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Case report
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Use of preputial skin as cutaneous graft in post excision of a verrucous hemangioma of the thumb

Başparmağın verrükoz hemanjiom eksizyonu sonrası kutanöz greft olarak Sünnet derisinin kullanılması

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Abstract

Although the use of preputial skin as cutaneous graft is not new, it was seldom been utilized and reported by surgeon all over the world. The preputial skin graft has many advantages when use as a full thickness skin graft to cover a defect over certain importance area. Herein, we reported a case of a rare vascular anomaly of thumb; Verrucous Hemangioma when a surgical excision was indicated, but the skin defect would have been too large to be directly closed, therefore the foreskin was taken as a full-thickness skin graft to cover the cutaneous defect of the thumb. The procedure was taken in a single stage with the ritual circumcision. The graft intake was favourable and provided a good functional repair with satisfactory aesthetic characteristic. Similar cases have not been reported before in the literature or at least in author's origin of country.

Keywords: Preputial, Foreskin, Skin graft, Verrucous Hemangioma, Circumcision

Öz

Sünnet derisinin deri grefti olarak kullanımı yeni olmamasına rağmen, tüm dünyada cerrah tarafından nadiren kullanılmış ve bildirilmiştir. Sünnet derisi grefti belirli bir alandaki bir kusurun üstesinden gelmek için tam kalınlıkta bir deri grefti olarak kullanıldığında birçok avantaja sahiptir. Burada, nadir görülen bir vasküler anomali olgusu bildirildi; Verrükoz hemanjiom, cerrahi eksizyon gerekli görüldü, ancak deri defekti direkt olarak kapanmak için yeterli olmayacaktı, bu nedenle sünnet derisinin kutanöz defektini kaplamak için tam kalınlıkta bir deri grefti olarak alındı. İşlem, ritüel sünnetiyle tek bir aşamada alındı. Greft alımı elverişli ve tatmin edici bir estetik özellik ile iyi bir fonksiyonel onarım sağladı. Benzer vakalar daha önce literatürde veya en azından yazarın ülkesinin orijininde bildirilmemiştir.

Anahtar kelimeler: Preputial, Sünnet derisi, Deri grefti, Verrükoz hemanjiom, Sünnet

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Introduction

Verrucous hemangioma (VH) is an uncommon capillary vascular malformation, frequently clinically mistaken for Angiokeratoma. About 95% of the cases arose from the lower extremity and these are commonly unilateral. It might involve unusual anatomic positions such as the abdomen, arm, and glans penis [1]. VH if left incompletely excised have a great chance of recurrence. The prepuce skin is a good autologous full-thickness skin graft in some conditions and most frequently been used in urethral reconstruction for congenital or acquired penile defects, in burn reconstruction, eyelid resurfacing, and in syndactyly repair [2,3]. However in the last 20 years, use of preputial skin graft (PSG) has been increasingly described for many more conditions such contracture release, eyelid and anal canal reconstruction, intraoral burn reconstruction, defect closure post nevus excision and penile skin defect repair [4].

To the best of our knowledge, there was no literature reported a VH located at the thumb in children. In this report, we demonstrated the excision of VH of the thumb and the use of foreskin following a ritual circumcision as a full thickness skin graft for coverage. We also discussed the comparison between the preputial graft to its counterpart and a brief literature review.

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Case presentation

Two years and six months old boy was brought to Out-patient Clinic with a complaint of swelling of his right thumb since at birth. The lesion gradually increased in size, painless and occasionally bled upon contact. Physical examination revealed a well-defined 2 cm x 2 cm hard dome shape with multiple dark bluish spots located at the skin overlying interphalangeal joint of right thumb on the ulnar aspect (Figure 1). The mass was firm, immobile, non-tender, non-pulsatile, no ulceration and no bleeding. Based on clinical findings a provisional diagnosis of a low flow vascular malformation was made.

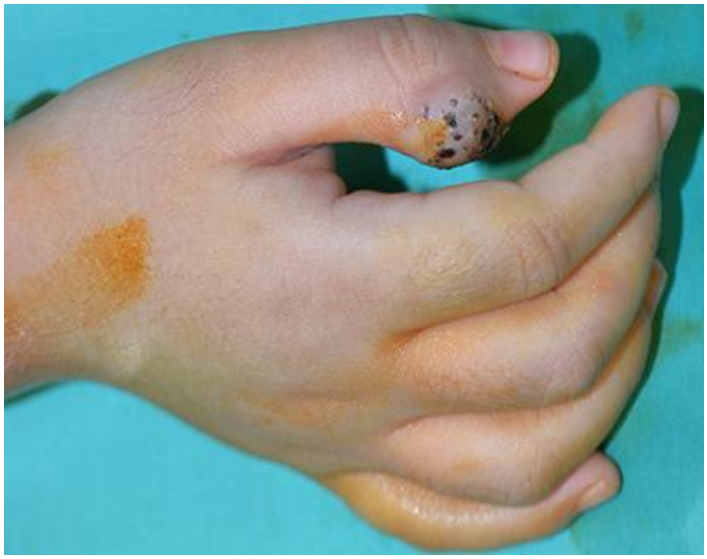

Figure 1: Vascular malformation of the right thumb

The child was scheduled for an excision with a cutaneous graft as coverage for the defect. The parent had decided that the child to undergo the ritual circumcision in the same setting as the excision surgery and had agreed that to use the excised foreskin as the cutaneous coverage of the said defect. The lesion was excised and the circumcision was performed using the dorsal slit technique. The graft was fixed to the defect with an absorbable suture. Histopathology examination revealed a proliferation of anastomosing capillaries with focal cavernous dilatation. The epidermis layer showed hyperkeratosis and acanthosis. There was intact basal granular layer with elongation of the rete ridges. Vascular proliferation was also seen at mid-dermis and these capillaries were lined by flattened endothelial cells without nuclear atypia and no mitosis was seen (Figure 2). Hence the final diagnosis was VH. Six month post operation showed the skin graft has healed well with no functional limitation (Figure 3). There was also no residual or new vascular anomaly.

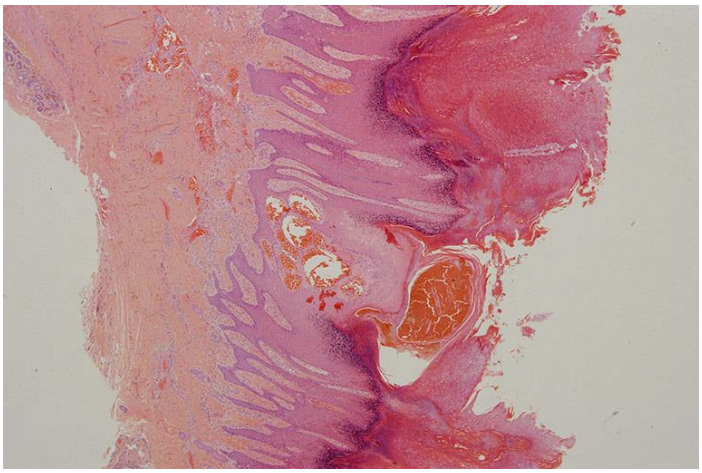

Figure 2: The epidermis layer showed hyperkeratosis and acanthosis with an intact basal granular layer with elongation of the rete ridges. There was proliferation of capillaries within epidermis and dermis without nuclear atypia and no mitosis was seen (H&E: 40x)

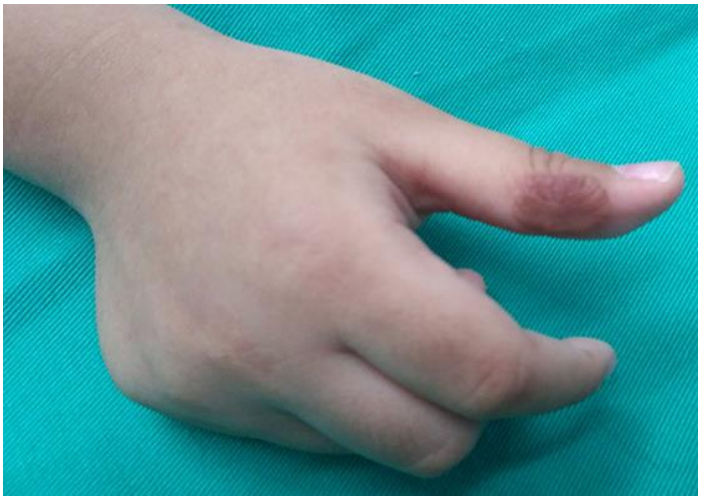

Figure 3: Six months post operation with skin graft healed well and stable. No new or residual vascular anomaly was noted.

Discussion

The clinic-pathologic characteristics of VH were first reported by Imperial and Helwig in 1967 in a study of 21 cases and distinguished it from its imitator, Angiokeratoma. VH signifies a rare congenital vascular anomaly that presents at birth or in early childhood. It is usually unilateral and localized to the lower limbs. VH in upper limb is very unusual and although few had described it on fingers but no literature had ever reported VH at thumb in children population as in our case. It also does not involute spontaneously and can persist after inadequate excision [2]. With time it darkens, thickens to become hyperkeratotic, and occasionally bleeds with ulceration, without regression [5]. Contrarily to other forms of vascular anomalies, in VH categorization as a vascular neoplasm or vascular malformation cannot be established [6]. Regardless of its misrepresentative name, VH is considered by most a malformation rather than a true hemangioma [7] and it was lately been categorized as ‘unclassified’ in the 2014 International Society for the Study of Vascular Anomalies (ISSVA) classification. However, North et al. [5] textbook on vascular anomalies pathology as well as per McCuaig CC proposal (2017), suggested that Verrucous Venulocapillary Malformation is a more appropriate designation for this lesion.

By histopathology examination, it should be distinguished by the presence of epidermal verrucous hyperplasia along with dilated capillaries and venules extending deep into the dermis with a low mitotic rate [1,5]. It also marks proliferative response of the epidermis highlighting hyperkeratosis, irregular epidermal acanthosis and papillomatosis. There is presently no specific immune-histochemical marker for VH and therefore the diagnosis should be considered after vigilant clinic-pathological correlation [7]. Early diagnosis and intervention is vital in selected patients for timely surgical excision and better cosmetic result as they do not involute spontaneously [8,9]. Inadequate excision of VH would lead to potential recurrence of the lesion from the deeper components [6,9]. As in our case, early diagnosis and surgical excision is essential as to prevent further growth and recurrence. However the excision of the lesion to a free margin had left the defect too large to be closed primarily therefore a full thickness skin graft (FTG) from the excise foreskin following a circumcision was utilized to cover the defect, hence avoiding unnecessary donor site scar.

Skin grafting originated 2500 to 3000 years ago, but it was not until the 19th century that it was presented again as a reconstructive option [4]. Baronio described the first skin graft application in 1804 on animal model whereas in humans, Bunger was the first to demonstrated it. A graft is the simplest way to cover skin defect. It comprises of the transfer of a part of skin, of variable thickness and size, which is completely separate from its original site and moved to cover the area to be repaired. Based on the thickness of the explants, skin grafts are categorized as FTG and split thickness Graft (STG) [8]. The pros and cons of FTGs over their STGs counterparts are well documented in various literature [10]. Because of poor acceptance of the resulting scars, STGs are generally reserved for deep and full thickness dermal burns, extensive skin losses in areas other than the face, and where the recipient bed is poorly vascularized. Full thickness skin grafts commonly applied to close small areas, offer solid elastic material, producing satisfactory scarring results not subject to retraction. They are particularly valuable for repair of skin loss on the face and fingers, as an alternative to local flaps [4]. Their composition of full thickness of epidermis and dermis make them able to deliver excellent color, texture and thickness matches for facial defects [8]. Most FTGs are harvest from above the shoulders, whose color, texture and thickness best match to the tissue adjacent to facial defects [11].

In last decade, an extraordinary type of FTG; PSG has been used as an alternative graft source and its application is well described in hypospadias surgery until now [4]. But PSG is still rarely used as a routine FTG among surgeons given to its easy to harvest and utilize [2,4]. Histopathologic study has revealed the prepuce mucosa to be highly vascular and to have epithelial characteristics akin to mouth, vaginal, and esophageal mucosa and very advantageous in intraoral, eyelid, and urethral repairs [10]. A study done by Mcheik et al [4], concluded that keratinocyte resulting from foreskin have a high capacity of division which enabled them to propose with their patients for wound healing especially for burns in children. The unique gains of PSG over other FTG donor sites consist of; expendable, thin and pliable; almost no donor site morbidity and no need for donor site care; very low tendency to contract; good adaptation and natural color matching, especially along the mucosal side; absence of hair follicles; hidden donor site; can be harvested with simple instrument & technique and perhaps the most significant, being an extra graft reserve site [2,4,10].

Contraindications to the use of foreskin are similar for circumcision in general, including prematurity, or a family history of bleeding disorders [12]. The main disadvantages are its only applicability to the male circumcised population and the pigmentation changes seen in the reconstructed area which restricts its usage in face and neck area [2,10]. But it can be used in extremity and scalp defects [2]. Mohammed et al [12] made a research on the use of PSGs for post-burn contractures of digits in children found that all children did not have graft loss, and all the wounds healed within 2 weeks. However hyperpigmentation of the grafts was observed, which was satisfactory well accepted.

As for in our case who happen to be a Muslim boy where ritual circumcision was a common practice had made it easier and justified to conduct the procedure. When Islam was established, the ritual of circumcision became ensconced in it. The practice was widely spread among the Arabs even before their conversion to Islam. Whether or not it was to be an absolute commitment was disputed by numerous schools of thought [13]. Circumcision is the surgical method for harvesting PSG. As with any surgical procedure, bleeding and infection are possibly the most common complications. Urethral injury and penile necrosis are extraordinary complications [14]. In some countries, circumcision is the most common surgical procedure in boys and regularly performed with very low complication rates [4].

Castellsague et al [15] reported that, with circumcision, the incidence of human papilloma infections in men and cervical cancer in their partners has decreased significantly and this procedure has become famous in some non-majority Muslim state in Asia like China and South Korea. Pang et al [16] discovered 60% of the South Korean population to be circumcised while in Turkey, circumcision is conducted generally as a symbolic ritual and a preventive medical procedure. For the similar reasons, it is recognized that 48% of infants in Canada are circumcised [17]. Moreover, there are also some absolute medical indications for circumcision, such as phimosis secondary to balanitis xerotica, obliterans and recurrent balanophosthitis, while the relative indications are paraphimosis, phimosis, redundant foreskin, and hypospadias surgery [18].

With Muslim dominance society, and circumcision being a common ritual procedure, we utilised the foreskin of children to fulfil both aims: the ritual was performed and, at the same time, the foreskin was used for coverage of the excision, obviating the need for a new donor site and second exposure to general anesthesia. The use of foreskin for male children with localized vascular malformation of the thumb has proved to be a successful method for the coverage of the hand defect with no donor site morbidity. In our case the lesion was surgically excised and showed no relapse at 6 months, indicating that surgery remain as a primary modality of treatment for a localized VH.

In conclusion, although PSG is still not in routine use, the acceptance of the prepuce skin as an alternative full-thickness donor site needs some special attention and reported series have shown promising results. Every surgeon must keep PSG in mind

as an option for donor site. VH or Verrucous Venulocapillary Malformation is a rare condition, must be distinguished for an early diagnosis for surgical intervention and to have a good cosmetic outcome. We present this case for its sheer rarity of its presentation, its notoriety for recurrence and the use of PSG as an adequate coverage after an excision.

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