Angiofibroma of the Cheek Mimicking Lipoma: A Diagnostic Challenge
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Abstract
Extranasopharyngeal angiofibroma are rare and the most common site of presentation is the maxillary sinus. We report a case of angiofibroma of the buccal mucosa to highlight an unusual presentation of angiofibroma in the oral cavity as reports of such cases involving the oral cavity is still very few in the literature.

Keywords: Extranasopharyngeal; Angiofibroma; Maxillary Sinus; Lipoma; Buccal Mucosa; Immunohistochemistry

Introduction
The head and neck region is the common site for vascular and fibrous tumours whether benign or malignant. The benign ones reported in the literature include angiofibroma and solitary fibrous tumor. Malignant types include low-grade fibromyxoid sarcoma (LGFMS), low-grade myxofibrosarcoma, lipomatous (myxoid liposarcoma). Differentiating between these morphologically confusing neoplasms is of considerable clinical significance because of different treatment options.

Angiofibromas are highly vascular benign tumours that characteristically arise within the nasopharynx. The tumor affects almost exclusively males and the symptoms occur at the mean age of 13–15 years. The incidence of angiofibroma in females and other age groups has been reported, but still raises controversy. Although it is the most common benign neoplasm of the nasopharynx, it accounts for 0.05-0.5% of all head and neck neoplasms. Angiofibroma has been reported to occur in other areas apart from the nasopharynx. This has been termed extranasopharyngeal angiofibroma. As at 2009, 56 extranasopharyngeal angiofibromas have been reported, with the most common site of presentation being the maxillary sinus. Other extrapharyngeal areas reported in the literature include: soft tissues of the extremities, often in relationship to joints or fibro-tendinous structures, the orbit and the heart. The buccal space is an uncommon location for these tumors, with only three previously reported cases of buccal space extranasopharyngeal angiofibromas. With this knowledge gap, more new cases of extranasopharyngeal angiofibromas need to be reported once they are diagnosed. We, therefore present a rare case of angiofibroma in the upper buccal sulcus presenting as a cheek swelling.

Case Report
A 15 year old otherwise healthy boy presented in our department with a three years history of right facial swelling. No history of trauma. The swelling was painless, soft to firm in consistency and not attached to underlie structure or the overlying skin (Figure 1). Evidence of tumour association with a peripheral nerve was also not identified. Intraoral examination revealed swelling in the right buccal mucosa, about 5x4 cm with slight bluish tinge extending from posterior region of the maxillary tuberosity till about 2-3 cm from corner of mouth. It had a lobulated surface. His past medical history was unremarkable. An impression of pleomorphic adenoma of minor salivary gland was made to rule out lipoma because of the lobulated surface. He was planned for excisional biopsy under general anaesthesia. Fine needle aspiration cytology reveals a benign tumour. Preoperative Packed Cell Volume (PCV) was 34%, platelet count was 795×10⁹/l, Partial Thromboplastin Time (PTT) was 22sec with control 16sec, Partial Thromboplastin Time in Kaolin (PTTK) was 30sec with control 38sec and International Normalised Ratio (INR) was 1.5. Blood chemistry was within normal range.

He had excisional biopsy done under GA on the 13th of March 2015 via intraoral approach (Figure 2). Gross specimen was lobulated giving an impression of a lipoma, however, because of the estimated blood loss of 1.5 litres we doubted the impression.
of a lipoma. He was transfused with two pints of whole blood and nursed in the immediate post-operative period in the Intensive Care Unit. His postoperative recovery was uneventful.

Figure 1: Extraoral preoperative photograph

Figure 2: Intraoperative photograph showing lobulated specimen

Macroscopic features of the excised tumour showed a multinodular irregular grey tissue with varying nodular size. The largest nodule measured 4.5×5cm and weighs 150g. Cut surfaces shows grey-white solid areas. Microscopy (H and E) of the excised tumour showed a benign mesenchymal neoplasm that is composed of proliferating spindle shaped cells interposed by slit-like vascular channels (Figure 3). Immunohistochemistry of the tumour showed positive CD 34 staining of vascular endothelium (Figure 4) while vimentin staining (Figure 5) showed positive fibroblastic stroma. Histological diagnosis of Angiofibroma was made.

Figure 3: Sections of angiofibroma showing vascular channels of varying sizes (black arrows) and loosely fibrous stroma (green asterisk). H & E X 100

Figure 4: Immunohistochemical stain of angiofibroma showing CD34 positive vascular endothelium (black arrows). X 100 magnification

Figure 5: Immunohistochemical stain of angiofibroma showing Vimentin positive fibroblastic stroma (black arrows). X 100 magnification
DISCUSSION
Angiofibromas was first described by Hippocrates in 5th century BC and Friedberg first used the term angiofibroma in 1940. They are histopathologically benign but potentially locally destructive vascular tumors. They are un-encapsulated neoplasms composed of a rich vascular network within a fibrous stroma. The smaller vessels in the central portion of the lesion typically lack muscular elastic laminae and the absence of muscular coat contributes to the capacity for massive bleeding that occurs with angiofibromas. This feature of massive bleeding in angiofibromas was encountered intraoperatively in our case by blood loss of about 1.5 litres which necessitated transfusion of 2 units of whole blood.

In a review of 704 cases of angiofibroma by Tasca and Compadreti (2008), 13 cases presented outside the nasopharynx suggesting extranasopharyngeal possibility. Windfuhr and Remmert (2004) reviewed the literature and compiled 65 cases of extra nasopharyngeal angiofibromas in which four cases had oropharyngeal origin and the maxilla was the most commonly affected site. Extranasopharyngeal angiofibromas do not present clinically in terms of symptoms, age and sex as nasopharyngeal angiofibroma. Because of these reasons, there is controversy whether extranasopharyngeal angiofibromas should be considered as a separate entity from nasopharyngeal angiofibroma. Celik et al opined that this type of angiofibroma should be termed “atypical angiofibroma”. In our study, the patient is a male and 15 years of age, which is in agreement with male predilection and age bracket reported in the literature. However, other studies have given a female predilection and other age brackets but these are still controversial.

The present case did not present with nasal symptoms such as rhinorrhea, epistaxis, snoring, foreign body sensation, halitosis, headache, post nasal drip and loss of sense of smell typical of nasopharyngeal angiofibroma, we then speculate that this may be another case of buccal space extra nasopharyngeal angiofibroma.

In conclusion, extrapharyngeal angiofibroma should be considered as part of differentials of lobulated intra oral swellings.

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