

Case Report

Resection of localized cheek infantile hemangioma in the proliferative phase: a case report

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ABSTRACT

Infantile hemangioma (IH) is the most common vascular tumor which is characterized by proliferative phase, involuting phase, and involuted phase. As the regression will ultimately occur at the end of the phase, the choice of treatment become controversial. Although surgical management often advocated for complicated and conservative therapy failure cases, early resection may give some benefits both to the patients and parents. This case report described a 9-month-old female with cheek IH who underwent tumor resection followed by cheek flap as the procedure to close the defect. After two days post-operation, the patient was discharged from hospital without any complication observed. The surgical scar was favorable in the first month after surgery. Early resection can be established as one of the treatment choices for fast-growing hemangioma in the proliferative phase to avoid undesirable aesthetic sequelae in the future.

Keywords: Cheek flap, Hemangioma, Resection

INTRODUCTION

Infantile hemangioma (IH) is classified among the vascular tumors group based on the most widely accepted vascular anomalies classification by The International Society for the Study of Vascular Anomalies (ISSVA) 2014.^{1,2} It is characterized by 3 phases of life cycle comprise of proliferative phase, involuting phase and involuted phase that distinguish it from another form of vascular anomalies. However, there are commonly no clear boundaries between proliferative phase and involuting phase in clinical setting; involuting features typically coexist with proliferative features during the process.^{1,3}

Histopathology examination has been an immense help to diagnose IHs in daily practice. It reveals early proliferative phase IHs are usually consists of well-circumscribed, un-encapsulated masses of capillaries

composed of plump endothelial cells and pericytes that are arranged in lobules and separated by delicate fibrous septae. Later in the involuting phase, the lesions become less well-defined as lesional capillaries start to disappear and apoptotic bodies, as well as mast cells, increase in number. As the lesion can regress spontaneously after a complete involution phase, management of IH become controversial. Watchful-waiting used to be prevailed in many clinical settings unless proliferation could arise some complications that unlikely to resolve without treatment.^{1,3}

Life-threatening or function-threatening complication of IH is an uncommon entity. Although many children have a complete regression into a normal skin, 40-50% IH cases leave permanent changes in the skin such as hypopigmentation, excessive fibrofatty tissue, telangiectases, skin alteration, and atrophy. For these reasons, early resection in the proliferative phase has

been proposed in some complicated cases to improve residual scarring.^{2,4} Surgical management of IHs can be challenging depends on the tumor size and extent as well as the anatomical site. Therefore, this case report is aimed to describe the resection of localized IH in the proliferative phase as well as the reconstruction of the defect.

CASE REPORT

A 9-month-old female patient was brought to the plastic surgery department with a chief complaint of a lump on the right cheek since two weeks after birth. The lump had developed from a red spot to an elevated red lump that progressively increased in size. The parents said there were no breathing difficulty, tendency of bleeding and lump on the other part of the body.

The patient was a full term (37 weeks) normal weight (2500 gram) infant. The maternal age was 37 years old without any history of pregnancy complications. There was no family history of the similar symptom.

On physical examination, a well-demarcated concave red tumor with grayish discoloration at the center of the lesion was detected with a diameter of 3 cm (Figure 1). Routine laboratory investigations such as complete blood count, biochemical parameters, and hemostasis test were within normal limits. The treatment plan was tumor resection followed by local cheek flap to close the defect.

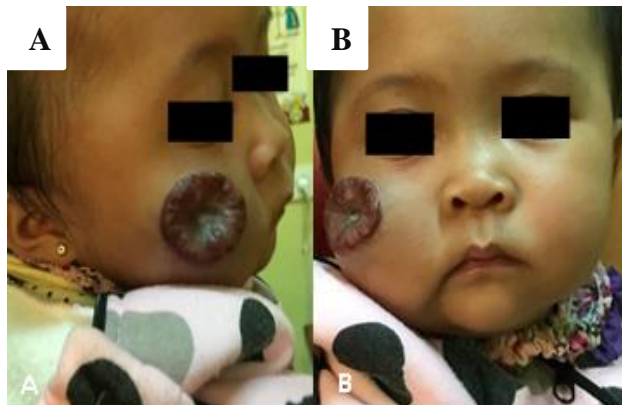


Figure 1: Preoperative photograph of the cheek hemangioma; A) Lateral view, B) Anterior view.

Surgical procedure

The procedure was performed under general anesthesia and was started by drawing a demarcation line around the tumor. Vasoconstrictor was injected subcutaneously around the tumor to minimize bleeding. Several sutures were placed along the demarcation line to prevent bleeding as the vessels surrounding the tumor were ligated. In addition, the sutures helped to define the tumor border allowing more accurate excision. The tumor then excised by making a circular incision along the

demarcation line while performing hemostasis carefully. After excision, a 3 x 2.7 x 1.5 cm specimen was sent to the histopathology department (Figure 2). Histologic examination demonstrated features of highly proliferating hemangioma (Figure 3).

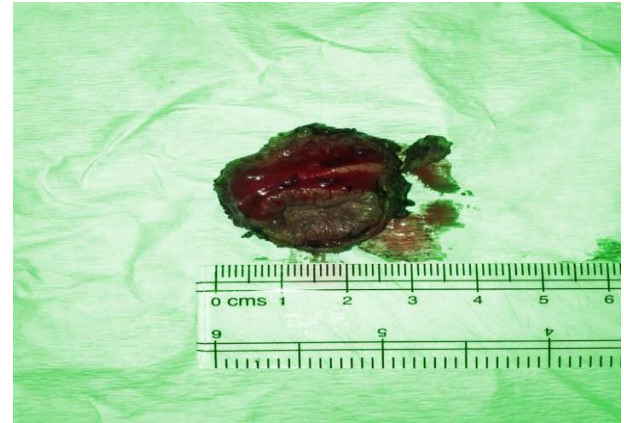


Figure 2: Specimen of hemangioma mass.

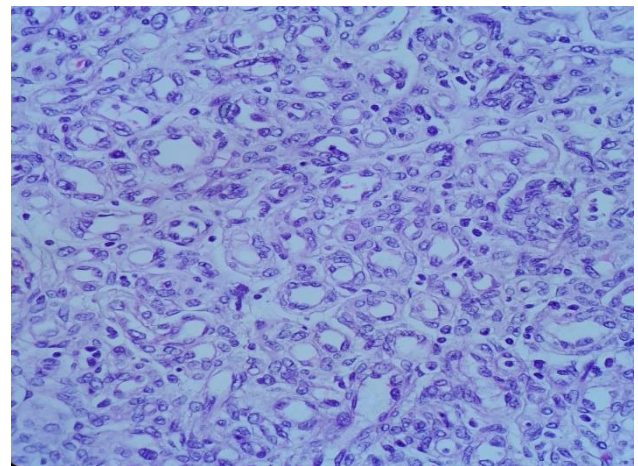


Figure 3: Histopathology of proliferative IH, endothelial cells proliferation arranged in lobules.



Figure 4: A) Raw surface post hemangioma resection; B) The defect was closed with local cheek flap.

Reconstruction with local cheek flap was performed after the raw surface was measured (Figure 4). It was started by making cheek flap design over the skin. The incision was made along the designated line, ran transversely from the defect to the anterior temporal hairline. The cheek flap then incised and elevated above the Superficial Muscular Aponeurotic System (SMAS). It was advanced to the defect area while preserving the subdermal plexus. Any formation of dog ear was removed. The wound then closed by layers. The overall intraoperative bleeding was minimum.

Postoperative and follow up

On the first day after surgery, the flap site was slightly bluish, edema and warm by palpation. The patient was discharged on the second day and followed up on the fifth day, second week and the first month at the outpatient department. On the fifth day after surgery, the edema was reduced and there was slight wound dehiscence occurred in the former tumor location, re-suturing and debridement were immediately done in the operating room. The surgical scar was favorable in the first month after surgery (Figure 5).



Figure 5: One month after surgery photograph.

DISCUSSION

Infantile hemangioma (IH) has various morphology, growth rate, size and involuted tumor appearance. The patient was a 9-month-old female which is an expected age for a proliferative phase tumor that typically occurs in the first 6 to 8 months until one year of age. A red spot mark presence on the second week after birth resembles an early form of IH that commonly appears on the first few weeks after birth.^{1,3,5} Although the exact cause and pathogenesis of IH remain unclear, there are some associated risk factors mentioned. Two related risk factors were present in this patient such as gender (female) and older maternal age (37 years old).^{6,7}

About 24% IHs end up with some complications such as ulceration and scarring.^{5,8} Facial IHs are 1.7 times more

likely to develop complications as well as larger IHs.⁶ The patient in this case report demonstrated a relatively large proliferating tumor on her midface with grayish discoloration at the center of the lesion. Central graying can be present as the result of the regression that begins to occur while the periphery is still proliferating. It usually leads to impending ulceration that represents necrosis of the epidermis. Scarring can eventually occur when ulceration extends to the dermis or deeper.⁵ Early resection followed by cheek flap reconstruction was performed in this patient with the aim of preventing the possible occurrence of unfavorable scar that can interfere with her appearance.

Choosing the best reconstructive technique was crucial in this patient as the defect was located on her midface and the remaining tissue was insufficient to be closed primarily. The ideal result of face reconstruction should have no distortion and good agreement of the skin tone. Local skin flap was chosen due to its similar features to the missing tissue according to “replace like with like” principle which results in more superior aesthetic outcome compared with the other reconstructive techniques. Cheek flap was used in this case considering the defect location and the availability of surrounding skin. The advantages of using this flap are its sufficient blood flow, flexibility, and ease of manipulation.⁹

The previous perception about surgical management of proliferative phase hemangiomas is a difficult procedure due to the highly vascular lesion of the tumor that can cause inconvenient intraoperative bleeding.^{8,10} However, it is actually more beneficial as there is a “pseudocapsule” formation as the result of the tumor rapid growth that generates a clear delineation between the lesion and surrounding subcutaneous fat. The excision can be essentially bloodless by remaining within this subcutaneous plane. Minimum intraoperative bleeding was also achieved by vasoconstrictor subcutaneous injection and vessels ligation around the tumor. Surgical management in this patient was quite challenging as the tumor location was in the one of the facial danger zones. Therefore, a meticulous facial dissection was done in order to keep the dissection plane from passing through the SMAS, avoiding vessels and nerves injuries.¹⁰

The patient was followed up on the fifth day, second week and the first month after surgery. There was no clinical sign of important structures injuries that were evaluated as soon as the facial edema subsided. Slightly surgical wound dehiscence (SWD) occurred on the fifth day after surgery. SWD is one of the possible complications after surgery that can be caused by several factors such as inadequate wound undermining, excessive hematomas formation and excessive tension of the wound. This condition can be treated by secondary intention or re-suturing the wound with adequate debridement.^{11,12} The result was favorable scarring in the first month after surgery. Some retrospective studies also reported successful outcome of facial hemangiomas early

resection and suggested the procedure for IH management as it is proffering the possibility to be lesion free with one procedure, reducing the occurrence of aesthetic sequelae and preventing the psychological impact.^{11,13,14}

CONCLUSION

Early resection can be established as one of the treatment choices for fast-growing hemangioma in the proliferative phase to avoid undesirable aesthetic sequelae. Delaying the procedure may allowing further enlargement of the tumor and ultimately a larger scar in the future.

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