

MUCORMYCOTIC LATE SEROMA AFTER POLAND SYNDROME CORRECTION WITH IMPLANT-ENHANCED LATISSIMUS DORSI FLAP – A CASE REPORT

Maria Giuseppina Onesti, Davide Marino, Alice Patrignani,
Diego Ribuffo

*Department of Plastic Reconstructive and Aesthetic Surgery, Policlinico Umberto I, Sapienza
University of Rome, Rome, Italy*

Summary

Introduction. Breast malformation correction with implants is a common procedure, and a potential complication is the development of a late seroma, defined as occurring more than one year post-implantation. Etiologies include infectious, auto-inflammatory, and neoplastic factors. This case report presents an unusual case of seroma due to mycosis emerging 15 years after amastia correction using a latissimus dorsi flap (LD) and implants.

Materials and methods. In 2006, a 44-year-old patient underwent amastia correction with an LD flap and implants in Ecuador. She presented with sudden breast volume increase, tension, and pain in the left breast. Urgent intervention involved implant removal, collection evacuation, and total capsulectomy. Cytological/histological analysis ruled out neoplasia, while microbiological examination identified *Mucorales* as the cause. After an uneventful postoperative period, a definitive breast prosthesis was implanted two years later, following ultrasound evaluation and inflammation indices.

Discussion. *Mucorales* are opportunistic pathogens, particularly in immunocompromised or uncontrolled diabetes patients, causing infections in sinuses, brain, and lungs. Mucormycosis linked to breast implants in immunocompetent individuals is rare and poorly defined in literature.

Conclusions. Late seroma onset post-breast implants is rare, and a prompt differential diagnosis is crucial due to potential malignancies like BIA-ALCL. Mucormycosis, a potentially lethal condition, requires medical and sometimes surgical intervention. The incidental discovery of *Mucorales* infection facilitated complete eradication of the pathology, resulting in full recovery.

Key words: mucormycosis, Poland syndrome, breast reconstruction, breast surgery

Received: August 5, 2024
Accepted: September 26, 2024
Published online: September 30, 2024

Correspondence

Davide Marino

E-mail: davide.marino@uniroma1.it

How to cite this article: Onesti MG, Marino D, Patrignani A, et al. Mucormycotic late seroma after Poland syndrome correction with implant-enhanced latissimus dorsi flap – A case report. PRRS 2024;2:46-49. <https://doi.org/10.57604/PRRS-606>

© Copyright by Pacini Editore Srl



OPEN ACCESS

This is an open access article distributed in accordance with the CC-BY-NC-ND (Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International) license. The article can be used by giving appropriate credit and mentioning the license, but only for non-commercial purposes and only in the original version. For further information: <https://creativecommons.org/licenses/by-nc-nd/4.0/deed.en>

INTRODUCTION

Poland syndrome (PS) is a congenital disorder characterized by a series of

defects in the embryonic development of the breast, thoracic wall, and the ipsilateral upper limb¹. It may manifest with a broad spectrum of morphological alterations, from mild to severe. In females, it generally affects the breast parenchyma with hypoplasia or aplasia, while the nipple-areolar complex may be hypoplastic or absent¹. The correction of this condition is challenging and is primarily aimed at improving the contour of the breast mound and chest wall². Numerous corrective strategies have been described over the years, ranging from implant-based to autologous techniques^{2,3}. The first may use standard breast implants or custom-made pectoral implants, while the second include fat grafting procedures, latissimus dorsi flap, and different types of free flaps such as abdominal-based or thigh-based flaps³. Recently, FALD flap has been introduced, enabling reconstruction with an adequately thick flap, while avoiding the use of prostheses and their potential complications⁴. Similarly to any surgical procedure, there are several potential complications, ranging from common issues like hematoma or infection, to more uncommon occurrences such as Breast Implant-Associated Anaplastic Large Cell Lymphoma (BIA-ALCL)⁵. In this study, we analyze the surgical history of a woman with PS who has developed a late seroma 23 years after corrective surgery. She underwent to urgent breast implant explantation, with microbiological findings suggesting a Mucorales infection.

CASE PRESENTATION

A 29-year-old Ecuadorian woman affected by Poland syndrome (PS), without other specific comorbidities, underwent corrective surgery in 2008. She presented with left-sided aplasia of the pectoralis major, pectoralis minor, and absence of the ipsilateral breast gland. The operative strategy consisted in harvesting a latissimus dorsi (LD) flap to cover a unilateral breast implant positioned on the thoracic wall. 13 years after surgery, following an accidental fall, the patient developed a late seroma, spreading within the previously treated breast mound. On the 20th day after the fall, she sought emergency care due to discomfort and increased volume in the left hemithorax.

Ultrasound imaging revealed a periprosthetic fluid collection. She was initially treated with conservative management consisting of antibiotics and pain relievers over a course of a few days which saw a partial improvement, which warranted her discharge with oral treatments. Over the subsequent 30 days, the inflammation did not subside and periprosthetic effusion volume increased (Fig. 1), accompanied by complaints of significant pain, leading to a second visit to the emergency department. This time, the patient was admitted in the Plastic Surgery Department of our institution. An ultrasound-guided



Figure 1. Preoperative picture of the patient at the time of hospital admission, showing evidence of inflammation and periprosthetic effusion.

sampling of the periprosthetic fluid with cytological examination was conducted as per guidelines^{6,7} on the first day of admission. However, the clinical evaluation of the patient indicated the need for emergency surgery the following day. The intraoperative results and those from the needle aspiration were received simultaneously and showed overlapping findings. The patient did not undergo a preparatory MRI due to the high risk of skin tearing. Urgent surgery was deemed necessary to mitigate the risk of skin disruption and potential implant exposure. This resulted in explantation surgery with en-block capsulectomy. During the implant removal the pocket has been inspected, to search for any necrotic tissue. Fortunately, none was found, eliminating the need for further tissue debridement. Additionally, the latissimus dorsi muscle has undergone fibroadipose involution.

Due to the deterioration of the prosthesis, identifying the make and model was not possible. Samples of the periprosthetic fluid collection were sent for microbiological examination as well as cytological analysis, along with the periprosthetic capsule which was sent for histological and immunohistochemical analysis. Diagnostic exams returned negative for malignancies but were positive for a fungal infection of the Mucor spp.

Patient's history was negative for penetrating trauma to the interested breast side. The fungal contamination is believed to have occurred during the initial surgery when amastia performed, while the accidental fall led to the spread of seroma.

Following surgery, infective diseases consultancy was requested which deemed antifungal unnecessary therapy due the absence of systemic and local symptoms.



Figure 2. Postoperative picture, two years after the explant, with no signs of inflammation and/or infection.



Figure 3. Postoperative picture 3 months after reconstruction with prosthesis and contralateral mastopexy.

The post-operative course was uneventful with resolution of inflammatory signs and pain. Patient returned every 3 months for scheduled follow-up visits where she received chest ultrasound scans and monitoring of inflammatory markers, to exclude recurrence of seroma and infection. Two years after initial presentation (Fig. 2), reconstructive surgery planning became feasible. In September 2023, the patient underwent received the submuscular placement of a 175cc smooth round breast implant (Mentor, Johnson & Johnson, New Brunswick, New Jersey, USA) associated with 200 cc fat transfer injected in all aspects of the breast mound to enhance the breast contour. In the same setting, she also received contralateral balancing with a mastopexy. Post-operative course was uneventful, and the patient returned and scheduled follow-ups, the latest of which was at 3 months, where she expressed satisfaction with the current results (Fig. 3).

DISCUSSION

The surgical treatment of PS requires meticulous planning, especially in women, where restoring the harmony

of a physiological breast mound is crucial. When planning for surgery, patients should always be informed that any surgical procedure may be associated with specific complications, either immediate or delayed⁸⁻¹⁰. Late-onset seromas in patients with breast implants often raise red flags as they are the most common presentation of BIA-ALCL⁹, as well as the BIA-SCC¹¹, newly described entity.

Nevertheless, reactive causes (including infections) are far more common than malignant ones¹². In our case, we speculate that the patient sustained a direct trauma to the breast, potentially representing the breach through which contamination might have occurred. Typically, these processes are sustained by bacterial agents that commonly constitute the skin flora, more rarely by fungal agents^{10,12}. However, our patient sustained an infection from an unlikely culprit which was mucormycosis. This infection is caused by fungi of the Mucorales order, which usually result in opportunistic infections, primarily affecting individuals who are immunocompromised, diabetic, or have predisposing factors such as chemotherapy or organ transplant recipients¹⁰. In our case, the patient was young and healthy, with no relevant personal or family history. Generally, mucormycosis affects the airways, central

nervous system and skin, giving rise to a destructive inflammatory process which frequently requires aggressive debridement procedures for eradication¹³. The absence of risk factors, as seen in the presented case, make this occurrence quite unique. Of note, our patient received final reconstruction after a long period of time. Indeed, because of the known aggressiveness of mucormycosis and the high rates of recurrence¹⁴, we have decided to err on the side of caution, delaying reconstruction in accordance with the patient.

Due to the higher susceptibility of macrotexturized breast implants to infections, then a smooth breast implant has been employed for the delayed reconstruction⁸.

CONCLUSIONS

Mucormycosis is a condition with a natural history that is challenging to predict. Treatment is often elusive and sometimes non-resolving. The occurrence as a late seroma in a patient without risk factors is an unusual event, adding complexity to the diagnosis and, consequently, the treatment. When a severe infective complication is suspected, erring on the side of caution and ensuring patient safety is paramount. Following our approach, mucormycosis was resolved and the patient received optimal aesthetic results despite the delay. As the field of plastic and reconstructive surgery continues to advance, ongoing research and technological innovations promise even more refined and effective solutions for addressing the challenges associated with PS treatment and its possible complications.

Conflict of interest statement

The authors declare no conflict of interest.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Author contributions

OMG: A

MD: S

PA: W

RD: O (supervision and review)

Abbreviations

A: conceived and designed the analysis

D: collected the data

DT: contributed data or analysis tool

S: performed the analysis

W: wrote the paper

O: other contribution (specify contribution in more detail)

Ethical consideration

Not applicable.

References

- 1 Delay E, Nachaoui H, Frobert P. Syndrome de Poland [Poland's syndrome]. *Ann Chir Plast Esthet* 2022;67:358-373. <https://doi.org/10.1016/j.anplas.2022.07.020>
- 2 Chiummariello S, Pica A, Guarro G, et al. Poland syndrome: an algorithm to select the appropriate chest wall surgical reconstructive treatment. *Ann Ital Chir* 2014;85:237-243.
- 3 Seyfer AE, Fox JP, Hamilton CG. Poland syndrome: evaluation and treatment of the chest wall in 63 patients. *Plast Reconstr Surg* 2010;126:902-911. <https://doi.org/10.1097/PRS.0b013e3181e60435>
- 4 Santanelli di Pompeo F, Sorotos M, Paolini G, et al. The use of Fat-Augmented Latissimus Dorsi (FALD) flap for male Poland syndrome correction: a case report. *Case Reports Plast Surg Hand Surg* 2022;9:197-202. <https://doi.org/10.1080/23320885.2022.2117701>
- 5 Matar DY, Wu M, Haug V, et al. Surgical complications in immediate and delayed breast reconstruction: a systematic review and meta-analysis. *J Plast Reconstr Aesthet Surg* 2022;75:4085-4095. <https://doi.org/10.1016/j.bjps.2022.08.029>
- 6 Longo B, Di Napoli A, Curigliano G, et al. Clinical recommendations for diagnosis and treatment according to current updated knowledge on BIA-ALCL. *Breast* 2022;66:332-341. <https://doi.org/10.1016/j.breast.2022.11.009>
- 7 Kazzam ME, Ng P. Postoperative Seroma Management. [Updated 2023 Aug 14]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing;2024 Jan (<https://www.ncbi.nlm.nih.gov/books/NBK585101>).
- 8 Meresse T, Chaput B, Grolleau JL, et al. Complication des lambeaux en reconstruction mammaire [Complications of autologous breast reconstruction]. *Ann Chir Plast Esthet* 2019;64:594-619. <https://doi.org/10.1016/j.anplas.2019.07.015>
- 9 Paolini G, Firmani G, Briganti F, et al. Assessment of risk factors for rupture in breast reconstruction patients with macrotextured breast implants. *Aesthetic Plast Surg* 2023;47:517-530. <https://doi.org/10.1007/s00266-022-03118-9>
- 10 Steinbrink JM, Miceli MH. Mucormycosis. *Infect Dis Clin North Am* 2021;35:435-452. <https://doi.org/10.1016/j.idc.2021.03.009>
- 11 D'Orsi G, Giacalone M, Calicchia A, et al. BIA-ALCL and BIA-SCC: updates on clinical features and genetic mutations for latest recommendations. *Medicina (Kaunas)* 2024;60:793. <https://doi.org/10.3390/medicina60050793>
- 12 Washer LL, Gutowski K. Breast implant infections. *Infect Dis Clin North Am* 2012;26:111-125. <https://doi.org/10.1016/j.idc.2011.09.003>
- 13 Dessy LA, Corrias F, Marchetti F, et al. Implant infection after augmentation mammoplasty: a review of the literature and report of a multidrug-resistant *Candida albicans* infection. *Aesthetic Plast Surg* 2012;36:153-159. <https://doi.org/10.1007/s00266-011-9777-x>
- 14 Santanelli di Pompeo F, Clemens MW, Atlan M, et al. 2022 Practice Recommendation Updates from the World Consensus Conference on BIA-ALCL. *Aesthet Surg J* 2022;42:1262-1278. <https://doi.org/10.1093/asj/sjac133>