

ISLAND ABDOMINAL SKIN FLAP FOR RECONSTRUCTION POST EXCISION DEFECT OF ARTERIO-VEINUS MALFORMATION IN LABIA MAJORA: A CASE REPORT

Jenny Indah Haryanti¹, Ahmad Fawzy Mas'ud²

1. General Practitioner; Intern Participant in Plastic Reconstructive and Aesthetic Department, Prof Dr Margono Hospital, Purwokerto
2. Consultant of Plastic Reconstructive and Aesthetic Department of Prof Dr Margono Hospital, Purwokerto

ABSTRACT

Background : Arterio-venous malformations (AVMs) defined as high-flow vascular malformations of dysmorphic arterial and venous vessels. And connected directly to one another without an intervening capillary bed. The incidence is higher in females than male (3-5:1). Globally, there are only 5 cases of genital hemangiomas reported. Various treatment has been investigated, a recent topical treatment is the application of timolol. It has minimal adverse effects, easy administration, and good cosmetic outcomes. However, the effectiveness of timolol in vulvar cases remains unclear.

Case Report : A one-year-old girl was brought by her mother with complaint of a lump in her child's genitals. The lump was appeared during the early days of her life and it was getting bigger gradually. A plastic surgeon performed excision and reconstruction procedure using the island abdominal flap technique.

Discussion : Preserving vaginal function and perineal integrity is quite challenging, and cosmesis correspondingly less important. The patient underwent surgical excision and primary closure procedure in the labia majora region, followed with abdominal flap tunneled through the mons pubis.

Conclusion: Surgical excision and primary closure are the most recommended procedure for overcoming genital lesion. A reconstruction using flap following the procedure resulted in a normal contour and well functioned genital.

Keywords: Vascular Malformation, Abdominal Flap, Island Flap

Latar Belakang: Arterio-venous malformations (AVMs) didefinisikan sebagai vaskular malformasi tipe high-flow dari gangguan pembuluh darah arteri dan vena. Dan terhubung langsung satu sama lain tanpa intervensi dari dasar pembuluh darah. Insidensinya lebih tinggi pada wanita daripada pria (3-5:1). Secara umum, hanya ada 5 kasus hemangioma di genital yang telah dilaporkan. Berbagai pengobatan telah diselidiki, pengobatan secara topical yang terbaru adalah penggunaan timolol. Penggunaan timolol memiliki efek samping minimal, mudah pengaplikasiannya, dan menghasilkan hasil yang baik. Namun, efektivitas dari timolol belum dapat dijelaskan dalam kasus-kasus vulva.

Laporan Kasus: Seorang anak perempuan berusia satu tahun dibawa oleh ibunya dengan keluhan benjolan di alat kelamin anaknya. Benjolan tersebut muncul pada saat lahir dan secara bertahap semakin membesar. Ahli bedah plastik melakukan prosedur eksisi dan rekonstruksi menggunakan teknik island abdominal flap.

Diskusi: Untuk mempertahankan fungsi dari vagina dan integritas perineum merupakan hal yang cukup menantang, dan secara kosmetik kurang bermanfaat. Pasien menjalani eksisi pembedahan dan prosedur penjahitan primer di sekitar labia majora, dilanjutkan dengan abdominal flap yang disalurkan melalui mons pubis.

Kesimpulan: Eksisi pembedahan dan penjahitan primer merupakan prosedur yang paling direkomendasikan untuk mengatasi lesi pada genital. Rekonstruksi menggunakan prosedur flap memberikan hasil dan fungsi yang baik pada alat genital.

Keywords: Vascular Malformation, Abdominal Flap, Island Flap

Received: 15 Desember 2019, Revised: 20 Desember 2019, Accepted: 27 Desember 2019

ISSN 2089-6492 ; E-ISSN 2089-9734

This Article can be viewed at www.jprjournal.com

BACKGROUND

Malformation Vascular

The International Society for the Study of Vascular Anomalies (ISSVA) Classification System classify the pathologic and hemodynamic features of vascular anomalies into two type: vasoproliferative or vascular neoplasms such as hemangioma, and vascular malformations ⁽¹⁾. Malformation Vascular is a rare vascular anomaly with inappropriately connected vasculature ⁽²⁾. Malformation Vascular are comprised of abnormally formed channels within a vascular apparatus that are lined by endothelial cells and do not undergo abnormal cellular turnover, congenital in nature but often go unnoticed at birth, never regress and grow proportionally with the individual. ⁽¹⁾

The most commonly found vascular malformations are Lymphatic Malformations (LMs), Capillary-venular Malformations (CM) Venous Malformations (VMs) and Arteriovenous Malformations (AVMs). Arteriovenous Malformation (AVMs) is congenital high flow vascular malformation of anomalous capillary beds, which shunts blood from the arterial system to the venous system ⁽²⁾. The incident rate of venous malformations are 1 to 2 out of 10.000 people per year and prevalence of 0.1-1% ⁽³⁾. The overall incidence of congenital vascular malformations in general population is 1.5% ⁽⁴⁾. They are usually located in head and neck (40% of cases), trunk (20%), and extremities (40%), the common site involving the female pelvis are rare and usually extending to the labia majora ⁽⁵⁾. Vulvar hemangiomas are often present at birth. The median age for vulvar hemangiomas patient is 2 weeks; however, 30-50% of the disease presents at birth. The incidence is higher in females than male (3-5:1). Puberty and trauma trigger the growth of the lesion and manifestation of its troublesome symptoms ⁽²⁾. They are infiltrative causing destruction of local tissue and often life-threatening secondary to massive bleeding ⁽²⁾. However, hemangiomas are rarely found in genitalia (1% of all hemangiomas) and only 5 cases of genital hemangiomas have been reported worldwide ⁽⁶⁾.

In general, AVMs can be diagnosed based upon clinical examination, bruit or thrill can be detected, Supporting examination such as ultrasonography with color Doppler and magnetic resonance imaging (MRI) is useful to detect vascular malformations and to determination of the nature and extent of a deep

vascular malformation ^(2,7). Clinically, AVMs can appear in soft tissues or bone and are typically not accompanied by pain, but rather frequent episodes of bleeding ⁽¹⁾.

Island Flap

Flap is a mass of tissue for grafting including skin, which only removed partially from one part of the body, retaining its own blood to the new location. Island flap is an advancement flap of skin and subcutaneous tissue with a pedicle of subcutaneous tissue, which constitutes the sole vascular supply ⁽⁸⁾. Island flaps is a transposition of an island of skin, including its vascularization, which replacing the defect area. Whereas the donor site is closed primarily, tunneling the flap under adjacent skin on its vascular pedicle ⁽⁹⁾. Epidermis and dermis provide flaps with significant mechanical rigidity and elimination of their connection to adjacent tissue allows significant movement along the extensible subcutaneous tissue ⁽¹⁰⁾

Case Presentation

A one-year-old girl, taken to a plastic surgeon due to a reddish lump at her left labia majora. The lump appeared since birth as a red dot and grew gradually. Upon clinical examination, the lump is occupying her left labia majora completely. It was bright red colored, soft in palpation, mobile, and painless. The superficial skin was intact, no ulcerations nor wounds. Bruit sound was negative. Considering the condition of her wound, the patient was taken to the plastic surgeon for further treatment. Laboratory blood test were within normal limits. According to the examination, this patient we diagnosed with vascular malformation. We performed excision of the vascular lesion and the post-excision defect was reconstructed with island abdominal random skin flap for her lower abdomen. Pathological analysis from excised specimen showed: arterio-venous malformation. The flap successfully provides the appropriate structure for labia majora. No clinical problems founded at the donor site

Disclosure: *The authors have no financial interest to disclose.*



Figure 1: Patient condition (A) pre surgery (B) durante surgery (C) after excision and reconstruction with Island Skin Flap.

DISCUSSION

Arterio Venous Malformations on labia majora is a rarely found, AVMs marked by early proliferation, stabilization, followed by spontaneous involution⁽¹¹⁾. AVMs of the vulva may present as a lump prior to puberty, due to physiological increase in blood flow on the vulva⁽¹²⁾. A vascular malformation is a structural anomaly which can be easily differentiated from a capillary hemangioma by anamnesis and clinical exam. AVMs may grow larger under a few circumstances including infection, trauma, or hormonal influence⁽⁶⁾.

A one-year-old girl, taken to the plastic surgeon with a reddish lump at her left labia majora. The lump showed up since birth as a red dot and grew slowly ever since. At time of clinical examination, the lump already occupied her entire left labia majora, bright red colored, soft in palpation, mobile and painless. The superficial skin was intact, no ulcerations nor wounds. Bruit sound was negative. Laboratory blood test was within normal limits. In theory, the clinical symptoms above are clinical symptoms of AVMs. This is consistent with the results of pathological examination that described arteriovenous malformation. However, there are some symptoms that are not fit with the theory, which are an absent of bruits and pain.

The remaining defect was reconstructed with the island abdominal flap method. This method has many advantages, first it closes some wounds with one cosmetic unit, maximizing the concordance of tissue quality, color, texture, and thickness. Another advantage is excellent blood supply provided by vascular pedicle⁽¹⁰⁾. According to reconstruction ladder, when treating a soft tissue injury a surgeon should initially consider primary closure. If that solution is not feasible, then local flaps and advancements from adjacent tissue could be considered. If skin grafting does not adequately provide durable coverage in the areas, then a so-called distant flap such as a multiple stage pedicled groin flap might be proposed⁽¹³⁾.

A skin flap has its own blood supply. Flaps are usually required for covering recipient beds that have poor vascularity, covering vital structures, reconstructing the full thickness of the eyelids, lips, ears, nose and cheeks and padding body prominence. preferable when it may be necessary to operate through the wound at a later date to repair underlying structures⁽¹⁴⁾. The choice of flap depends on wound location, defect size, quality of the surrounding skin, location of adjacent excess tissue. One should anticipate the appearance of the donor site scar and when possible, plan to leave the scar in a natural crease line⁽⁹⁾. Flap may be chosen because the aesthetic result will be superior. A skin flap consists of skin and subcutaneous tissue transferred from one part of the body to another with a vascular pedicle preserved for nourishment⁽¹⁴⁾.

The island pedicle flap provides a number of advantages. First, it makes closure of some wounds with one cosmetic unit, maximizing the concordance of tissue quality, color, texture, and thickness. Similarly, maintenance of sebaceous quality can significantly improve the final cosmetic result of closure. Another advantage is the excellent blood supply provided by vascular pedicle, no need to remove surgical dog-ears or Burrow's triangles, also minimize tension on the cutaneous wound. In instance, any reconstruction requires restoration not only with skin but also with tissue bulk, subcutaneous island pedicle flap can be used to fill deeper defects⁽¹⁰⁾.

CONCLUSION

Island abdominal flap is a good, robust flap to cover a defect which need excellent blood supply. However, the disadvantage is the cosmetic appearance of the triangular scar after an island flap is far superior to that of a skin graft because of retention of the skin's texture and quality.

Corresponding author :

Jenny Indah Haryani

General Practitioner; Intern Participant in Plastic
Reconstructive and Aesthetic Department.

Prof Dr Margono Hospital, Purwokerto.

jennyindah@gmail.com

REFERENCES

1. Joshua A Cox, Erica Bartlett, Edward I Lee. Vascular Malformations: A Review. Thieme Med Pub. 2014; 28:58-63.
2. Richter, Gresham T.; Friedman, Adva B. Hemangiomas and Vascular Malformations: Current Theory And Management. Int Journal of Pediatrics. 2012 January.
3. Beata Durcanova, Alvin Y. Chan, Joeseeph A. Osorio, Cynthia Chin, Sean P. Ferris, Andrew W. Bollen, Line Jacques. Venous Vascular Malformations and Compressive neuropathy. elsevier. 2019 January; 16: 113-116.
4. Sandra Eifert, J. Leonel Villavicencio, Tzu-Cheng Kao, Bettina M. Taute, and Norman M. Rich. Prevalence of deep venous anomalies in congenital vascular malformations of venous predominance. J vasc surg. 2000; 31(3): 462-471.
5. Brian M. Christenson, Matthew G. Gipson, and Mitchell T. Smith. Pelvic Vascular Malformations. Sem Intervent Rad. 2013; 30:364-371.
6. Pankaj P. Dangle, Lopa L. Pandya IV, Casimir F. Firlit. Bilateral Asymptomatic Incidental Vulvar/Labial Hemangioma: Kissing Hemangiomas. Page Press. 2011; 25:e10.
7. Burrows PE. Vascular Malformation Involving The Female Pelvis. Sem Intervent Rad. 2008; 25(4):347.
8. Dorland WAN. Dorland's Illustrated Medical Dictionary. In Flap. Philadelphia: Elsevier Sndrs; 2011. p. 715.
9. Mary Tschoi, Erik A. Hoy, Mark S. Granick. Skin Flaps. Clin In Plast Surg. 2005; 32: 261-273.
10. Arash Kimyai Asadi, Leonard H. Goldberg. Island Pedicle Flap. Dermatol Clin. 2005; 23: 113-127.
11. P.E. Nichols, J.E. Michaud, M.H. Wang. Infantile Hemangioma Of The Clitoris Presenting As A Clitoral Mass: A Case Report. Elsevier. 2017 September; 15: 44-45.
12. Julie Beveridge, Frankie OG Fraulin, Ulrich Amendy. Management Of A Rare Case Of Multiple Arteriovenous Malformations In The Context Of Neurofibromatosis Type 2. Plast Surg Case Studies. 2015; 1(1): 15-18.
13. Fu-Chan Wei, Samir Mardini. Flaps And Reconstructive surgery. 2nd ed. Wei FC, editor. British: Saunders; 2009.
14. Thorne CH. Grabb And Smith's Plastic Surgery. 6th ed. Thorne CH, editor. New York: Lipponcott Williams and Wilkins; 2007.