Case Report

Unmasking the uncommon: Two case reports of emerging *Streptococci*

Sunayana Mukesh Jangla[1], Uma Pankaj Chaturvedi[2], Shifa Firoz Khan[3]

From [1]Consultant Clinical Microbiologist, [2]Pathologist and Head, [3]Pathologist, Department of Pathology, Bhabha Atomic Research Centre Hospital, Mumbai, Maharashtra, India

ABSTRACT

Streptococci, especially a few rare species, are evolving into true human pathogens beyond their traditional clinical spectrum, causing serious human infections. These were once popular as animal pathogens and known to be part of the normal flora of skin and mucosa in humans. We report two cases, one of an infected pilonidal sinus in a young healthy male and another of right leg cellulitis in an elderly diabetic male, both caused by Gram-positive cocci. Former was identified as *Streptococcus pluranimalium* and the latter as *Streptococcus dysgalactiae* ssp *equisimilis*. Both were treated as per the antibiotic susceptibility report. These cases highlight the transforming epidemiology of *Streptococcal* infections, their potential for affecting unusual sites, ability to cause severe complications and the need for heightened microbiological and clinical vigilance. In addition, it emphasizes the importance of advanced diagnostic systems in detecting such unusual organisms.

Key words: *Dysgalactiae*, Emerging, *Equisimilis, Pluranimalium, Streptococcus*

Genus *Streptococcus* is a diverse group of Gram-positive bacteria having similar spherical microscopic morphology and is mainly classified based on their cell wall surface antigens. It comprises various representatives that are part of the normal flora of mucosal membranes of the mouth, upper respiratory tract and lower genital tract, and causative agents of many purulent infections varying in severity [1]. Based on Lancefield's classification, they are classified as Group A, which consists of *Streptococcus pyogenes*, Group B *Streptococcus agalactiae*, Group C or G *Streptococcus dysgalactiae* ssp *equisimilis* (SDSE) and Group D Enterococci. There are some, like *S. pneumoniae* and the Viridans group that are non-typeable [2,3]. *Streptococcus pneumoniae, S. pyogenes,* and *S. agalactiae* are established pathogens responsible for causing morbidity and mortality worldwide [1]. *Streptococcus pluranimalium* is an atypical member of this genus known to cause solely animal infections, and is recently being isolated from humans causing either primary or secondary infections like sinusitis, endocarditis, and brain abscesses [4]. However, cases of *S. pluranimalium* causing pilonidal abscess are few and far between. *S. dysgalactiae* ssp. *equisimilis*, a normal human commensal, is increasingly being recognised as an opportunistic pathogen causing human infections ranging from mild infections to invasive diseases [5].

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This case series is about the changing spectrum of these two budding species of *Streptococci* and their emergence as infectious entities in humans.

CASE SERIES

Case 1

An 18-year-old male presented with swelling and discharge from the crease of the buttock since last 2 years. The swelling was aggravated over the last 6 months. There was no history of trauma, fall, fever, pain in the abdomen, nausea, vomiting, constipation or similar complaints in the past. He did not have a history of contact with animals or pets and has no comorbidities. On examination, there was swelling and discharge over the natal cleft with erythema and edema of the surrounding skin. His blood investigations were normal. Magnetic resonance imaging of the perineum showed a blind ending tract opening externally at the skin of the gluteal region and internally going towards the gluteal cleft. The dimensions of the tract were 4.7 × 1.1 cm. There was no communication with the bowel loop. It was diagnosed as a pilonidal sinus abscess.

Incision and drainage were done, pus was drained, and the tissue was excised. Pus was sent for culture and antibiotic sensitivity, and the tissue for culture and histopathological examination. Gram stain from both

Correspondence to: Dr. Sunayana. Jangla, Consultant Clinical Microbiologist, Department of Pathology, Bhabha Atomic Research Centre Hospital, Mumbai - 400-094, Maharashtra, India. E-mail: sunayanajangla79@gmail.com

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showed plenty of pus cells and Gram-positive cocci in pairs and short chains (Fig. 1a). Pus and tissue were inoculated on 5% sheep blood agar (SBA), MacConkey agar (MA) and Chocolate agar and incubated for 24 h at 37°C in a CO_2 incubator. Both samples showed growth of small grey beta-hemolytic colonies on SBA (Fig. 1b) and no growth on MA. Gram stain from the growth showed Gram-positive cocci in pairs. The organism was identified as *S. pluranimalium* using GPID and ST03 cards on the Vitek 2 compact system from both samples. Antibiotic susceptibility was done using the Kirby-Bauer disc diffusion method, which showed the isolate to be resistant to erythromycin and susceptible to amoxycillin-clavulanate, ampicillin, cloxacillin, clindamycin, cefazolin, cefuroxime, ceftriaxone, and ciprofloxacin. Histopathological examination revealed a sinus tract lined by infected granulation tissue and squamous epithelium (Fig. 1c). The patient was started on oral amoxycillin-clavulanic acid 625 mg once a day for 7 days. His wound healed as seen on regular follow-up, and there was no recurrence at the end of 3 months.

Case 2

A 74-year-old male presented with redness and pain in the right lower limb and fever since one day. There was no obvious discharge, history of trauma, recent travel, nausea, vomiting, or similar complaints in the past. He is a known case of diabetes mellitus and hypertension on treatment. On examination, he was afebrile. The right lower limb was edematous, erythematous, tender, and pulsations were not palpable. He was advised hospitalisation. His C-reactive protein was 152 mg/dL and total white blood cell count was 13,000. Other investigations were normal. Venous color Doppler showed diffuse subcutaneous edema in the right leg. It was diagnosed as right lower limb cellulitis. Three small knicks were given at different sites on the lower limb, and the exudate along with tissue was sent for culture and antibiotic sensitivity. Gram stain from both samples showed plenty of pus cells and Gram-positive cocci in pairs and short chains (Fig. 1a). Pus and tissue were inoculated on 5% SBA, MA and Chocolate agar and incubated at 24 h at 37°C in CO_2 incubator. There was growth of pinpoint beta-hemolytic colonies on SBA (Fig. 2) and no growth on MA. Gram stain from the growth showed Gram-positive cocci in pairs. The organism was identified as *S. dysgalactiae* ssp. *equisimilis* using GPID and ST03 cards on Vitek 2 compact system from both samples. Antibiotic susceptibility was done using the Kirby-Bauer disc diffusion method, which showed the isolate to be resistant to erythromycin and susceptible to amoxycillin-clavulanate, ampicillin, cloxacillin, clindamycin, cefazolin, cefuroxime, ceftriaxone, and ciprofloxacin.

The patient was started empirically on intravenous vancomycin and later shifted to oral amoxycillin-clavulanate 625 mg once a day for 7 days. In addition, limb care was also explained to the patient. His wound healed as seen on regular follow-up, and there was no recurrence at the end of 3 months.

DISCUSSION

S. pluranimalium, a unique member of this genus, is extremely rarely isolated from humans and causes primary and secondary infections in patients with sinusitis, endocarditis, and brain abscesses [4,6]. *S. pluranimalium* has animal reservoirs in the form of infected milk and other animal secretions [7]. It is known to infect a wide variety of animal hosts and causes mastitis in milk cows, septicemia and endocarditis in chickens, and vaginitis and abortion in cattle [4,8]. Transmission to humans takes place when consuming thermally unprocessed dairy products obtained from infected cows or as a result of direct contact while handling animals. Hence, it is a known causative agent of zoonosis. As it possesses certain virulence factors, this microorganism is capable of causing pathological processes in the body of animals and humans. It can cause infections not only in immunosuppressed but also in healthy individuals [4,9]. Our patient was a young adult male without comorbidities. Although he did not give an obvious history of contact with animals, he could have possibly gotten an infection by infected milk consumption or unnoticed animal contact. It produces small grey colonies, generally alpha or occasionally beta hemolytic colonies on blood agar, and on Gram stain

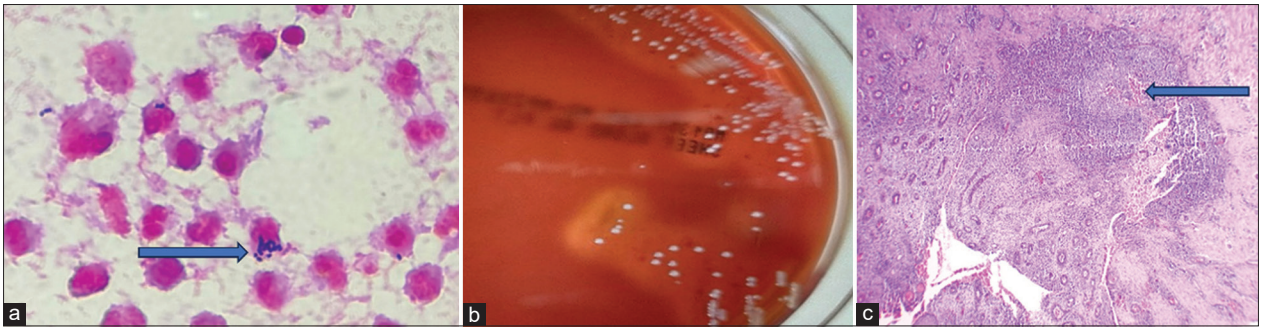


Figure 1: (a) Gram stain from samples of both cases showing pus cells and Gram-positive cocci in pairs and short chains (×100); (b) pin point beta-hemolytic colonies *Streptococcus pluranimalium* on 5% sheep blood agar; (c) Histopathological examination of tissue of pilonidal sinus showing sinus tract lined by infected granulation tissue and squamous epithelium (×40)

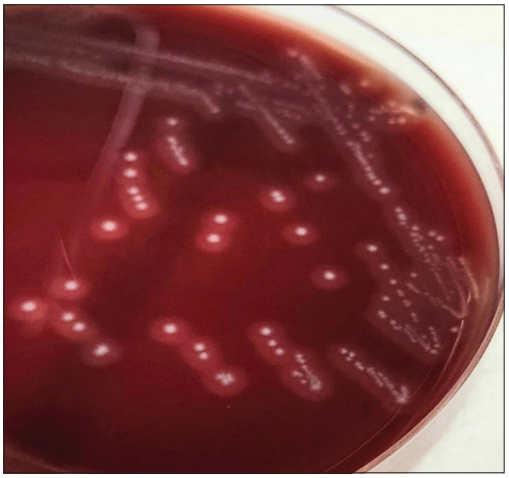


Figure 2: Growth of *Streptococcus dysgalactiae* ssp. *equisimilis* on 5% sheep blood agar

appears as Gram-positive cocci in chains and pairs [4]. Growth was similar in our case except that beta hemolysis was seen instead of the regular alpha hemolysis. In addition, cases of *S. pluanimlaium* causing pilonidal sinus infection are seldom reported in the literature. The drug of choice for *S. pluranimalium* infections are vancomycin, aminoglycosides, and cephalosporins [9]. He was treated with amoxycillin-clavulanate based on antibiotic sensitivity report.

S. dysgalactiae subsp. *equisimilis* is normal flora of human skin, pharynx, gastrointestinal tract, and female genital tract [10]. It is becoming evident as an opportunistic pathogen in humans, causing infections like pharyngitis, cellulitis, and invasive diseases. It has recently been recognized to cause life-threatening infections such as necrotizing fasciitis and toxic shock syndrome [10]. SDSE infections are increasingly identified in India [5]. It produces large beta-hemolytic colonies on blood agar and is universally susceptible to penicillin but has high-level macrolide resistance [5,11]. Our isolate also demonstrated these properties. A consistently increasing incidence of invasive SDSE infections is being observed in elderly males [12]. In India, this organism causes soft tissue infections in males with comorbidities like diabetes, as this may predispose them to skin breakdown [2,5]. Likewise, our patient also was an elderly male with a long-standing case of diabetes mellitus. He was initially treated with vancomycin, but the antibiotic was later changed as per the antibiotic sensitivity report.

CONCLUSION

Emerging *Streptococci* represent a shifting epidemiologic landscape with increased clinical and public health relevance. *S. pluranimalium* and SDSE are agents of the same genus with a wide spectrum of diseases and incurring as human pathogens. Their host range includes both healthy as well as immunosuppressed patients. To the best of our knowledge, cases of *S. pluanimlaium* causing pilonidal sinus infection in humans are rarely reported in the literature. With the help of advanced diagnostic tools, their recognition till the species and subspecies level is possible. Hence, making optimum use of such tools for identifying such atypical organisms, along with clinical correlation and not disregarding them as contaminants, goes a long way in optimum patient management.

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