

Case of Ludwig's angina due to *Corynebacterium diphtheriae* from western Rajasthan, India-A case report of an uncommon presentation

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ABSTRACT

Corynebacterium diphtheriae is a toxin producing, classically noninvasive bacteria that causes diphtheria a vaccine-preventable disease mainly in children. With increasing vaccine cover, new spectrum of infections is increasingly seen involving invasive infections and nontoxigenic strains of *C. diphtheriae*. Here, we present a case of Ludwig's angina caused by *C. diphtheriae* in a 45-year-old female. Only *Corynebacterium* spp. have been previously reported in Ludwig's angina patients.

Keywords: *Corynebacterium*, *Corynebacterium diphtheriae*, Diphtheria, Ludwig's angina, submandibular abscess

Introduction

Corynebacterium diphtheriae, a gram-positive, rod-shaped bacterium is the causative organism of diphtheria, a vaccine-preventable disease.^[1] The bacterium primarily infects the upper airway and produces a toxin that is responsible for the systemic manifestations of the disease.^[1] Although classically this bacterium is known to be noninvasive, recently *C. diphtheriae* has been reported to cause other forms of invasive infections.^[2] Here we report a case of Ludwig's angina caused by *C. diphtheriae* in a patient from western Rajasthan, India.

Case Report

A 45-year-old lady presented to the hospital with a complaint of pain in the right lower jaw 7 days, associated with a rapidly

progressive, painful right-sided neck and facial swelling for the past 2 days. There was a history of decreased mouth opening with drooling of saliva. There was no history of change in voice, trauma, dental procedure, or toothache.

On clinical examination, the patient had a hard swelling on the floor of the mouth and bilateral submandibular regions with increased local warmth and tenderness. The overlying skin was erythematous without scarring or sinus formation. There were edema and crepitus in the right zygomatic, infrazygomatic, and infraorbital region. Mouth opening was reduced to half a finger width (Grade III trismus). The patient had poor oro-dental hygiene, pus was oozing out from the right lower third molar region with the presence of gingival edema. The posterior pharyngeal wall was not visualized because of decreased mouth opening.

On laboratory investigations, the patient was anemic (Hb 8 gm/dL) and total leucocyte count (TLC) was raised (17100/mm³). On ultrasonography, the right submandibular gland appeared bulky and heterochoic, measuring 2.5 × 1.9 cm with raised internal

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vascularity and a multiloculated collection, 2.8 × 1.4 cm was evident. Multiple discrete lymph nodes were found in the right IIB level with the largest measuring 9 × 4 mm. A contrast-enhanced computed tomography (CECT) was performed on the face and neck region. It showed evidence of multiple, small foci of collection and edema in the right submandibular gland region, floor of mouth, right submandibular space, and right masticator space with superior collection extending till right temporalis and infratemporal fossa.

A diagnosis of Ludwig's angina was established and approximately 5 ml pus was drained and sent to the microbiology laboratory for aerobic culture. The specimen, upon receipt in the microbiology laboratory, was inoculated on routine media used for aerobic bacterial culture i.e. 5% blood agar and chocolate agar. All culture media were incubated at 37°C under aerobic conditions. Gram stain was performed directly from the specimen that showed the presence of numerous pus cells per oil immersion field along with gram-positive bacilli. Culture on blood agar and chocolate agar plates showed bacterial growth after overnight incubation. The colonies were greyish in color, 1–2 mm in size, and did not exhibit any hemolysis on blood agar.

Bacterial morphology was seen on gram-stained smears prepared from these culture plates and it showed gram-positive bacilli. On performing an Albert stain, metachromatic granules [Figure 1] with cuneiform arrangement were seen. Subcultures were performed on Tinsdale medium [Figure 1] and tellurite blood agar. Black-colored colonies were observed on both these media. Further identification was done using conventional biochemical tests such as sugar fermentation using 1% carbohydrates in 25% serum medium, also known as Hiss serum sugars. The sugars tested were glucose, maltose, sucrose, trehalose, and starch. The isolate showed no urease activity and was identified as *C. diphtheriae* based on the biochemical reactions. Antimicrobial susceptibility testing was performed using disc diffusion testing as per the European Committee on Antimicrobial Susceptibility Testing (EUCAST) recommendations.

The patient was managed with both medical as well as surgical intervention. An emergency tracheostomy had to be performed

on the patient on the next day of admission. Incision and drainage of the collection of pus were done and a betadine soaked ribbon gauze packing was done. The patient was treated with intravenous (IV) antibiotics that included IV clindamycin and IV amoxicillin + clavulanic acid combination. Besides antimicrobials, the patient was prescribed IV analgesics and IV fluids as the patient was unable to take orally. The patient improved rapidly and tracheostomy tube was decannulated after 12 days. The patient was discharged from the hospital after a satisfactory recovery.

Discussion

This case shows the ability of *C. diphtheriae* to cause unusual clinical presentations in the patients. With increasing coverage using the diphtheria toxoid vaccine, the number of diphtheria cases went down significantly,^[3] but this success was followed by a new spectrum of *C. diphtheriae* infections.

A number of infections with non-toxigenic strains of *C. diphtheriae* were reported from many parts of the world, especially European countries.^[3] These nontoxigenic strains are serious pathogens as they are associated with high morbidity and mortality. These strains also have the ability to cause both localized and invasive systemic infections^[3] in patients such as infective endocarditis, bacteremia, septicaemia, osteomyelitis, septic arthritis, and splenic abscess.^[4] Cases of penicillin-resistant isolates have been reported in recent times.^[5] Apart from *C. diphtheriae*, other *Corynebacterium* species have been reported to cause both localized as well as invasive/systemic infections in humans.^[3,6]

Among cases of Ludwig's angina and localized abscesses, *Corynebacterium* spp. have been reported in up to 13.3% and 14.3% of cases in a study by Chettiar^[7] while a few other studies have also documented such association between *Corynebacterium* spp. and odontogenic abscesses.^[8-10] A limitation of our study is that toxigenicity testing could not be performed on the isolate.

The universal immunization program of India is pressing for sustained, high vaccination coverage against *C. diphtheriae* in an attempt to eliminate the disease. However, the invasive and new unusual cases are being reported so clinicians should be vigilant for early diagnosis and treatment of such cases, especially, at primary care level to prevent the case getting complicated as seen in this report.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

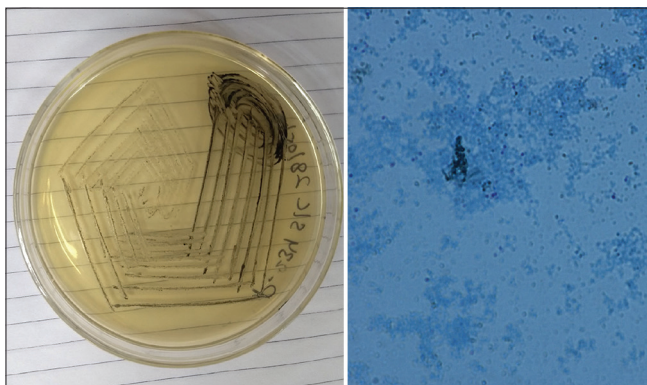


Figure 1: Black-colored colonies on Tinsdale Agar (Left) and metachromatic granules are seen in smear stained with Albert stain (Right)

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Conflicts of interest

There are no conflicts of interest.

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