Protocol for managing acute cancrum oris in children: An experience in five cases

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ABSTRACT

Background: Acute cancrum oris (NOMA) is a major health concern in most parts of northern Nigeria, affecting children aged 2-9 years. The management of the acute phase/condition poses a great clinical challenge. We present our protocol for the management of acute NOMA in a referral health facility in northern Nigeria. **Materials and Methods:** The management of five consecutive children with acute NOMA managed at the maxillo-facial department of Ahamdu Bello University Teaching Hospital, Kaduna, is presented. The initial management is conservative, followed later by multiple reconstructions. **Results:** NOMA thrives in children with poor nutritional status and/or compromised immune status. Aggressive wound debridement, antibiotic therapy and nutritional rehabilitation limited the spread of the orofacial tissue destruction and produced rapid wound healing. **Conclusion**: The management of acute NOMA should be nonoperative; surgery is only indicated to debride the ulcer. Efforts to boost patient's immune and nutritional status limit further tissue destruction and produced satisfactory wound healing.

Key words: Cancrum oris, children, Northern Nigeria

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INTRODUCTION

Cancrum oris (NOMA) was described by Tourdes (1848)[1] as "a gangrenous affection of the mouth, especially occurring children affected by serious illnesses, especially exanthematous fevers". The disease is caused by *Borrellia vincenti* and *Fusiformis fusiformis*.[2] It usually occurs in children aged 2-9 years. It rapidly results in serious destruction of the orofacial tissues, with attendant gross aesthetic and functional defects. Various predisposing factors believed to cause NOMA include malnutrition, poor oral hygiene and debilitating diseases such as malaria fever, measles and bronchopneumonia.[3]

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The natural history of NOMA consists of two phases; namely, an acute inflammatory phase with ulceration (acute NOMA) and the healed phase in which the ulcer is healed leaving behind varying sizes of orofacial defect (chronic NOMA). Acute NOMA is a major health concern in most parts of northern Nigeria.[4] In this report, we present our experience with the management of the acute phase of NOMA.

CASE REPORTS

Case 1

An 80-year-old woman was brought to the clinic with a week-long history of swelling involving the orofacial region. The swelling was followed a few days later with a black discolouration and consequent sloughing of the affected area. Six months earlier, she had diarrhoea and vomiting that persisted despite local medications. She was from a polygamous family of 15 children; her mother was a full-time housewife while her father was a peasant farmer.

On examination, she obviously looked malnourished, with a distended abdomen, generalized muscle wasting and inability to move the limbs. There was a complete loss of the lower lips, the chin, left mandible, left check and one-third of the right check [Figure 1]. A big sequestrum of the left mandible hung loose in the mouth; most of the teeth in the lower jaw were already lost.

A diagnosis of acute NOMA was made. The laboratory data showed haemoglobin of 7 gm/dl. She reacted positively to human immunodeficiency virus (HIV) type I and II screening by enzyme-linked immunosorbent assay.

She was admitted and managed for acute NOMA. She was placed on high protein diet and was transfused with packed red blood cells. The orofacial ulcer was debrided twice daily with hydrogen peroxide solution and normal saline, after which the wound was packed with gauze soaked in Euzol solution.

Eight-hourly daily course of amoxicillin and metronidazole was administered for 1 week. The patient's parents could



Figure 1: An eight-year-old malnourished girl with acute cancrum oris, showing a complete loss of the lower lips, the chin, left mandible, left check and one-third of the right check. A big sequestrum of the left mandible hung loose in the mouth, most of the teeth in the lower jaw were already lost



Figure 2: A five-year-old girl with acute cancrum oris showing complete loss of the tissues of the left check, eyelids, eyeball, half of the upper lip, one-third of the lower lip, part of the nose and complete loss of the teeth



Figure 3: A five-year-old boy acute cancrum oris extensive ulceration, necrosis of the left side of the face involving the lower border of the chin and the supra-orbital region with complete destruction of the left eye, mandible, pre-maxilla, check and half of the nose



Figure 4: A three and half year-old boy with acute cancrum oris showing gangrene of the upper lip, the right check, the pre-maxilla, part of the nose, left corner of the mouth as well as a part of the lower lip



Figure 5: Circum-oral ulceration involving the right and left check, with complete destruction of the pre-maxilla, anterior portion of the palate, two-thirds of the nose and lateral third of the lower lip in a six and half year-old boy with acute cancrum oris

not afford retroviral drugs and while an effort was being made to raise money for her, she died of complications arising from acquired immunodeficiency syndrome.

Case 2

A 5-year-old girl presented with a 10-day history of left facial swelling, which later burst, resulting in an extensive facial defect. She had measles 2 weeks earlier and was being managed at home until the orofacial destruction intervened.

Examination showed a malnourished girl with abdominal distension and paedal oedema. Maxillofacial examination showed complete loss of the tissues of the left check, eyelids, eyeball, half of the upper lip, one-third of the lower lip, part of the nose and complete loss of the teeth [Figure 2].

A diagnosis of acute NOMA was made. The patient was placed on a high protein diet, transfused with two units of packed blood cells, and had debridement of the orofacial defects with thrice-daily dressings of the ulcer using dilute hydrogen peroxide solution and normal saline. She had a course of antibiotics (ampicillin–cloxacillin combination, metronidazole), multivitamin preparation and antiseptic therapy for 1 week. The wound healed spontaneously after 2 weeks of therapy. The patient was discharged home for monthly review in the clinics.

Case 3

A 5-year-old boy was brought to the hospital with a 5-day history of facial swelling. He had had fever and rashes 2 weeks previously. The grandmother noticed a swelling around the mouth only 5 days later. The swelling was associated with an offensive mouth odour. This was later followed by blister formation and black discolouration of the area involved.

Clinical examination showed a malnourished, irritable child gangrene of the upper lip, the right check, the premaxilla, part of the nose, left corner of the mouth as well as a part of the lower lip [Figure 3]. A diagnosis of acute NOMA was made and the patient was admitted and blood was taken for investigations. The haemoglobin level was 7.5 g/dl, the blood film showed malaria parasite (+ +) and stool microscopy showed some cysts of ascaries.

The patient was transfused with one unit of packed cells and was rehydrated with intravenous fluid. Debridement of the defect was commenced after 2 days of admission and by the fifth day, the whole necrotic tissues had sloughed off completely. He had a 2-week course of ampicillin, metronidazole and multivitamin preparation and a high-protein diet. The resultant ulcer was dressed with hypochlorite (EUSOL: Edinburgh University Solution of Lime) solution and the patient was discharged home after 2 weeks.

Case 4

A 3 1/2-year-old boy was brought to the hospital with a week-long history of extensive necrosis and eventual ulceration of the left side of the face. He had suffered from an attack of measles 3 weeks earlier and was being managed at home when the present condition was noticed by the grandmother. He was the fourth of five children from the same mother. The father was married to four wives and had 13 children. The father was a messenger in one of the state ministries.

Maxillofacial examination revealed extensive ulceration, necrosis of the left side of the face involving the lower border of the chin and the supraorbital region, with complete destruction of the left eye, mandible, premaxilla, cheek and half of the nose [Figure 4]. He was admitted and routine investigations were carried out. The haemoglobin was 8 g/dl; white blood cell count and differential count showed elevated lymphocytes, eosinophil and basophil.

He was maintained on high-protein diets, regular debridement with dilute hydrogen peroxide and normal saline and twice daily with EUSOL solution and a weeks' course of ampicilin–cloxacillin and metronidazole; miltivitamin preparation was added twice daily for 2 weeks. The patient healed his ulcer and was discharged home after 3 weeks in the hospital.

Case 5

A 6 1/2-year-old boy was brought into the hospital with a 2-week history of swelling around the mouth, which progressively ulcerated, leaving behind a large defect. He had been ill in the previous week with fever and rashes and was treated at home. He had not fully recovered from the fever and loss of appetite when the grandmother noticed some swelling around the mouth. The swelling later formed blisters and ulcerated, resulting in an extensive defect.

Examination showed circumoral ulceration involving the right as well as the left cheek [Figure 5]. There was complete destruction of the premaxilla and the anterior portion of the palate. Two-thirds of the nose and the lateral third of the lower lip were destroyed. He was admitted and treated for acute NOMA.

A nasogastric tube was passed for feeding with high-protein diets. The ulcer was debrided with dilute hydrogen peroxide and with normal saline regularly until all necrotic tissues were removed. Twice daily EUSOL dressing was commenced for 2 weeks. He had ampicillin–cloxacillin and metronidazole for 1 week and multivitamin preparations for 2 weeks. Wound healing progressed rapidly and healed completely within the 2 weeks of treatment. The patient was discharged for further treatment of the healed ulcer at a later time.

DISCUSSION

NOMA has been known since the time of antiquity,[4-6] and has a worldwide recognition,[4-8] including Nigeria.[9-14] Starting initially as an innocently benign swelling, a boil or rash, NOMA tends to affect both hard and soft tissues around the mouth, except the tongue, leaving its victim aesthetically unacceptable.

The predisposing factors for NOMA include malnutrition, poor oral hygiene and debilitating diseases, especially measles and malaria fever. All the five cases in this report had suffered from measles and/or malaria shortly before the onset of this disease. Measles is the highest known predisposing condition in our environment followed by malaria fever.[4] Other conditions that contribute to the occurrence of NOMA include poor or lack of maternal care, HIV infections and following some courses of cytotoxic drugs used in the management of Burkitt's lymphoma.[4,15] NOMA also predominates in the low socioeconomic groups, as was the case in this study.

Childhood NOMA is common in northern Nigeria as a result of lack of maternal care due to sociocultural practices that enables mothers to leave their babies in the care of their grandmothers as soon as they are weaned to enable them to compete with their mates for their husband. Most of these grandparents lack knowledge of balanced diets, with the result that the babies are usually severely malnourished, as was the cases in this study. Another predisposing factor to childhood NOMA is debilitating disease. Before this study, four children with acute NOMA had presented in our clinic with HIV infection. This necessitated the screening of every child presenting with acute NOMA.

Bacteriology of acute NOMA is cumbersome, especially because of secondary infection. It is a general belief that *Fusiformis fursiformis* and *Borrelia vincenti* are the main causative organisms.[16] Other bacteria associated with acute NOMA are bacteroides and nonhaemolytic Streptococcus.

The reported sex incidence is about equal,[4,15,16] although in this report the male to female ratio is 1:5:1. The occurrence of acute NOMA is seasonal and is favoured by dry season (October to April). This incidentally is the peak period for measles and malaria in Nigeria. We observe that the incidence of acute NOMA is on the decline in our setting, probably because many more parents now embrace routine immunization of their children against the common childhood communicable diseases.

Out protocol for the treatment of acute NOMA involves (i) general investigations, (ii) local management of the ulcer, (iii) drug therapy, (iv) dietary supplementation and (v) treatment of immediate complications.

The usual routine investigations include full-blood count with white blood cell differentials, stool microscopy and cultures, serum protein level and HIV screening. In all cases of NOMA, hypoproteinaemia and anaemia (usually of nutritional origin) are common findings. The stool examination usually reveals worms, especially hook worm.

Effective local management of the ulcer involved regular/repeated debridement and frequent dressing using dilute hydrogen peroxide or EUSOL. All necrotic tissues, including shaky teeth and sequestrum, must be removed.

Drug therapy of choice in our setting is penicillin (plus cloxacillin) and metronidazole. The patients are routinely treated for malaria and dewormed using appropriate antimalaria and antihelmintic drugs to boost their immunity.

The most important aspect of the management protocol is dietary supplement. The patients are fed with a high-protein diet. Multivitamin preparations help boost the nutritional levels. Intravenous fluids are carefully selected to correct dehydration and electrolyte imbalance when indicated.

The immediate complications in acute NOMA include formation of sequestrum and formation of fibrosis leading to ankylosis of the temperomandibular joint with limitation of mouth opening. The sequestrum is allowed to form fully before removal. All areas of fibrosis are completely exercised under local anesthesia.

The orofacial defect is usually very extensive and requires multiple and highly specialized surgical reconstructions. Reconstruction is delayed until the patient is matured enough because the patient's cooperation for effective postoperative management is required. Delayed surgical reconstruction also ensures adequate tissue for reconstruction. It also allows the defect to contract and reduce in size.

Temporomandibular joint ankylosis and fibrosis are released as soon as possible.[12] Masticatory problems and speech defect can be aided with the use of prosthesis.

In conclusion, NOMA is a disease of children who are afflicted with malnutrition, debilitated premorbid condition and poor immune status. The initial management of acute NOMA should be conservative (nonoperative). Apart from ulcer debridement, surgery should be reserved for reconstructive surgery at a later time. Efforts to boost the patient's immune and nutritional status limit further tissue destruction and produce satisfactory wound healing.

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