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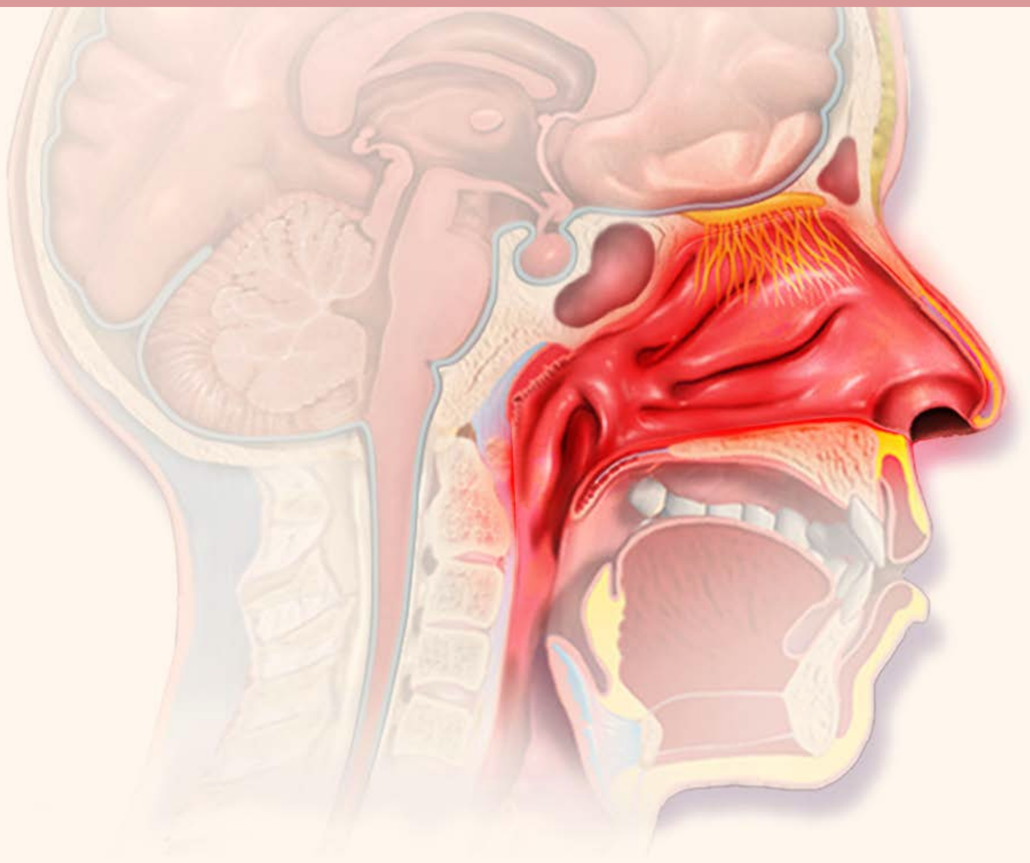

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Case Report

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Clear Cell Myoepithelioma - A Rare Presentation in Nasal Cavity

Arpit Saxena, MS[1*]; A. V. Ramesh, MS[1]; Poonam Raj Mehra, MS[1]; Nikhilesh Kumar, MD[2]; Richa Ranjan, MD[2]

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ABSTRACT

Myoepitheliomas are predominantly tumors of salivary glands constituting less than 1% of all salivary gland tumors. A 65 year old women presented with bleeding unilateral mass for 03 months. Contrast Enhanced Computed Tomography (CECT) revealed a heterogeneously enhancing mass lesion arising from right anterior ethmoid air cells and filling the right nasal cavity entirely. Endoscope guided endoscopic biopsy was performed. Histopathological examination showed a well circumscribed tumour arranged in small glands and sheets. Tumor cell were immunopositive for S100 (Ib), Cytokeratin (CK) and Vimetin (focally) while negative for chromogranin, synaptophysin, CD 10 and Smooth Muscle Actin (SMA). MIB-Labelling Index was <5%. With these features a diagnosis of clear cell myoepithelioma was made. Nasal myoepithelioma is an extremely rare low-grade neoplasm. Nasal myoepitheliomas are composed of myoepithelial cells with solid, myxoid or reticular patterns of growth. The cells themselves may be clear-cell type, spindle-shaped, plasmacytoid and epithetlioid. In all reported cases of myoepithelioma, surgery was the mainstay treatment. Partial maxillectomy via a lateral rhinotomy approach, Caldwell-Luc procedure has been recommended for patients who were suspected to have a low-grade sarcomatous neoplasm. Only five cases have been reported in sinonasal region. We report the second case to be managed endoscopically. We report an extremely rare benign tumor of nasal cavity (clear cell myoepithelioma) which was managed endoscopically with recurrence within 6 months. The recurrence was also managed endoscopically. This case highlights the varied malignancies which may be encountered in sinonasal region.

KEYWORDS: Myoepithelioma; Clear cell tumor; Endoscopic sinus surgery.

INTRODUCTION

Myoepitheliomas are predominantly tumors of salivary glands constituting less than 1% of all salivary gland tumors.[1] Only five cases of sinonasal myoepithelioma have been reported in literature till today's date. We present the first case of clear cell sub type of myoepithelioma in nose and the second case to be managed endoscopically.

CASE REPORT

A 65 years old lady presented with complains of intermittent bleeding for 3 months and nasal obstruction on the right side for 2 months. There were no known comorbidities. On examination patient had a bulge over right lateral wall of nose seen externally. Anterior rhinoscopy revealed a fleshy smooth mass filling right nasal cavity. No blood clots or secretions were seen. Probing revealed the mass to be painless, soft in consistency, arising from right lateral wall of nose and bleeding on touch. Posterior rhinoscopy revealed normal nasopharynx. Contrast Enhanced Computed Tomography (CECT) revealed a heterogeneously enhancing mass lesion arising from right anterior ethmoid air cells and filling the right nasal cavity entirely (Figure 1). No features of bone erosion were seen. Hematolgical and biochemical parameters of patient were normal. It was planned an endoscope guided excision biopsy of mass. Intraopera-

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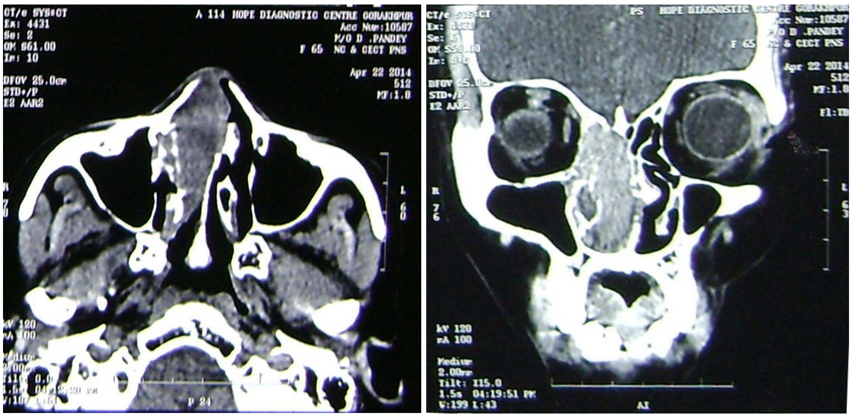

Figure 1: CECT Paranasal Sinuses: Heterogenously enhancing soft tissue mass in right nasal cavity and anterior ethmoid air cells.

tive, mass was seen arising from sphenoethmoidal recess. Brisk bleeding was encountered coming from sphenopalatine artery which was controlled with bipolar cautery. Post op uneventful.

Histopathological examination showed a well circumscribed tumour arranged in small glands and sheets. The individual cells were bland with clear cytoplasm. No layering was seen. Mitosis was scant and no necrosis was seen. Few dead bony spicules were noted (Figure 2). Special stains revealed glycogen in clear cells. Tumor cell were immunopositive for S100 (Ib), Cytokeratin (CK) and Vimetin (focally) while negative for chromogranin, synaptophysin, CD 10 and Smooth Muscle Actin (SMA). MIB-Labelling Index was <5%. With these features a diagnosis of clear cell myoepithelioma was made.

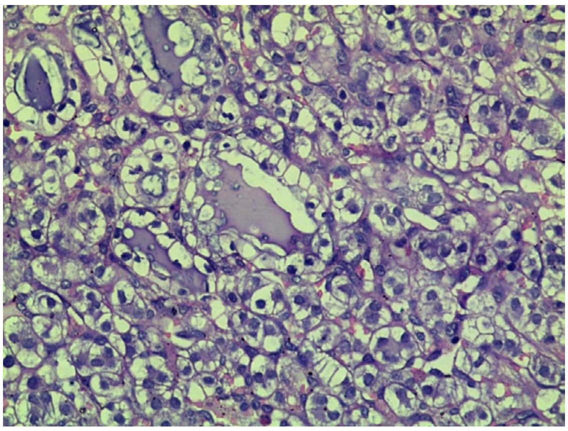

Figure 2: H&E microphotograph showing clear cell in tubules. No mitosis or atypia noted.

Patient was kept on follow up with no further episodes of epistaxis. Endoscopy after 6 months showed fleshy mass arising from middle meatus. Repeat tomography was done which showed recurrent mass lesion arising from right anterior ethmoid air cells. Endoscopic excision of middle turbinate and anterior ethmoid air cells was done and specimen sent for histopathology which also showed clear cell myoepithelioma with no evidence of malignant transformation. There has been no further recurrence during a follow up period of 1year after second surgery.

DISCUSSION

Primary pleomorphic adenomas of the nasal cavity constitute around 18% of sinonasal nonepithelial neoplasms.[2,3] Nasal myoepithelioma is an extremely rare low-grade neoplasm. The main symptoms of nasal myoepithelioma are rapid enlargement of the tumor mass with nasal obstruction and epistaxis for periods varying from 3 month to 3 years.[4,5] The imaging appearance of a myoepithelioma is usually nonspecific.[5]

Nasal myoepitheliomas are composed of myoepithelial cells with solid, myxoid or reticular patterns of growth. The cells themselves may be clear-cell type, spindle-shaped, plasmacytoid and epithelioid.[6] Myoepitheliomas are usually devoid of ductal elements. However in three out of five cases of nasal myopepitheliomas small amounts of ductal elements have been reported. Variable degree of nuclear atypia, often mixed with a population of cells with eosinophilic cytoplasm has been noted. Frankly malignant change has not been seen in nasal myoepitheliomas. Immunohistochemistry (IHC) is an important adjunct in differential diagnosis of myoepitheliomas (Figures 3 and 4). The possible differential diagnosis and their features are as in Table 1.[7]

In all reported cases of myoepithelioma, surgery was the mainstay treatment. Partial maxillectomy *via* a lateral rhinotomy approach, Caldwell-Luc procedure has been recommended for patients who were suspected to have a low-grade sarcomatous neoplasm.[4,8] With the advent of nasal endoscopes Fijukura and Okubu removed a 10 mm tumor endoscopically.[9] In our patient, CECT showed tumor arising from anterior ethmoid air cells, restricted to nasal cavity with no bony erosion. Hence, an endonasal endoscopic approach was planned.

CONCLUSION

We report an extremely rare benign tumor of nasal cavity (clear cell myoepithelioma) which was managed endoscopically with recurrence within 6 months. The recurrence was also managed endoscopically. This case highlights the varied malignancies

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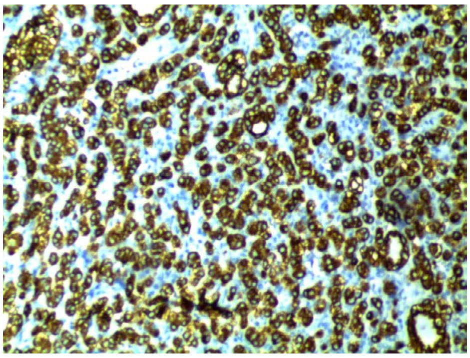

Figure 3: IHC showing membranous cytokeratin positivity.

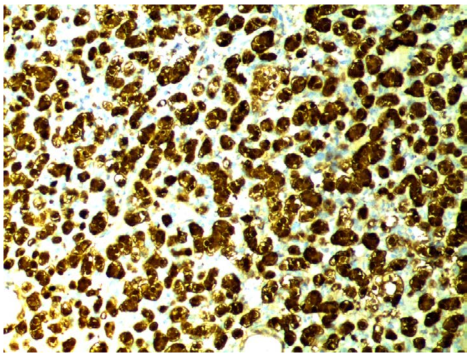

Figure 4: IHC showing diffuse, strong nuclear positivity for S100.

S.No.	Tumor	Morphology	IHC & Special stain
1	Clear cell oncocytoma	Trabecular pattern in cell with central round nuclei and granular eosinophilic cytoplasm	PTAH + Vimentin +/-
2	Epithelial myoepithelial Carcinoma	Bilayer ductal structure, inner cuboidal & outer clear cell	Inner layer – CK + Outer cells – S 100 +, Actin +
3	Mucopidermoid Carcinoma	Squamoid and intermediate cell along with clear cells	Clear cell resistant to diastase, Mucicarmine +
4	Acini cell carcinoma	Microcystic pattern cells with peripheral nuclei and basophilic cytoplasm	PAS + (not sensitive to diastase), Amylase +, CEA +
5	Clear cell carcinoma NOS	Usually diagnosis of exclusion	Negative myoepithelial marker (SMA, S100, Calponin, P63 – ve)
6	Metastatic RCC	Sinusoidal pattern clear cells, Hemosiderin deposits, Haemorrhage present.	CK +, Vimentin +, CD 10 +

Table 1: Histopathological and IHC characteristics of differential diagnosis of myoepitheliomas.

which may be encountered in sinonasal region. An in-depth knowledge, high index of suspicion and use of IHC is required for an accurate diagnosis. Endoscopic excision is likely to be the mainstay of management in the future.

CONFLICTS OF INTEREST: None.

CONSENT

Authors obtain written informed consent from the patient for submission of this manuscript for publication.

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Research

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Cortical Auditory Evoked Potentials in Persons Using Hearing Aids

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ABSTRACT

Introduction: A review of literature on usefulness of Cortical Auditory Evoked Potentials (CAEPS) in verifying the usefulness of hearing aid shows equivocal results and a majority of the studies are carried out in a research laboratory.
Objective: The aim of the present investigation was to investigate the usefulness of recording CAEPs for verification of hearing aids in a clinical set up.
Material And Methods: CAEPs to stimulus /ma/, /ga/ and /ta/ were recorded from 14 persons with normal hearing and nine persons with mild to moderately severe sensorineural hearing loss. For persons with hearing impairment, the testing was carried out without a hearing aid (unaided) and with a hearing aid (aided) programmed based on NAL NL 1 prescriptive formula.
Results: The results revealed that in aided condition, the detectability of CAEP responses was more when compared to unaided condition in persons with hearing impairment. There was a significant difference between the unaided CAEP responses of persons with hearing impairment and CAEP responses of persons with normal hearing. However, no such difference was observed between aided CAEPS responses of persons with hearing impairment and those of normal hearing.
Conclusions: CAEPs can be reliably recorded in a clinical set up from individuals using hearing aids. The detectability of responses increases when a person is wearing hearing aid. CAEPs will be helpful in verification of hearing aids especially in persons with moderately severe to severe hearing loss.

KEYWORDS: Long latency response; Aural rehabilitation; Hearing aid fitting.

INTRODUCTION

The advancement in the field of pediatric audiology has resulted in early, efficient and objective measures of hearing threshold estimation for infants. This has provided with the ability to fit appropriate hearing aid at a very young age. Verification of the selected hearing aid in infants and small children is a challenging task as it is difficult to obtain reliable behavioral measures from them. There is a need to use electrophysiological measures for such population.

A review of literature shows that investigators have studied the usefulness of various auditory evoked potentials such as auditory brainstem response (ABR), auditory steady state responses (ASSR) and cortical auditory evoked potentials (CAEPs) as a tool for verification of selected hearing aid. ABR and ASSR are best elicited by click and tonal stimuli and these stimuli gives very limited information regarding speech perception thus their use is limited. CAEPs can be elicited using speech stimuli and hence can be more useful in verification of hearing aids. CAEPs recorded in persons using hearing aids will also verify if the sounds are sufficiently amplified and processed in the auditory pathway till cortex.

Rapin, Graziani[1] were the first to study the effect of sensorineural hearing loss and personal hearing aids on CAEPs. They found that a majority of their participants (5 out of 8) had aided cortical responses better than the unaided cortical responses to clicks and pure tones,

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however, two of the infants did not show any changes in cortical responses for aided *versus* unaided condition. Though attempts to record CAEPs in persons wearing hearing aid started 50 years back, it is still not a proven measure of validating hearing aid use in the clinical set up. Some of the investigators have reported that CAEPs demonstrate benefit of hearing aids. It has been reported that use of personal hearing aid substantially improve the detectability of CAEPs and a majority of individuals with hearing impairment showed reduced latency, increased amplitude and improved morphology when tested in with their hearing aids, The improvement in detectability was especially observed in individuals with higher degree of hearing impairment.[2]

Recent research has also focused on investigating the usefulness of CAEPs in assessing the benefit from hearing aid in different frequency regions. It has been suggested the recording CAEPs for /m/,/g/ and /t/ stimuli will check the hearing across the speech spectrum, as each of the stimuli represent low, mid and high frequency region respectively.[3]

Contrary to the studies which support use of CAEPs in hearing aid validation, some researchers reported that CAEPs do not reflect the change in hearing aid gain. Tremblay, Kalstein, Billings, Souza[4] observed very subtle enhancement in amplitude of CAEPs when the hearing aid provides mild high frequency gain. Similarly, Billings, Tremblay, Souza, Binns[5] reported no significant difference in latency and amplitude of CAEPs when the hearing aid gain was changed by 20 dB.

Thus, though there is evidence in literature suggesting that CAEPs can be recorded reliably from persons using hearing aid, there is variability in the results observed in different studies. This variability may be due to the variations in the test protocol and the amplification devices used. It has been well established that both stimulus related and acquisition related factors have an effect on CAEPs. In addition the effect of hearing aid related variables on aided cortical potentials is yet to be completely explored. It has been reported that hearing aid processing alters the acoustic properties of the signal used for eliciting CAEPs and the aided CAEPs may not reflect accurately reflect the signal amplified from a hearing.[6,7] Also, CAEPs may not reliably reflect hearing aid gain as amplification alters the signal to noise ratio which in turn can affect the CAEPs.[8] The effect of amplification on hearing aid output is complicated as it depends on the amplification device or the hearing aids used. Easwar, Purcell, Scollie[9] compared the hearing aid processing of phonemes in running speech and phonemes used for recording CAEPs. There was a difference in processing of the two signals by hearing aids. In addition, they observed that the output from the hearing aid varied depending on the hearing aid used.

Thus, it can be inferred from these studies, that the latency and amplitude of aided CAEPs may not be good parameters to measure the benefit from hearing aid/s. However, the presence or absence of waveforms may be better indicator of hearing aid benefit in a clinical situation. Glista, Easwar, Purcell, Scollie[10] investigated the reliability of recording and interpreting CAEPs using a commercially available clinical instrument to assess the benefit from hearing aid technology. They observed that for some children frequency compression hearing aids increased audibility in certain frequency regions which in turn increased the detectability of tone burst CAEPs. An investigation by Billings, Papesh, Penman, Baltzell, Gallun[11] corroborate this. They reported that CAEPs are helpful clinically in determining whether audible signals are detected physiologically.

The aim of the present investigation was to probe the feasibility and usefulness of recording CAEPs from persons using hearing aids, in a clinical set up using a commercially available auditory evoked potential system. The present research also investigated if there is a difference in the CAEP responses recorded from persons with hearing loss and those of normal hearing. All the three measures, the latency and amplitude of peaks as well as the detectablity of waveforms were considered for analysis.

MATERIAL AND METHODS

Participants

Nine individuals with hearing impairment and 14 individuals with normal hearing in the age range of 60-70 years participated in the study. Pure tone average for 500 Hz, 1000 Hz and 2000 Hz was less than 25 dB HL and immittance evaluation indicated no middle ear pathology for participants with normal hearing. For participants with hearing loss, pure tone average ranged between 41 to 70 dB HL in the better ear with an air-bone gap of less than 10 dB and immittance evaluation revealed no middle ear pathology. Retro-cochlear pathology was ruled out based on the clinical history and the results of the audiological test battery including pure tone audiometry, speech audiometry, immittance evaluation and auditory brainstem responses. All the participants with hearing loss benefitted from the hearing aid used in the study. Sound field behavioral thresholds with the hearing aid programmed based on NAL NL formula was less than 55 dB HL. Participants were in good general health, with no report of any otologic or neurologic disorders. The study was approved by the Research and Ethics Committee of Bharati Vidyapeeth University, Pune and informed consent was taken from all the participants before collecting data.

Stimuli For Recording Caeps

Stimulus for CAEPs was natural speech sound /ma/, /ga/ and /ta/ recorded in a computer using adobe audition software, version 2.0. The sampling frequency was 48,000 Hz with 16 bit resolution. The sound was spoken by a native, male Marathi speaker into a unidirectional microphone connected to the computer. The duration of each stimulus was approximately 350 msec. The stimuli were loaded into Biologic auditory evoked potential system for CAEP recording.

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Hearing Aid

A digitally programmable behind-the-ear hearing aid coupled to an open tube was used throughout the study for all the participants. According to the manufacture's published specifications the frequency range of the hearing aid extended from 100 to 6000 Hz. The hearing aid had a maximum output of 133 dB SPL with a gain of 0-100 dB. The hearing aid had 4 channels and 8 bands. Hearing aid used for the research was checked for the electroacoustic characteristics using Fonix 7000 hearing aid analyzer. The hearing aid was programmed using NOAH software and hearing aid programmer, HI-PRO.

Procedure

Biologic auditory evoked potential system (Navigator pro) with auditory evoked potential software version 7.0.0 was used to record CAEP. Participants were instructed to sit on a chair in relaxed and comfortable position. Silver coated disc electrodes were placed on testing sites after cleaning the site with skin preparing gel. Conduction paste was used to increase the conductivity of the signal. The electrodes were securely placed using a medical tape. The inverting electrode was placed on the mastoid of the test ear; non-inverting electrode was placed on vertex (Cz), with the common electrode on low forehead (Fpz). It was ensured that electrode impedance and inter-electrode impedance was less than 5 kΩ and 2 kΩ, respectively. CAEPs were recorded using the protocol given in Table 1. CAEPs were recorded twice to ensure replicability and the waveforms obtained in two recordings were then added to improve the morphology. P1, N1, P2 and N2 peaks were marked independently by two audiologists who were unaware of the test conditions.

Stimuli	/ma/, /ga/, /ta/
Stimulus intensity	60 dB SPL
Repetition rate	1.1/sec
Polarity	Rarefaction
Filter	0.1-30 Hz
No. of channels	Single channel
Amplification	30,000
No. of sweeps	300

Table 1: Protocol for CAEP recording.

For persons with hearing impairment, testing was carried out without a hearing aid (unaided) and with a hearing aid (aided). The hearing aid programmed based on NAL NL 1 prescriptive formula was fitted to the better ear and the poorer ear was blocked during testing. The obtained data from behavioral and electrophysiological measures were tabulated and statistical analyses were carried out using Statistical Package for Social Sciences (SPSS) version 16.

RESULTS

CAEPs could be reliably recorded for all the three stimuli, however all the peaks were not present in all the individuals. The most consistent peaks were P2 and N2. All the four peaks (P1, N1, P2, N2) could be recorded for /ma/ in 13 individuals. For /ga/ sound P2 and N2 were present in all 14 individuals. P1 could be identified in eight individuals and N1 could be identified in only 9 individuals. Responses for /ta/ sound showed P1 and N1 in all 14 individuals, P2 in 13 and N2 in 12 individuals. Figure 1 shows representative waveforms recorded from participants with normal hearing for the three stimuli.

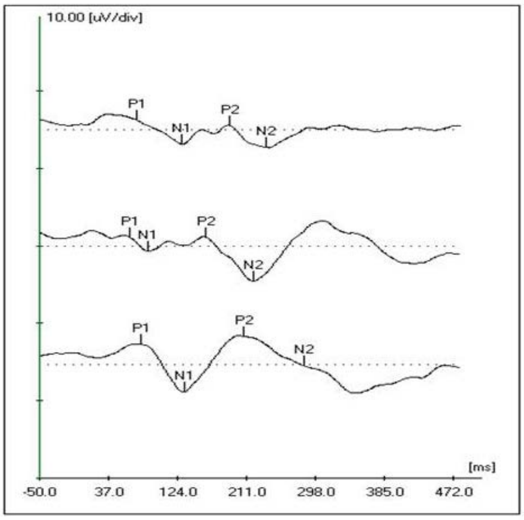

Figure 1: CAEPs for /ma/, /ga/ and /ta/ stimuli in normal hearing individuals.

CAEPs were obtained from individuals with hearing impairment without a hearing aid (unaided) and with a hearing aid (aided condition). The responses obtained were compared with those recorded from persons with normal hearing. P2 was the most consistent response and was present in a majority of individuals with hearing impairment. Two individuals showed no response to all the sounds. For /ma/ sound, CAEPs could be recorded from 7 individuals whereas for /ga/ and /ta/ sound, responses could be obtained only from 6 individuals. Figure 2 shows samples of waveforms obtained from persons with hearing impairment. Detectability of responses increased in aided condition. However, CAEPs could not be recorded from all the individuals with hearing impairment even in aided condition. A lot of individual variability was observed. Some persons showed improvement in morphology with a hearing aid while a few did not show any improvement. Figure 3 shows CAEP responses for the three stimuli in an individual with hearing impairment who showed improvement with hearing aid while Figure 4 shows responses for a person who did not show any improvement in CAEPs with a hearing aid.

Table 2 shows the mean and standard deviation of the latencies (P1, N1, P2, and N2) for individuals with normal hearing and for those with hearing impairment. The table shows latencies in both unaided condition and aided conditions for those with hearing impairment. Overall, the mean latencies were longer for individuals with hearing impairment when compared to

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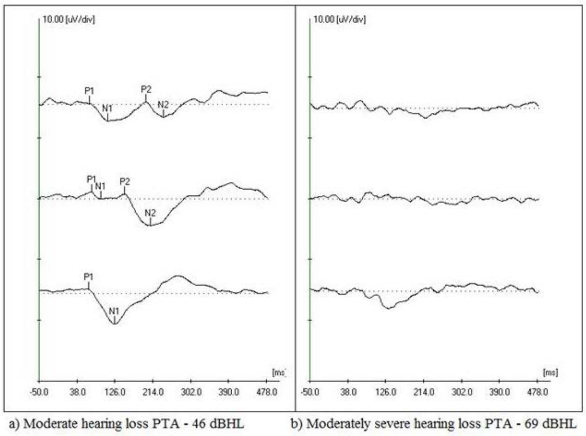

Figure 2. Representation of CAEP responses in individuals with hearing impairment..

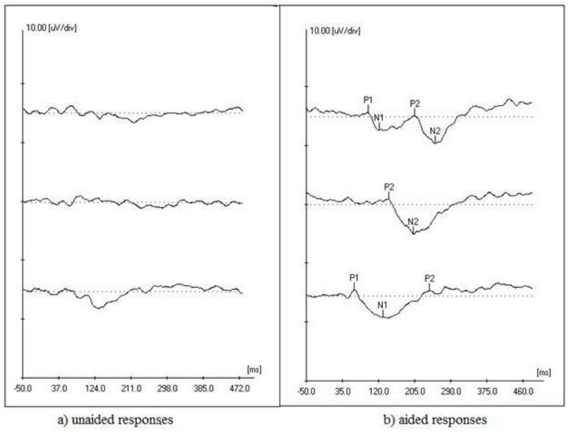

Figure 3. CAEP responses for /ma/, /ga/, and /ta/ stimuli in an individual with hearing impairment who showed improvement with hearing aid.

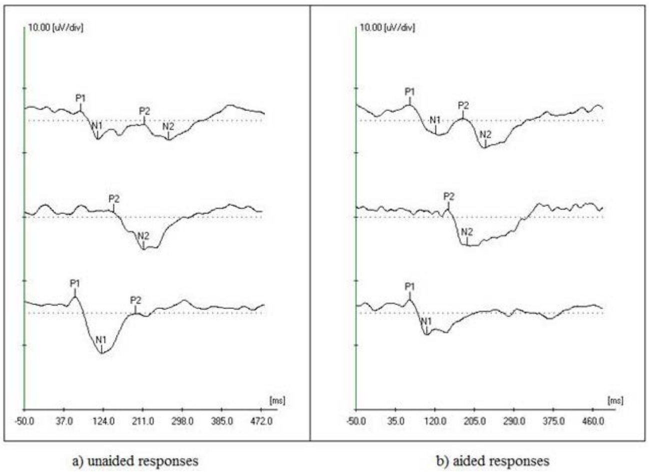

Figure 4. CAEP responses for /ma/, /ga/, and /ta/ stimuli in an individual with hearing impairment who did not show any improvement in CAEPs with a hearing aid.

			/ma/			/ga/			/ta/	
		N	Mean	SD	N	Mean	SD	N	Mean	SD
P1	NL	13	69.28	10.99	8	76.35	13.4	14	74.18	11.30
	HI	5	81.58	9.43	0	-	-	6	69.86	11.33
		6	70.06	9.06	0	-	-	6	77.00	9.66
NI	NL	13	114.74	11.79	9	103.53	20.26	14	127.05	12.80
	HI	5	132.83	14.50	1	122.82	-	6	133.33	11.59
		6	120.38	17.70	1	129.05	-	7	135.45	16.39
P2	NL	13	189.43	9.90	14	163.56	16.82	13	211.09	26.10
	HI	7	214.12	37.90	6	158.54	11.28	6	225.35	26.94
		8	198.41	10.90	7	154.03	8.08	6	233.50	20.60
N2	NL	13	232.45	15.77	14	223.92	21.68	12	251.20	36.90
	HI	5	247.09	12.48	6	219.62	18.29	2	263.35	17.67
		7	245.05	10.79	7	225.87	21.77	3	282.43	5.91

Note: 'NL' refers to normal hearing individual 'HI' refers to individual with hearing impairment.

Table 2: Mean and SD of latency (in msec) of CAEP peaks in individuals with hearing impairment in unaided condition and those with normal hearing.

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those with normal hearing. The latency of peaks in persons with hearing impairment was lesser in aided condition when compared with the unaided condition. Table 3 shows the amplitude (P1N1 & P2N2) for the two groups. The amplitude was larger for /ma/ and /ta/ sound and smaller for /ga/ in individuals with hearing impairment when compared to those with normal hearing except for P2N2 amplitude for /ta/. With a hearing aid, there was an increase in amplitude of P2N2 of /ta/ and /ga/ sound.

Mann Whitney U test was carried out to check if the latency and amplitude observed in persons with hearing impairment was significantly different from those observed in persons with normal hearing. Comparison between unaided responses of individuals with hearing impairment and those of normal hearing showed that for /ma/ sound, the latency of all the peaks was significantly different from those of participants with normal hearing but there was no significant difference in the amplitude of the response. For /ga/ sound the latencies of P1 and N1 as well as amplitudes of P1-N1 and P2-N2 differ significantly from those of persons with normal hearing. There was no significant difference between the two groups for latencies and amplitude of all the peaks of /ta/. It can be observed from the table that the latencies and amplitudes of responses obtained in aided were not significantly different from those obtained for individuals with normal hearing except for latency of N2 and amplitude of P2-N2 for /ma/ sound (Table 4).

To summaraise, the result revealed that in aided condition, the detectability of CAEP responses was more when compared to unaided condition in persons with hearing impairment. There was a significant difference between the unaided CAEP responses of persons with hearing impairment and CAEP responses of persons with normal hearing. However, no such difference was observed between aided CAEPS responses of persons with hearing impairment and those of normal hearing.

DISCUSSION

The aim of the present study was to investigate the usefulness of CAEPs in verification of hearing aid. The speech stimuli used in the present study were consonant vowel (CV) syllables with consonants representing low, mid and high frequency region. The duration of all the three stimuli was 350 m sec with a SD of 12 msec. Two of the consonants were voiced (/m/ and /g/) and one was voiceless (/t/), the vowel /a/ was kept constant.

Statistically significant difference observed between CAEP responses of individuals with hearing impairment in unaided condition and those of normal hearing can be attributed to the loss of audibility in persons with hearing impairment.

			/ma/			/ga/			/ta/	
		N	Mean	SD	N	Mean	SD	N	Mean	SD
P1N1	NL	13	3.77	1.16	8	1.71	1.17	14	6.33	1.80
	HI	5	4.40	1.58	0	-	-	6	6.76	4.66
		6	4.40	3.02	0	-	-	6	5.88	3.90
P2N2	NL	13	2.32	0.69	14	5.33	1.76	12	1.75	1.20
	HI	5	3.97	1.92	6	4.69	2.46	2	1.11	0.45
		7	3.25	1.00	7	4.92	2.13	3	2.27	1.92

Note: 'NL' refers to normal hearing individual 'HI' refers to individual with hearing impairment.

Table 3: Mean and SD of amplitude (in μV) of CAEP peaks in individuals with hearing impairment in unaided condition and those with normal hearing.

	/ma/		/ga/		/ta/	
	Unaided	Aided	Uniaded	Aided	Unaided	Aided
P1	2.80**	-0.48	-2.66 **	-	-0.56	-0.54
N1	2.79**	-0.48	-2.43*	-1.22	-0.76	-1.68
P2	2.36*	-1.82	-0.76	-1.27	-1.23	-1.71
N2	2.77**	-2.58*	-0.82	-0.78	-0.37	-1.16
P1N1	1.54	-0.09	-2.00*	-	-0.30	-1.32
P2N2	0.51	2.02*	-1.96*	-1.04	-0.73	-0.72

Note: *=significant at 0.05 level; **=significant at 0.01 level.

Table 4: Results of Mann-Whitney U test (z values) comparing CAEPs of persons with hearing impairment with those of individuals with normal hearing.

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Similar results have been reported by earlier investigators.[12,13] No significant difference was obtained between aided responses of individuals with hearing impairment and those of individuals with normal hearing indicates that the audibility has improved with hearing aid. However, the latencies in the aided condition were longer than those obtained for persons with normal hearing. Korczak, Kurtzberg, Stapells[13] also reported, prolonged latencies in aided conditions in comparison to the mean latencies obtained in the normal hearing individuals in persons who were benefitting from hearing aids. They concluded that despite of the benefits provided by the hearing aid, individuals with hearing impairment process speech in less effective manner than their normal hearing counterparts.

Inspection of individual data showed that for /ma/ sound, 8 participants showed improvement in aided condition. For /ga/ 6 participants showed improvement and for /ta/ only 4 individuals showed improvement with hearing aid. Individuals with severe hearing loss (pure tone average greater than 71.6 dB HL) showed marked improvement in CAEP responses when unaided and aided responses were compared. These findings suggesting detectability of CAEPs improve when the degree of hearing loss is high as compared to lesser degree of hearing loss, is similar to the finding's reported by earlier investigators.[13] These results suggest that CAEPs can be used to assess the usefulness of a hearing aid in those who cannot give a voluntary response. Recording aided CAEPs in infants and children can assure the clinician and the parents/caregivers that the child is hearing with the hearing aid.

Longer latency observed in aided condition when compared to unaided conditions for some responses could be attributed to fact that CAEP's are sensitive to the changes in temporal features within milliseconds[14] and hearing aids alter the acoustics of speech stimuli and thus CAEPs.[4,5] Billings, Tremblay, Miller[8] studied the effect of hearing aid gain settings on latency and amplitude of P1, N1 and P2 waves. They reported that hearing aid modifies stimulus characteristics such as SNR, which in turn affects CAEP in a way that does not reliably reflect hearing aid gain.

CONCLUSION

To conclude, the results of the present study reveal that the CAEPs can be reliably recorded in a clinical set up from individuals using hearing aids. The detectability of responses increases when a person is wearing hearing aid. CAEPs can be used for verification of hearing aids in difficult-to-test population who are not able to give reliable behavioral responses. CAEPs may be helpful in verifying the usefulness of hearing aids in persons with severe hearing loss.

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CONFLICTS OF INTEREST: None.

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Research

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The Relationship Between the Degree of the Mastoid Pneumatization and Mean Platelet Volume

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ABSTRACT

Objectives: To explore the relationships between mean platelet volume (MPV) values and the degree of the mastoid pneumatization.
Study Design: A retrospective clinical chart review.
Methods: In total, 189 patients (130 females and 59 males; average age, 36.50±15.62 years; age range: 18-65 years) were included in the study. The patients were divided into three groups in terms of the degree of the mastoid pneumatization. The mastoid pneumatization was measured between 0 and 5 cm^3 for group A, between 5 and 10 cm^3 for group B and 10 cm^3 and above for group C, respectively. The MPV values of each groups were compared.
Results: The mean mastoid pneumatization in group A, B and C was 3.96±2.72 cm^3, 8.93±2.14 cm^3 and 11.40±1.36 cm^3, respectively. The mean MPV values of group A, B and C were 7.80±1.22 fl, 8.12±1.46 fl and 7.78±1.26 fl, respectively. The mean MPV values did not differ between males and females ($p>0.05$). The mean mastoid pneumatization was higher in males than in females ($p=0.024$, $p<0.05$). The mean MPV values did not differ significantly between the groups ($p>0.05$).
Conclusions: The degree of the mastoid pneumatization did not affect the MPV values. Further studies with larger numbers of patients are needed to evaluate the relationship between the degree of the mastoid pneumatization and MPV values.

KEYWORDS: Mastoid pneumatization; Mean platelet volume; Chronic hypoxia.

INTRODUCTION

The precise functions of the mastoid air cell system are a current and controversial theme. The mastoid air cell system is adopted as an air reservoir for the middle ear. However, knowledge of the physiologic functions of the mastoid air cell system remains unsatisfactory. The potential functions of the mastoid air cell system are:

- protection of the sensitive inner ear structures from external temperature changes,
- pressure regulator by impact of the large surface area in accordance with gas exchange.[1]

The mastoid air cell system enlarges variably to all regions of temporal bone which has a pyramidal shape. The pneumatization of mastoid bone varies individually and its development alters with age. The mastoid pneumatization has been measured in cadavers *via* cross-sectional histological analysis. In 1940, Diamant[2] is the first to report the mastoid pneumatization in literature. The mean size of normal adolescent's mastoid was reported as 12.07 cm^3 by him. The development of mastoid air cell system is completely mature at approximately 15 years of age in males and 10 years of age in females.[2,3] However, the determination of the exact mastoid pneumatization is difficult although all air cells are interrelated. The mastoid

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pneumatization has been calculated quantitatively by several methods including such as water-weight method,[4] an acoustic method[5] and a pressurized transducer.[6] Recent and significant advances in computed tomography (CT) provide better images of the anatomical features of the temporal bone.[1] Multiplanar reconstruction (MPR) is used to this end. Recent advances in CT allow simple and accurate measurement of the degree of mastoid pneumatization.[7] Cadaver studies is associated with more errors than are computer-assisted anatomical approaches. Especially, measurements derived from CT images with the aid of MPR afford objective and reliable values. Only a few studies have measured the mastoid pneumatization using this technique.

Mean platelet volume (MPV) is used as a parameter of platelet functions. In literature, increasing MPV levels have been associated with the prognosis of some diseases including such as hypertension, unstable angina pectoralis, neurological diseases, autoimmune diseases and obstructive sleep apnea.[8-15] MPV may be used as a marker that indicates chronic intermittent hypoxia. In the study of Somuk et al.[16] reported that MPV parameter was found high in the children with chronic effusion otitis media. According to Wittmaack's endodermal theory,[17] middle ear diseases in infancy and early childhood are reduced the pneumatization of the mastoid bone. Therefore, hereditary and environmental theories proposed that a small mastoid air cellular system predisposes to chronic or acute otitis media. To our knowledge, there is no reported study that exploring the relationships between MPV values and the degree of the mastoid pneumatization. We address this topic in the present study. We explored the relationships between MPV values and the degree of the mastoid pneumatization.

MATERIALS AND METHODS

We retrospectively reviewed data collected from January 2013 to January 2016 on patients which were referred to the Department of Otolaryngology, Head-and-Neck Surgery, of our hospital for trauma. In total, 189 patients were included in the study. Patients with ossicular chain defects, a cholesteatoma, tympanosclerosis, atelectasia, a history of previous ear surgery, history of chronic otitis media and temporal bone fractures were excluded from the study. All patients underwent CT imaging to exclude temporal bone fractures. No any temporal bone fracture was determined on CT imaging in all of the patients. The side-effects of radiation were explained to all patients prior to CT, as was the reason why CT was planned. All patients were told of the purpose of the study and written informed consent was obtained from the patients. The study was conducted in accordance with the principles of Helsinki Declaration. The study protocol was approved by our local Ethics Committee. A multidetector CT system (Siemens Sensation 40, Erlangen, Germany) was used for CT imaging. Imaging parameters included a slice thickness and reconstruction interval of 0.5 mm and a field of view of 21.8×28.8 cm; we took at least 150-400 images, which were reconstructed using a classical filtered-back projection. Temporal CT imaging was performed using a Med plus Dicom Wiever system (Med plus Ltd., High Wycombe, UK). No contrast material was injected. The images were evaluated on a workstation (Leonardo; Siemens) by two experienced radiologists. In this volumetric procedure, mastoid air cells with a gray-scale level similar to air in the temporal bone were determined on the CT imaging. After image processing, only the volumes of the extracted pneumatized parts were measured. The right and left sides were calculated separately in each patient (Figure 1A, 1B and 1C). Routine blood samples were taken from the antecubital vein into tubes with ethylene-diamine-tetracetic acid (EDTA) by a nurse. MPV was measured by hematology analyzer machine. Normal values for MPV were accepted as 6, 0-11, 0 fl. The patients were divided into three groups in terms of the degree of the mastoid pneumatization. The mastoid pneumatization was measured between 0 and 5 cm^3 for group A, between 5 and 10 cm^3 for group B and 10 cm^3 and above for group C, respectively. The MPV values of each group were compared.

Statistical Analysis

Number Cruncher Statistical System (NCSS) 2007 software (Kaysville, UT, USA) was used for all statistical analyses. Descriptive statistics (means and standard deviation, medians with interquartile range) were derived. The significance of each intergroup difference was analyzed using Student's t-test, and the significance of any difference in median values was explored with the aid of the Mann-Whitney U-test and Chi-square test. Qualitative data comparisons were performed using the Pearson χ^2 test. A $p<0.05$ was considered to reflect statistical significance.

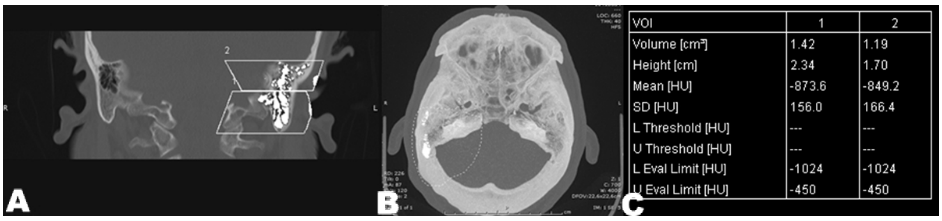

Figure 1A: The measurement of left mastoid air cell volume on the CT coronal slice with a gray-scale similar to air.
Figure 1B: The measurement of right mastoid air cell volume on the CT axial slice with a gray-scale level similar to air.
Figure 1C: Calculation of the areas and the volumes of the extracted pneumatized parts of mastoid bone.

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RESULTS

We included 189 patients: 130(68.8 %) females and 59(31.2 %) males. Their average age was 36.50±15.62 years (range: 18-65 years). The mean mastoid pneumatization in group A, B and C was 3.96±2.72 cm^3, 8.93±2.14 cm^3 and 11.40±1.36 cm^3, respectively (Table 1). The mean MPV values of group A, B and C were 7.80±1.22 fl, 8.12±1.46 fl and 7.78±1.26 fl, respectively (Table 2). The mean MPV values did not differ between males and females ($p>0.05$). The mean mastoid pneumatization was higher in males than in females ($p=0.024$, $p<0.05$) (Table 3). The mean MPV values did not differ significantly between the groups ($p>0.05$) (Table 4).

	Mastoid volume (cm^3)	Patient number	%	Mean volume (cm^3)
Group A	Between 0 and 5 cm^3	59	31.2	3.96±2.72
Group B	Between 5 and 10 cm^3	84	44.4	8.93±2.14
Group C	10 cm^3 and higher	46	24.4	11.40±1.36

Table 1: Summary of group characteristics.

Groups	Mean Platelet volume (fl)
Group A	7.80±1.22 (5.50-10.92)
Group B	8.12±1.46 (6.22-11.60)
Group C	7.78±1.26 (6.24-12.50)

Table 2: The mean platelet volume of groups.

	Mastoid pneumatization (cm^3)
Males	9.50±2.53 cm^3 (2,96-14,89 cm^3)
Females	8.69±1.82 cm^3(3,02-13,66 cm^3)
*p	0.024

*Mann-Whitney U-test.

Table 3: Comparison of the levels of mastoid pneumatization between males and females.

*p			
	Group A	Group B	Group C
Group A	x	0.258	0.622
Group B	0.258	x	0.496
Group C	0.622	0.496	x

*Mann-Whitney U-test.

Table 4: Comparison of the levels of mean platelet volume of each groups.

DISCUSSION

The degree of mastoid pneumatization plays a crurical role in middle ear physiologic functions. The development of mastoid pneumatization varies between individuals.[1] Two hypothesis have been propounded among inter-individual variations of the degree of the mastoid pneumatization. The first hypothesis is that the degree of mastoid pneumatization is determined genetically. In study of Sade et al[18] reported that patients with otosclerosis have larger temporal bone pneumatization than do healthy subjects. In another study, Pata et al[19] investigated the relationship between presbycusis and mastoid pneumatization considering the etiologies of both are reflected to have genetic factors. The cited authors found no differences between the presbycusis subjects and normal subjects in terms of the volume of mastoid pneumatization.[19] Todd et al[20] explored the reason why cystic fibrosis patients had significantly less otitis than the normal population. Cystic fibrosis patients frequently have nasal polyps and sinusitis, but interestingly are spared from an increased occurrence of otitis media. This condition legitimized the authors. The authors reported that mastoid pneumatization of cystic fibrosis patients was larger than the normal population.[20] The second hypothesis is that the status of the middle ear cavity affects the degree of mastoid pneumatization. The degree of pathologic involvement of the middle ear cavity among childhood states the size of the mastoid pneumatization. Increasing the number of pathologic involvement of the middle ear cavity among childhood decreases the degree of the mastoid pneumatization. Therefore, impact of the degree of mastoid pneumatization on hematological parameters remains unclear. Is there any relationship between the poorly mastoid pneumatization and systemic chronic intermittent hypoxia? Or is there any predictive value in hematological parameters for defining poorly mastoid pneumatization? These questions remain unclear. Also, it was the consideration that legitimizes the present study. MPV may be used as a marker that indicates chronic intermittent hypoxia. In the present study, the patients were divided into three groups in terms of the degree of the mastoid pneumatization. The mean mastoid pneumatization in group A, B and C was 3.96±2.72 cm^3, 8.93±2.14 cm^3 and 11.40±1.36 cm^3, respectively. The mean MPV values of group A, B and C were 7.80±1.22 fl, 8.12±1.46 fl and 7.78±1.26 fl, respectively. The mean MPV values did not differ significantly between the groups. To our knowledge, the present study provides the first report of explored the relationships between the mastoid pneumatization and MPV values. However, the relationship between systemic chronic hypoxia and mastoid pneumatization remains unclear. The discrepancies among previous studies with our study may be attributable to the imaging parameters used, subject data and sample size. Although previous radiological studies have been measured the two dimensional size of mastoid pneumatization, in the present study we measured the degree of mastoid pneumatization using a three-dimensional computer-based image reconstruction technique. The value of the technique used in the present study is its high accuracy and easy-to-use. The limitations of our study include the small sample size and the lack of randomization, the lack of assessment of other systemic chronic hypoxia parameters. If assessment of other systemic chronic hypoxia parameters were performed, the study may be more valuable and effective.

CONCLUSIONS

In conclusion, the MPV values did not affect the degree of the mastoid pneumatization. Further studies with larger numbers of patients are needed to evaluate the relationship between the degree of the mastoid pneumatization and MPV values.

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CONFLICTS OF INTEREST

No author has any potential conflicts of interest.

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Case Report

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Rare Localization of Lymphoma

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ABSTRACT

Objective: Primitive Thyroid Lymphomas (PTL) are rare tumors. Women in the sixth or seventh decade of life are more commonly affected. In the present study, we report a case of primitive thyroid lymphoma and we review the epidemiology, the clinical presentation, the diagnosis, and the treatment of this rare disorder.
Case Report: A 47-year-old woman presented to our department reporting a recent-onset neck mass since 3 months. Clinical examination revealed an enlargement of the thyroid gland with a 2.5 cm-firm left nodule. Cervical ultrasound was done. The patient had a thyroidectomy associated with bilateral Central Lymph Node Dissection (CLND). The diagnosis was a transformation of a Mucosa-associated lymphoid tissue (MALT) lymphoma into an aggressive Diffuse Large B-Cell Lymphoma (DLBCL).
Conclusion: The most common type of primary thyroid lymphoma (PTL) is diffuse large B-cell lymphoma, which behaves in a more aggressive manner than mucosa-associated lymphoid tissue lymphoma. Treatment and prognosis of PTL depend upon the histology and stage of the tumor at diagnosis.

KEYWORDS: Thyroid; Lymphoma; Mucosa-associated lymphoid tissue (MALT); Diffuse large B-cell lymphoma.

ABBREVIATIONS: PTL: Primitive Thyroid Lymphomas; CLND: Central Lymph Node Dissection; MALT: Mucosa-associated lymphoid tissue; DLBCL: Diffuse Large B-Cell Lymphoma; FNA: Fine Needle Aspiration; NHL: Non-Hodgkin lymphoma.

INTRODUCTION

Primary Lymphoma (PL) of the thyroid is a very rare disease. It is less than 2 to 5% of malignant neoplasms of the thyroid.[1] Involvement of the thyroid gland may occur in the context of a systemic disease or rarely be primitive. The most common histological sub-type is a diffuse large B-cell lymphoma (DLBCL) followed by Mucosa-associated lymphoid tissue (MALT) lymphoma.

CASE REPORT

A 47-year-old woman presented to our department reporting a recent-onset neck mass since 3 months. There were no local obstructive symptoms associated such as dyspnea or dysphonia. Her medical past history was unremarkable. She had no history of personal or family thyroid disease or radiation exposure. Clinical examination revealed an enlargement of the thyroid gland with a 2.5 cm-firm left nodule. No cervical nodes were palpable.

Cervical ultrasound showed multinodular goiter with hypoechogenes heterogeneous nodules containing microcalcifictions. Infracentimetric lymph nodes in levels ll, III, IV of the both lateral neck were detected. The patient was in biological eutyhroidie. She underwent thyroidectomy associated with bilateral central lymph node dissection (CLND).

On macroscopic appearence, the thyroid gland was solid, nodular white-gray colored

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with a fish-flesh. On microscopic examination, the findings showed a marked diffuse lymphocytic infiltration destroying thyroid follicles. Large lymphatic cells with a typical and large nuclei containing central nucleoli were observed.

Elsewhere, we found a typical lymphocytes with small and centrocyte-like cell. The neighboring thyroid parenchyma contained lesions of thyroiditis. The metastasis of central neck node was evident. Immunohistochemistry showed CD20 positivity and CD15, CD3, CD20 negativity. The diagnosis was a transformation of a MALT lymphoma into an aggressive diffuse large B-cell lymphoma (Figure 1).

Total body Computed Tomography (CT) scan was realized to complete staging according to Ann Arbor classification. It did not document any pathological finding. The diagnosis of primitive thyroid lymphoma was then made. The patient was at stage IIE. A combination of chemotherapy and radiotherapy treatment was adjuncted post-operatively. No tumor recurrence was observed after a mean follow-up of five years.

DISCUSSION

Primitive thyroid lymphoma (PTL) represents 2 to 7% of all extranodal primitive lymphomas. It mainly occur in middle- to older-aged patients with a predilection for females in the 6th decade of life.[2]

Most of the patients with PTL have a previous history of auto immune thyroid it is with or without hypothyroidism. In fact, Hashimoto's thyroiditis co-exists in 83% of patients with PTL. Furthermore, in patients affected by chronic autoimmune thyroid it is, the probability of developing a PTL is 20 times greater than in the general population.[2]

Non-Hodgkin lymphoma (NHL) is the most common PLT (93%). Two sub-types are frequent: Diffuse large B-cell lymphoma is the most encountered accounting for more than 50% of cases, followed by mucosa-associated lymphoid tissue (MALT) lymphoma.[3] Upto 40% of all diffuse large cell lymphomas appear to have undergone transformation from a MALT lymphoma.[4]

Clinically, general symptoms associated with lymphomas, such as fever, excessive perspiration and weight loss, are present in only 10-20% of patients.[2] A rapidly growing (usually within 1-3 months), painless thyroid enlargement, either in the form of goiter or discrete nodule, is the most common clinical presentation in PTL.

Diffuse large B-cell lymphoma (DLBCL) thyroid lymphoma is considered as a high grade lymphoma with a more aggressive clinical course. They present as a painless fast-growing mass causing compressive symptoms like dysphagia, hoarseness or dyspnea. These symptoms overlap with that of the anaplastic thyroïd carcinoma.[5]

MALT lymphoma is considered low grade tumor with an indolent natural history and presents as a slow-growing tumor with early stage disease confined to the thyroid. DLBCL can develop from MALT lymphoma, and these two subtypes can be detected in the same gland. The mixed MALT and DLBCL sub-type shows the same clinical behavior as that of DLBCL.[6]

The diagnosis of PTL is not always evident. In fact, due to their rarity and clinical polymorphism the diagnosis is often made on definite histology after thyroid surgery as the case of our patient. Effectively, Fine Needle Aspiration (FNA) results are in-consistent due to the histopathological similarities between primary thyroid lymphoma and Hashimoto's thyroiditis.[7]

Once a diagnosis is made, total body CT scan should be performed to complete staging, according to the Ann Arbor classification. Large series of PTL revealed that about 50% of cases is confined to the gland (stage IE), 45% involved the gland and regional lymph nodes (stage IIE). lymph node involvement above and below the diaphragm (stage IIIE) or extranodal disease (stage IV) are found in only 5% of cases.[2,4]

Treatment depends on the histological subtype and the stage of the disease. Despite controversy regarding the optimal modality for the management of PTL, the combination of chemotherapy and locoregional radiotherapy is the standard treatment of localized aggressive lymphoma diffuse large B-cell lym-

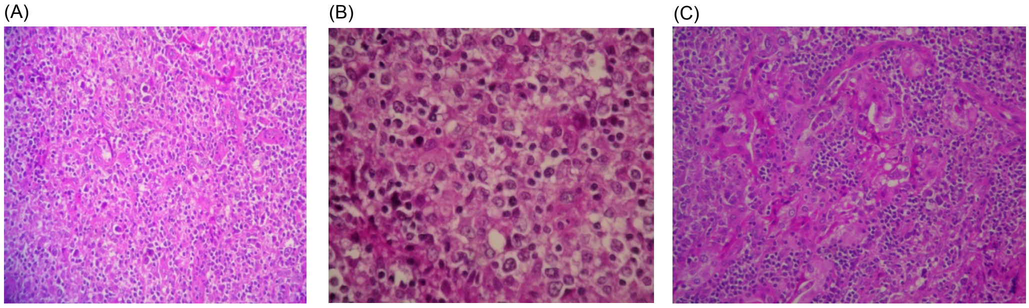

Figure 1: Microscopic finding of the thyroid mass showing. A) Marked lymphocytic infiltration of the thyroid parenchyma. B) Infiltration is made by large lymphocytes with voluminous nuclei. C) Lymphoid infiltration destroying folliclar thyroid realizing lymphoepithelial lesions.

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phoma.[8] The conventional chemotherapeutic regimen consists of cyclophosphamide, doxorubicin, vincristine, and prednisone (CHOP), and radiotherapy is used for local disease control.[6]

Surgery is the primary treatment of localized MALT lymphomas[1,9] in disseminated or aggressive disease, surgery may be indicated for alleviation of compressive symptoms or protection of the airway.[9] Surgical dissection may be more complicated than in standard cases of thyroïdectomy due to the possible tight adhesions existing between the gland's capsule and the surrounding structures.[2]

The prognosis of PTL is affected by disease stage. In localized tumors, it is usually favorable with a survival rate at 5 years from 70% to 80%.[10] However, the prognosis is very poor for lesions with extracapsular invasion (IIE) (20 to 50%). For stages IIIE and IV, the rates are 15 to 35%.[2]

CONCLUSION

In summary, PTL has excellent prognosis when it is confined to the regional neck area and treated properly according to histologic sub-type and stage. The diagnosis should be early evoked in abrupt thyroid enlargement or compression symptoms. Treatment of PTL requires a multidisciplinary approach in order to choose the most appropriate therapy.[4]

CONFLICTS OF INTEREST: None.

CONSENT

The authors article did not publish any personal photo or information regarding any of the patients in his manuscript. Thus, the consent is not required for the article publication.

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Research

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Imaging of Paranasal Sinus Mucoceles

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ABSTRACT

Introduction: Mucoceles are cystic masses developing after obstruction of the sinus ostium. The symptoms are not specific. Computed Tomography scan (CT scan) and Magnetic Resonance Imaging (MRI) confirm the diagnosis.
Objectives: We herein review the radiologic characteristics of mucoceles in CT scan and MRI.
Materials and Methods: We report a retrospective study of 43 patients diagnosed with paranasal sinuses mucoceles. CT scans were performed for all patients, but MRI was carried out only in selected cases.
Results: Our study was constituted of 27 males and 16 females with a mean age of 47 years. The CT scan appearence of mucoceles were in all cases as a well circumscribed expansile sinus mass with an effect on the neighbor bone structure. This mass was hypodense in 26 cases, isodense in 14 cases and hyperdense in 3 patients. The paranasal sinuses most frequently affected in our series were the fronto-ethmoidal sinuses. The most affected bone eroded was the lamina papiracea. Intracranial extension was seen in four cases. CT scan allowed to predict the cause of mucoceles in some cases and to provide information about anatomic variants. MRI was realized for 15 patients in addition to the CT scan. It allowed to study the extension of mucoceles to the neighboring organs especially orbital and endocranial ones.
Conclusion: The presentations of mucoceles on imaging are quite variable. CT scan provides precious information about the location, bone erosion and extension of the mucoceles. MRI is indicated in some cases especially in cases of orbital or cranial extension.

KEYWORDS: Mucoceles; Paranasal sinuses; Computed Tomography scan (CT scan); Magnetic Resonance Imaging (MRI).

ABBREVIATIONS: CT scan: Computed Tomography scan; MRI: Magnetic Resonance Imaging.

INTRODUCTION

Mucoceles are benign, slow-growing paranasal sinus lesions that develop after obstructions of the sinus ostium.[1] Symptoms are variable. The diagnosis is based on imaging. CT scan of the sinuses is the method of the choice. MRI is indicated in some cases and provides much information of mucocele extensions to adjacent compartments.[2]

The purpose of this study was to review the role of pre-operative imaging and to illustrate the main characteristics imaging findings of paranasal sinuses mucoceles.

MATERIALS AND METHODS

We conducted a retrospective review of the charts of 43 patients diagnosed with paranasal sinus mucoceles who were admitted to our Department of Otolaryngology, between January 1990 and December 2012.

Review of the patients' medical records including out-patient clinical records and reports of imaging were performed.

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CT scans of the head were performed for all patients. Axial, sagittal, coronal and contrast CT scan with 3 mm slice thickness were reviewed in all cases.

MRI was carried out only in selected cases for the evaluation of the extension of sinonasal mucoceles. MRI findings on coronal and axial views in T1, T2 weighted and contrast enhanced images were studied.

RESULTS

Clinical Features

Our study was constituted of 27 males and 16 females (sex ratio=1.68) with a mean age of 47 years (from 14 to 77 years).

Rhinosinusitis past history was present in six patients, a facial traumatism in seven patient and eleven patients have undergone prior sinus surgery.

The most commonly reported symptoms were ophthalmologic one (n=24, 56%), including proptosis (n=16), chronic lacrimation (n=7), diplopia (n=3), visual acuity reduced (n=2) and ptosis (n=1).

Headache was present in twenty three patients. Rhinologic symptoms were reported in 20 patients and were dominated by chronic discharge (17 cases).

On examination, we noted a face swelling in 17 cases, a proptosis in 16 patients and ophtalmoplegia in two cases.

Endoscopic nasal examination revealed an obstructive deviation of nasal septum in 10 cases, a filling of middle meatus in six patients and adhesions between the middle turbinate and the nasal septum in three cases.

Radiologic Findings

CT scan: The CT scan appearence of mucoceles were in all cases as a well circumscribed expansile sinus mass with an effect on the neighbor bone structure. This mass was hypodense in 26 cases, isodense in 14 cases and hyperdense in 3 patients. After injection of contrast agents, we saw a poor enhancement in three cases and a peripherally enhanced image in all others cases.

The paranasal sinuses most frequently affected in our series were the fronto-ethmoidal sinuses (Figure 1). Mucoceles involved both the frontal and ethmoidal sinus in fourteen cases, ten mucoceles were located in the ethmoid sinus, and five were located in the frontal sinus (Figures 2, 3 and 4).

Bone erosion was noted (Figures 5 and 6). The most affected was the lamina papiracea which was eroded in 27 cases (Table 1).

Intracranial extension was seen in four cases and was

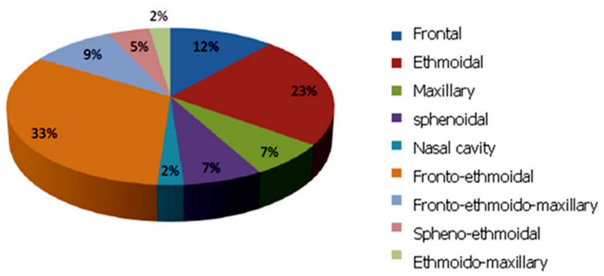

Figure 1: The location of the mucoceles in our patients.

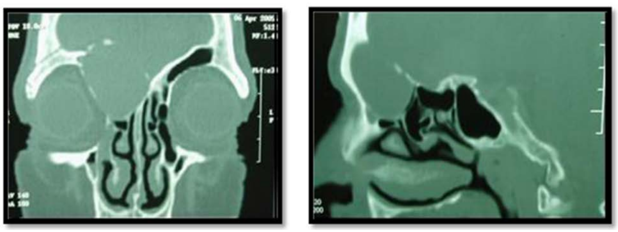

Figure 2: CT scan imaging (coronal and sagital sections) showing an expansile mass in the fronto-ethmoidal sinuses with orbital involvement.

Figure 3: Axial CT scan of a mucocele involving the right ethmoid.

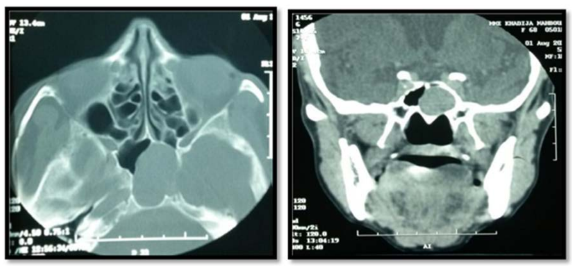

Figure 4: Axial and coronal CT scan imaging demonstrating a mucocele of the sphenoid sinus.

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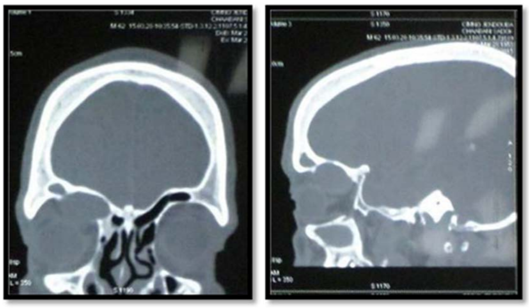

Figure 5: CT scan with coronal and sagital reconstruction: Mucocele within the left frontal sinus with erosion of the roof of the orbite and extension into the orbital cavity.

Figure 6: Axial computed tomographic image of the paranasal sinuses showing a completely opacified right maxillary sinus with a medial bulge of the wall of sinus and the septum.

Bone erosion	Number of cases	Mucocele location							
		F	E	M	S	FE	FEM	SE	EM
Lamina papiracea	27	2	7	_	_	12	4	2	_
Orbital roof	9	2	_	_	_	5	2	_	_
Ethmoidal roof	6	_	_	_	_	4	2	_	_
Anterior wall of frontal sinus	5	2	_	_	_	2	1	_	_
Posterior wall of frontal sinus	5	2	_	_	_	2	1	_	_
Postero-superior wall of sphenoidal sinus	3	_	_		1	_	_	2	_
Medial wall of maxillary sinus	2	_	_	2	_	_	_	_	_
External wall of maxillary sinus	1	_	_	1	_	_	_	_	_

F: Frontal sinus; E: Ethmoidal sinus; M: Maxilary sinus; S: Sphenoidal sinus.

Table 1: Bone erosion in our study.

consequently to a frontal mucocele in one case, fronto-ethmoidal mococele in two cases, spheno-ethmoidal mucocele in one other case (Figure 7).

Extension into the orbite was observed in 27 cases and concerned predominately the medial wall of the orbite. Optic nerve was repulsed in two cases of fronto-ethmoidal mucoceles and one case of fronto-ethmoido-maxillary mucocele. It was compressed in others three cases of: Ethmoidal, Sphenoidal and Spheno-ethmoidal mucoceles. We noted one case of atrophic optic nerve in a sphenoidal mucocele (Figures 8, 9 and 10).

CT scan allowed to note a proptosis in 17 cases (grade 2 in 16 patients and grade 3 in one patient with a fronto-ehtmoidal mucocele associated with an important orbital extension) Others lesions were observed some predicted of the cause of mucoceles:

*Adhesions between the middle turbinate and lateral nasal wall were observed in three patients who had prior sinus surgery and were seen respectively in an ethmoidal, fronto-ethmoidal and fronto-ethmoidal maxillary mucoceles.

*Calcifications of the frontal recess in one patient having a frontal mucocele and history of craniofacial traumatism.

*Sinus retention were noted in three cases of maxillary mucoce-

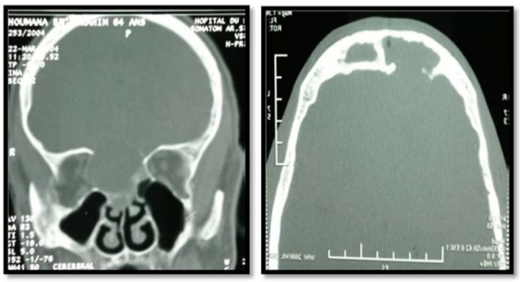

Figure 7: Axial and coronal CT scan imaging of the sinuses demonstrating a mucocele of the frontal sinus and the ethmoid with important endocranial extension.

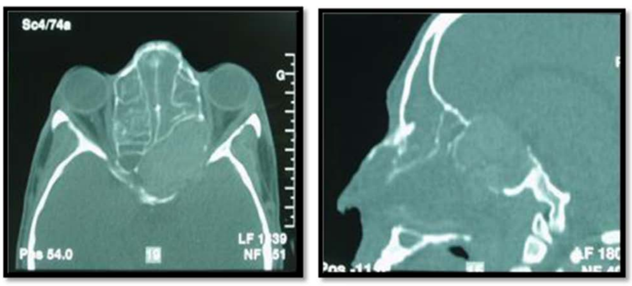

Figure 8: Axial and sagittal images of CT scan demonstrating a mass consistent with a left spheno-ethmoid mucocele. The medial orbital wall is expanded and we note a mass effect on the medial rectus and optic nerve with endocranial extension.

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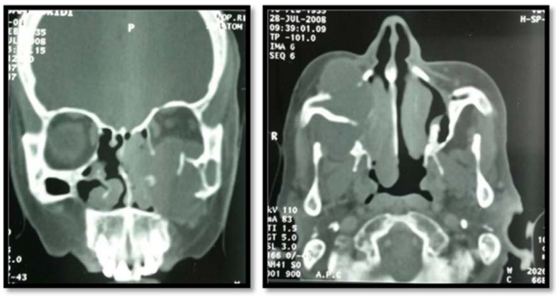

Figure 9: Axial and coronal CT scan imaging demonstrating a mucocele of the right maxillary sinus with important erosion of the adjacent walls and extension to the orbite and infratemporal fossa.

Figure 10: Coronal CT scan imaging showing a mass of the ethmoid with large erosion of the lamina papyracea and compressing of the orbite contents.

le, fronto-ethmoidal and fronto-ethmoido-maxillary mucocele.

*Concha bullosa was present in four patients presenting: Ethmoidal mucocele, Fronto-ethmoidal mucocele, Maxilary mucocele and Nasal cavity mucocele. (Figure 11)

*Hypoplastic frontal sinuses was noted in one case of ethmoidal mucocele.

*Agenesis of the frontal sinus in one case of ethmoidal mucocele. (Figure 12)

*Presence of a frontal cell (kuhn's cell) in a frontal mucocele. (Figure 13)

*Paradoxical middle turbinate in one patient having a fronto-ethmoidal mucocele.

*Septum deviation was observed in 17 patients and were obstructive in 10 cases.

CT scan provided also information about anatomic variants. We noted:

*Procidence of the canal of the carotid artery in four cases. It was bilateral in three cases (frontal, maxillary and sphenoidal mucocele) and unilateral in one case of fronto-ethmoidal mucocele associated with the presence of a sphenoid sinus septa attached to the artery canal.

*Procidence of the optic nerve canal in one case of a frontal mucocele.

*Asymmetry in the height of the ethmoid roof in two cases of a fronto-ethmoidal mucoceles.

MRI: MRI was realized for 15 patients in addition to the CT scan. It was indicated in four cases of sphenoid mucoceles (Figure 14) and in 11 cases presenting orbital or endocranial extension on CT scan.

Mucoceles appeared as a homogenous round mass who was in:

-Hypointense signal on T1-weighted images, hyperintense signal on T2-weighted images in nine cases.

-Hyperintense signal on T1-weighted and T2-weighted images in two cases.

-Hypointense signal on T1-weighted images, isointense signal on T2-weighted images in two cases.

Figure 11: Axial CT scan imaging showing a mucocele developing from a concha bullosa of the left middle turbinate.

Figure 12: Coronal CT scan shows ethmoidal mucocele associated with an agenesis of the left frontal sinus and a pneumatization of the right frontal sinus and the crista galli.

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Figure 13: Axial CT scan imaging showing a left frontal mucocele associated with a left frontal cell (Kuhn's cell).

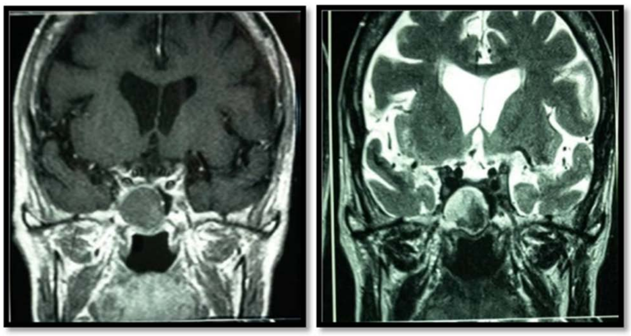

Figure 14: Coronal MRI imaging demonstrating a sphenoidal mucocele (isointense T1 weighted images and hyperintense T2 weighted images).

-Isointense signal on T1-weighted images, hyperintense signal on T2-weighted images in one case.

-Isointense signal on T1-weighted and T2-weighted images in one case.

In all cases, we have noted no enhancement after injection of gadolinium.

MRI allowed to study the extension of mucoceles to the neighboring organs especially orbital and endocranial ones. We observed that:

-The orbite contents was repulsed in two cases of fronto-ethmoidi-maxillary mucoceles and one case of fronto-ethmoidal mucocele. (Figure 15)

-Optic nerve was pushed in three patients who had respectively sphenoid mucocele, spheno-ethmoidal mucocele, fronto-ethmoido-maxillary mucocele.

It was stretched in one case of fronto-ethmoidal mucocele with important orbital extension.

-Endocranial extension was noted in three cases: Spheno-ethmoido mucocele, fronto-ethmoidal mucocele and fronto-ethmoido-maxillary mucocele. (Figure 16)

DISCUSSION

Paranasal sinus mucoceles are cystic masses filled with mucous and lined by respiratory epithelium which are capable of expansion by bone resorption and new bone formation.[1]

They occur principally in the third or fourth decades of life with a male predilection.[1]

Mucoceles result from the blockage of the sinus drainage secondary to inflammation, trauma, anatomical aberrations, tumours. chronic rhinosinusitis, allergic disease and craniofacial disease are the most common cause of mucocele formation.[2,3]

Symptoms depend on the location of the mucocele and may vary from rhinological, neurologic, or ophthalmologic ones.[1-3]

CT scan is considered to be the complementary method of choice in the investigation of mucoceles. It allows to evoke the diagnosis of the mucocele and determine its location, to study the neighboring structure (bone erosion, extension), to presume an aetiology of the sinus ostium obstruction and to research the anatomic variants of the sinuses.[3,4]

Mucocele appears on CT as a homogenous well circumbscribed expansible cyst involving one or much sinuses and

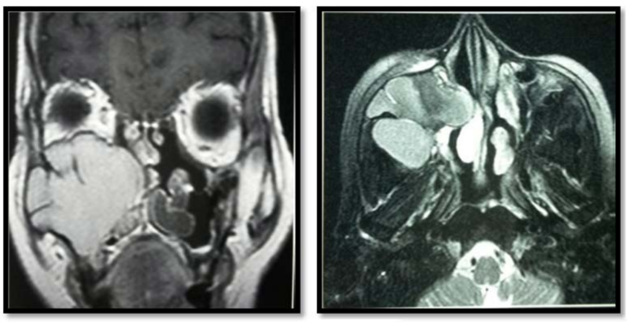

Figure 15: Axial and coronal MRI imaging showing a maxilary mucocele with extension toward the orbite and the infra temporal fossa.

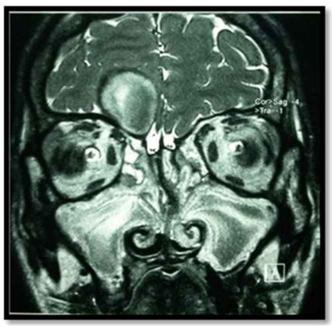

Figure 16: Coronal T2-weighted MRI showing a fronto-ethmoidal sinus mucocele with endocranial extension.

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may expand to neighboring structure. they may appear either hypo- or hyperdense.[4] This variable densities depend on their protein content and possible infection. Mucoceles do not show contrast enhancement centrally but a thin peripheral enhancement, can be seen and is suggestive of encapsulation.[5]

The differential diagnosis for mucocele is made with inflammatory, congenital cystic and neoplastic lesions of skull base, facial sinuses and nasopharynx.[4-6]

Mucocele location depends on anatomic conditions that favorise its formation. The frontal sinus is most commonly affected followed by the ethmoid sinuses, mucoceles occur in these locations in 70 to 90%. Sphenoid mucoceles are rare. The maxillary antrum is a relatively unfrequent site for mucocele formation, accounting for 10 percent or less of mucoceles.[7,8]

In our study, mucoceles were seen frequently in fronto-ethmoidal sinus followed by ethmoidal location then frontal sinus.

The neighboring bone structure is remodeled with areas of thickening and erosion.[2,4,5] In some areas of greater fragility, we may observe herniation into adjacent structures such as the orbite and the endocrine.[4,9]

CT scan provides also information about existence of factors favorising mucoceles formation as: Osteoma, Fibrous displasia, Facial traumatism, Sinonasal polyposis. It can visualizes anatomic abnormalities that may cause a blockage of sinus drainage.[10-12] For our patients, CT scan has allowed to evoke the cause of mucoceles in some cases as adhesions, concha bullosa, septum deviation, paradoxical middle turbinate.

Furthermore, this exam is essentially to define anatomic variants prior to surgery.[13]

MRI is an excellent exam in mucocles. It allows to differentiate and to assess any extension into the orbit or intracranial compartment, but, unfortunately it can't study bone detail.[14,15]

Its indications are in complement of the CT scan in case of diagnosis doubt in CT scan between mucocele and other tumors or inflammatory lesions, in case of sphenoid location and in endocranial or orbital extension.[14]

The MRI appearance of paranasal sinus mucoceles is quite variable, depending on the composition of the mucocele.[16] The usual signal characteristics are a low intense T1 and a high intense T2 but any combination of signal intensity may be seen depending on the presence of blood products or the degree of hydration of the contents.[5,14,17,18] The appearance consisted of hyperintense signal on both T1 and T2 type images, corresponding to more hydrated secretions, which are also high in protein content.[17-20]

We have noted variable presentation of mucocele in MRI from hypo to hyperintensity T1 and T2 but in all cases, we have noted no enhancement after injection of gadolinium.

CONCLUSION

The presentations of mucoceles on imaging are quite variable depending on its contents. Practicians should know the typical as well as atypical findings. Special care should be taken with regard to differential diagnosis and associated cases.[16]

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

CONSENT

As our article is a retrospective study and did not publish any personal photo or information regarding any of the patient in our manuscript thus the consent is not required for the article publication.

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