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Otolaryngology International

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Otolaryngology International
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Engaging Doctors in Tobacco Control

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Introduction

For nearly a half century we have been struggling with the 20th century's brown plague: tobacco use. As we began this new century, we faced both a grim forecast, and a new hope. The grim forecast - This voracious devourer of health and life threatens hundreds of millions of new victims, especially in the developing world. The source of hope - We have now learned through our failures as much as our successes how to fight against tobacco. These lessons were hard won. At first, we believed that the verdict of science, and public awareness of that verdict, would compel tobacco users to quit, and governments to take appropriate action to control tobacco use. But we were wrong. We did not, could not, imagine the depths to which the international tobacco industry would descend to deny, deceive, bully, undermine, and confuse public understanding and government action.

Tobacco is the leading preventable cause of death and more than five million people die globally from the effects of tobacco every year-more than that of HIV/AIDS, malaria and tuberculosis. Tobacco is a serious threat to health and a proven killer and ranks second as a cause of death in the world. Tobacco use is an emerging pandemic marching forward relentlessly. Evidences accumulating since early 1950s indicate that more than 25 diseases are now known or strongly suspected to be causally related to smoking. WHO estimates that unless current smoking pattern is reversed, tobacco will be responsible for 10 million deaths per year, by the decade 2020-2030, with 70% of them occurring in developing countries. In India tobacco kills 8-10 lakhs people each year and many of these deaths will occur in people who are very young. Deaths attributable to tobacco are expected to rise from 1.4% of all deaths in 1990 to 13.3% in 2020. Currently about one- fifth of all worldwide deaths attributed to tobacco occur in India, more than 8,00,000 people die and 12 million people become ill as a result of tobacco use each year in India, as per WHO projection and will have the highest rate of rise in tobacco-related deaths during this period compared to all other countries/ regions. Youth in general and adolescents in particular fall prey to this deadly habit with severe physical, psychological, and economic implications. Among the youth, students are particularly involved due to increasing academic pressures and uncertain career. Encouragement from peer group, the lure of popularity, and easy availability of tobacco in different forms make a teenager an easy prey. In India, approximately 5500 children and adolescents start using tobacco products daily, some as young as 10 years. The majority of users have first used tobacco prior to the age of 18 years. It has been reported that there is an increased trend of tobacco habits among the healthcare students pursuing healthcare education like any other youths. Little attention has been given to the context of when and how healthcare students undergo attitudinal and behavioral changes with respect to their own smoking habits Teaching about the effects of use of tobacco is essential for medical college students, because these would be physicians, future teachers, and responsible citizens and will hold, key positions to lead tobacco cessation programs in our community. So they should not be sanctimonious. Health professionals serve as role models for healthy behavior to the public. During routine visits, health professionals can counsel patients on dangers of smoking and the importance of quitting; and such counseling is one of the most cost-effective methods of reducing smoking. The medical students who are future health professionals ignore epidemiological evidence and continue to use a substance that is harmful to health. Moreover, health professionals, who happens to be health promoters/educators and health role models

for society in future can have negative impact on society if they themself are involved in tobacco consumption practice . In developing nations where reductions in tobacco use have not been realized, it is critical that health professionals be encouraged to abstain from tobacco use. Data on tobacco use among health professionals in India are limited.

Health Professionals Against Tobacco

The detrimental effects of smoking constitute one of the most urgent health problems . Individual doctors have a clear duty and responsibility toward their patients in this respect . Campaigns to inform the public of the damage to health caused by smoking will not be convincing if doctors as individuals and as a profession are seen as smokers. Doctors are in a unique position to help their patients stop smoking because their advice on health matters is trusted more than anyone else. Many smokers want to stop smoking, and others may be receptive to encouragement to stop. A brief intervention by a doctor has been shown to increase the chances that a smoker will successfully stop smoking. In smoking cessation, the decisive value is assigned to the physician's approach and his/her assistance, mere advice from the physician not to smoke increases the likelihood of successful quit rate in the patient by about 10%. Doctors need to understand that their interventions can have a powerful impact, not only in cessation counseling and treatment, but also in policy advocacy.

- Evidence shows that doctors remain among the most respected and trusted community voices on matters related to health.
- An oncologist may save more lives by counseling for tobacco control for few hours than by treating lung cancer patients for a lifetime.

What should be our goal?

Our goal should be to motivate more doctors to become active in the full range of tobacco control activities.

According to *Doctors and Tobacco: Medicine's Big Challenge,* by David Simpson, medical professionals probably have the greatest potential of any group in society to promote a reduction in tobacco use, and thus, in due course, a reduction in tobacco induced mortality and morbidity. Unfortunately, too many doctors in too many countries do not consider tobacco control to be part of their professional responsibilities. Instead they simply treat the illnesses tobacco use causes. They have a unique potential to contribute to tobacco control in several complementary ways:

- As role models in not smoking, or quitting smoking.
- In counseling patients not to smoke.
- In providing smoking cessation treatment.
- In organizing and speaking out publicly and lobbying for comprehensive public policies to control tobacco use.

In countries with the highest smoking rates, doctors smoke even more than the general public and, as a result, serve as negative role models. Getting doctors to quit smoking can have profound effects on tobacco control. They become positive role models for patients and are far more likely to advocate for tobacco control than those who still smoke. In those nations in which the tobacco epidemic appears to have peaked and begun to ease, a retrospective view reveals that it was, in nearly all cases, physicians who led the way by changing their behavior from being one of the groups with the highest smoking prevalence to being one of, if not the, lowest. Therefore, in any nation where the tobacco epidemic has not peaked, or has not yet taken hold, focusing on reducing smoking among physicians and involving them in tobacco control activities by appealing to and educating them personally and through their medical societies may be the most important action a national tobacco control movement can take.

Who do we need to persuade in order to motivate doctors to become more engaged in tobacco control?

Among doctors, two key target audiences are medical students, who are among the most open to a new understanding of doctors responsibilities. Doctors who quit are more likely to become engaged in both cessation counseling and advocacy. Our target audience should include those who have the most professional influence with doctors, including deans of medical schools, faculty, and other respected, award-winning doctors and medical scientists. Even in countries without active tobacco control efforts, many doctors are informed of smoking's health risks but often fail to take responsibility for tobacco control. They do not recognize that their professional responsibility extends beyond the treatment and cure of tobacco-caused disease to include prevention and cessation. Emphasis should be to encourage medical professionals to give up smoking and to embrace the key role they play in helping others curtail their tobacco use.

According to the World Health Organization (WHO)

" Health professionals are encouraged to

personally exhibit and promote a tobacco-free lifestyle. The advice and treatment given by health professionals can be a major factor in whether or not a person tries and succeeds in quitting smoking."

According to the WHO's Tobacco Free Initiative (T.F.I.)

"Health workers function as exemplars and educators for their patients, and consequently should set an example by abstaining from tobacco. When this point is emphasized in professional organizations and through the education system that trains professionals, their tobacco use rates decline. If health professionals and researchers focus as much on efforts to prompt attempts at tobacco cessation as on creating new approaches to treatment, many additional tobacco users will be motivated to quit."

- Doctors should avoid smoking and the use of tobacco products in their personal lives.
- Assess and document smoking and tobacco use status as part of the medical history for every patient.
- Provide cessation counseling and other proven therapy to all patients who use tobacco.

Doctors need to hear messages from other doctors who are already active in tobacco control.- "Doctors believe doctors."-

Doctors will be receptive to messages that come from other medical professionals, medical societies, and other leaders of the medical community.

- Doctors who have already become tobacco control advocates are perhaps the most powerful messengers.
- Medical school professors have a prime opportunity to educate young doctors about the hazards of tobacco use. Their curricula can introduce prospective doctors to tobacco control activities and can make them aware of their obligation to participate as members of the medical profession.
- Leading physicians have access to the mass media as guest experts on news programs and talk shows or with their own health guidance programs. They can use these media opportunities to encourage their colleagues to get involved in tobacco control.
- Medical societies / Associations are in a unique position to influence the behavior of their members. They can:
- Conduct surveys of their members, which would include questions about their smoking patterns, the extent to which they provide tobacco use counseling and cessation treatment, and their willingness to become engaged in tobacco control advocacy.
- Organize plenary speakers and panel discussions on Tobacco control at conferences and workshops.
- Place regular articles in the association's journal on the risks of smoking.

Medical Schools

Medical schools have a critical opportunity to educate and motivate emerging doctors. Medical school deans and professors can take several complementary approaches:

- Courses in tobacco control treatment can be offered and even mandated within the curriculum.
- Tobacco control responsibility can be incorporated into orientation lectures and brochures for incoming students.
- Medical school students can be recruited for diverse tobacco control projects, from working in cessation clinics to conducting surveys of tobacco use by doctors in medical facilities.
- Professors can use their status to speak out about the importance of tobacco control.

Others

Voluntary organizations, NGOs dedicated solely to tobacco control, and other national NGO coalitions have played, and can continue to play, a strong role in recruiting doctors to support tobacco control. They can:

- Develop and maintain a list of doctors who are active in tobacco control and who will recruit other doctors, lecture at medical forums, and speak with the media about the need for doctors to be engaged.
- Hold workshops for doctors.
- Help organize talk show debates about doctors' responsibilities in Tobacco control.
- Encourage medical leaders to write letters and editorial articles to newspapers.
- Promote newspaper editorials about the connection between medical ethics and doctors' tobacco control responsibilities.

Appeal

By taking advantage of this unique opportunity , we want to ensure that more doctors and prospective doctors will join in the fight to eradicate tobacco use as an international public health pandemic.

Original Article

Otolaryngology International
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Role of FNAC in Diagnosing Tubercular Lymphadenitis: Our Experience

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Abstract

Aim: To study the accuracy, advantages and limitations of FNAC in diagnosing tubercular lymphadenitis in cases of cervical lymph node swellings. *Materials and Methods*: Retrospective study was conducted over a period of 1 yr which included 72 patients who presented with lymph node swelling in the neck to the ENT outpatient department in our institute.Both males and females from all age groups were included in the study. In all 72 patients FNAC was done. Out of the 72 patients 28 patients agreed to undergo excision biopsy and their histopathological results were compared with the preoperative FNAC results in order to determine the accuracy of FNAC in correctly diagnosing tubercular lymphadenitis in particular. As 28 patients underwent excision biopsy following FNAC, the study group included the above 28 patients only. *Results:* Out of the 28 patients, incidence of males was slightly higher than females and all age group were included, the youngest was a 12 yr old and the oldest was 75 yr old. Out of the 28 patients with lymph node swelling, there were 10 tubercular lymhadenitis, 9 chronic non specific lymphadenitis, 5 metastatic carcinoma and 4 lymphomas proved hystologically. The diagnostic accuracy of aspiration cytology was 100% for tubercular lymphadenitis with all the 10 cases proved positive by FNAC. *Conclusion:* FNAC is a simple, quick, economical and least traumatic procedure that can be easily carried out on outpatient basis with high degree of accuracy. As the sensitivity of FNAC in diagnosing tubercular lymphadenitis is extremely high and in our study 100%, this investigation is alone enough to start the patient on medical line of treatment for tuberculosis which helps in reducing the inpatient burden for diagnosing the disease especially in counties like ours where the incidence of tuberculosis is still remains high.

Keywords: Fine Needle Aspiration Cytology (FNAC); Accuracy; Excision; Tuberculosis.

Introduction

Tubercular lymphadenitis is a very common cause of superficial cervical lymphadenopathy in our country. FNAC has evolved as one of the most cost effective investigations to diagnose tubercular lymphadenitis. It provides an alternative to excision biopsy for lymph nodes and is an easy procedure for collection of material for cytomorphological and bacteriological examination [1].

The well defined role of FNAC in investigation of lymph node has previously been studied [2,3]. Our study aims at determining the accuracy of FNAC in diagnosing tubercular lymphadenitis in particular by comparing the cytological findings with post operative histopathological findings. FNAC does not give the same architectural detail as histology but it can provide cells from the entire lesion, as many passes through the lesion can be made while aspirating [4].

Materials and Methods

A retrospective study of 72 patients who presented with neck swelling to the ENT outpatient department in our institute over a period of 1 yr was done. Detailed clinical history was taken and examination done and all the 72 patients were subjected to FNAC.

FNAC was done using 22-23 gauge, 3-5cm long needle with 10 ml syringe. All 72 patients were advised to undergo excision biopsy so that the cytology report could be compared with the histopathology report and thereby the accuracy of FNAC could be determined.

However out of the 72 patients 58 patients were medically fit to undergo excision biopsy but out of this only 28 patients were willing to undergo the procedure. Therefore the histopathology and FNAC report were compared in these 28 patients.

Result

Study results compared the preoperative FNAC report with the post-operative histopathology report in 28 patients who underwent excision biopsy. Patients were in the age group between 10 to 80yrs (Table 1). Youngest patient was 12 yrs old who had tubercular lymphadenitis and oldest patient was 75yrs who had chronic non specific lymphadenitis.

On Clinical Examination

Only 6 out of 28 patients had tenderness over the lymph node swelling. On palpation 16 neck nodes were firm, 5 were hard and 4 rubbery in consistency. Another 3 showed variable consistency.

Classification of Cytology Reports (Table-3 & Fig 1)

1. In cases where the FNAC features were similar to HPE report –these were cosidered Positive.
2. In cases where the cytological features were suspicious of a lesion which was confirmed by histopathology these were cosidered as Doubtful or Inconclusive.
3. In cases where the cytological features did not correlate with histopathological features these were considered as Negative.

Diagnostic Accuracy of FNAC in Lymph Node Swellings (Table 4 & Fig 2)

Out of 28 lymph node swellings, there were 10 tubercular lymphadenitis 9 chronic non specific lymphadenitis, 5 metastatic carcinoma and 4 lymphomas proved histologically. The diagnostic accuracy of aspiration cytology was 100% for tubercular lymphadenitis with all the 10 cases proved positive by FNAC.

False Negative Report

Only in 1 case false negative report was obtained by FNAC. Metastatic squamous cell carcinoma was reported as reactive node. Thus the percentage of false negative report was as low as 3.57%.

Inadequate Smears:

Smears in which only blood or clear fluid without any cells was seen even after repeated aspiration were considerd as inadequate smears. In our study only in 1 case inadequate smear was reported.

Table 1:

Age in years	Number of cases with cervical lymph node
11-20	6
21-30	7
31-40	4
41-50	3
51-60	4
61-70	3
71-80	1
Total	28

Table 2:

Sex	Number of patients	Percentage
Male	15	53.57%
Female	13	46.43%
Total	28	100%

Males were slightly more in number than females (table 2)

Table 3:

Neck swelling	Number of cases who undervent both FNAC and Excisio biopsy	Positive	Doubtful	Negative
Lymph node	28	20 (71.43%)	6 (21.43%)	2(7.14%)

Table 4:

Types of Lymph node swelling	No. of cases Histopathologically proved	No. of case proved positive by FNAC	Percentage
Tubercular lymphadenitis	10	10	100
Chronic non specific lymphadenitis	9	7	77.78
Metastatic carcinoma	5	2	40
Lymphoma	4	1	25
Total	28	20	71.43

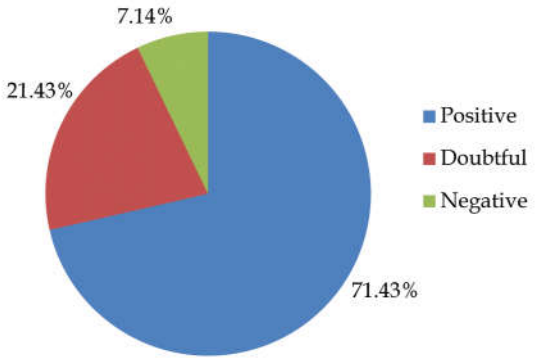

Fig. 1:

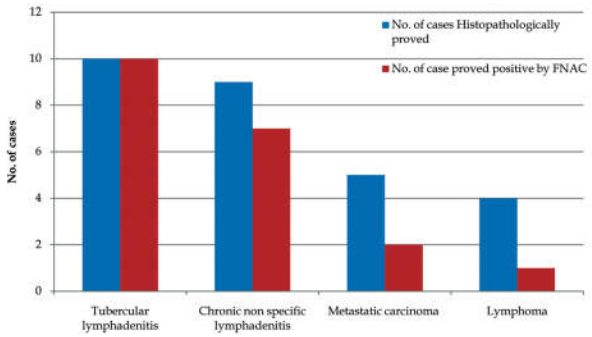

Fig. 2:

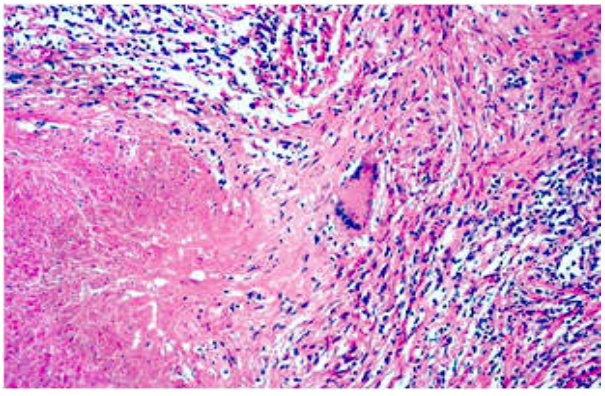

Fig. 3:

Inconclusive Smear

FNAC suspicious of a lesion but not definite and the suspicion proved correct after histopathology.

In 6 cases inconclusive smear was reported in our study-1 chronic non specific lymphadenitis, 2 metastatic carcinoma and 3 lymphomas.

Discussion

Cervical lymphadenopathy is a very common clinical finding and its etiology can be suspected by clinical signs and symptoms. However in suspected cases of tubercular lymphadenopathy a morphological diagnosis is essential to start antitubercular treatment. FNAC thus has contributed in a big way in diagnosing tubercular lymphadenitis as it is a simple to perform, non invasive inexpensive test with high sensitivity in diagnosing tubercular lymphadenitis and can replace excision biopsy, especially in developing country like ours where the burden of disease is still high.

With prevalence as high as 1.5% [5], tuberculosis is still rampant in this part of the world and the tuberculosis involvement of lymph nodes is the most common form of extrapulmonary tuberculosis responsible for 30-40% of cases [6]. FNAC has contributed a great deal to transform cytology from primarily a screening tool to a powerful diagnostic technique [7].

FNAC not only offers tissue diagnosis but serves as a preliminary screening procedure for a number of clinical considerations example lymphoma, leukemia, metastasis, tuberculosis and lymphadenopathy not othewise specified [8,9,10].

In our study out of 28 cases of lymph node swelling who underwent FNAC and excision biopsy, 10 cases of tubercular lymphadenitis, 9 nonspecific lymphadenitis, 5 metastatic carcinoma and 4 lymphomas were proved histologically. The diagnostic accuracy of aspiration cytology was 100% for tubercular lymphadenitis with all 10 cases proved positive by FNAC thereby none of the tubercular lymphadenitis cases were misdiagnosed or left undiagnosed. Most common cytological pattern was epitheloid clusters with or without langhan's giant cells with necrosis (Figure 3), which is similar to the study by Gupta et al [11]. Only one case out of 28 was reported as false negative by FNAC which was a case of metastatic squamous cell carcinoma and was reported as reactive node.

1- Tubercular granuloma

2 – Lymphocytes)

The diagnostic accuracy of lymph node swellings of neck varies with different authors. Janes Thamsen et al (1973) reported 90% accuracy in 47 cases, S.K Lau et al (1990) reported 93% accuracy in 68 tubercular lymphadenitis, Yadav S.P.S et al (1991) reported 89.8% accuracy in 50 cases.

In our study, the results are extremely encouraging as diagnostic accuracy of lymphadenitis is 89.47% and all the 10 tubercular lymphadenitis cases were reported positive by needle aspiration making it 100% accurate.

Conclusion

FNAC is a safe, well accepted procedure by patients, very cost effective and requires minimum instrumentation in comparison to excision biopsy. Diagnostic accuracy is 100% in tuberculous lymphadenitis and therefore in most remote areas FNAC can be used for diagnosing tubercular lymphadenopathy accurately.

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Case Report

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Neurofibroma of Nasal Cavity

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Abstract

Neurofibromas are benign peripheral nerve sheath tumors which originate from Schwann cells. They are known to occur in extremities as a local manifestation of Von Recklinghausen's disease, but isolated neurofibroma occurring in the nasal cavity is an rare entity. Here we report one such case in a 44 yr old man who, presented with a unilateral nasal mass. The mass was histologically confirmed to be a neurofibroma. The clinical presentation, histological features and management of nasal neurofibroma are discussed.

Keywords: Neurofibroma; Schwannoma; Endoscope.

44 yr old male presented with history of mass left nasal cavity for 1 year. On examination there was obliteration of left alar groove. A whitish firm mass filling the nasal cavity was seen which appeared to arise from left lateral nasal wall was sensitive to touch and there was no change in size on vasoconstriction. General examination did not reveal any abnormality.

CECTPNS (Figure 1) showed heterogeneously enhancing mass lesion completely filling the left nasal cavity with dimensions of 14mm*38mm* 59mm. There was opacification of the left maxillary, frontal, ethmoid and sphenoid sinuses. Mass effect was noted in the form of focal indentation and bulge of the nasal septum to the right. No gross bony erosion/sclerosis was seen around the mass. Investigations endoscopic biopsy was suggestive of neurofibroma.

Patient was planned for excision of mass via a lateral rhinotomy incision approach. Flaps were elevated and the left wall of maxilla was exposed. Medial maxillectomy done by taking cuts medial to inferior orbital foramen and along the nasal floor anterior wall of left maxilla was drilled out. Mass was visualised in left maxillary antrum (Figure 2). Mass was carefully dissected from its attachments along the roof and floor of maxillary antrum. Mass was removed in toto after freeing from attachments along ethmoids, sphenoid. Using a 30 degree endoscope through the incision entire cavity was inspected. Small mucoceles were found and drained from posterior ethmoid and sphenoid sinuses. Sphenoidotomy was done and left frontal recess cleared. Left nasal packing was done. Incision was closed in layers. His postoperative recovery was uneventful.

Histopathology showed soft tissue mass 5.5*4.5*1cm (Figure 3) with a smooth and shiny external surface with multiple areas of haemorrhage. Histopathology sections (Figure 4) showed cells are arranged loosely and diffusely composed of spindle shaped cells with wavy comma shaped nuclei scant cytoplasm. The nuclei are seen separated by collagen fibres and myxoid material. No mitotic activity, verocay bodies, palisading of nuclei or hyaline thickening of vessel wall was seen. No atypia/ granuloma seen. Opinion was of Neurofibroma left nasal cavity. On IHC staining S100 was positive.

Patient was kept under regular nasal endoscopic followup for a period of 2 years and no recurrence was noted.

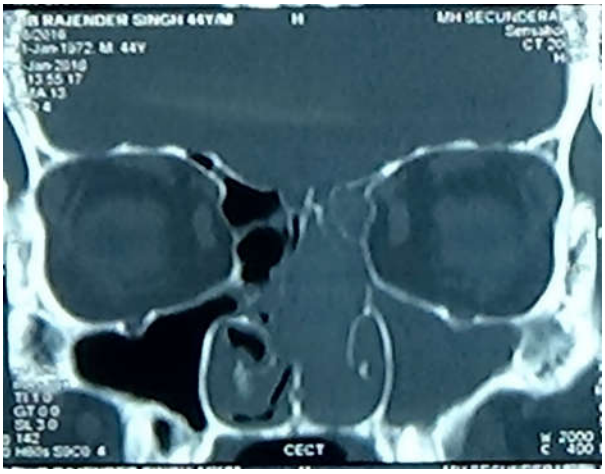

Fig. 1: CT scan PNS showing opacification of left maxillary sinus and nasal cavity

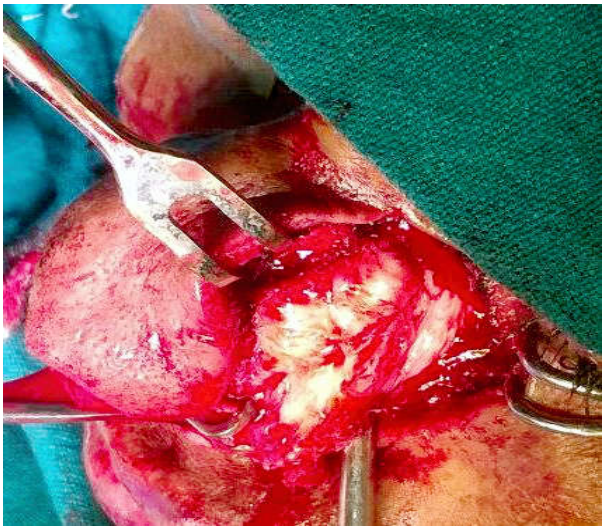

Fig. 2: Exposure of the neurofibroma using the lateral rhinotomy approach

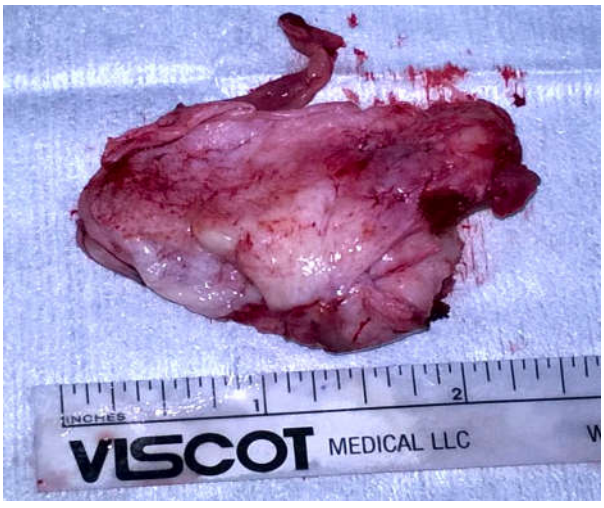

Fig. 3: Photograph of the excised specimen

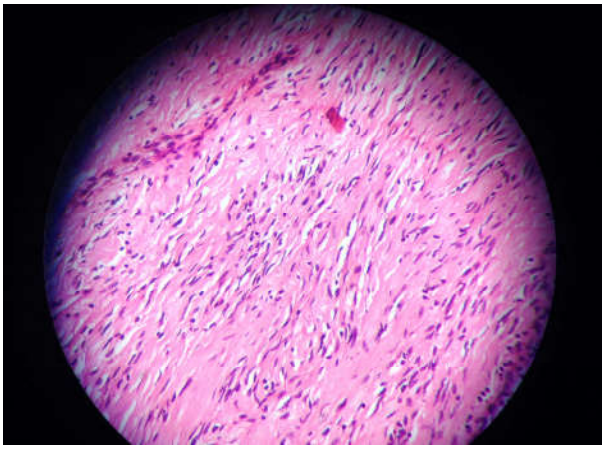

Fig. 4: High power H&E Stain of the neurofibroma showing characteristic spindle cells

Discussion

Neurofibromas are benign peripheral nerve sheath tumors that originate from the nonmyelinating Schwann cells which are derived from neuroectoderm [1]. Neurofibromas can be categorized into dermal and plexiform subtypes. Dermal type is associated with individual peripheral nerves and plexiform types are usually associated with many nerve bundles [1]. The plexiform tumors can rarely undergo malignant transformation [1].

Neurofibromas are usually found in individuals with neurofibromatosis, which is an autosomal dominant condition. There are two types of neurofibromatosis: type 1 (von Recklinghausen disease), which is more common, and type 2, which typically has a more severe course due to central nervous system tumors [2].

Occassionally an isolated neurofibroma can occur without being associated with neurofibromatosis in the gastrointestinal system [3] and very rarely they have been reported to occur in the paranasal sinuses. They have a malignant potential of 2.6% and if associated with von Recklinghausen's disease, the chances for malignant transformation are 3-15%. Neurofibromas commonly occur in third or fourth decades of life.

25% to 45% neurofibromas arise in the head and neck region of which only 4% involve the nasal cavity and paranasal sinuses [4]. Neurofibromas have been reported to occur in the nasal cavity [5], ethmoid sinus, maxillary sinus and sphenoid sinus. In the area of the nose and paranasal sinuses neurofibroma arise from the first and second division of trigeminal nerve and the autonomic plexuses.

Neurofibromas are slow growing benign tumors, however can become very large leading to

compression of local structures, including expansion and erosion of adjacent bone and bone resorption [4]. The clinical features of a neurofibroma depend on the site of the tumor and subsequent involvement of surrounding structures [4]. Imaging for these tumors include CT scanning and magnetic resonance imaging (MRI) to demonstrate the extent and involvement of the tumor. MRI allows better differentiation of the tumor from adjacent soft tissues and better evaluation of any intracranial and intraorbital extension [4] whereas CT scan is useful for bone destruction. The differential diagnosis of neurofibromas include benign tumors of nose like fibroma, papilloma, leiomyoma, and schwannoma [4,5].

The characteristic histologic features of neurofibromas are spindle cells with wavy nuclei, and wavy collagen fibrils. There is a 10% malignant transformation reported in neurofibromas hence it is essential to distinguish it from schwannoma and malignant peripheral nerve sheath tumor [6]. While schwannomas have a capsule; neurofibromas are generally not encapsulated and usually interdigitate with adjacent tissue. In neurofibromas axons are seen to transverse the tumor mass whereas it is not so in schwannomas. Immunohistochemistry is useful in differentiation as neurofibromas show reaction with S-100, NSE Neuron specific enolase and Vimentin [7] but not for desmin or smooth muscle actin which helps in differentiating between neurofibroma and other tumors.

The mainstay of the treatment is complete surgical excision because neurofibromas may infiltrate extensively. The type of approach for removal is dependent on the extent and location of the tumor. Transnasal endoscopic resection can be done if neurofibroma is solitary and located in the nasal cavity and if the origin can be identified. It is important to visualise the origin of the tumor which allows macroscopic complete resection under endoscopy. Open surgical procedures such as lateral rhinotomy, extended ethmoidectomy or partial maxillectomy are required for extensive tumors.

Recurrences are infrequent however rarely tumors with locally invasive tendencies are known to recur after incomplete removal[4]. With a 2-year follow-up, no recurrence was noted for the present case.

Conclusion

A rare case of isolated neurofibroma of the nasal cavity is reported which presented as a unilateral nasal mass and was diagnosed histopathologically as neurofibroma of the nasal cavity. Immunohistochemistry may play an important role in diagnosing nasal neurofibromas.

Conflicts of Interest

The authors have no conflicts of interest to declare

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Case Report

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Fibrous Dysplasia of Temporal Bone with Secondary Cholesteatoma: A Rare Presentation

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Abstract

Fibrous Dysplasia is a benign, non-familial condition in which normal bone is replaced by fibrous tissue and woven bone. It rarely effects temporal bone. The diagnosis is usually based on clinical, radiographic, and histopathologic features. We here present a case of secondary cholesteatoma caused by external auditory canal occlusion by fibrous dysplasia which was successfully treated by radical mastoidetomy and partial cavity reconstruction by muscle periosteal flap with adequate meatoplasty.

Keywords: Fibrous Dysplasia; Monostotic; Polystotic; Temporal Bone.

Introduction

Fibrous dysplasia (FD) is a congenital skeletal disorder characterized by thinning of the cortex and replacement of the marrow with fibrous tissue that demonstrates characteristic ground-glass appearance on radiography with X- Rays and High Resolution Computerized Tomographic (HRCT) scanning. Von Recklinghausen first recognised fibrous dysplasia as a clinical entity in 1891. The term "fibrous dysplasia" was coined by Lichtenstein in 1938, although, it was first described by McCune and Bruch in 1937. There exists a mainly self-limiting form of fibrous dysplasia classified as monostotic (unifocal), which is characterized by dysplastic bone in a single location and a polyostotic (multifocal) form, which can exhibit aggressive growth placing adjacent structures at risk for compressive sequel.It may sometimes present as McCune-Albright syndrome in which it is accompanied by precocious puberty, endocrine disorders and ''cafe au lait'' skin pigmentation [1,2]. The preferred sites include the diaphyses and metaphyses of long bones, ribs, pelvis, shoulder and craniofacial skeleton. The lesions have been reported to account for 2.5% to 7.0% of all benign bone tumors, with an equal predilection for both sexes [3-5]. In head and neck region, the skull and facial bones are involved in 10-25% cases of monostotic and in 50% cases of polystotic FD. Out of all craniofacial involvement temporal bone is involved in 24% of cases [6,7].

The numbers of case reports of fibrous dysplasia of temporal bone (FDTB) with secondary cholesteatoma are very limited. A fewer cases have been reported from India. We here report a case of FDTB presenting with cholesteatoma and subsequent hearing loss.

Case Report

A 30-year old male presented in outpatient department with the history of canalplasty for fibrous dysplasia in right ear about 12 years back. The patient had lost the complete records of previous treatment. After surgery patient was apparently fine for about 2 years after which he developed complaints of gradually progressive loss of hearing, pain and discharge from right ear. There was no

history of tinnitus, vertigo, headache, facial asymmetry, swelling anywhere else in the body or any other neurological deficit. On external examination of the ear the pinna, pre and post-aural regions were normal. On otoscopic examination, a bony hard swelling was seen obliterating the right external auditory canal. Left ear was normal. HRCT scan of temporal bone revealed the characteristic ground glass opacification mainly of squamous and mastoid portion of the right temporal bone, and to a lesser extent the petrous part. This appearance was consistent with that if fibrous dysplasia. The bony labyrinth was normal. The condition did not involve any other bone of skull. The cortex appeared thinned out and external auditory canalwas obtruded by the bony mass. The canal medial to the obstruction, the middle ear cavity, aditus and the antrum were all expanded, their landmarks were distorted and they were occupied with a soft tissue mass. No ossicles were seen except the head of malleus [Figure 1, 2, 3]. Pure tone audiogram showed severe mixed hearing loss with air-bone gap of 54 decibel in right ear. In left ear hearing was normal. All other haematological and biochemical investigations were normal.

Patient's consent for surgery was taken after he was explained prognosis and the possibility of recurrence. Patient was taken up for surgery by post-auricular approach. EAC was obstructed by a bony hard swelling. Skin over the swelling was raised and the bone underneath was drilled. Medial to the bony swelling, cholesteatoma sac was found which eroded the posterior canal wall and extended into antrum, attic, peri-sinus air cells, hypotympanum, Eustachian tube area and peri-facial cells [Figure 4].

All surgical landmarks in the middle ear appeared displaced. Roof and floor of canal, tegmen tympani, and anterior wall of middle ear were pushed and partially eroded. Only head of malleus was found in attic, incus was completely eroded and stapes footplate present. Mastoid cavity and posterior wall reconstruction was done with inferiorly based musculo-periosteal flap (Singapore flap). Temporalis fascia was placed over foot plate of stapes and type III tympanoplasty was done. Total ossicular replacement was not tried at this stage keeping in mind the extent of cholesteatoma and chances of its recurrence. Wound was closed in layers and post-op period was uneventful. Some of the bone from cortex was sent for histopathologic evaluation which revealed irregular trabeculae of woven bone intermixed with a connective tissue stroma in haematoxylin-eosin, which is consistent with fibrous dysplasia [Figure 5].

Post-operative period was uneventful and patient was discharged on seventh day in a satisfactory condition. After 6 weeks fallow up, post-operative cavity was well epithelised [Figure 6], there was only mild hearing improvement, though the remnant air

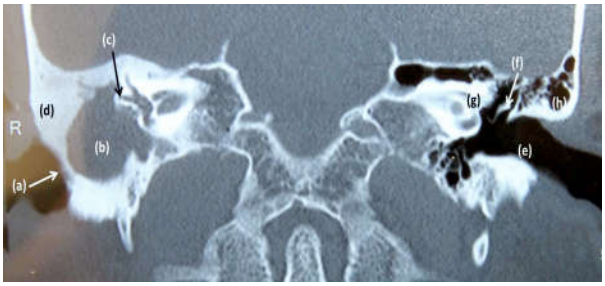

Fig. 1: CT Scan in coronal section showing **(a)** Obstructed external auditory canal of right ear, **(b)** Cholesteatoma cavity, **(c)** Right lateral semicircular canal, **(d)** Fibrous dysplasia in right temporal bone, **(e)** Patent external auditory canal of left ear, **(f)** Incus, **(g)** Cochlea, **(h)** Normal air cells in temporal bone

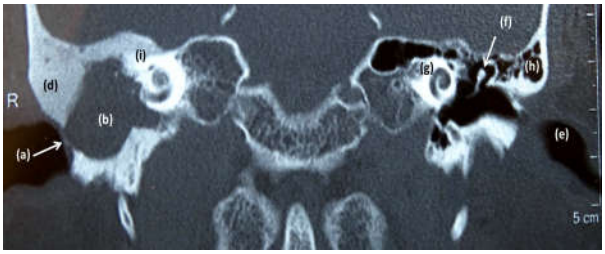

Fig. 2: CT Scan in coronal section showing **(a)** Obstructed external auditory canal of right ear, **(b)** Cholesteatoma cavity, (c) Right lateral semicircular canal, **(d)** Fibrous dysplasia in right temporal bone, **(e)** Patent external auditory canal of left ear, **(f)** Incus, **(g)** Cochlea, **(h)** Normal air cells in temporal bone

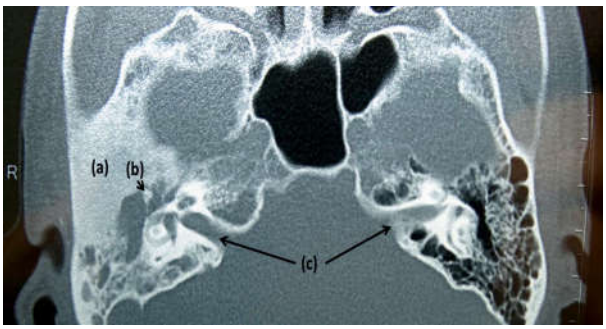

Fig. 3: CT Scan in axial section showing **(a)** Fibrous dysplasia in right temporal bone , **(b)** Head of malleus in attic, **(c)**Internal auditory meatus

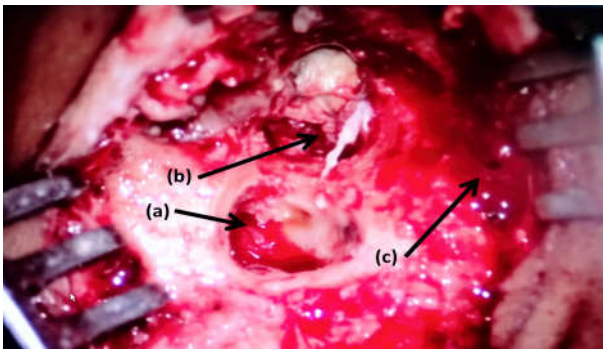

Fig. 4: Per-operative picture showing **(a)** Mastoid Antrum, **(b)** Cholesteatoma in external auditory canal and middle ear, **(c)** Tip of mastoid

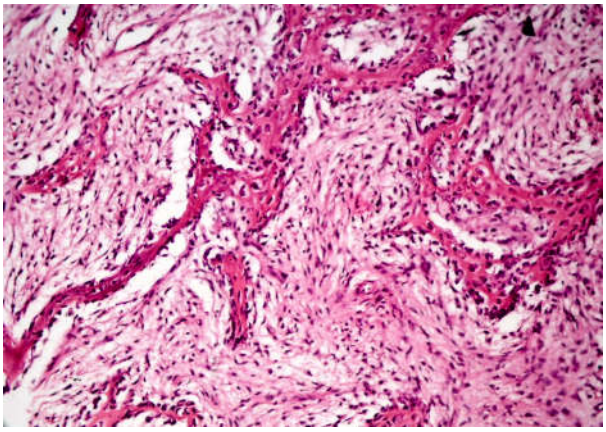

Fig. 5: Histopathological picture showing irregular trabeculae of woven bone intermixed with a connective tissue stroma (haematoxylin-eosin, original magnification X 40)

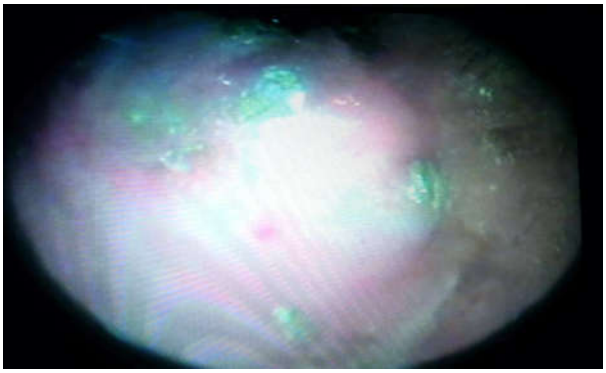

Fig. 6: Well-epithelised post-operative cavity

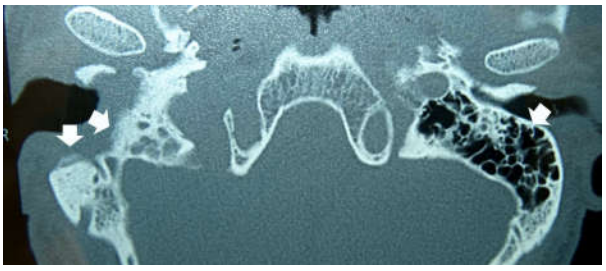

Fig. 7: Dissected mastoid cavity (compare white arrows on both sides) with soft tissue reconstruction of posterior wall

bone gap persisted. CT scan performed after surgeryshowed patent external auditory canal [Figure 7]. Patient was asymptomatic in follow-up forup to 6 months.

Discussion

FD exists in three forms out of which monostatic variety accounts for about 75-80%, polyostotic for about 20-30%, and McCune Albright Syndrome for about 3% cases. Out of all the craniofacial involvement, the ethmoids were the most commonly involved (71%), followed by the sphenoid (43%), frontal (33%), maxilla (29%), temporal (24%), parietal (14%), and occipital (5%) bones [7]. The temporal bone may occasionally be involved, and it may be the site of monostotic, less frequently, of polyostotic FD. The underlying defect in fibrous dysplasia is a mutation of the *GNAS1* gene, which leads to constitutive activation of gene products that preclude the maturation of osteoprogenitor cells and lead to development of abnormal bone matrix, trabeculae, and collagen, produced by undifferentiated mesenchymal cells. Although malignancy is rare (<1%), there is a risk of sarcomatous degeneration, which is increased by exposure to ionizing radiation [1].

The diagnosis of FD is usually based on clinical, radiographic, and histopathologic features. Clinically, the most common presentation is swelling; other manifestations include weakness, localized pain, deformity, fractures, and compromised vision or hearing. Involvement of the temporal bone results in painless progressive enlargement of the squamosal and mastoid, which may manifests as progressive conductive hearing loss due to stenosis of the external auditory canal, sometimes leading to a secondary cholesteatoma and facial nerve palsy, while sensorineural hearing loss and vestibular disorders usually follow otic capsule involvement which occurs due to spreading infection or penetration of inflammatory product and toxins through the round window into the inner ear. This in turn results in cochlear destruction, internal auditory canal stenosis or vestibular fistulisation [7-10]. A clinical staging had been suggested by Barrionuevo et al, in accordance to the progression of the disease. Stage 1 is the latent or asymptomatic phase, where by the management is conservative with regular follow-up. Stage 2 is the symptomatic phase and stage 3 is for those with complications [11]. The case reported here had stage 2 manifestations.

However, the majority are asymptomatic and discovered incidentally on radiographs as lesions with the characteristic ground glass appearance. A radiographic diagnosis is usually sufficient and a subsequent bone biopsy may not be required. However when performed histological examination reveals disorganized bony trabeculae and spindle cells surrounded by a fibrous matrix. A bone scan may be considered to rule out polyostotic FD, and follow-up radiographs recommended every 6 months to ensure there has been no progression. Computed tomography (CT) and magnetic resonance imaging are additional modalities for further elucidating the extent of bony and neurovascular involvement, and total body bone scintigraphy can determine the extent

of skeletal disease and predict functional outcome. CT scan often assists with differentiating fibrous dysplasia from other osteodystrophies of the skull base including otosclerosis, osteogenesis imperfect, Paget's disease, osteopetrosis, hemangioma, meningioma and mucocele [1-4].

Appropriate treatment of FD is often highly individualized and based on presentation. Observational studies report bisphosphonates (like pamidronate) help improve function, decrease pain, and lower fracture risk. Surgical intervention is aimed at preventing functional complications while improving regional asthetics [10]. In the present case, stenosis of the external auditory canal with resultant cholesteatoma was a definitive indication for surgical intervention. The patient underwent radical mastoid exploration with partial reconstruction of the cavity with inferiorly based musculoperiosteal flap and temporalis fascia which healed and epithelized well in the post-operative period.

Conclusion

FD is a benign, non-familial condition which can affect temporal bone and can lead to formation of secondary cholesteatoma. Diagnosis can be made mainly on the basis of radiographs and CT scans, though histology can further confirm the diagnosis. Surgical treatment is directed towards excision of all the resectable bone, choleasteatoma and restoration of as much hearing as possible. Partial obliteration of cavity with inferiorly based musculoperiosteal flapwith adequate meatoplastycan help in achieving shallow and dry cavity. Periodic CT can be used to follow the progression of the disease in post-operative period.

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