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Case report

Melolabial interpolated island flap for reconstruction of an anterior oronasal fistula after horseshoe Le Fort I osteotomy with iliac bone grafts interposition - A case report

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ABSTRACT

Introduction: Horseshoe Le Fort I osteotomy (HLFO) in combination with iliac bone grafts interposition is an established and very effective procedure for reconstructing the severely atrophic maxilla. However potential complications connected to this method, such as oronasal fistula (ONF), have not been described in the literature to date.

Case presentation: We report the case of a female patient with severe atrophy of the edentulous maxillary alveolar ridge with type 2 diabetes (T2D). Initially, a sinus floor augmentation was performed, followed by a failed placement of dental implants. Afterwards, HLFO with simultaneous interposition of iliac bone grafts was conducted. Subsequently, an oronasal communication occurred in the antral maxilla. As several local flaps had not achieved sufficient results, a melolabial interpolated island flap was carried out, yielding satisfactory results. Discussion: Failed implant treatment or bone augmentation procedures in combination with T2D may have resulted in significant tissue irritation and subsequent wound healing complications in the antral maxilla, leading to an ONF.

Conclusion: In this case, an ONF occurring after HLFO was described for the first time. The melolabial interpolated island flap proved to be an outstanding long-term solution for the management of an anterior ONF occurring after HLFO over a period of 10 years. Less invasive treatment options including zygomatic implants should be taken into consideration for the treatment of such patients.

1. Introduction

An oronasal fistula (ONF) is an abnormal epithelialized track communicating between the oral and the nasal cavity with multifactorial etiology [1]. Oronasal fistulas (ONFs) are very common following the failure of cleft palate repair with an incidence reported up to 68% [2,3]. ONFs may also occur during iatrogenic maxillectomies for tumor excision [4], surgical removal of ectopic teeth [5], postoperative infections [6], subperiosteal dental implantation [7], orthognathic surgeries [8], septoplasty [9], segmental Le Fort I osteotomy [10], carbonion radiotherapy [11], may be attributed to chronic cocaine abuse [12]

and finally, in some cases of medication-related osteonecrosis of the maxillary bone [13].

Based on their size, three types can be categorized: small (<2 mm), medium (3–5 mm), and large (>5 mm) ONFs [14]. Further, they can be described as anterior fistula, midpalatal fistula, fistula at the junction of the soft and hard palate, and soft palate fistula, depending on their location [6].

Due to the interconnection of the oral and the nasal cavity, clinical features such as nasal regurgitation of food and fluids, defective speech, bad taste, fetid odor, upper respiratory tract, and ear infection may occur [15].

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Abbreviations: ONF, oronasal fistula; ONFs, oronasal fistulas; HLFO, horseshoe Le Fort I osteotomy; AKH, Vienna General Hospital; OPG, panoramic radiograph; T2D, Type 2 Diabetes; aPDT, antimicrobial photodynamic therapy.

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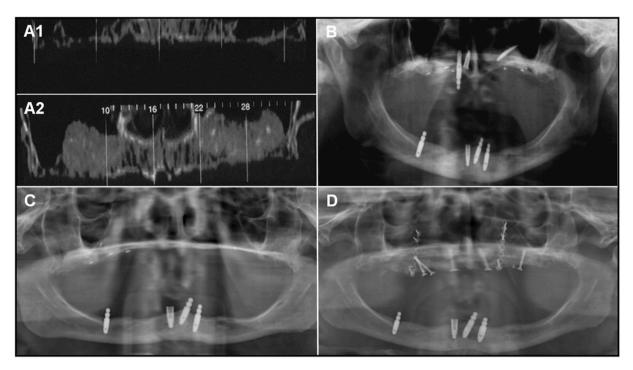


Fig. 1. A1: Dental CT before sinus floor elevation; A2: dental CT after sinus floor elevation; B: OPG showing displaced dental implants; C: OPG before HLFO; D: OPG after HLFO.

Although the literature of the past decade includes numerous cases of ONFs, e.g., occurring as a result of injuries in the maxillofacial region, cleft palate surgery, oncological surgeries, or extraction of teeth retained in the maxilla, to date no publication describes any case of an ONF after horseshoe Le Fort I osteotomy (HLFO) and iliac bone grafts interposition for maxillary rehabilitation.

HLFO is a safe and reliable surgical method for the reconstruction of the severely atrophic maxillary alveolar ridge [16].

This technique is based on the Le Fort I level osteotomy first described by Bell et al. in 1977 [17]. However, a decisive disadvantage of this method was the remaining flat palatal shelf. Therefore, dental prostheses were not retained against anterior displacement and could not gain sufficient retention [18]. In 1980, Härle and Ewers first described a modification of the horseshoe-shaped osteotomy in the Le-Fort-I level [19]. The first clinical long-term results of this method were published by Fermand et al. [20] and Sailer et al. [21]. HLFO alongside endosseous implantation was first introduced by Ewers et al. [22] and Watzinger et al. [23]. In 2004, Yerit et al. [18] demonstrated that the HLFO, in combination with harvested iliac bone grafts for sandwich interposition, is an effective pre-prosthetic procedure for the treatment of severe atrophy of the maxilla prior to implant placement. Furthermore, it leads to satisfying survival of implants and rehabilitation of function. Delayed implantation after HLFO is described as the method of choice and enhances the likelihood of successful treatment [18]. Moreover, it is a useful technique for repositioning of the maxilla with high accuracy and postoperative stability [24].

Additional modifications of the HLFO, in which the palatal component is further divided, can be successfully applied for treating scissor bites [25], cases of severe gummy smile, or asymmetry of the maxilla [26] as well as cases of vertical maxillary excess [27].

Finally, despite recent literature indicating that the HLFO and its modifications are safe and reliable methods devoid of severe complications (e.g., hemorrhage, avascular necrosis, devitalization of teeth, or oroantral/oronasal fistulas) [27], as described in this case, a persistent anterior ONF occurred after HLFO with the interposition of iliac bone grafts for the first time.

Even though in some cases, non-invasive treatment of ONFs e.g., due

to obturator dentures may be sufficient [28], the final management of ONFs usually requires surgical treatment [15]. The chosen method of surgical treatment depends on the size and site of the defect, the age of the patient, and the associated comorbidity [15,29]. The recurrence of ONFs after closure remains a challenging problem, with a rate of 33% to 37% [29].

While a wide range of flap techniques for the repair of ONFs have been described, e.g., local, regional flaps, free vascularized or non-vascularized flaps [30–33] or multilayered closing via endoscopic technique [34], as yet there is no consensus regarding the most effective treatment method [35]. Since each method has benefits and weaknesses, the selection strongly relies on the characteristics of the defect.

Melolabial interpolated flaps, which have frequently been used for reconstruction of nasal defects [36], can be successfully used to treat anterior ONFs [31]. They are vascularized from perforating branches of the facial artery with venous drainage from the facial angular vein. Melolabial cutaneous flaps are harvested from the cheek lateral to the nasolabial crease, may be either superiorly or inferiorly based, and they can be differentiated between peninsular flaps or island flaps. In this case, we used a superiorly based melolabial interpolated island flap.

The patient was managed at the Vienna General Hospital (AKH) and all procedures at this facility were performed by experienced board-certified maxillofacial surgeons. This case report has been reported in line with the SCARE Criteria [37].

2. Case report

We report the case of a 50-year-old female Caucasian patient. In 2007, she was referred to the AKH, Department of Maxillofacial Surgery, by her dentist with severe atrophy of the edentulous maxillary alveolar ridge and the request for a fixed restoration of the maxilla. She was suffering from Type 2 Diabetes (T2D) controlled by oral medication, hypothyroidism and mild hypertension, but apart from that had unremarkable family and psychosocial histories and was a non-smoker. A dental CT was taken and in order to prepare a condition for implant placement, a bilateral sinus floor elevation using Algipore® (Dentsply, Germany) bone grafting material was performed under local anesthesia.

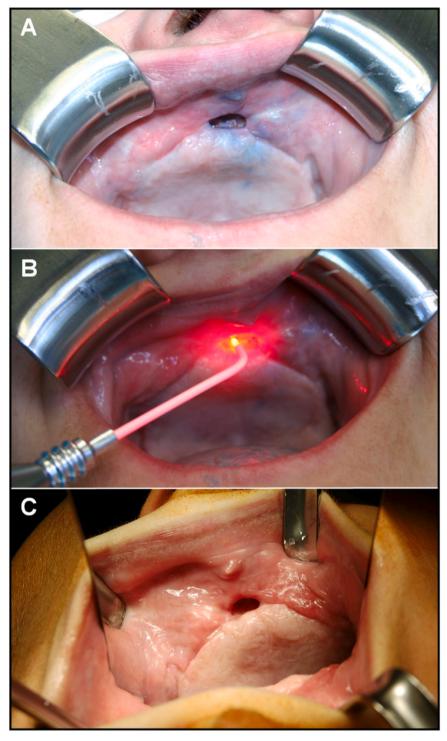


Fig. 2. A: ONF after application of the photosensitizer for aPDT; B: laser light radiation for aPDT: Slightly reduced ONF after several local flaps and aPDT.

The patient remained under supervision and was referred back to her dentist for implant placement (Fig. 1).

In 2008, the patient returned to the AKH describing symptoms of paranasal pain. A panoramic radiograph (OPG) was taken, which showed displaced dental implants, one located sagittally at the nasal floor and one cranially in the nasal cavity. The implants at regions 11 and 24 were removed endoscopically, the implant at region 13 was also removed using a trepan drill and a postoperative OPG was taken (Fig. 1).

Due to the remaining high degree of atrophy, 10 months later, HLFO with iliac bone graft interposition was conducted under general

anesthesia, in order to provide implant restoration at the second attempt. Surgical access to the maxilla was provided by circumvestibular and median palatal incisions. By raising bilateral mucoperiosteal flaps up to the palatine arteries, the palatal roof was exposed and then osteotomized medially to the palatine arteries. Afterwards, conventional Le Fort I osteotomy was carried out and the horseshoe-shaped alveolar process was carefully downfractured. The hard palate remained pedicled to the nasal septum, vomer, and nasal mucosa. By analyzing cephalometric radiographs and models the desired position of the alveolar crest was determined. Corticocancellous bone grafts, harvested

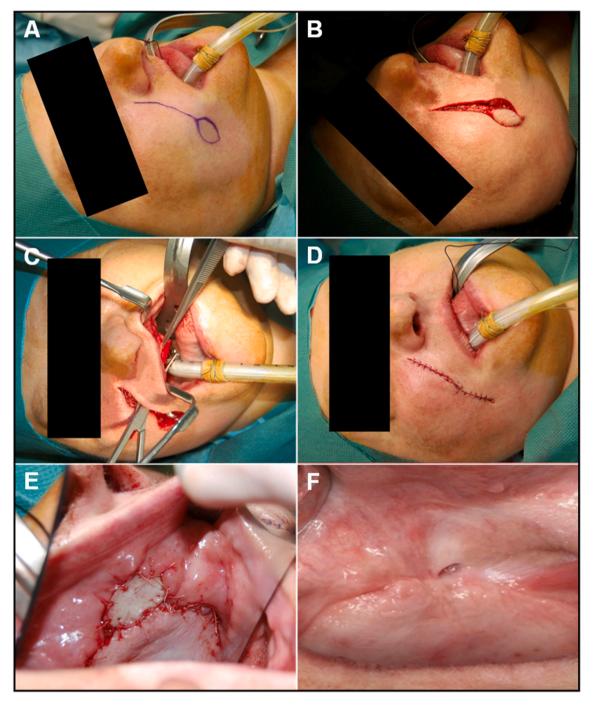


Fig. 3. A: Preoperative design of the flap; B: harvested melolabial interpolated island flap; C: tunneling and partial de-epithelialization of the flap pedicle; D: wound closure of the donor site; E: carefully sutured skin island covering the defect; F: clinical situation 10 years after the melolabial interpolated island flap.

by use of a medial approach to the anterior iliac crest, were taken simultaneously with the horseshoe osteotomy and used for sandwich interposition. The remaining space was filled with bone substitute Bio-Oss® (Geistlich, Germany) and the alveolar ridge was fixed with microand mini plates (Fig. 1).

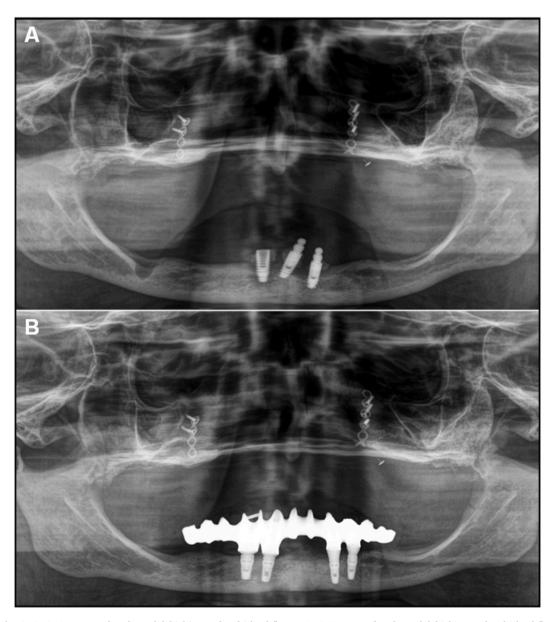
The patient remained under supervision and was given postoperative antibiotics, a non-steroidal anti-inflammatory agent and a proton pump inhibitor and was discharged in stable condition.

Nine months after the HLFO, the patient returned reporting food leftovers in the oronasal cavity. During the medical examination, an oronasal fistula (ONF) was probed. Due to an infection, partial removement of the iliac bone grafts and osteosynthesis material was performed under general anesthesia. To close the ONF, several attempts with local

flaps were performed.

Furthermore, antimicrobial photodynamic therapy (aPDT) was applied in order to reduce bacterial occurrence and accelerate wound healing. Although the ONF reduced in size, there was still communication between the oral and the nasal cavity (Fig. 2).

The following approach for treatment was a melolabial interpolated island flap. At that time, the patient received perioperative antibiotics and surgery was performed under general anesthesia. A superiorly based melolabial interpolated island flap was harvested from the right side, supplied by the facial artery. The circular-shaped flap design was based on the size of the defect and the incision length based on the rotation needed to reach the defect. A thick base of the flap pedicle was maintained in order to avoid trauma and bad vascularization. Access to the



 $\textbf{Fig. 4.} \ \, \text{A: OPG 5 years after the melolabial interpolated island flap; B: OPG 10 years after the melolabial interpolated island flap.} \\$

oral cavity was achieved by preparing a tunnel through the upper lip located opposite to the ONF. The parts of the flap pedicle that are in the tunnel have been partially de-epithelialized. Finally, the skin island covering the intraoral defect was carefully sutured with interrupted resorbable sutures (Vicryl® 3-0; Ethicon, Germany) into its final position. Wound closure was performed without tension on the local flap. The donor site was directly sutured in layers. The patient remained under ongoing supervision and was discharged in a stable condition given postoperative antibiotics, a nonsteroidal anti-inflammatory agent, and a proton pump inhibitor.

Postoperative follow-up showed good wound healing with no further complications and the integrities between the oral and the nasal cavity were successfully restored (Fig. 3). The patient remained under supervision and regularly came for routine check-ups to the Center for Oral and Maxillofacial Surgery at the Danube Private University. No further clinical abnormalities could be detected in the following check-ups and during the most recent appointment, ten years after the closure of the ONF, clinical and radiological examination showed the melolabial interpolated island flap was successful and no ONF was probed (Figs. 3 + 4). Moreover, the patient is clinically symptom-free and is satisfied with

the outcome of the treatment. Finally, the patient was prosthetically restored with a removable full denture. She is pleased with this solution and refuses implant placement or any other surgical interventions in the maxilla due to the previous procedures.

3. Discussion

As mentioned above, the HLFO is an established and very effective procedure for the reconstruction of the highly atrophic maxilla. To our knowledge, complications of this method have not been described to date [27].

In our opinion, the reason for the anterior ONF might be a combination of T2D and the prior failed implant treatment, which may have led to significant tissue irritation and therefore, subsequent wound healing disorders in the antral maxilla. Diabetes can be a risk factor for complications such as delayed and impaired wound healing after dental implant placement or augmentation surgeries [38]. Furthermore, diabetes can be considered an independent risk factor for surgical wound infections after reconstructive procedures [39].

The melolabial interpolated flap is a proven technique for

reconstruction of nasal defects and covering anterior ONFs [31,36]. In our case, a melolabial interpolated island flap proved to be a successful long-term solution with minimal loss of function and aesthetic impairment of the donor site to cover an anterior ONF occurring after a HLFO in a TD2 patient over a period of ten years.

We were aware of the fact that an invasive treatment could trigger postoperative complications, however, due to the extreme maxillary atrophy, surgical intervention was essential to provide a sufficient bone level for implants or retention for a dental prosthesis in order to improve the patients' overall quality of life. Furthermore, as suspected by Yerit et al. [18], even as implants were not inserted, due to the HLFO the patient benefited from great retention of the denture, restored facial esthetics, better speech and masticatory function.

4. Conclusion

Although the HLFO is a widely used method for reconstruction of the highly atrophic maxilla, complications such as an ONF have been described here for the first time. Furthermore, the melolabial interpolated flap proved to be an outstanding long-term solution for the management of an anterior ONF occurring after HLFO. Therefore, anterior ONFs occurring after HLFO should be treated primarily with a melolabial interpolated island flap since local flaps showed inadequate results. Finally, less invasive treatment options including zygomatic implants, short implants, custom-made subperiosteal implants or customized allogenic bone blocks should be taken into consideration for the treatment of such patients.

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Ethical approval

The ethical approval has been exempted by our institution.

Consent

Written and informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Benedikt Schneider and Florian Pfaffeneder-Mantai: study concept and design, writing the paper. Pascal Grün, Oliver Meller and Katharina Dobbertin: data collection, analysis and discussion of data. Dritan Turhani: final approval of the version to be published. All authors read and approved the final manuscript.

Registration of research studies

Not applicable.

Guarantors

Dritan Turhani, Benedikt Schneider.

Provenance and peer review

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Declaration of competing interest

The authors declare that there is no conflict of interest.

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