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Case Report

Pediatric cervicofacial actinomycosis disclosing an underlying congenital dermoid cyst

Santwana Verma[1], Ghanshyam Kumar Verma[2], Vinay Shanker[2], Anil Kanga[1], Gagandeep Singh[4], Neelam Gupta[3], Geeta Ram Tegta[2], Jatin Sharma[2], Aarti Garg[1]

[1]Departments of Microbiology, [2]Dermatology, [3]Pathology, Indira Gandhi Medical College, Shimla, [4]Department of Medical Microbiology, PGIMER, Chandigarh, India

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Address for correspondence:
Dr. Santwana Verma,
Department of Microbiology,
IGMC, Shimla - 171 001, India.
E-mail: drsantwana@gmail.com

ABSTRACT

Pediatric cervicofacial actinomycosis is a rare occurrence consequent to dental infections and manipulations or maxillofacial trauma. The clinical presentation ranges from multiple draining sinuses to swellings resembling tumors and cysts. The present unusual case had congenital dermoid cyst of mid upper lip with *Actinomyces israelii* infection identified on microscopy, culture, and histopathology. A successful outcome in the present case was obtained using combination of medical and surgical treatment.

Key Words: Actinomycosis, cervicofacial, dermoid cyst, pediatric

INTRODUCTION

Actinomycosis is a rare, chronic, suppurative, endogenous infection presenting with multiple draining sinuses. The commensel anaerobes of *Actinomyces spp*. become pathogenic subsequent to breach in mucosa. Dental infections or extraction and maxillofacial trauma pre-dispose to cervicofacial actinomycosis.[1-4] Facial swelling without discharging sinuses, resembling desmoid tumor, cysts involving salivary glands or bone are rare entities.[5,6] We report a rare case of cervicofacial actinomycosis in a female child with multiple discharging sinuses of upper lip mucosa. Microscopy of pus revealed grains and culture grew *Actinomyces israelii*. Tissue histopathology revealed chronically inflamed dermoid cyst. The child was managed successfully on amoxicillin/clavulanic acid and surgical excision.

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CASE REPORT

A 6-year-old female child presented with an upper lip swelling [Figure 1a] discharging pus from the oral mucosa underlying central part of lip with mild pain exacerbating while eating. The swelling progressed from few millimeters to about 2 cm × 2 cm over 1 year, started discharging thick, pus with white granules. No antecedent external trauma or dental ailments and procedures or self-manipulation were reported. Lesion resolved partially following administration of antibiotics at peripheral health facility. Relapsing symptoms prompted specialist consultation.

The child was a term baby, delivered vaginally without instrumentation during labor. The milestones were average, but Bacillus Calmette Guerin (BCG) vaccine was not administered. The past medical and family histories were non-contributory and other siblings were in good health.

Examination revealed a soft, fluctuant, erythematous, cystic midline upper labial swelling, 2 cm × 2 cm with three openings on the mucosal surface discharging, granular white pus, and a single central sinus opening on the overlying skin discharging serous grey pus [Figure 1b]. Physical and systemic examinations were normal, dental hygiene good, and no congenital

anomalies apparent. Vitals were normal and X-ray of maxilla ruled out bone involvement. The possibilities of actinomycosis, infected congenital upper lip commissure and tubercular pathology were considered. Mantoux test was negative. The wet mount preparation of pus sample demonstrated granules and gram stained smears showed gram positive cocco-bacilli and polymorphonuclear cells. Aerobic culture of pus on blood agar and Mac Conkey agar were sterile after 24 h of incubation at 37°C. Culture on brain heart infusion blood agar in candle jar grew pale cream, gritty, colonies after 1 week incubation at 37°C [Figure 2a]. Smear from culture showed characteristic branching filamentous gram positive beaded rods, which were non-acid fast on Ziehl Neelsen and modified ZN staining. Growth characters, microscopic morphology and standard biochemical reactions revealed *A. israelii*.[7]

Therapy was started with amoxicillin/clavulanic acid and surgical excision done with continuation of antibiotics post-operatively for 4 weeks. We achieved success with a combination treatment. Tissue histopathology revealed a cyst measuring 1.2 cm × 0.6 cm × 0.5 cm grossly, with keratinizing stratified squamous epithelial lining microscopically. Sub-epithelia showed fibro-collagenous tissue, dilated congested blood vessels, hair follicles, skeletal muscle, and sebaceous glands entrapped in the muscle layer with chronic inflammatory cell infiltrate [Figure 2b]. Characteristic microcolonies of *Actinomyces* were not seen. These features suggested an inflamed dermoid cyst.

DISCUSSION

Actinomycosis is seen once annually in major medical centers with a male to female ratio of 3:1. The ambit of clinical disease encompasses cervicofacial actinomycosis, abdominopelvic, thoracic and cerebral involvement. The disease is uncommon in children and cervicofacial actinomycosis is rarer still.[1,8,9]

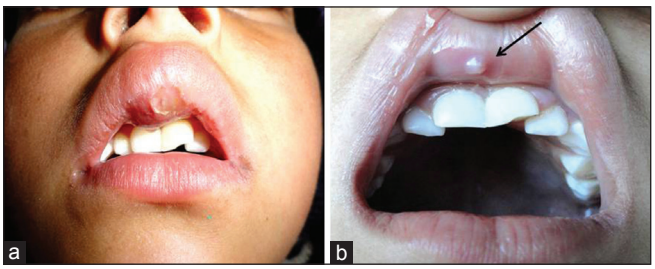

Figure 1: (a) Midline swelling of upper lip with sinus openings. (b) Labial mucosa showing granular pus discharging sinus (arrow)

The causative bacteria, *Actinomyces spp*. constitute normal endogenous flora in oropharynx, gastrointestinal and female genital tracts and are non-virulent.[10,11] A breach in mucosa provides a portal of entry.[1,11] The antecedent factors for actinomycosis include poor dental hygiene and caries, extractions or other traumatizing procedures, perforating injuries, human bites and compound fractures.[2,8,11] This marks the onset of a chronic, suppurative, granulomatous, diffuse swelling with multiple discharging sinuses.[1,2,4]

This child with good oral and dental hygiene and absence of previous trauma had a midline upper lip congenital defect diagnosed as dermoid cyst on histopathology, which came to light due to an actinomycotic infection. Actinomycosis is an enigmatic, masquerading infection suspected in 10% of clinical situations.[12] Diagnosis in typical cases usually relies on the clinical picture. The atypical cases reported in medical literature emphasize the role of laboratory work up. Neck swellings due to tonsillar mass, isolated mid-cervical tumor like granuloma without discharging sinuses and ruptured cyst or desmoid tumor-like facial actinomycosis are reported in adults. Cervical mass, painless, non-tender, non-inflammatory thyroid or expansile mandibular masses, salivary gland, and mandibular bone involvement are documented during childhood.[1,2,5,6,13-15] An unusual pediatric case of facial actinomycosis with intracranial lesion symptomatic with headache, cranial nerve dysfunction, and ataxia is also reported.[11]

Actinomycosis may mimic osteomyelitis, granulomatous disease or neoplasia and those with nodules and fistula resemble tuberculosis, sporotrichosis and nocardiosis.[8,14] We rejected tuberculosis and

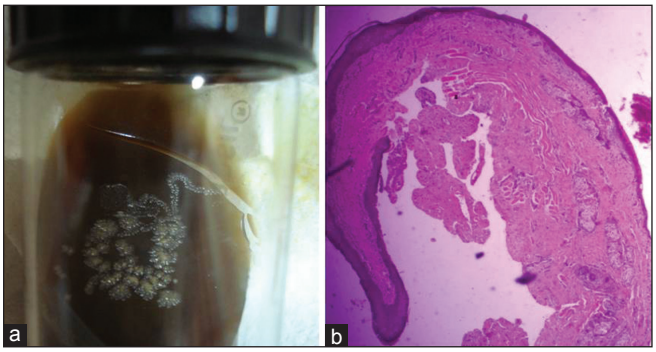

Figure 2: (a) Brain heart infusion blood agar slant showing straw colored colonies of *Actinomyces israelii*. (b) Photomicrograph of dermoid cyst showing keratinized stratified squamous epithelium with sebaceous glands, hair follicles, and skeletal muscle in sub-epithelial tissue (H and E, ×40)

nocardiosis due to a non-indurated Mantoux test and absence of acid-fast bacilli in pus and mycobacterial culture. There was no growth in fungal cultures.

In the present case, visualization of granules macroscopically and microscopically substantiated by culture of *A. israelii* clinched the diagnosis. Gram stain of crushed granules is more sensitive than culture. Though difficult, yet culture is by far the most accurate method of diagnosis and requires repeated attempts.[2,14] The pathologist opined a congenital dermoid cyst, which is characterized by sac like birth defects containing hair, fluid, teeth, and skin glands although the specific identification of *Actinomyces spp.* remained crypt on histopathology. Clinically, dermoid cysts present as non-progressive midfacial swellings without inflammation. On the contrary, inflammatory swellings progress rapidly.[16] In our case, progression over a year, positive microscopy and culture, and the presence of inflammatory cells were evidence of an infective process in a congenital cyst. The child was managed successfully on combined medical-surgical approach. Such adjunctive is a proven therapeutic modality.[9]

CONCLUSION

This case is a rare case report, which highlights the occurrence of actinomycosis complicating an undiagnosed congenital defect. It also emphasizes the importance of collaborative efforts of clinicians in suspecting and laboratory physicians in confirming atypical cases.

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