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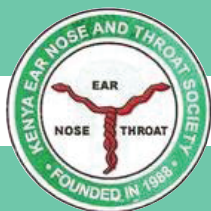
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BEYOND THE OTOSCOPE: CHALLENGES IN OTOLARYNGOLOGY PRACTICE IN THE DEVELOPING WORLD - NOW AND BEYOND

Otolaryngology – The Practice of Ear Nose and Throat, Head, Neck and related Conditions

Sharon Pearson once said “It isn’t the challenge that defines you, its what you do with it”

Developing countries are facing unprecedented burdens to their capacity to deliver optimum health care outcomes to their population. This is especially so in specialist practice like otolaryngology which may have to compete for attention with communicable diseases such as malaria and tuberculosis for scarce resources allocated by governments to health.

Article 43 of the Kenya constitution states that “Every person has the right to the highest attainable standard of health”.

Gaps in global health that manifest more clearly in developing and emerging economies, present a modern challenge to otolaryngologists and allied professionals, as they seek to find their place among other medical professionals in the modern healthcare framework.

A survey by the World Health Organization global action plan, found that non communicable diseases accounted for over 60% of global mortality in 2008, and disabling hearing loss is the most common disability internationally. Disabling hearing loss presents a significant challenge to practicing otolaryngologists and allied professionals in the developing world.

A report published by the global burden of diseases and risk factors disease control priorities ranks hearing disability third on the list of most common non fatal conditions in low and medium income countries.

A report by the World Health Organization deafness and hearing impairment report noted that the developing world accounts for 80% of the worlds 278 million people with bilateral conductive hearing loss, yet only 1 in 40 people who would benefit from hearing aids has one.

Gaps in the availability of equipment that are cost effective and of acceptable quality and difficulties in the access to high quality and relevant targeted training, still remain a challenge to otolaryngologists and allied professionals in the developing world.

Finally, there is need to implement policies, laws and programs that aim to deliberately reduce gaps in healthcare funding and especially for otolaryngologists in the developing world. Implementation of the Abuja declaration that seeks to allocate 15% of every countries’ annual budget to health infrastructure would be a viable starting point.

We must free ourselves from viewing healthcare as a hierarchical privilege, and place health on a more equality based trajectory. We must deliberately create implementational matrices that are equitable and achievable, for short, medium and long term goals that aim to mitigate the challenges of access of quality healthcare, especially in otolaryngology practice.

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OUTCOME OF THYROIDECTOMIES IN A HOSPITAL SETUP IN KENYA: A PROSPECTIVE CROSS-SECTIONAL ANALYTICAL STUDY

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ABSTRACT

Background: Thyroidectomy is a surgical procedure usually done under general anaesthesia. Diagnosis, treatment planning and surgical intervention in thyroid disease are critical in determining good outcomes post surgery.

Objective: The purpose of the present study was to present our experience and evaluate the outcomes of thyroid surgery under general anaesthesia.

Methodology: This was a cross-sectional analytical study carried out primarily at Makueni County Referral Hospital, Makueni, a 150 bed general referral hospital, from January 2019 to December 2021. One hundred and one patients underwent thyroidectomy for benign and malignant diseases under general anaesthesia. The patients ages ranged from 9 years to 67 years. After appropriate pre-operative investigations were done, all patients underwent various types of thyroidectomy procedures.

Results: Ninety nine were female while two were male. Female to male ratio was 98:2. The median age was 42.0 (IQR 33.0 – 51.0) years. Forty three point six percent presented with right thyroid mass, 37.6% presented with left thyroid mass. Ten point eight percent presented with median isthmus mass, while 8.0% presented with diffuse bilateral thyroid mass. Surgery included left hemithyroidectomy 35.7%, right hemithyroidectomy 39.6%, isthmusectomy 11.9%, sub total thyroidectomy 4.0% and total thyroidectomy 3.0%. Histological diagnosis included a diagnosis of benign colloid goitre (77.2%), follicular adenoma (8.9%), follicular carcinoma (7.9%), papillary carcinoma (4.0%) and lymphocytic thyroiditis (2.0%). Post operative complications (5%) included change in voice pitch (1 patient), difficulty breathing (1 patient), expanding haematoma (1 patient), prolonged hoarseness of voice (1 patient) and persistent hypocalcemia (1 patient). Average length of stay was 2.0 (SD 0.4) days. While three patients (3.0% n = 101) had post operative surgical drains.

Conclusions: In our study, thyroidectomy is an effective and safe surgical procedure to address various lesions within the thyroid gland. There is a high prevalence of thyroid conditions in the female population as compared to the male population. Complications are low when proper pre operative evaluation is done and meticulous surgical technique is employed.

Key words: Thyroidectomy, General anaesthesia, Thyroid surgery, Thyroid gland, Surgical drains

INTRODUCTION

The thyroid gland is a butterfly shaped organ located in front of the neck under the voice box. It overlies the second and third tracheal rings. It is about 5cm each in transverse length and 2 to 3cm length in the antero posterior dimension. A superior extension of the thyroid gland, the pyramidal lobe is found in 70% of cases. The normal size of a thyroid gland in adult men is 20 to 60grams and adult female is 20 to 40 grams¹.

Diseases and disorders of the thyroid gland may develop and may be related to production of too little

or too much thyroid hormone, thyroid nodules or lumps, abnormal thyroid growths or thyroid cancers.

A goiter is an abnormal enlargement of the thyroid gland and may be one sided, diffuse, or confined within part of a lobe or isolated within the isthmus of the thyroid³. Simple goiters may be 60grams or more and may eventually convert to a multinodular goiter. Thus, multinodular goiter is the result of repeated hyperplasia and involution with degeneration of a simple goiter. When the thyroid gland weighs 100grams or more, the cause is likely due to heterogenous response of follicular epithelium to Thyroid Stimulating Hormone (TSH).

Goitre affects women at a proportionately much higher frequency with a female to male ratio of 9:1. It may also develop more frequently in adolescence and pregnancy. Thyroid enlargement may be due to a multitude of factors which may include increased TSH secretion mainly due to iodine deficiency or autoimmune thyroiditis (Hashimoto's disease), oxidative damage to follicular cells causing hyperplasia, genetic factors with concomitant risk factors including Sertoli Leydig tumours, drug induced, including amiodarone treatment which may interfere with thyroglobulin synthesis and goitrogens, found in cassava, cabbage cauliflower and other vegetables and may interfere with iodine transport within the gland³.

Diagnosis includes clinical exam which may reveal an enlarged median or para median thyroid gland. Majority are euthyroid in nature. Hypothyroid goiter may be due to decreased production of T3 (Tri iodothyronine) T4 (Thyroxine) hormones while hyperthyroid goiter may be due to autonomous transformation. Compression of trachea and esophagus is uncommon as enlargement is usually toward the strap muscles and sub cutaneous tissue. Rarely there may be compression of the recurrent laryngeal nerve and phrenic nerve leading to paralysis and hoarseness of voice or diaphragmatic paralysis respectively.

Ultrasonography may reveal an enlarged iso or hyper echoic gland with surrounding hypo echoic halo⁴. There may also be a sponge like or a honey comb pattern. Anechoic areas may also contain colloid with areas of internal calcification. Thyroid ultra sounds are generally reported using the Thyroid Imaging Reporting and Data Systems (TIRADS). Thyroid nodules are present in about 50% of the population in the United States. About 95% of thyroid nodules are benign⁵. Fine Needle Aspiration (FNA) may be indicated if there is a history of rapid growth, pain or tenderness, unusual firm areas in the nodules or suspicious areas on sonography. Characteristics of thyroid nodules that raise suspicion include central vascularity, more rounded, more than 1cm in diameter, micro calcifications and irregular borders. FNA reports were classified according to the Bethesda system. Pathological characteristics include Hurthle cells, Hurthle edges, Lymphocytic infiltration, and shrunk follicular cells.

Treatment options may include medication which includes Carbimazole for primary hyperthyroidism with or without propranolol for symptom control through their inhibitory action of adrenergic effects⁶. Radioiodine treatment may also be employed.

Surgical options include lobectomy, partial or total thyroidectomy. Surgical approaches depend on

clinical suspicion, FNA and ultra sound findings.

Thyroidectomies are one of the most common procedures in endocrine surgery⁷. In recent years, the number of thyroidectomy procedures has increased in number due to a rise in the incidence of thyroid malignancies, which now account for 1.7% of the total number of malignancies worldwide⁸.

Makueni County Referral Hospital is the primary level 5 referral hospital in Makueni County, lower eastern region serving a population of about 1.3 Million people. Makueni County is located at about 3,117 feet above sea level and has a geographical area of 8,008 km². Majority of patients seek tertiary services including surgical procedures. Majority of these surgeries were performed at Makueni County Referral Hospital.

MATERIALS AND METHODS

This study sought to audit the outcome of thyroidectomies performed by an otolaryngologist primarily at the county referral hospital and analyse the outcomes and histological characteristics. The baseline results will be useful for future analysis of this procedure and for future surgical planning for thyroid surgery.

A prospective data analysis and follow-up was performed over a three year period on patients who underwent thyroidectomies by the author between January 2019 and January 2022. The principal author is the consulting Otolaryngologist Head and Neck Surgeon attached to Makueni County Referral Hospital. Data on type of goiter, type of surgery, length of stay, complication and histological analysis were recorded. Hypocalcemia was defined as serum calcium levels of less than 2.20mmol/l following thyroidectomy. Recurrent laryngeal nerve palsy was defined as hoarseness of voice immediately post operatively and lasting for more than one week and failing to resolve after 6 months and confirmed on indirect laryngoscopy. Bilateral recurrent laryngeal palsy was defined as immediate hoarseness of voice requiring emergent intervention and lasting for more than 24 hours. Expanding haematoma was defined as immediate post operative difficulty in breathing and drop in oxygen levels in the background of an expanding anterior neck mass lesion.

Type of study

The mean age of the patients was 41.9 (SD 13.0) years, where the minimum age was 9.0 years, and the maximum age was 67.0 years. The median age was 42.0 (IQR 33.0 – 51.0) years.

The complications as experienced from the 5 patients were change in voice pitch (1 patient), difficulty breathing (1 patient), expanding

haematoma (1 patient), hoarseness of voice for 2 months (1 patient), and persistent hypocalcemia (1 patient).

Table 1: Patients characteristics

		Frequency (n=101)	(%)
Sex	Male	2	2.0
	Female	99	98.0
Age (years)	≤20	6	5.9
	21-30	10	9.9
	31-40	28	27.7
	41-50	31	30.7
	51-60	17	16.8
	>60	9	8.9

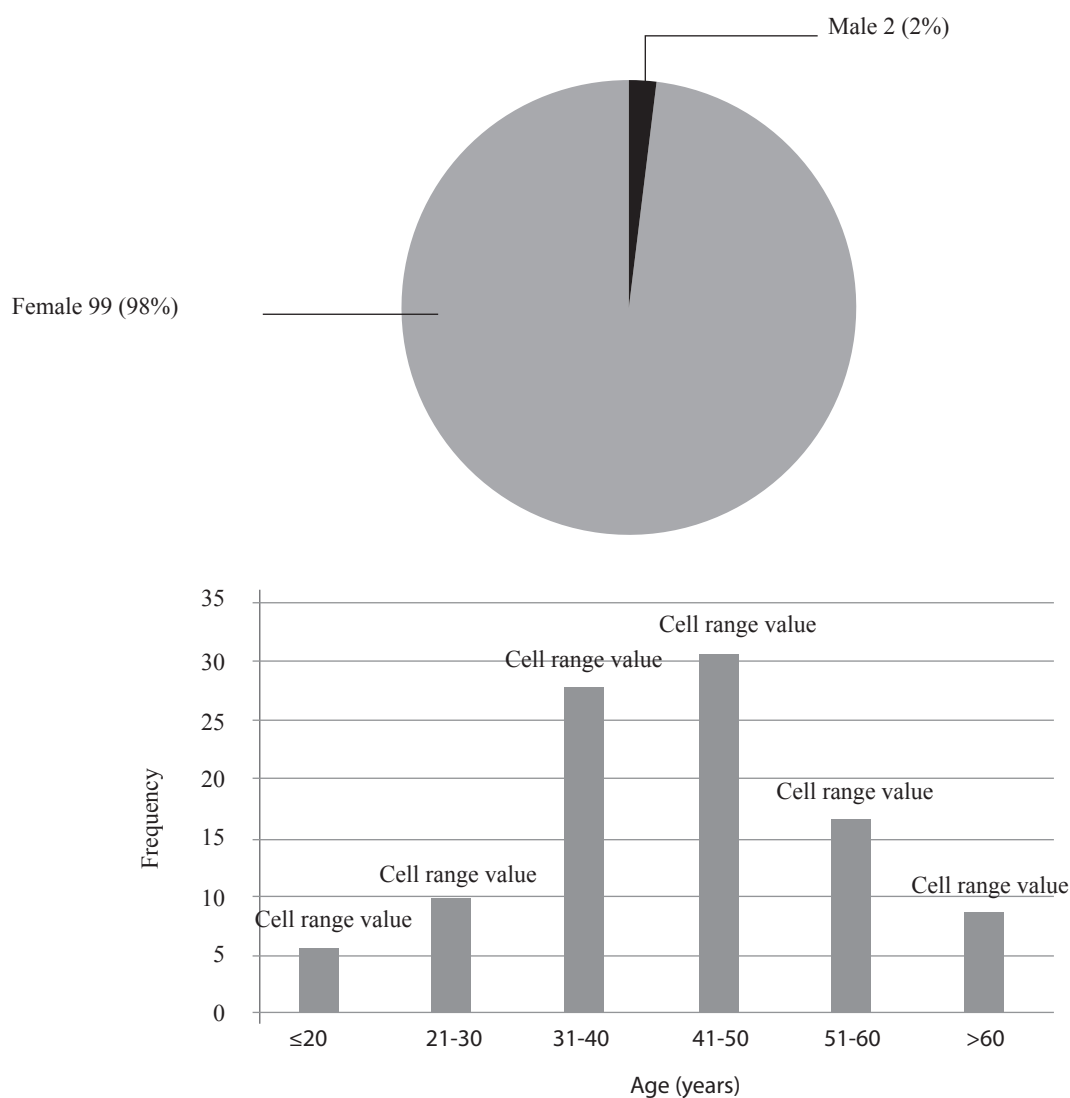


Table 2: Laterality

	Frequency (n=101)	(%)
Right	44	43.6
Left	38	37.6
Median isthmus mass	11	10.8
Diffuse bilateral	8	8.0

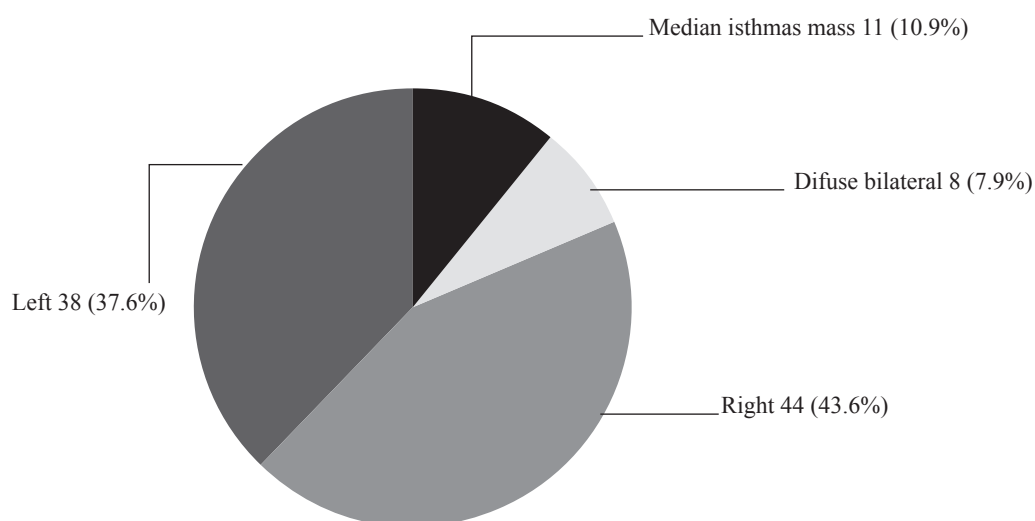


Table 3: Surgery

	Frequency (n=101)	(%)
Hemithyroidectomy with parathyroidectomy right	1	1.0
Isthmusectomy	12	11.9
Left hemithyroidectomy	36	35.7
Right hemithyroidectomy	40	39.6
Right lobectomy	3	3.0
Right lobectomy with isthmusectomy	1	1.0
Sub total thyroidectomy	4	4.0
Total thyroidectomy	3	3.0
Total thyroidectomy with central neck dissection	1	1.0

Table 4: Diagnosis

	Frequency(n=101)	(%)
Benign colloid goitre	78	77.2
Follicular adenoma	9	8.9
Follicular carcinoma	8	7.9
Papillary carcinoma	4	4.0
Lymphocytic thyroiditis	2	2.0

Table 5: Complications

	Frequency(n=101)	(%)
Yes	5	5.0
No	96	95.0

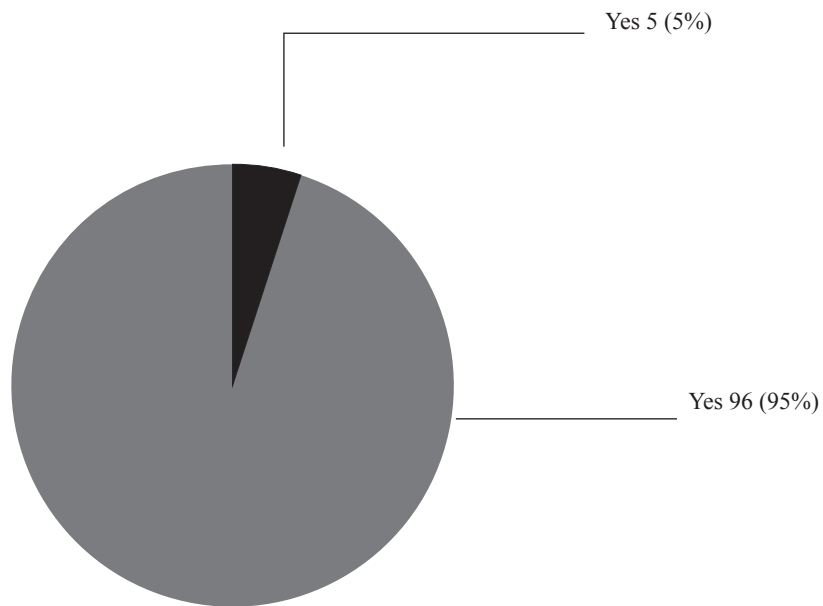


Table 6: Length of hospital stay

	Frequency (n=101)	(%)
1.5	24	23.8
2.0	71	70.3
3.0	5	5.0
4.0	1	1.0

The mean length of stay was 2.0 (SD 0.4) days

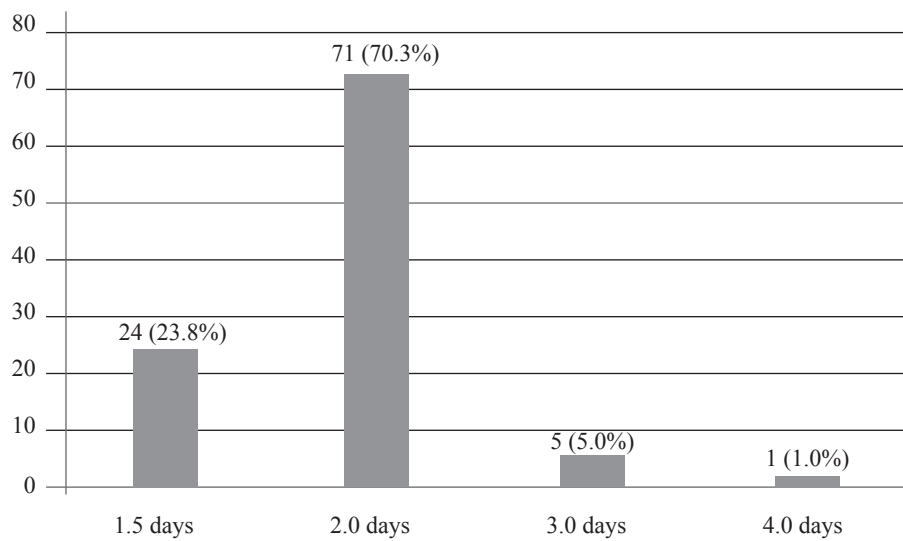
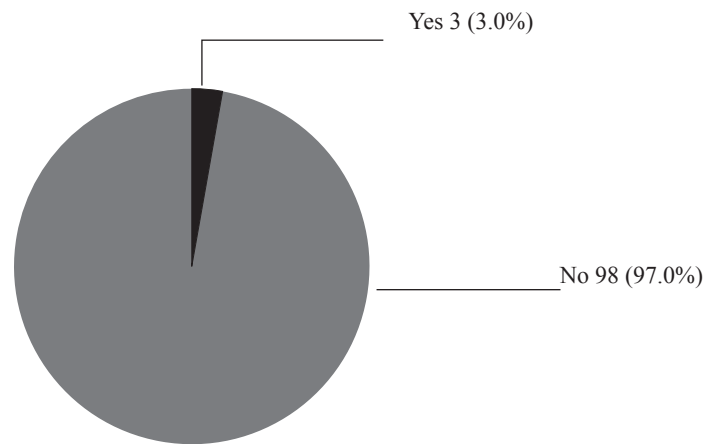


Table 7: Surgical drains

	Frequency (n=101)	(%)
Yes	3	3.0
No	98	97.0



DISCUSSION

Thyroid surgery represents one of the common head and neck surgeries performed in patients. Females are much more commonly affected by thyroid conditions than men⁹. Approximately 1 in 8 women will be affected by a thyroid condition at some point in their lives. The risk for women is about 10 times higher than for men. One reason for this is that thyroid disorders are often triggered by autoimmune responses, which happen when the body's immune system starts to attack its own cells. We still don't understand what causes these responses, but we do know that autoimmune conditions are more common in women than in men.

Another reason for the prevalence of thyroid disorders in women is that there is an interplay between thyroid hormones and the hormones that fluctuate during the menstrual cycle.

One hundred and one patients were scheduled for elective thyroidectomies at our hospital between January 2019 and December 2021. They were reviewed and pre operative work up was done for thyroidectomy, and met the criteria for surgery. Laboratory and clinical workups were done for each patient. Ninety nine thyroidectomies were female while two patients were male. Female to male ratio was 98:2. The median age was 42.0 (IQR 33.0 – 51.0) years. Forty three point six percent presented with right thyroid mass, 37.6 % presented with left thyroid mass. Ten point eight percent presented with median isthmus mass, while 8.0% presented with diffuse bilateral thyroid mass. Masses were clinically identified by palpation and confirmed by ultrasound. Any patient who met criteria for fine needle aspiration cytology workup was offered the service. Bethesda system was used to classify thyroid nodules. Ultrasound criteria was based on the TIRADS (Thyroid imaging reporting and data systems) reporting tool to determine risk stratification for malignancy¹⁰. Various thyroidectomies were

performed based on the clinical, radiological and pathology criteria. Type of thyroidectomy performed was based on the 2015 American Thyroid association guidelines on thyroid nodules and thyroid cancer management¹¹. The most common procedure was a hemi thyroidectomy (75.3%). This represented the commonest procedure performed in this setup. Further stratification into a hemithyroidectomy (35.7%), and right hemithyroidectomy (39.6%) was analysed. Other procedures included isthmusectomy (11.9%), sub total thyroidectomy (4.0%) and total thyroidectomy (3.0%). The commonest histological diagnosis was benign colloid goitre (77.2%). This was followed by follicular adenoma (8.9%), follicular carcinoma (7.9%), papillary carcinoma (4.0%) and lymphocytic thyroiditis (2.0). This result shows that approximately 17% to 20% of histological diagnosis may reveal a histological carcinoma. Most are low risk tumours. Post operative complications were reported in five (5%) patients. This included change in voice pitch (1 patient), difficulty in breathing (1 patient), expanding haematoma (1 patient), prolonged hoarseness of voice (1 patient) and persistent hypocalcemia (1 patient). The incidence of bleeding after thyroid surgery is low (0.3-1%), but an unrecognized or rapidly expanding haematoma can cause airway compromise and asphyxiation. Recurrent Laryngeal Nerve (RLN) injury results in true vocal-fold paresis or paralysis. Deliberate intraoperative identification and preservation of the RLN minimizes the risk of injury¹². The average length of stay was 2.0 (SD 0.4) days. This is mainly due to hospital based protocols on admissions and discharges that may lead to varying duration of hospital stay. Average length of stay may vary from 1 day to 4 days in hospital based setups¹³.

Post operative surgical drains may be preferred in certain setups or thyroid conditions. In many facilities, drains are routinely inserted after thyroidectomy with the aim of preventing haematoma formation and accumulation of seroma. The continued use of

drains may be based more on tradition rather than proven scientific evidence¹⁴. Three patients (3.0% n = 101) had post operative surgical drains. In one study, it was found that mean duration of hospital stay after thyroidectomy was significantly higher among the patients with surgical drains as compared with the patients with no drain. In a similar study, the incidence of higher pain scores were found among the patients with surgical drain after thyroidectomy than those without surgical drains¹⁴.

CONCLUSIONS

This study shows that thyroid enlargement is mainly predominant in females. It also displays that the commonest presentation of thyroid enlargement is a unilateral thyroid lobe enlargement. Guidelines on thyroidectomy procedure was according to the 2015 American Thyroid Association guidelines on thyroidectomy for nodules and tumours. Thyroid carcinoma is also prevalent among presentations of thyroid masses and should be given special attention. Majority of thyroid cancers in this study were low risk thyroid cancers. Post-thyroidectomy drain insertion was not frequently performed as this is mainly a surgeon's preference and is not based on any existing guideline. Further research may be required to formulate recommendations and practice guidelines regarding the insertion of postoperative drains following a thyroidectomy. Our study therefore reveals that thyroidectomy is an effective and safe surgical procedure to address various growths within the thyroid gland.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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No funding was received for this study.

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VALIDATION OF THE USE OF HEAR SCREEN, A MOBILE PHONE APPLICATION AS A SCREENING TOOL FOR HEARING LOSS AT THE KENYATTA NATIONAL HOSPITAL

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ABSTRACT

Background: Conventional Pure-Tone Audiometry (PTA) test is considered the gold standard hearing test. The World Health Organization (WHO) has recognized mobile applications as having potential for audiometry screening.

Objective: To validate the mobile phone based application hear screen as a screening tool for hearing loss at the Kenyatta National Hospital (KNH).

Methods: This was a prospective study conducted at KNH on 40 patients referred for conventional PTA. Convenient sampling was done. This was an experimental study where each ear was tested and analyzed separately. Hearing was compared across all frequencies in all modalities with proportion and 95% confidence interval determined. Regression analysis, T-test and Fischer's exact test were used for comparison between smartphone and conventional, mean testing time and preference of use respectively.

Results: There was no statistical difference between smartphone and conventional PTA across all frequencies with a regression coefficient of 1.26 and a $p < 0.01$. Time taken to do the tests had no statistical difference $p < 0.01$. Majority of the respondents preferred to use conventional audiometry with no correlation ($p=1.00$) between this preference and their age or level of education.

Conclusion: Hear test the threshold version of hear screen provides thresholds comparable to conventional PTA in both the quiet office set up and normal clinic set up and can thus be used reliably as a screening tool.

Key words: Hear screen, Pure tone audiometry, Screening, Smartphone

INTRODUCTION

Approximately 466 million people in the world have disabling hearing loss with the highest burden being in Pacific Asia and Sub-Saharan Africa where the prevalence of hearing impairment in Sub-Saharan Africa is estimated at 9%¹. Hearing loss has a great impact in the social, cultural, developmental and economic aspect of the individual thus early detection and timely intervention can help curb these effects. Availability of Ear Nose Throat (ENT) healthcare professionals in developing countries is limited with an estimate of less than one audiologist for every one million people and less than one ENT specialist for every one million people in Sub-Saharan Africa².

Increase of innovative technology and global connectivity has resulted in mobile health being widely proposed as an affordable acceptable option to combat the shortage of and access to skilled health

care professionals. The WHO has put a lot of focus on implementing primary health care services in hearing services and use of mobile health has been one of the strategies³. The Pure Tone Audiometry (PTA) test is considered the gold standard for detection of hearing loss. This test requires an audiologist who is a skilled health worker and equipment in the form of an audiometer and should be carried out in a modified environment which is a sound proof room or booth. These services are not readily available in our country and the developing world at large due to the lack of infrastructure, equipment and enough audiologists². Innovative use of technology and mobile phone applications would serve to bridge this gap.

There are applications developed to test hearing either by air conduction or speech audiometry and are readily available in the market. One such application is hear screen application which offers

user friendly and affordable hearing tests for children and adults⁵. This validated application is designed to detect hearing problems and provide referrals by linking one up to the nearest health care providers and helps to keep records. It has a threshold version hear test which is an air conduction pure tone test that tests 500 to 8 KHz frequencies at intensity levels between 10 to 100dB. It works on android and iOS devices coupled with calibrated ear phones and the average test takes 12 minutes. Several countries have adopted hear screen and hear test and integrated it into their National ear screening programmes. In this study we will be validating the threshold version of hear screen as a screening tool in Kenyatta National Hospital.

Objectives

Research question

Can Hear Screen, a smart phone based mobile application be used as a screening tool in KNH?

Primary objective

To validate the threshold version of the application Hear Screen, as a hearing screening tool in Kenyatta National Hospital

Secondary objectives

- To compare the hearing threshold smartphone based PTA in a normal clinic setup with conventional PTA.
- To compare the hearing thresholds of smartphone based PTA in an office setup with conventional PTA.
- To determine the time frame taken to carry out each of the tests
- To determine the preference of use of the tests

Null hypothesis

Pure tone audiometry can only be done in a soundproof booth to get accurate results.

Alternative hypothesis

Pure Tone Audiometry can be done in any environment as long as it's done using automated audiometry.

MATERIALS AND METHODS

This was a prospective experimental study where a repeated measure within subject design was employed. Participants were conveniently sampled for PTA and afterwards underwent the hear screen test both in a quiet office and in the normal clinic environment. The sample size was calculated using the equation stated by Brujang and Baharum 2017. Alpha was set at 0.05 and power at 0.9. A sample size of 40 participants was determined and each ear was analyzed separately resulting in a total sample of 80. Convenient sampling was used to select participants. All patients sent for pure tone audiometry aged above 18 years were included in the study while any deaf patients or patients with single sided deafness were excluded

The study was carried out in the ENT Department in Kenyatta National Hospital among patients who were sent for PTA. The conventional PTA was carried out in the sound proof booths within the audiology section of the clinic which have an ambient noise level of 21dB as per the ISO 1989 standards. The smartphone test was carried out in a quiet room and in the clinic. The quiet room had very little traffic thus had very little interference from outside noise and had an average ambient noise level of 35dB. The clinic set up was in one workstation within the ENT filter clinic with an average ambient noise level of 45dB.

Standard otoscope and speculums, 512Hz tuning fork, mobile phone Samsung galaxy A3, Sennheiser HD280 PRO supra aural earphones, Clinical Audiometer – Interacoustics AC33, and Telephonics TDH39 supra aural earphones for PTA were used for the study

Univariate analysis was carried out to determine the mean age of the participants and their gender distribution. The mean durations with standard durations of length for testing procedure between all arms of the study was calculated. Proportion with 95% confidence intervals was calculated to determine grading of hearing loss. For the comparison of repeated measures the Wilcoxon signed rank test was used. Students' tests were carried out to determine if the mean time taken was significantly different between the tests. Fishers exact test was used to determine whether degree of education or age in years impacted on the choice of modality between conventional PTA and smartphone PTA on each ear was analysed separately. Two way scatter plots were derived after regression fit analysis (with

95% confidence interval) to compare agreement of smartphone in clinic setting and office setting versus gold standard PTA.

RESULTS

The study had a total of 40 subjects with a mean age of 42.8 years (18 to 82 years). There were 14 (35%) males and 26 (65%) females. Majority of the

participants 19 (47.5%) had tertiary education, 14 (35%) had secondary education and 7 (17.5%) had primary education. Thirty (75%) respondents had subjective hearing loss while (25%) did not have subjective hearing loss. Weber's was central in 23 (57.5%) and lateralized in 17 (42.5%) participants and Rhine's positive in 37 (92.5%) ears and negative in 3 (7.5%) ears.

Mean thresholds across all frequencies in all the three tests

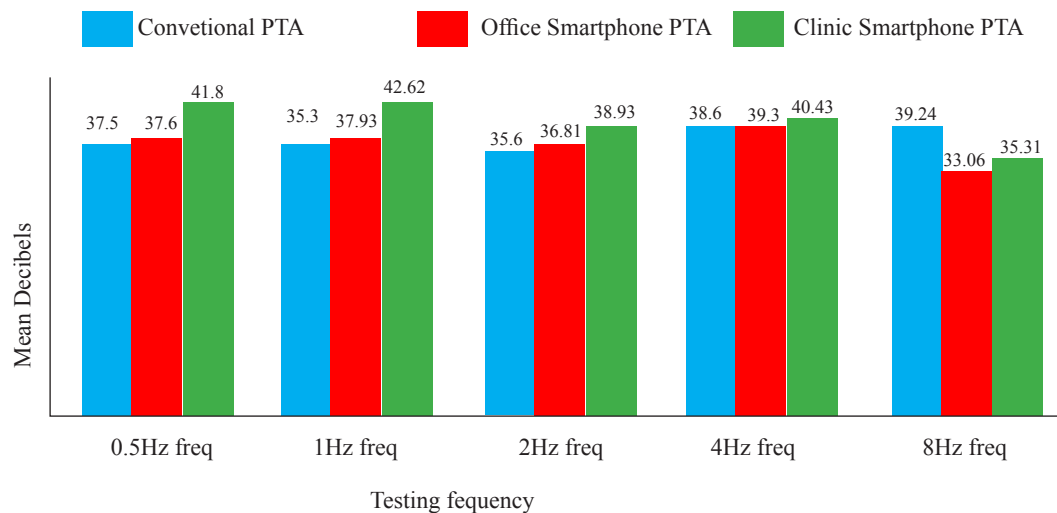


Figure 1: Bar chart showing mean thresholds across all testing frequencies

The mean decibels for each test carried out on each frequency were almost similar and within 5dB of the conventional PTA except in the 8Hz frequency which was within 10db. All were within the accepted audiological limits.

Distribution of the degree of hearing loss from pure tone average (95% confidence interval)

As shown in Table 1, pure tone averages were obtained from the mean thresholds of 0.5, 1, 2, 4kHz.

This was done to determine the level of hearing, all the three tests were comparable with a difference that was not statistically significant with p value of < 0.01. However it should be noted that the threshold increases with the increase in the ambient noise levels as depicted in the clinic based smartphone test result.

Table 1: Distribution of the degree of hearing loss from pure tone average (95% confidence interval)

	Conventional PTA		Smart phone office		Smart phone clinic	
	Right (%)	Left (%)	Right (%)	Left (%)	Right (%)	Left (%)
Normal	52.5 (36.6-67.9)	50 (34.3-65.8)	42.5 (27.7-58.7)	47.5 (32.1-63.3)	37.5 (23.5-53.9)	35 (21.4-51.5)
Mild	12.5 (5.1-27.5)	7.5 (2.3-21.7)	20 (10.0-36.0)	10 (3.6-24.6)	22.5 (11.7-38.7)	22.5 (11.7-38.7)
Moderate	22.5 (11.7-38.6)	15 (6.6 – 30.4)	15 (6.6 – 30.4)	17.5 (8.2-33.2)	22.5 (11.7-38.7)	10 (3.6-24.6)
Severe	2.5 (0.3-16.9)	12.5 (5.1-27.5)	12.5 (5.1- 27.5)	12.5 (5.1-27.5)	10 (3.6-24.6)	20 (10-35.9)
Profound	10 (0.3-24.6)	15 (6.6 -30.4)	15 (6.6-30.4)	12.5 (5.1-27.5)	7.5 (2.3- 21.7)	12.5 (5.19-27.5)

Comparison of smartphone PTA in office setup at 35 dB ambient noise level and conventional PTA

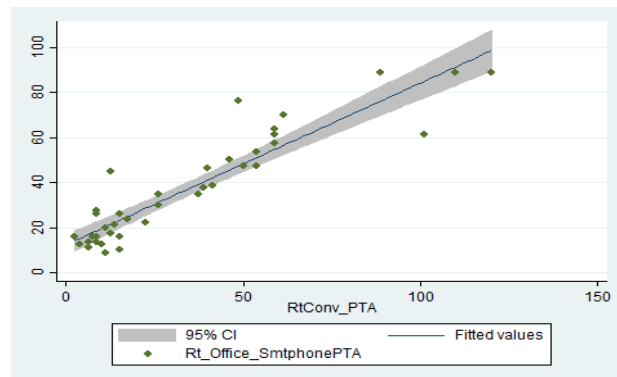


Figure 2: Two way scatter plot with regression fit comparing conventional PTA to clinic smartphone Right ear Regression coefficient is 1.18 (95% CI 1.00-1.34) p-value <0.001

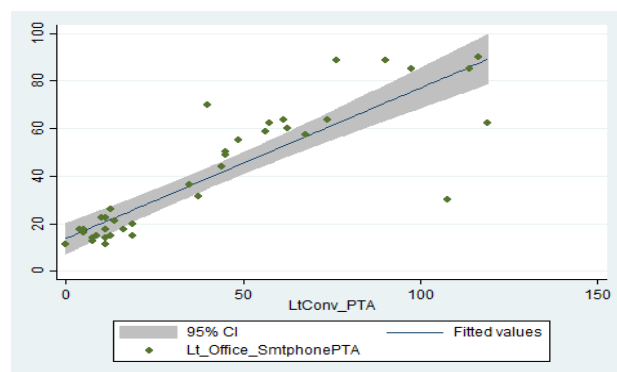


Figure 3: Two way scatter plot with regression fit comparing conventional PTA to office smartphone left ear Regression coefficient is 1.18 (95% CI 0.96-1.41) p-value <0.001

When smart phone based PTA done in an office with ambient noise level of 35dB was compared with conventional PTA, based on the line of regression with a 95% confidence interval there was a strong agreement between conventional PTA and smart phone PTA both in the left and right ear.

Conventional PTA compared with smartphone PTA in clinic set up 45dB ambient noise level

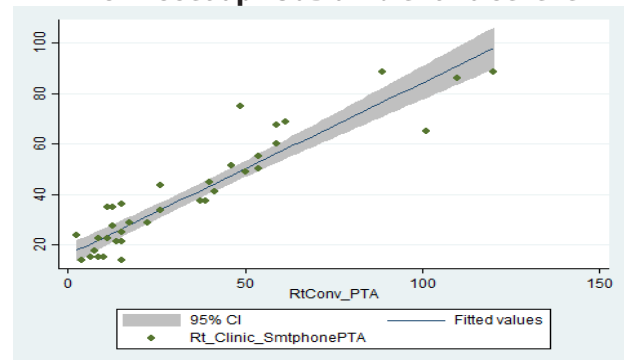


Figure 4: Two way scatter plot with regression fit comparing conventional PTA to clinic smartphone Right ear. Regression coefficient of 0.90 (95% CI 0.81-0.99) p value of <0.01

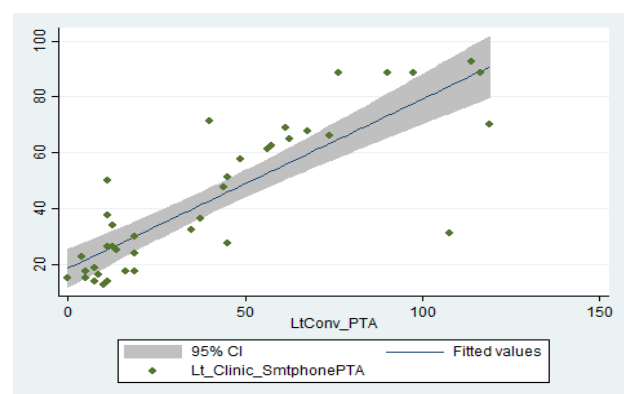


Figure 5: Two way scatter plot comparing conventional PTA to clinic smartphone left ear. Regression coefficient 0.93(95%CI 0.83-1.03) p value <0.01

When smart phone based PTA done in the clinic with ambient noise level of 45dB was compared with conventional PTA, based on the line of regression with a 95% confidence interval there was a strong agreement between conventional PTA and smart phone PTA in both the left and right ears.

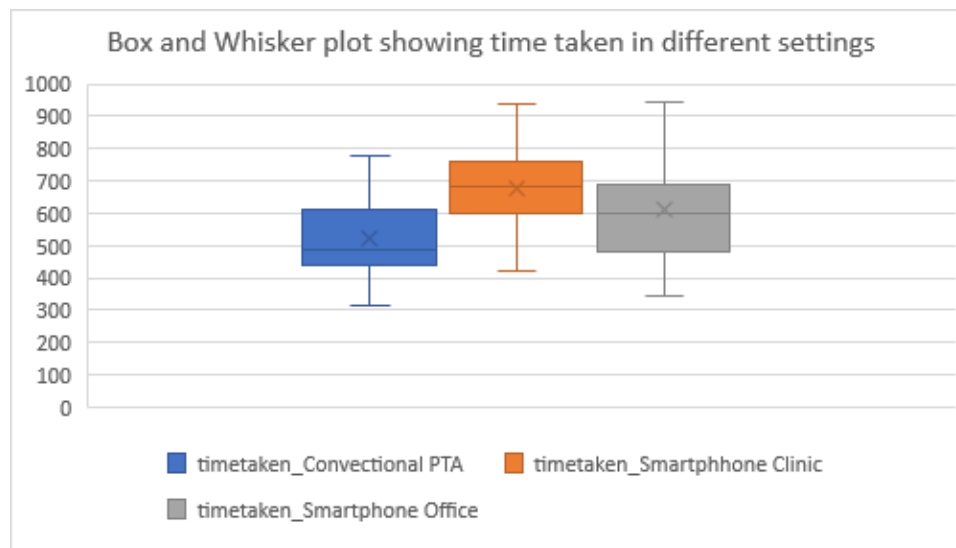


Figure 6: Comparison of time taken to do each of the tests

The mean time taken to complete the conventional PTA test was 522.9 (SD 172.4) sec, 678 (SD 133.9) sec for smartphone clinic test and 609.8 (SD 148.2) sec for smartphone office test. There was a significant difference ($p=0.04$) between time taken to complete the conventional PTA test and the office smartphone test.

In comparing smartphone clinic PTA conventional PTA the $P > 0.01$. The time taken for conventional PTA was significantly less. As for the office smartphone test compared to the clinic test the $P > 0.05$ meaning the clinic time had a significantly greater mean time taken. However there was a significant overlap within the 95% confidence interval therefore ignoring the null hypothesis thus no statistical difference in the time taken to do the tests.

Comparison of preference between smartphone PTA and conventional audiometry

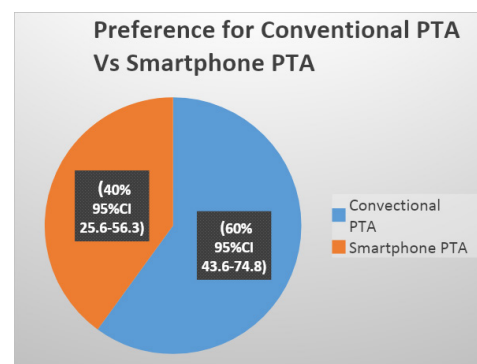


Figure 7: Preference for conventional PTA vs smartphone PTA

Table 2: Correlation between the age and the preference of the mode of PTA

Age (years)	Ease of use of PTA (n=40)		Total
	Conventional PTA	Smart phone PTA	
Young adults (18-35)	2	1	3
Middle Age 36 – 59)	21	14	35
Elderly (>60)	1	1	2
Total	24	16	40

Fishers exact $p=1.00$

Fishers exact test with a p value of 1.00 show no correlation between the age and the preference of the mode of PTA.

Table 3: Correlation between the level of education and the preference of the mode of PTA

Education level	Ease of use		Total
	Conventional PTA	Smart phone PTA	
Primary	5	2	7
Secondary	8	6	14
Tertiary	11	8	19
Total	24	16	40

Fishers exact - P = 1.0

Fishers exact test with a p value of 1.00 shows no correlation between the level of education and the preference of the mode of PTA.

Twenty four (60%) participants preferred conventional PTA while 16 (40%) preferred smartphone PTA.

DISCUSSION

Validating a method of screening for hearing loss requires it to be compared to conventional PTA. Hearing thresholds of 10dB or less between two methods is accepted as subclinical in clinical diagnostic audiometry (OSHA 1983). In spite of this it is important to keep in mind that in children a difference of 10dB can make a major clinical difference and is considered to be significant. This study compared thresholds obtained from hear test the threshold version of hear screen (smartphone based mobile application) carried out in two different setups (quiet office 35dB, normal clinic 45 dB) to the conventional PTA done in a soundproof booth (21dB). There was no statistically significant differences between smartphone and convectional PTA thresholds across all frequencies. Majority of the thresholds obtained via smartphone clinically differed from conventional PTA by 5dB or less except at 8khz which was 10dB and were all within our 95% confidence interval. Other studies done comparing mean threshold difference between conventional audiometry and automated audiometry using the same smartphone application agree with this study Van tonder *et al*²⁰. The results from the study also tallied with the original clinical validity study done on the application by Swanepoel *et al*⁸ where the thresholds obtained from smartphone audiometry and manual audiometry were within a 5 dB difference both in normal ears and diseased ears.

Pure tone average is important as it helps to determine the level of hearing in an ear. Though there was no study found that compared the pure tone average obtained from the different tests, we calculated the means of the pure tone average obtained from the different tests as shown in Table

1 and all were within the 95% confidence interval. This shows that results from pure tone audiometry are comparable to conventional audiometry. In spite of there being no statistical difference, results showed that an increase in the ambient noise led to higher thresholds. One advantage of the smartphone test is its accuracy in areas with high ambient noise levels, it has a real time noise monitoring which automatically leads to repetition of any pure tone presented at ambient levels above 40dB. With this in mind the increase in threshold with the rise in ambient noise levels could be attributed to other factors such as patient distraction or poor concentration and not to the test accuracy.

There was no statistically significant difference in the time taken to do the tests though overall the conventional PTA took a shorter time than the automated audiometry. Generally this could be due to the fact that automated audiometry in the application comes with a standard waiting period between the results and presentation of the next tone in order for it to upload and save the results. Conventional audiometry on the other hand relies on the speed of the response from the participant and the familiarity of the test protocol by the audiologist. The results of this study compared to a study done by Van tonder *et al*²⁰ which showed no significant difference in time though the smartphone test took longer. Contrary to this study, Stephanie *et al*²⁴ showed that smartphones were more efficient and took less time compared to conventional audiometry. The average time taken to do the smartphone test in ambient noise of 35dB was shorter than the one done in 45dB. This could be explained by the fact that the test repeats presentation of any tone presented at ambient noise levels above 40 dB and there is an increase in the number of false responses thus it took much longer to do the test in the normal clinic set up due to the higher ambient noise levels.

Majority of the participants preferred the use of conventional audiometry to the smartphone based

test. This differed from studies done by Yousuf *et al*²² and Stephanie²⁴ where participants preferred smartphone audiometry. This preference was probably due to the fact that the smartphone test was conducted twice and thus took longer time and hence creating a bias. There was a standard protocol of carrying out the study where all the conventional tests were done first. There was no correlation between the preference of mode of test with level of education or age of participants.

CONCLUSIONS

Hear test the threshold version of the hear screen provides hearing thresholds comparable to conventional PTA when done both in a quiet office and in a normal clinic set up. It can thus be used reliably as a screening tool to identify patients with hearing loss in a community level with limited human resources and infrastructure. It can also be efficiently used in follow up and monitoring of threshold changes in patients attending oncology clinics or the TB clinics where they are exposed to ototoxic medication. The application can help in the monitoring of patients with fluctuating hearing loss and those exposed to occupational noise. It provides a solution in the achievement of most of the objectives in the Kenya National Strategy for ear and hearing care 2016 - 2020.

ACKNOWLEDGEMENT

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AUDIOPHONIOLOGICAL RESULTS OF CONVENTIONAL CHILDREN'S HEARING AIDS AT THE INTERNATIONAL CENTER FOR HEARING CORRECTION OF ABIDJAN (CICA)

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ABSTRACT

Objectives: To determine the audiophonological results of the children's hearing aids at the international Center for Hearing Correction of Abidjan (CICA).

Methods: We carried out a prospective analytical study from 25th September, 2019 to 31st December, 2020 at CICA. It included patients aged under 15 years fitted with a conventional CICA hearing aids for sensorineural hearing loss. The parameters studied were epidemiological, audiometric and audiophonological. Data were analyzed using Excel version and Epi info version 7.1 software.

Results: The prevalence of hearing aids was 53.84%. The average age at the time of fitting was 7.4 ± 2.97 years. The sex ratio was 1.05. Deafness was post lingual in 70% of cases. It was of sensorineural hearing loss in 95.65% on the right and in 97.05% on the left. The degree of hearing loss was severe and symmetrical in 34.28%. The hearing aid was bilateral in 95.71%. The APCEI profile had highlighted a level of acceptance of 82.85%, a level of perception of 77.14%, a level of understanding of 74.28%, a level of expression of 80% and an intelligibility level of 78%. The audiophonological result was good in 68% of cases. The factor influencing the audiophonological result was the laterality of the fitting.

Conclusion: The hearing aid has improved the audio phonological capacities in a little more than half of the patients. The factor influencing the audiophonological result was the laterality of the fitting.

Key words: Hearing loss, Conventional hearing aids, Audiophonology

INTRODUCTION

Deafness is the most frequently encountered neurosensory deficit in children, it is declared as a public health problem in several countries. Its prevalence is estimated to be between 1 to 3.4 per thousand at birth and could reach 1 per 100 in the event of severe perinatal pathology¹. Child deafness differs from adult deafness, first, by the aetiologies and then by the fact that it prevents the normal acquisition of speech and language¹. The aetiologies of the child are dominated by congenital causes which will generate profound and sensorineural hearing loss essentially. In the event of a delay in diagnosis and treatment, deafness leads to a delay in speech or language, behavioral disorders and cognitive disorders, with knock-on effects such as learning difficulties at school². Deafness poses a diagnostic and management problem. The management of sensorineural hearing loss is essentially based on

hearing aids or cochlear implants. The most used means in Côte d'Ivoire is the hearing aid with a prevalence of 45.17% according to the study by Foua Bi³ in 2017. However, we do not have any work concerning the audiophonological results of the hearing aid in children. It is with this in mind that we conducted this study to analyze the results of hearing aids for children in our work context. This would improve the indications for hearing aids in children as well as the management of deafness. The objectives were to determine the audiophonological results of the children's hearing aids

MATERIALS AND METHODS

A prospective analytical study was carried out at the International Center for Hearing Correction in Abidjan (CICA) from 25th September, 2019 to 31st December, 2020. Children wearing a conventional hearing aid with minimum fitting period of 6 months

were included. A questionnaire was administered to the patients or their relatives after which the patient underwent tonal and vocal audiometry. An audiophonological evaluation was then carried out. Parameters studied were audiological (The type of hearing loss⁴, the degree of hearing loss⁵) and audiophonological performance using the rating scale: APCEI⁶. Five areas were assessed: A = Acceptance of the device and/or implant; P = Auditory perceptions thus fitted; C = Understanding of the perceived oral message; E = Oral expression, use of voice; I = Intelligibility of the child (quality). Each of these areas was scored between 0 (no performance) and 5 (maximum performance requested in the domain). We defined a good result as a score greater than or equal to 3 for all the different items. The comparison of the proportions was carried out using the Chi square test. When

one of the theoretical numbers of a proportion was less than 5, the Fisher test was then used. For all statistical tests, the significance level (p-value) used was set at 0.05.

RESULTS

Seventy patients were fitted out of 130 patients (prevalence of 53.84%). The average age at the time of fitting was 7.4 ± 2.97 years with a range from 2 - 15 years (Figure 1).

The study population was composed of 36 male and 34 female patients (sex ratio of 1.05). The educational level of the patients was primary in 60.29% of cases, secondary in 32.35% of cases and pre-kindergarten in 7.34% of the cases.

The degree of deafness is as shown in Figure 2. The intelligibility threshold was determined in 40

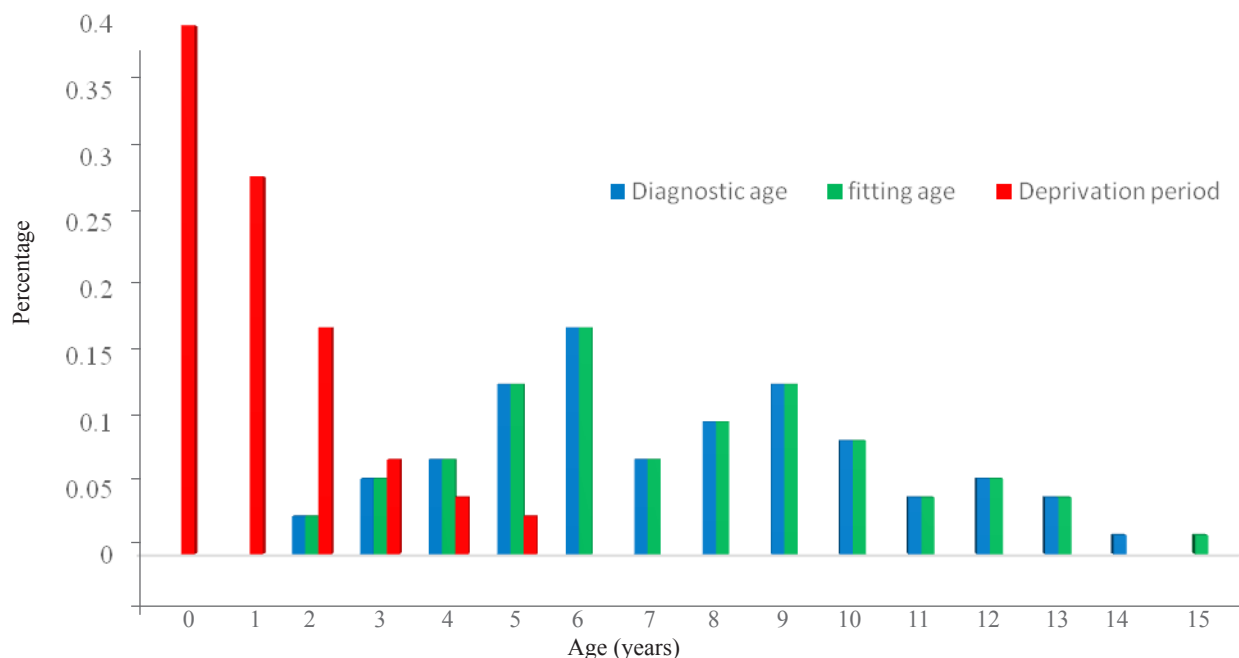


Figure 1: Patients age and management period (n=70)

cases. It was at 120dB in 37.5% of cases. Behind-the-ear (BTE) hearing aids were used in 99% of cases and canal hearing aids in 1% of cases. The fitting was bilateral in 67 cases or 95.57% and unilateral in 3(4.28%) cases. The prescription of hearing aids (unilateral or bilateral side wearing) were respected in 78.57%. The cost of the prostheses varied between 300.64 and 1277.753 USD per unit. The patients' individual APCEI scores are as shown in Figures 3 and 4. The need for all-day use of the hearing aid was noted in 58 cases, i.e. 82.85%

of cases. The perception of the normal voice was raised in 54 cases or 77.14%. A good understanding of simple everyday oral instructions was observed in 52 cases, i.e. 74.28%. The syntax was correct in 47 cases or 67.14%. The understanding was possible for everyone in 24 cases, or 48%. We observed a good APCEI profile in 34 cases or 68% of cases and bad in 16 cases or 32% of cases. There is a statistically significant link between the APCEI profile and the laterality of the hearing aid (Table I).

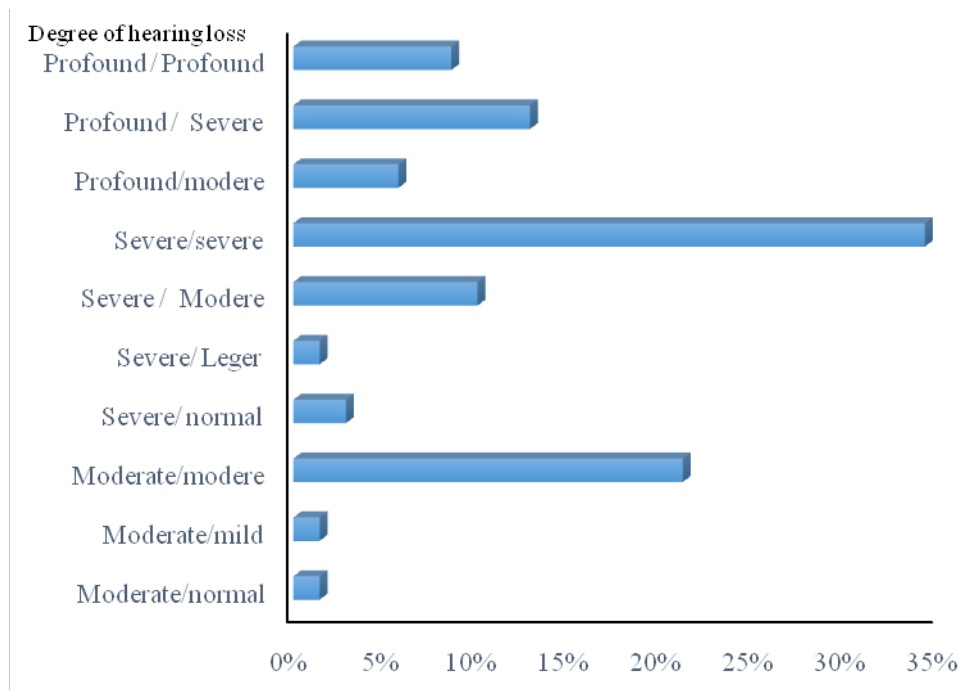


Figure 2: Degree of hearing loss in fitted patients (n=70)

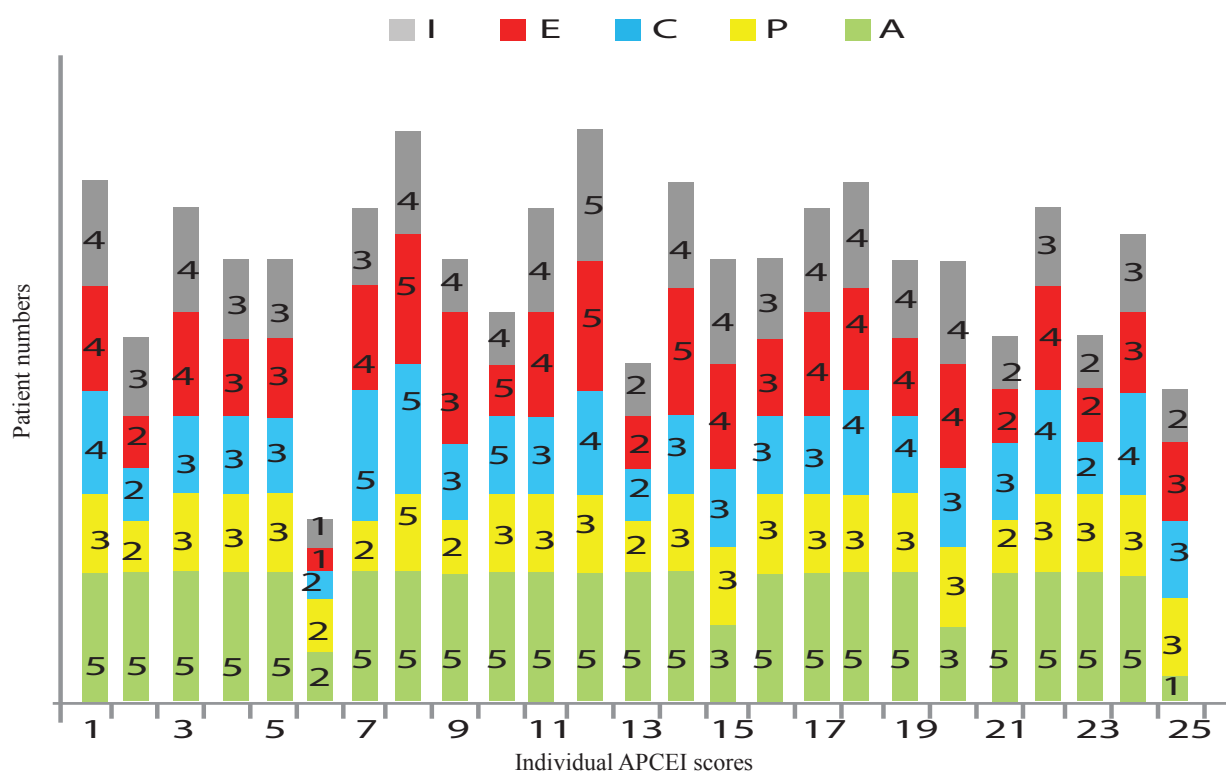


Figure 3: Satisfaction of fitted patients (number 1 to 25).

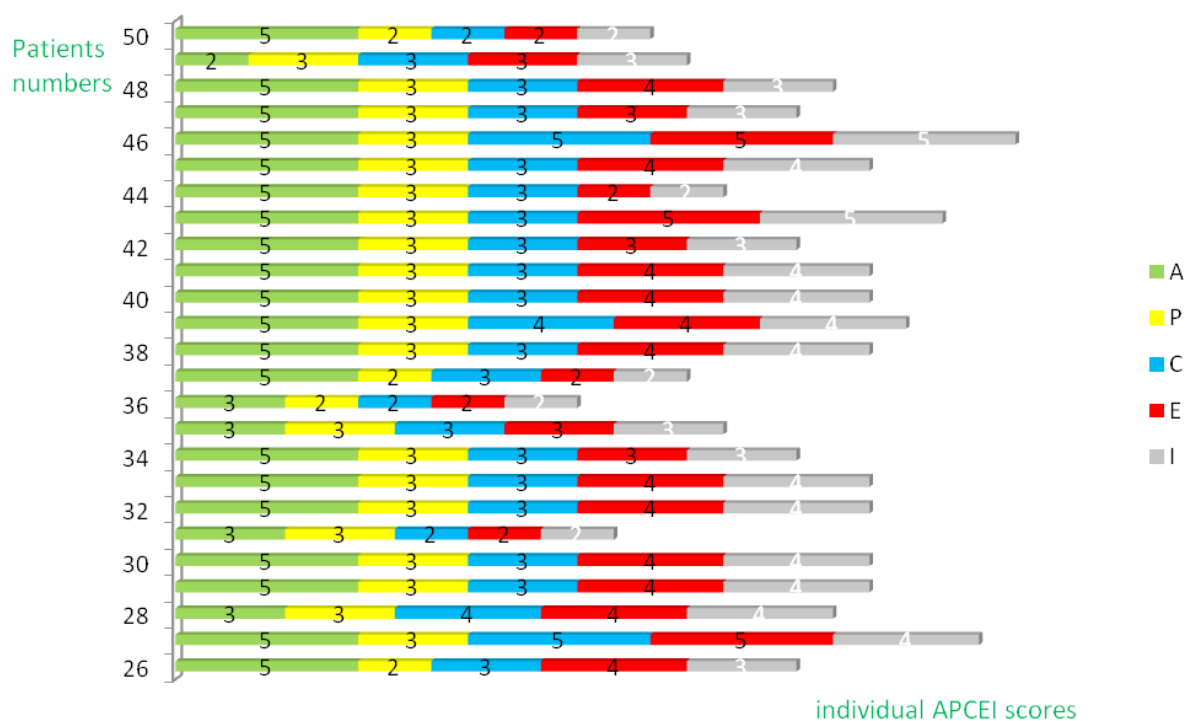


Figure 4: Satisfaction of fitted patients (number 26 to 50)

Table 1: Factors influencing the APCEI profile

		Bad	Good	Total	P
Fitting age	< or = 3	02	04	6	0.12
	> 3	20	44	64	
Gender	Male	15	21	36	2.69
	Female	07	27	34	
Deprivation period	< or =1	17	33	50	0.20
	>1	5	15	20	
Degree of hearing loss	Moderate	08	11	19	0.3696
	Severe	09	28	37	
	Profound	05	09	14	
Bare-ear intelligibility threshold	=< 60 dB	05	14	19	0.96
	> 60 dB	02	19	21	
Laterality of the hearing aid	Unilateral	05	13	18	0.0086
	Bilateral	17	35	52	

DISCUSSION

Wearing the prosthesis was frequent and regular in the vast majority of the cases in our study. This demonstrates the benefits of the hearing aids. Indeed, Souza and Iorio⁷ reported in their study that frequent daily use of the hearing aid was associated with better performance in children fitted. The hearing aid enabled the perception of the human

voice in most patients in our study. This helps to break out of social isolation and participate in a life of communication.

According to the works of Eman Abdel-Fattah Said⁸, awareness of surrounding sound in children with hearing aids was significantly linked to the degree of deafness. This allows communication with the environment. This communication requires an understanding of one's interlocutor. This was

the case for most patients regarding simple daily oral instructions in our study. But only about a third of the patients were able to fully understand the interlocutor with colloquial language in our study. The majority of patients managed to build a sentence. But it was well structured in half of the cases in our study. This could be related to poor lexicon (vocabulary) as illustrated in the study by Penna *et al*⁹ where 65% of children with hearing aids had a lower than normal level of lexicon. Thus, the understanding of the majority of patients was easy when the interlocutor was accustomed to his way of expressing himself. Because these patients have difficulty with phonation as reported in 88.8% of patients in the study by Penna *et al*⁹. For a quarter of the patients in our study they could be understood by all people. It could be the consequence of the alteration of phonation secondary to a difficulty of perception and discrimination of sound resulting from the reduction of hearing acuity in children with deafness. What motivated patients with a significant degree of deafness to favor non-oral communication in his study (sign language)⁹.

The audiophonological results were influenced only by the laterality of the hearing aid fitting in our study. The laterality of the device impacted the audiometric result in our study. This therefore led to a disturbance of the audiophonic loop in the child; when a sound message is delivered: the child picks it up (hears it), decrypts it (understands it) and is able to respond orally if necessary (oral production) while being understandable (intelligibility). This loop therefore requires Auditory Perception (P), which depends on the state of the peripheral auditory pathways⁶. The small sample size may explain this sole association

CONCLUSIONS

The hearing aid has improved the audiophonological capacities in a little more than half of the patients. The factor influencing the audiophonological result was the laterality of the fitting. The hearing aids fitting must be supported by the state.

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ENT PRACTICE IN KENYA; ASSESSMENT OF ACCESS TO MICROSCOPES AND ENDOSCOPY UNITS

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ABSTRACT

Background: The scope of Ear Nose and Throat (ENT) practice entails the use of endoscopy units and microscopes and their related surgical sets. There is no current data on their availability to ENT surgeons practising in the public sector in Kenya.

Objective: The primary objective of this study was to assess the availability of endoscopy units, microscopes and related surgical sets for endoscopic and microscopic surgery to ENT surgeons in the public sector in Kenya. Study design: This was a prospective cross-sectional study carried out between the months of February and June 2022.

Methods: A survey was distributed among ENT surgeons working in the public sector through the Kenya Ear Nose and Throat Society (KENTS) members' WhatsApp group. Data on availability of endoscopy units, microscopes and related surgical sets, referrals of procedures/ conditions that the ENT surgeons can perform if they had the equipment and the availability of anaesthesia providers and other surgeons who would benefit from the same equipment was collected.

Results: Majority of ENT surgeons did not have access to microscopes and endoscopy units (52% and 64% respectively). Majority also did not have the related surgical sets necessary to perform the surgeries and would have to borrow the equipment. The majority referred otology (78%) and micro laryngeal surgeries (68%) "most of the time" or "always" (which they were capable of performing) due to lack of the same equipment.

Conclusions: The scope of ENT practice is limited in the country due to lack of endoscopy units and microscopes with their accompanying surgical sets. Investing in the availability of the equipment will improve the level of ENT care provided and hence ease the burden to patients and referral institutions.

Key words: Ear Nose Throat (ENT), Operating microbes, Endoscopic unit

INTRODUCTION

The number of Ear Nose and Throat (ENT) surgeons in Kenya has steadily risen over the years and is currently estimated to be 113, serving a population of 47.5 million¹. The scope of ENT practice ranges from open surgery to endoscopic surgery including endoscopic sinus surgery, endoscopic ear surgery and endoscopic laryngeal surgery. Laryngeal and ear surgery also entails the use of operative microscopes.

Kenya has a high burden of ENT diseases that require the use of microscopes and endoscopy units to manage. The prevalence of chronic otitis media among children aged 2 – 15 years is estimated at 15 per 1,000². These are children who potentially will require ear surgery with the use of a microscope

for definitive treatment. Children with recurrent acute otitis media and persistent Otitis Media with Effusion (OME) with hearing loss also require ventilation tube insertion to prevent the sequelae of Chronic Otitis Media (COM) and delayed speech respectively; a procedure that requires either the use of a microscope or an endoscope. Furthermore, COM and OME lead to hearing loss; which has been identified by WHO as the 4th leading cause of disability in the world³. Access to microscopes and endoscopes may reduce this burden.

Sinonasal surgery has evolved from the use of headlights and open techniques to the use of endoscopes. This has enabled advancement in minimally invasive techniques with shorter hospital stay and more cosmetically appealing results.

Suspected nasal and nasopharyngeal masses can quickly be assessed and biopsies taken. Definitive surgery for sinonasal pathologies including sinonasal squamous cell carcinomas may be performed endoscopically with the same results as open techniques but with less morbidity⁴.

Availability of endoscopes and endoscopic units also facilitates rapid assessment of suspected laryngeal pathology. Intervention for visualised laryngeal lesions e.g. Polyps, nodules, papillomas requires the use of an operating microscope or endoscopes to safely remove the pathology.

Certain microscopes and endoscopy units can also be multipurpose with use across other surgical disciplines. For example, urology requires endoscopic units to perform cystoscopies, transurethral resection of bladder tumours, transurethral resection of the prostate among other procedures. General surgeons perform laparoscopic procedures with the use of endoscopy units. They can also perform oesophagogastroduodenoscopies and colonoscopies with the same units. Certain multi-use operative microscopes can be used by ophthalmologists, plastic surgeons and neurosurgeons to magnify small structures to perform fine surgery. Availability of multi-use endoscopy units and microscopes will thus benefit patients across the multiple surgical specialities.

Whereas availability of these equipment would help address the burden of ENT diseases in the country, there is no data on their availability to ENT surgeons in Kenya. The aim of this study was to assess the availability of endoscopy units, microscopes and related surgical sets for endoscopic and microscopic surgery to ENT surgeons in Kenya.

MATERIALS AND METHODS

A survey was carried out among ENT surgeons practising in the public sector in Kenya after ethical approval from Kenyatta National Hospital-University of Nairobi's ethics committee. The survey was a google forms online questionnaire. Survey questions included county of practice, years of practice, availability of endoscopy units and microscopes and related surgical sets, referrals of procedures/ conditions that they can perform if they had the equipment and the availability of anaesthesia providers and other surgeons who would benefit from the same equipment.

The survey was distributed through the Kenya Ear Nose and Throat Society (KENTS) ENT consultants WhatsApp group, a group with 92 consultants practising in the country. The group has ENT surgeons in both private and public practice. Inclusion criteria was ENT surgeons practising in a public facility. Reminder messages to fill the questionnaires were sent on the 2nd and 4th week after the first message was sent. Results from the survey were collated and analysed using google forms and Microsoft excel.

RESULTS

There were 25 responses from ENT surgeons practising in the public sector from various counties. The average duration of practice was 7.1 years with a range of 1-22 years. Most of the respondents (14/25) practised in level 5 facilities (Figure 1).

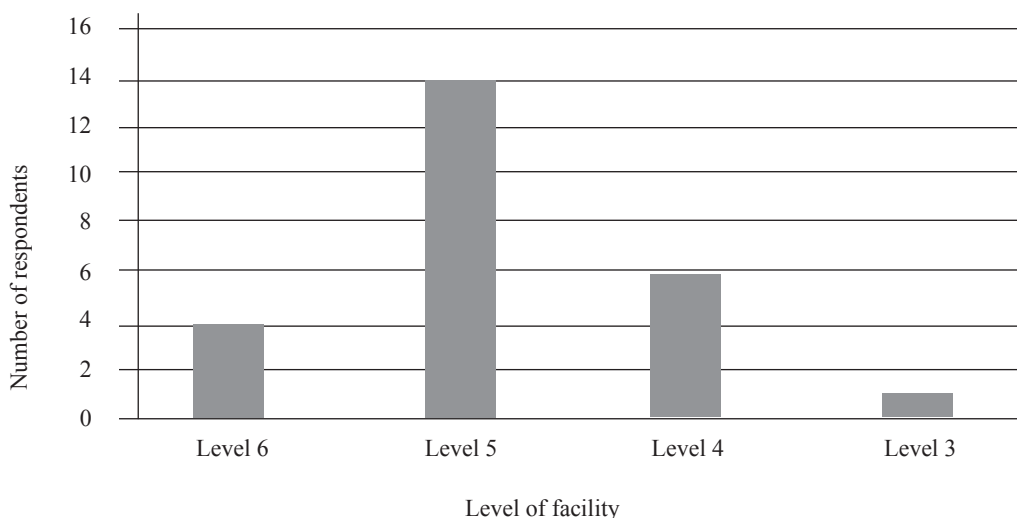


Figure 1: Respondents per facility level

Majority of the practitioners had no access to microscopes or endoscopy units. 52% and 64% respectively (Figure 2).

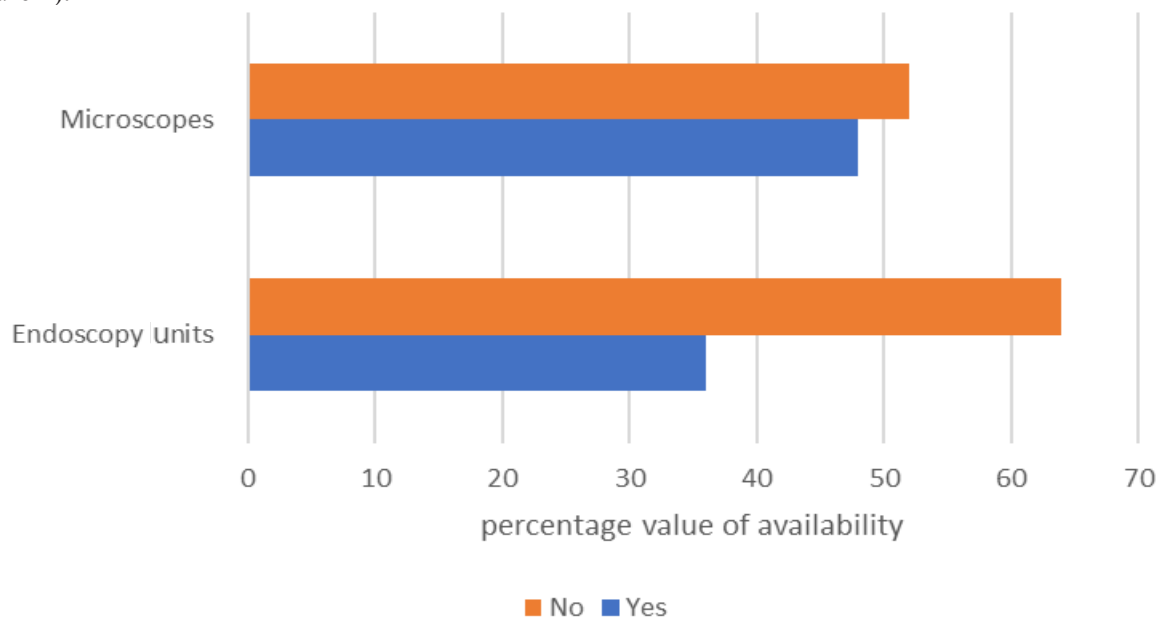


Figure 2: Access to microscopes and endoscopy units, with the sets

Majority of the practitioners also did not have the necessary surgical sets to perform microlaryngeal, otological or sinus surgery (Figure 3).

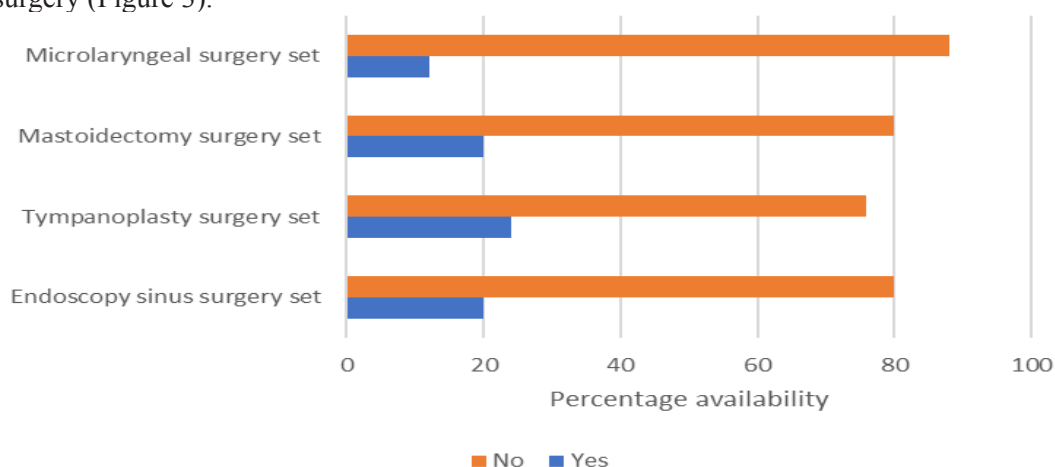


Figure 3: Availability of surgical sets.

Otology and microlaryngeal surgeries were “most of the time” to “always” referred out due to lack of equipment. Seventy two percent and 68% respectively. Sixty percent of ENT surgeons would

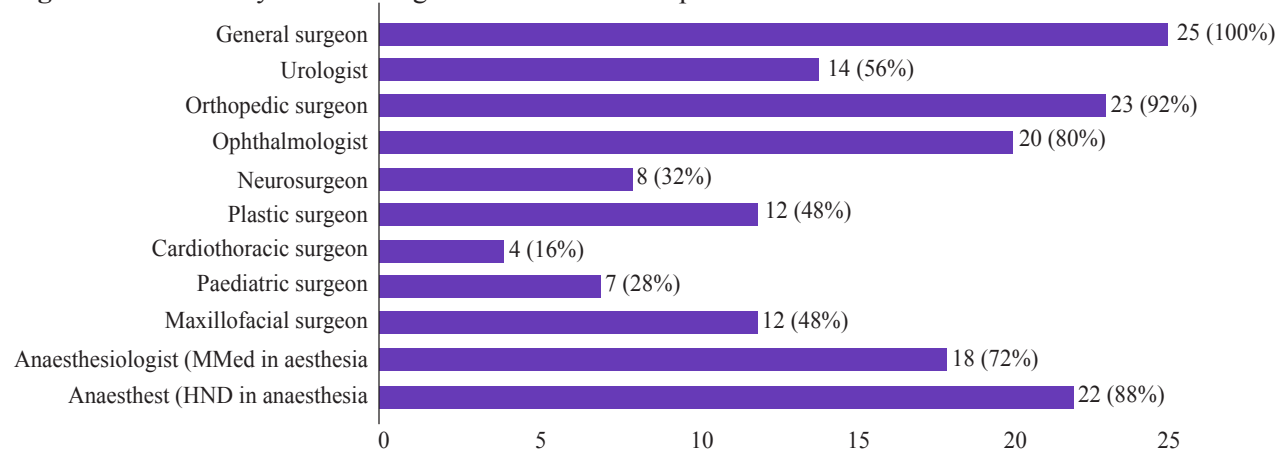
also borrow surgical instruments to perform ear, sinonasal or laryngology surgery within the public facility (Table 1).

Table 1: Referrals of surgeries out of the facility due to lack of equipment and frequency of borrowing of equipment

Referral of cases that once can perform (%)	Always	Most of the time	Sometimes	Never
Otology	28	44	12	16
Microlaryngeal surgery	48	20	16	16
Frequency of borrowing equipment (%)	16	44	20	20

Availability of other surgeons and anaesthesia providers is highlighted in Figure 4.

Figure 4: Availability of other surgeons and anaesthesia providers



DISCUSSION

This is the first study evaluating access to operating microscopes and endoscopy units to ENT surgeons in the public sector in Kenya. The findings show a large deficit of the equipment and relevant surgical instruments. This would in turn mean that a significant part of the country's population lacks access to adequate treatment of ENT diseases due to the lack of equipment. Furthermore, given the scope of practice of ENT surgeons, we note that ENT surgeons are underutilised with the lack of equipment in the public sector.

The majority of the ENT surgeons surveyed worked in level 5 facilities and they would refer ENT cases that they can perform at their facility due to lack of equipment. This applies pressure to the few level 6 facilities that have the equipment. This leads to delays in treatment of patients and a diversion of resources in the level 6 hospitals to treat diseases that can be readily addressed at the peripheral facilities. It also has an economic implication on the patients and their families who have to bear transport and accommodation costs in addition to more expensive medical costs to attend referral facilities far away from their homes.

Given the lack of equipment, more than half of the ENT surgeons surveyed have resulted to borrowing equipment from other institutions. This however is not sustainable without a formal structure. Creative ways to address the lack of equipment is needed. Surgical training camps like Operation Ear Drop (OED) have helped to bridge the gap of lack of ear surgery at peripheral facilities due to lack of

microscopes⁵. Donations of equipment by partners in high resource centres can also assist bridge the gap of the equipment needed. Such negotiated donations however have been reported to attract large import fees once they arrive in the country with the equipment stalling in storage facilities further incurring storage charges. Lobbying of the government institutions involved during importation with formulation of a framework to subsidise or grant waiver of the import fees would make the donated equipment readily available.

Leasing of the microscopes and endoscopy units may be another alternative. In 2015, the Government of Kenya established leasing of medical equipment through the MES program⁶. This program has made imaging and dialysis equipment available in the peripheral facilities. Inclusion of microscopes and endoscopy units to this list might make them more available. Given the unavailability of these equipment, specialists in the facilities who are likely to benefit from the use of similar microscopes and endoscopy units should interact and lobby for the purchase of equipment that can be used across their specialties.

CONCLUSIONS

The scope of ENT practice is limited in Kenya by the lack of endoscopy units and microscopes with their accompanying surgical sets. Investing in the availability of the equipment will improve the level of ENT care provided and hence ease the burden to patients and referral institutions.

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LARYNGOPHARYNGECTOMY WITH PECTORALIS MAJOR MYOCUTANEOUS FLAP RECONSTRUCTION WITHOUT USE OF A SALIVARY BYPASS TUBE FOR HYPOPHARYNGEAL CANCERS: REPORT OF TWO CASES

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ABSTRACT

Hypopharyngeal cancer is a subtype of head and neck tumour that carries a grave prognosis. Most of these tumours are diagnosed late largely due to nonspecific symptoms that are initially ignored. The best outcomes are achieved by a combination of surgery and chemoradiation. Surgical resections result in large defects that are a challenge during reconstruction. Various options for reconstruction are available depending on the size and location of the defect. This is a report of two cases managed with surgery and postoperative chemoradiation giving details on the surgical steps for total laryngopharyngectomy using the pectoralis major flap for reconstruction without use of salivary bypass tube.

Key words: Laryngopharyngectomy, Pectoralis, Salivary bypass tube, Hypopharyngeal cancers

INTRODUCTION

Hypopharyngeal cancer is the most aggressive of head and neck cancers because of anatomic and tumour physiology factors that are peculiar of tumours in this subsite. The hypopharynx having a large space allows for tumour to expand to great dimensions before obstructive symptoms arise. Most patients thus present with an advanced T stage and are also likely to present with nodal metastasis due to the rich lymphatic network in this anatomical region. These kinds of tumours also have a predilection for extensive submucosal spread especially inferiorly¹. Due to the factors above, surgical resection is likely to be extensive in most patients if oncologic margins are to be achieved. This poses a challenge in reconstruction due to the size of the defect as attempts to restore the aero digestive tract are made. Several options are available for such reconstruction each with its own merits and demerits. These cases describe a technique using a pectoralis major flap in reconstruction in hypopharyngeal surgery without use of a salivary bypass tube.

CASE REPORT 1

A 42-year old male was seen as a referral from a peripheral facility with a 3 month history of sore throat, painful swallowing and hoarseness. Direct laryngoscopy revealed a left side piriform tumour involving both the medial and lateral walls. The larynx was free of invasion by tumour. There

were no tumours in the esophagus or stomach at esophagogastroduodenoscopy. CT scan of the neck revealed a left piriform mass with erosion of the thyroid cartilage. There were no evidence of enlarged cervical nodes or chest metastasis on a chest CT scan. Biopsy taken was reported as invasive squamous cell carcinoma.

Total laryngopharyngectomy, total thyroidectomy and bilateral level II to VI selective neck dissection was done. Reconstruction was by use of the pectoralis major myocutaneous flap. The resultant chest defect was closed primarily after insertion of vacuum drains which were removed in 4 days. The patient developed a pharyngocutaneous fistula that closed spontaneously in 44 days.

He however presented with odynophagia shortly after discharge. Oral examination revealed a right side tongue base ulcer that was confirmed to be squamous cell carcinoma on biopsy. Partial glossectomy and level I neck dissection was done 40 days after initial discharge. Follow up PET scan done three months after completion of chemoradiation had no evidence of local or systemic disease.

Laryngopharyngeal specimen pathology

Histology of the primary tumour was Basaloid squamous cell carcinoma. There was no lymphovascular or perineural invasion. Two of the 24 lymph nodes had metastasis without extracapsular spread. Margins were free of tumour with the closest margin being 3mm.

Tongue specimen pathology

Invasive moderately differentiated squamous cell carcinoma. Margins free of tumour with the deep margin were less than 1mm. There were seven nodes harvested in the neck dissection all of which were negative of metastasis.

CASE REPORT 2

A 44-year old female presented with a 3-year history of sporadic painful swallowing and foreign body sensation which had been treated as pharyngitis. Progressive dysphagia was reported with the patient being able to take semisolids at the time of presentation. There was in addition loss of 12 kg in weight over the preceding 6 months. There was no history of alcohol or tobacco use. There was family history of gastric cancer which had claimed the life of her mother. She had suffered a stroke and pulmonary tuberculosis from which she had fully recovered. Direct laryngoscopy revealed a postricoid tumour biopsies of which were reported as invasive squamous cell carcinoma.

Total laryngopharyngectomy, left hemithyroidectomy and bilateral level II to VI selective neck dissection was performed. Reconstruction was done by use of pectoralis major myocutaneous flap. The chest defect was closed primarily over a vacuum drain. There was no evidence of pharyngocutaneous fistula or any other complication in this patient. Discharge was 30 days post-surgery with the patient able to solid feed orally.

Pathology

Moderately differentiated squamous cell carcinoma with all margins being clear. Nearest margin was 3mm. There was lymphovascular invasion present. No perineural invasion was evident. One of the 42 nodes had metastasis with no extracapsular spread.

Description of the surgical procedure

The patient was positioned supine with the neck extended and prepped from the mandible to the lower rib cage. An endotracheal tube replaced the tracheostomy tube in one of the patients and was secured using Nylon 2.0 sutures to the chest away from the operation side. The procedure started with raising an inferiorly based flap in a subplatysmal plane leaving the submandibular skin intact. The superior edge of the flap was approximately the level of the hyoid bone. Bilateral neck dissection was then performed leaving the fibrofatty tissue

attached to the larynx anteriorly. The sternal heads of the sternocleidomastoid were then transacted at their attachments to the sternum. Dissection was then carried out medial to the carotid arteries on both sides to reach the prevertebral space. Blunt dissection with a finger was then used to elevate the pharyngolarynx off the prevertebral fascia. Clinical assesment for prevertebral invasion was done at this point. Attention was then turned to the suprahyoid region where cuts were made superior to the hyoid to enter the vallecula. Perpendicular cuts were continued towards the posterior pharyngeal wall which was then transected to amputate the laryngopharynx superiorly.

A stomal opening was then made on the inferiorly based flap the lower margin being about 2cm above the suprasternal border. The circumference of the stoma was roughly the same as that of the trachea. The anterior half of the trachea was then incised between the cartilages guided superiorly by the site of the tracheal stoma.

Care was made to ensure that a good enough tracheal stump was available so that it's attachment to the skin was not under tension. The anterior portion of the tracheal stump was then sutured to the inferior portion of the skin stoma ensuring that the edge of the skin aligned to cover the raw area of the tracheal stump. Two other sutures of the same nature were made on either side of the first. These served to prevent tracheal retraction once the circumferential cuts were made. A new endotracheal tube replaced the previous one through which anaesthesia was given henceforth. Circumferential cuts were made to amputate the trachea. The esophagus was then clumped with two non- crushing intestinal clamps and transected in between to allow delivery of the surgical specimen. A nasogastric tube was then passed through the nose into the stomach through the esophageal introitus after release of the intestinal clamp.



Figure 1: Skin surface markings

Inferiorly based flap with the submental flap marked. The size is dictated by the pinch test that assesses amount that can be harvested and allow for primary closure of the resultant defect. Blue arrow

indicates the site of the hyoid which is the superior edge of the flap. Neck dissection performed with fibrofatty tissue attached to the laryngopharynx that is retracted using the Allis forceps marked by the blue arrow. The Phrenic nerve [P] with the Scalenes [S] below it and internal jugular vein [I] exposed after neck dissection (Figure 2).

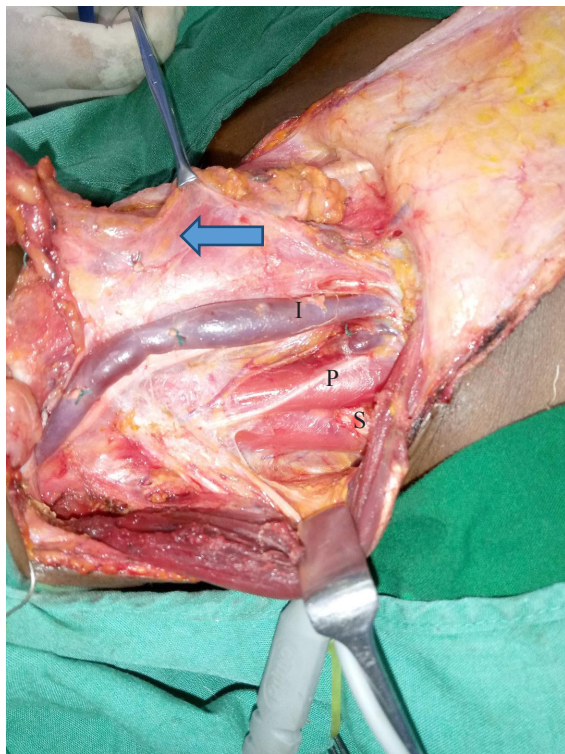


Figure 2: Surgical site after neck dissection

Entry in the retropharyngeal space [black curved arrow] by dissecting bluntly medial to the carotid artery pointed by the blue arrow (Figure 3).

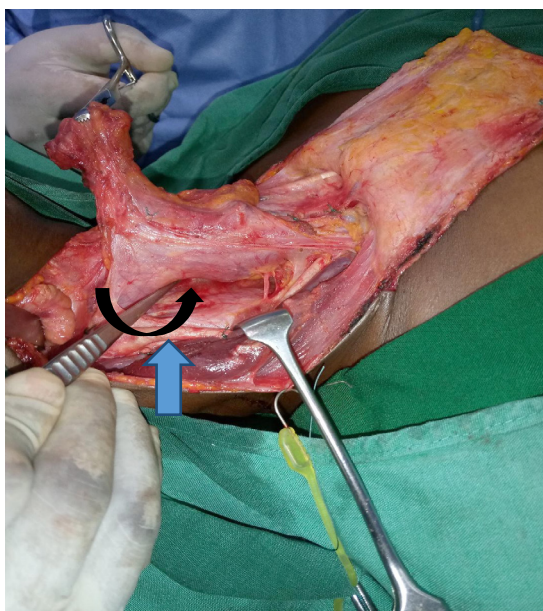


Figure 3: Entry into the retropharyngeal space

Laryngopharynx [L] transected superiorly at the suprahyoid region and deflected inferiorly to expose the prevertebral fascia [P] (Figure 4).

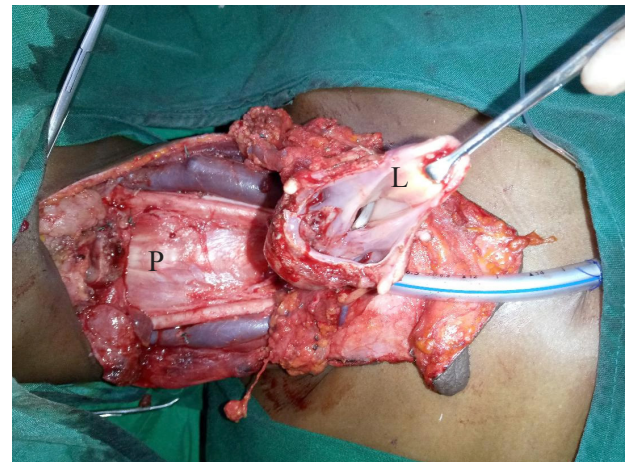


Figure 4: Superior transection of the laryngopharyngeal specimen.

Non-crushing intestinal applied on the cervical esophagus [E] prior to transection (Figure 5).



Figure 5: Clamping of the esophagus

The tracheal stump [T] with the endotracheal tube positioned inferiorly over the subplatysmal flap. The clumped cervical esophagus [E] and posterior pharyngeal wall [P] well exposed (Figure 6).

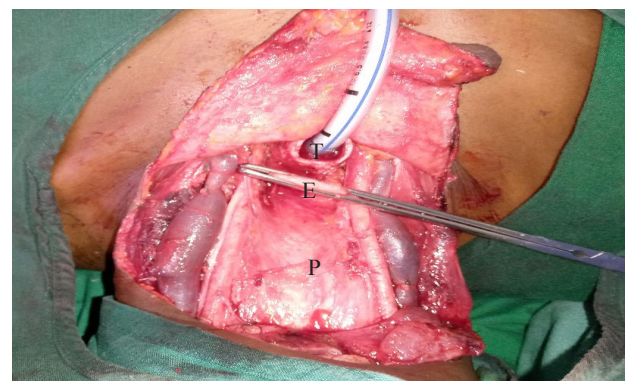


Figure 6: Surgical site after resection of laryngopharynx

A pectoralis major flap was harvested on the non-dominant hand side taking care that enough skin was available for reconstruction. The point of rotation of the pectoralis major was about the midclavicular point from which the lowermost point of the flap was estimated to be equidistant with the most superior part of the defect to be reconstructed namely the tongue base. Estimation of the adequate size was then done from assessment of the length of the defect which informed the height of the flap while the width was dictated by the pinch test which allowed primary closure of the chest defect after harvesting the flap. Surface marking of the pectoral branch of the thoracoacromion artery which supplies the flap was mapped by intersection of a line drawn from the acromion to xiphoid and another perpendicular one from the mid clavicle. The skin should lie above the pectoralis major muscle along the line described above. The sternocostal joints formed the medial border of the flap. Once fully mapped out, incision of the skin around the paddle was done and deepened to reach the pectoralis major muscle fibres. The skin was then undermined to the clavicle, anterior axillary line and sternocostal region. The lateral border of the pectoralis major muscle was then identified and elevated off the pectoralis minor using blunt finger dissection done. The muscle fibers were then incised at the lower and medial edges of the skin paddle extending superiorly to the clavicle cauterizing the perforating branches of the internal mammary artery. This allowed the muscle to be elevated superiorly to expose the vascular pedicle. The humeral attachment of the Pectoralis major muscle was then released. Part of the clavicular attachment was also released with the vascular pedicle in direct view to avoid accidental transection. Once fully mobilized, the myocutaneous flap was tunneled into the neck for reconstruction taking care not to kink the pedicle. The tunnel was a minimum of four finger breaths to avoid strangulation of the vascular pedicle.

Reconstruction of the neopharynx started by suturing the posterior esophageal wall and superior posterior oropharyngeal mucosa to the prevertebral fascia. The skin of the fasciocutaneous flap was then sutured to the prevertebral fascia starting in the superior region adjacent to the lateral part where the oropharyngeal mucosa was sutured to the prevertebral fascia to the esophageal repair inferiorly. This portion of repair was medial to common carotid and internal jugular vein. The skin was then rolled over in an inverted U as the suture proceeds over the anterior oropharyngeal mucosa and esophagus. The last portion of repair was on the prevertebral fascia on the opposite side completing the repair of the neopharynx. Completion of the tracheal stoma

was then carried out before closure of the neck flap over a vacuum drain. The chest defect was likewise closed primarily over a drain.

The posterior esophagus and posterior mucosa of the oropharynx is stitched to the prevetebral fascia marked with the big and small arrows respectively. The Pectoralis Major Myocutaneous [PMF] flap ready for reconstruction (Figure 7).

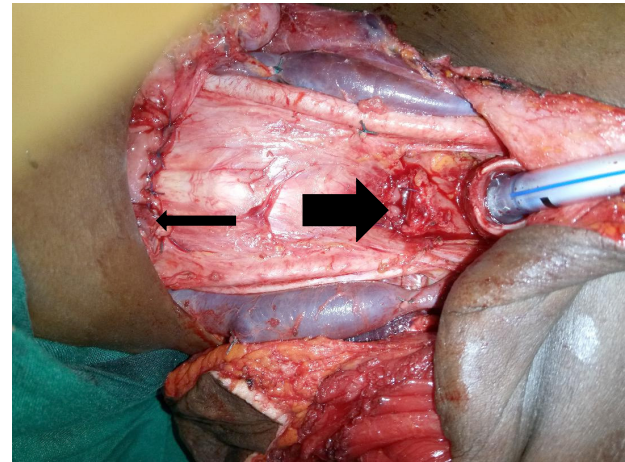


Figure 7: Reconstruction of the neopharynx

Suturing of the pectoralis major flap skin to the prevertebral fascia medial to the Carotid and Internal jugular vein from the oropharyngeal mucosa superiorly to the esophageal region inferiorly marked by blue arrow (Figure 8).

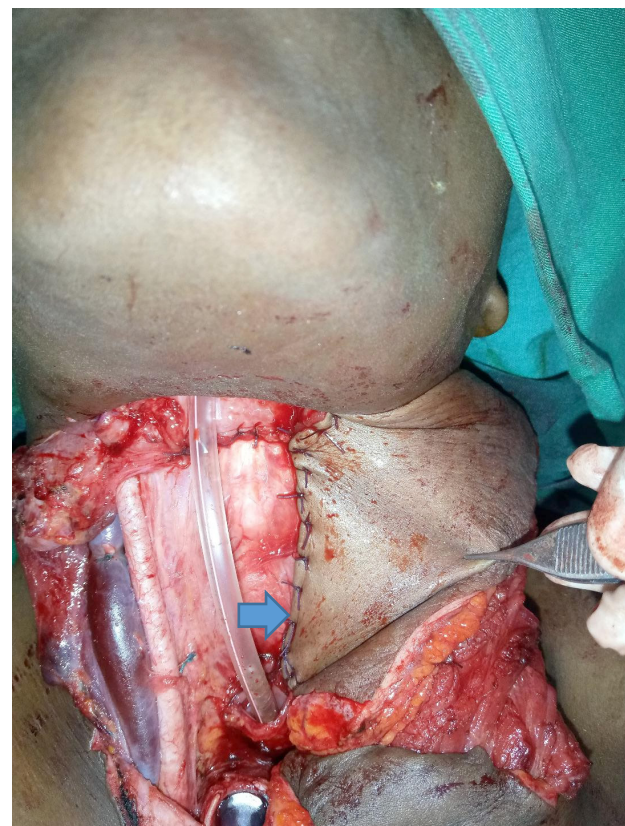


Figure 8: Suturing the PMF to the prevertebral fascia

Pectoralis Major Flap [PMF] being sutured on the anterior oropharyngeal mucosa and esophagus effectively forming an inverted U with the prevertebral fascia [P] forming the posterior part of the neopharynx (Figure 9).

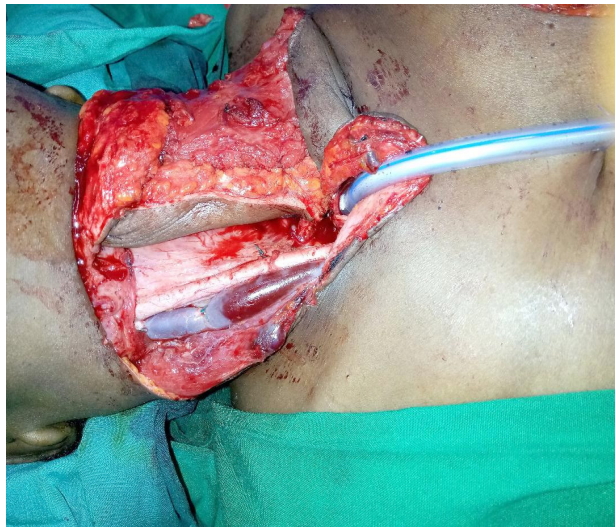


Figure 9: Fashioning the neopharynx

DISCUSSION

Hypopharyngeal cancer is one of the most aggressive of head and neck cancers carrying a poor prognosis. Most patients with hypopharyngeal cancer present in the 6th decade at an advanced stage. Late presentation is largely because the initial symptoms are non-specific and may therefore be ignored or misdiagnosed for other conditions that may present in a similar manner. The hypopharynx is anatomically spacious allowing for tumour expansion to large dimensions before obstructive symptoms develop. Hypopharyngeal carcinomas unlike other head and neck cancers have a propensity for submucosal spread especially in the inferior direction. This kind of tumour extension has been found to be more in irradiated tumours¹. Surgical oncologic resections therefore leave extensive defects that pose challenges during reconstructions. Salvage surgery is more challenging as the margins are less well defined, resections are wider due to more extensive submucosal extension and healing is poor with a higher incidence of pharyngocutaneous fistula.

Due to the abundance of lymphatics in this anatomical region, there is a high propensity of nodal metastasis. Tumours of the hypopharynx will have bilateral nodal spread except those that are well lateralized in the piriform fossa². Studies have shown that tumours of the hypopharynx will

rarely metastasize to level 1 making dissection unnecessary in this level unless there are palpable nodes or evidence of involvement in imaging studies. This specific pattern of drainage allows for use of the submental flap in reconstruction based on submental branch of the facial artery. The pedicle however allows for limited arc of rotation making it appropriate for reconstruction of lateral defects of the hypopharynx after partial pharyngectomy and or partial laryngopharyngectomy where the defect is not circumferential³.

Various options are available for reconstruction of circumferential defects after hypopharyngeal carcinoma resection. These include free jejunal flaps, gastric pull through, tubed anterolateral thigh flaps and radial forearm flaps. The pectoralis major flap has been used for reconstruction after total laryngopharyngectomy as described by Spriano *et al*⁴. In his technique, the pectoralis major flap was used in a U-fashion in reconstruction of total laryngopharyngeal surgery. A salivary by-pass tube is used to reduce the incidence of pharyngocutaneous fistula⁵. The salivary bypass tube is used mostly in cases of total laryngopharyngectomy and salvage laryngectomy where a general trend of reducing the incidence of PCF has been observed though most studies have not found statistical significance in studies. No salivary by-pass tube was used in both patients presented in this study. One patient presented with a PCF that closed spontaneously in 5 weeks while the other one didn't present with one. The salivary bypass tube is not universally used by most surgeons and unavailability of one should not be a cause for concern in total laryngopharyngectomy.

The pectoralis major flap is relatively easy to raise and tunnel to the neck. It brings with it a large amount of skin and muscle. The skin is used as the replacement of the anterolateral mucosa of the neopharynx while the prevertebral fascial forms the posterior part. The muscle is important as it offers a cover for the major neck vessels that are exposed during dissection. This offers protection to the vessels in the setting of a pharyngocutaneous fistula and prevents carotid blow out. Use of the PMF has been described in salvage surgery for laryngeal cancer to reduce chances of pharyngocutaneous fistula formation. This flap may be used for reconstruction for partial pharyngectomy. Using a tubed PMF may result in dysphagia due to the bulk of the muscle with a narrow lumen of the neopharynx and stenosis at the anastomotic sites⁶. Most of hypopharyngeal cancer patients present at an advanced stage with

most not being in the best of performance status. Use of the PMF allows for reduction of surgical time for patients who may be frail. Use of a free flap in these patients requires additional time of operation carrying with it increased anaesthetic risk. The chest wall defect is closed primarily negating the need of a skin graft that would result in an added wound for the patient. The patients did well post-surgery excellent swallowing being reported in both patients. The patients did well despite not using a salivary bypass tube.

CONCLUSIONS

The PMF is a relatively easy flap to raise and use for reconstruction in reconstruction of total laryngopharyngeal defects. It offers a good option as it restores the aerodigestive tract in a short time with minimal donor site morbidity at the donor site as primary closure is possible despite the large amount of skin that can be harvested.

Conflict of interest: There was no conflict of interest in the study.

Informed consent: Written informed consent was acquired from the patients prior to commencement of the study.

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COVID SWAB AS A FOREIGN BODY IN THE RIGHT MAIN BRONCHUS: CASE REPORT

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ABSTRACT

Performing a mucosal swab for SARS-CoV-2 in a patient with a tracheostomy can be hazardous. There are no guidelines on the subject matter. A case report of a Covid-19 swab stick retained as a foreign body in the right main bronchus is presented.

Key words: Coronavirus, HDU, SARS-CoV-2, Covid-19, Covid-19 swab, Tracheostomy, Bronchoscopy, Foreign body, Airway

INTRODUCTION

Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) is the virus responsible for Coronavirus disease 2019 (Covid-19)¹. The main mode of spread is direct person-to-person transmission through close range contact via respiratory particles. It is mainly a respiratory disease; however, it can affect any part of the body. The recommended test is performed via a mucosal swab of the nasopharynx or oropharynx^{2,3}. The spectrum of disease ranges from asymptomatic to critical, including mortality. Although the greater majority are mild cases, the elderly or those with pre-existing conditions are at risk of severe disease. Personal preventive measures to reduce the risk of transmission like mask wearing, social distancing and diligent hand washing are important. Guidelines highlighting safety precautions during aerosol generating procedures and conditions are available⁴⁻⁷; however, there is paucity in the guidelines when it comes to testing laryngectomy and tracheostomy patients. A case report that highlights the perils of taking a Covid-19 swab through the tracheostomy is presented.

CASE REPORT

A 76-year-old man with hypopharyngeal cancer and a tracheostomy due to upper airway obstruction was in the high dependency unit awaiting insertion of a gastrostomy tube for enteral feeding. Prior to the surgery, a swab was to be taken to test for SARS-CoV-2 as per hospital protocol prior to any elective surgery. An oropharyngeal swab was used through the tracheostomy tube to take a tracheal specimen.

During the process, the medical officer noted the distal end of the swab stick had fallen into the trachea. The patient coughed but the vital signs remained stable with no change in oxygen saturations.

The culture swab that was used is the Nest disposable sampler from Wuxi NEST Biotechnology Co., China. The oropharyngeal swab has two breakpoints, one at 32mm and another at 80mm. The most distal break point at 32mm is what broke off.

Suctioning through the tracheostomy tube was attempted but failed to bring up the detached swab. An immediate change of tracheostomy tube confirmed the distal end was in the trachea. The patient was subsequently taken to theatre and rigid bronchoscopy done through the tracheostoma. Due to the aerosolizing risk of the procedure, a nasal rapid antigen test was performed prior to the procedure and the results were negative for SARS-COV-2. A 30° rigid telescope of diameter 5.5mm and working length 489mm was used. The swab was identified in the right main bronchus (Figure 1) and was subsequently removed with a peanut grasping forceps without any damage to the mucosa.

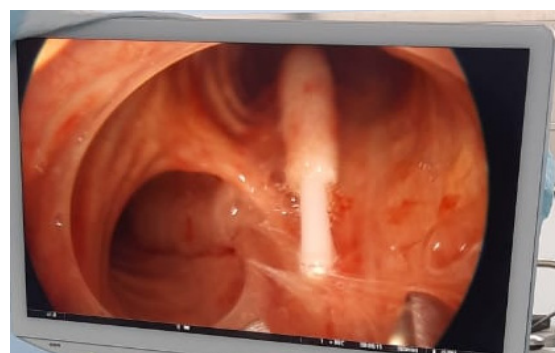


Figure 1: Covid-19 swab lodged in right main bronchus

DISCUSSION

This case highlights the danger of performing a tracheal mucosal swab through the tracheostomy. It also highlights the paucity of data of Covid-19 sampling in laryngectomy and tracheostomy patients. This has not been documented in Africa; however, in other countries there are rare case reports³.

Accurate diagnosis of SARS CoV-2 is the cornerstone of treatment. Real time Reverse Transcription-Reverse Polymerase Chain Reaction (RT-PCR) are highly specific and highly sensitive tests for diagnosing SARS CoV-2¹. They detect the ribonucleic acid (RNA) genes. The primary target tissue for the SARS-CoV-2 is the ciliated respiratory mucosa. The highest accuracy is obtained with specimen from the lower respiratory tract followed by the upper respiratory tract. There are studies that demonstrate the virus is detected in other tissues like the gastrointestinal tract¹.

In laryngectomy patients or those with tracheostomies, where the upper and lower respiratory tract are separated, there is paucity in the guidelines discussing testing⁴⁻⁶. Due to the possibility of a Covid-19 swab breaking off, a tracheal or bronchial aspirate is preferable to an oropharyngeal or nasopharyngeal swab. Using the standard technique, gently introduce the suction catheter into the tracheostomy tube or laryngotomy. Gently rotate the catheter while withdrawing. Cut the catheter tip into a viral testing tube.

This highlights the challenge of knowledge and skill gap in healthcare workers performing tracheal aspirates. Furthermore, there is a dearth of information on the distribution of SARS-CoV-2 in the nasopharynx and oropharynx when the upper and lower airway are separated such as in tracheostomy and laryngectomy patients.

CONCLUSION

Laryngectomy patients and those with tracheostomies are a special population and therefore guidelines are required to provide safe and accurate outcomes in in sampling of Covid-19.

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HAEMATOHIDROSIS: A RARE CLINICAL ENTITY: CASE REPORT

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ABSTRACT

Haematohidrosis is a rare clinical condition where a patient presents with spontaneous, non traumatic bleeding from various sites. A number of cases have been reported but it still remains a condition that not much is known about its aetiology or management. A 15 year old female presented with haematohidrosis and was diagnosed on the basis of the clinical presentation and investigations. The management varies and for this patient beta blockers and anxiolytics helped control the symptoms.

Key words: Haematohidrosis, Stress, Propranolol

INTRODUCTION

Haematohidrosis also termed as haematidrosis or haemidrosis is a rare clinical condition but a well-recognized diagnosis according to the International Classification of Diseases (ICD-9-CM: 705-89).

The main presentation is spontaneous bleeding from the skin without any sign of trauma. Various factors have been attributed to such as extreme stress, systemic disorder such as primary thrombocytopenic purpura or vicarious menstruation have been proposed in terms of aetiology.

CASE REPORT

A 15 year old AO female presented to the clinic with history of acute, spontaneous right bloody otorrhea from the external auditory meatus and bilateral nose bleeds. At no point was she on her menstruation. The patient had 3-4 bleeds in a day. All the bleeding episodes were witnessed by either friends or relatives.

There is childhood history of left chronic otitis media and underwent a cartilage tympanoplasty at the age of 10 years and had been asymptomatic since then (Figure 1). Reports to have mild allergic rhinitis with occasional sneezing and uses antihistamines as per needed with good symptomatic control. No other systemic diseases are documented or a positive history or family history of bleeding tendencies. On examination she was noted to have blood clots at the EAC which were easily removed using a probe and the underlying skin was noted to be normal. There was no evidence of any trauma, purpura, echymosis or scratch marks (Figures 2-4). Video otoscopy was done and no abnormalities were noted (Figures 5 and 6).

The nose bleeding was noted to be around the columella and the anterior nasi. Rigid nasoendoscopy

findings were normal with a normal little's area, mild hypertrophy of the inferior turbinate and no post nasal pathology (Figure 7). The rest of the physical exam was normal. The patient was admitted for further evaluation and noted to have periorbital bleeds (Figure 8). Basic investigations such as total blood count, a peripheral blood film, coagulation profile, liver function tests all of which were normal. A skin biopsy was suggested to rule a dermatological pathology but the parents declined. A temporal bone CT scan and paranasal sinuses CT scan was noted to be normal. A psychiatric evaluation elaborated on the fact the periorbital bleeding was triggered by the illness of a loved one and the other stressor identified was social stigma due to her hearing disability. She exhibited no other signs or symptoms of neurotic behavior or severe anxiety disorder. A treatment protocol on stress management mechanisms and breathing exercises with alprazolam 0.25mg once a day for 2 weeks and propranolol 10mg daily for one month. She did well after 2 weeks with only 4 episodes of bleeding. Alprazolam was withdrawn. There was no further psychiatric review done.

A review done after one month, indicated no bleeding and the patient was carrying out regular exercises to manage her stresses and propranolol was tapered down to alternate days for 2 weeks. Another review was done after six weeks' and it indicated no bleeding and the patient reported to have been able to control her anxiety with breathing exercises and reported she was able to enjoy school without fear of the bleeds and also felt her confidence levels were better. The parents reported that she did demonstrate more positive attitude towards school. The patient re-bleed twice post the withdrawal of propranolol and was restarted on it again.

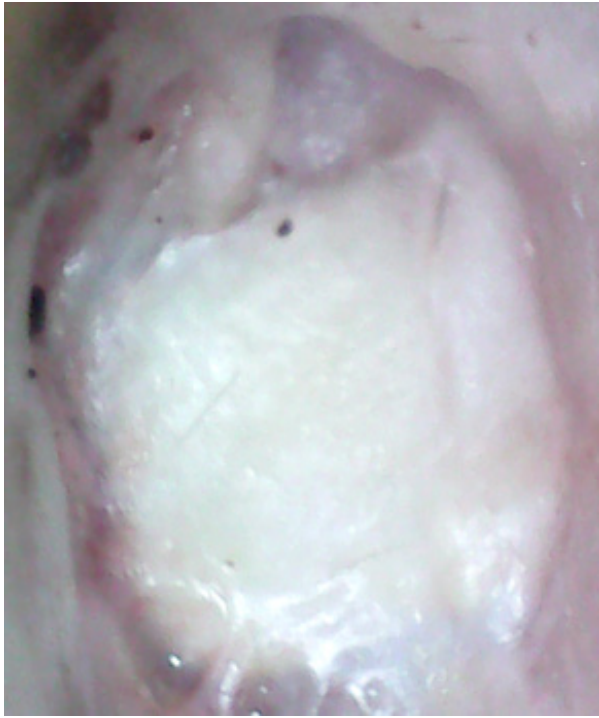


Figure 1: Cartilage tympanoplasty



Figure 4: Right EAC bleed



Figure 2: Left EAC bleed



Figures 5 and 6: Magnification of the EAC skin where the blood clot was removed



Figure 3: Nose bleed

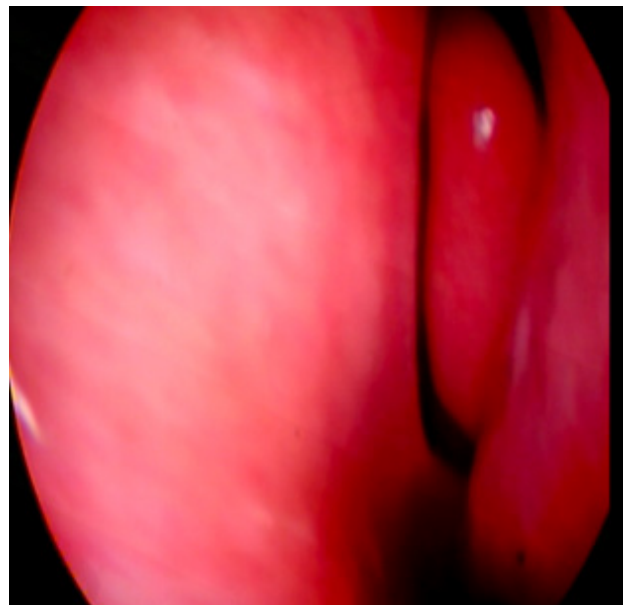


Figure 7: Rigid nasoendoscopy of the left nostril



Figure 8: Periorbital bleed

DISCUSSION

Haematohidrosis is a rare condition that is characterized by spontaneous oozing of blood from the skin and mucocutaneous surfaces¹. Approximately about 80 cases have been reported in the literature, with few reports within the 20th century, and these have usually been diagnosed by the clinical events alone without laboratory investigations. The age distribution varies with most cases noted in the pre-puberty /puberty age groups.

A number of theories have been postulated about the possible pathogenesis. It has been documented in the ancient scriptures; Leonardo Da Vinci described a soldier sweated blood before battle. Jesus Christ experienced haematohidrosis while praying in the garden of Gethsemane before his crucifixion as mentioned in the Defenders Bible by Physician Luke as “and being in anguish he prayed more earnestly and his sweat was like drops of blood falling to the ground”¹.

A major postulated pathogenesis is that severe physiological stress results in cutaneous vasoconstriction secondary to the sympathetic system overdrive after which there is rebound vasodilation and this results in rupture of the tiny blood vessels into the sweat glands causing the blood to mix with the sweat. The sympathetic nervous system is activated by psychological stressor as a stress-fight or flight response and hence a release of excess norepinephrine by the nerves is noted².

Manonukul *et al*³ proposed that there may be some defects in the dermis which cause stromal weakness and these areas provide a communication

with vascular spaces in the dermis. The defects then dilate and enlarge as blood-filled spaces when the blood comes in and then extrudes the blood out by follicular canals or directly onto the skin surface. After the extrusion/emptying the spaces collapse leaving no scar. This phenomenon acts like a balloon, waxes and wanes explaining why these bleedings are sometimes intermittent and self-limiting. He recommended immediate skin biopsy because a late biopsy, after these spaces collapse, will not help in identifying them.

Skin pathohistological study by Zhang *et al*⁴ revealed some intradermal bleeding and emphyseated (obstructed) capillaries. No abnormality was found in sweat glands, hair follicles, and sebaceous glands. They concluded that pathological basis for haematohidrosis might be a distinctive vasculitis.

Various causative factors, like it being a component of systemic disease - One case report has attributed platelet factor-3 dysfunction as a cause^{5,6}.

Vicarious menstruation (a condition in which bleeding occurs from a surface other than the mucous membrane of the uterine cavity at a time when normal menstruation should take place) have also been documented⁷.

Psychogenic purpura is supposed to be caused by hypersensitivity to the patients' own blood or auto erythrocyte sensitization and is characterized by repeated crops of ecchymosis, gastrointestinal bleedings and haematuria⁸.

Another type of bleeding through skin is psychogenic stigmata; a term used to signify areas of scars, open wounds or bleeding through the unbroken skin. Patients belonging to this group were found to be frequently neurotic. The clinical findings of this type are a slight elevation of skin before prolonged oozing of blood, a pea-sized bluish discoloration on patient's palm and erysipelas-like lesions. In another study, a patient developed bleeding from her old scars whenever she had severe anxiety³.

Treatment protocols vary with varied responses. Literature review indicates use of anxiolytics to manage the stress. Beta-blockers has also been used in such cases to help inhibit the normal sympathetic effects². The dosage of the anxiolytics and betablockers are adjusted as per the age and weight of the patient and eventual tapering off is recommended⁹. Transdermal atropine patches over the affected bleeding areas for one-month duration and gradual improvement was noted¹⁰. In some patients there was simply emphasis on stress management, counseling and behavioral interventions¹¹.

CONCLUSION

Rare medical entity with no proper management outline. The key to successful treatment includes convincing the parents about the nature of this illness, its aggravating factors and the possible treatment of this condition.

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EARLY THYROIDECTOMY IN THE CONTEXT OF MULTIPLE ENDOCRINE NEOPLASIA TYPE IIA: CASE REPORT

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ABSTRACT

Background: Multiple endocrine neoplasia type 2 A (MEN 2 A) is an autosomal dominant abnormality combining Medullary Thyroid Carcinoma (MTC), pheochromocytomas and parathyroid tumours.

Objective: To highlight the importance of early screening in a context of at-risk families and to emphasize the concepts of prophylactic surgery and early surgery in patients with RET gene mutation.

Results: This was a 28-years-old female patient (M.D.C), with a family history of medullary thyroid carcinoma, hyperparathyroidism and pheochromocytoma. Given the mutation of the RET gene in our patient and in some members of her family and given a high level of thyrocalcitonin presented by M.D.C, she benefited from a preventive total thyroidectomy associated with a bilateral neck lymph node dissection. The postoperative course was uneventful. The histological results were in favor of microcarcinoma.

Conclusion: Prevention of MTC in a family at risk context is based on genetic screenings. In case of mutation of the RET gene, early thyroid surgery or prophylactic surgery may be recommended.

Key words: MEN 2A, Medullary thyroid cancer, Prophylactic thyroid surgery, Early thyroid surgery

INTRODUCTION

Medullary Thyroid Cancer (MTC) is a special entity within thyroid pathology. It secretes a hormone: calcitonin which dosage allows diagnosis and follow-up. There are sporadic forms and familial forms with autosomal dominant inheritance, occurring in isolated form or integrated as part of multiple endocrine neoplasia type 2 (MEN2). Patients with MEN 2A develop MTC, pheochromocytomas and parathyroid tumors¹. MEN 2A is related to active mutations of the proto-oncogene RET (Rearranged during Transfection) which encodes a transmembrane receptor for tyrosine kinase.

Prophylactic surgery consists, in case of mutation of this proto-oncogene RET, in a total thyroidectomy associated as the case with a bilateral neck lymph node dissection and aims to prevent the occurrence of Medullary Thyroid Cancer (MTC). At this stage, the thyroid gland is healthy, without pathology of C cells¹.

Early surgery is when thyroidectomy is performed on a gland that already carries C cell pathology (C cell hyperplasia and/or microcarcinoma)².

The objective of this study was to emphasize, through the study of an observation, the importance of early diagnosis in a context of at-risk family and to insist on the notions of prophylactic thyroid surgery and early surgery in carriers of the mutation of the RET gene.

CASE REPORT

This was a case of a 28-year-old female patient (M.D.C), with a family history of Medullary Thyroid Carcinoma (MTC), hyperparathyroidism (HPT) and pheochromocytoma (PHEO). Indeed, her mother and two of her maternal aunts presented a MTC associated with an HPT; a cousin was carrying a MTC associated with an HPT and a PHEO and another cousin presented a MTC alone (Figure 1: Family Tree).

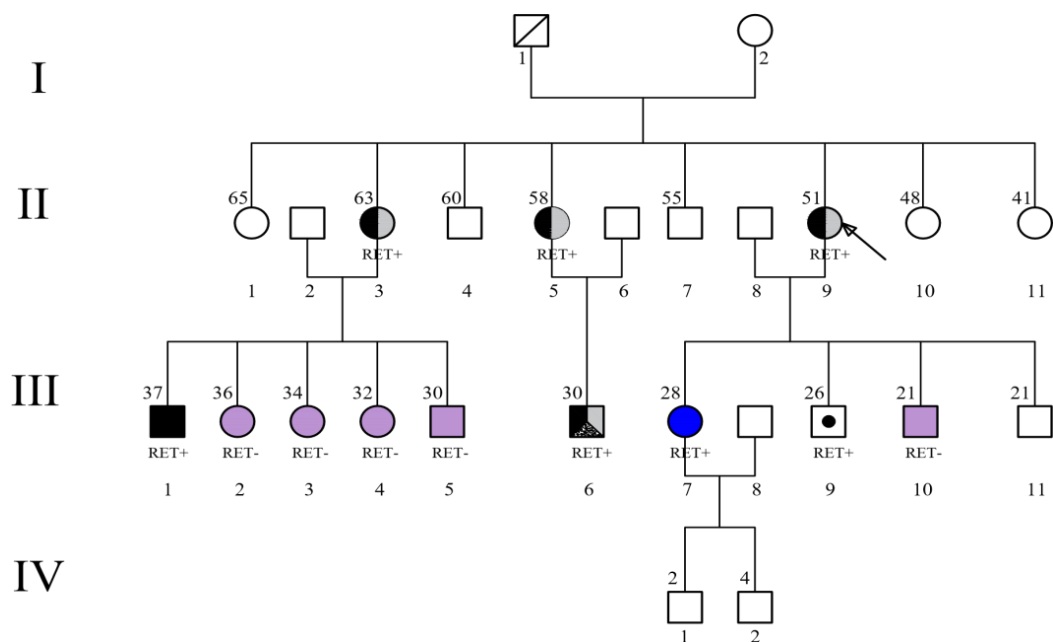


Figure 1: Our patient's family tree

□

○

Subjects unexplored male and female respectively

◻

◌

Deceased subjects

◐

◑

Index case

◑

RET+

◑

Subject carrying a mutation but not explored on the injury level

RET-

No mutation

RET+

Mutation identified

◑

Our patient

◐

◑

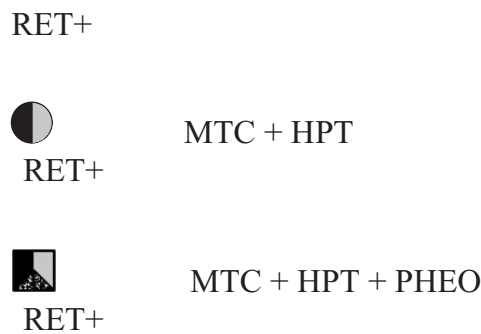
Subjects explored and without mutation

RET-

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Medullary thyroid cancer



The numbers above the symbols indicate the age of the patients.

Figure 1: Our patient's family tree

The numbers above the symbols indicate the age of the patients.

Genetic tests were performed on 12 consenting family members and found a mutation in the RET gene in 7 members (the mother, 2 of her aunts, a brother and 2 cousins) as well as in our patient. In our patient, genetic analysis after sequencing exon 11 of the RET gene had highlighted a point mutation of codon 634 responsible for the substitution of a cysteine by a tyrosine at the protein level.

Faced with this mutation and a high level of thyrocalcitonin, M.D.C was referred to the ENT Department of the Military Hospital of Ouakam (HMO) by his endocrinologist for a prophylactic thyroidectomy as part of a family carrying a NEM2A. At her consultation in ENT, Mrs. M.D.C showed no functional signs, no complaints. The physical examination found a right lobar thyroid nodular mass of 1 cm, hard, non-sensitive and mobile, without palpated cervical lymphadenopathy. The rest of the examination was normal, especially abdominal palpation.

Preoperative serum calcium (87mg/l [84 – 102]) and ultrasensitive TSH (0.95 micro IU/ml [0.35 – 4.94]) were normal. The blood level of base calcitonin was elevated: 124ng/L [reference value less than 10]. The pentagastrin stimulation test was not performed because it was not available in our structure. Blood levels of parathyroid hormone were normal: 60.3 pg/ml [15.0 – 68.3], as were levels of free methoxyamines:

Normetanephrine less than 0.25nmol/l [N inf 0,94]

Metanephrine less than 0.14 nmol/l [N inf 0,37].

The chest X-ray was normal.

At the cervical ultrasound, the right thyroid lobe had two hypoechoic, heterogeneous, lower polar, anterior 12mm and posterior 11mm nodular

formations, presenting an internal color Doppler signal. Abdominopelvic ultrasound was normal.

Given the family history of MTC, the positivity of genetic tests (mutation of the RET gene), clinical and ultrasound thyroid nodules and elevated calcitonin, the decision of prophylactic total thyroidectomy was made. After informed consent, Mrs. M.D.C. underwent a total thyroidectomy associated with a bilateral mediastino-recurrent and jugulo-carotid lymph node dissection. Immediate postoperative course was uneventful. There was no dysphonia and postoperative serum calcium as measured on the third postoperative day and one month after the procedure was normal. The anatomo-pathological results were in favor of medullary microcarcinoma of the thyroid and the dissection reported 10 lymph nodes all free of metastasis.

The level of basal calcitonin (or thyrocalcitonin) normalized postoperatively with the following values recorded:

(i) at 6 months postoperatively: 1.8 ng/l [N inf 10]

(ii) at 12 months: 4.8 ng/l

(iii) at 18 months: 5.7 ng/l

Serum calcium, parathyroid hormone and blood and urinary catecholamines measurements, carried out as part of postoperative follow-up, are normal. The current follow-up time is 6 years, without clinical and/or biological recurrence.

DISCUSSION

The incidence of MTC in nodular thyroid pathology is around 1–2%². The MTC comes in two forms:

(i) a sporadic form in the majority of cases;

(ii) a familial form in almost 30–35% of cases. It is then integrated into multiple endocrine neoplasia type 2A (MEN 2A).

It is a carcinoma for which there is a good correlation between the level of thyrocalcitonine (TC) and the tumor mass³. The finding of a major basal hypercalcitoninemia (in rule > 100 pg / ml) associated with a thyroid nodule is very strongly suggestive of the diagnosis of MTC. This was the case in our patient with a basal CT level of 124 pg/ml and a centimetric clinical thyroid nodule and two ultrasound nodules.

The pentagastrin (Pg) stimulation test is the most widely used for the diagnosis of MTC. It confirms the diagnosis of C cell pathology (MTC or HCC C cell hyperplasia). It is useless if the basal CT value is very high (> 100 pg/ml) with a highly probable CMT diagnosis on clinical or cytological arguments^{4,5}. This was the case with our patient.

Mutations of the proto-oncogene RET located on chromosome 10 have been identified in the NEM 2A. The identification of at-risk relatives carrying the RET mutation allows the diagnosis and early or even prophylactic management of MTC^{6,7}.

Prophylactic surgery consists, in case of mutation of the RET gene, in a total thyroidectomy associated with a bilateral neck dissection and aims to prevent the occurrence of Medullary Thyroid Cancer (MTC). At this stage, the thyroid gland is healthy, without pathology of C cells. Early surgery is when thyroidectomy is performed on a gland that already carries C cell pathology (C cell hyperplasia and/or microcarcinoma)².

In practice, it appears relatively rare for the thyroid body to be free of pathology, and surgery is in fact rarely truly true prophylactic^{7,8}. Our patient had been referred to us by his endocrinologist for prophylactic thyroid surgery. However, histological examination after total thyroidectomy and lymph node dissection had found medullary microcarcinoma confirmed by immunohistochemistry. So it was an early thyroid surgery.

It should be noted that, despite late thyroidectomy, at 28 years of age, we did not find clinical, radiological or histological lymph node metastasis. Long-term healing can only be assessed on the results of annual CT concentration checks associated with a Pg test every year at first, and then every 3 to 5 years⁹.

CONCLUSIONS

Medullary Thyroid Cancer (MTC) is a special entity within thyroid pathology. Its prevention in people from families with NEM 2A is essentially based on screening by molecular studies (research of the mutation on the proto-oncogene RET). In case of mutation, early or even prophylactic thyroid surgery may be proposed.

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