Haemangioma in the Oro-Facial Region: A Report of Fifteen Cases and Review of Literature

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ABSTRACT

Objective: A haemangioma is a benign vascular tumour consisting of an abnormal overgrowth of tiny blood vessels. Haemangioma may not be present at birth but may appear at 6 months of life. This vascular tumour is not common. We therefore report fifteen cases of haemangioma occurring in the oro-facial region: eight occurred in the cheek, one in the floor of the mouth, four in the lower lip and two in the maxillary gingivae. The objective of this study was to outline the clinical presentation and management of haemangioma in the oro-facial region treated in our Centre.

Methods: This is a retrospective study of all the patients that were treated from July, 2009 to July, 2019. Information was extracted from case files of patients. Data collected included: age at presentation, sex, location of the tumour, treatment given, findings and histological diagnosis.

Results: The result showed fifteen cases, nine (60.0%) were males and six (40.0%) females. Male to female ratio is 1.50 to 1.00. Our diagnostic tool was the aspiration of frank blood from the tumour which had similar clotting time with the normal blood and our mode of treatment included: injection of sclerosants and excision of the fibrous tissues.

Conclusion: It is essential to treat the tumour as early as possible especially in large lesion with disfigurement or where it is complicated by haemorrhage or infection.

Keywords: Cheek, haemangiomas, oro-facial, sclerosants, tumour, vascular

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INTRODUCTION

A haemangioma is a benign vascular tumour consisting of an abnormal overgrowth of tiny blood vessels.[1] They are believed to be harmatomatous proliferations of vascular endothelial cells.[2] It is an uncommon vascular tumour and is benign endothelial cell neoplasm, that is typically absent at birth but may appear at 6months of life. However, of the entire patients who eventually develop haemangioma, 30% of them have evidence of their presence at birth, while 100% have manifested by age 6 months.[3] Characteristically, they have rapid growth in infancy with spontaneous involution later in life.[2] This contrast with another known group of childhood vascular anomalies;- such as capillary and venous malformations, lymphangiomas and arterio-venous malformations which are present at birth and are characterized by very slow growth with persistence into adult life. Haemangiomas are classified on the basis of their histological appearance as capillary, mixed cavernous or a sclerosing variety that tends to undergo fibrosis.[4] They are more common in premature infants and in girls.[2] Rosca et al.[3] in their study among Caucasians concluded that female patients were more affected than male patients by ratio of 3:1. It could be found anywhere in the body, but the skin, subcutaneous tissues, eyelids, cheek, tongue, mucous membranes of oral cavities are common site of involvement[4], the liver, spleen and kidneys can also be involved.[5] About 50% of haemangioma occurs in the head and neck region.[4] Jennifer et al.[6] in their study also stated that

haemangiomas are commoner in the head and neck region. It is also the most common tumour of orbit and peri-orbital areas in childhood.[3]

However, systemic involvement of haemangioma can be a significant source of morbidity and mortality.[5] The purpose of this study is to elucidate the clinical presentation and management of haemangioma in the oro-facial region that were treated in our hospital. Fifteen cases were reported and their common complaint was facial disfigurement.

MATERIALS AND METHODS

A retrospective study of patients who were treated for haemangioma in the oro-facial region at Barau Dikko Teaching Hospital, Kaduna State University, Nigeria, over a period of ten years from July 2009 to July 2019 was done. Fifteen cases were included in this study. Only those that accepted our mode of treatment were included, two adult female patients with lower lip tumour, and one adult male with cheek tumour who declined treatment were excluded. The patients were analyzed for age, sex, site of the tumour, treatment modalities, histological findings and recurrences. Data were sorted, organized and entered into SPSSS version 20 (IBM SPSS statistics Armonk New York, United States) for analysis. Frequency statistics and cross tabulations were done and chi-squared test was used to test for significance between variables at the critical $p<0.05$. Ethical approval was obtained from the ethical committee of the hospital.

RESULTS

Fifteen patients were treated, nine (60.0%) were males and six (40.0%) were females. The ratio of males to females is 1.5:1.00. Age range of patients was 4months to 39years (Table 1). Eight (53.3%) of the patients had the tumor in the cheek unilaterally (cases 1,3 8, 9,12-15), four (26.7%) had it in the lower lip (cases 4,5,10,11), two (13.3%) had it in maxillary gingiva (cases 6,7) and one (6.7%) had it in the floor of the mouth (case 2). The histology showed capillary type (n=11, 78.6%), cavernous type (n=3, 21.4%).

Table 1: Age, sex and other parameters

Cases	Age	Sex	Site	Treatment	Histology
1.	4months	Male	Cheek	*Sclerotherapy and Excision*	Capillary
2.	3years	Male	Floor of the mouth	Sclerotherapy only	------
3.	6 years	Female	Cheek	*Sclerotherapy and Excision*	Capillary
4.	16 years	Female	Lower lip	*Sclerotherapy and Excision*	Cavernous
5.	24 years	Female	Lower lip	*Sclerotherapy and Excision*	Capillary
6.	26 years	Female	Gingiva	Excision	Lobular capillary
7.	27 years	Female	Gingiva	Excision	Lobular Capillary
8.	29 years	Male	Cheek	*Sclerotherapy and Excision*	Capillary
9.	30 years	Male	Cheek	*Sclerotherapy and Excision*	Capillary
10.	31 years	Female	Lower Lip	*Sclerotherapy and Excision*	Capillary
11.	32 years	Male	Lower lip	*Sclerotherapy and Excision*	Capillary
12.	36 years	Male	Cheek	*Sclerotherapy and Excision*	Cavernous
13.	37 years	Male	Cheek	*Sclerotherapy and Excision*	Capillary
14.	39 years	Male	Cheek	*Sclerotherapy and Excision*	Cavernous
15.	39 years	Male	Cheek	*Sclerotherapy and Excision*	Capillary

Extra oral examination of the cheek tumour revealed a solitary, pedunculated, spherical and baggy shaped, reddish pink swelling with distinct order and irregular surface (Figure 1A). On palpation, the swelling was non-tender, soft to firm in consistency, not blanching on pressure. The outcome post-surgery (Figure 1B). Extra oral examination of the lower lip tumor revealed enlarged lip with baggy and spherical shaped, tensed and reddish. The tumour in the floor of the mouth bulges out from the floor the mouth and the submandibular region extra-orally (Figure 1). Aspiration biopsy of all the tumours yielded frank blood which had similar clotting time with normal blood, and a diagnosis of haemangioma was made. The two in the gingival presented as haemorrhagic tumour and the histology showed

lobular capillary haemangioma. All our patients had haematological and biochemical investigations done and their values were within normal range. The surgical procedures were done under local and general anaesthesia.

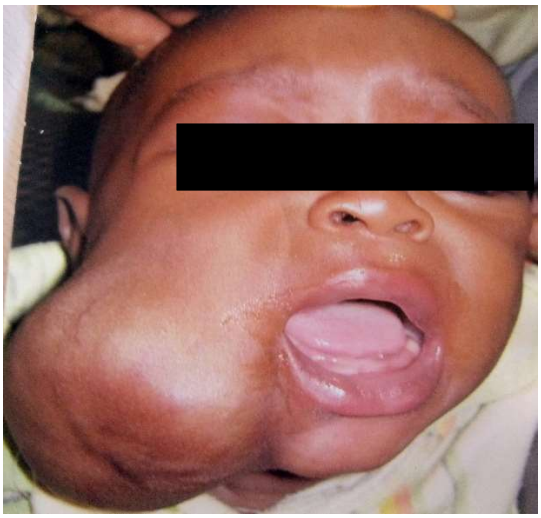

Figure 1A: A 4month old boy with cheek tumour.

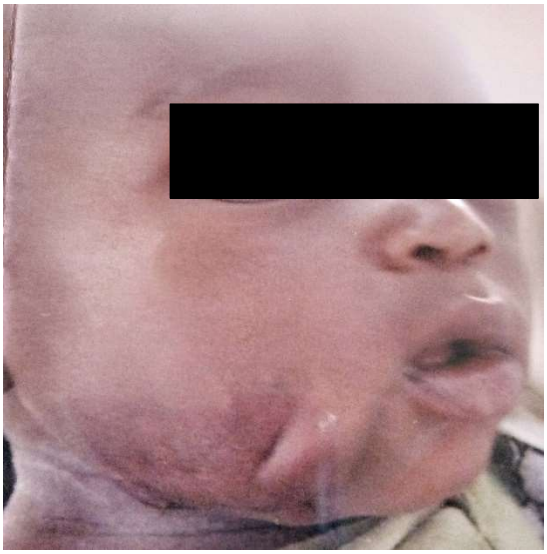

Figure 1B: The boy after surgery

Figure 2: A 3 year old boy with the tumour in the floor of the mouth.

Surgical Procedure

Sclerotherapy which involves the intra-lesional injection of boiling normal saline after withdrawal of blood from the lesion, an amount which is more than the blood withdrawn was injected into the lesion. This was repeated two or three times at three weeks intervals until it was only little or no blood that was aspirated. Then surgical excision of the fibrous tissues was done for the tumour, in lower lip and cheek. Excisions of the gingival tumours were done under local anaesthesia. The excised tissues were sent for histology and the report showed proliferation of endothelial lining of the blood vessels and encapsulated aggregates of closely packed, thin walled capillaries with endothelial linings and perivascular fibrosis, a diagnosis of capillary haemangioma was made for cases 1,3,5-11,13,15) (Figure 3). For cavernous haemangioma the histology showed thick walled blood vessels and anastomosing vascular channels of cavernous

pattern (cases 4,12,14) (Figure 4). The patients were reviewed after 6months of surgery and there was a recurrence in case 3 only and was rescheduled for excision surgery.

The outcomes of the treatments were satisfactory.

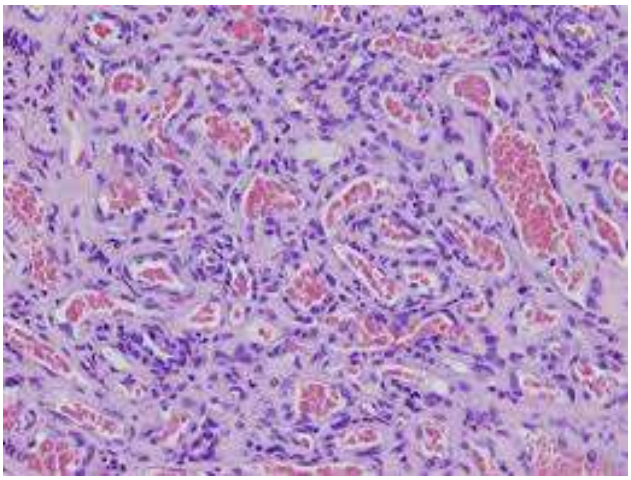

Figure 3: Photomicrograph of capillary hemangioma.

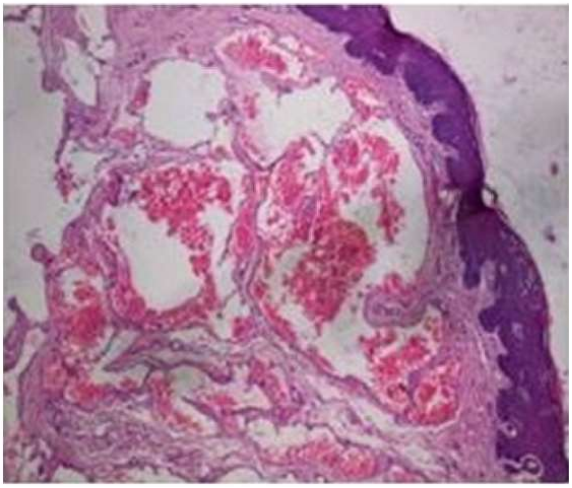

Figure 4: Photomicrograph of cavernous hemangioma.

DISCUSSION

A haemangioma is a benign tumor of infancy. It is a tumour consisting of an abnormal overgrowth of tiny blood vessels, common in premature infants, and girls are most affected.[2] It is typically absent at birth and characteristically has rapid growth in infancy with spontaneous involution later in life.[3] The cases 1-5 reported underwent rapid growth in three months. Haemangiomas are now thought to be of placental origin due to a unique micro-vascular phenotype shared by juvenile haemagiomas and human placenta.[6]

Haemangiomas generally exhibit two phases of growth: a proliferative phase and an involution phase. The proliferative phase of rapid growth typically occurs from 8-18 months. The involution phase is characterized by slow regression of the haemangioma.[7] Cases 8-15, who are adults claimed the tumours regressed after they crossed puberty. One half of all lesions will involute by age 5years, and 75% will involute by age 7years, during this stage the mast cells numbers decrease to normal and there is decrease in endothelial and mast cell activity.[8]

Epidemiology shows that as many as 50 percent of systemic haemangioma occurs in the head and neck region.[4] The cases in our study occurred in the cheek, floor of the mouth, maxillary gingivae and lower lip in the head and neck region. Of all the patient who eventually develop haemangioma, about 30% of them have evidence of their presence at birth, while 100% have manifest them by age of 6months.[4] Cases 1-3, patients first noticed the tumour when the child was born, and cases 4-5 when the child was two weeks old.

However, systemic Involvement with haemangioma can be a significant source of morbidity and mortality.[6] The cases reported did not have any systemic involvement, with the full blood count of the patients within normal limit.

Rosca et al.[3] in their study of vascular tumours in the orbit reported female predominance at ratio 3:1[3] in contrast to this study where ratio of male to female is 1.50 to 1.00. This may be because majority of our patients were adults who cared about facial disfigurement at later age. Radiology is usually only required when the diagnosis is unclear, and this is most frequent in lesion with a deep component. Histology of haemangiomas shows proliferations of endothelial cells of small blood vessels.[4] Yusuf et al.[9] in their study of vascular tumours in Northern Nigeria reported that capillary haemangioma accounted for 40.3% and is the most common of the entire vascular tumours studied. In our study it accounted for 80%, this is because we studied only haemangioma not all vascular malformations. The two gingival tumours in this study occurred in adult females and were lobular capillary haemangioma otherwise known as pyogenic granuloma. Haemangioma has

demonstrated predilections for head and neck region and structures there are specially affected are: lips, nasal, and oral cavities.[9] It has tendency to affect children and young adults.[9] This study also showed the same. Ultrasonography of haemangioma is characteristics of hyperechoic and compressible lesion with high peak intra-tumoral vessels on arterial shift and is most useful for smaller, limited lesion.[8] Ultrasound of cheek tumors in our study showed hyperechoic and compressible lesion. Computerized tomography appearance is that of a strongly enhancing homogenous lobulated mass with thin septa.[8] Magnetic resonance imaging is usually hypotensive in Ti, Iso to hyper intense on T2 with multiple serpiginous flow voids.[8] Indications for treatment of haemangioma includes: functional impairment such as visual or feeding compromise, bleeding which is potentially life threatening and risk of long term permanent disfigurement. Facial disfigurement and undue pressure on contiguous tissue prompted our patients to seek treatment, except case 2 with the tumor in the floor of the mouth that found swallowing difficult (Figure 3).

Treatment of haemangiomas consists of injection of corticosteroids, intratumoural laser therapy has been used for longer lesion and recalcitrant cases, interferon or vincristine can be considered.[9, 10] Sclerotherapy, embolization and surgery have been found useful.[9, 10] Some of our cases had sclerotherapy with injection of hot normal saline and excision of fibrotic tissue (cases 1,3,4,5,8-15) under general anesthesia, Cases 6,7 had excision of gingival tumor done under local anaesthesia, case 2, a 3year old boy had sclerotherapy only, excision of tissue was not necessary because the fibrotic tissue had fused into the mandible. Review of our patients after six months showed evidence of recurrence only in case 2, who later had excision of the tumour.

CONCLUSION

Haemangioma in the head and region need urgent attention to avert long term permanent disfigurement.

Source of Support
Nil.

Conflict of Interest
None declared.

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