Aesthetic Breast Augmentation Mastopexy Followed by Postsurgical Pyoderma Gangrenosum (PSPG): Clinic, Treatment, and Review of the Literature

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Abstract

Introduction Pyoderma gangrenosum (PG) is a rare autoinflammatory neutrophilic ulcerative skin disease, often developing after a trauma or surgical wounds. In the literature there are several reports of post-surgical PG (PSPG) of the breast. The authors of this article experienced an impressive case of PSPG after an aesthetic breast augmentation mastopexy. PSPG is a rare but severe complication in this elective aesthetic surgical procedure.

Method A systematic review of the literature was performed, focusing on PSPG after aesthetic breast surgery (augmentation mammoplasty/mastopexy). The online databases Pubmed, Medline, and Cochrane were used and additionally a Google search was conducted. We compared the data obtained from a systematic literature review to an index case of PSPG after esthetic augmentation mammoplasty.

Results The literature search identified seven articles describing eight cases of PSPG after aesthetic breast surgery. In four of these cases augmentation mammoplasty had been carried out, in two cases mastopexy and in two cases augmentation mammoplasty and mastopexy (augmentation mastopexy). The patient we treated and describe in this paper underwent an augmentation mastopexy outside our clinic. Eight patients suffered from local disease, at the site of surgical wounds, one patient had disseminated disease. Leukocytosis was present in five cases (out of nine). Eight patients had received corticosteroid treatment, one patient refused such treatment. The duration of corticosteroid treatment was on average for 41 days (range 21–60 days). In all cases, the areola had been spared. Complete healing of PSPG was observed on average after 5 months (range 1.5 months–1 year).

Discussion PSPG of the breast after aesthetic breast surgery is rare, but every plastic surgeon should consider this possibility, especially if skin disease develops post-surgery, mimicking wound infection that does not respond to broad-spectrum antibiotic treatment.

Conclusion Although the literature does not recommend this step, implant removal is recommended by the authors because bacterial wound infection normally cannot be ruled out definitely in the early stages of disease. Additional surgical intervention should be limited to the absolute necessary and performed only under adequate systemic immunosuppressive therapy.

Level of Evidence V This journal requires that authors assign a level of evidence to each article. For a full description of these Evidence-Based Medicine ratings, please refer to the Table of Contents or the online Instructions to Authors www.springer.com/00266.
Introduction

Pyoderma gangrenosum (PG) is a rare ulcerative skin disease, which belongs to the spectrum of neutrophilic dermatoses [1] and was first described in 1930 by Brunsting et al. [2]. The incidence is low, estimated at about three cases per million per year in the United States [3]. Pathogenesis includes a misdirected immune response to an underlying disease or trauma. It is associated with several diseases—the most common ones are ulcerative colitis and rheumatoid arthritis (Fig. 1). Diagnostic criteria include the presence of a rapidly progressive, painful necrolytic ulcer with an irregular, violaceous, and undermined border and exclusion of other causes of cutaneous ulceration and the presence of at least one of the following: history suggestive of pathergy or clinical finding of cribriform scarring, systemic diseases associated with PG, histopathologic findings (sterile dermal neutrophilia, mixed inflammation, lymphocytic vasculitis), or treatment response (rapid response to systemic steroid treatment) [1].

As surgeries represent a trauma to the patient’s body, PG can also be triggered by them. This relatively newly described entity of pyoderma is called post-surgical pyoderma gangrenosum (PSPG) and has been reported in recent years, also after breast surgery [4–7]. Our department also experienced an impressive case of PSPG after an elective augmentation mammoplasty and mastopexy.

![Algorithm: clinical diagnostic approach to PSPG](image-url)

Post Surgical Pyoderma Gangrenosum (PSPG)
(augmentation mastopexy). After facing this first case of PSPG ever at our department we did a literature search which showed this to be a relatively rare complication. We focused on elective, aesthetic breast surgery because colleagues working in an outpatient setting should also be aware of this rare complication. Especially when there is no multidisciplinary setting with limited diagnostic and interdisciplinary opportunities.

**Review of the Literature**

**Patients and Methods**

A broad search of the Pubmed, Medline, and Cochrane databases and additionally a Google search was conducted using the terms “Pyoderma gangrenosum breast”, “Pyoderma gangrenosum breast reduction”, “Pyoderma gangrenosum mastopexy”, “Pyoderma gangrenosum mamma”, “Pyoderma gangrenosum mamma augmentation”, “Pyoderma gangrenosum breast augmentation”, “Pyoderma gangrenosum mammoplasty”, “Postsurgical Pyoderma gangrenosum breast”, “Pyoderma gangrenosum breast surgery”, Pyoderma gangrenosum breast reconstruction”, “Pyoderma gangrenosum mamma reconstruction”, which yielded 79 papers, published between 1967 and 2014. The titles and abstracts were screened individually. If the articles seemed relevant or lacked adequate data in the title or abstract to indicate their relevance, the full-text articles were obtained and fully analyzed. Study references were cross-checked and screened manually to identify potential citations not captured by the initial database search. Our search included papers written in English, French, Spanish, and German.

**Selection of Articles**

We focused only on papers in which PSPG was described after augmentation mammoplasty and/or mastopexy with an aesthetic indication. Out of the pool of the primary selected articles, we identified seven articles, in which these surgical procedures where reported and PSPG developed. These articles were fully analyzed.

**Results**

We could identify eight cases (seven articles) of PSPG after aesthetic breast surgery, reported in the literature. In four cases, augmentation mammoplasty had been carried out, in two cases mastopexy and in two augmentation mammoplasty and mastopexy (augmentation mastopexy). The patient whose case is reported in this paper also had augmentation mastopexy and is included in the data evaluation. The average age of patients was 36 years (range 21–56 years); the average time from surgery to development of pyoderma symptoms was 17 days (wide range from 3 days to 5 weeks). The time from initial presentation with symptoms to correct diagnosis lasted from 9 days to about 1 month, but this information is available only in four described cases. All cases showed the typical areola sparing. In eight patients, disease was restricted to the site of surgery, one patient had disseminated disease. Leukocytosis was present in five cases (out of nine), in one case there was definitely no leukocytosis, and three articles gave no information on this point. Fever was reported in four cases. All cases were treated unsuccessfully with antibiotics and had negative bacteriology. All authors did a biopsy, 4 observed an interstitial neutrophilic infiltrate, 1 observed an aspecific inflammation, 1 observed purulent necrosis, and in 3 cases the histopathological appearance is not mentioned exactly. Eight patients received corticosteroid treatment, one patient refused this therapy. The average duration of corticosteroid treatment was 41 days (range 21–days). Only one patient received an additional drug together with corticosteroids. This patient was primarily treated with the immunosuppressive drug cyclosporine; corticosteroids were added secondarily to the therapy regimen because of coexisting ulcerative colitis. Additional surgical intervention together with systemic immunosuppressive therapy at the time of diagnosis of
PSPG was performed in six cases (Fig. 2). Two patients underwent debridement and received tissue coverage (skin grafts and local flap coverage). Our patient, too, received secondary split unmeshed skin grafts. Two patients were additionally treated with Eosin and 1 patient was treated with Integra® (J&J Medical, Skipton, UK). Implants were removed in four out of seven cases. Complete healing of PSPG was observed on average after 5 months (range 1.5 months–1 year). One patient suffered from coexisting ulcerative colitis, two patients had pleural effusion (Fig. 3).

Case Report

A 26-year-old healthy woman, non-smoker, presented at our department with pain and implant exposure on both breasts after an aesthetic breast augmentation mastopexy performed 7 days earlier in a private clinic. There was no fever, but the blood samples showed leukocytosis (22.8 × 10^9/l). Bacterial wound infection was suspected and treatment with broad-spectrum antibiotics was started (moxifloxacin and cefuroxime) (Fig. 4). After removal of both implants under general anesthesia and debridement, the wounds progressively worsened from dehiscence into larger skin ulcers with violet discolored skin and involving subcutaneous tissue. Wound cultures for bacteria, mycobacteria, and fungi were negative and the biopsies taken showed a dense, interstitial neutrophilic infiltrate. Painful wound deterioration was progressive on both breasts sparing both nipple-areola complexes. Lack of success with antibiotic treatment raised suspicion and hardened diagnosis of PSPG so that immunosuppressive therapy with corticosteroids (50 mg/day prednisolone starting dose) was initiated. Fat gauzes with antiseptic solution were used as wound dressings and changed daily. Finally, resolution of wound ulceration could be achieved and pain vanished. Corticosteroid therapy was tapered gradually and stopped after 3 weeks. After remission of PSPG, the patient received tissue coverage with unmeshed split-skin grafts harvested bilaterally from the inner face of the medial thigh, which was of course a salvage procedure to achieve wound closure with the less invasive surgery available (under pre-peri and postoperative immunosuppressive therapy with corticosteroids) to avoid a reactivation of pyoderma (Fig. 5).

Discussion

In 1999 Sotillo-Gago et al. [8] reported one case of PSPG following bilateral augmentation mammoplasty with silicone prostheses. PSPG manifested 5 days postoperatively bilaterally with pustules and ulcers on the surgical wounds. Initially these changes were misdiagnosed as wound infection and treatment was started with antibiotics and surgical debridement was performed. There was no improvement, after removing both prostheses the patient was sent to the authors’ department where PSPG was diagnosed and treatment with cyclosporine was started (5 mg/kg/day).
Forty-five days after beginning of the therapy, a complete healing resulted. Also ulcerative colitis was diagnosed as a coexisting pathology and treated with corticosteroids. In 2004, Pouke et al. [9] published a case of a 41-year-old woman suffering from PSPG after mastopexy (Fig. 6). Three days postoperatively the wounds became red and swollen and the patient was in a serious health condition. Also in this case, bacterial wound infection was suspected and treated with high-dose antibiotics and hyperbaric oxygen. As there was no improvement in the clinical course, the authors suspected PSPG and treated the patient with high-dose corticosteroids (40 mg methylprednisolone three times a day intravenously). They also administered additionally fluconazole and piperacillin/tazobactam to treat possible fungal and bacterial infections. Careful wound care was done, and after decrease of the inflammatory parameters, the ulcerated wound surface was treated with Integra® (Johnson and Johnson Medical, Skipton, UK), a template that acts as a bilayer matrix that provides a scaffold for dermal regeneration. Corticosteroids were continued for 2 months.

In 2007 Ouazzani et al. [10] reported a case of a 21-year-old woman who underwent bilateral augmentation mammoplasty with silicone implants because of Poland Syndrome. After 6 days wound dehiscence occurred, and the patient developed low-grade fever and PG. The clinical course continued to worsen and 25 days postoperatively the implants were exposed. The authors did surgical revision, but there was no improvement and re-exposure of the implants was noted. At this point, they suspected PSPG, and started treatment with methylprednisolone (32 mg/day, intravenously) and covered the wound defects with local flaps taken from the aponeurosis of the rectus abdominis. The implants were removed in this case. Corticosteroid treatment was tapered gradually and stopped after 4 weeks. In 2008 Bonamigo et al. [11] published a case of a patient in whom PSPG developed after facial plastic surgery and augmentation mammoplasty with silicone prosthesis. Ulcerating PSPG developed on both sites, in the face and on the breasts, beginning 5 days postoperatively. Since there was no improvement after extensive antibiotic therapy with ciprofloxacin, ampicillin, sulbactam, oxacillin, and linezolid, PSPG was suspected. Surgical debridement was performed, the silicone implants were removed and treatment was initiated with prednisone (70 mg/day) and dapsone (100 mg/day). Under this therapy, 4 months later, the ulcers healed.

Duval et al. [12] published a case of PSPG after bilateral mastopexy in 2011 in a 28-year-old woman. Also in this case initially bacterial wound infection was suspected; however, antibiotic treatment and surgical debridement proved to be unsuccessful. The patient then was transferred to the authors’ department where PSPG was diagnosed; treatment with corticosteroids was started (1 mg/kg/day) and surgical debridement was carried out once again. As there was improvement in the clinical course within 24 h, the dose of corticosteroid was slightly reduced and treatment was stopped after 1.5 months. The patient had

Fig. 6 One year post PSPG—Re-mastopexy without implant with first partial removal of the skin grafted area on both breasts (3 weeks post OP)
<table>
<thead>
<tr>
<th>Authors and year published</th>
<th>Patient age</th>
<th>Surgical procedure</th>
<th>Location (local vs. disseminated)</th>
<th>Areola sparing</th>
<th>Time from surgery to presentation</th>
<th>Time from initial presentation to right diagnosis</th>
<th>Leukocytosis</th>
<th>Fever</th>
<th>Unsuccessful Antibiotic treatment</th>
<th>Bacteriology</th>
<th>Biopsy</th>
<th>Corticosteroid treatment</th>
<th>Duration of corticosteroid treatment</th>
<th>Other systemic, immunosuppressive therapy</th>
<th>Additional surgical intervention together with systemic immunosuppressive therapy at time of right diagnosis</th>
<th>Implant removal</th>
<th>Duration from right therapy start until complete healing</th>
<th>Coexisting ulcerative colitis</th>
<th>Pleural effusion</th>
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</thead>
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<tr>
<td>Sotillo-Gago et al. (1999)</td>
<td>43</td>
<td>Augmentation</td>
<td>Local—breast</td>
<td>yes</td>
<td>5 days</td>
<td>Not mentioned</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Negative</td>
<td>Yes—not mentioned</td>
<td>No</td>
<td>60 days</td>
<td>Cyclosporine</td>
<td>Debridement and skin grafts Debridement and local flap coverage</td>
<td>Yes</td>
<td>1.5 months</td>
<td>Yes</td>
<td>No</td>
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<td>Poucke et al. (2004)</td>
<td>41</td>
<td>Mastopexy</td>
<td>Local—breast</td>
<td>yes</td>
<td>3 days</td>
<td>Not mentioned</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Positive</td>
<td>Yes—not reported</td>
<td>No</td>
<td>30 days</td>
<td>Integra® (J&amp;J Medical, Skipton, UK) Debridement and local flap coverage</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
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<td>Ouazzani et al. (2007)</td>
<td>21</td>
<td>Augmentation</td>
<td>Local—breast and face</td>
<td>yes</td>
<td>6 days—face</td>
<td>About 1 month</td>
<td>No</td>
<td>Yes</td>
<td>No</td>
<td>Negative</td>
<td>Yes—not reported</td>
<td>No</td>
<td>30 days</td>
<td>No—refused</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
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<td>Bonamigo et al. (2008)</td>
<td>56</td>
<td>Mastopexy</td>
<td>Local—breast</td>
<td>yes</td>
<td>3 days</td>
<td>About 15 days</td>
<td>Yes</td>
<td>No</td>
<td>No</td>
<td>Negative</td>
<td>Yes—not reported</td>
<td>No</td>
<td>45 days</td>
<td>No—refused</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
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<td>Duval et al. (2011)</td>
<td>28</td>
<td>Augmentation</td>
<td>Local—breast</td>
<td>yes</td>
<td>5 weeks</td>
<td>About 14 days</td>
<td>Yes</td>
<td>No</td>
<td>No</td>
<td>Negative</td>
<td>Yes—not reported</td>
<td>No</td>
<td>About 7 weeks</td>
<td>No—refused</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
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<td>González de Vicente et al. (2011)</td>
<td>32</td>
<td>Augmentation Mastopexy and Mastopexy (augmentation mastopexy)</td>
<td>Local—breast</td>
<td>yes</td>
<td>3 weeks</td>
<td>Not mentioned</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>Negative</td>
<td>Yes—not reported</td>
<td>No</td>
<td>About 7 weeks</td>
<td>No—refused</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
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<tr>
<td>Gonzalez de Vicente et al. (2011)</td>
<td>36</td>
<td>Augmentation</td>
<td>Disseminated—breast and shins</td>
<td>yes</td>
<td>1 month—breast and months—shins</td>
<td>Not mentioned</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>Negative</td>
<td>Yes—not reported</td>
<td>No</td>
<td>No—refused</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td></td>
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<tr>
<td>Apalla et al. (2013)</td>
<td>39</td>
<td>Augmentation</td>
<td>Local</td>
<td>yes</td>
<td>7 days</td>
<td>Not mentioned</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>Negative</td>
<td>Yes—not reported</td>
<td>No</td>
<td>No—refused</td>
<td>Secondary unmeshed split skin</td>
<td>No</td>
<td>No</td>
<td>No</td>
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</table>

Table 1 Systematic analysis of literature
complete healing after 4.5 months. González de Vincente et al. [13] reported in a Spanish article in 2011 two cases of PSPG. Both patients had undergone surgical procedures, which were in fact revisions of augmentation mammoplasties, done years ago in other departments (Table 1). One was a 32-year-old woman who was treated with augmentation mammoplasty and areolar and slight mammary reduction and the other patient was a 36-year-old woman who received mastopexy and implant replacement. In the first case reported, the first four postoperative weeks were uneventful and in the fifth week blistering and PSPG developed. Antibiotic therapy resulted in no improvement and PSPG was diagnosed and treated with corticosteroids (initial dose 40 mg/day). Corticosteroid therapy could be stopped after about 7 weeks. In the second reported case, PSPG developed 3 weeks postoperatively. PSPG was diagnosed and treated the same way as the other case with corticosteroids. Healing was accomplished in both patients after 2 months of therapy. Apalla et al. [14] reported in 2013 a case of disseminated PSPG involving parts of the body other than the surgical sites. A 39-year-old woman presented to them with ulcerating lesions on the surgical wounds of the breasts appearing 1 month after silicone breast implant procedure. She also complained of mild arthralgia and had a history of ineffective antibiotic therapy. Two months after surgery she also developed progressively tender, ulcerating pustules on the shins. Bacteriology was negative and PSPG was suspected. The patient refused any treatment and disease improved gradually over the next 6 months.

PSPG of the breast after aesthetic breast surgery is rare, but the surgeon should keep this possibility in mind, especially if skin disease develops shortly after surgery mimicking wound infection that does not respond to broad-spectrum antibiotic treatment. As mentioned in the introduction, diagnostic criteria also include histopathological findings, rapid response to steroids, systemic diseases associated with PG and cribriform scarring. These points, however, can only be analyzed retrospectively and since treatment needs to be initiated immediately, it might not be wise to wait too long for findings from histopathology and bacteriology. So if these criteria are excluded, the surgeon’s diagnosis is in fact based on the clinical appearance and lack of response to antibiotics. Clinical appearance also differs among patients; Powell and Collins [15] reported four different variants of PG, namely an ulcerative, bullous, pustular, and vegetative form.

To summarize, based on the characteristics identified in all the cases analyzed in this review, the primary prospective diagnostic approach to PSPG of the breast is as follows: skin changes with areola sparing near the surgical wound mimicking bacterial infection, no response to antibiotic treatment, immediate response to corticosteroid therapy.}

<table>
<thead>
<tr>
<th>Authors and year published</th>
<th>Other Symptoms</th>
<th>Arthralgia</th>
</tr>
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<tbody>
<tr>
<td>Poucke et al. (2004)</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Ouazzani et al. (2007)</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Bonamigo et al. (2008)</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Duvale et al. (2011)</td>
<td>No</td>
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<tr>
<td>Gonzalez de Vicente et al. (2011)</td>
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<td>No</td>
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<td>Gonza´lez de Vicente et al. (2011)</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Apalla et al. (2013)</td>
<td>No</td>
<td>No</td>
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Table 1 continued |
Once diagnosed, the treatment of choice is starting therapy with corticosteroids. The initial dose, application form (oral or intravenous), schedule of steroid tapering and steroid derivatives used were different, as reported in the literature. It would appear that these specific aspects of steroid administration can be neglected in the clinical setting, especially because all patients analyzed responded immediately to steroids and showed disease improvement. If breast implants should be removed or left is a challenging question. Literature analysis revealed implant removal in four out of seven cases. Apalla et al. [14] suggest that implants do not play a significant role in the development of PSPG because there were no cases in the literature in which implant removal was followed by disease improvement. This hypothesis may be right and implant removal might be ineffective to treat PSPG itself, but as PSPG is an exclusion diagnosis, implant removal should be done, because normally a potential bacterial wound infection cannot be ruled out definitely in early stages of the disease. If PSPG can be ascertained, then proceeding without implant removal would seem reasonable, however, if there is any doubt then implants should be removed as a precaution and secondary breast reconstruction should be done if necessary after PSPG is under control. We think that an underlying implant especially in a breast augmentation mastopexy with implant can be the cause of an additional wound tension contributing further development of a PSPG. Additional surgical intervention together with systemic immunosuppressive therapy was initiated once PSPG was diagnosed in 6 cases (out of 9).

Conclusion

According to our experience, once PSPG has developed, surgical interventions should be limited to the absolute necessary in the early stages of the disease and only performed after control with corticosteroids. Surgery should be minimally invasive, as further traumatization might re-activate PSPG. Secondary surgical interventions after complete healing would be the most preferable approach in patients developing PSPG, but always under immunosuppressive therapy due to the fact that PSPG has a high tendency of recurrence once diagnosed in a patient. Antibiotics should be added in order to prevent any kind of bacterial infection in these high risk patients. It is also important to keep in mind that this immune reaction can also appear at the donor sites of e.g., split-skin grafts (used for defect closure) or at other sites of the body, thought to be never involved in the disease. Nevertheless, we suggest doing these surgeries, if these, often young, patients suffer from sequelae of PSPG and have a disastrous result without any further surgical intervention.

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References