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CASE REPORT

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Orofacial haemangioma: Series of two cases managed at University of Maiduguri Teaching Hospital and review of literature

Kefas Mari Mbayas[1], Mujtaba Bala[2], Chubado Tahir[3], Ahmed Ahidjo[4], Audi I Kwari[1], Umar Hassan[4]

[1]Department of Surgery, University of Maiduguri Teaching Hospital, Maiduguri, Nigeria
[2]Department of Oral and Maxillofacial Surgery, Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria
[3]Department of Surgery, University of Maiduguri, Maiduguri, Nigeria
[4]Department of Radiology, University of Maiduguri Teaching Hospital, Maiduguri, Nigeria

ABSTRACT

Haemangioma is a benign vascular proliferation of blood vessels commonly found in children. Head and neck haemangioma constitute about 60%–70% but those in the orofacial region are relatively rare. Whereas some categories of these lesions involute over time, some definitely require medical or surgical intervention or both. We present two cases of vascular lesions in two patients who presented with a history of discoloration and painless progressive facial swelling clinically, radiologically, and histologically diagnosed as haemangioma. Both cases were successfully treated with sclerosant therapy as well as surgical excision. There was uneventful healing and favorable clinical outcome during the postoperative period and subsequent follow up. Additionally, relevant literature in terms of clinical presentation and management has also being highlighted. Surgical treatment following oral propranolol and intralesional triamcinolone has shown to be a very effective treatment modality.

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Haemangioma; sclerosant; vascular tumour; buccal mucosa

Introduction

The vascular lesions are mostly grouped into two major groups namely; haemangioma and vascular malformation [1]. A haemangioma is a benign and a more common vascular lesions consisting of an abnormal overgrowth of tiny blood vessels [2]. Haemangioma may not usually appear at birth but could be seen from 6 months of life [2,3]. Haemangioma can occur in all parts of the body, however, in the head and neck regions, they are relatively common [4]. The presence of these lesions in the oral cavity is rare and most commonly occurs on the lips, tongue, palate, and buccal mucosa [5].

The diagnosis of this condition has been through history, clinical examination, radiological and histological findings [4]. Haemangioma present with serious challenge in management and requires careful evaluation and prompt medical and surgical management.

Case 1

An 18-month-old baby girl presented to the Department of Plastic and Reconstructive Surgery, the University of Maiduguri Teaching Hospital with purplish discoloration and swelling of the left cheek and both upper and lower lips since birth. The swelling was ovoid, non-ulcerated, and measures about 7 × 6cm in diameter with extension to the upper and lower lip (Fig. 1) The swelling was said to progress with associated distortion of the face. However, there were no associated difficulties in feeding or breathing. A diagnosis of haemangioma was made based on clinical examination and confirmed through contrast-enhanced computed tomography (CT) scan and histological investigation of the excised mass. Oral propranolol was initiated augmented with intralesional injection of triamcinolone. The patient presented 3 months later to the accident and emergency with associated profuse bleeding which was controlled by the application

Contact Mujtaba Bala ✉ mujtababala@yahoo.com Department of Oral and Maxillofacial Surgery, Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria.

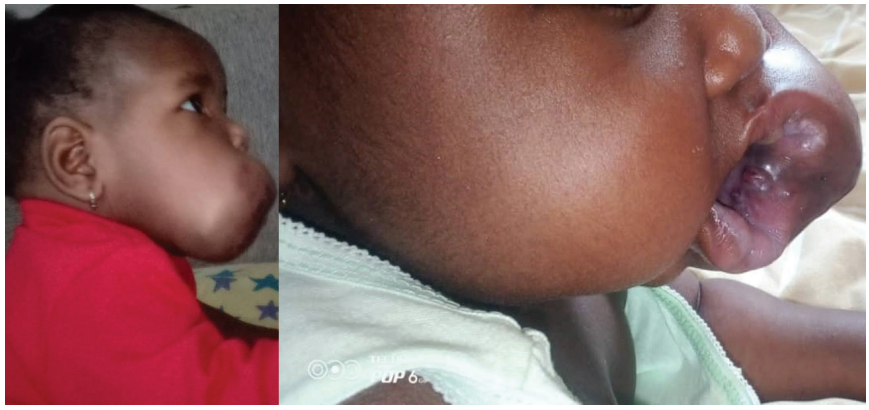

Figure 1. Preoperative photograph showing the cheek lesion (Right) and upper and lower lip extension (Left).

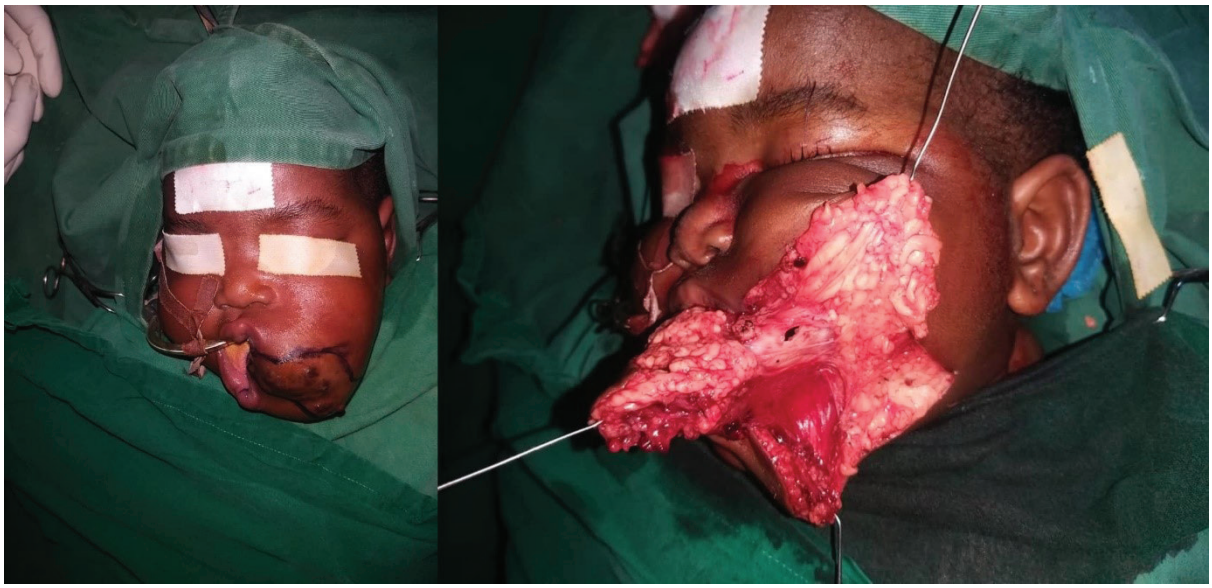

Figure 2. Intraoperative clinical photographs; the surgical marking (right) and flaps (left).

of pressure and suture ligation. At this juncture, a decision for surgical intervention was made and counseling was done. The patient had excision of the haemangioma with direct closure of the cheek, upper and lower lips (Fig. 2), and left commissuroplasty after 7 months of the initial surgery. The patient had 5 courses of intralesional triamcinolone postoperatively and had uneventful follow up thereafter. Figures 3 and 4 show the immediate postoperative photographs of the patient and at 5 months follow up, respectively.

Case 2

A 26-year-old female presented with a 26-year-old history of left cheek discoloration and painless progressive swelling which gradually involved both the upper lower lips. There was a previous history of recurrent bleeding that warrant blood transfusion about 7 years ago. However, there was an associated history of difficulty in chewing and recent occasional pain. On examination, there was a diffuse discolored cheek swelling measuring about 6–8 cm with extension into the upper and lower lip and a CT scan showing evidence of the mass (Fig. 5). A provisional diagnosis of haemangioma was made based on the clinical judgement which was confirmed with imaging and histological examination. The patient had six sessions of embolization by interventional radiologists using 99% ethanol and doxorubicin. She also received 5 courses of intralesional triamcinolone to the lips and later had lesional excision of left cheek, upper and lower lip with cheek and lip reconstruction with favourable follow up outcome (Fig. 6). Shows a section of tissue composed of poorly circumscribed collections of large ectatic vessels lined by endothelium and containing red blood cells. Shows the postoperative

photograph of the patient at immediate post operation and at 6 months post-operative period.

Surgical technique

All the 2 cases were operated under general anaesthesia following consent taking and detailed explanation of the procedure and possible complications to the caregivers. After induction, 2% lignocaine with 1:100,000 adrenaline was injected at the area of planned incision and waited for about 7 minutes. Stay sutures were applied throughout the circle of the incision (Fig. 5). A size 15 blade was used to initiate skin incision and there after deepened by a combination of blunt and sharp dissection taking vital structures in to cognisance. Stenson's duct, parotid gland tissue, and healthy mucosa were preserved. Bipolar diathermy was used to achieve haemostasis and direct closure was done (Fig. 6). Additional commissuroplasty was done in case 1 after 7 months.

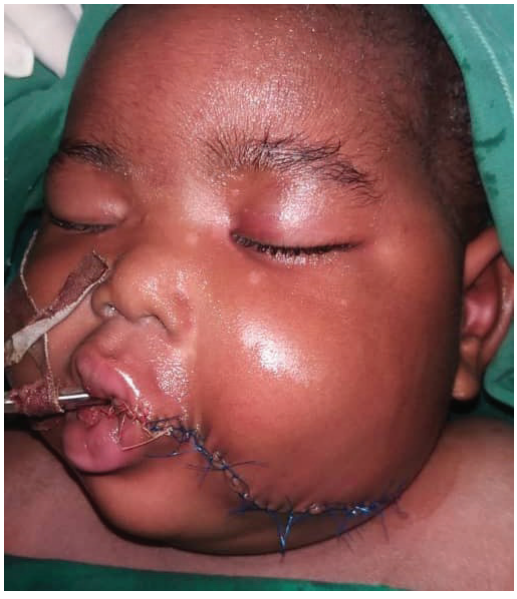

Figure 3. Immediate postoperative clinical photograph.

Discussion

Orofacial haemangioma is a benign lesion that develop due to endothelial cell proliferation of blood vessels in and around the oral cavity [6]. Haemangioma usually becomes apparent few

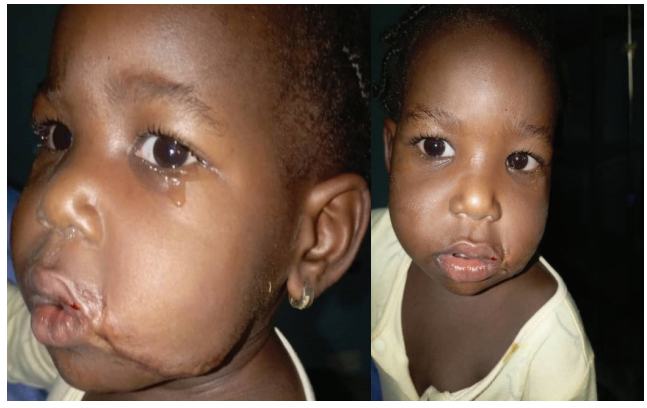

Figure 4. Postoperative photographs at 5 months follow up.

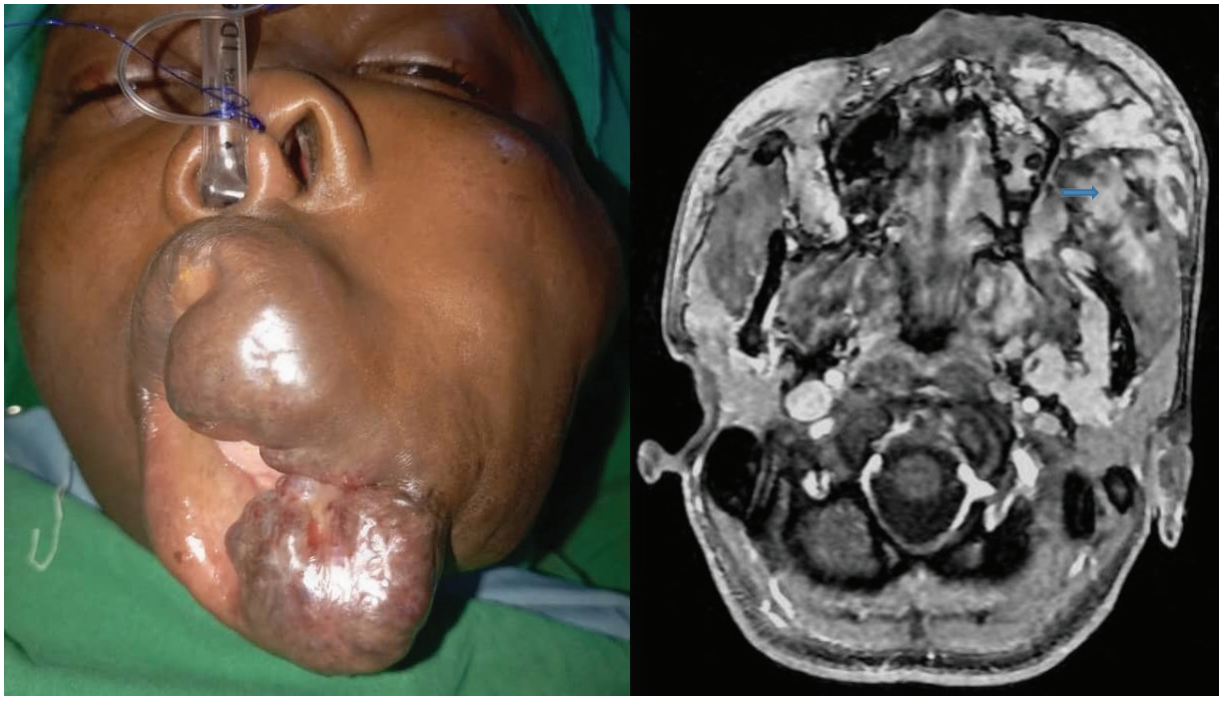

Figure 5. Preoperative photograph of the case 2 patient showing the extent of the lesion (Right) and axial magnetic resonance imaging (MRI) showing hyper intense mass (arrow) at the left face with facial asymmetry (Left).

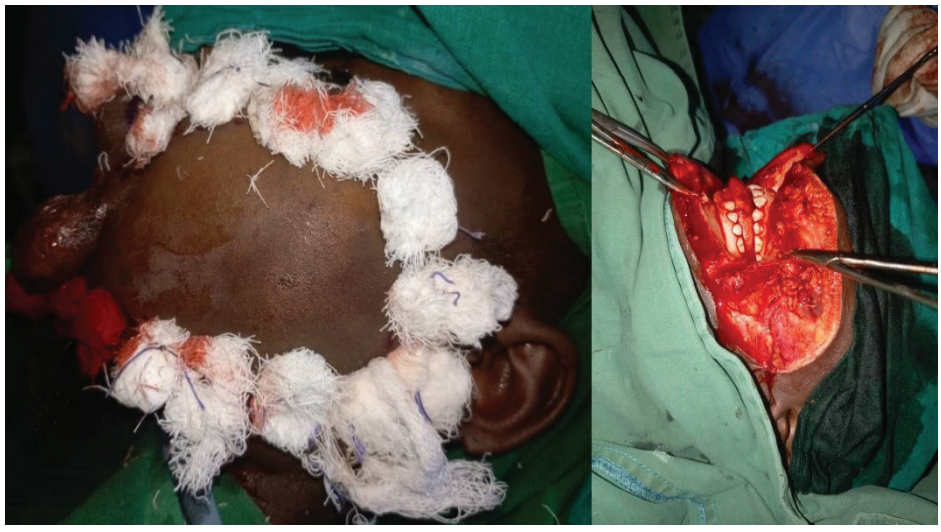

Figure 6. Intraoperative clinical photographs showing stay suture (Right) and the site after the tumour excision (Left).

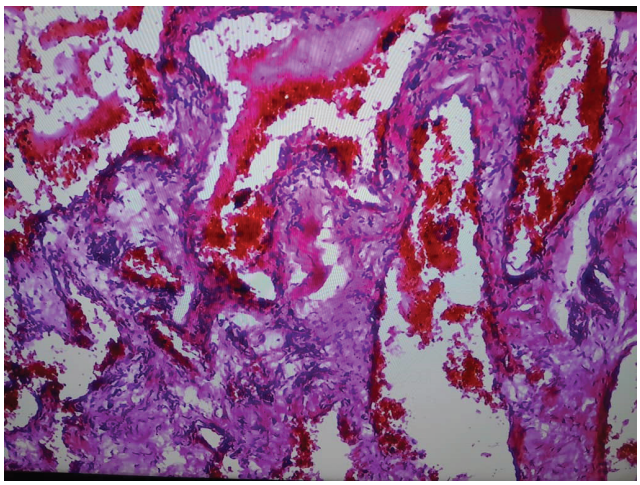

Figure 7. Section shows tissue composed of poorly circumscribed collections of large ectatic vessels lined by endothelium and containing red blood cells H & EX 10 magnification.

months after (Figs. 7–9) birth and continues to progress or regress depending on the type [7]. About 60% of all cases of haemangioma occur in the head and neck region with relatively rare occurrence in the oral cavity [8]. The sites of occurrence include gingiva, tongue, lips, buccal mucosa, palatal mucosa, salivary glands, alveolar ridge, and jaw bones. In some of the cases multiple sites are affected just like the two current cases in which buccal mucosa, cheeks, upper and lower lips are all affected.

Several classifications of haemangioma exist in the current literature [9]. Clinically, haemangiomas can be classified as juvenile (infantile) or congenital. Infantile hemangiomas arises during the first 2 months of child life and shows rapid proliferation between 6 and 12 months of age, followed by a period of slow involution [10] Majority of infantile haemangiomas spontaneously regress between 6 and 9 years of age [10,11]. On the contrary, congenital hemangiomas are usually present at birth, do not exhibit a proliferative phase, and may rapidly involute [11]. The majority of haemangioma will completely involute, with 10%–20% persisting into adolescence or adulthood [9].

Based on the depth of involvement, it can be classified as superficial (those occuring on skin surface), deep (those occuring under the skin surface) and combined [11]. Based on the size of the blood vessels, haemangioma can be histologically classified into capillary and cavernous type [9,11]. The capillary type is characterized by small thin-walled vessels of capillary size that are lined by a single layer of flattened endothelial cells and bounded by a discontinuous layer of pericytes and reticular fibres [10,11]. The cavernous type is composed of large blood-filled endothelial tubes that are separated by a scanty connective tissue stroma [10].

The diagnosis of haemangioma has been majorly based on clinical evaluation. A color-doppler ultrasound is usually ordered especially if treatment is desired [12]. Ultrasonography is non-invasive, non-radiation, and cost-effective modality that is capable of providing vascular and its morphological information of the lesion [13]. For deeper tissue involvement, CT angiography and a contrast-enhanced MRI imaging modality are considered desirable and capable of revealing fine details in intramuscular and intraosseous haemangioma [12]. In the two cases presented here, both ultrasonography and contrast-enhanced MRI were done and

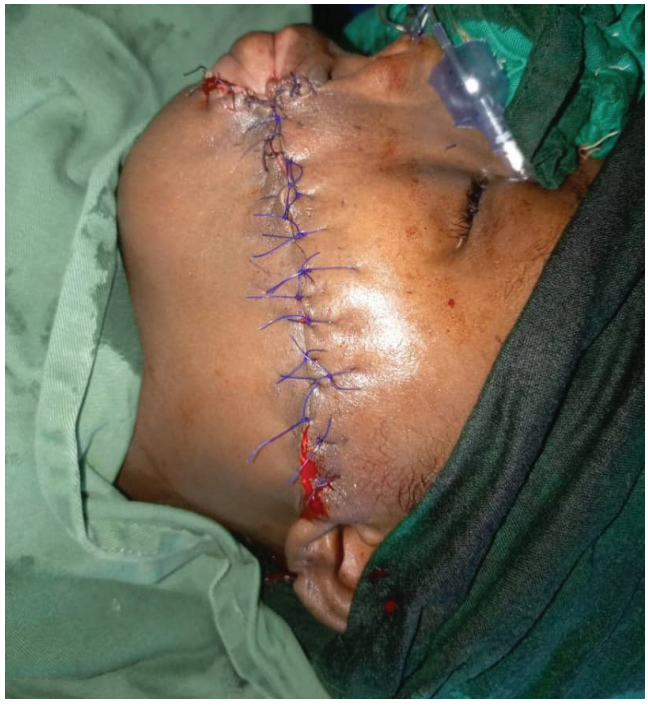

Figure 8. Immediate postoperative photograph.

Figure 9. Clinical photograph showing 6 months postoperative photograph of the patient Surgical technique.

proved effective in the management of the cases. Due to the high chances of bleeding, an incisional biopsy is avoided. It may be done with great caution especially in superficial lesions, if malignancy is strongly suspected, or in case of fibrosis secondary to sclerotherapy. Histological examination of the two excised lesions revealed large blood-filled spaces lines by flat endothelial cells with an incomplete advential rimming in keeping with cavernous haemangioma.

The treatment of haemangioma include conservative (non-interventional), medical, and surgical options [14]. However, there is no standardized treatment protocol at our end. Due to unnumbered possible complications, treatment of haemangioma is typically not chased unless functional impairment exists. The medical treatment involves oral propranolol, or sclerotherapy in form of intralesional steroid, boiling water, or sodium tetradecyl sulphate. Sclerotherapy may be to reduce the size of the lesion before surgical intervention [15]. Surgery is indicated in case of non involuting haemangioma or cases with functional and aesthetic impairment [16]. The present cases were managed by both medical and surgical models. Stay sutures were found very useful in achieving haemostasis and vital structures were also preserved. The healing occurred uneventfully and no complications recorded during the intraoperative or postoperative period. The surgical treatment provides definitive for symptomatic haemangioma located on the lips, cheek, and buccal mucosa. Some literature do not support resection of large lesions that may cause significant impairment rather advocated sclerotherapy [17,18]. Thus studies have demonstrated minor adverse reactions with sclerotherapy treatment, but the risk of thrombosis with embolization has been a concern [18].

Conclusion

Orofacial haemangioma can present with varying symptoms and complications warranting surgical treatment. Surgical treatment following oral propranolol and intralesional triamcinolone has shown to be very effective modality.

Conflict of interest

None declared.

Source of funding

Nil.

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