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대퇴부에 발생한 재발성 이소성 피부 선암: 양성 낭종성 병변과의 임상적 유사성으로 인한 진단 및 치료의 지연

(Recurrent Extrasalivary Cutaneous Adenoid Cystic Carcinoma of the Thigh: Clinical Mimicry of a Benign Cystic Lesion Leading to Delayed Diagnosis and Management)



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Purpose: Adenoid cystic carcinoma (ACC) is an exceptionally rare malignancy in extra-salivary sites, frequently presenting a diagnostic trap. Due to its indolent progression and unremarkable appearance, cutaneous ACC often disguises as common benign lesions, such as sebaceous cysts. This clinical mimicry leads to low index of suspicion, resulting in repeated incomplete excisions and significant delays in definitive diagnosis. We report a recurrent cutaneous ACC of the thigh, emphasizing the necessity of high clinical suspicion in such pitfalls.

Methods: A 49-year-old female presented with a recurrent right posterior thigh mass, previously misinterpreted as a benign cyst and excised twice without histopathological confirmation. A third recurrence finally revealed ACC with positive margins. Preoperative MRI for remnant tumor mapping showed focal T2 high signal intensity with enhancement. [Figure 1] Based on these findings, we performed a definitive wide radical excision with 1.5 cm safety margin, ensuring tumor-free boundaries via intraoperative frozen sections, followed by adjuvant radiotherapy. [Figure 2]

Results: Final pathology confirmed ACC with clear margins. Although lymphovascular and perineural invasions were absent, the history of multiple recurrences necessitated aggressive multimodal therapy. The patient has remained disease-free for 24 months. [Figure 3] However, due to the tumor's propensity for late-onset systemic spread, rigorous long-term surveillance remains mandatory.

Conclusion: The primary challenge of extra-salivary ACC lies in its benign clinical appearance, which delays early oncologic intervention. Recurrent or atypical cutaneous masses must be approached with a high degree of suspicion and mandatory biopsy. Radical surgical clearance combined with adjuvant therapy is essential to manage this persistent malignancy.

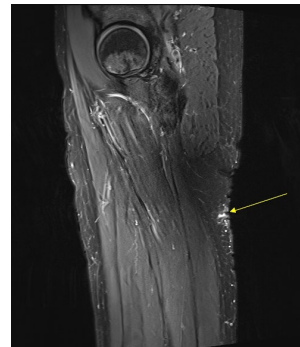


Fig. 1. Preoperative MRI findings. Contrast-enhanced MRI reveals ill-defined enhancement in the subcutaneous and skin layers of the right proximal thigh. Although suspicious for postoperative changes, the focal T2 high signal intensity indicates potential remnant tumor.

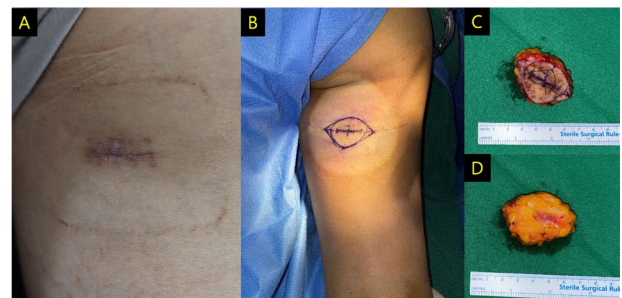


Fig. 2.

(A) Preoperative photograph of the patient's right posterior thigh shows the old scar from a previous excision, which was subsequently diagnosed as adenoid cystic carcinoma with positive resection margins on the third excision. (B) Intraoperative photograph showing the planned incision lines designed to achieve a wide excision with a sufficient safety margin around the previous scar. (C, D) Photographs of the resected specimen. The images show the excised skin and soft tissue, confirming the planned safety margins of 1.0 cm laterally and 1.2 cm deep. The gross appearance of the specimen indicates a successful wide excision for the recurrent tumor.



Fig. 3. A photograph of the well-sutured surgical site on post-operative day 3, on which the patient was discharged. Prophylactic radiation therapy was initiated 1.5 months later in consultation with the Department of Radiation Oncology. The patient has remained tumor-free for 2 years since, with no signs of local recurrence or distant metastasis.