CASE REPORT

Acute Necrotizing Sinusitis

Caused by *Staphylococcus lugdunensis*

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ABSTRACT

Staphylococcus lugdunensis is most commonly associated with infections arising from the inguinal region, but we here 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 admitted to hospital 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 one week of treatment with oral co-amoxiclav (625mg three times daily), his symptoms had worsened.

The diagnosis of adenocarcinoma of the prostate (Gleason score 9, reflecting poorly differentiated disease) had been made three years previously, and he had undergone trans-urethral resection of the prostate and received hormonal treatment with an anti-androgen (Bicalutamide), LHRH 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 out-patient chemotherapy with prednisolone 5mg 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 oedematous 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* infection. There were areas of deep, painful ulceration of the hard palate with surrounding mucosal erythema and oedema (Fig. 1A,B). He was afebrile and haemodynamically stable (blood pressure was 135/70 mmHg, heart rate 70 beats per minute).

Baseline blood tests showed a normocytic anaemia (Hb 9.3 g/dl, MCV 96.9 fl) and thrombocytopenia (94 x 10^9/l), but the white cell count was within the normal reference range (5.8 x 10^9/l). C-reactive protein (CRP) was raised at 74 mg/l. Blood cultures were sterile. Magnetic Resonance Imaging (MRI) with gadolinium enhancement demonstrated marked right-sided proptosis, with only subtle signs within the orbit to explain this; periorbital cellulitis was confirmed, with inflammatory change extending into the nasolacrimal sac, nares and philtrum. There was swelling and enhancement of the right lacrimal 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, a diagnosis was made of right-sided necrotising maxillary and ethmoid sinusitis, complicated by peri-orbital cellulitis and conjunctivitis with co-existing disseminated zoster. The patient was treated with empirical intra-venous co-amoxiclav (1.2 g three times daily) and aciclovir (10 mg/kg three times daily), and chloramphenicol eye-drops (0.5%, four times daily).
venous liposomal amphotericin (AmBisome® 5mg/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 anaesthetic. 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, 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 (BSAC disc diffusion method). No other organisms, including fungi, were grown from operative samples despite incubation on Sabouraud’s agar slopes for 21 days.

Histopathology examination demonstrated extensive inflammation, haemorrhage 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 for fungal
infection, given the clinical suspicion for mucormycosis, anti-fungal treatment was continued. He received twelve days of intravenous ambrisome and co-amoxiclav, before switching to oral posaconazole and clindamycin, for a total of six weeks of therapy. At outpatient follow-up one 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 from three independent biopsy samples was *Staphylococcus 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*, 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 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 species level is important to allow recognition of the organism as a likely pathogen, rather than being dismissed as contaminating commensal flora. However, based on the production of bound coagulase (5), *S. lugdunensis* can also easily be mis-identified in the laboratory as *Staphylococcus aureus* (8), as was initially the case in this instance. Because it shares certain virulence characteristics with *S. aureus*, *S. lugdunensis* is now well-recognised 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 subjects with inguinal skin breaks (vasectomy, or cardiac catheterization via a 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 limb (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 subjects 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 limb, *S. lugdunensis* has also been isolated from saliva and from nasal swabs (in 2% of saliva samples analysed, 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 osteomyelitis 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 patient 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.

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REFERENCES


FIGURE LEGENDS

Figure 1

A/B: Photographs of the hard palate in 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 Magnetic Resonance Imaging (gadolinium-enhanced) showing mucosal thickening in the nasal cavity and maxillary sinus on the right side (arrows). [R]/[L] indicate right/left laterality.