Original Contributions

Case Report

# Dental treatment of a rare case of pyoderma gangrenosum with aggressive periodontal disease

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## ABSTRACT

**Background and Overview.** Pyoderma gangrenosum (PG) is a rare neutrophil-mediated autoinflammatory dermatosis that can involve the oral mucosa. Dental surgery is a potential triggering factor for the onset of PG lesions. The authors describe and discuss the dental management of a rare case of aggressive periodontitis in a patient with PG, from multiple tooth extractions to prosthetic rehabilitation, including administration of systemic steroid prophylaxis before surgery to prevent the potential onset of PG-related lesions.

**Case Description.** A 22-year-old man who had a diagnosis of PG and who had aggressive periodontal disease underwent dental extractions, gingivoplastic surgery, and prosthetic rehabilitation. The patient received 8 milligrams of betamethasone intramuscularly 20 minutes before the oral surgery. The tissues healed perfectly, and no adverse effects were reported.

**Conclusions and Practical Implications.** For minor oral surgery, prophylactic corticosteroids might help reduce the risk of developing PG-related lesions. The clinician should plan the prosthetic devices to be as atraumatic as possible.

**Key Words.** Oral surgery; tooth extraction; prosthetics.

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First reported in 1930 by Brunsting,[1] to our knowledge, pyoderma gangrenosum (PG) to date is regarded as a neutrophil-mediated autoinflammatory dermatosis.[2,3] PG is a rare skin disease with an incidence ranging from 3 to 10 patients per million,[4] which may affect people at any age, with a peak at 20 to 50 years and with a slight predisposition for female patients.[4,5] PG involvement of the oral cavity is even more rare, with just a few cases reported in the literature.[6] In approximately one-half of the cases, a systemic disease associated with PG can be identified; often, these are inflammatory bowel diseases, endocrine diseases, rheumatoid arthritis, and hematologic disorders.[3,5] Otherwise, PG can be idiopathic or can contribute to defining rare syndromes, such as pyogenic arthritis, PG, and acne (known as *PAPA syndrome*).

The typical PG signs are sterile cutaneous pustules, in most cases localized at the legs, characterized by a neutrophilic granulocyte infiltrate.[7] In the ulcerating PG subtype, pustules result in painful ulcers with undermined purple borders and a central area of necrosis exuding pus, whereas in the vegetating PG subtype, the same pustules result in vegetating erosions.[7] Pustular forms of PG, either as bullous lesions (*bullous PG*) or as multiple pustules (*pustular PG*), are sporadic.[7] Further extracutaneous lesions can involve the subcutaneous adipose tissue in the form of panniculitis and the eye with corneal ulceration. Internal organs, such as bone or lungs, are affected rarely.[7] Infrequently, investigators have described intraoral and lip ulcers in patients with PG.[6-8]

The diagnosis requires the presence of both of the 2 major criteria (presence of clinical lesions and exclusion of other possible causative diseases on the basis of medical history and clinical findings) and at least 2 additional criteria, which include results from microbiological testing and histologic

examination, particularly pivotal for the differential diagnosis.[7] Histologic examination results contribute to excluding other diseases rather than to confirming the diagnosis because investigators have found that results from only 7% of biopsy specimens showed specific features, such as neutrophilic infiltration of the dermis, signs of vasculitis, and accumulation of immunoglobulins or complement factors beside the vessels.[7,9] PG standard treatment consists of administering systemic steroids, although dapsone is an alternative therapy in resistant cases.[7,9] Topical steroids are particularly useful in managing oral ulcers.[10]

Most patients report a triggering factor that initiated or worsened the disease. The phenomenon of pathergy, which correlates with an abnormal response to stimuli such as mechanical traumas (surgery, stings, or bites), is a typical finding in PG.[7,11] When surgery is required, evidence suggests that corticosteroid-based prophylaxis could reduce the risk of developing PG-related lesions triggered by surgical trauma,[12,13] although no specific guidelines for patients with PG who are undergoing dental surgery are available. In 2007, authors of a clinical case reported the onset of a PG lesion in the maxillary sinus after tooth extraction; the authors concluded that prophylactic corticosteroids could be of help in surgical interventions.[13] In 2017, Bissonnette and colleagues[8] reported a further recommendation; they stated that surgical debridement of PG lesions should be avoided without concomitant medically induced immunosuppression or preoperative corticosteroids because surgery can exacerbate cutaneous PG.

Given the rarity of this disease and the limited availability of established guidelines for oral surgery, in this clinical case we aimed at reporting the dental management of a case of aggressive periodontal disease in a young patient with a diagnosis of PG, who required multiple dental extractions and gingivoplastic surgery and who was not receiving concurrent immunosuppressive therapy at the time of dental surgery. We performed the surgery after intramuscular administration of corticosteroid prophylaxis to reduce the potential risk of triggering PG-associated oral lesions. We also describe the subsequent prosthetic oral rehabilitation, which resulted altogether in a clinically successful outcome.

## CASE REPORT

In October 2015, the patient's general practitioner referred a 22-year-old man to our clinic (Clinica Odontoiatrica Presidio Ospedaliero San Paolo, ASST Santi Paolo e Carlo, Università degli Studi di Milano, Milan, Italy) for periodontal evaluation. The patient had hepatitis B and 3 years previously had received a diagnosis of cutaneous PG. Since then, he had had a history of intermittent systemic steroid treatment with dexamethasone and dapsone. When he came to our clinic he was using dapsone, which his specialist suspended the use of a few weeks later.

At oral examination, the patient was partially edentulous, with few remaining teeth, which were in poor condition and characterized by Grade III mobility according to the Miller classification,[14] severe gingival inflammation, and bleeding on gentle probing. The patient had poor oral hygiene compliance and reported severe pain when toothbrushing (Figure 1A). The panoramic radiograph showed severe loss of alveolar bone, consistent with the clinical picture (Figure 1B). We formulated the following treatment plan: extraction of all of the remaining teeth in 2 stages (maxilla and, subsequently, mandible) and the delayed fabrication of maxillary and mandibular total dentures.

At the time of dental extraction, we premedicated the patient, who had not been receiving any systemic treatment for PG for a few weeks, with 8 milligrams of betamethasone (Bentelan, Sigma-Tau Industrie Farmaceutiche Riunite) administered intramuscularly 20 minutes before surgery. After the patient used a mouthrinse with 0.2% chlorhexidine gluconate (Clorexisan 0.2%, Olcelli Farmaceutici) immediately before the intervention, we induced local anesthesia by administering mepivacaine 2% plus epinephrine 1:100.000 (Optocain, Molteni Dental). We extracted all teeth without needing to open flaps or perform an osteotomy. We achieved a first intention suture with silk. The patient received postsurgical instructions, and we prompted him to rinse with 0.2% chlorhexidine gluconate twice per day for 2 weeks and apply 1% chlorhexidine gel (Corsodyl 1g/100g, GlaxoSmithKline) 3 times per day for 2 weeks on the wounds. We preserved the extracted teeth in 10% formalin for histologic analysis. We removed the sutures 1 week after surgery after complete healing of the soft tissues (Figure 2). At weekly follow-up visits, the patient did not complain of any pain or discomfort.

One month after the mandibular extractions, the soft tissues appeared to have healed perfectly, but, in the third quadrant, gingival remodeling resulted in a mucosal flap that would have hindered the correct fabrication of the mandibular denture. Therefore, we performed gingivoplastic surgery in the affected area (Figures 3A and 3B) and used the same premedication protocol with

**ABBREVIATION KEY**

**PG:** Pyoderma gangrenosum.