


# Release of Extra Articular Ankylosis of Jaws as a Sequelae of Cancrum Oris with Extensive Gingival Myiasis in a Scoliosis Patient: A Rare Case Report

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**Abstract** ‘Noma’ is an allusion to the rapidly destructive course of gangrene (Emmanuel et al. in *J Maxillofac Surg* 11:160–170, 1983). The pathogenicity is related to the high bacterial load of microorganisms resulting in gangrene that rapidly invades the facial tissues. The infection causes perforating wounds of the lips, cheek, nose, palate and bony fusion of the maxilla and mandible.

**Keywords** Extra articular ankylosis · Noma · Scoliosis · Cancrum oris · Forehead flap · Reconstruction

## Introduction

Cancrum oris is a disease of great antiquity described by Hippocrates and Galen [1]. Adult patients with noma sequelae display gross destruction of facial tissue. The disease is found in less developed regions of Africa, Asia, and South America [2]. In addition to having little access to modern medicine, these patients often battle the triad of malnutrition, poor oral hygiene, and periodontal disease [2]. Although a reconstructive surgeon may be keen to apply the principles of oncologic reconstruction to noma surgery, it must be noted that they are dissimilar and a slightly different approach is necessary [2]. Ankylosis of

the jaws and bony fusion of the maxilla and mandible are the most disabling sequelae of noma [3].

## Case Report

A 48 year old female reported to us with an unusual complaint of worms creeping out of her mouth for the last 15 days. She had been affected by polio and could not walk. She was not able to open her mouth from childhood. On clinical examination it was found that she had her anterior maxilla and palatal floor filled with larvae (Fig. 1). She was unable to open her mouth and her extraoral skin in the region of the left commisure and left anterior malar region was completely fused with the underlying bone. A roughly triangular shaped scar was present on the left lower third of the face, extending from left lip commisure to 3 cm in front of the left tragus, 2 cm below infraorbital rim and 2 cm above the lower border of the mandible. Orthopantomogram (OPG) showed (Fig. 1) the fusion of the maxilla and mandible in the left side. She had always been on a semi-solid or a liquid diet since her childhood days through a straw placed into small openings between the teeth on the right side of her mouth. Patients medical condition was very poor, accompanied with scoliosis and haemoglobin was 3.5 gm/dl. The patient was managed accordingly with erythropoietin injection, fresh frozen plasma infusion. Nutritional status was improved with the help of multivitamin infusion. Spirometry was started to improve pulmonary functions. Debridement of the oral cavity with oil of turpentine was done for getting rid of the larvae and Intra oral irrigation with povidone iodine was performed as regular interval for improvement of the oral hygiene. Surgery was performed after 14 days of admission when her haemoglobin was raised to 10 gm/dl. Fiber

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**Fig. 1** Preoperative view showed grossly decayed teeth with no mouth opening along with dead larvae & OPG showed the fusion of the maxilla and mandible in the left side

optic assisted intubation was performed. The extra oral cheek scar which was fused with the underlying bone was removed along with the teeth in the upper and lower arches. The defect was then resurfaced with lateral forehead flap and the donor defect was covered with partial thickness skin graft. Mouth opening was thus improved by removing the ankylositic mass (Fig. 2).

## Discussion

Cancrum oris or noma is still a major health problem in many developing countries. The triad of malnutrition, poor oral hygiene, and debilitating disease contributes to its continued presence [3]. In high-income countries, noma is now extremely rare. In contrast, noma still persists in the poorest developing countries, predominantly in Africa. The epidemiology of noma shows a similar pattern, although the mortality rate has been drastically reduced from 90% to approximately 8–10% due to modern antibiotics. Although noma can affect infants, children, and adults, a slight predilection may exist for females over males [3]. The most common predisposing factors are poor oral hygiene, dehydration, malnutrition, and concomitant illnesses, such as measles, scarlet fever, or tuberculosis, malignancies,

especially leukaemia, and immunodeficiency disorders, such as AIDS [4].

The main principle of facial reconstruction in noma patients is to excise all scar tissue, thus recreating the original tissue defect, and reconstruct with well vascularized flaps [5]. The extent of the loss of tissue is easily underestimated. For that reason the ‘Sokoto law of noma surgery’ has been formulated as follows: the extent of the flap you need is always larger than you planned, even if you take into account the ‘Sokoto law of noma surgery’. Because there is a kaleidoscopic variety in noma defects and the extent of surgical options is large, there are no clear guidelines which specific flaps are to be used in any specific case [5]. In ill-equipped local hospitals however facilities such as free flap surgery and intensive care are not available in general, and in those circumstances reconstructions have to be performed with local or pedicled flaps [5].

## Conclusion

There is both soft tissue and bony compromise as a sequelae of noma that affect both the aesthetics and function. Although complete correction may be planned to restore function and aesthetics, the noma patient may



**Fig. 2** Intraoperative & 3 months postoperative view

eventually be satisfied with a functional but less aesthetic outcome. Reconstruction surgery is performed by free flaps, local or pedicle flaps which are solely depends upon the facilities available in the treating centre of the country.

#### **Compliance with ethical standards**

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethical Approval** All procedures performed in this case involving human participant was in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards—ethical approval was taken by Ethics Committee of Guru Nanak Institute of Dental Sciences and Research.

**Patient Consent** Yes—written patient consent has been obtained to publish clinical photographs.

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