

Photodynamic Therapy as Palliative Therapy for Invasive Syringoid Eccrine Carcinoma of the Auricle: A Case Report

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ABSTRACT

Recurrent eccrine syringomatous carcinoma is an extremely rare adnexal tumor predominantly found in the head and neck region. We present a case of a 75-year-old woman with a 7-year history of recurrent syringoid eccrine carcinoma in the scalp and auricle. She had undergone five previous oncologic resections and declined further oncologic ablation of the auricle, including temporal bone and external ear canal resection. We performed five sessions of palliative photodynamic therapy using topical aminolevulinic acid HCL 20% (ALA) and between 25 and 33 pulses of Intense Pulsed Light (Quantum) with an intensity of 30 Joules. The ALA incubation time was 3 hours, while the time between each session was 4 weeks. This treatment helped to control ulceration, pain, and swelling. We saw the patient 1 month after the last treatment, by that time she moved and we lost contact.

Keywords: Syringoid Eccrine Carcinoma; Photodynamic Therapy

1. Introduction

Syringoid Eccrine carcinoma (SEC) is a very rare tumor. It is very locally invasive, destructive, and is often recurrent. It was originally described as a basal cell tumor with eccrine differentiation in 1969 [1].

SEC differs from basal cell carcinoma in its cytology and enzymatic patterns. Histologically, SEC resembles syringoma in its ductal, cystic, and comma-like epithelial components. It differs from syringoma in its cellularity, anaplasia, and deep invasiveness [2].

According to the Cruz Modified Classification System, SEC is also known as a basal cell tumor with eccrine differentiation, adenocarcinoma of the eccrine sweat gland, syringealhidradenoma, atypical syringoma, sweat gland carcinoma with syringomatous features, eccrinebasalioma and eccrine syringomatous carcinoma. With a predilection for the scalp, it clinically resembles basal cell carcinoma and tends to histologically invade beyond its gross margins [3].

The tumor presents itself in patients ranging from 1 to 86 years of age, with most cases occurring in the fifth to seventh decades of life. Overall, these tumors occur

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equally in both sexes, although certain subtypes appear to have a predilection for males or females. Most sweat gland carcinomas grow slowly, with patients frequently having a delayed presentation of 5 years or more. Lymphatic metastasis typically occurs at a rate of 20% to 24%. Regional cutaneous metastasis and hematogenous spread have also been noted. Local recurrence occurs in 14% to 20% of cases [4]. Most patients survive beyond 10 years and usually die of another cause, although notable exceptions of death within 6 months of diagnosis due to metastasis to the lymph nodes and lungs have been reported.

Wide local excision and evaluation of lymph node biopsy or regional lymphadenectomy samples are recommended. Radiotherapy is reserved for recurrence or metastatic lymph node involvement [5]. Chemotherapy is not commonly used, although administration of trastuzumab has been reported in a metastatic tumor with Her-2/neu gene amplification [6].

2. Case Report

A 75-year-old female patient developed a small (<1 cm) tumor in the right temporoparietal scalp that had been

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excised 7 years earlier and treated as a benign tumor without histologic evaluation. Several months after the first treatment, another tumor grew within the scar; it was reported to be a basal cell carcinoma and was resected with tumor-free edges. Two years later, it recurred, and the patient underwent wide resection and coverage with a skin graft. This time, it was diagnosed as an adenoid sclerosant ulcerated and multicentric basal cell carcinoma. One year later, it recurred, and the immunohistological report showed recurrent SEC. A year after that, it recurred again, and she underwent wide resection including the upper third of the right auricle with the same diagnosis of SEC. The tumor reappeared one year later, and the oncologic surgeon proposed wide resection with total amputation of the auricle and resection of part of the temporal bone followed by coverage with a free flap. The patient declined this ablation and requested palliative treatment without surgery or radiotherapy. After palliative treatment, she reported no systemic signs of illness, weight loss, or functional impairment. Her examination results were normal.

The patient rejected surgical treatment, including Mohs micrographic surgery (**Figure 1**). She underwent five sessions of palliative photodynamic therapy for recurrent SEC of the auricle using topical aminolevulinic acid HCL 20% (ALA) (Levulan Kerastick; Stiefel Coral Gables Fl USA) and between 25 and 33 pulses using Level Intense Pulsed Light (Quantum Lumenis SR 560). The ALA incubation time was 3 hours, and 4 weeks passed between each session. This treatment helped to control ulceration, pain, and swelling. We saw the patient 1 month after the last treatment (**Figure 2**), after which time she moved and we lost contact.



Figure 1. A 75-year-old female with a 7-year-history of recurrent ulcerated syringoid eccrine carcinoma. Pretreatment view.



Figure 2. After five sessions of palliative photodynamic therapy with satisfactory control of local ulceration and inflammatory reaction. One month after the last session.

3. Discussion

After many recurrences and extensive surgeries including partial auricle amputation, the patient declined further surgical treatment. Without therapy, the course of SEC is very slow and of long duration, but multiple recurrences are described [1,7].

Local recurrence occurs in 14% to 20% of cases. Most patients survive beyond 10 years and usually die of another cause, although notable exceptions of death within 6 months of diagnosis due to metastasis to the lymph nodes and lungs have been reported [4,8].

Therapy of SEC consist mainly of surgical excision [9-17]. Nowadays, Mohsmicrographic surgery is the method of choice for SEC [11.16.18.19] if there is no "skip" area or evidence of multifocality and no evidence of distant metastases. Nishiwara reports the use of chemotherapy as initial treatment in a case that exhibited multiple distant metastases at diagnosis. In general, chemotherapy and/or radiation therapy have been used for metastatic sweat gland carcinomas. Radiation treatment for sweat gland carcinoma has been reported to be ineffective, but more recent reports have found a role for radiation in the local control of this disease [20-24]. The role of chemotheraphy in sweat gland carcinoma is also under debate. Both single-agent and combination chemotherapy have been used infrequently and, at best, have shown only a temporary benefit [22-26]. There have been a few reports in which patients with SEC were treated with radiotherapy for local control and bone metastases [11,27]. Nishizawa et al. reported an extended SEC treated with seven cycles of combination chemotherapy [28].

Photodynamic therapy for superficial skin tumors, including basal cell carcinoma, following local application of ALA has been described by several authors [29-31].

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However, it has not been described for the treatment of SEC. We believe that this case shows the utility of photodynamic therapy as a palliative treatment for temporal control of tumor activity, even if it may not work in other clinical contest except the ones previously reported. This alternate approach was used because of patients' refusal to radical surgery, and there is not any previous report of palliative treatment in this rare neoplasm.

4. Disclosure

The authors have no commercial associations or financial disclosures with regard to this manuscript.

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