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TRANSORAL EXCISION OF HEMANGIOMAS AND LYMPHANGIOMAS OF THE LIPS AND CHEEK

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Hemangiomas and lymphangiomas are considered vascular malformations of either hamartomatous or neoplastic origin, that may involve arterial, venous, or lymphatic vessels. Hemangiomas and lymphangiomas constitute the largest group of tumors or tumor-like malformations in childhood with high prevalence in the periorbital region, the lips and cheek. Lesions of the lips and cheek represent a surgical challenge being related to important structures in addition to the risk of disfigurement following direct excision. Twenty five patients with hemangiomas and lymphangiomas of the lips and cheek were treated by transoral excision. The lesions affected the lower lip in 8 patients, the upper lip in 2 patients, the cheek in 11 patients, the cheek and extending to the upper lip in 2 patients and the angle of the mandible in 2 patients. Evertion of the lip or cheek and division of the gingivolabial sulcus, elevation of the buccal or labial mucosa, isolation of parotid duct and division of the buccinator muscle if needed. The whole lesion can be traced and excised even those reaching the lower eyelid. Complete excision of the lesion was feasible in 19 patients without residual swelling or discoloration. In patients with mixed hemangiomas, excision of the cavernous part was followed by fading of the capillary element within one year. In patients with cheek lesions, none had parotid duct or buccal nerve injury. One patient with ulcerated hemangioma of the lip had its ulcer remaining after disappearance of the swelling. Conclusions 1- This approach carried the least postoperative morbidity without scar on the skin. 2- Mixed hemangiomas should be treated by excision of the cavernous element only without increasing the risk of damaging the covering layer during excision of the capillary part as the later will fade by time. 3- Hemangiomas of the lip should not be treated conservatively as ulceration will compromise further surgical ablation.

INTRODUCTION

The hemangioma and lymphangioma are a common pediatric tumors consisting of a proliferation of blood vessels or lymphatics of variable diameter. The proliferation is considered to be hamartomatous rather than neoplastic in most cases. About 85% of these lesions develop by the first year of life, the head and neck being involved in 56% of cases [(1)]. They occur in 1% to 2% of neonates with higher incidences found in Caucasian infants and preterm babies. Females are more commonly affected than males. The lips, tongue and buccal mucosa are the most common oral sites. Hemangiomas and lymphangiomas, when considered together, they comprise about 30% of all oral tumors in children [(2)]. A minority of hemangiomas are associated with other congenital anomalies. To develop a rational approach to the management of hemangiomas, the individual practitioner must possess a thorough understanding of the natural history and complications associated with common vascular tumors. The practitioner also needs to be aware of the range of treatment possibilities and the potential problems that are associated with them. When treatment is indicated, general guidelines as to which lesions should be treated and how they should be treated are difficult to define. Radiation treatment to oral and paraoral structures can have serious sequelae. Successful treatment of oral hemangiomas was reported with cryotherapy [(3)]. Encouraging and often dramatic results were reported with the injection of sclerosing solutions [(4,5)]. Extreme surgical difficulty can be encountered because of hemorrhage and infiltration of hemangioma around vital structure in the head.

MATERIALS AND METHODS

Twenty five patients with hemangiomas and lymphangiomas of the lips and check were included in this prospective study. The patients' ages ranged from 8 months to 47 years (mean 16 years, 18 males & 7 females). The lesions were in the form of 4 lymphangiomas and 21 hemangiomas and involved a variety of anatomic locations in the head (Table 1). Seven hemangiomas were in the lower lip and varied in size from few millimeters (Fig 1) to several centimeters involving the whole lip. Five lesions of the lower lip seeked medical advise in their early life (8 months to 3 years) while the other two were examined beyond the 19th year of age. A case of hemangioma of the lower lip developed ulceration by the age of two years. Two hemangiomas of the upper lip were presented as elevation of the skin but the lesions and discoloration were more evident on the mucosal side (Fig 2). Nine hemangiomas and 2 lymphangiomas were observed in the cheek. Six of these hemangiomas were soft and compressible. One showed variable consistency with areas of calcification (Fig 3), and two have had discoloration of the overlying skin reaching the lower eyelid" strawberry mark" in addition to the cavernous part involving almost the whole cheek (Fig 4). One hemangioma and one lymphangioma affected the cheek and upper lip as a single continuous lesion, while the angle of the mandible was the site of two hemangiomas. All patient families had the procedure fully explained. Radiographic studies in the patients included ultrasonography with color doppler examination (n=14), computed tomography (n =2), magnetic resonance imaging (n=2).

Operative technique of transoral excision.

In lesions of the lip, identification of the extent of the lesion and its predominant structure is carried out while the patient is anaesthetized by digital compression or by using a glass slide as capillary hemangioma will blanch but not the cavernous type. Evertion of the lip until the mucosal reflection is visualized, the gingivolabial sulcus is divided transversely, the mucosal lining of the lip is dissected carefully till the lesion is reached. At this conjuncture, the lesion is traced at its peripheral limit, cauterizing all the feeding veins, separating the lesion out of the surrounding muscles of the lip stopping short of the mucosa usually leaving a thin rim of the capillary element attached to the mucosa (Fig 5). The redundant mucosa is then trimmed and the wound is closed without drain using 6/0 absorbable sutures. In lesions of the cheek, the angle of the mouth is retracted and the ckeek is everted to expose the lesion through the oral cavity (Fig 6). The parotid duct orifice is identified and the duct is cannulated to avoid its injury during the excision process. The buccal mucosa is divided transversely, extending the incision anteriorly if the lesion involves the upper lip, the buccinator muscle fibres are split apart if needed, and the lesion is picked up and traced sequentially in all directions (Fig 7), untill the whole lesion is taken out of the mouth (Fig 8). The wound is closed with a small Penrose drain at the anterior angle of the incision.

RESULTS

(Table 2), summarizes the clinicopathological types of the lesions. Five hemangiomas and one lymphangioma were of capillary origin. The capillary hemangioma was characteristically bright red, raised, well circumscribed and only partially compressible. None of the capillary hemangiomas was detected at birth but appeared several weeks later either as a red macular stain, telangiectases, or a pale anemic nevus. The hemangioma proliferated rapidly and extension occurred in multiple sites within the same area in all five patients. None of capillary hemangiomas but one developed ulceration at the age of two years and healed with scar in response to conservative measures. Complete excision of capillary lesion was feasible in the case of lymphangioma and 4 out of 5 hemangiomas without residual swelling or discoloration (Fig 9). Excision of the ulcerated hemangioma resulted in fading of the lesion but persistence of the scar (Fig 10). Seven cavernous hemangiomas and one lymphangioma were present in the current series. The cavernous hemangioma was nodular, large, deep, bluish purple and easily compressible. The lesions were localized mostly in the cheek extending either to the upper lip or extending beyond the edge of the mandible to the upper neck. One patient with cavernous hemangioma had his lesion extending to the eyelids and subconjunctival space.

Excision of cavernous lesions was complete in 7 of 8 patients (Fig 11), however, residual lesion of the eyelids remained after complete excision of the buccal hemangioma and required subsequent ophthalmological treatment (Fig 12). Large cavernous hemangioma was found to extend beyond the zygomatic arch reaching the temporal region in one patient and underneath the body of the zygoma in another patient. These bony fences represented the endpoint for further dissection as the feeding veins can not be secured and clamped. Although cavernous hemangioma at the angle of the mandile was far beyond the limits of buccal mucosa, however, careful dissection with division of the buccinator muscle and retraction of the anterior border of masseter allowed its complete excision (Fig 13). Five patients had mixed hemangioma, three of the lesions were in the cheek, one involved the cheek and upper lip and one localized solely in the lower lip. Although excision of the cavernous lesion only was followed one year later by complete resolution of the capillary element. However, one patient of such lesions developed induration at the original site of distribution and persisted for 18 months (Fig 15). Cavernous hemangioma and lymphangioma embedded in a pad of fat were

detected in 6 patients. The lesions were remarkably large and caused noticeable disfigurement even occluding the mouth, the nostril and were associated with prominence of the maxilla. Excision of such lesions transorally was a demanding procedure (Fig 15) and resulted in reduction of 70% of the prominence but further stages for maxillary bone reconstruction and lip elevation were needed (Fig 16). None of the patients developed injury of the parotid duct because it was cannulated and secured through the operation. None of the patients developed injury of the facial nerve branches. Two patients were excluded from the study either because of former laser therapy (Fig 17) or because of hemorrhage of the hemangioma before the operation (Fig 18). Although laser therapy resulted in some sort of lightening of the capillary element of the hemangioma but it induced hypertrophic scar and adhesions between the skin and the underlying cavernous part. Active hemorrhage of hemangioma interfered with the transoral route because the skin that we aimed to preserve has already sloughed and the bleeding was so severe that required preliminary facial artery ligation.

Table (1). ***Clinical data and anatomic locations of the lesions.***

Site	*Hemangioma*	*Lymphangioma*
	n=21	n=4
Lower lip	7	1
Upper lip	2	-
Cheek	9	2
Cheek and upper lip	1	1
Angle of the mandible	2	-

Table (2): ***Clinicopathological types of the lesions.***

	Hemangioma	Lymphangioma
Capillary	5	1
Cavernous	7	1
Mixed	5	-
Cavernous embedded in fat	4	2

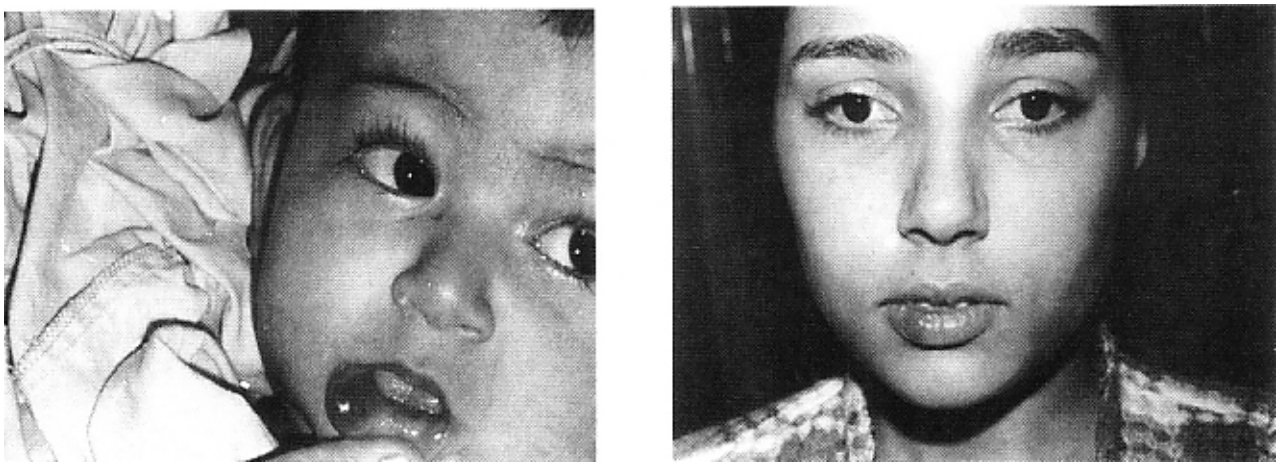

Fig (1 a,b). Small hemangiomas of the lower lip in different ages.

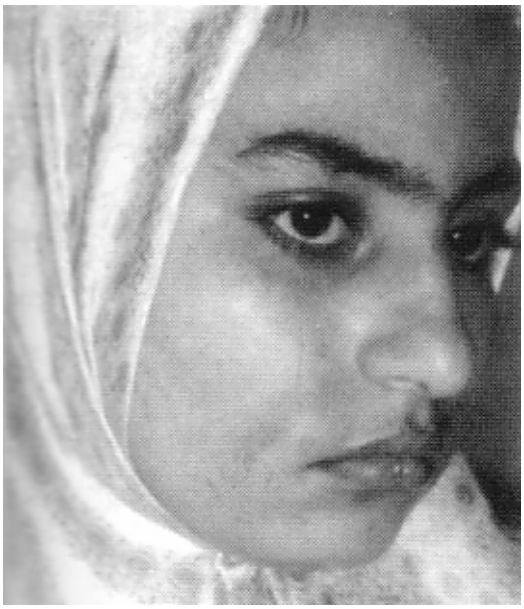

Fig (2a). Hemangioma of the upper lip presenting as elevation of the skin.

Fig (2b).Discoloration is more evident on the mucosal side.

Fig (3). Hemargioma of the cheek with areas of calcification.

Fig (4). Mixed hemangioma with the capillary element reaching the periorbital region.

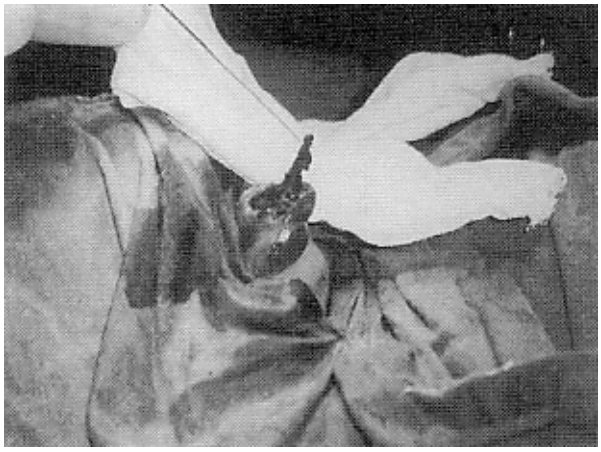

Fig (5). Shaving of hemangioma of the lip submucosally leaving a thin rim of the capillary element.

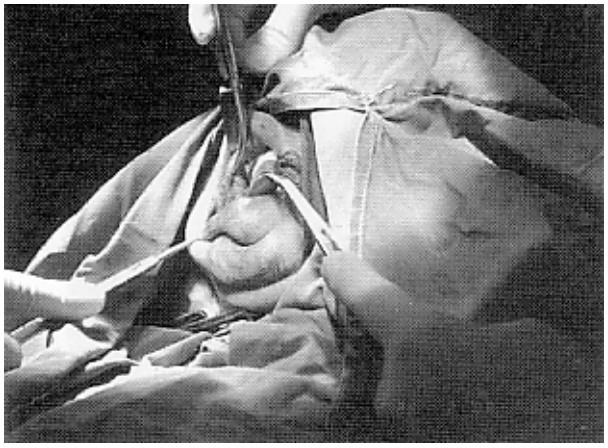

Fig (6). Exposure of the lesion through the oral cavity.

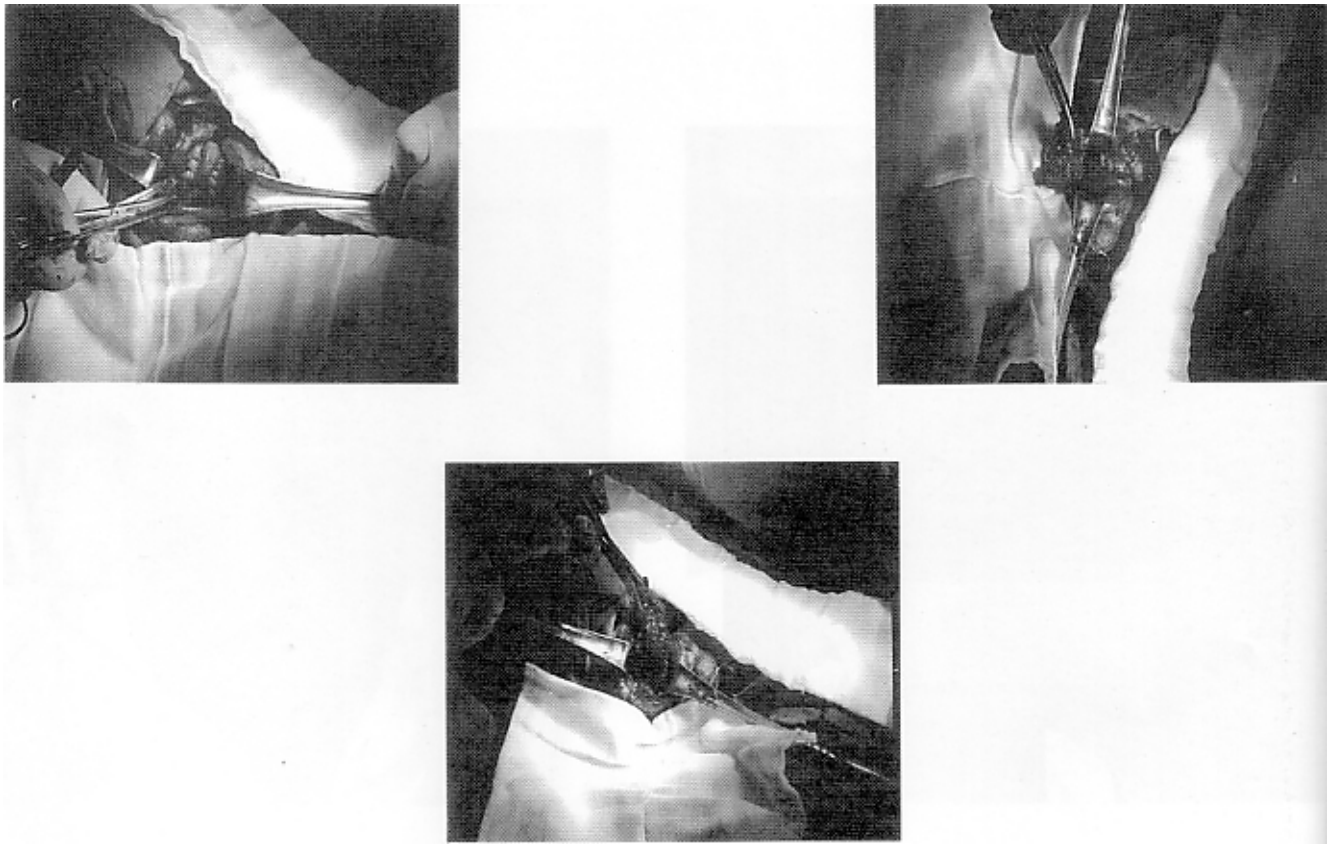

Fig (7a,b,c). Tracing the lesion in all directions after division of the buccinator muscle.

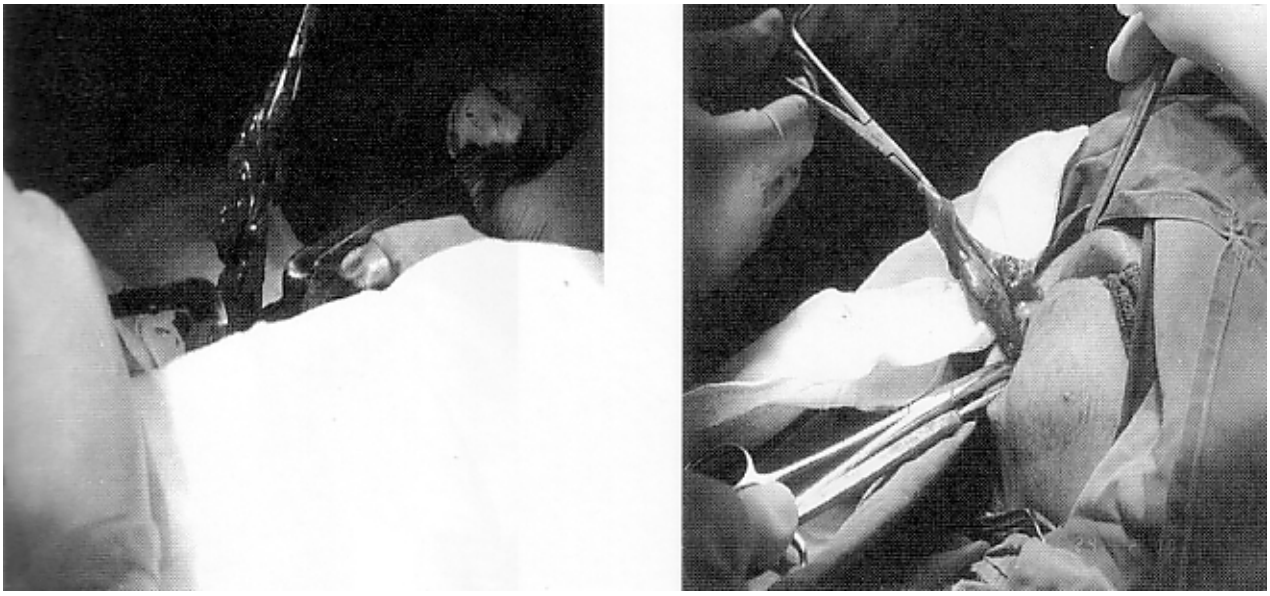

Fig (8a,b). Excision of the whole lesion transorally.

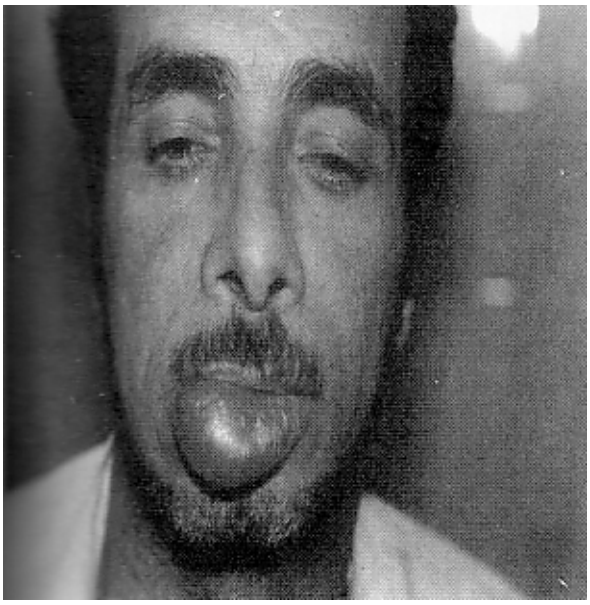

Fig (9a). Hemangioma involving almost the entire lower lip before surgery.

Fig (9b). Complete resolution of the swelling and discoloration after surgery.

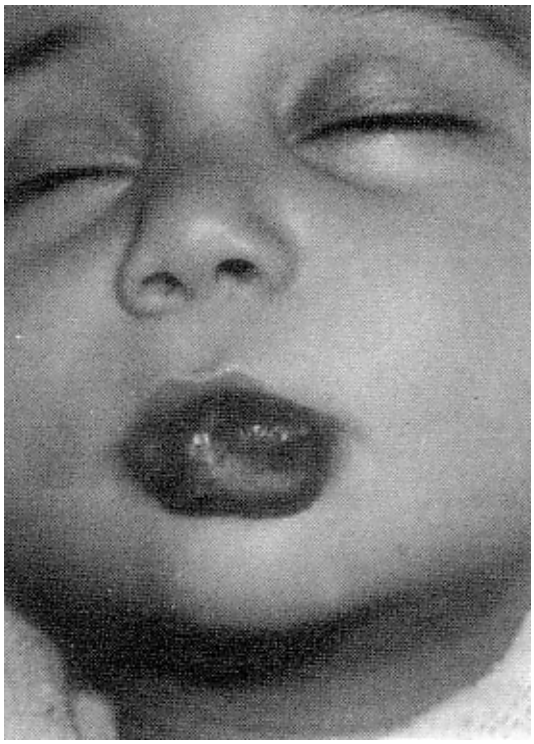

Fig (10a). Ulcerated hemangioma of the lower lip before surgery.

Fig (l0b). Fading of the swelling and color but persistence of the ulcer after excision.

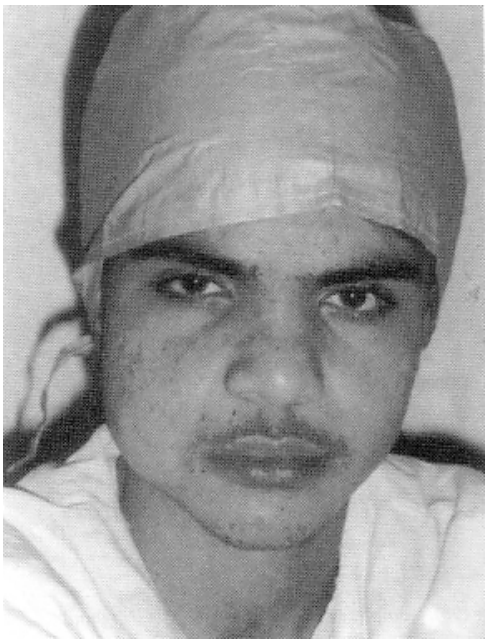

Fig (11a). Cavernous hemangioma of the cheek before surgery.

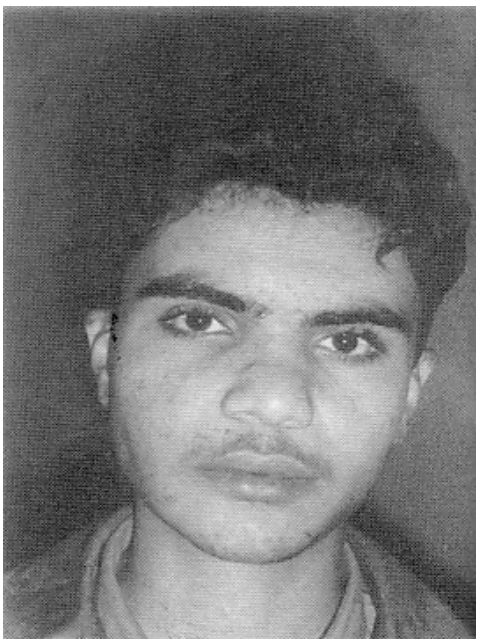

Fig (11b). Complete excision of the lesion without scar on the skin.

Fig (12a). Cavernous hemangioma of the cheek extending to the eyelids and subconjunctival space.

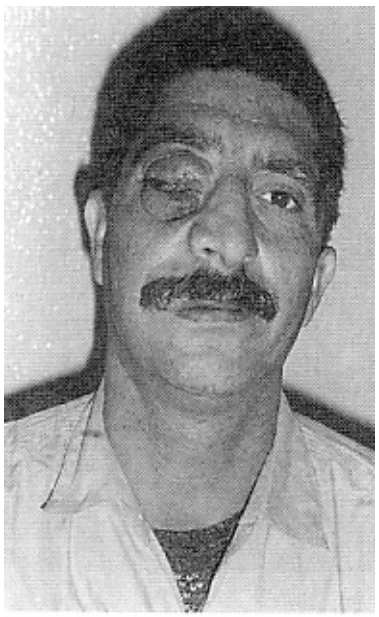

Fig (12b). Complete excision of the buccal part but persistence of the ophthalmic lesion.

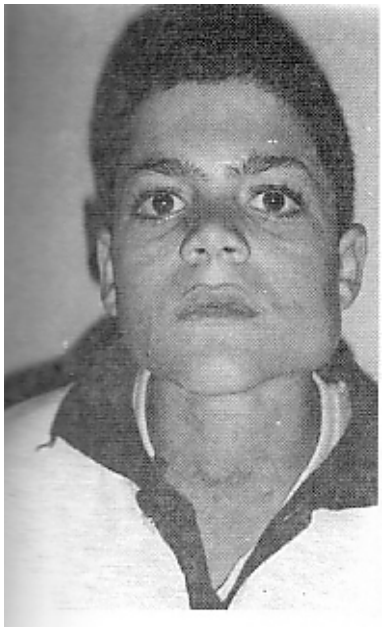

Fig (13a). Hemangioma at the angle of the mandible before surgery.

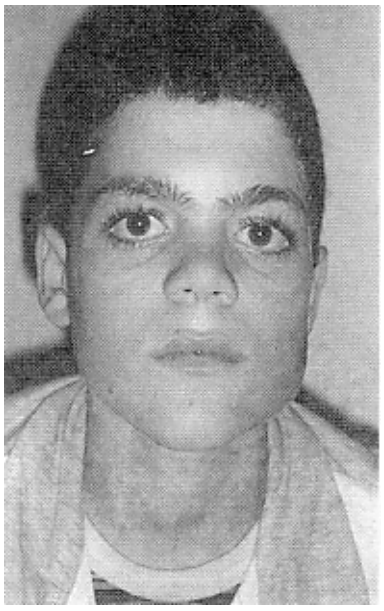

Fig (13b). After excision of the lesion transorally.

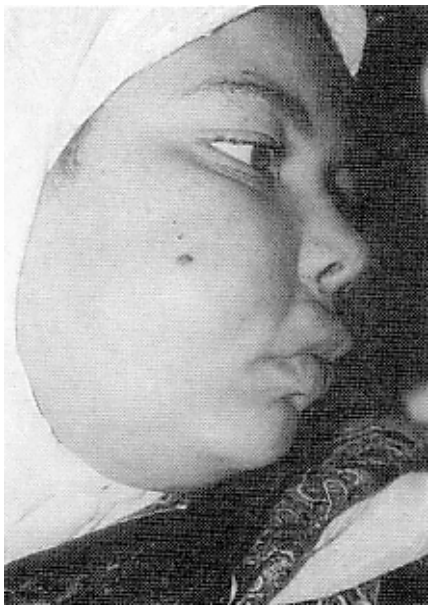

Fig (14a). Cavernous hemangioma of the cheek and upper lip.

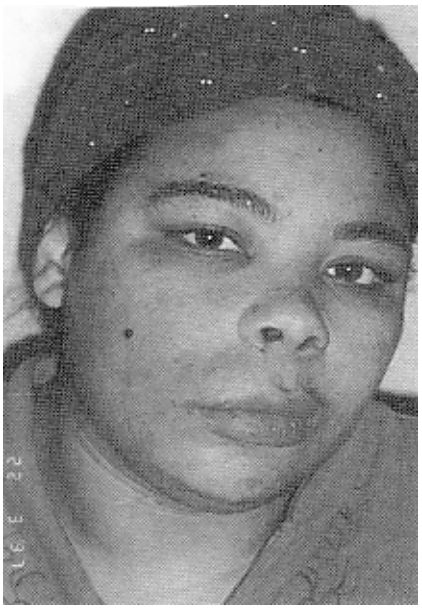

Fig (14b). Fading of the color and swelling but persistence of induration after surgery.

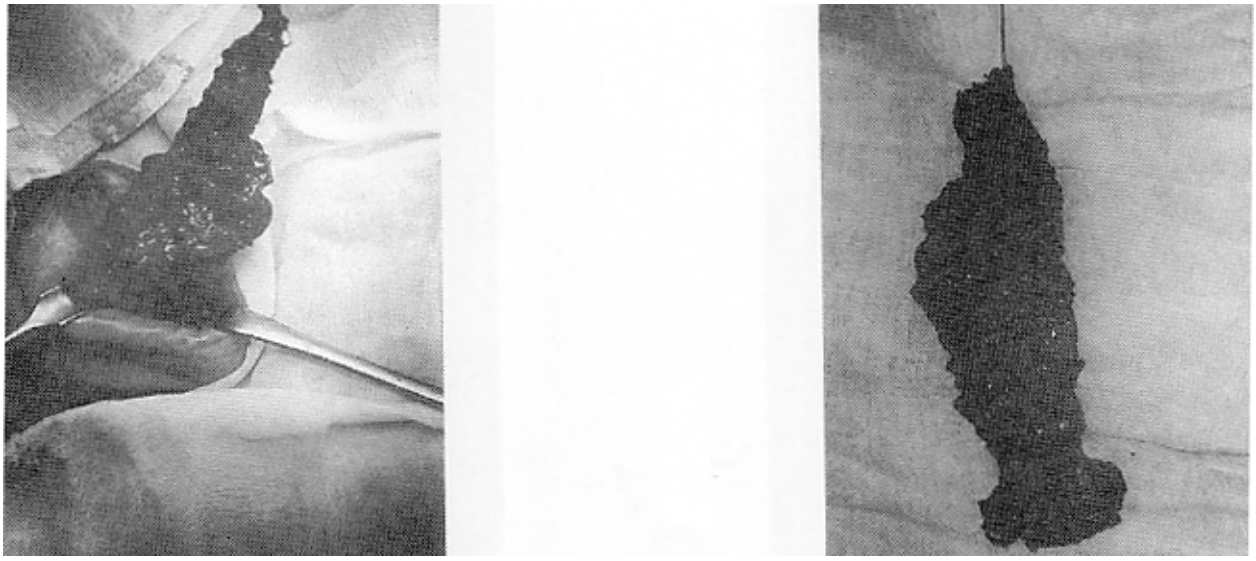

Fig (15a,b). Transoral excision of lymphangioma of the cheek.

Fig (16a). Lymphangioma of the cheek encroaching upon the upper lip, nostril and lower eyelid before surgery.

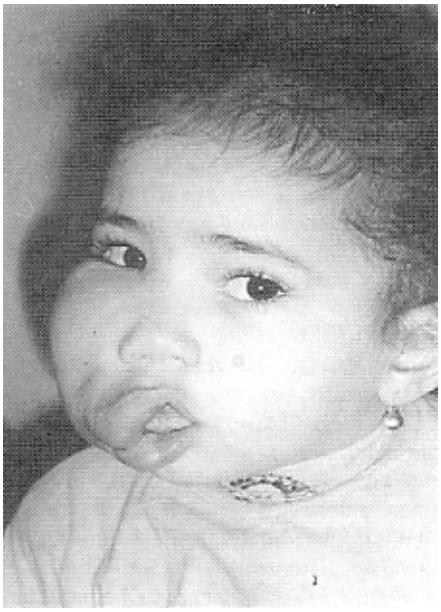

Fig (16b). Reduction of 70% of the swelling after resection of the lymphangioma.

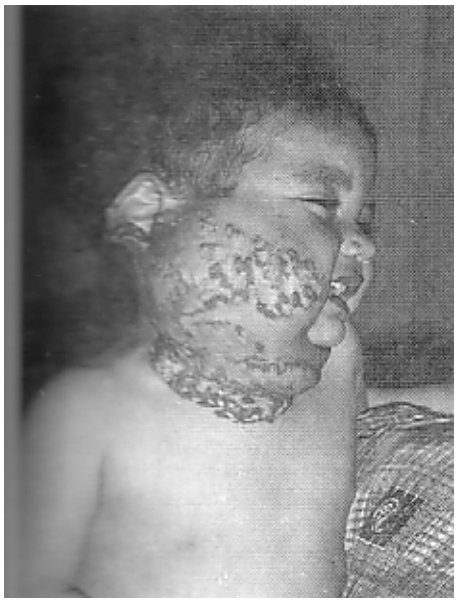

Fig (17). Hemangima of the cheek and neck after laser therapy.

Fig (18). Serious hemorrhage of ulcerated hemangioma of the cheek.

DISCUSSION

The proportion of females was lower in this group of patients with hemangioms of the lips and cheek than that reported in the literature: Five times versus two [6] or three times [7] more females than males. Since the majority of hemangiomas spontaneously involute, a major part of the clinical management involves reassurance and conservative observation. However, well- documented problems can occur, some with serious sequelae. The result after involution of a cutaneous hemangioma can be excellent or satisfactory with reparable skin changes (loose skin and telangiectasis). However, extensive hemangiomas often permanently alter the skin. The final result of involution is not influenced by the age of appearance of the vascular anomaly, the age of treatment onset, or the size and site of the cutaneous lesion [8]. This very high probability of uncomplicated regression can be potentially pernicious in that it might lure the clinician into rigidly noninterventionist postures. There are situations that mandate action. Even when the lesion is clearly a hemangioma with a future of regression, certain clinical situations have complications that require intervention. Ulceration and subsequent bleeding and infection are the most common local complications of hemangiomas. Ulceration occur frequently in mixed superficial and deep hemangiomas of the lip and neck. Ulcerated hemangiomas are quite painful and difficult to treat. The longer a hemangioma remains ulcerated, the higher the risk of infection [9]. We reported two patients with ulcerated hemangiomas, a strawberry hemangioma of the lip whose ulcer remained over 6 months then healed with scar. Although surgical excision succeeded to alleviate the bulk and color of the lesion, however, uncosmetic scar remained for several years afterwards. Another patient with mixed hemangioma of the cheek showed ulceration of the subdermal element with serious hemorrhage and was excluded from the study because of unfeasible transoral excision. The current observation, confirmed by others [10, 11], that ulceration often appears to initiate regression but it is in no way beneficial; as permanent scaring remains when the ulcer heals. These local complications in addition to the occasional incomplete involution leaving wrinkled skin and telangiectases and the potential enlargement of cavernous hemangioma after a long period of quiescence, raises the assumption that active surgical excision should be considered especially when it is harmless since it leaves no scar but otherwise a normal skin.

Buccal hemangiomas extending caudally toward the neck crossing the mandibular edge were easily excised. On the other hand, excision of lesions extending cranially toward the eyelids was hampered by the bones of midface. Intralesional corticosteroid injections are recommended for hemangiomas of the lower part of the face. Hemangiomas of the eyelids and orbits have feeding vessels arising from the ophthalmic artery. Retinal artery and choriocapillaris occlusion, causing loss of vision has occurred with the injection of a long acting corticosteroid suspension in an eyelid hemangioma [12]. For these function-threatening hemangiomas, systemic steroids are safer. Like systemic steroids, intralesional corticosteroids may elicit either a good or bad response. Therapeutic embolization has been described for deep hemangiomas. However, selective arteriography is difficult in infants whose vessels are short, thin, and prone to arterial spasm and thrombosis. Embolization needs to be done by a well - trained teams [13]. For the superficial hemangiomas, its real benefit remains to be proven. Radiotherapy should be dismissed unless other forms of treatment are ineffective or contraindicated. Irradiated lesions regress but the treatment is not free of long-term carcinogenic risk [14, 15]. Although laser therapy appears to be relatively safe, and may prevent enlargement, promote involution, or eliminate these vascular lesions, however, in view of the relatively high cost of laser therapy and its adverse effects [16, 17], it should be reserved for the use in carefully selected patients [18]. The concept that attempted surgical treatment of hemangiomas may cause adverse systemic or cutaneous side effects, particularly scarring, so intervention has to be reserved for patients with significant complications, is not applicable in the current study since this approach neither leaves a scar nor it causes any local injury to the ductular or neural structures in the assigned territory. In addition to the well-known fact that: although hemangiomas resolve eventually, the lesions persist in 35% to 50% of children who begin school [6]. Even after spontaneous involution of the lesions, 10% to 20% of children have residual skin changes, including telangiectasia, atrophy, wrinkling of the skin and cutaneous depression [19].

Lymphangiomas are congenital maldevelopment of the lymphatic system which may be seen initially in various forms and are usually present by the age of 2 years in 80% to 90% of patients. These lesions are anatomically complicated with multicompartmental distribution, making total excision virtually impossible without sacrifice of adjacent vital structures. Typically, the lymphangiomas cross fascial planes and extend along the neurovascular bundles. Although excision of lymphangiomas has been the primary method of treatment, partial debulking of the lesion is commonly the end result, often with recurrence of growth[20,21,22]. In many patients with congenital lymphangiomas, treatment is therefore palliative, aimed at preserving body function, sparing vital structures and maintaining an acceptable cosmetic status. In conclusion, we consider surgical resection as the mainstay of treatment for these lesions, in the light of overcoming the difficulties to clearly separate and spare normal structures from the abnormal tissue as well as the absence of scar on the skin which yields satisfactory cosmetic outcome. Putting into

consideration, the safety, feasibility and efficacy of this approach, most of these lesions should be actively surgically resected, in contrary to the former concept, before the development of complication that will otherwise preclude the surgical intervention.

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