Noma: Experiences with a Microvascular Approach under West African Conditions

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Noma (cancrum oris) is a serious ulcerating disease that generally begins in the gingival sulcus of children. One of the main areas of prevalence today is West Africa. If noma is survived, it results in disfiguring midfacial defects and intense scarring. Oral incompetence is often combined with trismus resulting from scar formation or bony fusion between the maxilla and the mandible. Reconstructive approaches with pedicled flaps from the head or shoulder area for closure of the outer defects have been prone to donor-site complications or have not properly addressed the trismus, leading to high recurrence rates. During three West African Interplast missions, a single-stage procedure was developed for reconstruction of the inner and outer linings of the oral, nasal, and paranasal cavities, with restoration of jaw function. The procedure consists of radical scar excision, placement of an external distractor for mouth opening, and primary closure of the defect with a folded free parascapular flap for full-thickness coverage. Twenty-three patients with various noma-related defects were treated with this procedure; two cases are described in detail. This combined treatment can be a safe successful procedure for patients with noma, especially those with severe soft-tissue destruction and profound trismus, even under demanding surgical conditions. (*Plast. Reconstr. Surg.* 112: 947, 2003.)

Noma (from the Greek word *nome*, meaning a pastureland or grazing; here, a continuously spreading ulcer[1]) is a well-known entity that was first described clinically by Carel Baten in 1595.[2] Synonyms are stomatitis gangrenosa and cancrum oris; the historical name water canker refers to the constant loss of saliva during the acute clinical stage of the disease.[1,3,4] The etiopathogenesis is not completely understood, but poor hygienic standards, malnutrition,[5] and immune system compromise,[6] which seems to be partially related to measles epidemics,[7] may lead to this multibacterial facial gangrene.[8–12] *Fusobacterium necrophorum* may play a decisive role,[5,13,14] and staphylococci and anaerobes contribute to the rapid decay of tissue, with a foul odor.[15,16] Noma affects almost exclusively children,[17–19] and extremely high mortality rates are observed in countries with poor medical, nutritional, and socioeconomic standards.[1,20,21] If noma is survived under these circumstances, it leaves mutilating defects of the lips, cheeks, nose, periorbital area, and underlying bony structures.[22–24] The combination of oral incompetence, constant salivation, and trismus caused by dense scar formation or even intermaxillary ankylosis[25–27] aggravates the poor nutritional status, and the sight of the facial defects completely alienates affected patients from their social surroundings. Efforts by the World Health Organization and related organizations to control the disease and its predisposing factors were initiated only recently and proved almost ineffective,[28] whereas facial reconstructive surgical treatment helps individuals regain social acceptance.[23,29]

Reconstructive surgical treatment of noma is usually performed by local surgeons, who are sometimes supported by plastic surgical teams participating in humanitarian projects.[23,29–32] Surgical treatment is always performed after the acute inflammatory stage.[24,33] Classically, only the outer lining is reconstructed with locoregional or pedicled flaps, depending on the depth and size of the defects.[23,34] Deltopectoral flaps,[35] pedicled latissimus dorsi flaps,[3] pedicled radial forearm flaps (J. Sluimers, Bronovo Hospital, Den Haag, The Netherlands, personal communication), and various forehead flaps[1,3] can be sufficient with respect to size,

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safety, and color-matching and are relatively easy and quick to use under local conditions.[36] The inner lining either is left to undergo spontaneous healing or is covered with a split-thickness skin graft.[22,23,30] Complications during healing or secondary scarring frequently lead to unsatisfactory results,[22] especially among patients for whom trismus was released. The restoration of jaw movement requires flexible soft tissue, as demonstrated by the promising results achieved by Adams-Ray and James[26] with a platysmal flap (in a multistage procedure, however). The results regarding the recurrence of trismus are often disappointing, primarily because of the lack of qualified physiotherapy for orthognathic training and the fact that patients could reduce their postoperative pain after trismus release simply by closing their mouths.[3,27] Preventive intraoral (distractors or wooden or acrylic screws) applications have proved to be effective, if adequate dental support is left and the patient is compliant,[25,32] but in our opinion they are rarely helpful in severe cases with extensive postinfective disruption of dentoalveolar integrity.

In the past several years, the Noma Children Hospital in Sokoto, Nigeria, has become a referral center for noma patients in West Africa, treating a considerable number of patients.[20,29,31] On the basis of the knowledge of very experienced workgroups[20,29,31] and our own results on three Interplast missions, we think that two principles should be followed in major noma reconstruction cases. First, flap tissue is required for the outer and inner linings. In severe cases, locoregional flaps generally do not have adequate dimensions and pedicle length for simultaneous reconstruction. A free flap procedure has yielded excellent results under good medical conditions.[24,33] Second, an external stabilization device is temporarily required to keep the mouth in an open position, to prevent trismus recurrence and to provide safe flap healing, but should then allow sufficient motion in the full range between mouth closure and opening.

In our opinion, the free flap approach may be safely used in certain African settings, although to our knowledge there are no descriptions of a considerable number of noma reconstructions with free flaps or the use of an external distractor under such circumstances. We chose the parascapular free flap for soft-tissue coverage and developed an external distractor mounted on four 3-mm minipins in the zygomatic buttress and the mandibular body.

Patients and Methods

Patients

Between 1999 and 2001, 23 patients were treated with free parascapular flaps at the Noma Children Hospital in Sokoto, Nigeria, during three Interplast missions. The patients ranged in age from 5 to 55 years. All except one had developed noma during childhood. Eleven patients were male and 12 were female. All patients demonstrated negative human immunodeficiency virus test results, and their conditions were preoperatively classified with the standard NOITULP classification (nose, outer cheek lining, inner cheek lining, trismus, upper lip, lower lip, and particularities; Table I).[29] Only adequately nourished patients in stable condition underwent surgical treatment, after acute noma either was medically treated or had spontaneously subsided. On the basis of patient histories (established by a local translator), the findings of thorough physical examinations, and clinical judgment, all patients designated to undergo the free flap procedure were classified as American Society of Anesthe-

TABLE I
NOITULP Classification

	Loss (fraction of anatomical unit)				
	0	1	2	3	4
Nose	0	¼	½	¾	Complete loss
Outer lining	0	¼	½	¾	Complete loss
Inner lining	0	¼	½	¾	Complete loss
Trismus	Full opening	<4 cm	<3 cm	<2 cm	<1 cm or ankylosis
Upper lip	0	¼	½	¾	Complete loss
Lower lip	0	¼	½	¾	Complete loss
Particularities*	0				

NOITLUP, nose, outer lining, inner lining, trismus, lower lip, upper lip, particularities.
* Particularities represent pathological findings relevant for reconstruction (e.g., loss of palate or orbital floor).

siologists grade I or II. Preoperative small blood cell counts were performed, and patients with hemoglobin levels of less than 9 mg/dl were required to have one pint of compatible whole blood (donated by relatives) available. Despite careful hemostasis during the operation, all patients ultimately received the blood, generally because of diffuse bleeding during dissection of facial scars and release of trismus. Twelve patients with T3 or T4 trismus were also treated with an external distraction fixator.

Surgical Technique

All patients with considerable (T2 to T4) trismus underwent fiberoptic nasal intubation, with light sedation. Excellent results were achieved with coughing and forced inspiration by the patient after percutaneous cannulation of the trachea, with a 21-gauge needle, and intratracheal dispersion of 5 ml of 1% lidocaine. The fully anesthetized patient was then positioned prone on the side contralateral to the facial defect and underwent draping. Dissection of the deep neck vessels was performed through access in the carotid triangle, and the superior thyroid or lingual artery and concomitant veins were prepared under loupe magnification (4.5×). In each case, an enlarged postinflammatory lymph node overlying those vessels needed to be removed. Vessel loops were placed, and the wound was covered with a wet sponge during dissection in the face.

The facial area was injected with a 1:200,000 epinephrine solution for better hemostasis, and complete radical scar excision in all involved facial areas was performed. The bony intermaxillary bridge was chiseled away, and loose teeth were removed, especially when they seemed to impinge on the planned flap. Trismus release was performed with an interdental jaw distractor extending to an interincisal distance of 3 to 4 cm. The resulting soft-tissue defect was always larger than preoperatively anticipated.[20] Two pins of the special external distraction fixator (Fig. 1, *below, right*) were placed in the zygomatic buttress and the mandibular body. After completion of the external stabilization, a tunnel wide enough to accommodate parts of the flap and the pedicle was dissected from the facial defect to the neck area. Additional local flaps around the oral cavity (Gillies fan flap, Abbé flap, and others[1]) were used in several cases, to reconstruct the lips and oral commissures.

Meanwhile, the parascapular flap was elevated by a separate team, according to the size of the resulting defect. The flap was kept connected to the pedicle until the scar excision was complete, to reduce ischemia time. If the destruction attributable to noma involved the orbital floor or the palate, then a fascial sheet from the partially exposed latissimus dorsi at the donor site was sutured in place. For orbital support, the fascial sheet was combined with a polydioxanone sheet in two cases; for one palatal reconstruction, the sheet was folded for additional strength. The donor site on the back was closed primarily, with two drains in place. The flap was sutured into the orofacial defect, beginning with the caudal flap end in the oral cavity for inner lining. The flap was then folded, and the cranial flap portion was fit into the external facial defect. If an oral commissure was preexisting or had been reconstructed with local tissue, then the fold of the flap was deepithelialized and sutured under the lip tissue. In cases in which local lip tissue was missing, the fold of the flap was used as the commissure. The pedicle, with a short deepithelialized portion of the flap, was pulled through the skin bridge to the neck vessels. Drains were placed, and the microvascular anastomoses in the neck area were performed in either an end-to-end or end-to-side manner, depending on the situation, with 8-0 Vicryl microsutures. Finally, the neck wound was closed with 5-0 nylon sutures. No rheological measures were prescribed. All patients received an antibacterial mouthwash for 10 days and intravenously administered antibiotics (amoxicillin/clavulanic acid) for 5 days. If an external distraction fixator was mounted, it was kept static for 5 days and then carefully loosened for mouth closure, with retightening to the initial interincisal distance. This procedure was performed twice per day under surgical supervision, with adequate pain control.

CASE REPORTS

Case 1

An 8-year-old girl (120 cm, 20 kg) had a left-side facial scar resulting from noma (Fig. 1, *above, left* and *right*). The disease started at the age of 4 years and subsided after 1 year. Secondary scar remodeling caused the patient to be completely unable to open her mouth, because of bony bridging between the maxilla and mandible. The patient's condition was classified as NOITULP 0334110. Because of complete trismus (Fig. 1, *left, above* and *below*), fiberoptic nasal intubation was performed. After radical scar excision and removal of the bony bridge, the mouth could be opened intraoperatively to an interincisal distance of 3.5 cm. This result was stabilized

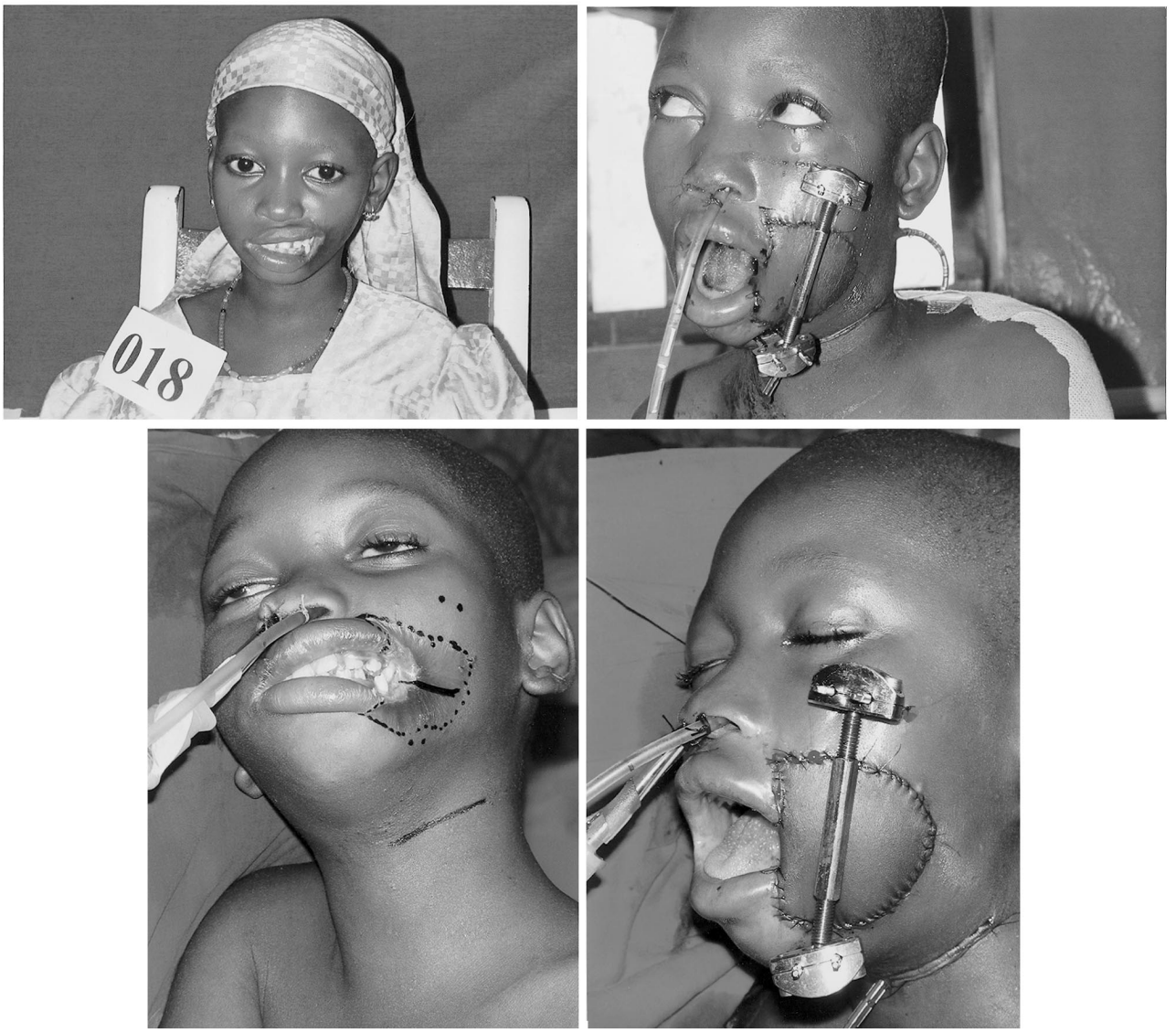

FIG. 1. Case 1. (*Above, left*) Left-side noma, with minor soft-tissue defects but complete trismus. (*Below, left*) Outer resection borders of the hyperpigmented dense scar, the zygomatic pin locations, and the neck incision for vessel preparation. (*Below, right*) Closure of the defect and partial reconstruction of the left commissure with flap tissue. (*Above, right*) Postoperative day 8. The interincisal distance was 4 cm.

with the dynamic external fixator, as described above (Fig. 1, *below, left*). A parascapular flap (15 × 5 cm) was then dissected from the right back and fit into the orofacial defect. Opening of the scar resulted in a left-side commissural defect of approximately 2.5 cm, which was closed with the fold of the flap. Vascular anastomosis to the superior thyroid vessels in the neck was performed. Eight days later, the flap was healing well, with the nasogastric tube still in place (Fig. 1, *above, right*).

Case 2

The patient was an 11-year-old boy in good medical condition (125 cm, 21 kg) who had developed left-side noma (NOITULP 133100, with the ectropion, maxillary sinus, and left lateral nasal wall gone) at the age of 6 years. Without medical treatment, the gangrene resulted in a disfiguring cavity on the left side of the face, although the other half was completely normal (Fig. 2, *above* and *center*). In this case, oral intubation was possible and a 16 × 6-cm parascapular flap, representing the new lateral wall of the left nasal cavity and the left upper gingival sulcus, was dissected from the left back. The flap was attached to the palate by means of a small deepithelialized strip. The flap was folded for extra bulk in the buccal area, deepithelialized at the nasolabial sulcus, and trimmed to fill the skin defect of the cheek (Fig. 2, *below*, and Fig. 3, *above*). No external jaw distraction was necessary. The microvascular anastomosis was performed as described above. Ten days later (Fig. 2, *below*, and Fig. 3), the flap was healing well, all sutures were removed, and a soft diet was prescribed.

RESULTS

Among the 23 patients with free parascapular flaps, 20 flaps healed without complica-

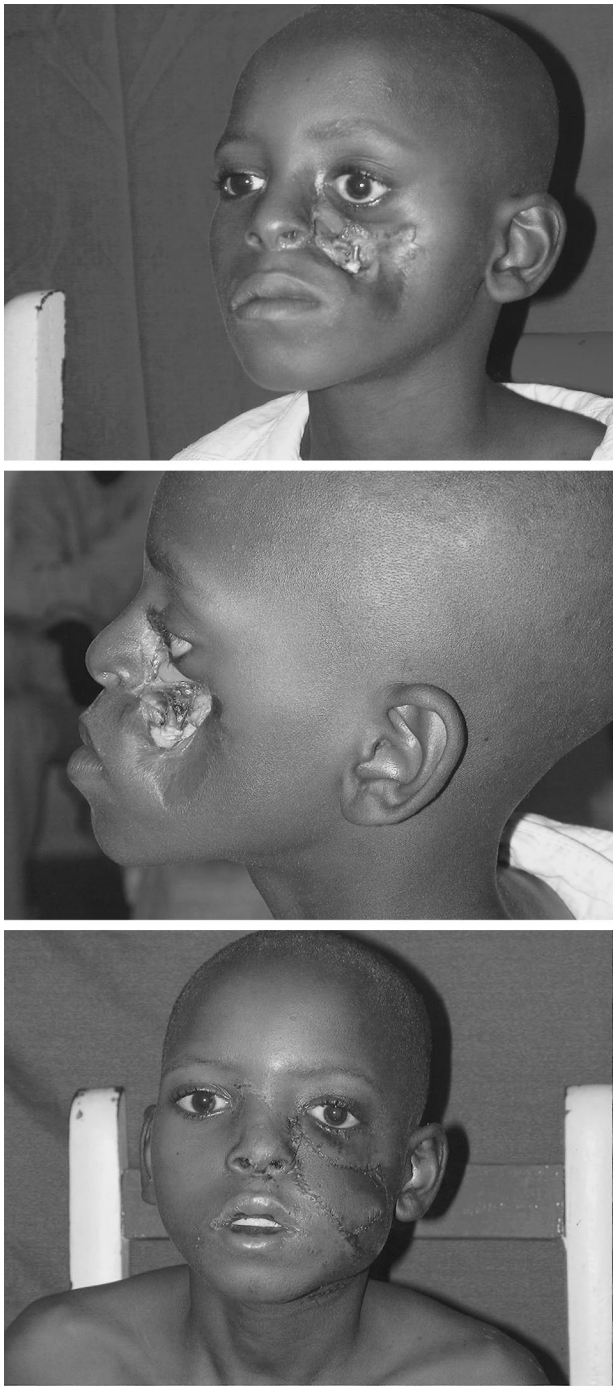

FIG. 2. Case 2. (*Above*) Scarring of the entire left cheek area, with an open maxillary sinus. (*Center*) *Anarchie dentaire*, with loss of cheek volume. (*Below*) Postoperative day 10. Good cheek volume and maxillary sinus closure can be observed.

tions. Extraoral sutures were removed after 10 days; intraoral sutures were dissolvable. We observed two complete flap losses and one partial flap loss. One flap was revised because of massive swelling observed in the recovery room, which was found to be attributable to arterial bleeding arising from the anastomosis in the neck. The bleeding was the result of a postoperative episode of agitation, which produced too much motion in the neck area. The anastomosis was revised, and further recovery was uneventful. Unfortunately, this flap developed partial intraoral tissue loss, necessitating subsequent grafting. Among the 20 successful cases, there was no wound dehiscence. One patient experienced a clinically apparent infection and fever on the third postoperative day. For that patient, several decayed teeth needed to be removed during the release of complete trismus (T4). Intravenous antibiotic therapy for 5 days led to complete resolution and uneventful healing. The two complete losses were proba-

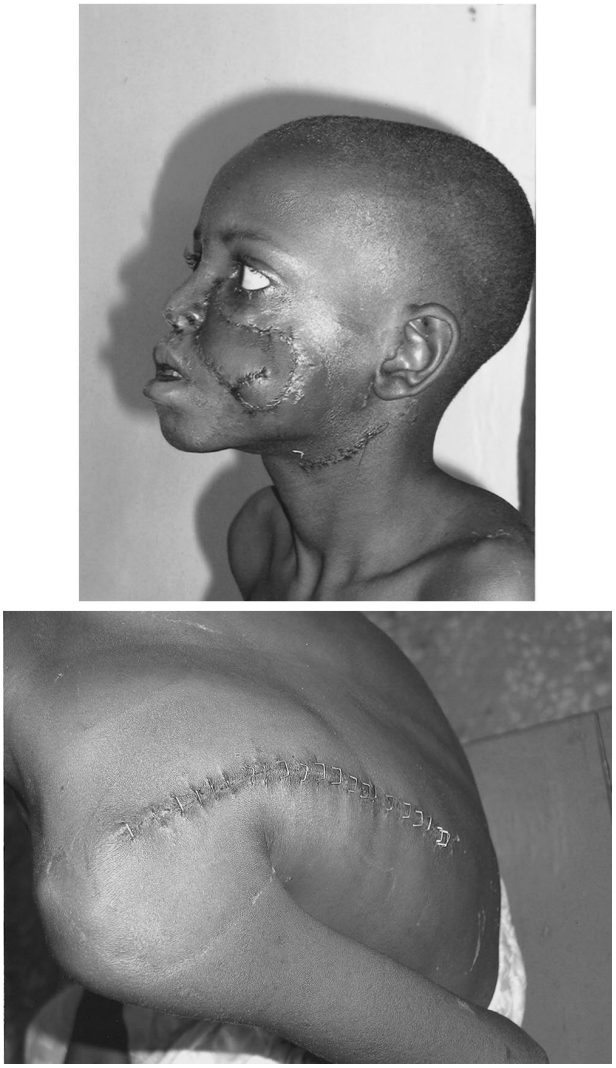

FIG. 3. Case 2. (*Above*) Postoperative day 10, side view. Some minor swelling was still present. (*Below*) Postoperative day 10, donor site after dissection of the 16 × 6-cm parascapular flap.

bly caused by arterial thrombosis at the anastomoses, and they required complete débridement. The defects were later closed with deltopectoral flaps and skin grafts. No complications were observed with respect to the donor site, with the exception of five patients with widened scars.

Interestingly, profound postoperative swelling of the flaps was present in all cases. Swelling is a well-known finding after reperfusion of free microvascular soft-tissue grafts, and it becomes even more evident in this procedure when the flap is doubled on itself and the facial skeleton resists the pressure. Therefore, care was taken during flap fitting to allow some swelling, without compromising the pedicle.

Discussion

Microvascular free flaps for the reconstruction of noma-related defects have been described in the literature, but only for individual patients who were transferred to countries with high medical standards.[24,33] We think that this approach is too expensive and benefits only a few patients; in addition, it endangers the sociocultural reintegration of the patients in their respective home countries. Our results demonstrate that microsurgical procedures can be safely performed in loco if performed by an experienced team in a hospital setting, such as in Sokoto.[29] The failure rate of approximately 9 percent is relatively high, compared with the usual Western standards. Because the surgical techniques do not differ, we explain the higher number of flap losses on the basis of several factors. First, preoperative coagulation evaluations were not available and rheologically active drugs (such as dextran, hydroxyethyl starch, aspirin, and heparin) were not prescribed to minimize postoperative bleeding complications, which would have been difficult to treat under those circumstances. Second, modern postoperative intensive care, with optimized fluid and electrolyte balances, control of coagulation parameters, and clinical monitoring and positioning of the patients, is not available in the Noma Children Hospital.

Since it was first described by Nassif et al.,[37] the parascapular flap has become a standard approach in many areas of reconstructive surgery. We chose it in the present situation for several reasons. The flap can be prepared in several shapes and sizes, and because it is associated with the subscapular artery system, it can be combined with muscle, bone, or fascia. The flap is thin and very flexible, especially among young slim patients. The flap is quick and easy to dissect, especially if another team is preparing the recipient site in the face of the prone patient. Furthermore, the pedicle vessels are large enough in caliber to be anastomosed with high-power loupes, even among 5-year-old children. For older patients, we preferred the larger lingual vessels to the smaller superior thyroid vessels because the pedicle artery and vein had considerably greater diameters. The long flap pedicle allows anastomosis of the vessels in a distant safe area of the neck. With flap folding, this results in positioning of the distal flap end in the oral cavity. Close perfusion control through postoperative intraoral inspection is required, because partial flap loss manifests there first. To gain more security in perfusion, it is possible to elevate the flap with its underlying fascia over its whole length, if fascia is not needed for orbital or palatal support.

A certain amount of subcutaneous fat can provide the bulk needed for reconstruction in the cheek area, and a skin surplus can serve as a tissue reservoir for secondary reconstructions of the nose or oral commissure. We tried to perform as much primary reconstruction as possible, to avoid second-stage operations, always taking into consideration the intraoperative status of the patient. In cases involving midfacial defects with partial or complete loss of the nose, it is tempting to include a nasal framework reconstruction in the procedure. We chose not to do so, because of the disappointing growth potential of free cartilage grafts among children (W. Muhlbauer, M.D., Ph.D., Städtisches Krankenhaus Bogenhausen, Munich, Germany, personal communication). Finally, it was easy to close the donor site primarily. We must admit, however, that the color matching was only moderately good, with the flap skin generally having a darker pigmentation than the facial skin (Fig. 1, *right, above* and *below*, Fig. 2, *below*, and Fig. 3, *above*).

In general, other free flaps would enable reconstruction of defects such as these.[24,38] Radial forearm flaps, for example, are thin enough for facial reconstruction and have been successfully used (as pedicled flaps) in the Noma Children Hospital (J. Sluimers, Bronovo Hospital, Den Haag, The Netherlands, personal communication). Unfortunately, the donor site of the radial forearm flap must be closed with a split-thickness skin graft, which is not acceptable for girls or for patients

involved in manual labor, who usually do not have their forearms covered. In addition, the classic flap might be too small for simultaneous closure of the inner and outer linings of larger defects. The same is true of the lateral arm flap, whose short pedicle can make the placement of the anastomosis very difficult. As mentioned above, the flap size is easily underestimated before trismus release.[33,39] Under other surgical conditions, combinations of two simultaneous free flaps for the outer and inner cheek linings or custom-made prelaminated flaps might be considered.[31,38,40] In our opinion, these approaches should not be used under the usual West African medical conditions. A multistage procedure, such as prelamination, represents a definitive disadvantage in this region, because follow-up monitoring, patient compliance, and repetitive plastic surgical treatment are problematic in these Third World countries.[22,33] Operating times should be minimized. Our average time for a reconstruction procedure, as described above (with an external distractor), was 4.5 to 5 hours. This value is comparable to the time for complex reconstructions with locoregional flaps in severe noma cases.

Whenever an external distractor was used, an arbitrary period of 3 months before removal was recommended. Because of a lack of patient compliance, most distraction devices were removed after 2 months. With follow-up periods of up to 24 months (three patients), we did not observe any recurrence of trismus. Obviously this period is sufficient, provided that the patient has enough teeth left to perform successful mastication and can thus maintain the temporomandibular joint in mobile condition. In our opinion, interposition of flexible, low-contractility, flap tissue between the maxilla and the mandible is the best way to prevent trismus recurrence.[22] We think that, with the reconstruction method discussed here, the free flap no longer represents "an unrealistic option"[26] for noma treatment. Further studies of postoperative distractor handling and long-term outcomes with respect to trismus are certainly required.[41]

CONCLUSIONS

The plastic surgical reconstruction of severe noma defects with a free folded parascapular flap may be a practical alternative to locoregional approaches even under certain African medical conditions. The flap eliminates the disadvantages of pedicled tissue transfer, has practically no donor-site complications, and provides important flexible flap tissue for the inner lining of the oral cavity. It should be combined with a distraction device in cases with severe trismus. Our experiences with an extraoral distractor demonstrated good results with respect to the prevention of trismus recurrence (up to 24 months).

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