Long Term Follow Up of Hemangioma on Bilateral Cleft Lip

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Background: Hemangioma is a common tumor of infancy that exhibits rapid postnatal growth and slow regression during childhood. Cleft lip is a common anomaly found in Indonesia. These entities are twice in common in the Asian population. However, simultaneous occurance of these two abnormalities is rare to be found.
Patient and Method: A case of bilateral cleft lip complicated with hemangioma on the left side of lip and the surrounding skin through the mucosa, and the prolabium, which had been performed cheiloplasty procedure at age 9 months old. Cheiloplasty design was made through the hemangioma area with a careful hemostasis to avoid massive bleeding.
Result: Hemangioma was partially left behind at the cheiloplasty procedure. There was no bleeding during and after the surgery. Two years following the surgery, there was a fine scar with good shape of the lip and no further growth of the hemangioma.
Summary: Hemangioma and cleft lip are very rare to be found in the same location. Because of the involution potential of hemangioma, timing of surgery is very important. Considering the psychosocial aspect, the surgery was performed at the age of 9 months. Two years after surgery, we didn't find any growth of hemangioma or deformity of the bone and lip. The scar growth is in good esthetic result, no difference with left lip child without hemangioma.

Keywords: *Hemangioma, cleft lip, scar, deformity*

Latar Belakang: Hemangioma merupakan tumor yang sering ditemukan pada bayi, bersifat tumbuh cepat setelah kelahiran dan regresi perlahan di masa anak-anak. Sumbing bibir merupakan kelainan yang umum ditemukan di Indonesia. Kedua kelainan ini banyak didapatkan di populasi Asia. akan tetapi, kejadian simultan kedua anomali ini jarang ditemukan.
Pasien dan Metode: Sebuah kasus sumbing bibir bilateral dgn hemangioma pada sisi kiri bibir dan kulit sekitarnya sampai ke mukosa dan prolabium yang telah menjalani operasi cheiloplasty di usia 9 bulan. Desain cheiloplasty dibuat melewati area hemangioma dan dengan hemostasis yang baik untuk mencegah pendarahan masif.
Hasil: Hemangioma ditinggal sebagian saat operasi cheiloplasty. Tidak ada pendarahan saat dan setelah operasi. Dua tahun pasca operasi, ditemukan garis parut yang halus dengan bentuk bibir yang baik dan tidak ada pertumbuhan hemangioma.
Ringkasan: Hemangioma dan sumbing bibir jarang ditemukan pada lokasi yang sama. Waktu operasi yang tepat sangat penting karena adanya potensi involusi hemangioma. Dengan mempertimbangkan aspek psikososial, operasi dilakukan di usia pasien 9 bulan. Dua tahun pasca operasi, tidak ditemukan pertumbuhan hemangioma atau deformitas tulang dan bibir. Jaringan parut tumbuh dengan estetik yang baik, tidak berbeda dengan pasien sumbing tanpa hemangioma..

Kata Kunci: *hemangioma, cleft lip, scar, deformity*

Received: 13 September 2013, Revised: 10 November 2013, Accepted: 20 November 2013.
(Jur.Plast.Rekons. 2013;3:120-124)

Hemangioma is a common tumor of infancy that exhibits rapid postnatal growth and slow regression during childhood. Approximately 30-40% of hemangiomas are nascent at birth, presenting as a premonitory cutaneous mark. Hemangioma grows rapidly during the first 6 to 8 months of infancy. It reaches its peak before the first year, and for a time thereafter, growth is proportionate to that of the child.

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Presented in the 17th IAPS Scientific Meeting, Bandung, West Java, Indonesia.

Disclosure: *The authors have no financial interest to declare in relation to the content of this article.*

The involuting phase continues until the child is 5 to 10 years of age. Regression is complete in 50% of children by age 5 years and in 70% by age 7 years, with continued improvement until age 10 to 12 years.

In the white population, cleft lip with or without cleft palate occurs in approximately 1 in 1,000 live births. These entities are twice as common in the Asian population.[2] However the concomitant occurrence of these relatively two common anomalies is extremely rare.[3]

PATIENT AND METHOD

A 9-month-old child with hemangioma on bilateral cleft lip was referred from another hospital in Jakarta because they were afraid that there would be complications during and post-operative. There was no prior therapy ever given to the patient. Hemangioma was on left side of lip and the surrounding skin through the mucosa, and the prolabium. There was no invation of hemangioma to the intraoral. The patient was otherwise healthy, no other congenital anomaly was found.

Cheiloplasty design was made through the hemangioma area. After sterile preparation and draping, the lip was infiltrated with 2% lidocaine containing 1:100.000 fresh epinephrine. The needle for injection was inserted from the healthy skin. After waited for the vasoconstrictor effect, about 5 minutes, incision was made. With a careful hemostasis, incision on the hemangioma did not cause massive bleeding. The wound was closed with interrupted 5-0 Ethilon in the mucosal and muscle, and 6-0 Vicryl to the skin.

Hemangioma on the lip was partially left behind while cleft lip was repaired. There was no complication occurred, such as bleeding during and after the surgery. Within one week follow up, there was no dehiscence and the wound was dry. Superficial sutures was removed. There was still hemangioma on the lip, prolabium and the surrounding skin. After two weeks, the appearance of the scar was quite similar with cleft patient without hemangioma. The pathology report confirmed a capillary and cavernose lymph-hemangioma with immature cells. After three months, the scar was not hiperemic, and there was still part of hemangioma on skin. Two years following the surgery, we found the fine scar with good shape of the lip and there was regression of the hemangioma.

RESULT

Hemangioma was partially left behind at the cheiloplasty procedure. There was no bleeding during and after the surgery. Two years following the surgery, there was a fine scar with good shape of the lip and no further growth of the hemangioma.

DISCUSSION

Hemangioma, often called infantile or juvenile hemangioma for clarity, is a benign tumor that exhibits early and rapid proliferation phase during the first year of life characterized by endothelial and pericytic hyperplasia, followed by a slower but steady involution phase that may last for years.[4] Hemangioma usually occurs a solitary, superficial lesion in the head and neck area, but it can be present anywhere in or on the body. Size can vary greatly, from nodular lesions several millimeters in diameter to plaque-like tumors covering the entire face or a quadrant of the body.[5]

Uncomplicated hemangioma can resolve spontaneously. By the age of 5 years, nearly 50 percent (and by age 7, 70 percent) of simple hemangiomas have involuted. When treatment is decided on, various methods can be use such as surgical excision, laser, corticosteroid injections, embolization, sclerothraphy, cryosurgery, interferon, radiotherapy, tattooing, cyclophosphamide, and aminocaproic acid. A treatment method that seems satisfactory for one hemangioma may not be appropriate for another. The age and sex of the patient, localization of the tumor, flow rate, and phase of the lesion are determinant factors for choosing the best method.[3]

Cleft lip is also a common condition.[3] The ratio of incidence of cleft lip varies with ethnicity, 0.41:1000 in African Americans, 1:1000 in whites, and 2.1:1000 in Asians.

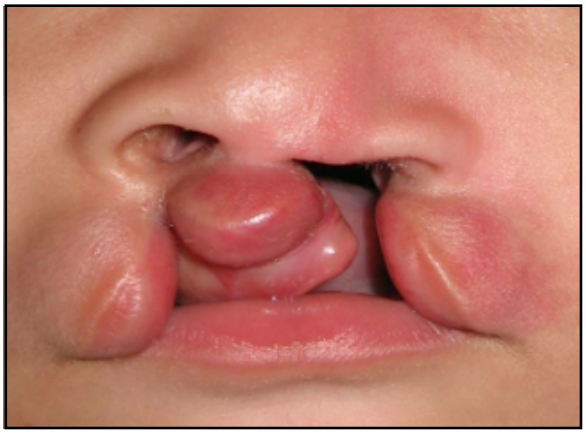

Figure 1. A 9-month-old baby with hemangioma on bilateral cleft lip.

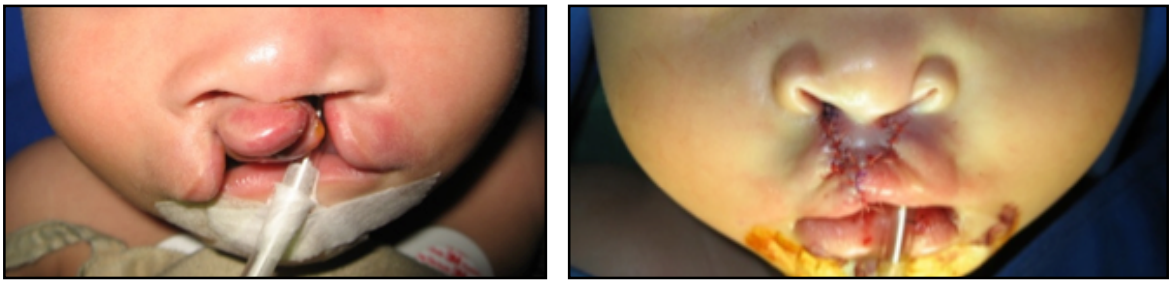

Figure 2. **Left :** Before surgery. **Right :** Immediately after surgery.

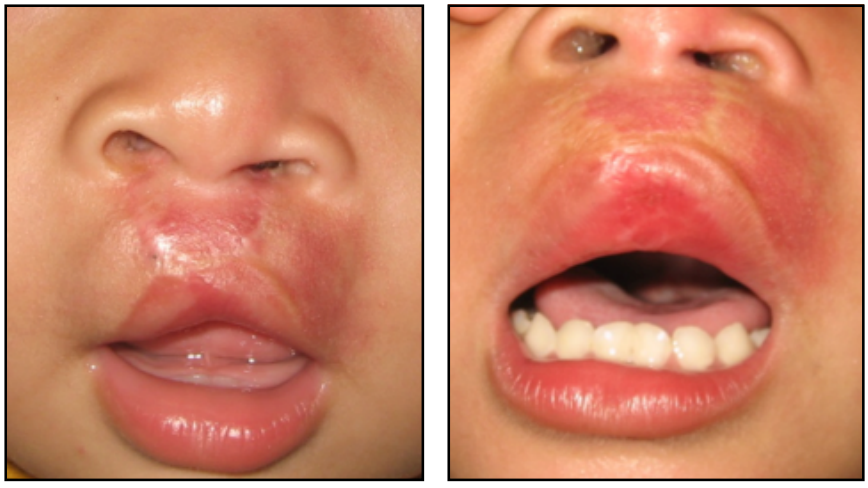

Figure 3. Patient after the surgery. **Left :** Three months after surgery. **Right :** Two years after surgery.

The etiology of cleft lip is regarded as multifactorial. Important potential risk factors include anticonvulsants, parental age, lower socioeconomic class, smoking, alcohol intake, and prenatal nutrition. Concomitantly, parents or siblings with cleft lip and palate predispose future children to an increased risk. In families of a one-cleft parent or sibling, there is a 4% risk of a subsequent child being born with cleft lip/palate.[6]

Hemangioma on cleft lip has been reported in several journals. The cases reported was unilateral cleft lip. The first case was reported in Atlanta, a 4-year-old girl with cleft lip on right side with hemangioma on almost the whole upper lip. Excision of hemangioma and cleft lip repair was performed in one stage. Williams suggest the cleft lip repair should be performed at age above 6 months old due to the potential to bleed from the hemangioma and the difficulty in obtaining reasonable aesthetic results. Moreover, endothelial proliferation seen in early hemangiomas would increase the chance of residual tumor growth after repair. In their case, the cleft lip repair was delayed until significant involution phase of the hemangioma. After the involution has occurred, a single-stage excision and lip repair can be performed. Nine months following the surgery, some hypertrophic scars exist along the philtrum.[7]

A case reported in Taiwan was a patient with Wolf-Hirschhorn syndrome. A 2-month-old boy diagnosed with cleft lip on right side and hemangioma on the medial side, columella and tip of the nose. Adhesion cheiloplaty was performed and there was complete separation one week after surgery. Later, the patient was given oral prednisolon and intralesion steroid injection. However, no response was observed. At the age of 1 year, the patient had another surgery on lip repair. Hemangioma was partly left behind. The result was good.[8]

A case of 3-month-old boy with left side cleft lip and hemangioma on vermillion was reported in Korea. Cleft lip repair was performed, leaving some part of the hemangioma on the lip. There were no complications such as bleeding and ulceration after the surgery. In their observation, when the patient was 5-year-old, the hemangioma had involuted. The lip has a good shape and color.[9]

Another case was reported in Turkey. A 3-month-old boy with left side cleft lip with hemangioma on columella, nasal sill, and most of the upper lip on the non-cleft side.[10] There was no report about the result after surgery.

Our patient had bilateral cleft lip with hemangioma on the prolabium, left side of the lip and the surrounding skin. We performed cleft lip repair using a labioplasty design through the hemangioma. We did not wait until the natural involution of hemangioma to occur considering further psychosocial problem if the lip repair was delayed. Vasoconstrictor was injected in similar dose and technique with other labioplasty. No massive bleeding occurred during the surgery. The skin was sutured using absorbable material with consideration of minimizing the risk of bleeding from suture removal.

Two weeks after the operation, no bleeding, dehiscence and ulceration was noticed. Three months after operation, the scar was not hyperemic and the shape of the lip was good. In our observation, two years following the surgery, there was a fine scar with good shape of the lip and the remaining hemangioma was partly regressed.

SUMMARY

Hemangioma and cleft lip are both very common separately, but very rare together, especially in the same location. Because of the involution potential of hemangioma, timing of surgery is very important. Definitive cleft lip repair can be performed without the risk of massive blood loss during the surgery. In our observation, within two years after the surgery, we didn't find any growth of the remaining hemangioma or deformity of the bone and lip. The scar growth was in good esthetic result, no difference with cleft lip patient without hemangioma.

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