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Acute Necrotizing Sinusitis Caused by *Staphylococcus lugdunensis*▽

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***Staphylococcus lugdunensis* is most commonly associated with infections arising from the inguinal region, but here we report this organism as a cause of bacterial sinusitis, highlighting its potential niche as a commensal of the upper airways. The severity of necrosis demonstrates the potential for destructive pathology mimicking *Staphylococcus aureus* disease.**

CASE REPORT

A 73-year-old Caucasian man with metastatic prostate adenocarcinoma was hospitalized 23 days following the administration of his 7th cycle of mitoxantrone chemotherapy. He reported a 9-day history of progressive right-sided facial and periorbital swelling, right-sided nasal blockage, serous nasal discharge, and visual blurring. He also described progressive discomfort and swelling in the roof of his mouth and had developed a widespread itchy, vesicular rash in the week preceding admission. Despite 1 week of treatment with oral co-amoxiclav (625 mg three times daily), his symptoms had worsened.

The diagnosis of adenocarcinoma of the prostate (Gleason score of 9, reflecting poorly differentiated disease) had been made 3 years previously, and he had undergone transurethral resection of the prostate and received hormonal treatment with an antiandrogen (bicalutamide), luteinizing hormone-releasing hormone blockade (goserelin), and diethylstilboestrol. Subsequently, he had undergone palliative radiotherapy for bony lesions in the right femur. At the time of admission, he was receiving outpatient chemotherapy with prednisolone (5 mg twice daily) and mitoxantrone, which he had been tolerating well.

On clinical examination, he looked unwell with marked swelling and erythema of the right eyelids and cheek. The right conjunctivae were injected and edematous, and there was mild right-sided proptosis, but no gaze palsy. He had a vesicular rash over the trunk and limbs, suggestive of disseminated varicella-zoster virus infection. There were areas of deep, painful ulceration of the hard palate with surrounding mucosal erythema and edema (Fig. 1A and B). He was afebrile and hemodynamically stable (blood pressure, 135/70 mm Hg; heart rate, 70 beats/min).

Baseline blood tests showed a normocytic anemia (hemoglobin, 9.3 g/dl; mean corpuscular volume, 96.9 fl) and thrombocytopenia (94×10^9/liter), but the white cell count was within the normal reference range (5.8×10^9/liter). C-reactive protein was raised at 74 mg/liter. Blood cultures were sterile. Magnetic resonance imaging with gadolinium enhancement demonstrated marked right-sided proptosis, with only subtle signs within the orbit to explain this; periorbital cellulitis was confirmed, with the inflammatory change extending into the nasolachrymal sac, nares, and philtrum. There was swelling and enhancement of the right lachrymal gland and marked mucosal thickening in the nasal cavity and throughout the ethmoidal sinuses on the right (Fig. 1C). The cavernous sinus and adjacent brain tissue looked normal.

Based on clinical and radiological findings, the diagnosis made was right-sided necrotizing maxillary and ethmoid sinusitis complicated by periorbital cellulitis and conjunctivitis with coexisting disseminated varicella-zoster virus infection. The patient was treated with empirical intravenous co-amoxiclav (1.2 g three times daily), acyclovir (10 mg/kg three times daily), and chloramphenicol eye drops (0.5%, four times daily). Intravenous liposomal amphotericin (AmBisome, 5 mg/kg once daily) was also commenced to cover the possibility of fungal sinusitis/mucormycosis.

The day following admission, he underwent endoscopic sinus exploration under general anesthesia. Necrotic tissue was debrided from the right maxillary antrum (via a right middle meatal antrostomy) and sent for culture and histopathological examination. An organism was identified in pure growth from all three samples and initially reported as *Staphylococcus aureus*, in that it was STAPHaurex positive (Remel, Thermo Fisher Scientific) and DNase positive. However, further laboratory tests (API Staph profile 6716150; bioMérieux Clinical Diagnostics) subsequently identified the organism as *Staphylococcus lugdunensis*. This identification was verified by the Health Protection Agency Respiratory and Systemic Infection Reference Laboratory using phenotypic methodology. The organism was sensitive to penicillin, oxacillin, erythromycin, gentamicin, tetracycline, and ciprofloxacin by disc testing (British Society for Antimicrobial Chemotherapy disc diffusion method). No other organisms, including fungi, were grown

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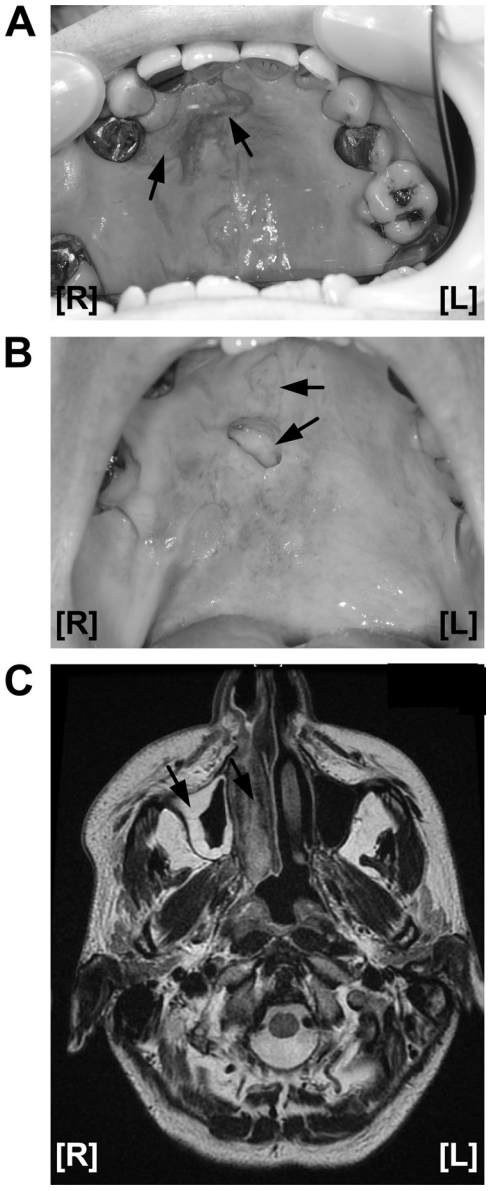

FIG. 1. (A and B) Photographs of the hard palate of a patient with necrotizing infection of the right maxillary sinus. Extensive mucosal defects (arrows) suggest ulceration through from the right maxillary sinus to the underlying palate. (C) Axial T2-weighted (gadolinium-enhanced) magnetic resonance image showing mucosal thickening in the nasal cavity and maxillary sinus on the right side (arrows). [R] and [L] indicate right-left laterality.

from operative samples despite incubation on Sabouraud's agar slopes for 21 days.

Histopathology examination demonstrated extensive inflammation, hemorrhage, and necrosis, suggestive of focal infarction; there was no evidence of malignancy, and no fungal elements were identified.

Over the course of the first week of treatment, his symptoms began to resolve. The proptosis resolved, and visual acuity improved. The rash crusted over, and antiviral therapy was discontinued after 7 days. Despite the lack of evidence of fungal infection, given the clinical suspicion of mucormycosis, antifungal treatment was continued. He received 12 days of intravenous ambisome and co-amoxiclav before switching to oral posiconazole and clindamycin, for a total of 6 weeks of therapy. At outpatient follow-up 1 month after surgery, he reported ongoing resolution of symptoms, including marked improvement of his nasal blockage. Further treatment for his prostate cancer was planned.

This patient presented with a severe acute necrotizing sinusitis complicated by periorbital cellulitis and ulceration through from the maxillary sinus to the hard palate. Macroscopic appearances of necrosis were present at endoscopic sinus exploration, as well as in microscopic examination of tissue samples. Despite the clinical suspicion of fungal disease, the only pathogen to be identified in three independent biopsy samples was *S. lugdunensis*.

The extent to which the presence of malignant disease and the use of cytotoxic chemotherapy contributed to this patient's presentation with infection is uncertain. The mitoxantrone and prednisolone treatment he was receiving is not thought to be heavily immunosuppressive, and he was not neutropenic at presentation. However, we postulate that disseminated varicella-zoster virus infection, arising as a result of defective cell-mediated immunity, could have led to cutaneous or mucosal lesions of the face or nose that provided a portal of entry for a superimposed staphylococcal infection.

S. lugdunensis is a coagulase-negative *Staphylococcus* that is a commensal of human skin (5). As it does not produce free or "tube" coagulase and DNase production can be weak or delayed, it may be reported by the laboratory simply as a "coagulase-negative *Staphylococcus*." This case highlights that correct identification to the species level is important to allow recognition of the organism as a likely pathogen, rather than dismissal as contaminating commensal flora. However, based on the production of bound coagulase (5), *S. lugdunensis* can also easily be misidentified in the laboratory as *S. aureus* (8), as was initially the case in this instance. Because it shares certain virulence characteristics with *S. aureus*, *S. lugdunensis* is now well recognized as a potential cause of invasive and destructive infections that can mimic *S. aureus* disease (1, 4, 7, 10, 14).

S. lugdunensis has been reported as an agent of endovascular infection, particularly in patients with inguinal skin breaks (vasectomy or cardiac catheterization via the femoral route [6, 8]) and as a cause of pelvic girdle abscesses (2), highlighting that the predominant ecological niche of the organism is likely to be the groin. *S. lugdunensis* has also been reported as a cause of skin and soft tissue infections, particularly involving the breast, abdomen, and lower limbs (1, 2, 4), and has been identified as an agent of prosthetic joint infection (10). In keeping with these reports, studies that have screened healthy persons for carriage have confirmed identification from skin of the abdomen, groin, and lower extremities (3, 12).

However, as well as being a skin commensal of the groin and lower limbs, *S. lugdunensis* has also been isolated from saliva and from nasal swabs (in 2% of saliva samples analyzed and in 6% of healthy adults with nasal carriage of staphylococci [9]). It has also been isolated in the setting of acute oral infection (13), and one case report documents it as an agent of osteo-

myelitis of the temporal bone in a patient with diabetes (11). These reports—and the case we describe here—confirm *S. lugdunensis* as a possible cause of head and neck infections.

To our knowledge, this case is the first reported instance of *S. lugdunensis* as a cause of facial cellulitis and sinusitis. The case is also striking in highlighting the potentially aggressive and destructive nature of infections with this pathogen, in this instance necessitating surgical debridement and prolonged treatment with antibiotics.

We are grateful to the patient for his permission to publish this report. The Health Protection Agency Reference Laboratory at Colindale provided confirmation of the identification of the bacterial isolates in this case as *S. lugdunensis*.

None of us has any conflicts of interest to declare.

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