

Case Report / Olgu Sunumu

A rare case of fistula: nasocutaneous fistula

Nadir bir fistül olgusu: Nazokütanöz fistül

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A three-year-old male patient was brought to our clinic by his family with the complaint of a non-healing wound with discharge on his forehead and the nasal region. He had a history of a fall one-year ago. The patient had recurrent complaints of swelling and hyperemia following this trauma. He had undergone an operation with the pre-diagnosis of foreign body in soft tissue six months ago. However, no foreign body had been detected and the inflamed tissue had been excised and closed again. The swelling and purulent discharge in the region between nose and eye continued after this surgery. The patient underwent an operation and a circular defect 0.5 cm in diameter was detected on the dorsum of the nasal bone and a fistula tract was found to have developed between this defect and the skin between the nose and the eye. A subcutaneous forehead flap without skin tissue was removed including the periosteum from the left part of forehead after excision of the fistula tract. The flap was transferred to the nasal dorsum and the component of the forehead flap including the periosteum was used for reconstruction of the nasal mucosa. The wound healed without any problems and no fistula or infection developed again during the six-month follow-up.

Key Words: Fistula; forehead flap; nasocutaneous fistula; subcutaneous flap.

Üç yaşında erkek hasta alın ve burun bölgesinde ivilesmeven akıntılı yara yakınmasıyla ailesi tarafından kliniğimize getirildi. Hastanın bir yıl önce düşme öyküsü vardı. Hastanın bu travmadan sonra burun bölgesinde tekrarlayan şişme ve kızarıklık yakınmaları olmuştur. Hasta altı ay önce yumuşak dokuda yabancı cisim ön-tanısıyla ameliyat edilmiştir. Ancak herhangi bir yabancı cisme rastlanmamış ve enfekte olan doku çıkarılarak yara kapatılmıştır. Burun ve göz arasındaki bu bölgede sisme ve pürülan akıntı ameliyattan sonra devam etmiştir. Hasta tekrar ameliyata alınmış ve nazal kemik dorsumunda 0.5 cm çaplı yuvarlak bir defekt olduğu tespit edilmiş ve bu defektten burun ile göz arasındaki cilde bir fistül yolunun geliştiği gözlenmiştir. Fistül yolu çıkarıldıktan sonra alnın sol yarısından deri dokusu içermeyen subkütan alın flebi periost ile birlikte kaldırıldı. Flep nazal dorsuma transfer edildi ve alın flebinin periost içeren komponenti nazal mukozanın onarımı için kullanıldı. Yara sorunsuz iyileşti ve hastanın altı aylık takiplerinde yeniden fistül ve enfeksiyonun oluşmadığı gözlendi.

Anahtar Sözcükler: Fistül; alın flebi; nazokütanöz fistül; subkütanöz flep.

A fistula is an abnormal canal or passage arising from the connection between a luminal organ and the body surface or another luminal organ, developing following trauma, illness or a congenital disorder.^[1-10] There are various types of fistulae, including oronasal,^[2] arteriovenous,^[3] enteroenteric,^[4] vesicovaginal^[5] and enterocutaneous.^[6] Although the causes of these fistulae are well known, sometimes a fistula can develop because of a rare condition.^[1] We present a patient with a very rare type of fistula, nasocutaneous fistula.

Received / Geliş tarihi: May 2, 2010 Accepted / Kabul tarihi: October 11, 2010

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Figure 1. Views of the patient at (a) the preoperative and at (b) the postoperative third month.

CASE REPORT

A three-year-old male patient was brought to our clinic by his family with the complaint of a nonhealing wound with discharge on his forehead and the nasal region. He had a history of a fall oneyear ago. The lacerated line on his nasal dorsum related to the fall had been sutured. The patient had recurrent complaints of swelling and hyperemia following this trauma. The patient had been admitted to our clinic because of these complaints six months after trauma and had undergone an operation with the pre-diagnosis of foreign body in soft tissue following a week of antibiotic therapy. However, no foreign body had been detected and the inflamed tissue had been excised and closed again. Swelling and purulent flow in the region between nose and eye continued. The complaints of hyperemia and swelling had extended to the forehead and the patient was brought to our clinic again because of a lesion causing a constant discharge at the nasomaxillary junction (Fig. 1). The patient underwent operative intervention after a

week of antibiotic therapy, and a defect 0.5 cm in diameter was detected on the dorsum of the nasal bone and a fistula tract was found to have developed between this defect and the skin between the nose and the eye. A subcutaneous forehead flap including periosteum was raised from the left part of forehead after excision of the fistula tract. The flap was transferred to the nasal dorsum and the periosteal component of the forehead flap was used for reconstruction of the nasal mucosa (Fig. 2). A dermofat graft removed from the right inguinal region during the same session was placed onto the flap donor area in order to prevent contour deformity that could later develop on the forehead. The wound healed without any problems and no fistula or infection developed again during the six-month follow-up period.

DISCUSSION

Fistulas opening to the skin or mucosa in the facial area generally arise from the frontal or maxillary sinuses.^[7,8] Other causes of fistula formation



Figure 2. Intraoperative closure of the defect. (a) Appearance of the defect in nasal bone, (b) appearance of the raised flap, (c) intraoperative final view of the patient.

in the head are oronasal fistulas, which is seen following cleft palate repair in general.^[9] However we encountered nasocutaneous fistula in only two cases in the English literature.^[10,11] A mucocutaneous fistula that developed following radiotherapy in a patient with Eales' disease was discussed in the first case reported by Fried in 1980.^[11] The nasocutaneous fistula of this patient had developed following high dose radiotherapy. The nasocutaneous fistula in the case in the current report developed from the nasal bone and the mucosal defect resulting from trauma. Fried used a forehead flap for the treatment of the case he reported.^[11] The second case reported in 1989 by Holds et al.^[10] was a nasocutaneous fistula that developed following dacryocystectomy performed on a patient with Wegener's granulomatosis.

The forehead flap used in our patient was an island flap that did not have skin tissue. The forehead flap was removed together with periosteum and periosteum was used for reconstruction of the nasal mucosa. Reconstruction of the mucosa using flaps consisting of only the fascial component without skin have been reported several times for defects of the oral cavity.[12-14] Uğurlu et al.^[12] proved that fascial flaps used for mucosa reconstructions had transformed to mucosa later. Although the use of fascial flaps in the repair of mucosal defects is not popular these days, reconstruction of a small mucosal defect surrounded by bone tissue in a profound location using a skin flap poses technical difficulties. Furthermore, had the reconstruction of the mucosa been completed using a skin flap under the flap, the greater risk for rejection of the skin flap could have caused a new infection or fistula recurrence. Thus, a fascial flap was used for mucosal reconstruction and the fistula did not recur after surgery. We did not find a case where the forehead flap without the skin island was used for mucosal reconstruction in the literature. Moreover in this case, we also aimed to decrease the risk of contour deformity in the donor site with a dermofat graft.

In conclusion, nasal mucocutaneous fistula developing after trauma is a very rare condition affecting the quality of life and damaging the anatomical structure of the face unless treated. We suggest that closure of the fistula and reconstruction of the mucosa using a flap may decrease the chance of fistula recurrence.

REFERENCES

- 1. Arora S, Garg LN, Julaha M, Tuli BS. Naso-oral fistula due to rhinolithiasis: a rare presentation. J Oral Sci 2009;51:481-3.
- 2. Sadhu P. Oronasal fistula in cleft palate surgery. Indian J Plast Surg 2009;42 Suppl:S123-8.
- 3. Ata Y, Turk T, Bicer M, Yalcin M, Ata F, Yavuz S. Coronary arteriovenous fistulas in the adults: natural history and management strategies. J Cardiothorac Surg 2009;4:62.
- 4. Jones J, Tremaine W. Evaluation of perianal fistulas in patients with Crohn's disease. MedGenMed 2005;7:16.
- 5. Kumar S, Kekre NS, Gopalakrishnan G. Vesicovaginal fistula: An update. Indian J Urol 2007;23:187-91.
- Otaigbe BE, Anochie IC, Gbobo I. Spontaneous enterocutaneous fistula-A rare presentation of enteric fever. J Natl Med Assoc 2006;98:1694-6.
- Marfatia HK, Muranjan SN, Navalakhe MM, Kirtane MV. Persistent frontal fistula. J Postgrad Med 1997;43:102-3.
- 8. Guthrie D. The operative closure of oro-maxillary fistulae. Proc R Soc Med 1932;25:1063-4.
- 9. Sadhu P. Oronasal fistula in cleft palate surgery. Indian J Plast Surg 2009;42 Suppl:S123-8.
- 10. Holds JB, Anderson RL, Wolin MJ. Dacryocystectomy for the treatment of dacryocystitis patients with Wegener's granulomatosis. Ophthalmic Surg 1989; 20:443-4.
- 11. Fried MP. Repair of nasal fistula secondary to radiation therapy for Eales's disease. Head Neck Surg 1980;2:509-12.
- 12. Uğurlu K, Hüthüt I, Ozçelik D, Ozer K, Sakiz D, Yildiz K, et al. Epithelialization process of free fascial flaps used in reconstruction of oral cavity mucosa defects in dogs. Plast Reconstr Surg 2004;113:915-23.
- Pinto FR, de Magalhães RP, Capelli Fde A, Brandão LG, Kanda JL. Pedicled temporoparietal galeal flap for reconstruction of intraoral defects. Ann Otol Rhinol Laryngol 2008;117:581-6.
- 14. Nayak VK, Deschler DG. Pedicled temporoparietal fascial flap reconstruction of select intraoral defects. Laryngoscope 2004;114:1545-8.