Nigerian Journal of Paediatrics 2002; **29 (4)**:119-21

Cancrum Auris in Children: Report of Two Cases

FO Ogisi*

Summary

Ogisi FO. Cancrum Auris in Children: Report of Two Cases. *Nigerian Journal of Paediatrics* **2002; 29:119.** Two cases of necrotising otitis externa (cancrum auris) associated with malnutrition in very young children are presented. *Pseudomonas aeruginosa* and Klebsiella spp were isolated from one of the children while *P. aeruginosa* and coliforms were cultured from swabs of the wound in the second case. Management consisted of wound debridement under local anaesthesia with application of both local and systemic antibiotics. Follow up showed good healing in one case and improvement in the other which was however, complicated by fibrosis and meatal stenosis. Malnutrition was most probably a predisposing factor in both cases.

Introduction

NECROTISING otitis externa, a rapidly progressive infection of the external ear canal, mastoid and base of skull, that was first described by Meltser and Keleman in 1959, is usually due to *Pseudomonas aeruginosa* in immunocompromised patients, especially, elderly diabetics.[1,2] This condition is very rare in young children, and since the first report of paediatric necrotising otitis externa in two non-diabetic infants by Joachim,[3] few cases have been reported. However, the more common necrotising and gangrenous condition affecting oro-facial structures known as cancrum oris or Noma has been widely reported in children mostly in underdeveloped countries[4-6] and is associated with malnutrition and immunosuppressive states.[7,8] A similar tissue-destructive process affecting the ear (cancrum auris) has, to our knowledge, been the subject of few reports in this country. In this communication, we present two cases of progressive necrotising external otitis in malnourished children.

Case Reports

Case 1

A nine-month old infant who had been having frequent stools and feeding difficulty due to "maternal deprivation" was brought to the hospital with a febrile illness diagnosed as septicaemia. While on admission, she was noticed to have a progressive inflammatory and ulcerating condition of the left external ear, with foul-smelling discharge and peri-auriular swelling. On examination, the infant was ill looking and marasmic. Her weight was 5kg. Examination of the left ear revealed purulent discharge and necrotizing ulceration of the external canal and pinna with exposure of the cartilage and marked inflammatory swelling in the peri-auricular area (Fig 1). Necrosis of the canal wall and concha had exposed the underlying bone with granulation tissue on the floor of the external auditory canal. The tympanic membrane was not evident at this stage. There was also a left facial paresis. Radiographs of the mastoids were not informative because of poor quality. Blood culture yielded no growth on two occasions. The haemoglobin level was 9gm/100ml. Urine examination was negative for sugar and random blood sugar level was within normal limits. Serum total protein level was 5.5gm/100ml (normal 6-8gm/100ml) with albumin 3.5gm/100ml (normal 3.5-4.5gm/100ml) and globulin 2gm/100ml (normal 2-3gm/100ml).

Debridement and excision of necrotic tissue and debris was carried out under sedation and local anaesthesia; necrotic soft tissue and cartilage with granulation tissue were excised from the posterior and antero-inferior walls of the outer external canal, down to the bony canal which was intact, and the lesion was irrigated with chlorhexidine. Packing was done with gauze wick impregnated with topical gentamicin. The child was placed on intramuscular ampicillin/cloxacillin and gentamicin; the latter of which was discontinued after two weeks. An ear swab taken initially yielded on

University of Benin Teaching Hospital, Benin City

Department of Surgery

* Associate Professor

Fig 1. The affected ear in Case 1, showing perichondritis, purulent discharge and ulceration of the pinna and outer ear

culture, a mixed growth of *Pseudomonas aeruginosa* and Klebsiella spp; a second specimen a week later grew Pseudomonas spp. only. Dressing and packing were carried out daily with topical gentamicin, while systemic antibiotic therapy was continued. Supplementary multivitamin therapy, high protein and calorie balanced diet and blood transfusion were administered. The infant's general condition gradually improved, with increase in weight from 5 to 5.5 kg in the first week. The aural lesion showed remarkable improvement with reduction in the purulent discharge and gradual healing and epithelialisation of the previously large area of necrosis. The external meatus, earlier occluded by infected granulations, gradually healed and the tympanic membrane was now visible and noticed to be intact. There was however, a moderate degree of stenosis after the healing. Systemic gentamicin was discontinued after two weeks while *Ampiclox* and topical gentamicin packing were continued. After six weeks, the child was discharged home with nutritional advice. Follow-up evaluation showed that the ear had healed almost completely, leaving only a small area of scarring.

Case 2

A two-year old female child was referred to the University of Benin Teaching Hospital with ulceration of the right ear which was noticed one month after a severe attack of measles. On examination, the child was alert but malnourished with angular stomatitis. There was marked oedema and inflammation of the pinna suggestive of perichondritis, and a fairly extensive ulceration of the concha and external meatus. Granulations in the canal wall partially obscured the tympanic membrane which however, appeared intact. A right facial nerve palsy was also present. Radiographs of the mastoids showed poor pneumatisation but no evidence of bone destruction. A full blood count showed haemoglobin level of 11gm/100ml and white cell count of 4,400/cmm. The erythrocyte sedimentation rate (ESR) was raised (78mm/hr Westergreen, normal range 0-10mm/hr). A biopsy taken from the edge of the ulcerated area showed on histology, inflammatory cells and pus cells only, while culture of a swab from the lesion grew *P. aeruginosa* and coliforms. There was no glycosuria.

Wound debridement was carried out with local anaesthetic; all the necrotic tissue and cartilage, as well as granulation tissue were excised and dressing done, initially with hydrogen peroxide and later, with gauze packing soaked in topical gentamicin/hydrocortisone lotion. The pack was changed daily. Systemic antibiotic therapy was instituted with intramuscular cloxacillin and gentamicin, the latter being discontinued after two weeks, while cloxacillin was continued for a further two weeks. The patient was nutritionally rehabilitated with balanced diet and vitamin supplements. The ulceration and inflammation slowly regressed during the next three weeks, while the child's nutritional status improved. The facial paresis improved but the healing process was accompanied by fibrosis and meatal stenosis (Fig 2).

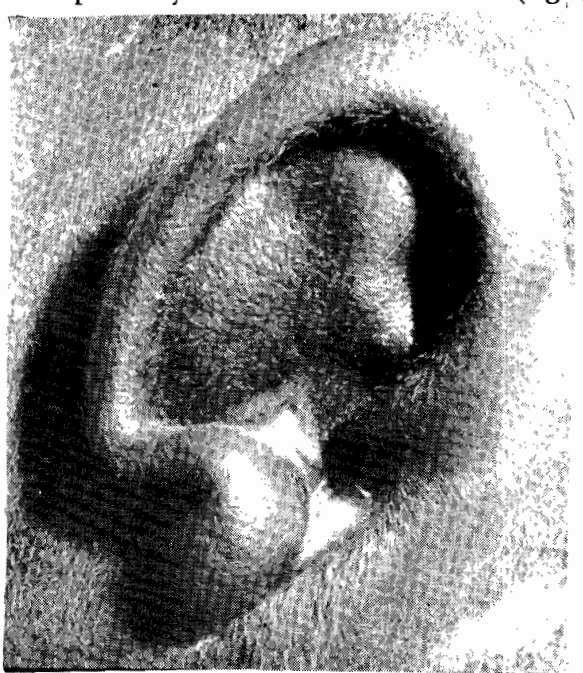

Fig 2. Photograph of Case 2, showing the affected ear after three weeks of treatment; there is healing with meatal stenosis.

despite prolonged packing. The child was discharged home after three weeks stay in hospital. She was subsequently lost to follow-up.

Discussion

The progressive necrotising inflammatory process affecting the external ear (cancrum auris) in these two cases is very similar to the gangrenous process of cancrum oris which affects the oro-facial tissues. A combination of infection and malnutrition predispose to the severe tissue destruction seen in both conditions. Malnutrition acts synergistically with endemic infection in promoting an immunodeficent state,[4,9] and a weakened immune system would appear to be a common denominator in both cancrum oris and cancrum auris. Anergy and depressed immunity promote overgrowth and invasion by an infective consortium consisting of anaerobic and Gram negative organisms such as Pseudomonas spp. as seen in the cases presented. The bacteriology as well as the spread of the inflammatory process through the meatal cartilage to involve the surrounding soft tissue bear some similarity to the necrotising ("malignant") otitis externa; however, this condition has been reported mainly in elderly diabetics.[10] In the two cases reported, there was progressive inflammation and necrosis of the external ear spreading through the cartilage to the surrounding soft tissue, with involvement of the facial nerve. Debridement combined with local and systemic therapy with gentamicin eventually led to satisfactory control and regression of the disease.

It would appear that the general debility and lowered resistance from malnutrition predisposed our patients to this severe infective process. There is controversy about whether or not, the synergism of infection and malnutrition is related to altered immunity. It has been observed that in the presence of protein energy malnutrition (PEM), there is usually a high rate of infection; unusual patterns of infections are also sometimes seen in children with PEM.[11] This could be attributed to impaired immunological status of the malnourished child, with particular reference to humoral and cellular immunity. Smythe[12] reported evidence of depressed cell-mediated immunity (CMI) in a group of children with PEM, although humoral immunity appeared normal. A depressed CMI would explain the high incidence of Gram negative infections in these cases. Other conditions often associated with PEM can cause depressed cell-mediated immunity. Measles, a common infection in PEM, is a potent cause of immuosuppression and may contribute to the serious infections that often follow an attack of measles in malnourished children[4,8] as in case 2. Furthermore, phagocytosis has also been shown to be impaired in the malnourished child.[12] It may be inferred that impaired immunological responses as often encountered in malnutrition are likely to have contributed to the aggressive infection in the two cases presentcd. It is therefore appropriate in the management of such cases to correct the nutritional deficiency.

References

1. Chandler JR. Malignant externa otitis. *Laryngoscope* 1968; **78**: 1257-94.
2. John AC, Hopkin NB. An unusual case of necrotising otitis externa. *Journal of Laryng & Otolaryngol* 1978; **92**: 259-64.
3. Joachims HZ. Malignant otitis in children. *Arch Otolaryngol* 1978; **102**: 236-7.
4. Enwonwu CO. Infectious oral necrosis (cancrum oris) in Nigerian children: a review. *Community Dent Oral Epidemiol* 1985; **13**: 190-4.
5. Smith I. Cancrum oris. *J Maxillofac Surg* 1979; **7**: 293-8.
6. Nash ES, Cheng LH, Smart K. Cancrum oris-like lesions. *Br J Oral Maxillofac Surg* 1992; **30**: 202.
7. Valadas G, Leal MJ. Cancrum oris (noma) in children. *Eur J Pediatr Surg* 1998; **8**: 47-51.
8. Lazarus D, Hudson DA. Cancrum oris - a 35-year retrospective study. *S Afr Med J* 1997; **87**: 1379-82.
9. Enwonwu CO, Falkler WA Jr, Idigbe EO, Savage KO. Noma (cancrum oris): questions and answers. *Oral Dis* 1999; **5**: 144-9.
10. Zaky DA, Bentley DW, Lowy K, Betts RF, Douglas GR. Malignant external otitis: A severe form of otitis in diabetic patients. *Am J Med* 1976; **61**: 298-302.
11. McLaren DS, Burtman D, eds. Textbook of Paediatric Nutrition. London: Churchill Livingstone (Publishers) Ltd, 1976: 91-145.
12. Smyth E. Thymolymphatic deficiency and depression of cell mediated immunity in protein calorie malnutrition. *Lancet* 1971; **ii**: 939-40.