

Surgical Correction of Nipple Hypertrophy after Nipple-sparing Mastectomy with Breast Reconstruction

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Summary: Nipple hypertrophy is a relatively common phenomenon, particularly in the Asian patient population. The incidence and prevalence or cause of nipple hypertrophy are not well defined in the literature. As survival rates for breast cancer patients continue to improve, there is an increasing emphasis on enhancing their quality of life. Treatment options, such as lumpectomy and radiation therapy or mastectomy, now prioritize preservation of the nipple-areolar complex (NAC) through techniques like nipple-sparing mastectomy (NSM). This approach has been shown to improve patient satisfaction and quality of life. However, it is important to note that NSM is associated with certain complications, including NAC necrosis, malposition, and local recurrence of the tumor. Among those complications, nipple hypertrophy is quite rare. In this report, we present a case of nipple hypertrophy that developed after breast reconstruction, using autologous tissue. The patient, a 48-year-old woman, underwent NSM for breast cancer, and had a tissue expander placed simultaneously. Following tissue expansion, breast reconstruction with a deep inferior epigastric perforator (DIEP) flap was performed. However, during the tissue expansion phase, the patient's nipple gradually enlarged, and the protrusion became more pronounced after the DIEP flap transfer. Nipple reduction surgery was subsequently performed for both diagnostic and cosmetic purposes. We present this case along with a review of relevant literature. (*Plast Reconstr Surg Glob Open* 2023; 11:e5134; doi: [10.1097/GOX.0000000000005134](https://doi.org/10.1097/GOX.0000000000005134); Published online 17 July 2023.)

A 48-year-old woman presented with right breast cancer in the B region, diagnosed as mucinous carcinoma. Preoperative imaging revealed the tumor to be 4 cm away from the nipple and measuring 20×13×25 mm. NSM was performed, and intraoperative frozen section confirmed the preoperative diagnosis. Neither the surgical margins nor the sentinel lymph node biopsy were positive for malignancy. Immediate breast reconstruction with a tissue expander was performed, placing the expander in a subpectoralis major muscle pocket and covering it with the muscle in a standard manner. No artificial materials such as acellular dermal matrix were used. The postoperative diagnosis was

mucinous carcinoma, and negative surgical margins were confirmed. No lymph node metastasis was identified. As the HER2 score was 1+, no adjuvant therapy or chemotherapy was administered. The postoperative wound healing was uneventful. Expansion of the tissue expander took place over a period of 3 months, during which the preserved nipple began to grow gradually, which seemed to be within normal physiological response. After full expansion, the tissue expander was removed, and breast reconstruction with a DIEP flap was performed. No unusual events occurred during or after surgery, with the exception of the growing nipple. The nipple continued to grow even after the autologous tissue transfer. At 9 months postoperatively, the nipple size was evident when compared with the normal side (Fig. 1). The shape of the nipple was spherical, and its size exceeded 2 cm in both diameter and height. Wedge-shaped reduction surgery was performed for both pathological diagnosis and aesthetic

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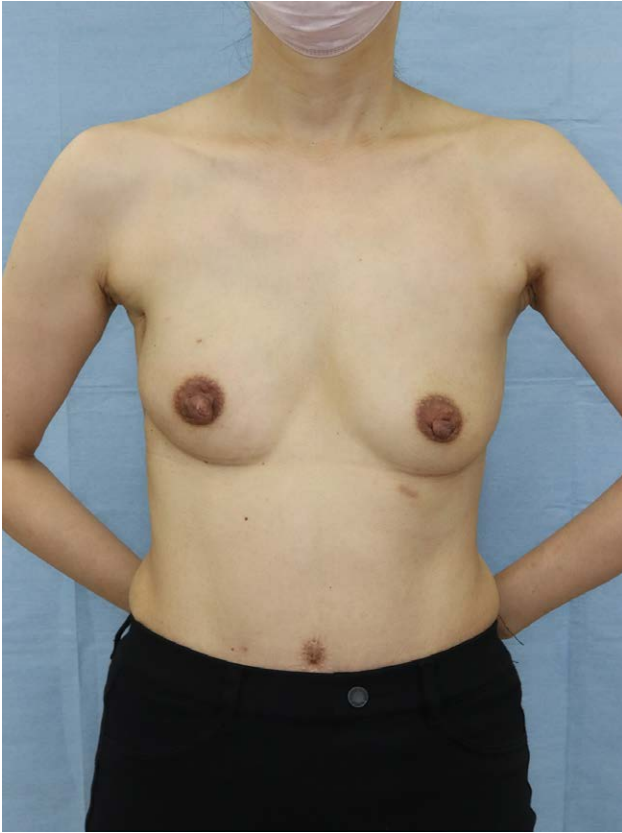


Fig. 1. This 48-year-old woman showed nipple hypertrophy after NSM and reconstructive surgery on her right breast. View of the breast 9 months after the reconstruction with DIEP flap.



Fig. 2. Intraoperative view of nipple reduction. Wedge-shaped excision was performed.

improvement (Fig. 2). The histopathological diagnosis was complex sclerosing lesion with multiple small epidermal cysts (Fig. 3). The patient was satisfied with the outcome of the reduction surgery and reported no loss of sensation during follow-up. Figure 4 shows the postoperative appearance of the nipple.

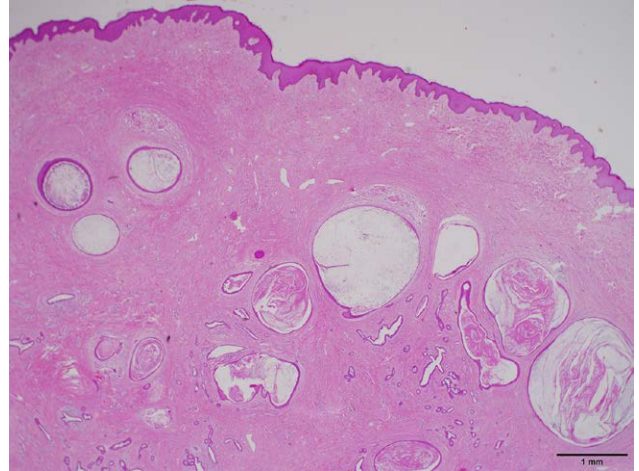


Fig. 3. Histopathological image of the resected specimen. Complex sclerosing lesion with multiple small epidermal cysts was seen.

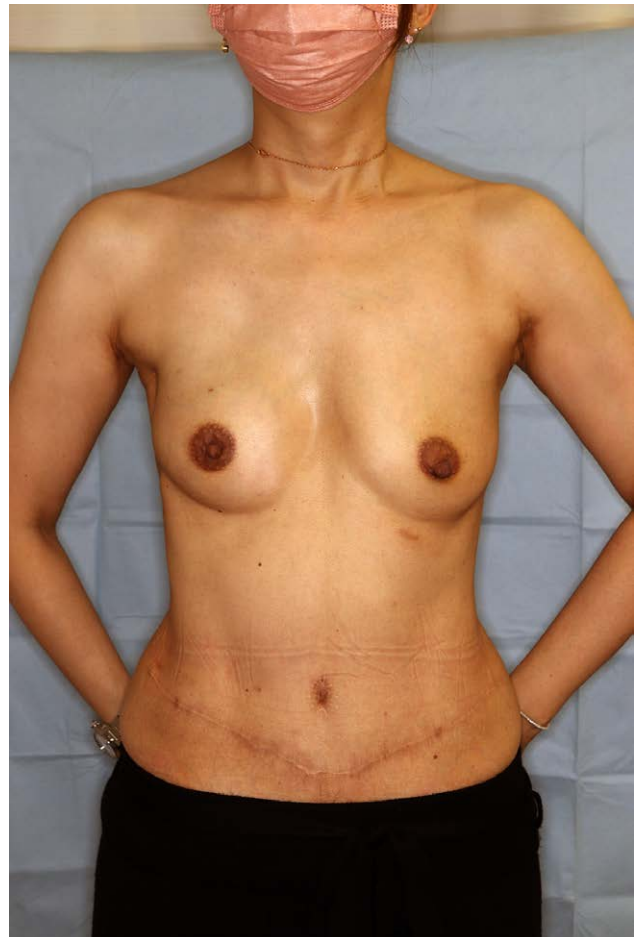


Fig. 4. Postoperative view of the breast one year and 6 months after nipple reduction.

DISCUSSION

There have been significant changes in the surgical treatment methods for breast cancer. (See figure, Supplemental Digital Content 1, which displays the types

of mastectomies: (A) Halsted radical mastectomy, (B) modified radical mastectomy, (C) total mastectomy, (D) skin-sparing mastectomy, (E) nipple-sparing mastectomy, (F) partial mastectomy. <http://links.lww.com/PRSGO/C668>.) Despite concerns about the potential risks of residual breast tissue harboring occult malignancy or future cancer, mounting evidence supports the oncological safety of NSM.^{1,2} Headon et al recommend that for NSM to be performed, tumors should be peripherally located, smaller than 5 cm in diameter, located more than 2 cm away from the nipple margin, and human epidermal growth factor 2–negative.² In the case reported in this article, the tumor was located 4 cm away from the nipple and was less than 5 cm in size.

Nipple necrosis is a common complication of NSM.³ As a result, growth of the nipple after NSM is unusual and requires consideration of tumor recurrence. On the other hand, physiological nipple swelling is common in Asian women, but not in White women.^{4,5} In the Asian population, bilateral nipple hypertrophy is not necessarily indicative of malignancy. However, unilateral nipple hypertrophy arising in the side that has a history of NSM is rare and should be evaluated carefully.

Macrothelia, or nipple hypertrophy, usually occurs after puberty and becomes more prominent after pregnancy. Hormonal changes during breastfeeding are considered to be a cause of this condition. However, there is limited literature on the epidemiology and cause of this deformity. Although most literature describes it as an aesthetic problem, it is also an oncological concern if it occurs on the side that has undergone NSM. In this case, the nipple enlarged slowly and had a smooth, spherical, regular shape, both of which were different from the typical features of malignancy, allowing for conservative follow-up. Nevertheless, careful and frequent follow-up is essential if the nipple starts to enlarge after NSM, even if it seems benign.

Although there is no formal definition of nipple hypertrophy,⁶ Lai et al proposed that the normal female nipple is approximately 1 cm in diameter, with an almost equal amount of anterior projection.⁷ Various techniques for nipple reduction have been described in the literature.^{4,5,7,8} These techniques can generally be divided into two groups: those that involve excision of the core of the nipple and those that involve circumcision of the nipple. In this case, we chose to perform reduction surgery using a wedge-shaped excisional design. One of the reasons for this choice is the possibility of malignancy. We were also concerned about preserving the blood supply to the nipple, as the NSM might have damaged the subdermal vascular plexus. Moreover, the patient no longer needed to lactate. We aimed to design inferior-based wedge-shaped resection to avoid compromising the vascular and neural networks. The patient was satisfied with the outcome.

Currently, the detailed mechanism of hypertrophic nipple formation is not well understood, making it difficult to conclude the reason for the occurrence of nipple hypertrophy in this case. As far as we are aware, this is the first report of a hypertrophic nipple arising from the

reconstructed breast that had undergone NSM. Given that the patient did not undergo any adjuvant therapies or chemotherapy, it is unlikely that these treatments had any impact on the development of nipple hypertrophy. We hypothesize that the resection of tissue located below the NAC and the compression by the tissue expander or DIEP flap may have reduced the local blood circulation to the nipple, leading to ischemia or congestion, edema, and inflammation, which may have resulted in hypertrophy. The growth of the nipple became apparent after DIEP flap transfer, suggesting that the flap may have played a role in this case. It is known that ischemia/reperfusion injury occurs during free flap transfer.⁹ Cytokines, growth factors, free oxygen radicals or nitric oxide, etc released from the flap may have induced the growth of the nipple. Alternatively, the multiple epidermal cysts could potentially account for the etiology of nipple hypertrophy.¹⁰

In conclusion, as NSM gains popularity, due to the concern of residual breast tissue harboring hidden malignancy or increasing the risk of future cancer, careful follow-up is crucial. Special care should be taken for the treatment of nipple hypertrophy after NSM, because blood supply to the nipple might have been compromised.

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DISCLOSURE

The authors have no financial interests to declare in relation to the content of this article.

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Data were used in accordance with the Declaration of Helsinki. Written informed consent was obtained from the patient for publication of this case report and any comparing images. The local ethics committee of Hyogo Medical University declared that ethical approval is not required for a case report.

REFERENCES

1. Costeira B, Silva FB, Oom R, et al. Locoregional recurrence in skin-sparing and nipple-sparing mastectomies. *Jso*. 2022;125:352–360.
2. Headon HL, Kasem A, Mokbel K. The oncological safety of nipple-sparing mastectomy: a systematic review of the literature with a pooled analysis of 12,358 procedures. *Archives Plastic Surg*. 2016;43:328–338.
3. Colwell AS, Tessler O, Lin AM, et al. Breast reconstruction following nipple-sparing mastectomy. *Plast Reconstr Surg*. 2014;133:496–506.
4. Marita A, Budiman B. Modified radial incision technique in reduction nipple hypertrophy. *Jurnal Plastik Rekonstruksi*. 1970;1:452–459.
5. Cheng MH, Smartt JM, Rodriguez ED, et al. Nipple reduction using the modified top hat flap. *Plast Reconstr Surg*. 2006;118:1517–1525.

6. Fanous N, Tawile C, Fanous A. Nipple reduction—an adjunct to augmentation mammoplasty. *Plast Surg-chir Plas.* 2009;17:81–88.
7. Lai YL, Wu WC. Nipple reduction with a modified circumcision technique. *Brit J Plast Surg.* 1996;49:307–309.
8. Regnault P. Nipple hypertrophy: a physiologic reduction by circumcision. *Clin Plast Surg.* 1975;2:391–396.
9. Siemionow M, Arslan E. Ischemia/reperfusion injury: a review in relation to free tissue transfers. *Microsurg.* 2004;24:468–475.
10. Naftali YB, Shoufani A, Krausz J, et al. Unusual presentation of epidermoid cyst mimicking breast cancer involving the areola—case report. *Int J Surg Case Rep.* 2018;51:17–20.