



CASE REPORT

Cryosurgery as a Minimally Invasive Alternative Treatment for a Patient with Erosive Adenomatosis of the Nipple

Kyung-Nam Bae^{1,2}, Kihyuk Shin¹, Woo-Il Kim¹, Min-Young Yang¹, Won-Ku Lee¹, Hoon-Soo Kim^{1,2}, Hyun-Chang Ko¹, Byung-Soo Kim^{1,2}, Moon-Bum Kim^{1,2}, Gun-Wook Kim³

¹Department of Dermatology, School of Medicine, Pusan National University, ²Biomedical Research Institute, Pusan National University Hospital, ³Department of Dermatology, College of Medicine, Dong-A University, Busan, Korea

Erosive adenomatosis of the nipple (EAN), also known as nipple adenoma, florid papillomatosis, or papillary adenoma of the nipple, is a benign neoplasm originating from a lactiferous duct of the breast. Although the potential for malignant change is invariably negligible, the nature of the disease is quite intractable despite several treatment methods. Surgical excision is known as the treatment of choice, but this invasive approach is generally not acceptable to the vast majority of patients due to the cosmetic outcomes. Cryosurgery could be an alternative choice to preserve the structure of the nipple-areola complex, though its application has not been studied due to the paucity of cases. A 22-year-old female presented with a unilateral, crater-like erosion of the left nipple with serosanguineous discharge. The skin biopsy revealed proliferation of tubular structures, which corresponded to EAN. She was treated with 4 sessions of cryosurgery (open cryospray with liquid nitrogen) over 6 months, and the skin lesion resolved completely without any recurrence for 12 months. Although further study is required to determine the optimal treatment regimen for EAN, cryosur-

gery should be considered as an effective option to surgical excision. (**Ann Dermatol 33(2) 182 ~ 185, 2021**)

-Keywords-

Apocrine glands, Cryosurgery, Erosive adenomatosis of the nipple

INTRODUCTION

Erosive adenomatosis of the nipple (EAN), a challenging disease for both physician and patient, is a rare condition occurring in middle-aged females¹. Although it is usually benign, its destructive process and frequent recurrence are considerably detrimental and negatively affect patient's quality of life. Complete surgical excision is the first choice in treatment, but the postoperative deformity and loss of lactation ability make patients reluctant to accept this invasive method². Some reports have suggested multipronged minimally invasive methods, however; these various approaches are not equally applicable because of limited feasibility for each case and the need for exquisite surgical technique. Furthermore, these methods are still classified as "surgical" treatment, which could present a psychological burden for the patient. An alternative choice could be cryosurgery, which could selectively destruct the lesional tissue and be suitable for patients with anxiety concerning surgery. Currently, only 2 cases of EAN treated with cryosurgery have been reported^{3,4}. Herein, we report a case of EAN in a young female patient successfully treated with cryosurgery without further recurrence.

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Corresponding author: Gun-Wook Kim, Department of Dermatology, College of Medicine, Dong-A University, 32 Daesingongwon-ro, Seo-gu, Busan 49201, Korea. Tel: 82-51-240-5435, Fax: 82-51-253-0787, E-mail: sunyanzi@hanmail.net

ORCID: <https://orcid.org/0000-0003-1599-7045>

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CASE REPORT

A 22-year-old female presented with a localized skin lesion on the left nipple. It initially appeared 5 years ago, beginning with focal erosion at the left nipple, and then gradually destroying almost half of the nipple structure. She had used various topical agents without sustained remission. Before visiting our clinic, a physical examination in the breast-surgery department identified an incidental breast mass. Core-needle biopsy revealed only fibrocystic changes, and the patient was referred to the dermatologic department for evaluation of the skin lesion on the left nipple. Physical examination revealed a widely sloughed-off nipple with a fresh red-colored wound base, showing as localized erosion with serosanguineous discharge (Fig. 1).

A skin biopsy revealed prominent hyperplasia of the dilated tubular structures from the eroded surface of the epidermis through the deep dermal layer. The tubular components were of various sizes and composed of layers of



Fig. 1. Localized erosion of the left nipple presenting with fresh red colored base with bloody discharge. We received the patient's consent form about publishing all photographic materials.

cuboidal cells with a few detached cell aggregates in the lumina, but no cellular atypia or pleomorphism was observed (Fig. 2). The patient was diagnosed with EAN. We proposed surgical excision, but the patient declined due to concern with unsatisfactory cosmetic outcomes and anxiety concerning the invasive procedure. Cryosurgery was selected as an alternative. Using open cryospray with liquid nitrogen, we utilized a double freeze-thaw cycle per treatment session. Each freezing cycle was applied for 30~35 seconds until lateral spreading from the central lesion appeared. After 2 sessions (3 months after the initial treatment), nearly half of the eroded surface was improved, showing reepithelialization without any discharge. Two additional treatment sessions were performed, and the skin lesion almost completely healed with only a tiny slit-like hole remaining four weeks after the final treatment (Fig. 3). The total treatment period was six months. The patient was cosmetically satisfied with the treatment outcome and there has been no recurrence for 12 months.

DISCUSSION

EAN was first described as "benign intraductal papilloma" in 1951 by Haagensen et al⁵. It is classified as an apocrine glandular neoplasm originating from a lactiferous duct in the nipple-areola complex⁶. Unilateral erosion with a serosanguineous or bloody discharge of the nipple is a characteristic clinical feature, with varying lesional size. In histologic findings, highly proliferating tubular structures are embedded mainly in the dermis, altering the epidermal structure by growing to the surface of the nipple. Though it has a locally destructive clinical nature, its malignant potential is not apparent and several studies showed controversial results^{1,7}.

Unlike our patient, most EAN has been reported to occur in middle-aged females⁸. However, appearance in ado-

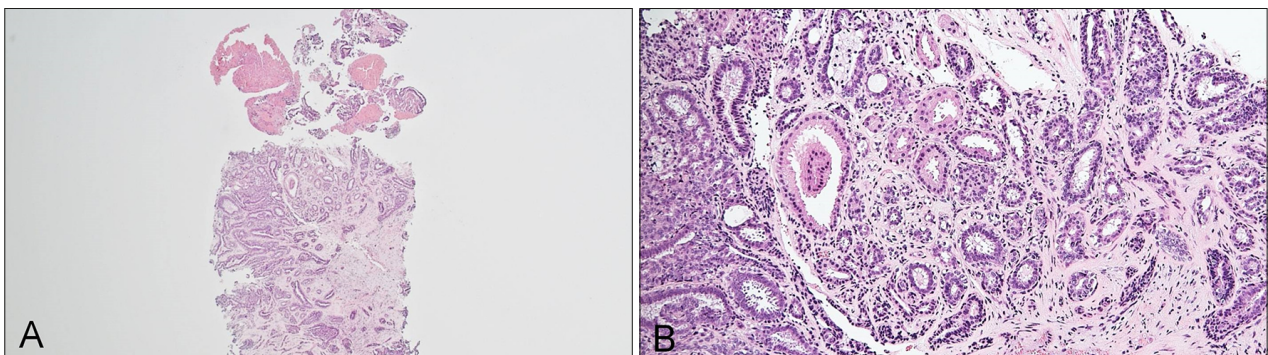


Fig. 2. (A) Prominent hyperplasia of the dilated tubular structures from eroded surface of epidermis through deep dermal layer (H&E, $\times 20$). (B) Numerous tubular components with various sizes but no atypia or pleomorphism, which composed of layers of cuboidal cells with a few of detachment of cell aggregates in lumina (H&E, $\times 100$).

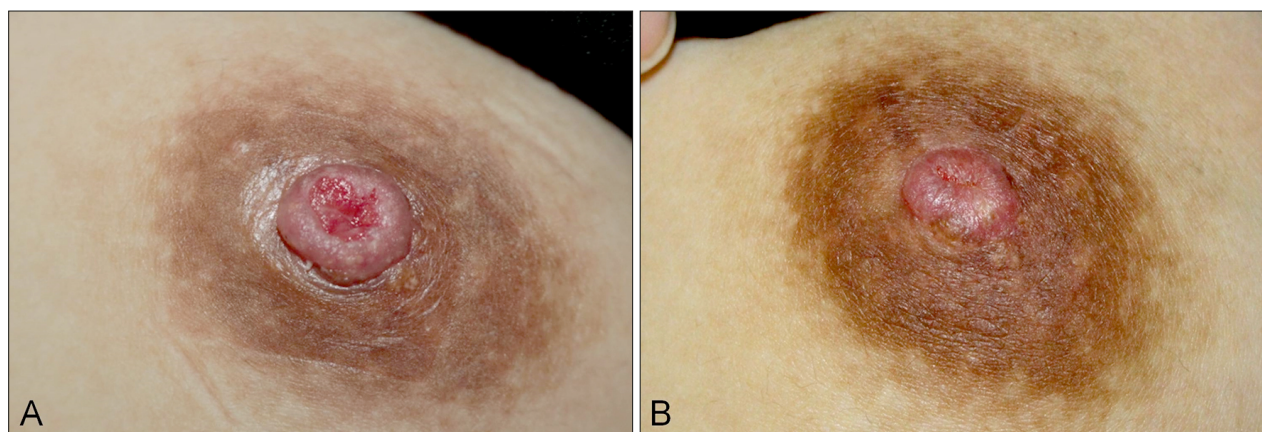


Fig. 3. (A) Improvement at 3 months after 2 sessions of cryosurgery. (B) Complete clearance with only a tiny slit-like opening remaining after 2 more consecutive cryosurgery sessions.

lescence or young adulthood is possible and there is even one report of EAN in an 8-year-old patient⁹. Furthermore, males may also be affected by EAN, although these are extremely rare cases, so the clinical or prognostic differences have not been studied yet¹⁰.

Considering its aggressive, recurrent features such as ulcer and bleeding, differentiation from other malignant disease should be clarified by histologic examination. In particular, Paget's disease and adenocarcinoma of the breast should be ruled out at initial evaluation. The microscopic findings are the most valuable evidence in differentiating EAN from these other diseases.

To date, only two reports have been found on the use of cryosurgery as a treatment option for EAN^{3,4}. The first case was a senile female patient who refused the surgical approach, so the author offered two sessions of cryosurgery, and the entire lesion healed without any further recurrence⁴. The other case was a young female patient successfully treated with only 1 session of cryosurgery³. Notably, the latter patient recovered lactational function and was able to breastfeed her baby. Given the good results and lack of long-term recurrence in these cases, we chose cryosurgery in this case and the outcome has been satisfactory for 1 year.

In addition to the benefit of the minimally invasive approach, cryosurgery may exhibit an advantage with selective tissue destruction. It has been demonstrated that adnexal structures (hair follicles, nerves, glandular structures) are more susceptible to freezing conditions^{11,12}. Considering the main pathologic feature of EAN, which is mainly composed of glandular epithelium, we speculated that cryosurgery could more selectively destroy the abnormal tissue.

The traditional surgical approach with wide excision of

the nipple-areola complex has limited usefulness due to unsatisfactory cosmetic outcomes and patient reluctance, especially in younger people. Various reports have introduced alternative surgical techniques using minimal wedge excision, purse string sutures, and Mohs micrographic surgery¹³⁻¹⁵. Although each method has potential benefits many patients have anxiety concerning any procedure described as 'surgical'. In our case, the initial skin lesion was quite large and the likely postoperative outcome did not meet with the expectations of our patient. Therefore, we concluded that cryosurgery would be the most appropriate and feasible approach in this circumstance.

Our case supports the favorable treatment outcomes of cryosurgery seen in previous reports of EAN. Of course, there is still no consensus or standardized procedure for cryosurgery and different procedures were reported in the two previous cases and in our case. Furthermore, assuming the deep infiltrative nature of EAN, the issue with recurrence after cryosurgery is unambiguously unsolved. However, cryosurgery could be an alternative treatment option and deserves further attention. Although the surgical treatment is rather straightforward and has the advantage of radical removal of the entire lesion, physicians need to be prudent and consider an individualized approach for each patient. In this regard, we carefully recommend cryosurgery as an alternative option for treatment of EAN.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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DATA SHARING STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID

Kyung-Nam Bae, <https://orcid.org/0000-0003-3254-8401>
 Kihyuk Shin, <https://orcid.org/0000-0001-8955-9828>
 Woo-Il Kim, <https://orcid.org/0000-0001-7991-2827>
 Min-Young Yang, <https://orcid.org/0000-0001-8994-8401>
 Won-Ku Lee, <https://orcid.org/0000-0002-4938-2455>
 Hoon-Soo Kim, <https://orcid.org/0000-0002-7649-1446>
 Hyun-Chang Ko, <https://orcid.org/0000-0002-3459-5474>
 Byung-Soo Kim, <https://orcid.org/0000-0003-0054-8570>
 Moon-Bum Kim, <https://orcid.org/0000-0003-4837-0214>
 Gun-Wook Kim, <https://orcid.org/0000-0003-1599-7045>

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