Journal of Plastic, Reconstructive & Aesthetic Surgery (2011) 64, 1528–1532

JPRAS
An International Journal of Surgical Reconstruction
www.JPRASurg.com

CASE REPORT

Pyoderma gangrenosum: A report of a rare complication after knee arthroplasty requiring muscle flap cover supplemented by negative pressure therapy and hyperbaric oxygen

D.S. Hill*, J.K. O'Neill, A. Toms, A.M. Watts

Department of Plastic Surgery, Royal Devon and Exeter Foundation Trust, Barrack Road, Exeter EX2 5DW, United Kingdom

Received 17 December 2010; accepted 12 March 2011

KEYWORDS
Pyoderma;
Gangrenosum;
Pathergy;
Knee;
Arthroplasty

Summary Pyoderma gangrenosum (PG) is rare ulcerating skin condition easily confused with wound infection following surgery. We report a complicated case of PG following knee arthroplasty where delayed diagnosis and repeated debridements lead to significant tissue loss. Successful reconstruction was achieved with a muscle flap, but subsequent reactivation of PG and superadded infection placed both the reconstruction and patient's life at risk. Prolonged combined use of negative pressure therapy (NPT), immunosuppression and hyperbaric oxygen (HBO) was successfully used to reduce the wound size, enhance wound granulation, promote re-epithelialisation, and provide pain relief. There is little or no published literature on these treatment modalities for the management of PG, with only one reported case using both NPT and HBO for PG (not following knee arthroplasty). More studies are necessary to determine the role of both modalities in the management of pathergy in large and complex wounds and the rare nature of this complication following knee arthroplasty explains the lack of evidence-based guidance.

In conclusion, we suggest a surgical algorithm. This is the first report of PG following knee arthroplasty with the use of both NPT and HBO in order to achieve soft tissue coverage.

* Corresponding author. Tel.: +44 7809 761746.
E-mail addresses: danielhill@doctors.org.uk (D.S. Hill), jenniferoneill@hotmail.com (J.K. O'Neill), andrew.watts@rdeft.nhs.uk (A.M. Watts).

1748-6815/$ - see front matter © 2011 Published by Elsevier Ltd on behalf of British Association of Plastic, Reconstructive and Aesthetic Surgeons.
doi:10.1016/j.bjps.2011.03.024

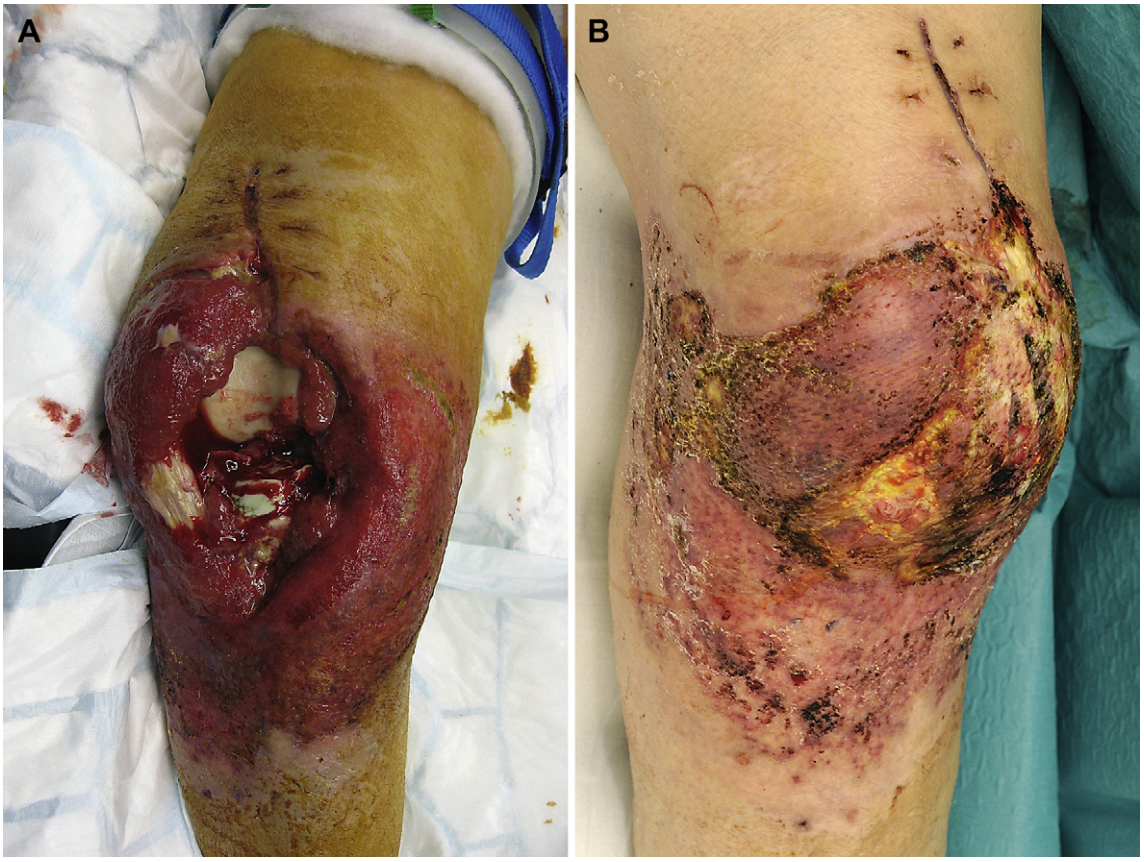

Figure 1 (A) Exposed knee joint. (B) Successful soft tissue coverage 14 days post operatively.

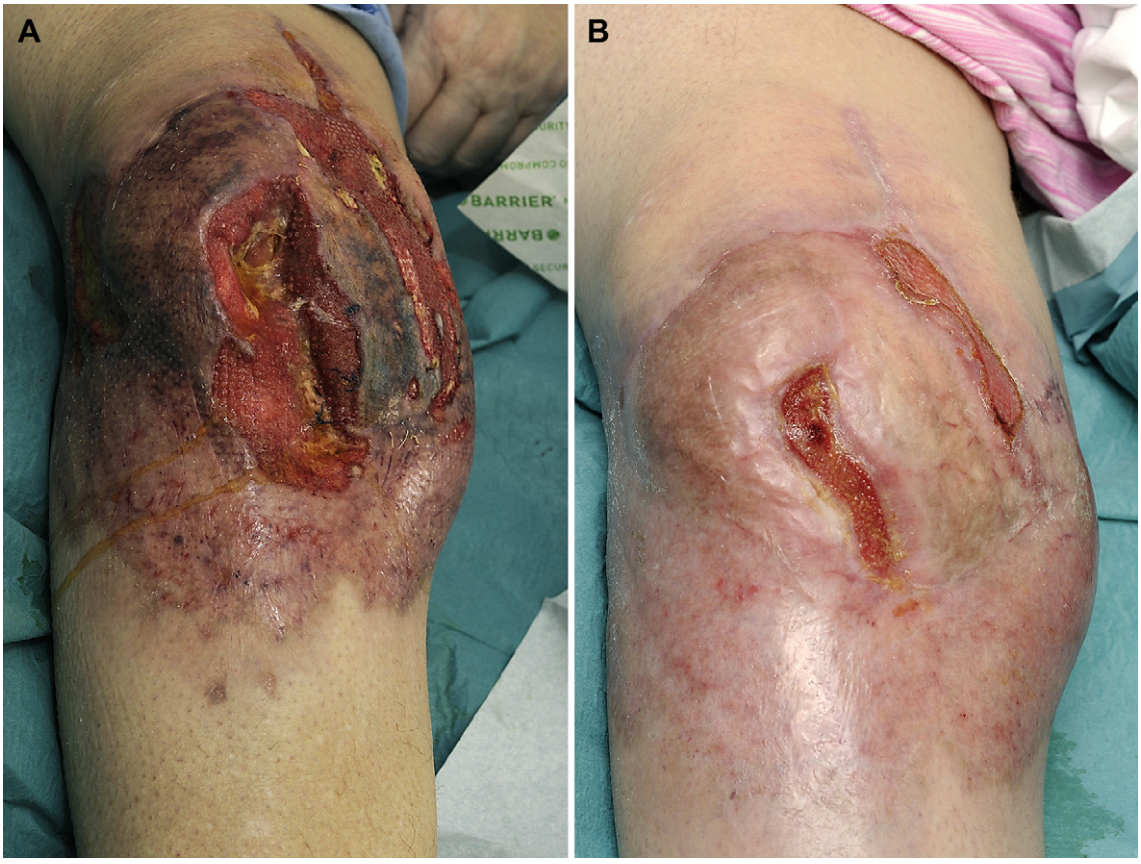

Figure 2 (A) Wound breakdown (post lavage). (B) Wound after HBO therapy.

Introduction

Pyoderma gangrenosum (PG) is a rare autoimmune disorder resulting in ulceration and necrosis of the skin. Underlying inflammatory, immunological, or neoplastic disorders occur in 50—70% of cases. Cutaneous trauma can lead to lesions in up to 30% of patients with PG — a concept termed pathergy. Only four cases following knee arthroplasty[1—4] and two following hip arthroplasty have been reported in the literature.[5,6] We describe a case where suppurative PG following knee arthroplasty required large soft tissue defect coverage. Negative Pressure Therapy (NPT) and HyperBaric Oxygen (HBO) were used alongside pharmacological treatments to control disease progression, infection and prevent disease reactivation. This is the first time all these modalities have been used to treat PG in the knee. We highlight the severe nature of this condition and postulate that multimodal treatment and multidisciplinary care offer the best outcome. We summarise our experience with a proposed surgical algorithm. The most important aspect is early recognition.

Case report

A 63-year-old woman was referred with a large full thickness defect over her right knee following a unicompartmental knee arthroplasty 26 days previously (Figure 1A). Her past medical history included Sjögrens disease.

Six days following arthroplasty the surgical wound broke down. This was initially thought to be a wound site infection, and treatment was irrigation and debridement with aggressive antimicrobial therapy. Subsequently, NPT was trialed for a five-day period to promote wound healing, but unfortunately wound breakdown progressed. Superficial wound swabs grew *Staphylococcus aureus* and *Enterobacter cloacae*. Deep tissue samples were negative.

12 days following initial surgery the exposed prosthesis was removed, but the progressive ulceration and wound breakdown continued and multiple debridements took place. Satellite lesions at IV cannulation sites (Koebner phenomenon) appeared nine days after initial wound breakdown. This raised suspicions of a systemic process, and a Rheumatologist diagnosed PG based on this together with the clinical features of the wound. Histological examination of tissues was non-specific.

Immunosuppression with high dose Prednisolone was commenced. After 12 days of treatment the erythematous—violaceous borders had regressed and the C-Reactive Protein (CRP) values had decreased from 291 to 11. 27 days following the initial arthroplasty the exposed femoral condyles and cement spacer were covered using a medial gastrocnemius muscle flap and split thickness skin graft. In the immediate postoperative period no pathergy developed at the donor site and seven days after surgery the knee remained covered with healthy muscle and approximately 80—90% graft take (Figure 1B). At this time intravenous antibiotics were changed to oral Rifampacin, but over the following two weeks, the CRP gradually climbed to 259. An initial full septic screen was returned as normal. 19 days post reconstruction the patient developed septic shock requiring ITU admission, inotropic support and immunoglobulin therapy. The knee wound had broken down, so the joint was irrigated and a large quantity of pus was drained from the donor site. Superficial tissue swabs again grew *Enterobacter cloaceae*, but deep samples failed to culture any organisms.

Surgical washout of the knee joint again exposed the femoral condyles (Figure 2A). NPT was used to assist with wound closure but was not successful, and the patient was referred for HBO treatment. After 30 daily HBO treatments in conjunction with NPT, the knee joint was no longer exposed and the wound was granulating and contracting well (Figure 2B). A reducing dose of Prednisolone was commenced and the patient remains on low dose therapy today.

Discussion

Postoperative wound breakdown is commonly caused by infection. Rarely, the same can result from surgically induced pathergy. Clinically differentiating these is difficult, requires a high index of suspicion, and is essential to avoid multiple surgical debridements. A severe inflammatory syndrome (despite aggressive antibiotic treatment) with no bacterial growth from wound swabs along with high WCC should raise the surgeon's suspicions. In addition to

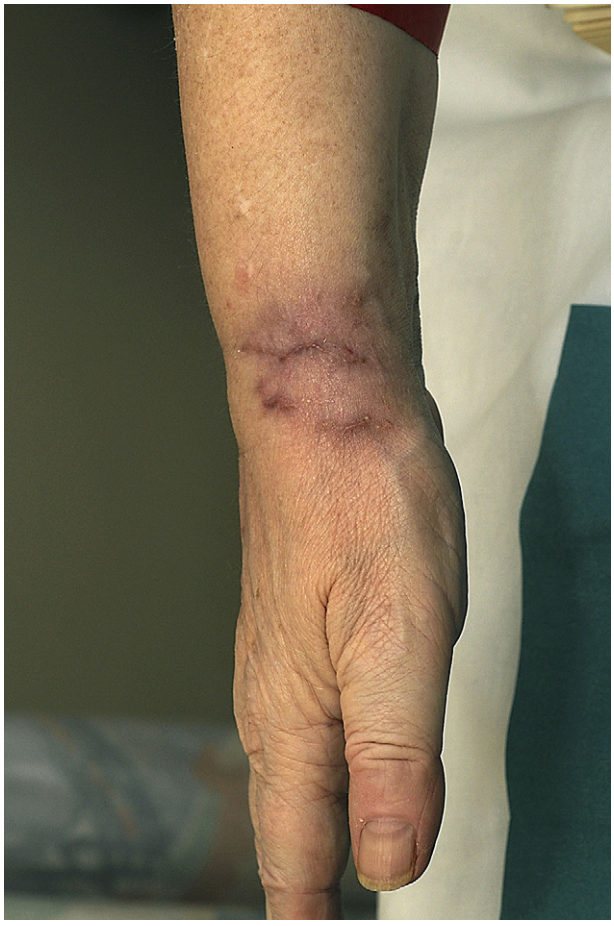

Figure 3 Reaction at peripheral venous cannula site (Koebner phenomeum).

the classic clinical appearance of PG, we propose that debridement resulting in dramatically increased wound breakdown alongside the presence of satellite lesions at multiple intravenous cannulation trauma sites should also raise suspicions (Figure 3). Histological examination of tissues is usually non-specific in cases of PG.[5]

Advances in the understanding of PG have lead to improved responses to medical treatments, including immunomodulatory and immunosuppressive therapies.[4] Mild cases can be successfully managed with pharmacological therapy alone. Despite this, resistant PG ulcers are encountered, requiring additional measures to achieve

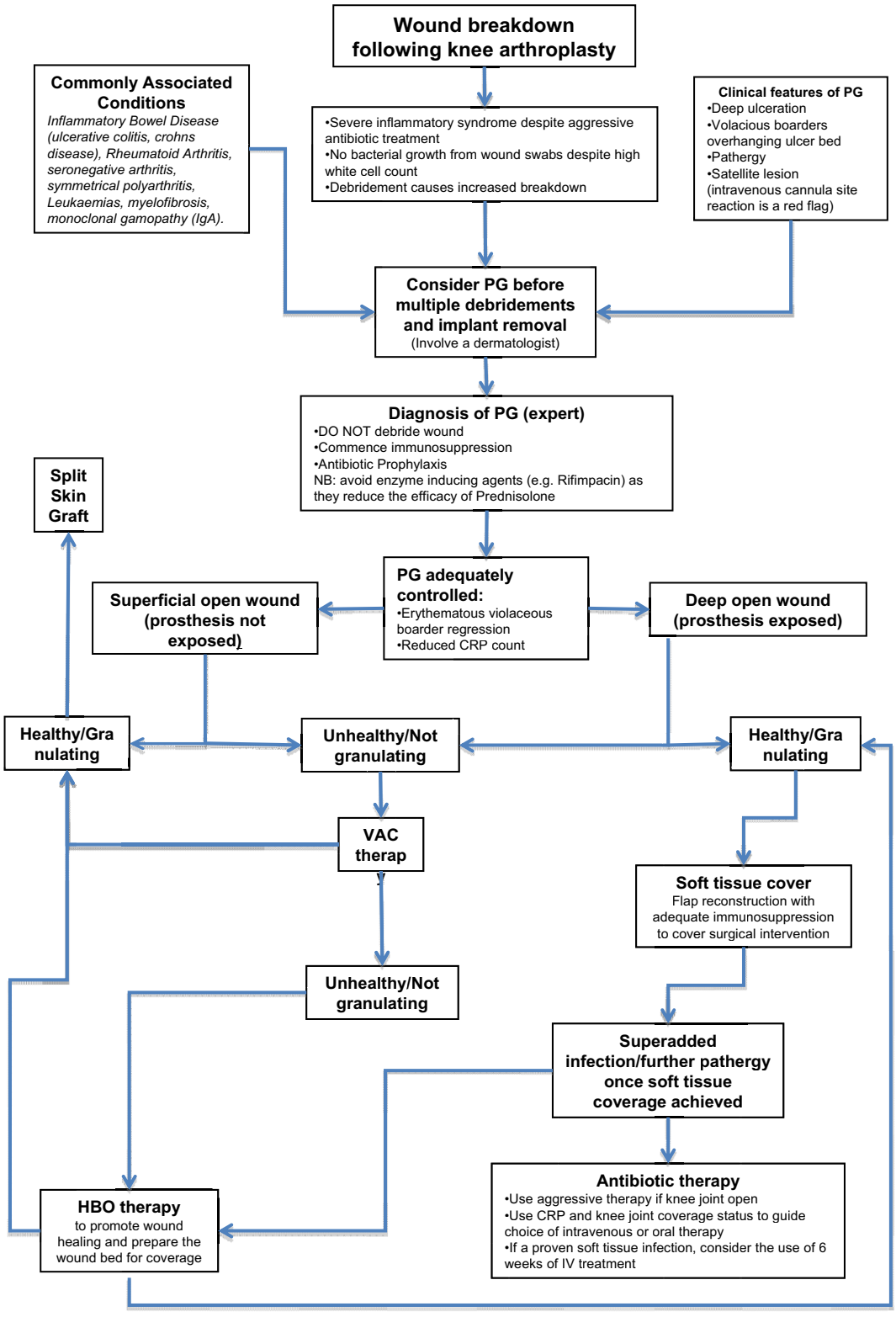

Figure 4 Surgical management algorithm for pathergy following knee arthroplasty.

wound closure. The surgical management of active PG ulcers remains discouraged owing to the risk of exacerbating the inflammatory reaction. In our case, exposure of the knee joint and the risk of developing osteomyelitis made the necessity for wound coverage a life and limb saving procedure. No pathergy occurred at the donor site. Two previous cases are reported of successful flap coverage of large soft tissue defects in patients with pathergy following arthroplasty. These were both in the knee and included; a latissmus dorsi free flap[3] and a medial gastrocnemius flap where NPT was used preoperatively.[1] In our case, the combination of reducing the does of Prednisolone and commencing oral Rifampacin (an enzyme inducer known to reduce the therapeutic dose of Prednisolone) may explain the reactivation of PG and flap breakdown.

NPT therapy has become an important tool for the management of complex wounds. In our case the use of NPT in the acute phase of wound breakdown caused significant skin maceration. Only six case reports have described NPT therapy use in the management of PG wounds. Ghersi et al.,[7] Steenbrugee et al.,[8] Kaddoura and Amm,[9] and Davis et al.[10] all reported the successful use on lower limb wounds, with Niezgoda et al.[11] reporting the successful use of NPT combined and HBO on a chronic superficial lower limb wound. Mandal[1] reported the only previously identified case of NPT used to prepare the wound bed of pathergy following a knee arthroplasty, before a medial gastrocnemius flap was used to achieve soft tissue coverage. We report that before the diagnosis of PG, 5 days of continuous NPT failed to improve the wound. We also report that NPT for a 12-day period after the previously successful soft tissue reconstruction had broken down did not result in improved wound healing.

The evidence for HBO in the management of PG remains sparse. HBO therapy is defined as *"inhalation therapy where the patient breathes 100% oxygen intermittently while inside a chamber at a pressure higher than sea level."* The rational of this therapy promoting wound healing is through; increased tissue oxygenation, reduced oedema, increased fibroblast activity, neovascularisation and bacteriostasis.[12] In a recent review, Tutrone et al.[12] identified only 13 cases in the literature with nine reporting a benefit. None involved pathergy following arthroplasty and none involved a secondary infection following successful flap coverage.

From our experience, a prolonged course of HBO combined with NPT in an immunosuppressed patient reduced the wound size, enhanced wound granulation, promoted re-epithelialisation, and provided high levels of pain relief. The use of NPT in the same patient earlier in their care, before PG diagnosis and immunosuppression, resulted in severe maceration and no benefit. Although as previously stated NPT has been reported as a successful tool in the management of PG ulcers, this was in combination with immunosuppressant treatment.

More studies are necessary to determine the role of both NPT and HBO in the management of pathergy in complex wounds. PG is a rare complication following knee arthroplasty, but should be considered before multiple debridements are performed.

We suggest a surgical algorithm (Figure 4) for managing these difficult cases.

Conflict of interest/funding

None.

Acknowledgements

Dr Marina Morgan, Dr Chris Bowers, Mr Colin Steinlechner.

References

1. Mandal A, Addison P, Stewart K, Neligan P. Vaccum-assisted closure therapy in pyoderma gangrenosum. *Eur J Plast Surg* 2006; **28**:529–31.
2. Wadia F, Malik M, Porter M. Post operative wound breakdown cause by pyoderma gangrenosum after bilateral simultaneous total knee arthroplasty. *J Arthroplasty* 2007;**22**(8):1232–5.
3. Nakajima N, Ikeuchi M, Izumi M, Kuriyama M, Nakajima H, Tani T. Successful treatment of a wound breakdown caused by pyoderma gangrenosum after total knee arthroplasty. *Knee*; 2010; doi:10.1016/j.knee.1010.06.005.
4. Verma SB. Atypical pyoderma following total knee replacement surgery – first report in dermatologic literature. *Ann Brazil Dermatol* 2009;**84**(6):689–91.
5. Armstrong PM, Ilyas I, Pandey R, Berendt AR, Conlon CP, Simpson AH. Pyoderma gangrenosum. A diagnosis not to be missed. *J Bone Joint Surg* 1999;**81**:893–4.
6. de Thomasson E, Caux I. Pyoderma gangrenosum following an orthopaedic surgical procedure. *Orthop Traumatol* 2010;**96**: 600–2.
7. Ghersi MM, Ricotti C, Nousari CH, Newman MI. Negative pressure dressing in the management of pyoderma gangrenosum ulcer. *Arch Dermatol* 2007;**143**(10):1249–51.
8. Steenbrugge F, Raaijmaakers M, Caekebeke P, Van Landuyt K. Pyoderma gangrenosum following trauma of the knee: a case of pathergy and review of orthopaedic cases. *Injury*; 2010; doi: 10.1016/j.injury.2010.06.015.
9. Kaddoura IL, Amm C. A rationale for adjuvant surgical intervention in pyoderma gangrenosum. *Ann Plast Surg* 2001;**46**:23–8.
10. Davis JC, Landeen JM, Levine RA. Pyoderma gangrenosum: skin grafting after preparation with hyperbaric oxygen. *Plast Reconstr Surg* 1987;**79**(2):200–5.
11. Niezgoda JA, Cabigas EB, Allen HK, Simanonok JP, Kindwall EP, Krumenauer J. Managing pyoderma gangrenosum: a synergistic approach combining surgical debridement, vacuum-assisted closure, and hyperbaric oxygen therapy. *Plast Reconstr Surg* 2006;**117**(2):24e–8e.
12. Tutrone WD, Green K, Weinberg JM, Caglar S, Clarke D. Pyogerma gangrenosum: dermatological application of hyperbaric oxygen therapy. *J Drug Dermatol* 2007;**6**(12):1214–9.