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Otolaryngology International

July - December 2017
Volume 2, Number 2

Contents

Review Article

Case Reports

Otolaryngology International

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Original Article

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Study on Eustachian Tube Dysfunction and Effect of Adenoidectomy on Hearing Threshold and Middle-Ear Pressures

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Abstract

Adenoiditis and Adenoid Hypertrophy are the most common health conditions afflicting the Paediatric population. Nasopharyngeal obstruction due to Adenoid Hypertrophy leads to Hyponasality, Mouth breathing, Snoring, and Sleep apnea. It also causes Eustachian Tube Dysfunction leading to increased negative middle ear pressure, which is an important etiological factor in the causation of hearing loss in children. This may result in serious consequences in the form of Speech impairment, Inattention, Poor performance in school, Behavioral problems and Impaired intellectual development. *Materials and Method:* In this study, one hundred children (200 ears) aged between 5 and 14 years who underwent Adenoidectomy were analyzed using Otoscopy, Pure Tone Audiometry and Impedance Audiometry pre-operatively and 6 weeks post-operatively. X-Ray Nasopharynx-Lateral view and Diagnostic Nasal Endoscopy was done to assess the grade of Adenoid Hypertrophy. *Results:* It was found that post-operative hearing threshold and middle ear pressures showed significant improvement following Adenoidectomy. The total percentage of ears with normal Type A impedance curve increased from a pre-operative 53.5% to a post-operative 91%. *Conclusion*: Our study concludes that Adenoidectomy has beneficial effects in children with Otitis Media with Effusion and Eustachian Tube Dysfunction.

Keywords: Adenoiditis, Adenoid Hypertrophy; Adenoidectomy; Hyponasality.

Introduction

Adenoiditis and Adenoid Hypertrophy are the most common health conditions afflicting the Paediatric population. Nasopharyngeal obstruction due to Adenoid Hypertrophy leads to Hyponasality, Mouth breathing, Snoring, Sleep apnea, Otitis Media with Effusion and Sinusitis.

Eustachian Tube Dysfunction can occur in Adenoiditis and Adenoid Hypertrophy. The functions of the Eustachian Tube include middle ear ventilation to Equalize the middle ear pressure with atmospheric pressure, Clearance of secretions produced within the middle ear into nasopharynx and Protection of middle ear from nasopharyngeal secretions. Hearing is optimal when pressure within the middle ear is relatively the same as that of the atmosphere. Adenoid Hypertrophy can obstruct the pharyngeal ostia of the Eustachian Tube by mechanical pressure and produce pressure on the lymphatics causing mucosal swelling. It can also act as a carrier of pathogenic bacteria and viruses leading to increased oedema and Eustachian Tube dysfunction.

The presence of Eustachian Tube Dysfunction leads to increased negative middle ear pressure which is an important aetiological factor in the causation of hearing loss. This may result in serious consequences in the form of Speech impairment, Inattention, Poor performance in school, Behavioral problems and Impaired intellectual development.

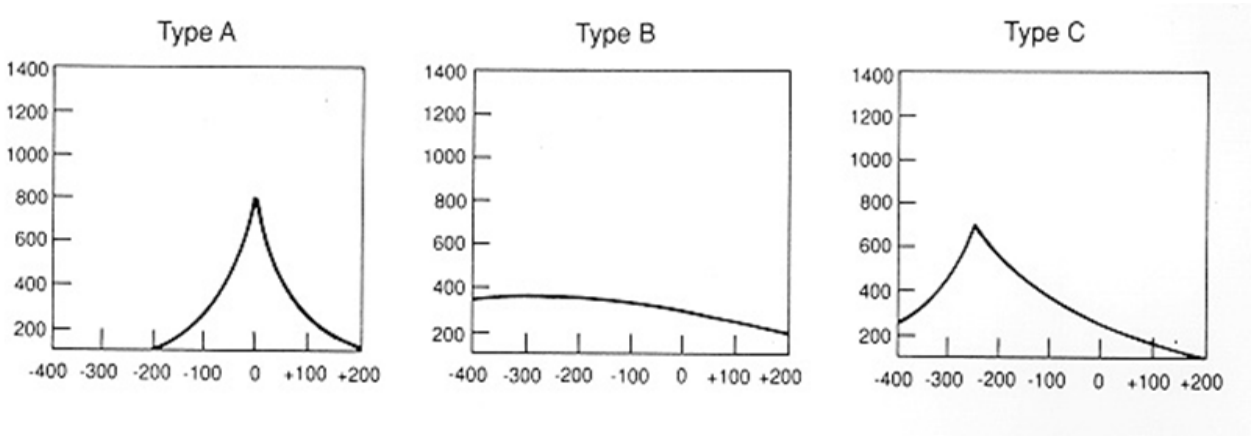
This study documented the presence of the Eustachian Tube dysfunction in children with Adenoiditis and Adenoid Hypertrophy and the

beneficial effect of Adenoidectomy on Hearing threshold and Middle ear pressures.

Materials and Method

Patients attending the Department of Otorhinolaryngology at Rajah Muthiah Medical College with symptomatic Adenoiditis and Adenoid Hypertrophy and undergoing Adenoidectomy between October 2015 to August 2017 were included in this study.

One hundred children, of either sex, in the age group between 5 and 14 years suffering from Adenoiditis and Adenoid Hypertrophy were included in the study. X-Ray Nasopharynx-Lateral view and Diagnostic Nasal Endoscopy was done to confirm presence of Adenoid Hypertrophy, to determine size of adenoids and to assess airway patency. Pure Tone Audiometry and Impedance Audiometry was done in all cases before surgery and 6 weeks after Adenoidectomy to assess the Hearing Threshold and Middle Ear Pressures and thereby Eustachian Tube Dysfunction.



Tympanometry was done in all patients pre-operatively and six weeks post-operatively and the results are noted as

Type A- Normal Compliance

Type B- Otitis Media with Effusion with reduced Compliance

Type C- Negative Middle Ear Pressure with normal Compliance

Endoscopic Grading of Adenoid Hypertrophy

GRADE	Percentage of Obstruction of Choana
I	< 25%
II	25 – 50%
III	50 – 75%
IV	>75 %

X-Ray Nasopharynx Lateral View Grading of Adenoid Hypertrophy

Grade	Soft Tissue Shadow in Nasopharynx (%)	Adenoid Hypertrophy
1	0-50	Mild
2	50-75	Moderate
3	75-100	Severe

Inclusion Criteria

1. Children between the ages of 5- 14 years
2. Children presenting with Adenoiditis and Adenoid Hypertrophy (Minimum three episodes/year)
3. Children presenting with complaints of Nasal Obstruction not influenced by decongestants, Mouth breathing, Snoring, Difficulty in swallowing and Hypoacusis.

Exclusion Criteria

Children with Cranio-facial anomalies, Nasal septal deviation and Sinonasal infection

Results

Table 1: Gender Incidence

Male	54 %
Female	46 %

Table 2: Presenting Symptoms

Symptom	Incidence
Dysphagia	100%
Odynophagia	100%
Mouth Breathing	52%
Snoring	52%
Hearing Impairment	18%
Ear Block	18%
Recurrent URTI	100%

Table 3: Examination Findings

Clinical Sign	Incidence
Open Mouth Breathing	16%
High Arched Palate	8%
Pinched Nostrils	8%
Crowded Teeth	12%

Table 4: Diagnostic Endoscopic Grading of Adenoid Hypertrophy in the Present Study

Grade of Adenoid Hypertrophy	Total (Number of Children)
Grade 1	0
Grade 2	48
Grade 3	36
Grade 4	16

Table 5: Diagnostic Nasal Endoscopic Finding of Abutment of Adenoid on the Torus Tubaris

Presence of Abutment on the Torus Tubaris (Percentage)	Absence of Abutment on the Torus Tubaris (Percentage)
36%	64%

Table 6: X-Ray Nasopharynx Lateral View Grading of Adenoid Hypertrophy in the Present Study

Grade	Incidence (Percentage)
1	48%
2	36%
3	16%

Table 7: Comparison Between Grade of Adenoid Hypertrophy and Pre-Operative Type of Curve

Grade of Adenoid Hypertrophy	Pre Op Type A Impedance Curve (Number of Ears)	Pre Op Type B Impedance Curve (Number of Ears)	Pre Op Type C Impedance Curve (Number of Ears)	Total
Grade 1	0	0	0	0
Grade 2	89	6	1	96
Grade 3	16	39	17	72
Grade 4	2	9	21	32
Total	107	54	39	200

Table 8: Comparison of Pre-Operative and Post-Operative Hearing Threshold

Hearing Threshold	PRE-OP (n = 200)	PRE – OP (PERCENTAGE)	POST - OP (n = 200)	POST-OP (PERCENTAGE)
	No of Ears		No of Ears	
0 – 25	148	74%	180	90%
26 – 40	36	18%	14	7%
41 – 60	16	8%	0	0%
61 – 80	0	0%	0	0%
> 81	0	0%	0	0%
Lost for Follow up	-	-	6	3%
TOTAL	200	100	200	100

Table 9: Comparison of Pre-Operative and Post-Operative Middle Ear Pressure

Middle Ear Pressure (mm H20)	PRE -OP (n = 200) No of Ears	PRE-OP (Percentage)	POST –OP n= 200 No of Ears	POST – OP (Percentage)
-400 to -350	9	4.5 %	0	0 %
-349 to -300	7	3.5 %	1	0.5 %
-299 to -250	5	2.5 %	1	0.5 %
-249 to -200	20	10%	3	1.5 %
-199 to -150	14	7 %	0	0 %
-149 to -100	22	11 %	8	4 %
-99 to -50	23	11.5 %	2	1%
-49 to 0	88	44%	159	79.5%
1 to 50	10	5%	18	9%
51 to 100	2	1 %	2	1 %
Lost for Follow - up			6	3%
Total	200	100	200	100

Table 10: Comparison between Pre-Operative & Post Operative Type of Impedance Curve

Type of Curve	PRE-OP (n = 200)	PRE-OP (Percentage)	POST-OP (n = 200)	POST-OP (Percentage)
Type A	107	53.5 %	182	91 %
Type B	54	27 %	13	6.5 %
Type C	39	19.5 %	5	2.5 %
Total	200	100 %	200	100 %

Discussion

Secretory Otitis Media also known as Otitis Media with Effusion has been identified as a common middle ear condition causing deafness in children. It affects the child's learning ability through recurrent temporary hearing loss, permanent hearing impairment and language disorders. A Child with an episode of Otitis Media with Effusion often experiences a mild to moderate fluctuating hearing loss, thus appreciating only partial or inconsistent auditory cues which may make it difficult for the ear to filter it from background noise. It has been hypothesized that this may affect the input to the knowledge base or to the neural substrate on which language learning is built. It also has been proposed that any difficulties attributable to Otitis Medis with Effusion associated hearing loss may become evident when a child reaches school age and faces the challenges of school environment. Academic skills particularly reading and other language based subjects may be affected when there is a high demand for attention to verbally presented information.

In my study following Adenoidectomy, Pure Tone Audiometry revealed that Hearing Threshold between 0 and 25 db increased from a pre-operative 74% to a post-operative 90%. Remarkable improvement in Middle Ear Pressure was also noted in the range of middle ear pressure between -99 to 99 mm H20, which increased from a pre-operative 61.5% to a post-operative 90.5%. The above findings substantiate the beneficial role of Adenoidectomy in improving Eustachian Tube Function and Middle Ear Pressure.

The main clinical problem in children with Adenoiditis and Adenoid Hypertrophy is that in some children the symptoms of Otitis Media with Effusion is occult. Some children may present with only poor expression and poor communication skills which parents may find to be trivial and hence neglected. Clinical examination is also difficult, especially in children with narrow ear canal, it is very difficult to examine the tympanic membrane. Children tend to be fearful of otoscopic examination, causing them to cry which in turn will cause tympanic membrane congestion resulting in a possible mis-diagnosis.

Thus diagnosing a child with Otitis Media with Effusion is a tricky task. Regardless of the difficulty in diagnosis, it is extremely important to subject a child with suspected Adenoiditis and Adenoid Hypertrophy to Otoscopic, Radiographic and Audiometric studies so that a diagnosis of Eustachian Tube Dysfunction and Otitis Media with Effusion is not missed. If a diagnosis of Otitis Media with Effusion is suspected, early diagnosis and prompt treatment will significantly improve the child's learning and cognitive abilities.

Conclusion

As Helen Keller once notably said, "*The problems of deafness are deeper and more complex, if not more important, than those of blindness. Deafness is a much worse misfortune, for it means the loss of the most vital stimulus- the sound of the voice that brings language, sets thoughts astir, and keeps us in the intellectual company of man*." Middle ear effusion and Eustachian Tube Dysfunction caused by Adenoid Hypertrophy and Adenoiditis will adversely affect hearing and interfere with the Child's Intellectual Development and Academic Performance.

Diagnosis of Otitis Media with Effusion in children is often delayed as they cannot complain of hearing loss. Most of the children with abnormal Tympanograms were regarded to have normal hearing by their parents. This may result in serious consequences in the form of Speech impairment, Inattention, Poor performance in school, Behavioral problems and Impaired intellectual development. Thus screening for hearing impairment should include Tympanometry, as Otitis Media with Effusion is one of the most preventable causes of Conductive Hearing Loss. As supported by the results seen in my study, Adenoidectomy has beneficial and constructive effects in children with Eustachian Tube Dysfunction and Otitis Media with Effusion.

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Case Report

Otolaryngology International
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Cemento Ossifying Fibroma of Maxilla: A Diagnostic Dilemma

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Abstract

An interesting case of cemento ossifying fibroma of maxilla in a 61 years old man presenting with ulcer over the left side of hard palate is discussed. The clinical features, investigations, differential diagnosis and management of cemento ossifying fibroma of maxilla is discussed.

Keywords: Cemento Ossifying Fibroma; Maxilla.

Case Summary

A 61 year old man presented to ENT OPD with complaints of pain over the left upper premolar tooth and ulcer in the left side of the hard palate for one month duration. Clinical examination revealed an ulcer over the left side of hard palate measuring about 1 x0.5 cm with irregular margins (Figure 1). The ulcer was 1 cm away from midline extending up to left upper pre molar. The margin of the ulcer was well defined edges and the floor of the ulcer showed granulation tissue. Rest ENT examination was NAD. Diagnostic nasal endoscopy showed normal study.MRI PNS (Figure 2) showed encapsulated fluid signal intensity area at the left side of hard palate with associated cortical breach of alveolar margin of the left side maxilla in the region between canine and first premolar. The differential diagnosis on MRI was osteomyelitis of hard palate with sub periosteal abscess and malignant mixed tumor of hard palate. Biopsy of the lesion showed low grade dysplasia.

All other hematological and biochemical investigations were within the normal range. Patient was advised surgical excision of the lesion. The patient underwent left upper alveolectomy (partial)(Figure 3) under general anaesthesia. The specimen was sent for histopathological examination which showed bony trabeculae in irregular shape without osteoblastic rimming surrounded by densely haphardly arranged fibrous stroma. Areas of psammomatous bodies around bony trabeculae are seen.There was no evidence of nuclear atypia,hyperchromasia or nuclear activity and the histopathological picture was suggestive of cemento ossifying fibroma of maxilla(Figure 4).

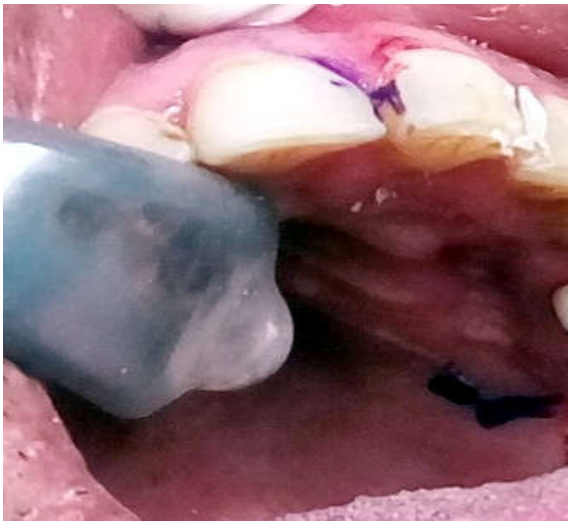

Fig. 1: Photograph of the lesion left upper alveolus

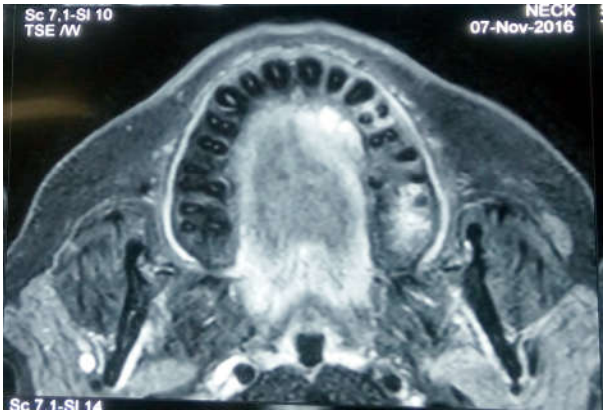

Fig. 2: MRI PNS(Axial) image of the lesion

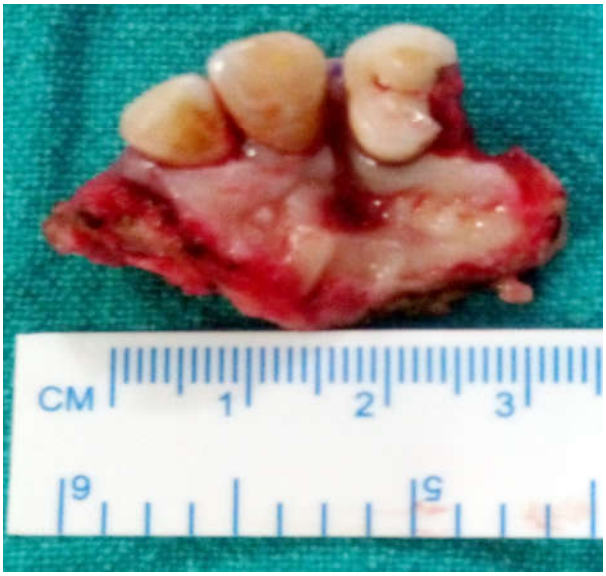

Fig. 3: Photograph of the resected specimen

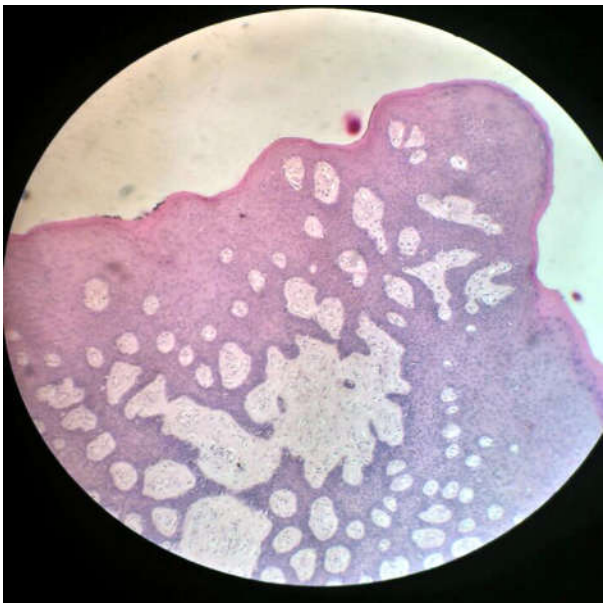

Fig. 4: Histopathological picture Haematoxylin and Eosin stain

Discussion

Ossifying fibroma forms a spectrum of fibro-osseous lesions of the jaws. They are rare, benign, non odontogenic tumors that are commonly seen in the head and neck region.

Cemento ossifying fibroma of the maxilla is an uncommon tumor. The tumor was first described by Menzel in 1872 [1] and the term ossifying fibroma was first given by Montgomery [2] in 1927 . It is considered as benign osseous tumor. Lesions with fibrous and osseous component include fibrous dysplasia (FD), ossifying fibroma (OF),cemento ossifying fibroma (COF) and cementifying fibroma(CF) [3].

According to the WHO classification [4] benign fibro-osseous lesions in the oral and maxillofacial region were divided into two categories osteogenic neoplasms and non neoplastic bone lesions. The differential diagnosis based on clinical and radiological examinations poses diagnostic challenges and only histopathological examination is confirmatory.

Ossifying fibroma is most commonly seen between the third and fourth decades of life. It's more frequent in women than men (4:1). The most common location is the mandible in 70 -90% of all cases [5]. Generally it is a slow growing tumor usually asymptomatic however the lesion can become large enough to present with facial deformity. Patients generally present with a history of painless expansion of a tooth bearing portion of the mandible. Lesions of the maxilla are less common.

Histologically these tumours are well vascularised consisting of fibrocellular tissue with capacity to form immature bone trabeculae and cementoid formations. These findings are not specific as it's seen in fibrous dysplasia as well. So a definitive diagnosis requires correlation of clinical, radiological and pathological evaluation [6].

Radiologically Cemento ossifying fibroma has different patterns based on the amount of mineralized tissue. It presents as demarcated unilocular lesion that might have a different degree of opacification. The differential diagnosis based on radiological evaluation included chondrosarcoma, osteosarcoma, fibrous dysplasia, odontogenic cysts , grolin's cyst and pindborg tumour. The well defined borders of Cementoossifying fibroma helps to differentiate from sarcoma and carcinoma. Fibrous dysplasia has a typical ground glass appearance.

The underlying cause of this condition is not known, there have been reports of past trauma in the area of lesion, postulating as a connective tissue reaction than a genuine neoplasm [7].

The treatment of ossifying fibroma is surgical with surgical options being ennucleation. curettage and radical sugery.

The recommended treatment of choice is excision of tumour including a rim of normal tissue. Management should be individualized based on size, location, benign nature and growth behavior of the lesion. In our case considering malignancy as a differential diagnosis we did a left upper alveolectomy (partial) under general anaesthesia.

Conclusion

The diagnosis of cemento ossifying fibroma can pose a diagnostic dilemma. The diagnosis should be carefully considered after ruling out malignancy. Imaging and histopathological examination play a crucial role in establishing the diagnosis.

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Case Report

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Life Saving Emergency Tracheostomy in Supervasmol Poisoning – A Clinical Experience!

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Abstract

Super vasmol 33 is a Hair dye also known as ParaPhenylenediamine (PPD) is a Common house hold Poison used as a suicidal agent because of its low cost and easy availability in the market. There are Many case reports of Paraphenylenediamine (PPD) poison used for deliberate self harm by adults and Accidental ingestion by children. We report a case of a 20 year old female who intentionally consumed Super vasmol poison, and presented to our causality within 6 hours of ingestion with Angioedema of face And Severe Respiratory Distress. Timely Tracheostomy after failed intubation proved to be a life saving Procedure in our patient.

Keywords: Super Vasmol Poison; Emergency Tracheostomy.

Introduction

Super vasmol also known as Paraphenylenediamine (PPD) is an aromatic amine which is used in variety of industrial products and by hair dye companies. The constituents of this hair dye are PPD, sodium lauryl Sulphate, EDTA, cestostearyl alcohol, resorcinol, propylene glycol, liquid paraffin, herbal extracts, Preservatives [1]. A rising trend of super vasmol poison ingestion both accidental and as a means of Deliberate self harm has been reported in India [2].

Paraphenylenediamine poisoning is reported globally, More so in developing countries. The first artificial dye was synthesized in the laboratory in 1856 [3].

In 1990 the number one leading Cause of poisoning in Morocco was found to be PPD [4] which caused Contact dermatitis in susceptible individuals, but its ingestion lead to Acute Angioedema of face and neck, Rhabdomyolysis and Acute Renal Failure [5,7].

Case Report

A 20 year old female was brought by her parents to casuality with complaints of Swelling over the face and inability to speak for 3 hours duration. On general examination patient was conscious but Unable to speak because of edema of the face and neck. Pulse 84/min regular, BP 110/70mmHg, RR16/min, SpO_2 92% on room air. Examination of oral cavity showed limited mouth opening and Edema of tongue. Neck examination revealed cervico facial swelling. Detailed history revealed that the Patient had orally consumed super vasmol hair dye as an act of suicide 7 hours prior to Reporting to casuality. 30 minutes after admission in the casuality the patient developed Severe Stridor And started to desaturate, SpO_2 dropped to less than 60%, patient was immediately shifted to emergency Operating room where oral intubation could not be attempted in view of the cervico facial and oral edema. Blind nasal intubation was tried which resulted in failure to intubate, flexible video laryngoscopy guided intubation was also attempted which showed edema

Fig. 1: Patient at the time of presentation to casuality, failure of flexible video laryngoscopy, intra operative tracheostomy and immediate post operative period

Fig. 2: Post operative follow up and ENT OPD review for video direct laryngoscopy

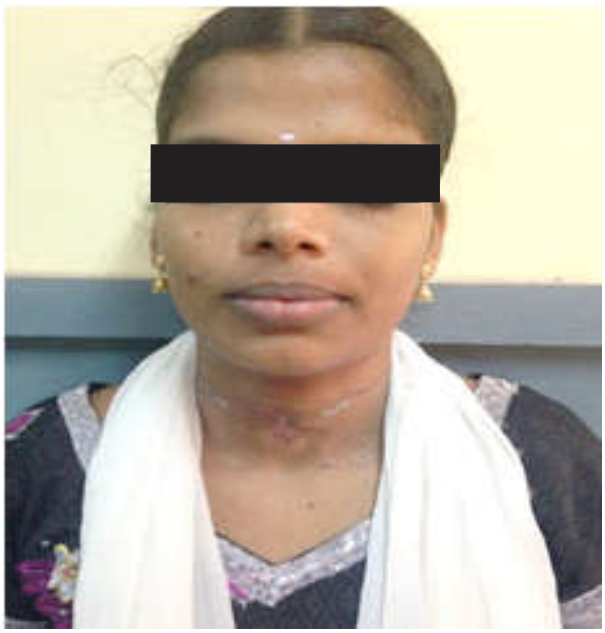

Fig. 3: Patient after post tracheostomy stoma closure

of epiglottis, false cords and true vocal cords, and the attempted intubation resulted in failure. Immediately a life saving Emergency tracheostomy was performed on this patient, portex tracheostomy tube 7 size was used to secure the airway and with ventillatory support the patient was shifted to intensive care unit. Initial blood parameters showed Hb –12.3gm%, wbc 9,600 cells/cumm, platlet-2.3lakh, serum urea-28mg/dl, serum creatinine 0.6mg/dl. Sodium 139, potassium 4.1 chloride 99 mmol/L. 18 hrs after admission the patients urine examination revealed proteinurea, haemoglobinuria and hemosiderinuria. CREATININE KINASE was 440 U/L. Liver function test was WNL. Patient was given symptomatic treatment and adequate i.v hydration, intake and output was monitored. On the 5 th day of admission, patient's blood and urine examination was WNL, facial edema reduced significantly, patient was discharged after 10 days with fullers metal tracheostomy tube insitu, and follow up in Psychiatry and ENT department.

Discussion

Suicidal tendencies and suicidal rates have drastically increased over the past few decades and the common available super vasmol hair dye has become one of the important house hold substance used as a means of poisoning [1]. In 1924 Nott documented the first case of PPD poison in the owner of a hair saloon [6]. Paraphenylenediamine (PPD) is

a derivative of paranitroanaline which on oxidation produces Bondrowski's base which is highly toxic, mutagenic and an allergen [7]. PPD is used along with ammonia and hydrogen peroxide in hair dying and also when added to henna it is used in tattooing for its darkening effects [9]. Onset of symptoms after ingestion of super vasmol poison is usually between 4 -6 hours. The symptoms are considered to be dose related and patients after ingestion of large quantity (7-10 gm of super vasmol) have higher morbidity and mortality [7]. Patient presents most commonly with Angioedema of face and neck, and Respiratory Distress. Organ damage caused by super vasmol poison may be assessed by appropriate tests in case with rhabdomyolysis, kidney and liver involvement. The effect of resorcinol is associated with seizure, coma, methaemoglobinemia, acute tubular necrosis, arrhythmias, intra vascular haemolysis, gastritis, vertigo, tremors, myocarditis and arrhythmias. The characteristic triad mentioned in literature is ANGIO NEUROTIC OEDEMA of face and neck leading to stridor, RHABDOMYOLYSIS with chocolate coloured urine and ACUTE RENAL FAILURE could be a confirmative evidence of Paraphenyldiamine poison even in the absence of laboratory facilities and when history is lacking in case of emergency [9]. Cervico Facial Oedema was the most marked presentation in our patient, severe enough to cause Respiratory Distress. Timely management to secure the airway was under taken and emergency tracheostomy was performed as a life saving procedure for the patient, Anti histamine and steroids were used to treat the patient although there is no therapeutic trials as to their benefit. Forced diuresis and maintenance of high urine output was done to augment the patient's renal function which improved in the following days. There is no specific antidote for PPD poison [2]. Treatment is mainly symptomatic and supportive. Early clinical diagnosis and timely intervention in securing an open airway, maintenance of adequate hydration and oxygenation along with good urine output is the corner stone of successful management and outcome.

Conclusion

Super vasmol poison is rapidly emerging as a suicidal poison because of its easy availability and Extensive use, hence awareness of its effects and side effects is important. The classical presentation of Cervico Facial Angioedema, severe Rhabdomyolysis and Acute Renal Failure warrants early diagnosis

And intervention to secure the airway and aggressive supportive is required to prevent organ failure.

Legislation is required to ensure that the hair dye is not freely available in the market.

Source of Support: Nil

Conflict of Interest: Nil

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Case Report

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Malignant Fibrous Histiocytoma of Right Upper Alveolus

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Abstract

Malignant fibrous histiocytoma (MFH) is a high grade and aggressive sarcoma accounting for around 20% of soft tissue sarcomas. These tumors rarely arise in the head and neck. A case of malignant fibrous histiocytoma of the right upper alveolus in a lady is described. The clinical features, investigative modalities, diagnostic features and management is described.

Keywords: Histiocytoma; Sarcoma; Alveolus; Fibrous; Recurrence.

Introduction

Malignant fibrous histiocytoma (MFH) is a high grade sarcoma originally described by O'Brien and Stout in 1964 [1]. It is the most common soft tissue sarcoma occurring in late adult life between 50 - 70 years and is extremely rare in childhood. MFH occurs most commonly in the extremities (70 - 75%) with lower extremities accounting for 59% of cases followed by the retroperitoneum [1]. Tumors typically arise in deep fascia or skeletal muscle and rarely arise in the head and neck.

Case Report

62 yr old lady presented with history of growth right upper alveolus for 6months with comorbid conditions of diabetes and hypertension. She was a known tobacco chewer for the last 20 years. On examination she had an proliferative 4*3 cm growth right upper alveolus extending from right lateral incisor and posteriorly till right 2nd upper molar involving right gingivo buccal sulcus and right hard palate (Figure 1). Neck Right level II and III nodes 1*1 cm were palpable. Biopsy was suggestive of a poorly differentiated malignant mesenchymal neoplasm.

CECT scan (Figure 2) showed enhancing soft tissue density noted involving the right alveolus of the maxilla measuring about 3x 1.2 cm with minimal bony erosion. Multiple enlarged cervical lymphadenopathy involving Ib, II, III, was seen on the right side. PETCT showed metabolically active disease right upper alveolus (SUV max 10.63) with enlarged cervical nodes were seen at level IB,II,III right side with the largest right level II (33*30 mm, SUV max 13.17). There were no distant metastasis.

Disease was initially staged as Malignant mesenchymal neoplasm Right upper alveolusT4 N2bM0. Patient underwent Right Infrastructural maxillectomy (Figure 3) with Right Modified Neck Dissection Type III with reconstruction by Anterolateral thigh flap (Figure 4). Histopathology on gross showed proliferative tumor hard palate 2.4*2.3*2 cm. All cut margins were free, 10/19 nodes were positive.

Microscopic examination (Figure 5) showed highly pleomorphic spindle to polygonal cells with marked variation in size shape, marked nuclear

atypia with enlarged hyperchromatin nucleus, irregular nuclear membrane, prominent nucleoli and moderate amount of eosinophillic cytoplasm. 2-3 mitosis/10 high power field. These cells were arranged in bundles, fascicles arranged in herring bone pattern and arranged in variable direction in fibromyxoid stroma. Increased angiogenesis was seen. On IHC vimentin was strongly positive. Rest markers cK, Cd31, Cd34, LCA, EMA, HMB, Desmin all were negative. On followup of 3 years patient is asymptomatic with no recurrence.

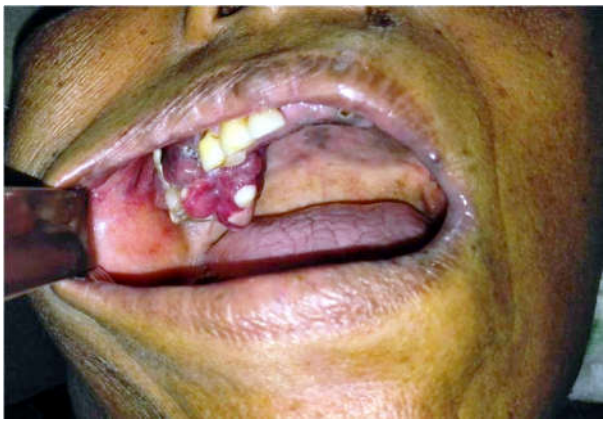

Fig. 1: Showing the tumor involving the Right hard palate and right upper alveolus

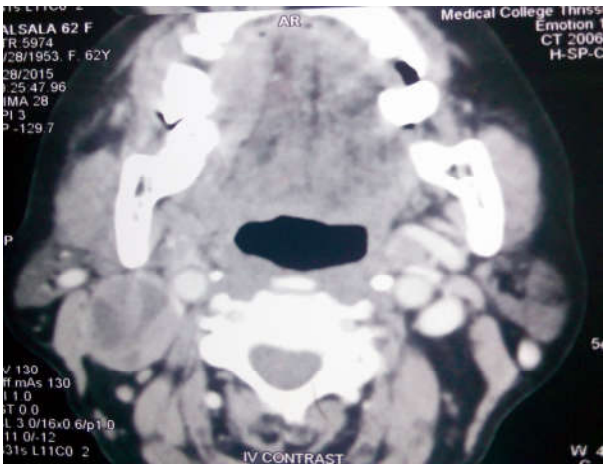

Fig. 2: CECT scan showing the tumor involving the right upper alveolus

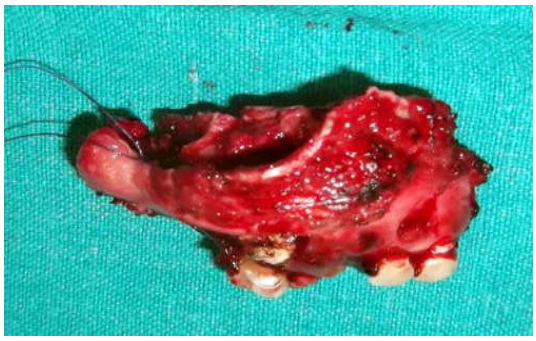

Fig. 3: Showng Right infrastructural maxillectomy specimen

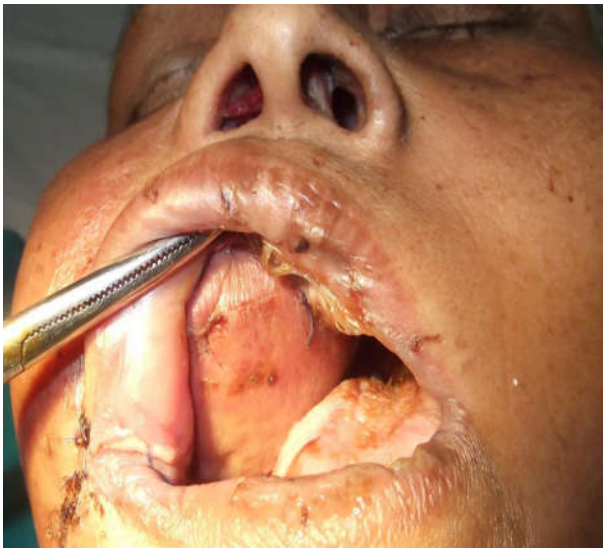

Fig. 4: Showing reconstruction of maxillarydefect by anterolateral thigh flap

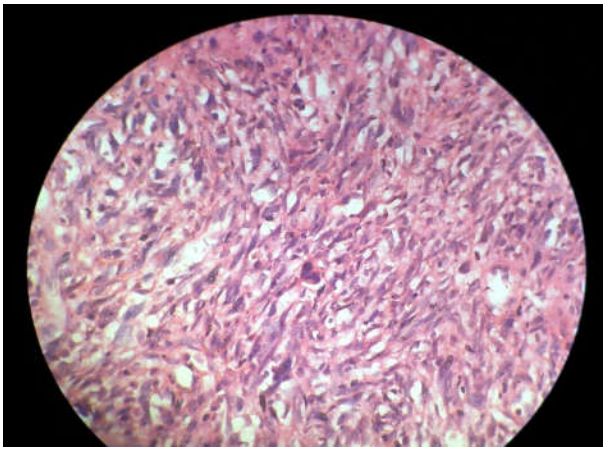

Fig. 5: Photomicrograph high power showing storiform pattern of MFH

Discussion

Malignant fibrous histiocytoma tumors (MFH) rarely occur in the head and neck with an incidence of 1-2% [2]. In this region, prevalence of the tumor in the nasal cavity and paranasal sinuses is 30%, craniofacial bones is 15-25%, larynx and soft-tissue of neck is 10-15% each, and oral cavity 5-15% [3]. In our case, it has affected the maxillary alveolar ridge and the hard palate.

Male predominance has been reported for this neoplasm with a male: female ratio of 2:1 However Chen et al [3] found a female predominance with male: female ratio of 1:2. In our case, it was a primary tumor reported in a lady.

MFH tumors can be either primary or secondary. Primary tumors are less aggressive and more

common than secondary tumors [4]. Secondary tumors are generally associated with underlying conditions such as prior radiotherapy prior trauma, fibrous dysplasia or benign bone tumors like enchondroma. MFH may occur at any age, with a peak incidence in the fifth to seventh decade. MFH is one of the most common Radiation-induced sarcoma of the head and neck. The period of latency between initial radiation therapy and diagnosis of Radiation-induced sarcoma ranged from 9 - 45 years with a median of 17 years [5].

The exact histogenesis of MFH remains controversial. The majority of investigators have suggested that histiocytic and fibrocytic cell lines are derived from the small numbers of undifferentiated mesenchymal cells [6]. In general, the tumor contains both fibroblast like and histiocyte like cells in varying proportions, with spindled and rounded cells exhibiting a storiform arrangement.

Enzinger and Weiss have defined five histological subtypes of MFH as follows: storiform-pleomorphic, myxoid, giant cell, inflammatory, and angiomatoid. The storiform-pleomorphic and myxoid variants are the most common type. MFH is generally seen between the ages of 50 and 70 years. An exception is the angiomatoid variant that usually affects individuals who are younger than 20 years old [1-3]. Tumors with angiomatoid and myxoid patterns are often associated with a more favorable prognosis as they metastasise late and respond well to surgery [2]. The inflammatory and, the pleomorphic types are more aggressive, metastasise early and respond less favorably to surgery alone [7]. Our patient showed malignant tumor arranged predominantly in form of in storiform pattern.

In recent times MFH is considered as a form of fibrosarcoma and the tumor is very likely over diagnosed by some pathologists. This is because the more pleomorphic the tumor, the more difficult is to distinguish from other types of sarcomas, such as spindle cell carcinoma, pleomorphic leiomyosarcoma and pleomorphic liposarcoma Distinction among these soft tissue tumors is best achieved by a joint immunohistochemical and ultrastructural study.

Fibrous histiocytoma is typically immunoreactive for vimentin, and sometimes for smooth muscle actin or alpha-1antitrypsin, but not for desmin, keratin, epithelial membrane antigen, S-100 protein, factor VIII-related antigen, CD34, or carcino-embryonic antigen, supporting the hypothesis that the tumor cells are of mesenchymal origin.

Surgery is the best treatment for this aggressive neoplasm. The advent of more advanced reconstructive techniques including free tissue transfer has made more aggressive surgical resection of these tumors possible. Local recurrence rate of MFH after initial local excision ranges between 16% and 52%. Block et al [7] reported local recurrence or distant metastasis in 55% of cases of MFH. Recurrence is related to size, depth of invasion, and microscopically positive surgical margins. The presence of positive surgical margins after definitive treatment is the single most important factor relating to local recurrence [8].

Owing to the high incidence of local recurrence, postoperative radiotherapy is generally advocated. Incidence of distant metastases is 25-35% in patients with MFH of head and neck region with pulmonary being most common (90%) followed by bone (8%), and liver [3]. The 5-year survival rate ranges from 35-60%. The primary tumor factors associated with a worse prognosis are the histiologic subtype, necrosis, a high mitotic count, and blood vessel invasion [9]. The clinical stage of the tumor, which is defined by tumor grade, size, and presence of distant metastases, is the most important prognostic factor. Other clinical predictors of a poor outcome include advanced age, male gender, underlying systemic illness, large primary tumors, tumors arising from the bones, deep-seated tumors, and a history of previous radiation [2,10].

Conclusion

Malignant fibrous histiocytoma of the head and neck is an aggressive tumor. Inadequate resection is related to a higher local recurrence rate and worse prognosis and hence these tumors need to be resected as extensively as possible.

Conflicts of Interest: Nil

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Case Report

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Pyogenic Granuloma - Presenting as Midline Swelling of Tongue

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Abstract

Pyogenic granuloma is an inflammation induced hyperplastic lesion involving oral cavity in response to various stimuli such as low grade local irritation, traumatic injury or hormonal factors. We report a case of 8 years old female patient with a midline lesion on the dorsum of tongue which was successfully excised under local anaesthesia andthe histopathological study revealed it as pyogenic granuloma.

Keywords: Pyogenic Granuloma; Tongue; Midline.

Introduction

Hullihen [1-8] reported the first case of Pyogenic granuloma in 1844. But Hartzell in 1904 [1,8] coined the current term, pyogenic granuloma or granuloma pyogenicum. Pyogenic granuloma is a commonlesion occurring on the skin [8] and extremely rare in the oral cavity where it is present over the keratinized tissue [1]. The name pyogenic granuloma is a misnomer since the condition is not associated with pus and does not represent a granuloma histologically [1,4,5]. The role of infective agents may play a part in recurrent Pyogenic granuloma.There are two types of Pyogenic granuloma, Lobular Capillary haemangioma (LCH) type and non-Lobular Capillary Haemangioma (non-LCH) type [1,8].

The pyogenic granuloma is a non-neoplastic [2,5] soft tissue tumour of oral cavity that is believed to be reactive to stimuli such as local irritation [2], traumatic injury, hormonal factors, and involves the gingiva most commonly [1,2]; rarely it can occur on lips, tongue, buccal mucosa and palate [3]. There is a high degree of occurrence in second [4], third, fourth decade [6], with male to female ratio of 1:1.7 [6,10]. Characteristically, pyogenic granuloma of tongue is more common on the lateral side of the tongue which may be related to trauma from adjacent sharp teeth or ill-fitting dentures [4,9].

The purpose of this article is to report an unusual case of pyogenic granuloma with the lesion occurring in the midline on the dorsum of the tongue.

Case Report

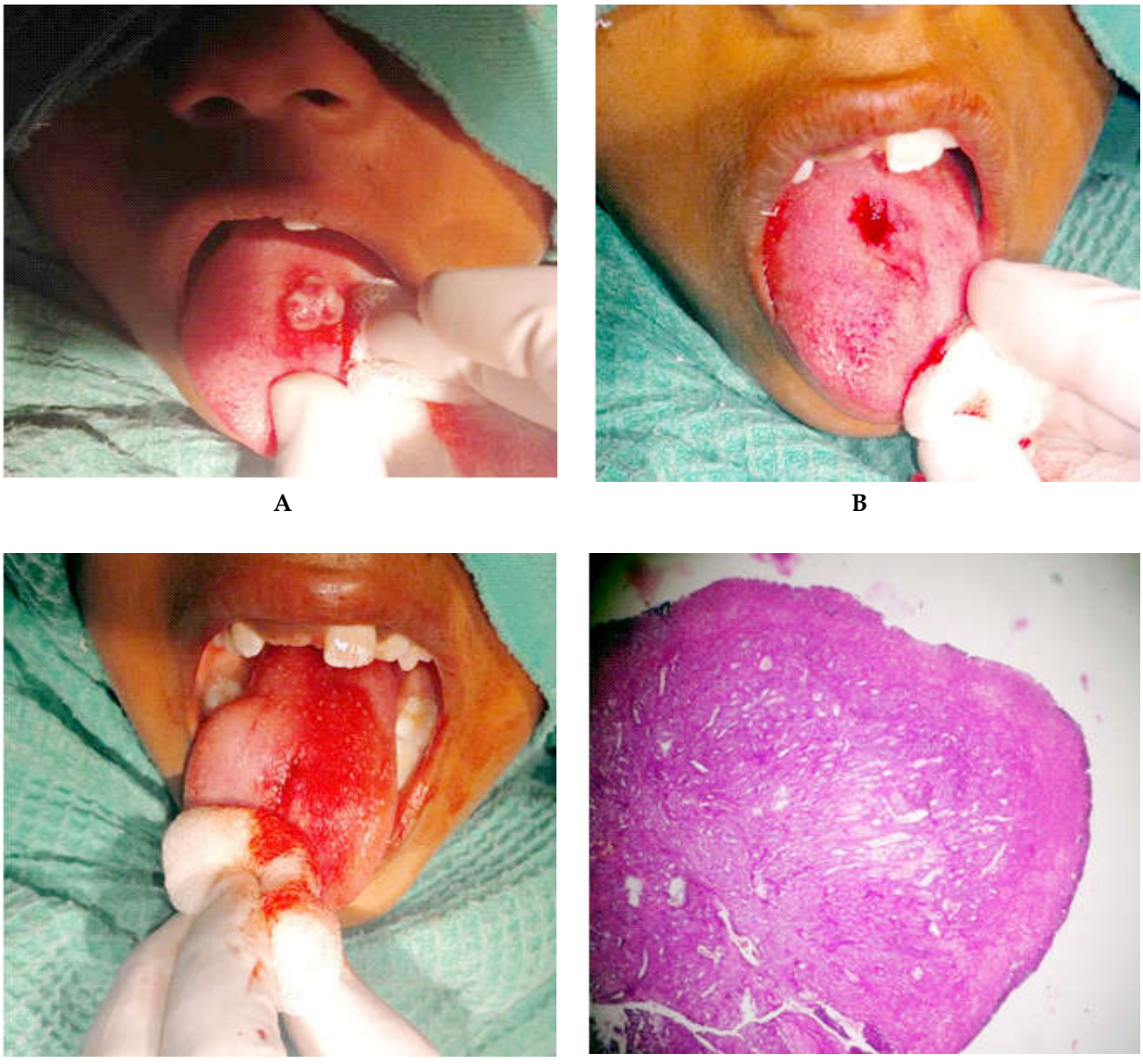
A 8 years old female child reported to Department of Otorhinolaryngology OP, Rajah Muthiah Medical college Hospital, with complaints of swelling over the dorsum of the tongue for the past two months with complaints of discomfort while eating and history of bleed from the lesion sometimes. Clinically, intra oral examination revealed, a1.0 x 0.8 x 0.5cms pedunculated growth, red in colour which was present in the midline on the dorsum of the tongue(Figure1 a). It was firm in consistency, non-tender, bled on touch. Routine laboratory investigations were normal. The lesion was excised

completely including a cuff of normal tissue (Figure1b)under local anaesthesia and biopsy specimen was sent for histopathological examination. The raw area was closed with chromic catgut (Figure 1c)

Histopathological Examination

HPE study shows tissue lined by stratified squamous epithelium showing mild hyperkeratosis, marked acanthosis and ulceration.

The underlying tissue shows a tumor mass comprising of numerous small capillary spaces lined by benign vascular endothelial cells, surrounded by fibrous stroma. Stroma is infiltrated with acute and chronic inflammatory cells (Figure 1d).



A **B**

C **D**

Fig. 1: (a) Shows red pedunculated lesion on the midline of the dorsum of tongue, **(b)** after removal of the lesion, **(c)** Sutured with catgut **(d)** Histopathological study section.

Discussion

Pyogenic granuloma is more commonly seen on the gingiva [1,10], particularly in the anterior segment [7], and uncommonly seen elsewhere in the mouth, such as the upper and lower lip, buccal mucosa, tongue,and the alveolar mucosa, particularly in edentulous regions [7]. The causative factors may be exaggerated localized tissue reaction to a trauma or any irritation like calculus, poor oral hygiene [8], nonspecific infection, overhanging

restorations, cheek biting, previous dental extractions, exfoliating primary teeth, bone spicules, root remnants, tooth brush trauma [5]. Characteristically, lateral side of tongue will be affected and may be related to trauma from adjacent structures [9]. In our case the lesion was found to arise unusually from one of the rare extra gingival site, on the midline on the dorsum of the tongue.

The pathogenesis of pyogenic granuloma can be attributed to an imbalance between inducers and inhibitors of angiogenesis. Excessive formation of Vascular Endothelial Growth Factor (VEGF), basic Fibroblast Growth Factor (bFGF) and lowering of angiostatin, thrombospondin-1 and Estrogen receptors leads to the formation of Pyogenic Granuloma [4].

Clinically these lesions usually present as a single red to purple nodule or sessile papule with smooth or lobulated surface depending upon the duration and vascularity of the lesion [5]. Younger lesions are usually red because they are highly vascular andare mainly composed of hyperplastic granulation tissue in which capillaries are prominent and older lesions are pink because they tend to become more collagenized [1,4]. Studies shows that there are two types of Pyogenic granuloma clinically, LCH type occurring as sessile mass and histologically it is characterised by proliferating blood vessels that are organised in lobular aggregates though the lesion does not undergo specific change such as edema, capillary dilation or inflammatory granulation tissue reaction. The lobular area of LCH type Pyogenic Granuloma, has a greater number of blood vessels with small luminal diameter [1,8]. The non-LCH type Pyogenic Granuloma occurs as a pedunculated mass and histologically it consists of highly vascular proliferation that resembles granulation tissue and the central area contains a significantly greater number of vessels with perivascular mesenchymal cells non-reactive for alpha- smooth muscle actin and muscle specific actin [8]. The non-LCH type Pyogenic Granulomais more frequently associated with etiological factors [1] like local irritation, trauma, hormonal imbalance.

Pyogenic granuloma varies in size from millimetres to centimetres but usually they are less than 2.5cms in size [1,5]. The lesion usually starts as small swelling and reaches its maximum size within weeks and remains stationary. It presents as a slow growing, asymptomatic and painless tumor [10]. It can be painful and may bleed if the lesion is present in areas where it subjected to repeated irritation.

The diagnosis can be made Clinically and confirmed by Histopathology [7]. Radiographs are suggested to rule out bony involvement, malignancy and to identify any foreignbody as an etiological factor [2,5].

When a mass lesion is found in the oral cavity, it is important to formulate a differential diagnosis, including Traumatic fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma, odontogenic fibroma, post extraction granuloma, metastatic cancer, kaposi sarcoma, non hodgkins lymphoma , haemangioma, basillary angiomatosis, angiosarcoma [4,5]. All excised Pyogenic granulomas should be sent for histopathological examination to rule out malignant changes and for the treatment and prognosis.

Management is based on the severity of symptoms [1]. If lesion is small, and there is no pain or bleeding, the removal of irritants and follow up can be advised. If the lesion is large,painful and bleeds, then complete surgical excision is the treatment of choice [3,5].

Other modalitieshave been proposed. Cryosurgery is the safe, easy and inexpensive procedure [6]. Nd:YAG laser excision is preferred than CO_2 laser because of its superior coagulation characteristics.In some studies, Flash lamp pulsed dye laser was used in masses which are concluded as resolute tissue, responded well to series of treatment.Ultrasonic scissors [4] is a newly developed technique and is also used in Pyogenic granuloma excision with the advantages of less blood loss and operative time. Sodium tetradecyl sulfate sclerotherapy is better alternative for excision as the technique is simple, produces less scarring even though multiple sessions are required. Series of Intra lesional corticosteroid injection, absolute ethanol]4] are used if there is any recurrence. Recurrence is believed to be due to incomplete excision, failure to remove irritants, re injury, poor oral hygiene.

Conclusion

Although the occurance of non-LCH type Pyogenic Granuloma in the Dorsum of the tongue is rare, poor oral hygiene could be a causative factor. Prevention , prompt diagnosis, management and treatment are important. Pyogenic granuloma occurs as a result of various stimuli such as low grade irritation, trauma, poor oral hygiene, hormonal factors, certain drugs, thus removal of cause is an important step in treatment. Although the diagnosis is made clinically, histopathological confirmation is mandatory for the final diagnosis and effective management.

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[1] Flink H, Tegelberg Å, Thörn M, Lagerlöf F. Effect of oral iron supplementation on unstimulated salivary flow rate: A randomized, double-blind, placebo-controlled trial. J Oral Pathol Med 2006; 35: 540-7.

[2] Twetman S, Axelsson S, Dahlgren H, Holm AK, Källestål C, Lagerlöf F, et al. Caries-preventive effect of fluoride toothpaste: A systematic review. Acta Odontol Scand 2003; 61: 347-55.

Article in supplement or special issue

[3] Fleischer W, Reimer K. Povidone iodine antisepsis. State of the art. Dermatology 1997; 195 Suppl 2: 3-9.

Corporate (collective) author

[4] American Academy of Periodontology. Sonic and ultrasonic scalers in periodontics. J Periodontol 2000; 71: 1792-801.

Unpublished article

[5] Garoushi S, Lassila LV, Tezvergil A, Vallittu PK. Static and fatigue compression test for particulate filler composite resin with fiber-reinforced composite substructure. Dent Mater 2006.

Personal author(s)

[6] Hosmer D, Lemeshow S. Applied logistic regression, 2nd edn. New York: Wiley-Interscience; 2000.

Chapter in book

[7] Nauntofte B, Tenovuo J, Lagerlöf F. Secretion and composition of saliva. In: Fejerskov O, Kidd EAM,

editors. Dental caries: The disease and its clinical management. Oxford: Blackwell Munksgaard; 2003. p. 7-27.

No author given

[8] World Health Organization. Oral health surveys - basic methods, 4th edn. Geneva: World Health Organization; 1997.

Reference from electronic media

[9] National Statistics Online—Trends in suicide by method in England and Wales, 1979-2001. www.statistics.gov.uk/downloads/theme_health/HSQ 20.pdf (accessed Jan 24, 2005): 7-18. Only verified references against the original documents should be cited. Authors are responsible for the accuracy and completeness of their references and for correct text citation. The number of reference should be kept limited to 20 in case of major communications and 10 for short communications.

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