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Case Report

Facia pyoderoma gangrenosum: A case report

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ABSTRACT

We present an unusual case of a 45-year-old lady presenting with rapidly progressing lesions affecting the face with underlying colitis. Following investigation and multidisciplinary involvement she was treated for pyoderma gangrenosum with resolution of her symptoms.

We outline the presentation and discuss the diagnosis and management challenges associated with such cases.

1. Introduction

Pyoderma gangrenosum is an uncommon, ulcerative cutaneous condition classified as a neutrophil dermatosis. The exact aetiology is unclear and often, but not invariably, reflects underlying systemic disease. Several variants exist and the condition rarely presents in the head and neck [1,2].

Misdiagnosis and inappropriate management have been quoted in the literature and can result in significant delay in appropriate treatment and increased morbidity. Given the rarity with which head and neck surgeons see this condition its resemblance to necrotizing fasciitis, severe bacterial, fungal and atypical mycobacterial infection especially in immunosuppressed individuals can lead to misdiagnosis and early aggressive surgical treatment [3]. Oral and maxillofacial surgeons should be conscious of this condition and acknowledge the propensity for rapid progression if early immunosuppression is not instituted.

2. Case report

A 45-year-old Caucasian female presented to the oral and maxillofacial surgery department with a 4-day history of increasing pain, swelling and erythema of the right peri-auricular tissues and left upper eyelid.

She had not suffered any previous episodes and gave no history of recent skin trauma, insect bite or foreign travel. She had been under the gastroenterologists 2 years prior for non-specific bowel symptoms. The symptoms were attributed to irritable bowel syndrome or gastrointestinal infection and no further follow up in secondary care was organized.

At presentation she was afebrile and haemodynamically stable. She had painful erythematous lesions affecting the right ear lobule, peri-auricular area (Fig. 1) and left upper eyelid.

There was no evidence of abscess formation requiring surgical drainage. Routine haematological investigations revealed only a mildly raised white cell count and C-reactive protein.

She was admitted for observation and broad-spectrum intravenous antibiotic therapy with a presumed diagnosis of severe facial cellulitis. There was no initial response and the lesions progressed to superficial ulceration with pustulation in 24 h (Fig. 2). Wound swabs showed no growth and computed tomography showed no orbital extension or collections.

The clinical picture and rapidly progressive nature of the lesions were consistent with a diagnosis of necrotizing fasciitis or severe fungal infection such as mucormycosis and aggressive surgical debridement was planned (Figs. 3 and 4). However, upon further questioning and systems review she described significant bowel symptoms. She reported multiple episodes of bloody diarrhoea with mild abdominal discomfort. She eluded to weight loss over the last few months but was unable to quantify the exact amount.

A full examination revealed erythematous nodules with central necrosis on her back and arms (Figs. 5 and 6). Repeat blood tests showed a mild drop in haemoglobin and hypokalaemia secondary to diarrhoea.

The maxillofacial team suggested a diagnosis of facial pyoderma gangrenosum associated with underlying inflammatory bowel

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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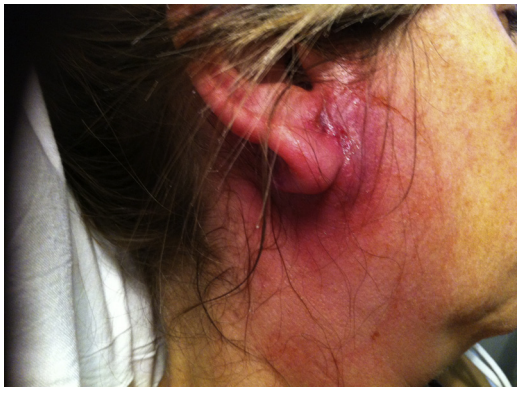

Fig. 1. Inflammation of peri-auricular skin.

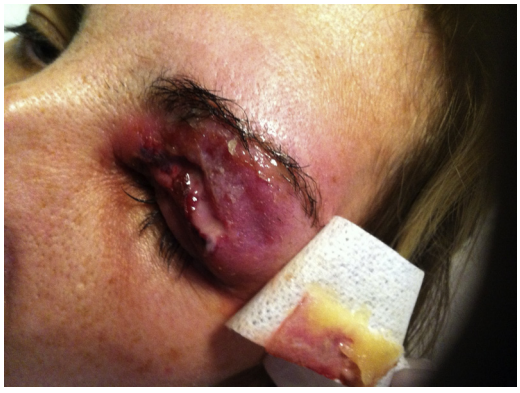

Fig. 2. Progression of lesion affecting upper eyelid.

disease. After involvement of the gastroenterologists and dermatologists she underwent a flexible sigmoidoscopy and multiple skin biopsies. Sigmoidoscopy revealed active colitis. Skin biopsies showed complete necrosis with neutrophil debris.

A diagnosis of pyoderma gangrenosum and ulcerative colitis was confirmed and the patient was commenced on oral azathioprine and pulsed intravenous methylprednisolone resulting in rapid improvement in both her gastrointestinal and skin conditions.

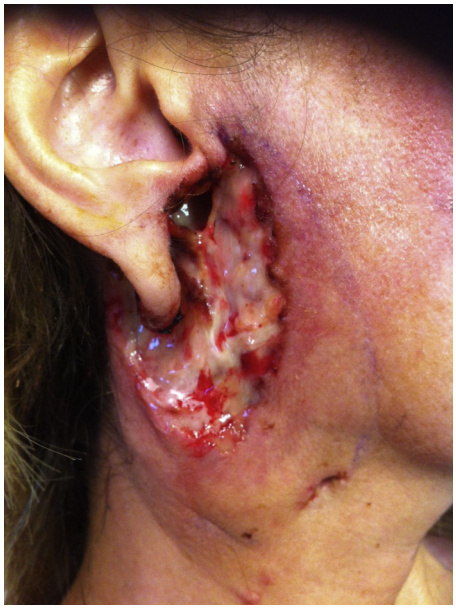

Fig. 3. Further necrosis of peri-auricular skin.

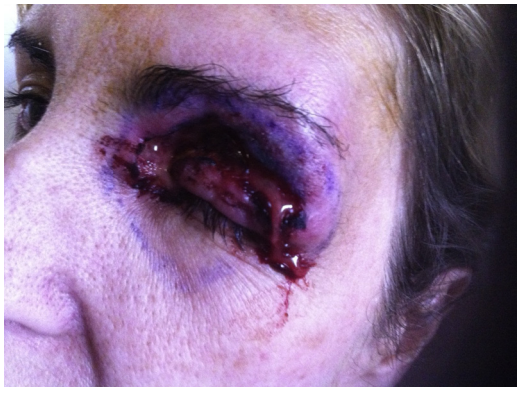

Fig. 4. Ongoing inflammatory process affecting upper eyelid.

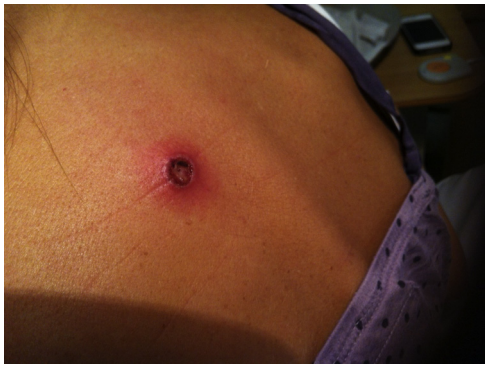

Fig. 5. Erythematous nodules with central necrosis.

3 months later, she remained controlled on oral azathioprine and prednisone and is currently being considered for infliximab infusions to hasten remission of her colitis. Her skin lesions have not completely healed but her eyelid function is not affected. She may well require reconstructive surgery to improve cosmesis once full remission is achieved (Figs. 7 and 8).

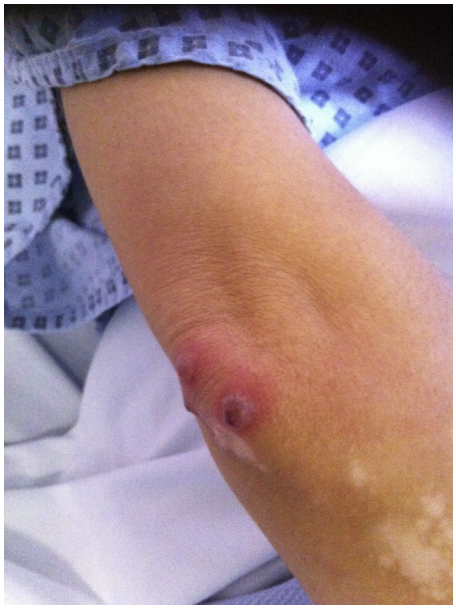

Fig. 6. Erythematous nodules with central necrosis.