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Unusual presentation of NOMA: a case report

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Summary

A case of noma with involvement of other parts of the body from extension and spread of cancrum lesion in the oral cavity and primary herpetic stomatitis in a two-year-old male patient is reported. The possible routes of infection to other parts of the body are discussed. It is expected that this case report will stimulate the awareness of health practitioners to this unusual presentation of cancrum oris.

Keywords: *Unusual, cancrum oris, Noma.*

Résumé

Un cas de noma affectant d'autres parties du corps humain de l'extension et la distribution d'une lésion cancreuse dans la cavité orale et la stomatite primaire d'herpe chez un garçon a été rapporté. Les routes possible d'infection d'autres parties du corps humain ont été discutées. Il est expecté que ce cas stimulera l'éveil des médecins a cette manifestation rare de l'oris cancreuse

Introduction

Cancrum oris had been defined by Tourdes (1848) as a gangrenous affection of the mouth that especially attacks children in whom constitution is altered by bad oral hygiene and serious illness especially the eruptive fevers [1]. The common presentation of this disease is an ulcer of the oral mucous membrane with oedema of the face extending from within out and rapid destruction of the soft tissues and bone with an outcome, which is almost always fatal.

Although the definition of Tourdes is still valid, the course of the disease has been altered by modern antibiotic therapy, improved nursing and advances in surgical techniques [2,3]. Cancrum oris is virtually non-existent in developed countries, but in developing nations where the triad of malnutrition, poor oral hygiene and debilitating disease is still common, it persists as a notable health problem [4].

Although the term "Noma" is generally used as a synonym for cancrum oris, however, there is a difference between the two. Noma includes lesions of similar nature, which appear in the prepuce, anus, genitalia and other parts of the body [5,6]. In this communication, we describe an unusual case of a two-year-old Nigerian male with "cancrum lesions" in other parts of the body. The likely path of infection to the other sites of the body is also examined.

Case Report

A 2-year-old male presented at the Oral Surgery clinic with a two-week history of soreness, pain and excessive salivation from the mouth. The patient's mother stated that the patient had a febrile illness a few days before the pain was felt in the mouth. The past medical, dental, family and social histories were non-

contributory except that the patient is the last child in a poor family with eight children.

Extra-oral examination revealed an ill-looking, apathetic, febrile patient with circular dome shaped vesicles on the lower lip. There were circular, sharply defined shallow ulcers with yellowish floors around the angles of the mouth and a perforation on the right cheek with exposure of the bone which drained saliva and fluids (Fig. 1). The right and left submandibular lymph nodes were enlarged and tender. There was also tenderness and redness of the prepuce and the scrotal skin (Fig.2). Intraorally, a full complement of the deciduous dentition was present; there was tenderness and redness of the gingiva with foul odour and a communication between the oral cavity and the skin of the cheek.



Fig. 1: Perforation of the right cheek with exposure of bone which drained saliva and fluids



Fig. 2: Redness of the prepuce and scrotal skin.

Based on the these findings, impressions of primary herpetic stomatitis, acute ulcerative gingivitis, cancrum oris, fourmier's gangrene of scrotum and acute neutropenia were made. On admission, bacterio-logical smears were taken from lesions on the cheek, eye, oral cavity and the scrotal skin and the patient was placed on intravenous fluids, high protein diet via nasogastric tube feeding, multivitamins, iron, ampiclox syrup, flagyl syrup and a strict oral hygiene regimen consisting of warm saline and hydrogen peroxide rinses. The wound on the cheek was dressed

with honey pack and chloramphenicol eye drops were instilled into the left eye. Radiological, haematological and urine investigations and HIV test were also requested.

The smears from the base of the cheek lesion, left eye, oral cavity and the scrotum showed overwhelming members of the fuso-spirochaetal complex (Fig 3). The other investigations were within normal limits and the HIV test was negative. A diagnosis of cancrum oris with involvement of other parts of the body (noma) and primary herpetic stomatitis was made.

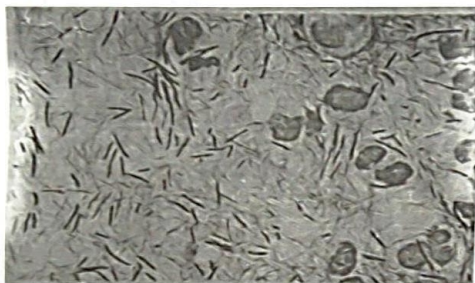


Fig. 3: Gram stained smear showing loose irregular shape of the spirochaetes bacteria among other bacilli and cocci

Recovery was gradual and patient was discharged home after resolution of infection to be reviewed on weekly basis. He did not keep his appointment and was however lost to follow-up.

Discussion

Noma is derived from the Greek verb 'to devour'. It is a rapidly spreading gangrenous affection of the mucocutaneous Orifices such as the lips, nostrils, eyelids and the genitalia [2]. It usually starts on the mucosal surface with the oral cavity being the most frequent site of involvement. Most authors believe that a careful history is important to obtain a clear picture [7]. Although, usually synonymously used with noma, cancrum oris is a gangrenous affection of the mouth and neighbouring structures only.

Since most cases of cancrum oris commence as acute ulcerative gingivitis there may be problems in differentiating it from primary herpetic stomatitis. The two have similar age brackets, oral features and occasional distribution of lesions on genitalia and eyes. Coexistence of acute ulcerative gingivitis and primary herpetic stomatitis had been reported in British children [8], which was also noticed in this patient. The distribution to the eyes and the genitalia sometimes seen in herpetic stomatitis was also observed in this patient, which necessitated the use of bacteriological smear to confirm the diagnosis. However, clinically, presence of dome shaped vesicles, severe systemic upset and widespread distribution of lesions are notable features of herpetic stomatitis.

The occurrence of cancrum oris in atypical sites is rare. Tempest [2] found that 11 (4.4%) in his study of 250 children

showed involvement of distant sites. Of these, 7 children had intra-oral disease in addition to the extra-oral lesions but 4 showed no sign of intra-oral disease. As in the case presented, typical Vincent's organisms were detected from all the extra-oral lesions.

Although the fatal course of noma has been altered by modern antibiotic therapy and improved nursing and paediatric care, malnutrition, poverty and debilitating diseases still common in developing countries account for varying clinical course of this disease in these nations. The disease is typically found in the low socio-economic groups, which the patient belongs.

The spread to the conjunctiva of the left eye might be as a result of self-inoculation of the eye by the child's uncontrolled habit. Hand already placed in the oral or cheek lesion can rub the eye. This could also account for the infection of the genitalia. The lesion on the cheek was as a result of direct contact of oral lesions, and is a good example of contact ulcers. However, extension through the blood stream should not be ruled out especially in cases of distant sites. Intestinal, pulmonary, intracranial and blood stream involvement have been reported but they are uncommon [5]. Lesions on the externalia genitalia can be due to autoinoculation or perverted sexual practices. The later mode of transmission is not likely because of the age of this patient.

The dentist should prevent iatrogenic transmission during treatment of fusospirochaetal stomatitis. Pressure sprays should be carefully used to prevent possibility of conjunctival infections.

Since most cases of noma start as acute necrotizing gingivitis, prevention of noma can be achieved by early and prompt remedial measures once a diagnosis is established.

References

1. Tourdes J: These de Strasbourg. 1848. Cited by Tempest, M.N. *Cancrum Oris*. *Br J Surg*. 1966; 53: 949 - 969.
2. Tempest M.N: *Cancrum Oris*. *Br J Surg*. 1966; 53: 949 - 968.
3. Adekeye E.O. and Ord R.A. *Cancrum oris: Principles of management and reconstructive surgery*. *J. Maxillofac Surg*. 1983; 11: 160 - 170.
4. Enwonwu C.O: Infectious oral necrosis (cancrum oris) in Nigerian children: A review. *Community Dent Oral Epidemiol*. 1985; 13: 190 - 194.
5. Burket L.W: *Oral Medicine-Diagnosis and Treatment*. (Sixth Edition) Philadelphia, J.B. Lippincott Company, 1971, p 43.
6. Kaimenji J.T. and Guthua S.W: Residual facial deformity resulting from Cancrum Oris: A case report. *East Afr. Med. J*. 1994; 71: 476 - 478.
7. Barrios T.J., Aria A.A. and Brahney C: *Cancrum Oris in an HIV - Positive Patient*. *J. Oral Maxillofac Surg*. 1995; 53: 851 - 855.
8. Cawson, R. A: *Essential of Dental Surgery and Pathology*. Third Edition Churchill Livingstone Edinburgh London and New York. 1978, p 85.