Case Report

Standing on the shoulder of giants: Tubed pedicle radial forearm flap reconstruction for cutis aplasia✩,✩✩

S.S. Jing*, K. Chakrabarty

Department of Plastic Surgery, Wythenshawe Hospital, Southmoor Rd, Wythenshawe, Manchester M23 9LT, United Kingdom

ARTICLE INFO

Article history:
Received 13 January 2020
Accepted 6 April 2020
Available online 16 May 2020

Keywords:
Tubed pedicle
Scalp
Reconstruction
Radial forearm flap

ABSTRACT

Introduction: Aplasia cutis congenita presents a reconstructive challenge. We report the use of a tube pedicle radial forearm flap for scalp resurfacing in a patient who previously had two failed free flaps.

Case report: A young male patient with cutis aplasia presented with a large full thickness defect of his occiput. He had multiple episodes of recurrent infections and a history of numerous surgeries as a child including tissue expansions. As a young adult he had two failed free flap reconstructions. A CT angiogram failed to demonstrate any adequate bloody supply to the scalp that would be suitable for another free tissue transfer. We successfully performed a tube pedicled flap from his forearm achieving good functional and aesthetic results.

Conclusion: Originally described by Filatov, the tube pedicle flaps remains a useful salvage option in the armamentarium of modern-day reconstruction.

© 2020 Published by Elsevier Ltd on behalf of British Association of Plastic, Reconstructive and Aesthetic Surgeons. This is an open access article under the CC BY-NC-ND license. (http://creativecommons.org/licenses/by-nc-nd/4.0/)

✩ Presentation: the case was presented at the Celtic BAPRAS Meeting, November 2019.
✩✩ Authorship: Jing SS- drafted and revised the article, and approved the version to be published. Chakrabarty K- conceptualised and revised the article, and approved the version to be published.
* Corresponding authors at: 30 Northiam, London N12 HA, United Kingdom.
E-mail address: shanshan.jing@gmail.com (S.S. Jing).

https://doi.org/10.1016/j.jpra.2020.04.001
2352-5878/© 2020 Published by Elsevier Ltd on behalf of British Association of Plastic, Reconstructive and Aesthetic Surgeons. This is an open access article under the CC BY-NC-ND license. (http://creativecommons.org/licenses/by-nc-nd/4.0/)
Introduction

Aplasia cutis congenita, described by Cordon (1767), is an uncommon condition characterised by an absence of skin, with or without the involvement of the underlying structures. The estimated incidence is 1/10,000. The scalp vertex is involved in 86% of the cases. Twenty percent of these patients have an underlying bony defect. The aetiology remains multifactorial. Attributed risk factors include teratogens, trauma, amniotic bands, neural tube defect, uterine compression and local ischaemia during pregnancy. Familial cases of aplasia cutis congenita have been reported but are rare. Occasionally it has been linked to trisomy 13, 4p syndrome and Adams Oliver syndrome. The presentation is variable. It can be associated with multiorgan malformations involving the cardiovascular, gastrointestinal, genitourinary, musculoskeletal and central nervous systems. Long term prognosis depends on the severity of these associated anomalies. Major complications can include sagittal sinus thrombosis, catastrophic haemorrhage, infection and abnormal neurological development. Currently, there is no consensus on its management, though this depends on the mode of presentation. Small areas heal by secondary intention. Large defects are a reconstructive challenge and require careful planning.

Case report

We report a case of adult cutis aplasia successfully reconstructed with a tubed pedicle radial forearm flap following previous failed attempts in childhood. A 32-year-old male with aplasia cutis congenita presented with a large full thickness defect involving the cranial vertex and dural exposure. He had a history of recurrent infections and had numerous unsuccessful surgeries as a child including cranioplasties and trials of tissue expansions. More recently he had two failed free flap reconstructions. In view of this, a computed tomography (CT) and CT angiogram were performed of his skull (Fig. 1). These showed a significant bony defect and no adequate recipient vessels to allow further microsurgical reconstruction.

Intraoperatively, the unstable scalp skin was debrided of any residual unhealthy soft tissue. The residual defect measured 17 × 10 cm. Fortunately, no dural reconstruction was needed. A tubed pedicle radial forearm flap matching the dimension of the defect was fashioned for scalp resurfacing with success (Fig. 2). The donor forearm was grafted. A bespoke thermoplastic splint was made prior to surgery and was fitted at the end of the procedure to ensure that no compression or stretch was imposed on the pedicle (Fig. 3). The patient tolerated the splint well for four weeks, after which, division of the flap pedicle and inset of the residual flap was performed. The patient received physiotherapy for his shoulder for a short duration. No complications occurred including a frozen shoulder. A good functional and aesthetic result meant that further cranioplasty was deemed unnecessary by the affiliated neurosurgeon. The patient was very satisfied with the outcome.

Fig. 1. CT of patient’s head and neck illustrating a large bony defect at the cranial vertex.
Discussion

Early definitive wound coverage is generally recommended to prevent fatal complications in severe cutis aplasia with large defects. Various reconstructive options have been reported including skin grafts, local and free flaps. Local flaps have higher failure rates in these cases due to large flap size required and the associated abnormal adjacent skin, which can lack dermal appendages. Authors have cautioned the use of such flaps unless they have been delayed. O’Neill et al. used a bi-pedicle flap based on the supratrochlear and occipital arteries with good results. In view of the proven poor blood supply to the scalp demonstrated in this patient, local flaps or another free flap were not good options. A distant flap such as a tubed pedicle flap was therefore the only good option remaining for this patient.

Some authors believe that spontaneous calvarial growth develops with time and that bony defects do not warrant reconstruction, or least it can be deferred until skeletal maturity has been reached. Options include cranioplasty using split ribs or peri-cranial transposition flaps. Raposo-Amaral and Raposo-Amaral successfully treated an infant with a full thickness scalp defect including the dura using a local temporal flap with a periosteal patch. Radiological bony ossification of the skull was demonstrated years later. This did not occur in our patient.

The tubed pedicle flap was originally described and performed by the Ukrainian ophthalmologist Filatov, and independently by Gillies a year later. They were popularised during the pre-antibiotic era and were the workhorse flaps for reconstructing battle injuries, especially of the face. With the advent of microsurgery in then 1970s, these flaps became largely obsolete. However, they remain a useful salvage option in the armamentarium of modern-day reconstruction. A century on, tubed pedicle flaps are still being used to cover hand and facial defects.
Having learned from the founding fathers of modern-day plastic surgery, we have successfully utilised the tubed pedicle technique in the treatment of a complex scalp wound where first choice options were no longer possible.

**Declaration of Competing Interest**

We, the authors, declare no potential conflicts of interest with respect to the research, authorship, and publication of this article.

**Funding**

We, the authors, declare that no funding was required for this article.

**Informed consent**

We, the authors, declare that consent for publication was obtained for this article.

**References**

1. O’Neill JK, Carter M, Warr RP. Aplasia cutis congenita. A case of scalp defect repair using two opposing bipedicled local flaps. *J Plast Reconstr Aesthet Surg*. 2010;3:e242–e244.
2. Koshy CE, Waterhouse N, Peterson D. Large scalp and skull defects in aplasia cutis congenita. *Br J Plast Surg*. 2001;54:276–277.
3. Raposo-Amaral CE, Raposo-Amaral CA. Aplasia cutis congenita: impact of early treatment on calvarial osteogenesis. *J Plast Reconstr Aesthet Surg*. 2011;64:e237–e240.
4. Marck KW, Palyvoda R, Bamji A, Van Wingerden JJ. The tubed pedicle flap centennial: its concept, origin, rise and fall. *Eur J Plast Surg*. 2017;40:473–478.