p = 0.002$) as a complaint. Abnormal audiometry was recorded in 12.3% of the cases and 3.1% of controls ($p = 0.067$) while abnormal tympanograms were recorded in 33.8% of cases versus 7.7% of controls ($p = 0.001$). There was no association found between duration of autoimmune disease and medication used with acquired ear disorders.

Conclusion: Patients with autoimmune disorders are more likely to have otologic disorders with resultant abnormal audiometric and tympanometry findings.

INTRODUCTION

Pathogenesis of autoimmune disorders arises from the activation of B and T lymphocytes reacting against antigens of the body's own tissues. An autoimmune response, usually induced by a triggering event, may be previously B or T cell mediated or both[1]. The most commonly diagnosed autoimmune diseases in Kenya are Rheumatoid Arthritis (RA) and Systemic Lupus Erythematosus (SLE)[2]. Rheumatoid arthritis is a common autoimmune systemic inflammatory disease affecting approximately 1% of the world population and involves many genes[2]. SLE is a chronic inflammatory disease that has protean manifestations and follows patterns of exacerbations and remissions[3,4]. Rheumatoid arthritis will present with symptoms of overt joint inflammation and swelling as persistent systemic polyarthritis[5]. Diagnosis is made according to the 2010 American College of Rheumatology/European League of Associations for Rheumatology (ACR/EULAR)[6] classification.

Various otologic manifestations can occur. They are common with severe active disease and have a higher incidence in men who have RA even though women are the most affected. Arterial stiffness and thickening of blood vessels is an important factor in RA[7]. For neuritis, it presents as a diffuse sensorimotor neuropathy with the underlying mechanism being small vessel vasculitis of the vasa vasorum of the nerves with ischemic neuropathy and demyelinization. Ototoxicity by drugs used in the treatment of RA has also been suggested though most researchers have failed to prove if use of salicylates independently causes hearing loss[8-10]. Systemic lupus erythematosus will have immune complexes which attack the ear. Wegener's granulomatosis causes serous otitis media in as high as 90% with symptoms of Conductive Hearing Loss (CHL), Sensori-Neural Hearing Loss (SNHL) (90%), otalgia, otorrhea being reported[11-13].

Several ENT manifestations in different autoimmune diseases have been described ranging from hearing and nasal disturbances, oral/mucosal ulcerations, laryngeal manifestations, salivary gland disorders and cranial nerve involvement. A review done by Papadimitraki *et al*[14] in 2004 describes various head and neck manifestations that occur in the setting of autoimmune diseases. Thirty-seven patients with RA were investigated with thirty-five controls in a study done to determine the prevalence and nature of HL in RA. This study found 35.5% had SNHL vs 5.7% of controls, 24.3% had CHL vs 5.7% of controls and 10.8% had mixed HL and none of the controls. Abnormal tympanograms were seen in 37.9% whilst the controls were 17.1%. Involvement of SNHL in RA was in the cochlea and CHL was due to discontinuity of ossicles[15]. In a study done in Turkey in 2011 where they had 44 patients with RA and 44 voluntary healthy controls, HL was found in 27.3% of patients compared with 15.9% of controls. Sub-clinical CHL was seen in 56.8% of the patients and 25% of controls and the reason was due to stiffness of ossicular chain[16]. This was in contrast to a previous study which had shown it was due to discontinuity of the ossicles[15]. RA even in its early stages when distinct hearing loss is not present may influence middle ear mechanisms and low frequencies by increase in stiffness rather than discontinuity.

Fifty patients with RA were matched with fifty controls for age and sex in a survey of relationship between RA and hearing disorders. PTA, speech discrimination, tympanometry, acoustic reflex decay and tonal decay were done. The evaluation of sensorineural hearing loss showed that the hearing loss was sensory. Patients were not aware of their hearing loss and did not have clinical complaints[17]. A comparative case control study done on hearing impairment in patients with RA was performed with 194 patients with 107 healthy controls and was by far the study with largest number of patients. The researchers averaged tonal audiometers for 7 frequencies. Hearing loss was detected in 42.7% of patients with RA compared to 15.9% with sensorineural type of hearing loss detected in 38.6% of the cases[18].

A case control study among 29 patients and 30 controls with RA was done with the disease duration being greater than 5 years. The percentage of patients with hearing loss was 45% vs 40% were found among controls with SNHL, 10% vs 7% were found among controls with CHL, 3% vs 0% were found among controls with mixed HL. Acoustic reflex threshold was 17% vs 7% were found among controls[19].

A study done prospectively evaluated 43 SLE patients in Greece. HL was found in nine patients (22.5%). More specifically 8 (21.5%) patients presented with SNHL and only one (2.63%) had conductive hearing loss. A quarter of SLE patients presented with SNHL, affecting mainly the middle and high frequencies, while only one had CHL[20]. Although most SLE patients were exposed to potentially ototoxic medication, the asymmetric results observed in 15/26 (56%) argued against ototoxicity as the cause of aural symptoms.

In a study done in patients with systemic lupus erythematosus, 20 female patients with SLE were selected and matched with 20 healthy subjects matched by age and sex. PTA was abnormal in 13 (65%) patients; 4 had tinnitus and 1 vertigo. All controls had normal hearing. The PTA results showed a highly significant statistical difference from the controls[21].

In an Iranian case control study on 45 patients, 5 patients (11.1%) complained of HL compared to 1 (2.2%) in the control group, 4 (8.9%) patients complained of otorrhea while none in the control group was found to have otorrhea, 3 (6.7%) patients had tinnitus whilst none in the control group had tinnitus. Twelve patients in the case group and 4 (8.8%) patients in the control group had SNHL and the difference was found to be statistically significant[22].

A study in asymptomatic sensorineural hearing loss in patients with SLE was done in thirty-one females matched to twenty-five healthy age matched women as controls. Twenty-one (70%) had impaired hearing, twenty (66%) had sensorineural loss at high frequencies in a bilateral and symmetric way and one had conductive alteration. Four controls had alterations in their audiometric tests, three had conductive alterations and one had sensorineural hearing loss[23].

The objective of this study was to determine prevalence of otologic manifestations (vertigo, otorrhea and tinnitus), demographic patterns associated with otologic manifestations and to determine if there is an association between duration of autoimmune condition and severity of otologic manifestations among patients with autoimmune diseases at a tertiary health facility in Kenya (Kenyatta National Hospital).

MATERIALS AND METHODS

This was a case-control study which was matched for sex and age. Participants were sampled from the Rheumatology clinic while controls were recruited from the Dental clinic. Sample size was calculated with a power of 80% and 95% confidence interval using the formula by Kirkwood[24]. Proportions were based on two studies carried out previously[25,26]. Calculated sample size was 130 patients (65 cases and 65 controls).

Inclusion criteria:

1. Patients with confirmed Rheumatoid Arthritis (RA) as per the 2010 American College of Rheumatology (ACR) classification and Systemic Lupus Erythematosus (SLE) as per the 1982 ACR classification.
2. Controls with no autoimmune disease.
3. Patients with autoimmune diseases and controls who consented to be included in the study.

Exclusion criteria

1. Patients with congenital hearing loss.
2. Patients with history of occupational noise exposure e.g. factory workers.

3. Patients with previously known systemic infections-meningitis, syphilis, VZV, herpes, mumps, measles.
4. Patients with known co morbid conditions – diabetes, hypertension, HIV, sickle cell disease, chronic renal failure.
5. Patients with previous ear surgery or temporal bone trauma.
6. Patients with previous known use of ototoxic agents – streptomycin, gentamicin, loop diuretics, cisplatin

A standardized structured questionnaire was used to record patients and controls bio-data and socio-demographic characteristics. Ear examination was carried out by the principal investigator which included inspection and tuning fork tests. Pure Tone Audiometry (PTA) was performed using Interacoustics Clinical Audiometer AC33. Threshold determination was done by beginning with the better ear at 1000Hz and a clear audible signal of 60db was presented to the ear. Bone conduction was done by placing a thin metal head band attached to a small bone vibrator on the patients head so that the vibrator part rested on the mastoid bone. This was done if there was a difference of 25db or greater in hearing levels between the two ears. Masking was done when indicated. Tympanometry was then done using the tympanometer AT235 Impedance Audiometer.

Data analysis: This was conducted using SPSS® version 18, and was developed in three stages; univariate descriptive, bivariate and multivariable analysis. During the initial descriptive analysis, each variable in the database was analyzed, in turn. For continuous data including age, mean and standard deviation was calculated. Frequency distribution was used to summarize the categorical variables (e.g. hearing loss). Bivariate analysis was then conducted to identify the manifestation of otologic features according to case and control status. Pearson's Chi square test for independence was used to determine which otological manifestations were associated with case control status (autoimmune disease). A cut off value of 0.05 was used to determine statistical significance. Finally, all otologic manifestations that were significantly associated with auto immune disease status in the bivariate analysis were included in multivariable logistic regression models to determine the independent otologic manifestations of auto immune disease status. The coefficients from the regression models were reported as Odds Ratios with the corresponding 95% confidence intervals.

Ethical consideration: Approval was given by the Kenyatta National Hospital (KNH) Ethics Committee (reference no: P579/08/2016). No extra cost was incurred by participants. Written and verbal consent was taken from each participant.

RESULTS

Demographic patterns: The sample comprised a total of 130 patients, 65 cases and 65 controls. The female to male ratio of cases of autoimmune diseases in KNH was 15:1 with 61 females (93.8%) and 4 males (6.2%) recruited as cases and followed by a corresponding ratio of age-sex matched controls. The mean age of the cases was 40.56 years similar to the controls. The modal age group was 41 – 64 years among both cases and controls (52.3%). The discrepancy of the case control modal group of 18 – 30 years and 31 – 40 years was due to overlap in matching ages of 29 -31 years. The majority of the cases were RA who were 33 cases (50.79%) followed by SLE who were 22 cases (33.84%). Ten cases (15.38%) represented other types of autoimmune disorders which were polyarthralgia, small cell vasculitis and systemic sclerosis. Average duration of disease was 4.67 years with a range of 1-22 years.

Medication: The medication commonly used was methotrexate with 37 (56.9%) cases , prednisolone with 35 (53%) cases , hydroxychloroquine sulfate 31 (47%) cases and mycophenolate mofetil 6 (7.6%) cases. NSAIDs were taken variably depending on whether they had flare ups of pain thus not consistently used.

Clinical evaluation: The history of otologic symptoms was reported in 20 (26.2%) cases compared to 3 (3.1%) controls (p = 0.029). Hearing loss was reported in 9 (13.8%) cases compared to 1 (1.5%) in the control group (p=0.002). Otalgia was reported in 1 (1.5%) case compared to 2 (3.1%) controls . Tinnitus was reported in 5 (7.7%) cases and vertigo in 5 (7.7%) cases with none in the control group. In clinical examination, 6 (9.2%) cases were found to have an abnormality in which 4 involved the tympanic membrane (3 had retracted TM, 1 had bulging TM). One case had external auditory canal stenosis and another had auricular skin hyperpigmentation. There were no abnormalities noted in the control group.

Table 1: Abnormal Pure Tone Audiometry in patients with autoimmune diseases and controls

	Cases	Controls
Conductive Hearing Loss (CHL)	5 (7.7%)	2 (3.1%)
Sensori-Neural Hearing Loss (SNHL)	1 (1.5%)	-
Mixed Hearing Loss	2 (3.1%)	-

Table 2: Abnormal tympanograms in patients with autoimmune diseases and controls

Type of tympanogram	Cases	Controls
A	40 (61%)	60 (92%)
As	13 (20%)	4 (6.1%)
Ad	3 (4.6%)	1 (1.5%)
B	4 (6.1%)	-
C	2 (3.1%)	-

Table 3: Types of autoimmune diseases with abnormal PTA and tympanograms

Cases	*Abnormal PTA	Abnormal tympanogram
Rheumatoid Arthritis (RA)	5 (15%)	9 (27%)
Systemic Lupus Erythematosus (SLE)	3 (13%)	10 (45%)
Other	-	3 (13%)

The association of developing abnormal tympanometry to duration of disease was not found to be statistically significant (p-value 0.996). The type of medication used was linked to increased risk of developing abnormal audiometric and tympanometry results. Prednisolone was 3.9 times (P = 0.002) more likely to cause an abnormal tympanogram and 2.8 times (P=0.113) more likely to cause an abnormal PTA. Methotrexate was 1.5 times more likely to cause an abnormal PTA (P=0.513) and 2.6 times more likely to cause an abnormal tympanogram (p=0.513). Hydroxychloroquine sulfate was 2.3 times more likely to cause an abnormal PTA (p= 0.223) and 2.8 times more likely to cause an abnormal tympanogram (p = 0.024).

An association between history of hearing loss to having an abnormal PTA was 2.3 times more likely (p = 0.341) and to having an abnormal tympanogram 2.87 times more likely (p=0.15). The association of having an abnormal clinical exam to an abnormal tympanogram was 4.5 times more (P=0.096) and an abnormal PTA 4.4 times more likely (P=0.125).

DISCUSSION

The study showed majority of the cases to be females compared to males in a ratio of 15:1 with 61 females and 4 males. In a study by Adelowo[29] looking at systemic autoimmune diseases in the African continent, majority of autoimmune diseases occurred in females compared to males. In Kenya, it was found to be at a ratio of 6.5:1. Similar local studies were done in KNH where SLE patients examined had female to male ratio of 30:1[28,29]. The mean age was 40 years ranging from 18-65 years with duration of disease ranging from 1year to 25 years. The most prevalent AI conditions were RA and SLE at 50% and 33% respectively with others at 10%. This is in keeping with Ekwom[31] where they stated the two most prevalent autoimmune conditions in Kenya were RA and SLE.

The duration of disease was calculated from the year of diagnosis rather than when the symptoms begun. The mean duration of disease was 4.7 years with a range of 1–23 years and no association between duration of disease and otologic manifestation could be established.. An association between age at disease onset and hearing loss was found by Mokbel *et al*[22] but there was no significant statistical association between duration of disease and hearing loss in this study. This is similar to Oczan *et al*[15] and Hussein and Doosti[17] who did not find any association between duration of disease and risk of otologic condition. This was similar to Kastanioudakis *et al*[20] study where they observed asymmetric results in 15 out of 26 SLE patients and unable to prove any association to ototoxicity.

The most common medication used were methotrexate, prednisolone and hydroxychloroquine sulfate. NSAIDs were not entered as they were used on a need basis and varied in types and duration. The medication was analyzed with likelihood of causing otologic manifestations and no significant association was found. Patients would have combinations of two or three medications therefore difficult to analyze them individually. All these drugs have been mentioned as having risk of ototoxicity in a study by Moskowitz *et al*[35]. Some have been mentioned to be beneficial[34-36]. Prednisolone is known to improve hearing in autoimmune ear disease together with methotrexate at low dosage[37].

Of the symptoms reported, 9 (13.8%) cases reported hearing loss compared to 1 (1.5%) control and this was statistically significant at 0.002. Otalgia was reported in 1 (1.5%) case, tinnitus in 5 (7.7%) cases, vertigo in 5 (7.7%) cases with none in the control group and were statistically insignificant. Mokbel *et al*[22] reported 4 cases having tinnitus and 1 case of vertigo. Abbasi *et al*[24] had 5 (11.1%) cases report of hearing loss compared to 1 (2.2%) case in the control group and this was statistically significant. Symptoms are subjective as patients are likely to perceive themselves as having otologic disturbances which may just be related to the overall ill health of the disease. Eight (12.3%) cases were found to have abnormal audiograms compared to 2 (3.1%) controls with a P value of 0.067 which was not statistically significant. Majority of the cases 5 (7.7%) had CHL compared to controls 2 (3.1%) and SNHL 1 (1.5%) case and mixed HL (3.1%). This is similar to Arslan *et al*[16] study where they found HL in 27.3% of the cases compared to 15.9% in controls with majority of them having CHL at 56.8% and controls at 25%. This is in contrast to some studies done where SNHL was found to be more prevalent like Oczan *et al*[15] who found 35.5% had SNHL compared to 24.3% who had CHL with controls at 5.7%, Garcia *et al*[18] who found 38.6% of SNHL predisposition and Roverano *et al*[25] who found 66% of cases had SNHL in SLE. This could mean majority of our cases would be having conductive anomalies involving the TM and ossicular chain rather than cochlea. RA has incudomalleolar and incudostapedial joint infiltration of T cells thus affecting the ossicular chain while SLE will have immune deposits within the middle ear.

Twenty cases were found to have abnormal tympanograms (33.86%) compared to five in the control group (7.7%) with a P value of 0.0001 which is statistically significant. Thirteen cases had type As (20%), 3 type Ad (4.6%), 4 type B (6.1%) and 2 type C (3.1%). Oczan *et al*[15] found abnormal tympanograms in 37.9% in cases compared to 17.1%. Halligan *et al*[19] found no difference in tympanometry studies done in their analysis

of 29 patients with RA compared to 30 control patients. Type As indicates either some fluid or ossicular fixation that partially decreases mobility. Synovial involvement in RA of the ossicular chain could cause fixation of the joints decreasing mobility. Type Ad indicates high static admittance from an overly mobile tympanic membrane caused by disarticulation of the bony structures of the middle ear which could result from immune complexes being deposited in middle and inner ear in SLE and RA synovial joint involvement which can cause separation of the joints. Type B shows low static admittance and in normal EAC canal shows middle ear fluid. Type C shows negative pressures indicative of ET dysfunction. AIs are multisystemic diseases and involve nasal structures as they can cause rhinitis and nasal obstruction therefore interfere with the Eustachian tube function.

CONCLUSION

The majority of AI conditions were RA and SLE with female preponderance and the complaint of hearing loss concurred with positive audiometric findings and abnormal tympanometry.

LIMITATIONS

This study selected patients at a tertiary health facility which might bias results as very often these patients are referred because of progression of symptoms or inability to diagnose at a smaller facility.

RECOMMENDATION

Patients with autoimmune diseases should undergo regular audiological assessments of PTA and tympanometry as they are more likely to have otological manifestations compared to the general population.

Conflict of interest: The authors declare no conflict of interest. The study recieved $US 2400 from the Kenyatta National Hospital Research fund for cost of performing audiometry and tympanometry and reimbursing assistants.

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RESEARCH ARTICLE

THE EFFECTS OF ADENOTONSILECTOMY ON PULMONARY PRESSURES FOR CHILDREN AT A TERTIARY HOSPITAL IN KENYA: A CROSS SECTIONAL STUDY

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ABSTRACT

Background: Patients with adenotonsilar hypertrophy have been shown to have raised pulmonary artery pressures due to upper airway obstruction. Adenotonsilectomy reduces these pulmonary arterial pressures.
Objective: The aim of this study was to determine if there is any difference between pre-and post-operative pulmonary artery pressures in patients with adenotonsilar hypertrophy by 6 weeks post operatively. However, there is a study that had assessed for changes at 1 and 3 months and found there was a significant drop in the mean pulmonary artery pressures (mPAP).
Methodology: One hundred and thirteen patients were consecutively recruited from the Ear Nose Throat (ENT) Department at a tertiary level hospital in Kenya. Nine were lost to follow up. Adenoid hypertrophy was graded using the Cohen and Konak scale and tonsilar hypertrophy graded using the Brodsky scale. Patients were assessed for level of awake partial pressures of oxygen, weight, height and body mass index and a complete cardiovascular and respiratory systems assessment. Echocardiograms were done pre-operatively and at 6 weeks post-operatively.
Results: There was a slight male preponderance while the age range was 2.5 years to 10 years. The age group of 2.5 to 4 years had the highest number of patients, while the symptom duration ranged from 1 to 9 years with the highest number of patients having symptoms of 1-3 years. The main symptoms included snoring (97.1%), mouth breathing (96.2%) and sleep disordered breathing with apneic attacks as seen by an observer (90.4%). The pulmonary pressures ranged from 0 to 24mmHg with 87% below 20mmHg and 10% between 20-24mmHg. Pre-operative and post-operative mPAP (mean pulmonary artery pressures) changed significantly (P value of <0.001).
Conclusion: There was an association between adenoid size and the level of mPAP, but no association between tonsillar size, history of symptoms, symptom duration and mPAP. Pre- operative mPAP dropped significantly post-operatively.

Keywords: Adenotonsilar hypertrophy, Adenotonsilectomy, Mean pulmonary artery pressures, Postadenotonsilectomy

INTRODUCTION

Adenoid hypertrophy refers to the hypertrophy of the nasopharyngeal lymphoid tissue while tonsillar hypertrophy refers to enlargement of the palatine tonsils. The adenoid has rapid growth in infancy but involutes from about age 8-10 years and is rarely seen in adults. The tonsil begins to involute during puberty[1]. Adenoid Hypertrophy (AH) and Adenotonsilar Hypertrophy (ATH) are known to cause Pulmonary Artery Hypertension (PAH) and eventually cor-pulmonale. This relationship was first noted and described in 1965 by Menashe *et al*[2]. PAH associated with AH is classified under the 3rd group as per the '2015 ESC/ERS Guidelines for the diagnosis and treatment of pulmonary hypertension' and has not been noted to have a genetic component associated with it[3]. Under normal conditions during nasal breathing, the air column has laminar flow. This is disturbed by ATH causing obstruction in the nasopharynx with a resultant rise in upper airway resistance in an attempt to raise air entry via the nasal route[4]. The hypertrophy of these tissues is associated with complications such as: nasal obstruction (snoring, mouth breathing, dry lips, obstructive sleep apnea)[5], cardiopulmonary symptoms (pulmonary hypertension), right ventricular hypertrophy and cor-pulmonale, ear and paranasal sinus complications (frequent otitis media and sinusitis), voice changes (flat and toneless)[6], failure to thrive (due to anorexia poor intake, increased energy consumption from increased work during breathing and alterations in nocturnal growth hormone patterns)[5,7]. In the physical examination, the grade of the tonsilar hypertrophy can be staged by the Brodsky tonsilar grading scale (Appendix I). It is important to assess patients for adenoid facies as well as other comorbidities like heart failure. Adenoid hypertrophy can be assessed using a soft tissue lateral neck radiograph and graded using the Cohen

and Konak grading system (Appendix I). The Cohen and Konak grading system compared to various radiological methods to assess adenoid hypertrophy was found to have 75% inter and intra- personnel reproducibility[8]. Of importance is that this method was also seen to have the best comparable results to endoscopy as compared to other radiological assessment methods[9]. Polysomnography is the gold standard to assess for sleep apnea and features of upper airway obstruction however these facilities are not available in our setup[5].

Pulmonary Artery Hypertension (PAH) can be inferred using Electrocardiograms (ECG) as well as chest radiographs which are cheaper modalities than ECHO, however negative findings in these modalities do not exclude the presence of disease. This is because the symptoms are more likely to be seen in severe as opposed to mild disease. PAH is defined as mean pulmonary pressures (mPAP) ≥25mmHg at rest as assessed by cardiac catheterization[10]. Normal pulmonary artery pressures fall within 14± 3mmHg range, with an upper limit of normal of approximately 20mmHg[5]. The patients with mean pulmonary pressure ranges between 21 and 24mmHg may not have significant features of cardiopulmonary failure[10]. Echocardiography has however been seen to be a useful noninvasive tool to diagnose pulmonary artery hypertension in comparison to cardiac catheterization which is the gold standard[10-12]. Multiple studies have been carried out to assess the prevalence of PAH in patients with Adenotonsillar Hypertrophy (ATH) and some of these studies compared pulmonary pressures pre-and post Adenotonsilectomy. These studies have been carried out in Egypt, Kenya and Nigeria. All these studies suggest an association with reduction in pulmonary arterial pressures post adenotonsilectomy.

The aim of this study was to find out whether there is any difference between pre-and post-operative pulmonary artery pressures in patients with adenotonsillar hypertrophy and to assess if there is an association between clinical grade of adenotonsillar hypertrophy and post-operative pulmonary arterial hypertension.

MATERIALS AND METHODS

This was a prospective cross sectional study conducted within the Ear Nose and Throat (ENT) clinic and ward at the largest tertiary health facility in Kenya (Kenyatta National Hospital-Nairobi). The study population comprised of 113 consecutively recruited patients already selected for adenotonsillectomy by the otorhinolaryngologist. Seven were lost preoperatively (5 unable to attend surgery and 2 unfit for surgery), 1 lost to follow up postoperatively bringing the sample size to 104. This sample size was estimated based on 'Sample Size for Testing Differences in Proportions for Paired-Samples' with a power of 80% and level of significance (α) of 5% . The ages ranged between 2 and a half years and 12 years. This study included patients who had already been selected for adenotonsillectomy and patient's whose parents/guardians consented to be included in the study. The exclusion criteria included patients with: congenital anomalies that would cause Upper Airway Obstruction (UAO) e.g. choanal atresia, Down's syndrome, muscular anomalies e.g. in patients with cerebral palsy, children who have chronic illnesses e.g. sickle cell disease, rickets, children with nasal obstruction due to other factors e.g. septal deviation, septal spurs, known cardiovascular diseases due to other factors other than ATH, tracheal malformations e.g. subglottic stenosis and patients who refused to participate.

Ethical approval was obtained from the Hospital Ethics Committee (KNH-UON Ethics Research Committee P409/05/2016) and both written and verbal informed consent was obtained from the parents/legal guardians of the participants.

Socio-demographic data was collected by means of a closed and open ended questionnaire. Pre-testing of this questionnaire was carried out on 10% of the sample size to validate the tool and test its reliability. Minor edits to language construct were required post-testing, so as to avoid bias and misinterpretations of the questions.

A general examination was done that included assessing a patient's functional status, vital signs e.g. heart rate SPO^2, obtaining patient weight and height, a full head and neck examination, cardiovascular, respiratory and abdominal examination.

A lateral soft tissue neck radiograph was done pre-operatively and used to measure the adenoid ratio using the Cohen and Konak grading scale (Appendix I) and confirm adenoid hypertrophy. Assessment of the radiograph was done using a ruler. Physical examination was used to grade the tonsilar hypertrophy using the Brodsky grading scale (Appendix I). A 2D Echocardiogram was also done pre-operatively and at 6 weeks post operatively by the same cardiologist. The cardiologist was blinded as to the severity of patient symptoms.

Continuous variables were analyzed for means and ranges and using a paired t-test for non-independent samples. Categorical variables were analyzed by calculating percentage of patients with a level of the variable preoperatively and comparing this with the postoperative proportion using a McNemar's chi square test for dependent proportions.

RESULTS

The sample comprised of a total of 104 patients of which there was a slight male preponderance (51.9% versus 48.1%). Majority of children were below 5 years of age (57.7%). The age group 1-3 years had the most persistent symptoms with 75% having quality of life affected.

There was a positive correlation between duration of symptoms and preoperative mean Pulmonary Artery Pressure (mPAP) (rho = 0.247, $p < 0.012$). Pre-operative mPAP also showed a strong positive correlation with post-op mPAP (rho = 0.676, $p<0.001$).

Snoring was found in up to 97.1% of children with mouth breathing in 96.2% and sleep disordered breathing with apneic attacks as seen by an observer was 90.4%. Only one child presented with symptoms of tachypnea and tachycardia and required to be managed for heart failure and stabilization before adenotonsillectomy. The weight of these children ranged from 10-40.6 kg with a mean of 17.8 ± 5.4kgs. The mean BMI was 15.4 ± 2.2 with a range from 10.2 to 21.93.

Tonsillar hypertrophy grade 3 was the most common with moderately sized adenoids being the highest in number. Those with small tonsillar size had more symptoms associated with chronic recurrent tonsillitis (Table 2).

Table 1: Summary of findings from studies comparing changes in pulmonary artery pressures post operatively in children with adenotonsillar hypertrophy

Author	Tests	Findings	Comments
Pac *et al,* 2005[13]	#Echo,$X-ray	No change in mPAP at 1mth postoperative	Small sample size
Yilmaz *et al,* 2005[14]	Echo, X-ray	Prevalence of PAH 59%, 21% change at 3 and 6mths post-operative	mPAP-cut-off 20mmHg
Abd El-Moneim *et al,* 2009[15]	Echo, X-ray	Reduced mPAP, improved Right ventricular (RV) output	Focus was on cardiac events
Marangu *et al,* 2011[16]	Echo, *ECG,		
	X-ray	mPAP > 21mmHg, 21.9% prevalence of PAH, cross sectional study	No pre-post Adenotonsillectomy analysis
Mossad *et al,* 2011[17]	Echo, X-ray	Significant improvement at 6mths	Noticed asymptomatic cardiac changes in AH
Koc *et al,* 201[21]	Echo, X-ray	Improved RV function and reduced mPAP at 3mths	Small sample size
Martha *et al.* 2013[18]	Echo X-ray	36% prevalence of PAH and reduction in mPAP by 26% post-operative	
Orji *et al,* 2017[19]	Echo, X-ray	mPAP >25mmHg, elevated PAP reversible by AT irrespective of symptom severity	Sample size of 39

#2D Echocardiogram, $Lateral neck radiographs, *Electrocardiograph

Table 2: Grading and size of adenotonsillar hypertrophy in children in KNH

	Frequency	(%)
TH Grade		
Grade 2	32	30.8
Grade 3	63	60.6
Grade 4	9	8.6
AH Size		
Large	39	37.5
Moderate	60	57.7
Small	5	4.8

Mean pulmonary arterial pressures were 9.34 ± 8.52 preoperatively, and 2.91 ± 4.21 in the post-operative period. There was a statistically significant decrease in pressure levels postoperatively and a significant difference in the pre-compared to post-operative values (Paired t-test, $p < 0.01$). Systolic PAP pressures pre-operatively had a mean of 13.50 ± 12.50mmHg and post operatively a mean pressure of 4.7 ± 6.12mmHg. The difference in means was statistically significant ($p<0.001$). There was no relationship between age and the mean pulmonary arterial pressure changes, both in the pre-operative and the post-operative stages (Figure 1).

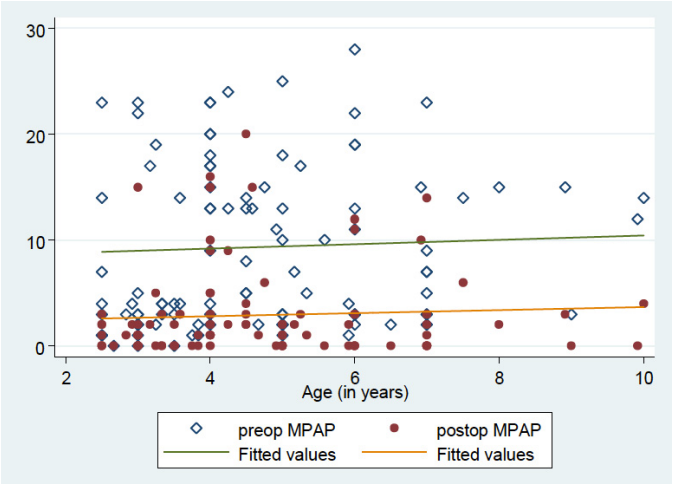

Figure 1: Pearson's co-relation between pre-and post-operative association of age and mPAP

History of symptoms was not statistically significant in association with the change in mean PAP. The BMI was not significantly correlated with either pre-op mPAP ($p = 0.862$) or post-op mPAP ($p = 0.609$).

Tonsillar size: There was no statistically significant association between tonsillar size and mPAP ($p = 0.323$) and the systolic PAP ($p=0.668$).

Adenoid size: There was a statistically significant association between adenoid size and the mPAP with a p value of <0.001. The systolic PAP was also statistically significant between adenoid size and systolic PAP (p value of 0.001). All three children with pulmonary artery hypertension (1 with moderate and 2 with mild), improved significantly post

operatively. They all had normal values of pulmonary artery hypertension by the 6th week post-operatively.

Echo: It was also noted there was a statistically significant drop in mean PAP between pre-and post-operative ECHOs a change from 9.3 to 2.9 a difference of 6.42 (p value <0.01). The change in systolic PAP pre- and post-operatively was also significant with a P value <0.001. There were three children with pulmonary artery hypertension. One had moderate PAH and required stabilization and management for heart failure prior to adenotonsilectomy. He had a large adenoid and a grade three tonsillar size. The second had mild PAH and had TH grade 4 and an AH graded as large. The third had mild PAH with a large AH but a grade 2 TH. A point to note is that only ten patients did not have a drop in the mPAP from pre-to postoperative periods, in fact one of these had an increase in the mPAP post operatively, one had no change, and 8 others had a very slight drop in mPAP and systolic PAP. These ten shall have follow up ECHOs again at 6 months.

DISCUSSION

There were 54 males (51.9%) and 50 females (48.1%). This is comparable to other studies which show a relatively greater proportion of male children[1,14]. The age of the patients ranged from 2.5 to 10 years despite the cut off being from 2.5 to 12 years, with the patients aged 2.5 to 4 years being the highest with 57.7%. The mean age was 4.5 years in this study which was different from Pac *et al*[13] who had a mean age of 7.3 ± 2.9 years (with a range of 3-14 years). Orji *et al*[19] had a similar mean of 4.6 years with a median of 3.4 years. This is in keeping with the age of those mostly afflicted by ATH. Marangu *et al*[16], found a mean age of 2.5 years but the study assessed patients with AH as well. The age group 2.5- 4 years also had the largest change in mPAP as compared to other groups. For Orji *et al*[19] the mPAP was recorded highest among the youngest participants who were aged <2 years (9 patients in their study), but the differences in the mPAP among the age groups were not significant. That was a finding similar to our study. There was however no association between age and mPAP in this study as seen by a weak Pearson correlation. Symptom duration ranged from 1-9 years with the symptom duration of 1-3 years being the largest percentage. This may also be associated with the largest age group of patients being 2.5- 4 years of age. Again, this is the age most afflicted by ATH. El-Moneim *et al*[15] found the duration of obstructive symptoms in years to be a median of 2.2 and a range of 1.2–9. Snoring was the most common symptom followed by mouth breathing and sleep disordered breathing. Martha *et al*[18] in 2013 found that all their subjects presented with snoring and oral breathing while 84.4% had restless sleep. In the study by Marangu *et al*[16] mouth breathing occurred in 74.8% while mouth breathing and restless sleep occurred in 88.5%[16]. In this study, Grade 3 TH had the highest in terms of percentage and TH Grade 2 was the lowest grade observed while moderate AH had the highest percentage. This is similar to other studies like that of Orji *et al*[19] whose study had a minimum TH of Grade 2. However, other studies like Martha *et al*[18] only found TH of Grade 3 and above. Marangu *et al*[16], found TH of Grade 1 to 4 with Grade 2 being the commonest. This is perhaps because the age group was mainly of younger patients.

Our study found a statistically significant association between adenoid hypertrophy and mPAP pre-operatively and no significant association between tonsillar hypertrophy and mPAP. This differs from Marangu *et al*[16] and Martha *et al*[18] who found no association between the size of adenotonsillar hypertrophy and the mPAP. However, Orji *et al*[19] found an association between AH and mPAP change. Preoperatively, most children had normal pulmonary pressures (range within 0-24mmHg), with 87% below 20mmHg and 10% between 20-24mmHg. Three percent had pulmonary artery hypertension (2 had mild pulmonary artery hypertension and 1 had moderate pulmonary hypertension). Yilmaz *et al*[14] had found raised mPAP of 27 (51%) of the 52 subjects. It is important to note that they used 20 mmHg as the cut off, above which was PAH in the study group preoperatively. Koc *et al*[11] also had significant drop in the mPAP pre-and post- operatively more observed after 3 months than the 1 month post- operative period. Abd El Moneim *et al*[15], found significant improvement in right ventricular output post- operatively. They felt the right ventricular output was the best measure for improvement post-operatively. There was a statistically significant change in mPAP between pre and post-operative ECHOs as observed in this study with a pre-operative mean of 9.34± 8.52 to a post-operative mean of 2.91± 4.21. There was thus a change of 6.42 in the mean and this was found to be statistically significant with a P value of <0.001. In the study by Abd El-Moneim *et al*[15], they found a calculated mPAP ranging from 30.6 mmHg ± 8.8 pre-operatively to 26.2mmHg ± 6.8 postoperatively which was statistically significant with a p value of 0.007. Martha *et al*[18] also found a change in systolic PAP with a significant drop in pre and post-operative values which is also similar to our study.

In this study, the systolic PAP was associated with AH but was not associated with TH, symptom duration, age and BMI, this is similar to Martha *et al*[18] who also had systolic PAP return to normal values after surgery. However, in their study, they had a reduced number of patients classified by the systolic PAP and this difference did not reach statistical significance.

CONCLUSION

There was an association between adenoid size and duration of symptoms, with the level of mPAP and systolic PAP. There was however no association between tonsillar size, BMI and mPAP. Pre-operative mPAP and systolic PAP dropped significantly 6 weeks post- operatively.

RECOMMENDATION

Patients with ATH who have an indication for adenotonsillectomy should undergo the surgery as soon as possible as the effects on the heart will have resolved by 6 weeks post-operatively. A larger prospective study should be conducted with post- operative assessment done at 3 and 6 months.

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Appendix I:

Appendix la) Cohen and Konak radiologic adenoid hypertrophy grading system

Air column (mm)	Distance between the posterior border of the soft palate 10mm away from the posterior nasal spine and the anterior curvature of the pharyngeal tonsil border
Air column/Soft palate ratio (AC/SPR)	Between air column and soft palate
Soft palate (mm)	Thickness of the soft palate measured 10mm away from the posterior nasal spine
Small	Air column/soft palate ratio ≥ 1-0
Moderate	An column/soft palate ratio≥ 0.5 but < 1.0)
Large	Air column/soft palate ratio ≥ 1.0

Appendix lb): The Brodsky clinical tonsilar hypertrophy grading system;

0-25c'c of the oropharynx i.tonsil just outside the tonsilar fossa)
25-50S: of the oropharyngeal width
50-~5fO- of the oropharyngeal width
75-100f;f of the oropharyngeal width

RESEARCH ARTICLE

CIGARETTE SMOKING AND ALCOHOL INGESTION AS RISK FACTORS FOR OROPHARYNGEAL SQUAMOUS CELL CARCINOMA AT THE KENYATTA NATIONAL HOSPITAL

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ABSTRACT

Background: Oropharyngeal cancer accounts for 10-12% of head and neck malignancies in Kenya. Squamous Cell Carcinoma (SCC) forms 90% of oropharyngeal malignancies. Tobacco smoking and alcohol intake have a strong interactive effect on the risk of squamous cell carcinoma of the head and neck.
Objectives: To determine the role of smoking tobacco and alcohol consumption as risk factors in causation of Oropharyngeal Squamous Cell Carcinoma (OPSCC) at Kenyatta National Hospital (KNH).
Methods: A hospital-based case control study that was conducted in three departments of the hospital. Data for 65 patients with confirmed OPSCC was selected from an ongoing prospective study while 130 controls matched for age and sex were recruited from the orthopaedic unit.
Results: Majority (53.9%) of the participants with OPSCC had multiple sub-sites involved, with the tonsils being the most affected sub-site (60%). Tobacco smoking was significantly associated with OPSCC ($P < 0.001$) and odds of OPSCC was 3.6 times higher among smokers compared to non-smokers. Stratification of smokers by current smoking status showed that compared to participants who had never smoked, current smokers were at highest risk of OPSCC (OR = 5.0; 95% CI 2.29-10.93) followed by those who had stopped smoking (OR = 2.85; 1.33-6.09). Alcohol consumptions did not have a significant association with OPSCC ($p<0.87$); however there was an exponential increase in risk with increased intake.
Conclusion: Cigarette smoking is a significant risk factor for OPSCC in patients seen at the Kenyatta National Hospital while alcohol consumption was noted to be a contributory factor.
Recommendation: Interventions targeted at reducing alcohol and cigarette consumption need to be emphasized since a reduction in risk was noted with cessation and lower consumption of the two exposures.

Key words: Kenya, ENT, Oropharyngeal carcinoma

INTRODUCTION

Cancer is a leading cause of death in both developed and developing countries; the burden is expected to increase due to growth and aging of the population as well as changing lifestyles. According to GLOBOCAN statistics estimate, 14.1 million new cases and 8.2 million deaths occurred in 2012 worldwide and the burden in low resource setting accounts for about 75% of cases and 65% of cancer deaths worldwide[1]. In Kenya, cancers account for up to 18,000 deaths annually with an incidence of 82,000 new cases per year and a prevalence of 82,500 cases[1,2]. Oropharyngeal cancers comprise 10-12% of head and neck neoplasms, with Squamous Cell Carcinoma (SCC) accounting for 90% of all histological types[3]. The role of Human Papilloma Virus (HPV) in tumourigenesis in OPC has been associated with change in trend where younger adults are increasingly being affected[4]. Several studies have demonstrated that alcohol and tobacco consumption increase the risk of oral cancer in a dose related fashion. A population-based case control study in Bhopal, India observed significant risk of bidi and cigarette smoking with a dose-response relationship for lung and OPC[5]. Studies have demonstrated that risk of OPC increase significantly with higher alcohol and tobacco consumption levels and duration[6,7].

The study aimed to determine the role of smoking tobacco and alcohol consumption in causation of oropharyngeal squamous cell carcinoma at Kenyatta National Hospital (KNH).

MATERIAL AND METHODS

Setting and design: This was a hospital based case control study which was matched for age and sex. This design was chosen as it is ideal for studying rare outcomes and thus was chosen due to the rarity of oropharyngeal carcinoma in KNH. Data on the cases who had histologically

confirmed oropharyngeal carcinoma was extracted from an on-going prospective study being conducted by ENT and radio-oncology unit within KNH. Controls were recruited from the orthopaedic unit in KNH.

Sample size determination: Sample size was calculated using Kirkwood Formula for difference in proportions with a power of 80% and 95% confidence interva[18]. Proportions were based on a study carried out previously[6]. Calculated sample size was 65 patients for cases and 130 for controls as a ratio of 1:2 was used.

Data collection: Cases; Data for the cases included demographic characteristics, alcohol consumption, tobacco smoking history and tumour characteristics.

Alcohol intake habits, including the type, duration and amount, were categorized as per NIAAA guidelines as to whether they are non-drinkers, social, moderate or heavy drinkers[9]. Cigarette smoking duration, type and amount were recorded and classified in pack-years.

The oropharyngeal sub sites affected with OPSCC were documented and differentiation characteristic also recorded. Due to the varying TNM stages noted, the cases were further stratified according to prognostic stages I-IV.

Controls: An informed consent was sought from selected controls from the orthopaedic unit. A physical examination of the oropharynx was done by the principal investigator to rule out suspect oropharyngeal lesions. A preformatted data collection questionnaire similar to the selected case population was filled by the principal investigator.

Similar to the cases, alcohol intake habits, including the type, duration and amount, were also categorized as per NIAAA guidelines as to whether they were non-drinkers, social, moderate or heavy drinkers. Cigarette smoking duration, type and amount was also recorded and classified in pack-years.

Statistical analysis: Mean, median and standard deviations were calculated for the continuous variables (age, duration of exposure to risk of OPC e.g smoking) in the univariable analysis. Counts and proportions were used to summarize categorical variables in the univariable analysis e.g describe the sex distribution of cases among other categorical demographic characteristics. In the bivariate analysis, Chi square and Fisher's exact tests were performed to detect associations between categorical variables and oropharyngeal cancer diagnosis whereas ANOVA and T tests were used to detect associations between continuous variables and oropharyngeal cancer. The analyses were completed using STATA version 18.0 (STATA Corporation, College Station, Texas, USA).

Multivariable logistic regression was used to detect independent predictors of oropharyngeal cancer. The factors included in the logistic regression were the primary exposures of tobacco smoking and alcohol ingestion and other risk factors that showed significant association with case-control status in the bivariable analysis. Statistical significance was based on a p value cutoff of 0.05

Review and approval of the study protocol, consent forms, and data collection instruments for this study was completed by the KNH and UoN Ethics and Research Committee (approval reference no: P626/08/2016).

RESULTS

The male-to-female ratio was 4:1 with 52 (80%) males and 13 (20%) females. The mean age of the cases was 58.6 (± 14.1) years. The modal age groups for cases of oropharyngeal cancer was 60-69 years (32.3%).

Table 1: Demographic characteristics of patients with OPSCC and controls

	Cases	Controls	OR (95% CI)	P value
Age (years)	7(10.8)	17(13.1)		
< 40				
40-49	7(10.8)	21(16.2)	0.81(0.24-2.76)	0.736
50-59	17(26.2)	37(28.5)	1.12(0.39-3.19)	0.838
60-69	21(32.3)	37(28.5)	1.38(0.49-3.86)	0.542
70 +	13(20.0)	18(13.8)	1.75(0.56-5.45)	0.331
Sex				
Male	52(80.0)	104(80.0)		
Female	13(20.0)	26(20.0)	1.00(0.48-2.11)	1.0

Presentation of oropharyngeal squamous cell carcinoma was as follows:

Majority of the cases presented with advanced disease.

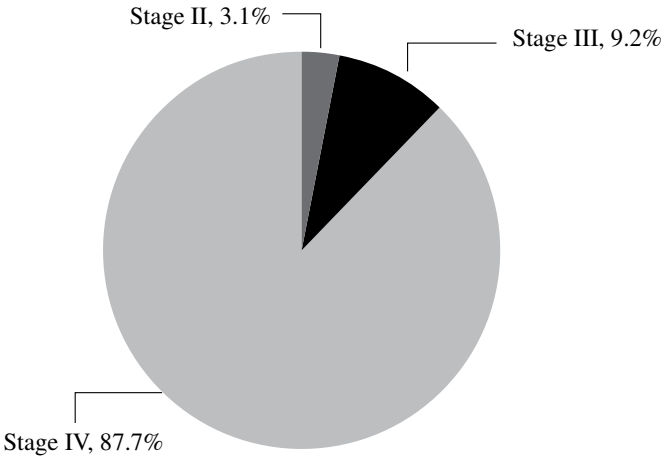

Figure 1: Stages of oropharyngeal squamous cell carcinoma in cases identified in KNH

Table 2: Histology of oropharyngeal SCC in cases identified in KNH

	Frequency (n)	(%)
Histologic findings of oropharyngeal SCC		
Undifferentiated	10	15.4
Poorly differentiated	9	13.8
Moderately differentiated	16	24.6
Well differentiated	30	46.2

Subsite involvement: Out of the 65 cases of OPC, 35 (53.9%) had cancers showing multiple subsite involvement. Palatine tonsils and tonsillar fossa was the most commonly involved site in oropharyngeal SCC in 60% of the cases (Figure 2). There were no cancers that involved the lateral and posterior oropharyngeal walls.

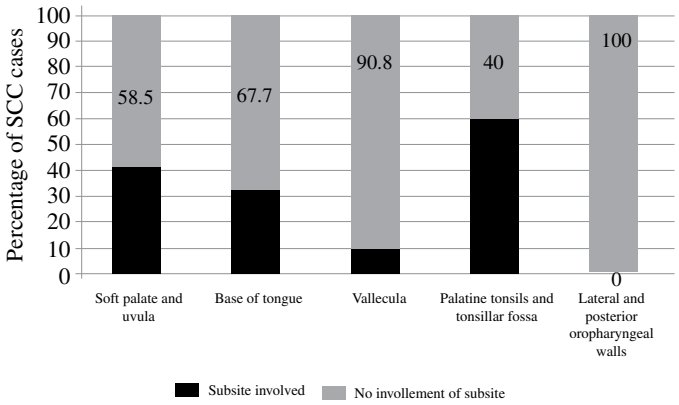

Figure 2: Subsites involved in oropharyngeal carcinoma among cases in KNH

Tobacco smoking as a risk factor to development of oropharyngeal squamous cell carcinoma: Tobacco smoking was significantly associated with oropharyngeal SCC development ($p < 0.001$) (Table 3). The likelihood of developing carcinoma was 3.6 times in a smoker as compared to non-smoker with OR of 3.6, 95% CI1.93-6.71). Current smokers were 5 times likely to develop SCC compared to non-smokers (OR = 5.0, 95% CI 2.29 - 10.93, p = 0.007). Participants who had quit smoking had a two to three-fold increased risk of SCC compared to non-smokers (OR = 2.85, 95% CI 1.33 – 6.09, $p < 0.001$).

Table 3: Tobacco smoking as a risk factor to development of oropharyngeal squamous cell carcinoma

	Cases	Controls	OR(95% CI)	P-value
Ever smoked				
No	25(38.5)	90(69.2)		
Yes	40(61.5)	40(30.8)	3.60(1.93-6.71)	<0.001
Current smoking status				
Never smoked	25(38.5)	91(70.0)		
Stopped smoking	18(27.7)	23(17.7)	2.85(1.33-6.09)	0.007
Current smoker	22(33.8)	16(12.3)	5.00(2.29-10.93)	<0.001

Alcohol ingestion as a risk factor to development of oropharyngeal squamous cell carcinoma: Overall, alcohol consumption was not significantly associated with increased risk of oropharyngeal SCC (OR = 0.87, 95% CI 0.47 – 1.63, p = 0.668). However, based on the NIAAA classification very heavy alcohol consumption significantly increased the risk of oropharyngeal SCC (OR = 5.22 CI 95% 1.80 – 15.12, p=0.002).

Table 4: Alcohol ingestion as a risk factor to development of oropharyngeal squamous cell carcinoma

	Cases	Controls	OR (95% CI)	P values
Alcohol ingestion				
No	23(35.4)	42(32.3)		
Yes	42(64.6)	88(67.7)	0.87(0.47-1.63)	0.668
Level of alcohol consumption[a]				
Light	4(6.2)	35(26.9)	0.22(0.07-0.71)	0.011
Moderate	10(15.4)	31(23.8)	0.63(0.26-1.51)	0.301
Heavy	12(18.5)	13(10.0)	1.81(0.71-4.59)	0.214
Very heavy	16(24.6)	6(4.6)	5.22(1.80-15.12)	0.002

[a]Level of alcohol consumption using NIAAA classification

Alcohol and tobacco as joint exposures as risk factors for oropharyngeal squamous cell carcinoma: More than half (56.9%) of the participants consumed alcohol and smoked tobacco and had nearly 2 times risk of OPSCC (OR = 1.95 95% CI 0.96 – 3.93, p = 0.063). Participants who consumed alcohol alone had a significantly reduced risk compared to those who only smoked (OR=0.20 95% CI 0.07-0.58, p=0.003), (OR=3.00 95% CI 0.46-19.43, p=0.249) respectively.

Table 5: Alcohol and tobacco as joint exposures as risk factors for oropharyngeal squamous cell carcinoma

	Cases	Controls	OR (95% CI)	P value
None	20(30.8)	40(30.8)		
Smoking & alcohol consumption	37(56.9)	38(29.2)	1.95(0.96-3.93)	0.063
Consumption of alcohol alone	5(7.7)	50(38.5)	0.20(0.07-0.58)	0.003
Smoking alone	3(4.6)	2(1.5)	3.00(0.46-19.43)	0.249

DISCUSSION

Among the cases recruited in our study, majority (87.7%) presented with advanced OPSCC stage disease according to the TNM classification. A similar study conducted in KNH found that 56% of patients with head and neck cancers presented with stage IV disease and this was attributed to hospital referral systems[10]. Majority of the cases recruited in this study were found to be predominantly elderly men in their seventh decade of life, similar to previous studies in KNH[10]. With the increasing rate of adoption of different lifestyles and cultural trends in developing countries, there has been a concurrent change in both tobacco smoking and alcohol consumption trends leading to concerns on the possible impact of these practices on oropharyngeal cancer epidemiology in Kenya.

Prevalence of lifetime smoking was higher among cases (61.5%) compared to controls (30.8%).The analysis showed that 28.3% of the burden of oropharyngeal cancer

was attributable to tobacco smoking. The increase in risk of oropharyngeal carcinoma that was attributed to smoking in the population was 34.8% implying a 34.8% reduction in the risk of oropharyngeal SCC in the population if smoking was eliminated. Other studies have demonstrated similar strong associations between smoking and OPC[6,7,11]. Being a current smoker was associated with a five-fold increase in the odds of OPSCC. This higher risk has been explained by the different tobacco consumption habits in Bhopal, India[5]. Cessation of smoking was noted to have reduced risk for oropharyngeal cancer. Participants who had quit smoking had a two to three-fold increase in risk of SCC compared to non-smokers, however compared to the current smokers; a decrease of risk was appreciated. Similar trend was noted in which reducing risk occurred with increasing smoking cessation period[6].

The prevalence of alcohol consumption was 64.6% among the oropharyngeal carcinoma cases compared to 67.7% among the controls. However, higher levels of alcohol consumption were noted amongst the cases compared to the controls. Alcohol consumption in this study did not have a significant association with OPSCC. However, an increase in alcohol consumption was noted to have a directly proportional increase in risk of OPSCC. Similar dose related trend has been reported by other studies[6]. In this study, heavy alcohol consumption was associated with five times higher odds of OPSCC as compared to participants who did not consume alcohol. Similarly, Altieri *et al*[12] demonstrated that the odds ratio for oral and pharyngeal cancers was directly proportional to the increasing level of alcohol consumption.

Most studies have demonstrated synergistic effects of joint exposures of alcohol and tobacco smoking[6,7]. A similar study done by Menach *et al*[13] on causation of alcohol consumption and tobacco smoking on laryngeal squamous cell carcinoma at the KNH also demonstrated synergism. In this study, surprisingly, synergism was not noted. As previously mentioned, tobacco smoking was noted to have a stronger association to OPSCC as compared to alcohol consumption. With joint exposure as compared to tobacco smoking, the risk reduces from 3 to1.95. Alcohol consumption amongst the participants varied greatly. The type of alcohol constituted beers, hard liquor, wine and traditional brews. The composition of the traditional brews is unknown and they differ from one culture to another. The effects of the constituents of the brews are thus unknown and further evaluation and analysis is needed to find out if they confer any protection.

The majority of tumours were well differentiated. This was also noted in retrospective cohort analysis carried out over 3 decades by Mehta *et al*[14] in which they found that most OPSCC were well differentiated. However, there was decline over time of incidence of well differentiated tumours. This decrease in tumour differentiation was attributed to influence of human papillomavirus in OPSCC. HPV induced SCC typically display non/poorly keratinization differentiation. HPV status was not assessed in this study and its influence cannot be ruled out. No association was noted between the disease stage and histologic grading.

The finding of the sample characteristics conforms to those documented in both local and international literature. Majority of the cases recruited in this study were found to be predominantly elderly men in their seventh decade of life. According to KDHS 2014, alcohol consumption and tobacco smoking was more common in males as compared to females and in addition, increased consumption was noted amongst the elderly group[14]. The role of tobacco and alcohol in tumorigenesis of head and neck cancers has been recognized for a long time now. One can then postulate, on the basis of available information that it is not surprising that a higher male prevalence was noted in this study.

In addition, the controls selected were from the orthopaedic unit in which most admissions are due to accidents which are alcohol related and thus this would then result in a higher prevalence of alcohol consumption amongst the controls.

CONCLUSION

This hospital based case control study strongly implicates tobacco smoking as a major risk factor for oropharyngeal squamous cell carcinoma. Heavy alcohol consumption had a contributory factor in OPSCC development. No synergism was noted with joint exposures.

RECOMMENDATIONS

(i) Measures to prevent OPSCC should be aimed at reducing intake of both alcohol and tobacco smoking.
(ii) Stringent measures by the government to ensure the Tobacco control bill 2012[15] and Alcoholic drinks control Act, 2012[16] are adhered to, with an aim of reducing prevalence of cigarette smoking and alcohol consumption.
(iii) Cancer Prevention and Control Act Cap 246B[17] needs to be executed so that an effective National Cancer Registry Institute is developed.
(iv) There should be public health awareness of symptomatology of OPSCC and continuous health care education to ensure early detection and management.

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RESEARCH ARTICLE

TRANSLATION AND VALIDATION OF A KISWAHILI VERSION OF 'OBSTRUCTIVE SLEEP APNOEA-18' QUALITY OF LIFE INSTRUMENT IN CHILDREN WITH SLEEP DISORDERED BREATHING DUE TO ADENOID AND TONSILLAR HYPERTROPHY: A CROSS SECTIONAL STUDY

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ABSTRACT

Background: Sleep Disordered Breathing (SDB) is associated with numerous cardiac, respiratory and behavioural complications. Adenotonsillar hypertrophy is a common cause of SDB in children. Given limitations in accessing polysomnography, 'Obstructive Sleep Apnoea 18'(OSA-18) quality of life questionnaire is used to assess quality of life in sleep disordered breathing.
Objective: This study aimed to translate and validate a Kiswahili (national Kenyan language) version of OSA-18 in children with sleep disordered breathing due to adenoid and tonsil hypertrophy.
Methods: A cross-sectional study carried out at the Ear, Nose Throat clinic at a tertiary health facility in Kenya. Children with adenotonsillar hypertrophy and clinical features of sleep disordered breathing were selected by convenience sampling. A translation and cross cultural adaptation of the OSA-18 was done using revised Brislin's method followed by testing and retesting of the Kiswahili version to 75 recruited patients/their guardians to assess reliability. The scores were tested against adenoid and tonsil size to determine the validity. Children underwent adenoid and tonsil surgery and Quality of Life (QoL) measured 4 weeks postoperatively.
Results: Good test-retest reliability and internal consistency achieved (Cronbach's Alpha 0.70). Spearman's correlation coefficient (rho) between scores and tonsil size was found to be r=0.21 which was similar to the original r = 0.29 against tonsil size but not against Apnoea Hypopnea Index rho =0.10 against rho = 0.43. The average preoperative score was 79.17 (SD13.58). The sleep disturbance domain had the highest scores. There was significant improvement in QoL following adenoidectomy and tonsillectomy with the average post-op scores being 22.20 (SD 6.58). Sleep disturbance showed the greatest improvement.
Conclusion: The Kiswahili version has shown good test-retest reliability, internal consistency and validity.

Key words: Sleep disordered breathing, OSA 18, Adenotonsillar hypertrophy, Adenoidectomy, Tonsillectomy

INTRODUCTION

Sleep-Disordered Breathing (SDB) is a term that encompasses a spectrum of sleep-related upper airway obstruction that ranges from primary snoring and Upper Airway Resistance Syndrome (UARS) to Obstructive Sleep Apnoea Syndrome (OSAS) to obesity-hypoventilation syndrome (Pickwickian syndrome).

Whilst thought to be benign, habitual snoring in children has been shown to lead to neurocognitive disorders and reduction in growth hormone secretion[1]. With SDB, disturbed sleep patterns in children lead to behavioral changes and poor concentration thus reduced Quality of Life (QoL)[1,2]. Long-standing sleep apnoea can result in irreversible pulmonary hypertension and if sustained this will lead to right heart failure and cor pulmonale. In Kenyan children with adenoid hypertrophy, 21.9% were shown to have pulmonary hypertension[3]. In the paediatric population, adenotonsillar disease is the commonest cause of SDB[4]. One to three percent of children are thought to have OSA whilst 7% are habitual snorers[5]. There has been no gender differences shown in pre-pubertal children[6]. Although SDB can occur at any age, it seems to present most commonly in 2 to 5 year olds[6,7]. For children with OSA that is caused by adenotonsillar hypertrophy, adenotonsillectomy is the treatment of choice[4].

The gold standard for diagnosing SDB is laboratory polysomnography (PSG). Given that PSG is expensive and often difficult to obtain, especially in low resource settings, primary care physicians look to other alternatives for diagnosis of SDB. However, polysomnography involves a detailed, laborious evaluation of cardiorespiratory and neurologic parameters in a sleep laboratory and, therefore, is not widely accessible. It is time-consuming, costly, and usually entails long waiting times[8,9].

The Obstructive Sleep Apnoea-18 (OSA-18), developed by Franco is the most widely used alternative tool to measure Quality of Life (QoL) for paediatric SDB, and has been validated as an evaluative and discriminative instrument[10]. The OSA-18 has been extensively used as a quality of life instrument in children with sleep disordered breathing and has been shown to be effective in this regard more so pre- and post adenoidectomy and tonsillectomy. This has been shown for both short term follow up (1-3 months)[11-13] and long term (6-24 months) post operatively[14,15].

Given the demographics of the Kenyan population regarding languages and multi ethnicity, and given that most of the population speak Kiswahili as the national language, cross cultural adaptation of OSA-18 into Kiswahili would be beneficial for monitoring outcomes of treatment, specifically adenoid and tonsillar disease related Sleep Disordered Breathing (SDB) and Quality of Life (QoL).

The aim of this study was to translate the OSA-18 Quality of life instrument into Kiswahili; To test the Kiswahili OSA-18 version for consistency, reliability and validity to assess QOL in children with SDB due to adenotonsillar hypertrophy and to assess quality of life before and after adenoid and tonsillar surgery using the Kiswahili OSA-18.

MATERIALS AND METHODS

This was a cross-sectional study carried out at the Ear, Nose and Throat (ENT) clinic at Kenyatta National Hospital (KNH), Kenya's largest tertiary health care facility. The study population comprised children under 12 years of age, who were attending the ENT clinic for adenoid and tonsillar hypertrophy with clinical features of sleep disordered breathing and scheduled for adenoid and/or tonsil surgery, and whose parents gave informed verbal and written consent and could understand Kiswahili. Those with prior adenoid and/or tonsillar surgery, craniofacial malformations, genetic disorders, cognitive disorder, mental retardation or neuromuscular disorders were excluded. Sample size was estimated with a one-sided percentage point of the normal distribution corresponding to power of 80% (μ =0.84) and two-sided percentage point of the normal distribution corresponding to 95% level of significance (v= 1.96). The proportion of positives detected with the gold standard (π0=97%) and the proportion hypothesized to be detected, given by (π= 88%)[16]. This came to 75 patients. An allowance of 5% was made or dropouts, and final sample size was 79. Convenience sampling was utilized.

The study took place in three stages. Stage 1 translated the OSA-18 questionnaire using the revised Brislin model for cross cultural adaptation[17]. This involved translation by 2 multi-lingual linguists into Kiswahili. They subsequently compared their versions to give the final Kiswahili version. Back translation to English was done by 2 blinded multilingual (English and Kiswahili speaking) otolaryngologists. Through this the Kiswahili version was assessed for accuracy and for any material lost in translation. A final panel meeting of all translators and the researcher was held to approve the final version. This version was then piloted (pre-tested) on 18 children (not part of the study) from different ethnic backgrounds to further refine it.

In stage 2, a patient biodata form and questionnaire were filled by the patient's guardian. The questionnaire asked about various symptoms related to SDB. A clinical examination was carried out to grade tonsillar size based on Brodsky's criteria[18]. A postnasal lateral soft tissue radiograph was taken to diagnose adenoid hypertrophy. Adenoid hypertrophy was graded using the Fujioka criteria with significant hypertrophy considered when adenoid nasopharyngeal ratio was ≥ 0.80[19].

Assisted filling of the Kiswahili version of OSA-18 by the caretakers of the children who fulfilled the inclusion criteria was done. This score was recorded and caretakers were asked to retake the questionnaire after 1week period just before their adenoid and tonsil surgery. The two results were analyzed for consistency, test – retest reliability and assessment for validity (using the tonsil size and the adenoid size as objective parameters).

Stage 3 involved filling the Kiswahili version of OSA-18 one month (4 weeks) after the adenoid and tonsil surgery. The results were analyzed to assess changes in the quality of life post intervention.

The study started after ethical approval by the Kenyatta National Hospital/University of Nairobi ethical committee. Written informed consent was obtained from each participant. All data on patient demographics, examination findings was analyzed using SPSS® version 20.0. The data was cleaned of errors, inconsistent or conflicting answers, as well as missing or duplicate entries. Descriptive statistics were calculated for the ages of children and caretakers, level of education by proportion and relationship to child. The reliability of the OSA-18 tool was calculated with a Cronbach's correlation coefficient alpha and validity by means of Spearman's correlation coefficient rho. Pre-and post-operative scores were compared by means of a paired t-test. Diagnosis of the patients and surgical procedure performed was summarized by frequency.

RESULTS

Forty (53%) were male children and 35 (47%) were female. The mean age of children was 50.84 months (SD 25.8 months) with a range from 9 to 138 months. The average age of the caretakers was 32.53 years (SD 6.5 years) with a range of 20 to 60 years. Forty percent of caretakers attained secondary level of education, 19% college certificates, and 8% university degrees. Majority of the caretakers were mothers (90%).

Reliability: Test- retest item correlations ranged from 0.64 to 0.99 (Table 1). All 18 items had excellent test-retest reliability, indicating good stability. The consistency

coefficient (Cronbach's alpha) ranged from 0.70 to 0.76 thus >0.70, indicating acceptable internal consistency.

Validity: The criterion validity between each test item and the tonsil grade and adenoid nasopharyngeal were examined. The total OSA-18 score, the emotional symptoms and caregiver concerns were significantly correlated with tonsil grade while the emotional symptoms and daytime functions were significantly correlated with the adenoid nasopharyngeal ratio (adenoid size). These results confirm the internal validity of OSA-18 with the proxy of tonsil grade (in place of a polysomnography) (Table 1). Spearman's correlation coefficients (rho) were considered significant when r= > 0.20 (p <0.05) with a p value of less than 0.05 considered significant. Table 1 contains all the correlation values and test retest reliability and internal consistency values (Cronbach alpha). The Tonsil Grade and ANR show the Spearman correlation coefficient.

Table 1: Reliability and validity of each Obstructive Sleep Apnoea -18 (OSA-18) item

OSA-18	$Consistency	Test-retest (p-value)	Tonsils grade	#ANR
OSA – 18 total scores	0.70	0.99 (<0.001)	0.21*	0.10
Sleep disturbance				
Loud snoring	0.74	0.79 (<0.001)	0.03	-0.02
Breath holding/pauses	0.75	0.78 (<0.001)	0.01	-0.10
Choking or gasping	0.75	0.69 (<0.001)	0.10	0.13
Fragmented sleep	0.74	0.64 (<0.001)	0.05	0.13
Physical symptoms				
Mouth breathing	0.74	0.76 (<0.001)	0.08	0.11
Frequent colds or upper Respiratory infections	0.76	0.76 (<0.001)	-0.06	0.06
Nasal discharge or runny nose	0.76	0.72 (<0.001)	0.03	0.10
Difficulty swallowing	0.74	0.74 (<0.001)	0.07	-0.06
Emotional symptoms				
Mood swings or tantrums	0.75	0.79 (<0.001)	0.18	0.10
Aggression/hyperactivity	0.74	0.76 (<0.001)	0.20 *	0.06
Discipline problems	0.76	0.83 (<0.001)	0.22*	0.21*
Daytime function				
Daytime sleepiness	0.75	0.80 (<0.001)	0.07	0.32*
Poor attention span or concentration	0.76	0.84 (<0.001)	0.05	0.19
Difficulty getting up in the morning	0.76	0.77 (<0.001)	0.06	0.09
Caregiver concerns				
Caregiver worried over child health	0.73	0.78 (<0.001)	0.09	-0.11
Caregiver concerned not enough air	0.74	0.72 (<0.001)	0.19	-0.13
Caregiver missed daily activities	0.74	0.83 (<0.001)	-0.03	-0.20*
Caregiver frustrated	0.73	0.87 (<0.001)	0.24*	-0.22*

Key: $Consistency based on Cronbach's correlation coefficient-alpha; *segments that show a significant Spearman's rank correlation coefficient (rho >0.20) when compared to tonsil size and adenoid size: #ANR- Adenoid nasopharyngeal ratio

The OSA-scores measurements: The average total OSA – 18 score in the patients at first test was 79.35 (SD 13.75) and at re-test it was 79.17 (SD 13.58). Post-operatively it was 22.20 (SD 6.58). The OSA-18 scores were distributed as follows during the pre-operative phase of the study: 9.33% had a small impact in health-related quality of life, 44% had a medium impact on health-related quality of life while 46.67% had a severe impact on health-related quality of life. In the post-operative phase of the study, all the 75 patients were graded as experiencing small impact on health-related quality of life due to adenotonsilar hypertrophy.

There is very little evidence (p=0.5091) against the hypothesis that there is a difference in average scores during the test and the retest phase. However, there was very strong evidence against the hypothesis that the test and the post-operative scores have a similar mean score (p<0.0001). This therefore means that there was a significant difference in OSA scores pre-and post operation. Table 2 shows the mean scores (standard deviation in brackets) and a p-value of the paired t-test between the test and the post-operative scores.

Table 2: The quality of life questionnaire mean scores and respective comparison

OSA-18	Test score	Retest score	Post-op score	*P-value
OSA – 18 total scores	79.35 (13.75)	79.17 (13.58)	22.20 (6.58)	<0.0001
Sleep disturbance	21.87 (4.26)	22.04 (4.38)	4.91 (2.52)	<0.0001
Loud snoring	5.90 (1.24)	5.85 (1.25)	1.36 (0.85)	<0.0001
Breath holding/ pauses	5.49 (1.52)	5.59 (1.48)	1.19 (0.67)	<0.0001
Choking or gasping	4.88 (1.87)	5.09 (1.78)	1.15 (0.59)	<0.0001
Fragmented sleep	5.59 (1.56)	5.51 (1.60)	1.21 (0.72)	<0.0001
Physical symptoms	20.75 (3.93)	21.15 (3.60)	5.12 (2.18)	<0.0001
Mouth breathing	6.04 (1.28)	5.96 (1.39)	1.21 (0.55)	<0.0001
Frequent colds or upper respiratory infections	5.64 (1.24)	5.80 (1.05)	1.36 (0.82)	<0.0001
Nasal discharge	4.91 (1.80)	5.19 (1.52)	1.41 (1.03)	<0.0001
Difficulty swallowing	4.16 (2.11)	4.20 (2.11)	1.13 (0.53)	<0.0001
Emotional symptoms	8.67 (5.06)	8.25 (5.28)	4.04 (2.13)	<0.0001
Mood swings or tantrums	3.43 (2.17)	3.20 (2.15)	1.52 (0.99)	<0.0001
Aggression/ hyperactivity	3.25 (2.29)	3.05 (2.22)	1.35 (1.01)	<0.0001
Discipline problems	1.99 (1.61)	2.00 (1.71)	1.17 (0.60)	<0.0001
Daytime function	8.67 (4.21)	8.50 (4.71)	3.64 (1.36)	<0.0001
Daytime sleepiness	2.71 (2.02)	2.81 (2.20)	1.07 (0.38)	<0.0001
Poor attention span or concentration	2.39 (1.82)	2.37 (1.89)	1.33 (0.86)	<0.0001
Difficulty getting up in the morning	3.57 (2.46)	3.32 (2.39)	1.24 (0.84)	<0.0001
Caregiver concerns	19.40 (7.18)	19.23 (7.38)	4.49 (1.59)	<0.0001
Caregiver worried over child health	4.97 (2.06)	4.93 (2.15)	1.24 (0.88)	<0.0001
Caregiver concerned not enough air	5.35 (1.94)	5.09 (1.98)	1.12 (0.68)	<0.0001
Caregiver missed daily activities	4.64 (2.23)	4.61 (2.12)	1.09 (0.44)	<0.0001
Caregiver frustrated	4.44 (2.30)	4.59 (2.28)	1.04 (0.26)	<0.0001

*p-value result from paired t-test of Test scores versus post-operative scores

The diagnostic outcome measures: The most common diagnosis was adenotonsillar enlargement (ATH) with 77.33% being diagnosed as having ATH. Consequently, the most common operation was ASTS (adenoid surgery and tonsil surgery) having the same proportion (77.33%). A majority of the patients had a tonsil grade of 3 (62.67%) while a minority had grade 4 tonsils (6.67%, n=5). There was no difference in scores between those diagnosed with ATH compared to those diagnosed with AH and CRT (p-value = 0.1341). It is important to note that a majority of the patients had enlarged tonsils (69.33%) while 30.67% did not have them. In our study, only 4% of our study subjects had an ANR of less than 0.80. The mean value was found to be 0.90.

DISCUSSION

The OSA-18 is the most widely studied measure of Quality of life associated with SDB. Whilst it cannot replace polysomnography in diagnosing obstructive sleep apnoea, it is useful in assessing for both the impact of SBD and response to treatment therefore overall Quality of life. Cross cultural adaptation, reliability and validity analysis is important to ensure that validated English language instruments are found specific to a particular social and cultural structure.

Franco *et al*[10] and Kang *et al*[16] in their studies in the original OSA-18 and in the Taiwan-Chinese versions respectively assessed the correlation coefficients of tonsil grade and adenoid size in addition to AHI from polysomnography to assess validity. The internal consistency coefficient for this study of 0.70-0.76 was comparable to other studies; Kang for the Taiwanese/ Chinese study (0.84), Fausto in the Portugese (0.821), Mousalidis in the Greek (0.951) and Kuptanon in the Thai study (0.77)[20-23]. The internal consistency is considered acceptable if 0.70 or greater[24].

Construct validity for our study was assessed using the adenoid nasopharyngeal ratio as a measure of adenoid size and tonsil size as adjuncts for polysomnography (which still remains the gold standard) given its unavailability. These were also used in the original study by Franco *et al*[10] and also by Kang *et al*[16] for the validation of the Chinese version of OSA-18. They also assessed the correlation coefficient using overnight polysomnography. The correlation coefficient vis a vis the polysomnography was noted to be at Spearman rank correlation r = 0.43 for the original study, r =0.48 for the Thai version, and r=0.40 for the Chinese version[10,20,23]. Concerning the relation with tonsil grade, Franco in the original study found a rank correlation coefficient factor r=0.29. Kang *et al*[16] found a correlation coefficient of 0.17 for the tonsil grade. This related to the total correlation coefficient for tonsil grading in our study of r = 0.21. With a coefficient factor of > 0.20 considered satisfactory. Adenoid correlation was found to be at r = 0.45 for the original OSA-18 version by Franco and r = 0.16 for the Chinese version[10,16]. This study had a coefficient of r = 0.10.

Franco *et al*[10] in their original study assessed adenoid size using fibre optic nasopharyngeal endoscopy assigning a grade on the basis of the percentage of choanal obstruction; 1+(0-25%), 2+ (26 -50%), 3+(51-75%), 4+ (76- 100%). Our study utilized Adenoid Nasopharyngeal Ratio (ANR) based on lateral soft tissue post nasal space radiograph. As shown by Fujioka *et al*[19], the significant level was taken to be an adenoid nasopharyngeal ratio of > 0.80. In our study, only 4% of our study subjects had an ANR of less than 0.80. The mean value was found to be 0.90. It was unclear whether the method of adenoid size assessment had a significant determinable effect on the eventual correlation coefficients as seen in our study compared with the original. Certain studies have indicated a preference for fibre optic naso-endoscopy compared with radiography citing an underestimation when using radiographs[24].

Concerning the scores seen on the OSA-18 preoperative scores, of the five subsections the section on sleep disturbance had the highest scores followed by physical symptoms and caregiver concerns; 21.81 ± 4.26 and 20.75 ± 3.93 respectively. Emotional symptoms and daytime function were noted to have the lowest scores. This was similar to the findings in the Greek and Portugese versions[20,21]. In the Thai and Chinese populations, however, it was noted that the section on caregiver concerns had the highest scores followed by sleep disturbance and physical symptoms[16,22]. According to Kang *et al*[16] (Taiwan/Chinese), this reflected the way culture of parenting in the Taiwanese population. He also indicated a selection bias as being a clinic based study, those bringing their children showed heightened concern. Emotional symptoms and daytime function still formed the least scores. In this study, most parents admitted that due to their children being at school or them at work, this did not allow for truly accurate scoring of daytime functionality.

In terms of impact on quality of life, more severe symptoms had a greater adverse impact on quality of life. It was noted that whilst the Taiwan Chinese OSA-18 showed higher scores in patients with severe OSA (78.2 ± 13.8) compared to mild (66.8 ± 19.6), the values were not noted to be statistically significant. In the study by Mitchell and Kelly[12], it was found that some children with severe OSA actually had better scores compared to some with mild ones. These findings confirm the findings by Constantin that whilst the OSA-18 is good at assessing the quality of life, its OSAS diagnostic capabilities are limited[25].

In assessing quality of life following adenoid and tonsilar surgery, the greatest changes were seen in sections of sleep disturbance, physical symptoms, caregiver concern, daytime function then emotional symptoms respectively. There was no noted significant difference noted between patients with adenotonsilar enlargement and those with adenoid hypertrophy alone. This may be due to the discrepancy in the sample size of the adenotonsilar patients, who were more than 3 times

that of adenoid hypertrophy patients. This may thus lend itself to further studies with a larger and more comparative sample size. The mean postoperative score was found to be 22.20. The changes noted were seen to be significant and was found to relate to previous QOL assessment studies[11-13] (1 to 3 months postoperatively) and Mitchel *et al*[11] and Flanary[14] for the long term (6-18 months). Whilst there was noted to be a comparatively slight difference in short term post-operative measures versus long term, this was not found to be statistically significant in a meta-analysis by Christina *et al*[26].

LIMITATIONS

In the domain of daytime function, it was noted that most parents did not spend time with their children throughout the day due to either the children being in school or the parents being at work. This may have led to some discrepancy in the scores of this domain. Kiswahili like most ethnic African languages is limited in its description of emotional disturbance disorders and hence there was noted limitation in answering the emotional disturbance segment. This may have led to the low change in scores.

The absence of a polysomnography posed some problems as pointed above; an inability to compare scores with severity of OSA. It also did not allow for assessment of cutoffs for OSA as demonstrated in other studies.

CONCLUSION

The Kiswahili version of OSA-18 was found to have good reliability, internal consistency. The validity was found to be satisfactory. There was significant impact on quality of life in the children with adenoid and tonsillar hypertrophy with the largest percentage exhibiting severe impact on quality of life preoperatively. Tonsillectomy and adenoidectomy resulted in a significant improvement in the quality of life with all the patients showing drastically reduced scores. The Kiswahili OSA-18 can be used for assessing impact on quality of life in children with sleep disturbed breathing due to adenoid and tonsil disease.

RECOMMENDATION

A study with a larger sample size and equal subjects to compare the impact on quality of life in patients with adenoid hypertrophy and in patients with both adenoid and tonsillar hypertrophy. Despite Kiswahili being a language used in East Africa, it varies in accents/pronunciations among various regions and cultural differences exist from one country to another hence cross cultural adaptation may be necessary in other countries.

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APPENDIX 1
FINAL KISWAHILI OSA 18 QUALITY OF LIFE QUESTIONAIRRE

UCHUNGUZI KUHUSU UBORA WA MAISHA (OSA-18)

	Hakuna wakati hata mmoja	Nadra kwa wakati huo	Kiasi kidogo sana cha wakati huo	Sehemu ndogo ya wakati huo.	Angalau sehemu kubwa kiasi ya wakati huo	Mara nyingi	Kila wakati
USUMBUFU WAKATI WA KULALA: Katika muda wa majuma manne yaliyopita ni mara ngapi mwanao amekuwa:							
Anakoroma kwa sauti ya juu?	1	2	3	4	5	6	7
Anazuilia pumzi, ama anakatiza katiza pumzi anapopumua usiku?	1	2	3	4	5	6	7
Anasakamwa ama kutoa sauti za ishara ya kukosa hewa anapo lala?	1	2	3	4	5	6	7
Ana usumbufu anapo lala ama kuamka amka kila wakati?	1	2	3	4	5	6	7
DALILI MWILINI: Katika muda wa majuma manne yaliyopita ni mara ngapi mwanao amekuwa na:							
Visa vya kupumua kutumia mdomo kutokana na kufungika kwa mapua?	1	2	3	4	5	6	7
Homa ya mafua, ama maambukizi katika sehemu ya mapua na koo?	1	2	3	4	5	6	7
Tatizo la kutokwa na makamasi?	1	2	3	4	5	6	7
Ugumu wa kumeza?	1	2	3	4	5	6	7
DALILI ZA KIHISIA: Katika muda wa majuma manne yaliyopita, ni mara ngapi mwanao amekuwa na:							
Mabadiliko ya mara kwa mara ya kihisia au vipindi vya hasira?	1	2	3	4	5	6	7
Tabia ya kukasirika kupita kiasi bila sababu au tabia ya kutotulia?	1	2	3	4	5	6	7
Amekuwa na tatizo la ukosefu wa nidhamu?	1	2	3	4	5	6	7
UTENDAKAZI NYAKATI ZA MCHANA: Katika muda wa majuma manne yaliyopita, ni mara ngapi mwanao amekuwa na:							
Shida ya kulala kupita kiasi nyakati za mchana?	1	2	3	4	5	6	7
Tatizo la kufuatilia jambo kwa umakini ama kutekeleza jambo kikamilfu?	1	2	3	4	5	6	7
Shida ya kuamka kutoka usingizini asubuhi?	1	2	3	4	5	6	7
WASIWASI WA MLEZI: Katika muda wa majuma manne yaliyopita ni mara ngapi mwanao:							
Amekufanya kuhofia hali yake ya kiafya kwa jumla?	1	2	3	4	5	6	7
Amekufanya uwe na wasiwasi kuwa hapati hewa ya kutosha?	1	2	3	4	5	6	7
Ametatiza uwezo wako wa kutekeleza majukumu yako ya kila siku ?	1	2	3	4	5	6	7
Amekuweka katika hali ya kutojua cha kufanya?	1	2	3	4	5	6	7
Jumulisha alama:							
0-60 = Madhara madogo kwa ubora wa maisha ki afya							
60-80 = Madhara ya kadri kwa ubora wa maisha ki afya							
80+ = Madhara makubwa kwa ubora wa maisha ki afya							

RESEARCH ARTICLE

THE SUBMANDIBULAR GLAND IN ADVANCED ORAL CAVITY CANCER

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ABSTRACT

Background: Treatment of oral cavity cancer results in xerostomia as the submandibular gland is usually removed or damaged by radiation therapy. Submandibular gland sparing in cancers with low incidence of level 1 nodal metastasis during surgery and intensity modulated radiation tomography have been instituted to reduce the detrimental effects of xerostomia. Sparing the submandibular gland in oral cavity is however challenging due to the high incidence of level 1 metastasis and proximity to primary tumour.
Objective: This study evaluated the submandibular gland in advanced oral cavity cancer aiming to elucidate the incidence and mechanism of involvement.
Methods: Medical charts of patients with oral cavity cancer T4a and T4b that were treated surgically including neck dissection were retrieved and analysed. CT scan images were also retrieved and evaluated for level 1b lymphadinopathy. Histopathology of the neck specimen was analysed. Analysis was done using Statistical Package for Social Science version 19(IBM Corporation, Armonk,NY,USA) (P value was set at 0.05).
Results: The study included 269 patients. A total of 394 neck specimens were analysed as 125 patients underwent bilateral neck dissection. Only 6 patients had pathological evidence of submandibular gland invasion giving an incidence of 1.52%. Only 6 of 11 radiological images with features of submandibular gland invasion had pathological evidence of involvement. There were no cases of bilateral or contralateral submandibular gland invasion. Significant parameters on univariate analysis were histology, neuroinvasion, lymph node metastasis, extracapsular node extension and radiological features of invasion.
Conclusions: Submandibular gland invasion is rare even in advanced oral cavity cancer. It is unlikely for contralateral or bilateral submandibular invasion in well lateralized oral cavity cancer. Manouver of submandibular gland sparing through surgery and radiotherapy are logical in the contralateral side even in advanced cancer.

Key words: Submandibular gland, Cancer, Oral cavity, Neck dissection

INTRODUCTION

The oral cavity is a common site of head and neck cancer with up to 40% developing lymph node metastasis[1]. Level 1 nodes are the echelon in oral cavity cancer and are invariably involved clinically or by occult metastasis. Neck metastasis is the single most important prognostic factor in oral cavity cancer with survival reducing in node positive patients[2]. Comprehensive treatment of oral cavity cancer thus entails management of the primary tumour and the neck due to the high incidence of metastasis. The submandibular gland is routinely removed during neck dissection of level 1 b despite its lack of intraparechymal nodes and therefore the oncological basis of its resection has been questioned.

Tseng-Chen Cheng *et al*[3] reviewed 383 neck dissection specimens of patients with oral cavity squamous cell carcinoma and found only 7 with submandibular gland involvement. Despite having included specimens from all stages, submandibular invasion could only be predicted inT4 and nodal status above N2b. They thus recommended submandibular gland sparing in oral cavity cancer T3 and below with N0 necks on preoperative evaluation to prevent xerostomia. Basaran *et al's*[4] review of 294 neck specimens had 13 with submandibular gland invasion. Most of the invasion was due to direct tumour invasion similar to Tseng-Chen's series. Four specimens had gland invasion from a periglandular metastatic lymphadinopathy. Most of the cases of gland invasion were in advanced tumours with lesser chances in the early stages. Only 2 cases of submandibular gland invasion were noted in 66 early cancers [stage 1 and 2] while11 cases were present in advanced disease [stage 3 and 4]. The early cases were both T2 with primaries in the tongue. There was one case of contralateral submandibular gland invasion from a metastatic periglandular lymphadenopathy. The author proposed intraoperative assessment of the submandibular gland with preservation of those not in close proximity to the tumour and without adherent nodes[4].

All the studies reviewed had included examination of neck specimen of all stages thus not giving a true incidence

of submandibular gland invasion in advanced oral cavity carcinoma where the greatest chance of involvement lies. Recommendations are made of submandibular gland preservation in early oral cavity squamous carcinoma when the gland is not grossly invaded either by tumour or node. Xerostomia for such patients who don't require postoperative radiation will be prevented. The idea of submandibular gland sparing in late oral cancers is an interesting subject as a balance must be made between tumour eradication and post treatment xerostomia as most of these patients will receive postoperative radiation which has adverse effects on salivary glands. Our study sought to investigate the incidence and nature of submandibular gland involvement in advanced oral cavity tumours thus elucidating feasibility of gland sparing in this stage of disease.

MATERIALS AND METHODS

The study was done at E-DA Hospital where medical charts of patients with advanced oral cavity squamous cell carcinoma managed surgically were retrieved. CT scan or MRI images were evaluated for lymphadenopathy in level 1b and submandibular invasion by tumour. Lymph nodes with the short axis greater than 1 cm on axial cuts were considered significant. Central necrosis of the nodes and direct invasion of the submandibular gland identified by loss of a plane between the gland and tumour were also evaluated. The parameters sought for were the T stage, N stage, histological differentiation, primary site, bone invasion, neural spread, lymphovascular invasion, skin invasion, muscle invasion, margin and lymph node status. P value was set at 0.05. Analysis was done using Statistical Package for Social Science version 19 (IBM Corporation, Armonk, NY, USA).

Only patient records with oral cavity squamous cell carcinoma stage T4a or T4b with radiological and complete pathological information conforming to the standard reporting format including submandibular gland status were included into the study. Patients with previous irradiation or chemotherapy, histology other than squamous cell carcinoma, tumours outside the oral cavity and incomplete pathological records were excluded from the study.

RESULTS

The study included 269 patient records that dated between 2007 and 2016. There were 256 males and 13 females. The age range was between 32 and 89 years with a mean of 54. Bilateral neck dissection was performed in 125 patients with the remaining being cases of unilateral neck dissection. A total of 394 neck specimens with submandibular reports were therefore included in the study.

The significant parameter on univariate analysis were histology, T stage, neuroinvasion, lymph node metastasis and CT features of direct invasion by tumour (Table 1).

Table 1: Representing different parameters in relation to submandibular invasion

Parameter	Item	Submandibular invasion		P value
		Negative	Positive	
Subsite	Floor of mouth	10	0	0.424
	Buccal	116	2	
	Tongue	85	1	
	Retromolar	21	1	
	Gingiva	126	1	
	Palate	17	0	
	Lip	11	1	
Histology	WDSCC	158	0	<0.001
	MDSCC	225	4	
	PDSCC	5	2	
T stage	T4a	342	2	0.003
	T4b	45	4	
Clinical nodal stage	N0	147	1	0.806
	N1	58	1	
	N2a	5	0	
	N2b	108	2	
	N2c	70	2	
Lymphovascular invasion	LV nil	317	4	0.308
	LV positive	71	2	
Bone invasion	Nil	201	4	0.687
	Positive	187	2	
Neuroinvasion	Nil	277	1	0.010
	Positive	111	5	
Skin invasion	Nil	298	4	0.627
	Positive	90	2	
Muscular invasion	Nil	81	0	0.353
	Positive	307	6	
Lymph node metastasis	N0	147	1	0.018
	N1	58	1	
	N2a –N2c	183	4	
Extracapsular spread	Nil	332	2	
	Positive	56	4	0.006
CT scan gland direct invasion	Nil	377	0	<0.001
	Positive	11	6	
CT scan nodes in Level 1B	Nil	261	1	1.000
	Positive	127	5	
CT scan nodal Necrosis	Nil	336	1	0.582
	Positive	52	5	

Six specimens had submandibular gland involvement by tumour giving an incidence of 1.52%. None of these had contralateral or bilateral involvement. Direct tumour spread was the most likely route of invasion of the submandibular gland as evidenced by CT scan images. Of the 11 cases with CT scan images with direct invasion of the submandibular gland by tumour, only 6 had pathological evidence of involvement. Five of these had enlarged nodes with 4 having CT scan features of extracapsular spread due to irregular margins.

Table 2: Representing characteristics of 6 pathology specimens with submandibular gland invasion

Serial	Tstage	cN-stage	Site	Histology	Bone	Nerve	LV	Skin	Mus-cle	Node METS	ECS
1	T4b	N2c	Tongue	MDSCC	-	+	-	-	+	+	+
2	T4b	N2b	Tongue	MDSCC	-	+	+	-	+	+	+
3	T4a	N2c	Buccal	PDSCC	+	+	+	+	+	+	+
4	T4b	N1	Retro-molar	MDSCC	-	-	-	-	+	-	-
5	T4a	N0	Buccal	PDSCC	+	+	-	-	+	+	-
6	T4b	N2b	Buccal	MDSCC	-	+	-	+	+	+	+

DISCUSSION

The submandibular gland is routinely removed during level 1b neck dissection with the perivascular and periglandular nodes. Metastatic disease from the oral cavity will most certainly involve this group of nodes making a supraomohyoid neck dissection the simplest of surgical neck treatment for oral cavity tumours. Unlike the parotid, the submandibular and sublingual glands have no intraparenchymal lymphnodes. Reports of metastatic spread to the submandibular gland from the oral cavity tumour has been reported in some series but is a rare occurrence. Tumour metastasis to the submandibular gland from the breast and genitourinary system is more likely. Most of the series investigating submandibular gland involvement from oral cavity tumours have shown local invasion and extracapsular spread from nodes are the most likely mechanisms. Local invasion is the predominant mechanism especially for advanced T stage. The incidence of submandibular gland invasion in early T1 and T2 oral cavity was found to be very low; 0.03% in the series by Ali *et al*[6] as only 1 in 253 cases was found. Tseng Cheng *et al*'s[3] series with 383 neck specimens of all stages had an incidence of 1.8%. The incidence when only T4 were considered was 6.67% as there were 90 specimens of this stage in this series. Our study focused on advanced oral cavity squamous cell carcinoma and had an incidence of 1.52%. Our series had 125 bilateral neck dissections while Tseng's had 41. It is likely that lower chances of involvement of the contralateral submandibular gland could account for the lower incidence in our study. Four of the 6 submandibular glands with carcinoma invasion were T4b while the remaining 2 were T4a. Previous studies had incidences much higher than ours despite having included all T stages. The mechanism of involvement of the gland in our series was by direct tumour invasion as was evidenced by CT scans as there was loss of the margin between the gland and tumour. Review also revealed 4 of the 6 had lymph node metastasis with irregular node margins. It is however difficult to conclude with certainty that the irregular margins of the nodes was due to extracapsular as it could as well be due to tumour erosion of node capsule as the primary tumour was in close proximity. This was easily identified in the CT scans which can be used to evaluate the status of the gland prior to surgery. Basaran *et al*[4] recommended that the decision to spare or extirpate the submandibular gland should be made intraoperatively after clinical assessment of invasion. This can be correlated in our study as 5 of 11 glands with CT features of invasion had negative pathology.

Other authors have proposed submandibular gland sparing in the contralateral neck of early stage oral cavity squamous cell carcinoma. This principle is supported by the fact that the gland is devoid of intraparenchymal lymph nodes. Muthuswarmy *et al*[5] demonstrated that it was possible to dissect the perivascular and periglandular nodes sparing the submandibular gland without compromising on oncological clearance. Contralateral submandibular gland sparing in early oral cavity squamous cell carcinoma has been recommended especially for those not receiving postoperative radiotherapy to prevent xerostomia. Parotid gland sparing with IMRT at levels less than 24Gy has been shown to reduce xerostomia as this gland is responsible for 90% of stimulated saliva. Sparing the submandibular gland is also of paramount importance as it is responsible for 70% unstimulated saliva. The submandibular gland is less radiosensitive than the parotid and thus higher levels of irradiations can be tolerated. Shielding of the submandibular gland in oral cavity squamous carcinoma is not recommended as it will compromise disease control due to proximity. Seikal-Jha procedure has been described of submandibular gland transfer to the submental region with subsequent shielding from radiation in tumours with low incidence of level 1 lymph node metastasis[7]. Prevention of xerostomia in 83% of the patients with preservation of one submandibular gland has been achieved by use of this technique. They however don't recommend the procedure for oral cavity squamous cell carcinoma. More recently, Hani *et al*[8] described a procedure to transfer the submandibular gland to the parotid region where it can be shielded from radiation and allow maximum radiation to the oral cavity. Though they report of the procedure in 9 patients only, it offers hope for oral cavity SCC patients if further prospective studies prove efficacious and of oncological safety.

Tumours of the floor of the mouth, lower alveolar and tongue have a higher incidence of local invasion into the submandibular gland due to proximity. The other subsites in their advanced stages have the potential to invade the gland as well. CT scan is able to demonstrate local extension of tumour to the submandibular gland and can thus be used to make preoperative planning regarding the extent of excision. Enbloc excision of the oral tumour with the neck specimen on the side of the lesion is recommended where possible to reduce incidence of recurrence through intransit lymphatics. Charles *et al*[9] found a significant poorer outcome in oral cavity cancer patients who had discontinuous neck dissection

when compared to those with a continuous enbloc neck specimen as the former experience a higher incidence of neck recurrence. The tongue and floor of mouth occasionally have in association the lateral or median lingual lymph nodes that are related to the genioglossus in the drainage path towards the submandibular triangle. The incidence is however rare with less than 1% of oral SCC having radiological evidence or involvement[10]. Routine neck dissection will not clear these nodes due to location. Enbloc excision with resection of the floor of the mouth muscles together with portion of the genioglossus could offer oncological clearance. Masahiro *et al*[11] report on their management recommend dissection only when presence is confirmed by CT scan or MRI. Where it is not possible to have an enbloc excision, marking the edge of the neck and oral cavity specimen for proper assessment of adequacy of resection is recommended. Our series did not have any case of bilateral or contralateral submandibular gland involvement. Oral cavity carcinomas that are well lateralized and not crossing the midline have a low incidence of bilateral neck metastasis.

CONCLUSION

The incidence of submandibular invasion in advanced oral cavity squamous carcinoma is low and is unlikely to have bilateral or contralateral lymph node metastasis in well lateralized tumours. We hypothesize that the contralateral submandibular gland even in advanced well lateralized oral cavity SCC if not grossly invaded by tumour or with adjacent positive nodes on frozen section can be transferred to the parotid region to allow shielding during radiation of the oral cavity. Further prospective studies are required to before adoption of the recommendations.

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CASE REPORT

COIL EMBOLIZATION FOR LINGUAL ARTERY PSEUDO-ANEURYSM CAUSING MASSIVE POST TONSILLECTOMY BLEEDING: A CASE REPORT

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ABSTRACT

Severe haemorrhage is a life-threatening complication of tonsillectomy. A case of a 44 year old male with severe post tonsillectomy haemorrhage due to lingual artery pseudoaneurysm is presented. The patient presented with recurring massive post tonsillectomy haematemesis occurring three times over a two month period. Intravenous fluid and blood transfusions were required in each of the epsiodes in addition to surgical control. An initial CT carotid angiogram was normal. A subsequent digital subtraction angiogram of the right common carotid artery showed an irregular lobulated pseudoaneurysm of the right lingual artery which was the cause of the bleeding. This was successfully embolized with coils and bleeding ceased immediately. No further bleeding was experienced in a two year follow up period .

Key words: Lingual artery pseudoaneurysm, Embolization, Post tonsillectomy haemorrhage

CASE REPORT

A 44 year old man presented with acute massive haematemesis two months after a successful tonsillectomy for recurring tonsillitis associated with tonsillar abcesses.The patient was in hypovolumic shock and required IV fluid rescuscitation, four units of blood transfusions and ICU care for stabilization. Once stable the patient was taken to theatre for examination under anaesthesia and tonsillar bed was surgically packed with surgicel. Subsequent torrential haematemesis occured on the third day of admission requiring further tonsillar bed cauterization and surgical packing which was partially successful in bleeding control and necessitated interventional radiology consult.

The patient had initially presented with similar complaints twice at another hospital facility over a two week interval. Intravenous fluid and blood transfusions had been given. In addition surgical packing with surgicel, diathermy coagulation and oversuturing of the tonsillar faunices had been done in an attempt at bleeding control.

In the current admission initial CT angiograms of the neck vessels were normal (Figure1). Subsequent DSA angiograms of the right common carotid artery showed an irregular lobulated pseudo aneurysm of the right lingual artery (Figure2a). This was successfully obliterated by coil embolization via trans femoral arterial route (Figure 2b,c). The entire artery from distal to proximal was coiled using one each of 4mm, 3mm and 2mm pushable embolization coils (Tornado, Cooks).

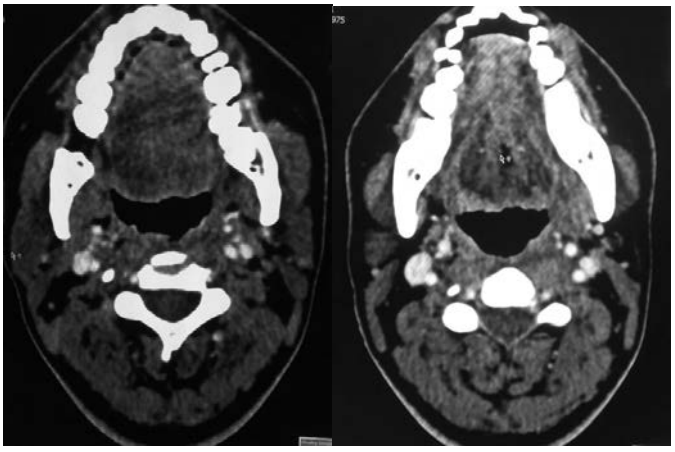

Figure 1a **Figure 1b**

Axial neck contrast CT scans of a 44 year old male with post tonsilectomy bleeding during the third hospital admission. Carotid vessels are normal. No vascular abnormality or active bleeding was seen in the shown axial images and further coronal and sagittal images not shown

Figure 2a **Figure 2b** **Figure 2c**

Figures 2a-2c DSA angiograms of the right common carotid artery of same patient shows an irregular lobulated pseudo aneurysm of the right lingual artery measuring about 4mm in diameter. This was successfully obliterated by coil embolization via trans femoral arterial route [2b]. The coils are seen along the extent of the artery beginning from the ostial origin (Figure 2c). Bleeding stopped immediately after the embolization with no recurrence. Thepseudo aneurysm was thought to have been caused by cauterization during the first two surgical procedures

Bleeding stopped immediately after the coil embolization and there was no recurrence in a four months follow up period. The patient presented to ENT emergency in the fifth month with foreign body sensation in the throat at swallowing. At direct laryngoscopy under anaesthesia one coil was found extruded into the oropharyngeal mucosa and was successfully removed under direct visualization. No further problems or bleeding were experienced in a further two year follow up period.

DISCUSSION

Tonsillectomy is a common surgical procedure representing approximately 20%-40% of ENT surgical procedures[1]. This procedure can be performed as a day case or admission depending on the severity of complications anticipated. Postoperative complications following tonsillectomy include haemorrhage, infection, vomiting, odynophagia, nausea, vomiting and pain. Dehydration may occur in children due to delayed or poor oral intake. Post-tonsillectomy haemorrhage remains the most serious complication of them all[2].

More than half (67%) of post tonsillectomy bleeding originates in the tonsillar fossa and the rest in the nasopharynx[3]. There are two major time frames for the post operative bleeding. Most often, the bleeding occurs within the first 24 hours after surgery and forms about 75% and is considerd primary bleeding. This is generally related to surgical technique and incidence has declined over the past few years due to improved surgical techniques. Secondary haemorrhage occuring more than 24 hours post tonsillectomy accounts for 25% of post tonsillectomy bleeds.The most severe haemorrhages are due to arterial dissections or pseudoaneurysms. Pseudoaneurysms can be caused by blunt or direct trauma during dissection or the placing of ligation sutures in the tonsillar bed and can be from facial or lingual arteries[4]. The pseudoaneurysm is actually a collection of blood that forms between the two outer layers of the artery, the muscularis propria and the adventitia. In the case presented, the pseudoaneurysm is believed to have been caused by trauma to the lingual artery during the initial surgery possibly from diathermy coagulation or suture ligation. Lingual artery pseudoaneurysms are quite uncommon though with sporadic case reports in the literature, and are associated with severe haemorrhage[2,5].

The diagnosis can be made by CT angiography although it can be misssed like in this case due to the small size. Digital subtraction angiography is more often required to establish the diagnosis and also guide endovascular management.

While surgery was the gold-standard treatment in the past, several less invasive treatment options are popular today[6]. Endovascular methods of managing haemorrhages caused by the pseudoaneurysm may include Ultrasound Guided Compression (USGC), thrombin injection, arterial embolisation, endovascular stent graft insertion among others. In this case, coil embolization was done. Three pushable coils of 4mm, 3mm and 2mm (Cooks) were used. The choice for embolization was due to failure of the initial two surgical management options. This may be expected as a pseudoaneurysm in the tonsillar region specifically should not be cauterized; infact the sac become worse with cauterization. Oversuturing of the tonsillar faunices help but the pseudoaneurysm is likely to bleed once the sutures absorb and edema of the tonsillar bed tissues resolve. Ultimately complete obliteration of the pseudoaneurysm is required and is best done with embolization which obliterates the sac, and if need be the parent artery to prevent further bleed by recanalization of the parent artery from distal collateral circulation.

CONCLUSION

Lingual artery pseudoaneurysms although rare can cause life-threatening post tonsillectomy haemorrhage and should be considered as cause in recurring posttonsillectomy haemorrhage. These are safely embolized in a minimally invasive endovascular procedure which needs to be considered in the initial stages of management and not as a last resort.

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