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Partial Loss of Nasal Tissue in a Facial Vascularized Composite Allograft Patient

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INTRODUCTION

Rejection remains a problem following facial vascularized composite allotransplantation.[1] Repeated T cell–mediated rejection and associated tissue insults were previously hypothesized to cause chronic skin changes in the absence of graft vasculopathy and donor-specific antibody.[2] We present a case in which repeated insults to the mucocutaneous junction of the nasal vestibulum caused a gradual loss of the right alar base of the nose.

A 57-year-old white woman received a full facial vascularized composite allotransplantation, including maxilla, in 2011.[3] Acute rejection episodes were treated in postoperative months (POMs) 2, 17, 30, 47, and 57. The patient showed pathologies of the donor-derived sinonasal tract. A septal gap was present at the donor–recipient interface, and bilateral maxillary sinusitis was treated by endoscopic release in POMs 26 and 38, respectively. Despite these procedures, the patient continued to suffer from nasal discharge and discomfort until drainage of an ethmoid mucocele in POM 58. Although mucosal rejection is poorly understood, we hypothesize that rejection of the mucosal lining may have contributed to the severity of these findings and to the changes illustrated in Figure 1.[4]

Itching of the nose was first reported in POM 57. An ulceration inside the right nasal vestibulum (mucocutaneous junction) was noted on inspection. Concomitantly, facial erythema and skin biopsy grade II confirmed acute allograft rejection. The findings progressed under low-dose prednisone. In POM 59, topical tacrolimus, steroid taper, and changes in maintenance immunosuppression were added to treat rejection. After treatment adjustment, the right nostril healed with a small defect. Around POM 70, the ulceration reappeared without obvious skin changes. In POM 74, the patient noted episodic nose bleeds from the right nostril and an urge to scratch her nose. Topical antibiotics (Bactroban) and moisturizing cremes were ineffective. Direct biopsy of the lesion ruled out malignancy and infection. The ulceration disappeared over time, leaving a noticeable tissue defect (Fig. 1). Additionally, a defect of the ipsilateral soft tissue triangle developed. We suspect a similar chronic process that went unnoticed on clinical inspection.

DISCUSSION AND VIEWPOINT

The lesion was localized to the mucocutaneous junction of the nose. A recent study shows that higher grade skin rejection (≥BANFF II) is almost always accompanied by equal or more prominent histological signs of rejection on mucosal biopsy.[4] Therefore, biopsy-proven grade II (sub)-clinical skin rejections in POMs 57, 59, 70, and 74 are likely a surrogate of rejection inside the mucocutaneous tissue of the nasal vestibule (Fig. 1) (**see figure, Supplemental Digital Content 1**, which displays normal nasal appearance in POM 12. The inflammatory process [first noticed in POM 57] reoccurred in POMs 59 and 70 (green outlines). In POM 60, notching of the alar base can be observed (red arrow). POM 96 shows a lateral view of the end result with a striking loss of nasal tissues, **http://links.lww.com/PRSGO/B446**). We hypothesize that repeated insults due to T cell–mediated rejection, perpetuated by recurrent mechanical trauma due to scratching,[5] caused a gradual contraction and loss of the right alar base. Persistent donor-specific antibodies were not seen at any of the time points.

Presented findings appear to be in line with previously reported data on mucous membranes as a target of rejection. Chronic changes such as these may need to be distinguished from watershed ischemia due to abnormal blood supply of the allograft, as well as potential small vessel chronic vascular changes related to rejection.

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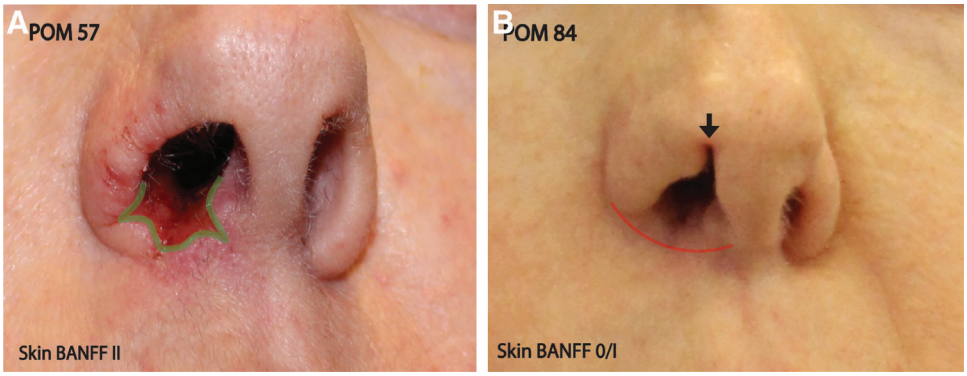

Fig. 1. First encounter of inflammation in POM 57. The process extends beyond the limen nasi and involves the mucocutaneous junction (A, green outline). Gradually, the patient shows loss of the right-sided alar base (B, red outline). Additionally, a defect of the right-sided soft-tissue triangle developed (B, black arrow-head). The defect developed gradually. (Additional pictures can be seen in **Figure**, **Supplemental Digital Content 1**.)

DISCLOSURE

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