

## Treatment of Buccal Lymph-hemangioma by Suction and Intralesional Steroid Injection

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**Background:** Lymphangioma is a tumor-like lesion of lymphatics that most often located in the head and neck area, usually causing marked cosmetic and functional problems. Challenge continues to find a safe and effective modality of treatment for anatomical and functional purposes. Until now, no satisfactory methods in lymphangioma elimination.

**Methods:** A 12 years old boy with lymphangioma of the left buccal was referred to the division of Plastic and Reconstructive Surgery in Cipto Mangunkusumo hospital. Suction and intralesional steroid injection was decided as a treatment in this patient. Pressure garment customized the size was made and applied to maintain and prevent healing in sagging position.

**Discussion:** Long term result should be evaluated, in this patient since the repair just only for 5 months. At follow up, 5 months post operatively after Suction and intralesional steroid injection, the buccal had improved significantly with normal anatomical and functional result.

**Conclusion:** In our experience, the correction of the buccal lymph-hemangioma with suction and intralesional steroid injection was achieved a satisfactory result.

**Keyword :** *Lymph-hemangioma, suction, intralesional steroid injection.*

**Latar Belakang:** *Lymphangioma* adalah lesi seperti tumor pada sistem limfatik yang paling sering terletak di daerah kepala dan leher, biasanya menyebabkan masalah nyata pada kosmetik dan secara fungsional. Tantangan untuk menemukan modalitas yang aman dan efektif dalam memperbaiki kelainan secara anatomi dan fungsional terus berlanjut. Sampai saat ini, belum ada metode yang memuaskan untuk mengeliminasi *lymphangioma*.

**Metodologi:** Seorang anak laki-laki usia 12 tahun dengan lymphangioma pada bucal kiri dirujuk ke divisi Bedah Plastik RSUPN Cipto Mangunkusumo. Dilakukan *suction* dan injeksi steroid intra lesi sebagai pengobatan pada pasien ini. *Pressure garment* yang disesuaikan dengan ukuran pasien dibuat dan dipakai untuk mempertahankan dan mencegah penyembuhan dalam posisi kendur.

**Diskusi:** Hasil dalam jangka waktu yang panjang harus dievaluasi pada pasien ini karena proses perbaikan hanya dipantau selama 5 bulan. Pada *follow up*, 5 bulan setelah dilakukan *suction* dan injeksi steroid intra lesi, buccal telah membaik secara signifikan baik pada segi anatomis dan fungsional.

**Kesimpulan:** Berdasarkan pengalaman kami, perbaikan pada *buccal lymph-hemangioma* dengan *suction* dan injeksi steroid intralesi telah mencapai hasil yang memuaskan.

**Kata Kunci :** *Lymph-hemangioma, suction, intralesional steroid injection.*

Lymphangioma is congenital malformation of lymphatic system. The common sites are cervico-facial region and axilla. According to Landing and Farber, those benign malformations are classified in four histological categories: capillary lymphangioma, cavernous lymphangioma, cystic lymphangioma (hygroma) and hemo-lymphangioma (combination of hemangioma and lymphangioma).<sup>1,2,3</sup>

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In most cases detected at birth or early in a child's life, usually before the age of two years. About 60% are seen at birth and 80- 90% manifest by the age of two years. The latter, congenital malformation, can remain asymptomatic for a long period of time. On the other hand, it may grow rapidly, surrounding or infiltrating the neighboring tissues or other major structures.<sup>4,5,6</sup>

Lymphangiomas are usually causing marked cosmetic and functional problems, re-

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currences are common. There are several treatment options, although some cases of successful surgical excision have been reported, complete resection of lymphangiomas is difficult due to their diffuse infiltration and intertwining with adjacent tissues, as well as their tendency to bleed. The risk of damage to surrounding structures (nerve and muscle) or poor cosmetic results (long scar) is high. Sclerotherapy agents have been shown to have less satisfactory re-sult, their use has been associated with severe systemic and cosmetic side effects. Systemic corticosteroid treatment has been reported to be an effective adjunct to surgery, but it may be in-effective as a sole agent. Challenge continues to find a safe and effective modality of treatment for anatomical and functional purposes.<sup>7,8,9</sup> Very few cases of lymph-hemangiomas have been reported in the literature. We refer to case report about the satisfactory result in treatment of lymphangioma circumscriptum by suction,<sup>10</sup> but there are no publications regarding suction therapy on lymph hemangioma in the face.

### CASE 1

A 12 years old boy was presented to the division of Plastic and Reconstructive surgery in Cipto Mangunkusumo hospital with complaints of increase in left buccal size since 3 days ago, palpable and painless, no teeth ache and no history of tooth ex-traction . He had a left buccal swelling since birth.

On clinical examination the left buccal was found oval in shape, 5x4x3 cm sized, no erythema, soft, compressible, no warmth and painless. The neurological examination was normal. The definitive histological diagnosis was lymph-hemangioma.

A decision to manage the buccal lymph-angioma with *suction* and intralesional steroid injection was made after informing the parents. benign tumor involving the infe-rior region of his left midfacial region (cheek). Intraoral view (right), the tumour does not protude and damage into the oral cavity (Figure 1)

### Operating Procedures

On the general anesthesia, an oval shape of lymph-hemangioma approximately 5x4x3 cm,



**Figure 1.** Preoperative appearance. Lateral view (left), tumour hangs below the inferior margin of the left mandible. anterior view, the benign tumor involving the inferior region of his left midfacial region (cheek). (1, right) Intraoral view, the tumour does not protude and damage into the oral cavity.

including the sternocleidomastoid muscle, the hyoid bone, the angle of the mandible and the incision sites are marked with gentian-violet.

We performed two incisions, in the submental crease and in the posterior lobular crease of the left ear. The incision is created and the imme-diate surrounding skin is sharply undermined with a small tenotomy scissors to allow appro-priate placement of the cannula in the proper plane and to prevent postoperative irregulari-ties at the incision site. The appropriate plane is just deep to the dermal-subcutaneous interface. Then without wetting, insert the suction cannu-la approximately 2mm in diameter from the in-cision, cannula was used to aspirate the serous fluid with fibrous tissue, avoid the branch of facial nerve.

Remaining lymph-hemangioma was injected with 1 cc triamcinolone acetone. The application of a under pressure bandage was performed postoperative then change with under pressure garment as soon as possible.

### RESULT

At follow up, 5 months postoperatively, the buccal had improved significantly with respect to size, color, and no pain was observed. Invisible scar, without sagging position and normal function of nervus facialis. (Figure 2)



**Figure 2.** (A, Left) before treatment. (B, Right) 5 months postoperatively, after suction of the left buccal and intralesional steroid injection.

### DISCUSSION

Lymph-hemangiomas are rare benign tumors that appear to arise from congenital malformation of the vascular system. The formation of that tumor may be explained by obstruction of the venolymphatic communication, between dysembrioplastic vascular tissue and the systemic circulation.<sup>3, 11</sup> The incidence of lymph-hemangiomas varies from 1,2 to 2,8 per 1000 newborns, both sexes get equally affected.<sup>12, 13</sup>

The clinical onset of lymph-hemangiomas can vary from a slowly growing cyst over a period of years to an aggressive enlarging tumor, without invasive ability. In untreated cases, complications such as haemorrhage inside cysts, spontaneous or traumatic rupture, infection nerve compression causing pain and paraesthesias, respiratory difficulty and disfigurement may occur. In our case, sudden increase in the left buccal size was not have any complication.<sup>3, 13, 15</sup>

The technique of suction presents a means of partially removing the deep cisterns while destroying their integrity. Suction has proven save in large number of patients. As in our patient, the small incision in the submental crease and in the posterior lobular crease required for insertion of the suction cannula presents an additional advantages over a surgical approach requiring a long incision. Permanent injury to the marginal mandibular branch of the facial nerve is rare as in

hypesthesia secondary to injury to the great auricular nerve. When paresis, paresthesia, or paralysis occurs, it is almost always short-lived and resolves with the tincture of time.<sup>16</sup> Furthermore, the technique offer a change to sculpt and taper the subcutaneous tissue so that any major contour defect is minimized. Post operative, the scar will be small and disguise, that very important in location that not be closed with clothes.<sup>10</sup>

Edgerton has shown that steroids tend to sensitize the vascular bed to vasoconstricting agents. Intralesional administration of triamcinolone devoid of systemic side effects. Corticosteroid treatment has been reported to be an effective adjunct to surgery, but it may be ineffective as a sole agent.<sup>17,18</sup> Furthermore, irregularities that become evident after suction procedure are almost always transient and treated with reassurance, gentle massage, and occasionally, dilute steroid injection.<sup>16</sup>

Other techniques available are surgical excision, injection of sclerotic agents, cryotherapy, Laser and radiotherapy (by radium, roentgen ray or radon seed), but none of them produced acceptable results. Recently, two sclerosing agents, bleomycin and OK-432, have been favored by some surgeons in the treatment of lymphangioma. However, the surgeon should be alert for the possible serious complications. The most serious adverse effect of bleomycin is interstitial pneumonia and pulmonary fibrosis. Mortality due to bleomycin sclerotherapy has also been reported. Complete surgical excision also presents the lowest recurrence rate, while complete surgical eradication may become more difficult and this procedure will remove a lot of tissue that is not a malignant tissue. The recurrence rates vary depending on the complexity of the mass, the anatomical location and the adequacy of the excision. However, lesions that have been completely excised, present 10-27% recurrence, while those being partially resected may recur in 50-100%. Although surgical excision was more effective than sclerotherapy, there was a high complication rate of 12% to 33% associated with surgical excision. Radiotherapy is used when surgical excision is not feasible; the radio-

sensitivity of hemo-lymphangiomas is not well understood.<sup>3, 19, 20, 21, 22</sup>

An important issue remains the continuous lymphorrhya during the instant post-operative period are the application of an under pressure bandage or the prolonged drainage.<sup>3</sup> The application of under pressure garment also maintain and prevent healing in sagging position.<sup>16</sup>

### SUMMARY

The challenges to find new ways to treat lymphangioma is needed because there is no satisfactory methods in lymphangioma elimination to date. Based on our experience the correction of the buccal lymph-hemangioma with suction and intralesional steroid injection will give satisfactory result.

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### REFERENCES

1. Deodhare SG, Sidiqi HM. Aggressive lymphangioma (report of an unusual case). *J Postgrad Med* 1983;29:255
2. Rattan KN, Kajal P, Kadian YS, Gupta R. Haemorrhage in a scrotal lymphangioma in a child: A rarity. *Afr J Paediatr Surg* 2009;6:110-1
3. Kosmidis I, Vlachou M, Koutroufinis A, Filiopoulos K. Hemolymphangioma of the lower extremities in children: two case reports. *Journal of Orthopaedic Surgery and Research* 2010, 5:56
4. Suzuki K, Tsuchida Y, Takahashi A, Kuroiwa M, Ikeda H, Mohada J, et al. Prenatally diagnosed cystic lymphangioma in infants. *J Pediatr Surg* 1998, 33:1599-604.
5. Issacs H Jr. Tumors of the fetus and newborn. Major problems in pathology Saunders Co. Philadelphia 1997, 35:69-72.
6. Cotran RS, Kumar V, Robbins SL. Tumors of lymphatics. Robbins pathologic basis of disease Saunders Co. Philadelphia, 5 1994, 512.
7. Harris GJ, Sakol PJ, Bonavolonta G, De Conciliis C. An analysis of thirty cases of orbital lymphangioma. Pathophysiologic considerations and management recommendations. *Ophthalmology* 1990;97 (12):1583-1592.
8. Sires BS, Goins CR, Anderson RL, Holds JB. Systemic corticosteroid use in orbital lymphangioma. *Ophthal Plast Reconstr Surg* 2001;17(2):85-90.
9. Tawfik HA, Budin H, Dutton JJ. Lack of response to systemic corticosteroids in patients with lymphangioma. *Ophthal Plast Reconstr Surg* 2005;21(4):302-305.
10. Manders EK, Egan N, Davis TS. Elimination of lymphangioma circumscriptum by suction-assisted lipectomy. *Annals of plastic surgery* 1986; 16:532-34
11. Balderramo DC, Di Tada C, de Ditter AB, Mondino JC. Hemolymphangioma of the pancreas: case report and review of the literature. *Pancreas* 2003;27:197-199.
12. Filston HC. Hemangiomas, cystic hygromas and teratomas of the head and neck. *Semin Pediatr Surg* 1994; 3:147-59.
13. Okazaki T, et al. Treatment of lymphangioma in children:our experience of 128 cases. *Journal of Pediatric Surgery* 2007; 42: 386- 389
14. Mansingani S, Desai N, Pancholi A, Prajapati A, Vohra PA, Raniga S. A case of axillary cystic hygroma. *Indian J Radiol Imag* 2005;15:517-9
15. Arola A, Narula MK, Sonkar P, Chada R. Cystic hygroma of chest wall. *Indian J Radiol Imag* 2003;13:120-1