

Capnocytophaga Canimorsus Sepsis Causing Sweet's Syndrome

Sir,

Capnocytophaga canimorsus, formerly called Dysgonic fermenter 2 (DF-2), is a thin Gram-negative rod found as part of the normal oral flora in saliva from dogs and cats. Transmission to humans is usually as the result of bites or scratches, but mere contact with infected animals can be enough to transmit the bacteria. Human infection with this bacterium was first reported in 1976 (1) and since then >100 cases have been reported.

In humans the bacteria can cause systemic infections but only rarely causes localized infections. Infections in healthy individuals are rare because of the low virulence of the bacteria. The majority of cases have been reported in individuals with a predisposing condition, most commonly splenectomy, alcohol abuse, chronic lung disease and treatment with immunosuppressive drugs.

Systemic infections mostly comprise septicemia, in some cases complicated by meningitis or infection localized to the heart. Peripheral gangrene is common but other skin symptoms, such as petechiae, purpura, erythema nodosum and macular-, papular- and erysipelas-like rashes, have also been reported. Reviews are given by Lion et al. (2) and by Griego et al. (3). Here we report for the first time a case of Sweet's syndrome (acute febrile neutrophilic dermatosis) following sepsis with *Capnocytophaga canimorsus*.

CASE REPORT

A 56-year-old otherwise healthy woman was admitted to our hospital with a 2-day history of headache, abdominal pain, rash and fever. The rash consisted of multiple, painful, sharply demarcated raised erythematous plaques on the face, neck, back and extremities (Fig. 1). The plaques were infiltrated, without pseudoblistering or pustules, and the mucous membranes were not affected. The patient's body temperature was 38.9°C and she was hemodynamically stable. Laboratory tests showed an elevated erythrocyte sedimentation rate of 76 mm/h, a C-reactive protein level of 267 mg/l and a neutrophilic leukocytosis of $15.9 \times 10^9/l$. Morphology of blood cells was normal. Bone marrow biopsy was not performed. Apart from a slightly elevated bilirubin level of 25 µmol/l all other blood tests were within normal limits. Abdominal ultrasound examination, chest X-ray and echocardiogram were normal.



Fig. 1. Sharply demarcated raised red plaques with a faint, central white-yellow discoloration were observed on the patient's neck and upper back.

Hematoxylin-eosin staining of a punch biopsy of the lesions showed a dermal neutrophilic infiltrate. The vessels were dilated, with a swollen endothelium, but without any signs of vasculitis. Gram staining showed no bacteria in the biopsy. The histological findings were interpreted as being in accordance with the diagnosis of Sweet's syndrome.

The patient was initially treated with prednisolone at a daily dose of 0.5 mg/kg. As a result she improved dramatically and the rash faded. On the third day after admission 2 sets of BACTEC aerobic blood cultures incubated at 35°C revealed thin, curved Gram-negative rods with tapered ends. *Capnocytophaga* species was already suspected at this time as a result of the morphology of the bacteria. On subculture the organism grew only on chocolate agar in an atmosphere of 7.5% carbon dioxide. The colonies were initially pinpoint but after further incubation they became larger, convex and smooth. After isolation by culture the bacteria were identified as *Capnocytophaga canimorsus* based on the following identification profile: gliding motility; facultative anaerobic; positive for catalase and oxidase; acid produced from glucose and maltose but not from mannitol and sucrose; no detection of ornithine and lysine decarboxylase activity; no reduction of nitrate to nitrite; and urea not hydrolyzed. Treatment with prednisolone was stopped and replaced with intravenous penicillin 5×10^6 IU 3 times daily and intravenous netilmicin 300 mg daily. Antimicrobial susceptibility testing was performed; as the isolated bacteria were susceptible to penicillin netilmicin was stopped. Treatment with penicillin was carried out for a total of 15 days. The patient recovered without any resultant disability and remained well 18 months after she was discharged from hospital.

The woman had never previously been hospitalized. She had a dog kennel, but had no memory of a recent dog bite.

DISCUSSION

Acute febrile neutrophilic dermatosis was first described in 1964 (4) and the entity was later termed Sweet's syndrome. Clinically the syndrome encompasses a condition with a characteristic rash on the face, neck, back, chest and extremities together with fever and leukocytosis. Other organs may be involved, such as the joints, eyes and rarely the kidneys, lungs, liver and pancreas. Depending on the associated condition Sweet's syndrome can be subdivided into 4 groups: idiopathic; para-inflammatory; paraneoplastic; and associated with pregnancy (5). It is most likely that our patient's rash was caused by the *Capnocytophaga canimorsus* infection as the patient recovered completely when she was treated with antibiotics. Examinations for other conditions associated with Sweet's syndrome were all normal as described in the case report.

Clinically the most obvious differential diagnosis in our patient was erythema multiforme and it has been suggested that there is an overlap between the 2 conditions. However, the biopsy showed no signs of changes in the interface between the dermis and epidermis as seen in erythema multiforme.

Our patient fulfilled the diagnostic criteria proposed by Su & Liu (6) for the diagnosis of Sweet's syndrome, and to our knowledge this is the first time a case of Sweet's syndrome in combination with *Capnocytophaga canimorsus* infection has been reported. The mechanism by which infection with *Capnocytophaga canimorsus* could have caused Sweet's syndrome remains, however, elusive.

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Sarcoidosis Developing after Stopping Oral Prednisolone for Systemic Lupus Erythematosus

Sir,

Although neither the pathogenesis of sarcoidosis nor that of systemic lupus erythematosus (SLE) has been elucidated, both are thought to result from abnormalities of the immune system. However, association of sarcoidosis and SLE is quite rare. We report here a patient who developed sarcoidosis after oral prednisolone therapy for SLE was stopped.

CASE REPORT

A 60-year-old Japanese woman was referred to our hospital in December 1999 for examination of a subcutaneous nodule on her left forearm that had appeared 1 month before. She had a butterfly rash, photosensitivity, leukopenia, antinuclear antibodies and anti-double-stranded DNA. She had been followed since the diagnosis of SLE in 1994, and oral prednisolone therapy for SLE had suddenly been stopped 2 months prior to that. A subcutaneous tumour subsequently appeared on her left forearm. She also noticed edematous erythema on both eyelids. Physical examination revealed the existence of multiple subcutaneous nodules on the extremities and buttocks.

Laboratory examination showed the following abnormalities: antinuclear antibody titre (320 ×) with a homogeneous and speckled pattern, positive antibodies to double-stranded DNA and SSA antigen, and reduced white blood cell count and hemoglobin. Antibodies to RNP, Sm and SSB antigens, LE test, rheumatoid factor, serum angiotensin-converting enzyme and serological tests for syphilis were negative.

A skin biopsy from the nodular lesion on her left forearm showed noncaseating epithelioid cell granulomas throughout the dermis, and a diagnosis of sarcoidosis was made. A biopsy specimen taken from the edematous erythema on her right eyelid revealed hydropic degeneration of basal cells and a dense superficial perivascular mononuclear cell infiltrate around superficial blood vessels, indicating SLE.

Chest X-ray and computed tomography demonstrated bilateral

hilar lymphadenopathy. Ophthalmic examination revealed endogenous uveitis.

The patient was diagnosed as having sarcoidosis with underlying SLE. She was treated with oral prednisolone (30 mg/day). The edematous erythema on the eyelids completely subsided in 1 week, and the subcutaneous nodules on the extremities and buttocks gradually became soft and small.

DISCUSSION

Teilum (1) first suggested that sarcoidosis and SLE might be involved in the same underlying immune mechanisms. David et al. (2) reported a patient with sarcoidosis who developed rapidly progressive SLE after steroid therapy was withdrawn. In our case, sarcoidosis occurred after cessation of oral prednisolone, and SLE was exacerbated simultaneously. Such cases suggest that once immunosuppression has occurred, steroid reduction may rapidly lead to immune dysfunction, resulting not only in recurrence of the disease but also in development of an allied disease.

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