Primary cutaneous nocardiosis: case report and review

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Abstract

Background: Infection caused by *Nocardia* species are uncommon and primary cutaneous nocardiosis is relatively rare. The lymphocutaneous pattern of the disease is the rarest type and commonly occurs in otherwise healthy individuals[1]. Clinically it can present as superficial acute skin and soft tissue infection (abcess or cellulitis), a lymphocutaneous infection and a deeper infection, mycetoma[9]. Mycetoma is commoner than other two variants. Here we report an interesting case of primary cutaneous nocardiosis (PCN) in posterior auricular, ear pinna and cervical region, progressing to lymphocutaneus nocardiosis which is a rare site.

Keywords: Nocardiosis, Children, Pedal edema, Bikaner

Case history: The child presented with bilateral pedal edema, loose motions, vomiting and a pustule behind right ear pinna and redness of inner aspect of pinna. The patient was diagnosed as PCN caused by Nocardia species grown on pus culture. Patient was a severe acute malnourished child and approximately treated according to sensitivity of antibiotics and drainage of abscess done.

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Introduction

Cutaneous nocardiosis presents either as a part of disseminated infection or as a primary infection resulting from inoculation. Primary cutaneous nocardiosis is relatively rare[2]. Disseminated nocardiosis is most commonly caused by N. asteroides and typically affects immunocompromised hosts. Nocardia brasiliensis is the main pathogenic organism for primary cutaneous infection, followed by N. asteroides[10]. Isolation of nocardia from clinical specimens and species identification is difficult and need expertise of microbiologist. Nocardiosis has been reported worldwide. Around 500-1000 cases of nocardiosis are recognized in US each year[12]. Nocadia infection is supposed to be uncommon in India, though it is reported from various parts of India like Himachal Pradesh in north[6], Mumbai in western part[7] and Karnataka[8] in southern part of India. Baxi and Mathur in their study of 73 cases of mycetoma from Rajasthan did not report a single case due to Nocardia[5]. Nocardia are aerobic, gram positive, partially acid fast filamentous bacteria found as saprophytic organism in soil and decaying plant parts. Human infection can occur either by inhalation of dust contaminated with nocardia which causes infection or by deep implantation due to thorn prick causing subcutaneous infection (mycetoma).Pulmonary infection is usually seen in immunocompromised whereas cutaneous infection is usually seen in immunocopetent persons[11]. From lung the infection can get disseminated to other organs causing systemic infection including skin. This paper presents a clinical case of this uncommon disease emphasizing nocardia as a causative agent in abscess of skin in posterior auricular and cervical region in children. Available literature showed that none of previous studies had reported PCN of posterior auricular and cervical region in children in North India.

Case Report

A one year old female child presented to us with complaints of bilateral oedema feet since last one month, fever for 6 days, loose motions and vomiting for 5 days, a pustule behind right ear pinna and redness of inner aspect of pinna, after 2 days of hospital admission. Child was full term and normal delivered in hospital, cried soon after birth, given goat milk for 3 days, breast feeding started late, mile stones were normal, mother had no antenatal complications. On physical examination revealed Child was irritable, pallor, febrile, pulse rate (130/minute), respiratory rate (32/minute), local examination revealed ill-defined tender, soft abscess with reddish pink colour overlying skin in right posterior auricular region and erythema and ulceration at inner side of right pinna, right posterior cervical lymph nodes were enlarged, tender and firm to hard in consistency. Anthropometry showed, length (67cm), weight (4.8kg), Z score (-4 SD), mid upper arm circumference (9.5 cm), she was a severely acute malnourished child, taken immunization appropriate for age. Hepatomegaly was present, other

systemic examination was normal. ENT surgeon reference was done and diagnosed as abscess with otitis externa, aspiration of abscess done, pus sent for culture and sensitivity, Fusidic acid ointment advised for local application. Patient had following laboratory parameters: Hb 5.6gm%, TLC-6900/ cumm, lymphocytosis (77%), platelets count 35000/cumm, reticulocytes (01%), ESR (70), blood sugar, urea and serum creatinine were normal. Liver function tests were normal except serum albumin (3.1gm%), serum electrolytes were normal, X ray chest normal, Mantaux test negative and HIV test was non-reactive. Aspiration of abcess yielded pus, which was sent for culture and sensitivity to microbiology department, which reported Nocardia species organism grown on culture, showing sensitivity to amikacin, ceftriaxone, cotrimoxa, levofloxacin, linezolid and intermediated sensitivity to vancomycin antibiotics. *Surgery:* Incision and drainage of abscess done by pediatric surgeon, pus drained out and dressing with povidone iodine gauze done, advised to review for dressing; tissue for histopathological examination could not be taken. Patient responded well to treatment with intravenous ceftriaxone and amikacin given for 2 weeks and oral co-trimoxazloe and linezolid antibiotics. Patient was discharged on oral antibiotics and advised to review after every 2 weeks.

Discussion

The genus Nocardia belongs to the order Actinomycetales, a group of aerobic, Gram positive, filamentous bacteria[10]. The first description of Nocardia, in 1888 by Edmond Nocard, a French veterinarian and human nocardiosis was described by Eppinger (1890)[10] as systemic infection. In a review of cases of pediatrics, nocardia asteroides infection between 1895 and 1981, 8% children had cutaneous infection.[3] About 70 cases of primary lymphocutaneous nocardiosis have been described in literature[13]. Most of reported cases were sporotrochoid forms on the limbs in otherwise healthy patients caused by N. brasiliensis (most common), N. asteroides (less common and true opportunistic agent) is exceptionally the causative agent in immunocompetent patients[1]. One of them was HIV infected.

In the pediatric cases reported, cervicofacial nocardiosis was caused by N. brasiliensis and only in two cases by, respectively, N. asteroides or N. caviae[3]. Cat scratches on the face and the children's playing habits were reported to have caused minor traumas and accidental contamination by soil. The usual sites of involvement are hands, feet, back, shoulder and scalp during agricultural work[15]. A Japanese fish handler suffering from primary cutaneous nocardiosis, without preceding injury, has been reported[1]. Pediatric cases can occur as early as 3 years of age[3]. In the case described here the age of child is one year, which may be the earliest age reported. Cutaneous nocardiosis presents as a primary cutaneous infection or as a secondary disease. The clinical presentations described are mycetoma, lymphocutaneous, superficial acute and secondary cutaneous infection with disseminated disease [14]. Mycetoma is the most common mode of presentation. Among several species of Nocardia causing cutaneous infection N. brasiliensis is the most common species isolated[(14)] and infection caused by it is more inflammatory, locally invasive and progressive rather than the self-limited course of lesion caused by N. asteroides[10]. Cervicofacial nocardiosis is a distinct subgroup of lymphocutaneous nocardiosis and only sporadic cases have been described in the literature[3]. It is considered a typical infection in children who develop a pustule in the nasolabial area and successively complain of regional submandibular or cervical lymphadenitis, fever and systemic symptoms. The suppurative evolution of lymphadenopathy may require surgical drainage. Clinically lymphocutaneous pattern of the disease simulates sporotrichosis but differs from this by its acute onset, erythema of the overlying skin, tenderness and a highly inflammatory course. Rarely, granules may be observed in the discharge from noduloulcerative lesions[4]. Superficial acute skin infections simulate those caused by common pyogenic organisms, occurring as a pustule, abscess, bulla, cellulitis or as a chronically draining ulcer. Unlike mycetoma, there is no granule formation[14]. The lesions are rapidly progressive with intense pain, erythema and oedema. One third of these cases may be transformed to lymphocutaneous disease[14].

The initial clinical diagnosis may be difficult due to the non-specific clinical picture. Demonstration of the organism from clinical specimens like granules, pus or aspirated fluid from nodule by Gram stain and modified kinyoun stain is the mainstay of diagnosis. Gram positive and acid fast, thin, beaded branching filaments are the characteristic appearance of the organism. Identification of the Nocardia species by culture is a tedious process. The organism is slow growing and may take up to 2-3 weeks for isolation from a clinical specimen[10]. Species identification can complimented by Western blot assay, DNA probing, ELISA (sero diagnosis)[16].

In our study, culture of pus aspirated from the abscess showed Nocardia species organism grown, we found the isolate to be sensitive to Amikacin, ceftriaxone, cotrimoxa, livofloxacin, linezolid and intermediate sensitive to vancomycin.The present case is unique as the primary site of infection is right ear pinna, posterior auricular and cervical region, which is an unusual site. This would have been commonly misdiagnosed clinically as a tuberculous lymphadenitis.

Patients with primary cutaneous nocardiosis respond very well to medical therapy. Surgical drainage of an abscess is the treatment of choice. Cotrimoxazole (TMP-SMZ) is the main stay of therapy. Other most active parenteral drugs are amikacin (95%), Imipenam (88%), ceftriaxone (82%) and cefotaxime (82%).

Linezolid is an effective alternate, orally[17]. Combination therapy with a carbapenem or a 3rd generation cephalosporin with or without amikacin is usually recommended for severely ill patients or CNS involvement. The mortality rate is 50% with cotrimoxazole alone. Superficial cutaneous infection is treated for at least one to three month, mycetoma, pulmonary or systemic nocardiosis for 6 months, and CNS infection for 12 months. Parenteral therapy can often be changed to oral therapy, such as high doses of cotrimoxazole, after 3-6 weeks and good clinical response. If HIV infection, treat indefinitely. The overall mortality rate is 75%, may be due to delay in diagnosis or debilitation of patients.

Primary cutaneous nocardiosis remains a diagnostic challenge. The majority of acute nocardial abscesses, lymphocutaneous infections, go unsuspected and undiagnosed because of nonspecific clinical picture and difficulties involved in isolation of the organism. A high degree of clinical suspicion is needed for the diagnosis of the condition along with stringent efforts of the microbiologist to isolate the organism.

Conclusion

Nocardia infection should be considered in the differential diagnosis of suppurative and granulomatous dermatitis presenting as nodule, abscess, and ulcer in the cervical region apart from tuberculosis.

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