

Cancrum nasalis: a case report

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Abstract

Cancrum nasalis, a variant of cancrum oris, is an oro-facial condition that is rarely reported in the literature. Predisposing factors include malnutrition, infection and reduced immunity. This paper reports a case of a 10 month-old child patient who presented with an isolated necrosis of the nose extending to the cutaneous part of the upper lip without oral manifestation, a rare clinical presentation. A maculo-papular rash which was diagnosed as measles preceded the necrosis of the nose and the child was also seen to be malnourished at presentation. Laboratory investigations and clinical management of the condition are described. It is also suggested that the public should be continuously educated on the importance of balanced diet and routine medical check for growing children.

Key words: Cancrum nasalis, malnutrition

Introduction

Noma (cancrum oris) has been described as an opportunistic infection promoted by extreme poverty and commonest in sub-Saharan Africa⁽¹⁾. The incidence peaks at 1-4 years and is a scourge in communities with poor environmental sanitation. A triple complex of malnutrition, infection and reduced immunity are usually present. Predisposing factors or antecedents of noma may include measles, malaria, severe diarrhoea or necrotising ulcerative gingivitis. Untreated cases progress to massive necrosis of oral and perioral tissues involving the face and leading to variable functional and aesthetic impairments, mid-facial defects, scarring, loss of mandibular movements, and social disintegration⁽¹⁾. These defects often require reconstructive surgery such as pedicled flaps or free microvascular flaps⁽¹⁾.

Though it has been reported that noma can involve oral, nasal, eyelid and scrotal tissues⁽²⁾, isolated nasal involvement without oral manifestation is rare. We present a case of isolated nasal involvement managed in our hospital, a similar case has been reported in India⁽³⁾. This paper lays emphasis on prevention, early diagnosis and treatment.

Case report

A 10 month old female child was brought into the paediatric out-patient clinic of our hospital by her mother with a history of fever and facial swelling of over 2 weeks duration. The patient developed high grade fever prior to presentation followed by discharge of 'white substances' in both ears, and boil-like multiple swellings in both nostrils. The child was taken to a health centre and Children Hospital in succession where, despite medications and transfusion of a pint of blood, the problem persisted. An

ophthalmologist attended to her on the eyelid involvement, and was later referred to Lagos University Teaching Hospital.

The patient was the third child of her parents who were not literate, and of low socio-economic status. Her mother commenced antenatal care at gestational period of 4-5 months and the child was a product of full-term normal delivery which was uneventful. Nutritional history showed exclusive breast feeding for 2 months after which water and pap was introduced which was gradually substituted with solid diet. Past medical and surgical history also showed that the patient was fully immunized for her age but with delayed developmental milestone, including eruption of primary dentition.

On Examination, the child was found to be malnourished, small for age and in respiratory distress, mildly pale, not jaundiced, not febrile, severely dehydrated but with no pedal oedema. The skin was dry and scaly, and the hair was thin and fluffy. Extra-orally, the necrosis of the nose extended to the cutaneous part of the upper lip, nasal septum and up to the fronto-nasal region sparing the alar. The right eye was swollen with copious excretion. The nose was gangrenous extending to the lower right eyelid. The affected tissues were necrotic, dark but not fully separated with suppuration from the eyes, which were also spared (**Figure 1**). The initial treatment included intravenous administration of ampiclox 200mg/kg/day, ceflogin 5mg/kg/day, vitamin A 50 w/u/day 1, 2 x14, Fortified meals, milk, paediatric protein supplements. Intra-orally, only the maxillary and mandibular central and lateral primary incisors were present. The oral mucosa, tongue, palate, gingivae and floor of the mouth were clinically normal.

Laboratory investigations showed low blood protein levels, 44g/l (normal range 66-78g/l); albumin, 14g/l (normal range 40-80g/l); haemoglobin, 7.1g/l; white blood cells of 12,800/cu mm; differentials were within normal range;

erythrocyte sedimentation rate was 120mm/hour. Electrolyte and urea results were: HCO_3^- , 23; Na^+ 138; Cl^- 100; K^+ 3.9; Ca 9.2; Phosphorus 4.1; Blood urea 44; creatinine 1.5mg.

An incisional biopsy showed chronic inflammatory reaction consistent with noma. Chest and abdominal radiographs were normal. Swabs for microscopy, culture and sensitivity were taken and results revealed isolation of *E. faecalis*, *E.coli* and *proteus Spp*. The organisms were sensitive to Amoxicillin + + +, Augumentin + + +, Ofloxacin + + +.

Based on the findings, a diagnosis of Cancrum nasalis secondary to protein-energy mal-nutrition (PEM) and dehydration with antecedence of measles was made. Patient was co-managed by the Oral and Maxillofacial Surgery and Paediatrics units. The patient was placed on Intravenous metronidazole 50mg, oral 125mg amoxicillin 8 hourly, both of which are active against *Borrelia* and *Fusobacterium spp*. which are common organisms in cancrum oris. In addition, metronidazole is effective against *Bacteriodes spp*. which has been implicated in the pathogenesis of Cancrum oris. Fortified meals, milk and paediatric proteinous supplements were also given.

Wound debridement was done for slough separation and subsequent removal using hibitane in water irrigation and dilute hydrogen peroxide. The wound was dressed with sulfatulle® gauze dressings were changed daily (Figure 2). When discharged home, patient attended out-patient clinic on alternate days for wound dressing. The condition improved rapidly as the residual wound healed quickly and the general condition of the patient improved (Figure 3).



Figure 2. Same patient after wound debridement.



Figure 3. Same patient with improvement 1 week after commencement of treatment



Figure 1. A 10 month old child with necrosis of right nose, nasal septum and part of upper lip.

Discussion

There have been varying incidences of cancrum oris reported in Africa and Nigeria, each characteristic to the country and communities (4,5,6,7,10). For instance, while Tempest⁽⁴⁾ found 250 cases of noma in University College Hospital, Ibadan within a 3 year period,(1962-1965), Adekeye and Ord⁽⁵⁾ in Kaduna Northern Nigeria found 140

cases over 4 years period,((1978-1982) while in North-western region of Nigeria, a noma incidence rate of 6.4/1000 has been quoted⁽⁶⁾. In recent times in Ibadan, a decline in incidence of noma was reported⁽⁷⁾. Previous workers in our centre, Sawyer and Nwoku⁽⁸⁾, reported 26 cases of cancrum oris in a year. In another study, Nwoku and Sawyer⁽⁹⁾ reported a fall in mortality rate due to the use of antibiotics as only one of their 14 new cases of cancrum oris had a fatal outcome. In the sub-sahara Africa, the frequency of noma is estimated to be between 1-7 cases /1000population in some communities and as high as 12 cases/1000 in most affected communities⁽¹⁰⁾. Some studies have also shown that nutritional cultures differ in different communities and countries and this might explain the variations⁽¹¹⁾. The patient in this report is from the south western part of Nigeria and presented with

protein energy malnutrition (PEM), infection and an antecedent or predisposing factor of measles (exanthematous fever). Among the vulnerable children, acute ulcerative gingivitis, traumatic oral lesions including tooth eruption are considered potentially capable of evolving into noma^(11, 12). The patient in this report though reported pre-existing measles and fever, did not present with any oral lesion, as the mucosa and gingival tissues were all healthy. This is the clinical significance of this paper. Thus our case is contrary to the general hypothesis that the interaction or virulence of certain micro-organisms in the oral and periodontal environment of a susceptible individual is a pre-requisite to noma^(4,10,13). It may be possible that micro-organisms of the nasal mucosa (where the lesion was located) was contaminated by oral microflora through the posterior nares or there may be similarities of organisms in both oral and nasal mucosal environments, or through yet to be identified means. Further studies in this area may be necessary.

This case is similar in presentation to noma neonatorum which usually involves oral, nasal, anal areas and occasionally eyelids and scrotum⁽³⁾. This is commonly associated with *Pseudomonas aeruginosa*. An association between noma and *Ps. aeruginosa* sepsis has been made with malnutrition playing a central role⁽³⁾.

The site of cancrum oris in this case (nasal soft tissues and bone) were not in favour of surgical closure of the nasocutaneous fistula, neither is prosthetic obturator possible until child is old enough to co-operate with such appliance.

In respect of surgical repair, most defects from noma involve lateral or antero-lateral aspects of the face. Our patient belongs to a subgroup called 'central noma' with defects composing of upper lip, maxillary soft tissues, pre-maxilla and nasal cartilaginous skeletons and soft tissues. There is no single standard surgical procedure that is sufficient for all types of defects. The common surgical approach to 'central noma' is a single stage reconstruction using loco-regional flaps which often leads to disappointing outcomes. An alternative is using a free flap reconstruction of upper lip as a versatile base for introducing loco regional flaps for functional and aesthetic refinements⁽¹⁴⁾. In this method, a secondary procedure includes total nose reconstruction with a free cartilage framework and forehead flaps. In their series,(n=53) free forearm, anterolateral thigh, para-scapular flaps all proved suitable for the central face in terms of pedicle length, tissue pliability, and bulk. Thus a micro-vascular tissue transfer preserves local and regional donor sites.

In lateral (buccal defects) of noma, temporalis muscle flap (loco-regional) has been found suitable and safe in a single stage procedure to repair buccal defects combined with release of bony or fibrous ankylosis of the temporomandibular joint (inter-positional arthroplasty). Even when medical condition is less ideal, satisfactory functional and cosmetic results are achieved by this method⁽¹⁵⁾. Other surgical procedures for lateral noma previously described include pedicled supra clavicular flap⁽¹⁶⁾, new split scar cheek flap⁽¹⁷⁾, use of naso-labial flap⁽¹⁸⁾.

Noma was once called 'face of poverty' in Europe as awareness that it was related to poverty, malnutrition, and preceding diseases such as measles increased in North-western Europe after the first half of the 18th century. However, noma has now disappeared in Europe due to

economic progress⁽¹⁹⁾. The developing nations are grappling with poverty and the only true effective approach to the problem of noma is combating poverty. It is advocated that Europe and the other economically advantaged countries should not forget their own history but rather should face the 'face of poverty' with eyes of mercy and concern in helping to combat poverty in the developing countries⁽¹⁹⁾.

Adekeye and others⁽⁵⁾ advocated the importance of educating the parents about feeding children with proteinous diets for prevention of noma. In a study in Ogbomosho involving well-nourished and cancrum- oris groups of children, total serum proteins levels were higher in the well nourished group and the difference was statistically significant⁽²⁰⁾. In the case presented, the albumin blood level was 14g/dl (normal range 40-80g/dl). The mother breast-fed the child for only two months and subsequently resorted to pap and water. The child suffered delayed developmental milestones due to protein energy malnutrition as the parents were un-informed about the importance of proteinous diets during weaning in a growing child. Public education on the need to use health facilities, provision of rural health centres, mobile clinics will bring health services closer to the people. It is interesting that the patient and the parents live in Lagos city, and yet remained un-informed of basic nutritious diet for the baby.

Further studies are suggested to clarify the possible inter-relationship between cancrum oris and cancrum nasalis. Finally, despite the rapid progress that has been made by the advent of antibiotics, the fact that this type of necrotic condition of the facial structures is still seen at this stage of our development calls for concern. It is also suggested that the public should be continuously educated on the importance of balanced diet and routine medical check for growing children.

References

1. Enwonwu CO, Falkler WA, Phillips RS. Noma (Cancrum oris). *Lancet* 2006; 368: 147-156.
2. Parikh TB, Nanavati RN, Udani RH. Noma neonatorum. *Indian J Pediatr* 2006; 73: 439-440.
3. Vaidyanathan S, Tullu MS, Lahirik R, Deshmukh CT. *Pseudomonas sepsis with noma, an association?* *Indian J Med Sci* 2005; 59: 357-360.
4. Tempest MN. Cancrum oris. *Br J Surg* 1966; 53: 949-969.
5. Adekeye EO, Ord RA. Cancrum oris: principles of management and reconstructive surgery. *J Maxillofac Surg* 1983; 11: 149-194.
6. Freger A, Marck K W, Busch R, Schmidt A. An estimation of incidence of noma in North-western Nigeria. *Trop Med Int Health* 2000; 5: 402-407
7. Denloye OO, Aderinokun GA, Lawoyin JO, Bankole OO. Reviewing the trends in the incidence of cancrum oris in Ibadan. *West Afr J Med* 2003; 22: 26-29.
8. Sawyer DR, Nwoku AL. Cancrum oris (noma): Past and present. *J Dent Child* 1981; 48: 138-141.
9. Nwoku AL, Sawyer DR. Cancrum oris mortality in Lagos, Nigeria. *Virginia Dent J* 1980; 57: 22-25.



10. Naidoo S, Chikte UM. Noma (cancrum oris): case report in a 4 year old HIV positive south African child South Afr Dent J 2000;55: 683-686.
11. Oji C. Cancrum oris: its incidence and treatment in Enugu, Nigeria. Br J Maxillofac Surg 2000;40: 406-409.
12. Enwonwu CO. Cancrum oris: Epidemiological and biochemical studies of necrotising ulcerative gingivitis and noma (cancrum oris) in Nigerian children. Arch Oral Biol 1972;17:1357-1371.
13. Yuca K, Yuca SA, Cankaya H, Caksen H, Calka O, Kiris M. Report of an infant with noma (cancrum oris). J Dermatol 2004;1:488-491.
14. Giessler GA, Cornelius CP, Suominen S, Borsche A, Fieger A J, Schmidt AB, Fischer H. Primary and secondary procedures in functional and aesthetic reconstruction of noma-associated complex central facial defects. Plast Reconstr Surg 2007;120:134-143.
15. Dammer R, Dunzl B, Kuhnel T. Therapy of bony and fibrous contractures and buccal defects after noma using the temporalis muscle flap. Mund Kiefer Gesichtschir 2005;9:225-232.
16. Hartman HM, Philip AV, Hartwig S, Sinikka HH. The use of pedicled supra-clavicular flap in noma reconstruction surgery. J Plast Reconstr Aesth Surg 2006;59:337-342.
17. Kuhmel TS, Dammer R, Dunzi B, Beule AG, Strutz J. New split scar cheek in reconstruction of noma sequelae. Br J Plast Surg 2003;56:521-620.
18. Tyran A, Kul Z, Turkasian T, Ozyigit T, Ozsey Z. Reconstruction of lower half defects of the nose with lateral nasal artery pedicle naso-labial island flap. Plast Reconstr Surg 2007;119: 1767-1772.
19. Marck KW. A history of noma, 'the face of poverty'. Plast Reconstr Surg 2003;111:1702-1707.
20. Sawyer DR, Nwoku AL. Malnutrition and oral health of children in Ogbomosho, Nigeria. J Dent Child 1985;52:141-145.