CASE REPORT

Microcystic adnexal carcinoma: A case report.

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ABSTRACT

Microcystic adnexal carcinoma is a rare malignant cutaneous adnexal neoplasm characterized by slow growth but locally aggressive behavior, with infiltrative growth and a high rate of local recurrence. Clinically, it may mimic other cutaneous neoplasms, particularly morpheaform basal cell carcinoma, posing a significant diagnostic challenge.

We present the case of a 73-year-old woman with a history of multiple basal cell carcinomas who presented with an infiltrated plaque located on the left malar region with an approximately one-year history of evolution. Dermoscopic examination revealed a central crust, adherent yellow keratin, and fine arborizing telangiectasias. Histopathological examination demonstrated an infiltrative epithelial neoplasm composed of small nests and cords of basaloid cells associated with ductal structures and keratin cysts embedded within a desmoplastic stroma, consistent with microcystic adnexal carcinoma. The patient was treated with Mohs micrographic surgery requiring four stages, with a favorable outcome and no evidence of recurrence to date. This case highlights the importance of clinicopathological correlation for the timely diagnosis of this uncommon adnexal neoplasm.

INTRODUCTION

Microcystic adnexal carcinoma (MAC) is a rare malignant cutaneous adnexal neoplasm first described by Goldstein et al. in 1982. It is characterized by slow growth but locally aggressive behavior, with marked infiltrative capacity and a tendency toward neurotropism. It predominantly affects the head and neck region, particularly in middle-aged and elderly patients.[1]

The diagnosis of MAC represents a challenge due to its low incidence and the significant clinical and histopathological overlap with other benign and malignant cutaneous proliferations.[2]

We present the case of a patient with microcystic adnexal carcinoma located in the left malar region, initially suspected to be morpheaform basal cell carcinoma, highlighting the clinical, dermoscopic, and histopathological findings that allowed establishment of the definitive diagnosis.

CASE REPORT

A 73-year-old woman with a medical history of arterial hypertension, type II diabetes mellitus, hypercholesterolemia, and multiple basal cell carcinomas presented with an approximately one-year history of a lesion located in the left malar region, which began as an erythematous macule with progressive growth and remained asymptomatic.

Physical examination revealed a solitary, erythematous, infiltrated plaque measuring 7 mm in diameter, with a hyperkeratotic surface, central ulceration, and adherent crust. On palpation, the perilesional skin felt indurated (Figures 1 and 2).

Dermoscopic examination revealed a central crust covered by adherent yellow keratin, peripheral whitish scale, and fine arborizing telangiectasias over a pinkish-erythematous background with shiny white structures. In addition, superficial irregular linear vessels were identified without evident melanocytic structures. (Figure 3)

A skin biopsy was performed, revealing skin extensively and diffusely infiltrated by a malignant epithelial neoplasm composed of small nests and cords of cuboidal basaloid cells with the formation of small keratin cysts showing focal calcification. The stroma exhibited desmoplastic fibrosis and marked solar elastosis in the superficial dermis. The tumor infiltrated the subcutaneous tissue. No perineural or lymphovascular invasion was identified (Figures 4 and 5).

Based on the clinical and histopathological findings, a diagnosis of microcystic adnexal carcinoma was established. The patient was treated with Mohs micrographic surgery, achieving tumor-free surgical margins after the fourth stage of surgery.

Figure 1.

Figure 2.

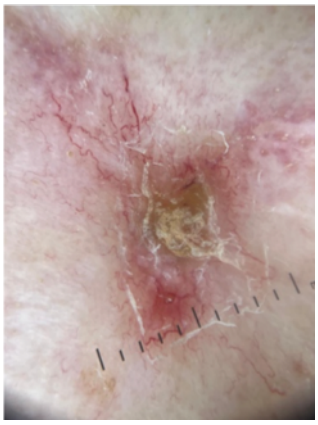

Figure 3.

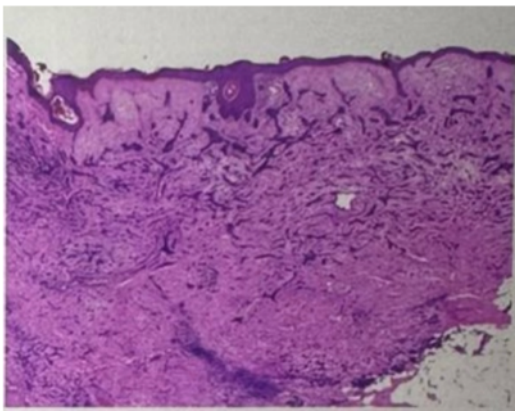

Figure 4.

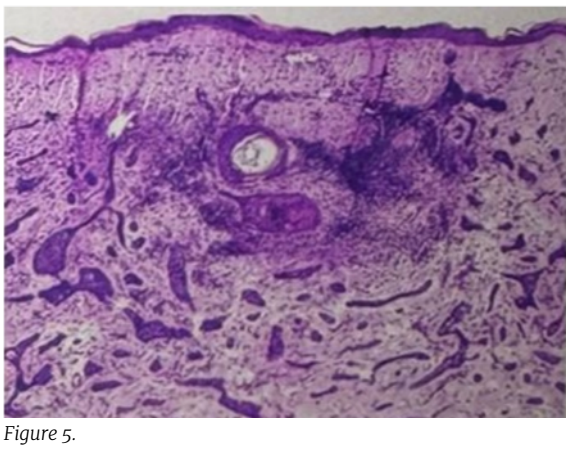

Figure 5.

DISCUSSION

Microcystic adnexal carcinoma (MAC) is an extremely rare neoplasm, with fewer than 700 cases reported worldwide. Its estimated incidence ranges from 1.6 to 6.5 cases per 10 million inhabitants. It occurs most frequently between the fourth and sixth decades of life, with a slight female predominance and a higher incidence among Caucasian and Asian patients, being uncommon in individuals of African descent.[1]

Factors associated with its development include exposure to ultraviolet or ionizing radiation, as well as states of immunosuppression. Clinically, it usually presents as an indurated plaque, nodule, or pseudocystic lesion that is generally asymptomatic and demonstrates slow, progressive growth. The presence of pain, paresthesia, or a burning sensation may suggest perineural invasion, a relatively frequent finding in this neoplasm.[5]

Most cases occur in the head and neck region, particularly on the left side of the face, with a predilection for the periorbital area and the nasolabial fold.[1]

Dermoscopically, microcystic adnexal carcinoma may share features with morpheaform basal cell carcinoma and desmoplastic trichoepithelioma, making clinical recognition challenging. Reported findings include fine linear telangiectasias, structureless white areas, and circular white structures, the latter being histologically related to keratin cysts and intratumoral calcifications. Costello et al. compared the dermoscopic characteristics of these entities and observed that circular white structures were more frequent in MAC and desmoplastic trichoepithelioma, whereas morpheaform basal cell carcinoma predominantly exhibited shiny white structures under polarized light.[6]

Microcystic adnexal carcinoma is characterized by locally aggressive behavior and marked infiltrative capacity, with the potential to extend into subcutaneous tissue, skeletal muscle, and even deeper structures such as the perichondrium, periosteum, and bone. Histopathologically, the epidermis is generally preserved, while the dermis shows a proliferation composed of small keratin cysts, islands of basaloid cells, and ductal structures embedded within a desmoplastic stroma.[1]

MAC represents a significant diagnostic challenge both clinically and histopathologically. Several series report that between 25% and 53% of cases are initially misdiagnosed, with morpheaform basal cell carcinoma being the most frequent differential diagnosis.[1] The principal histopathological differential diagnoses include syringoma, desmoplastic trichoepithelioma, morpheaform basal cell carcinoma, trichoadenoma, eccrine carcinomas, and metastatic breast carcinoma.[4] In addition to clinical similarities, histological diagnosis may also be challenging, particularly when superficial or shave biopsies are performed, as these may fail to adequately assess the deep infiltrative pattern characteristic of this neoplasm.[1]

In our case, the lesion exhibited clinical and dermoscopic features suggestive of morpheaform basal cell carcinoma, which was therefore considered the primary initial diagnosis. However, definitive histopathological examination demonstrated findings consistent with microcystic adnexal carcinoma.

Treatment options for MAC include surgery and radiotherapy. However, radiotherapy as monotherapy is not recommended because of its limited efficacy and the potential to induce more aggressive tumor variants. Currently, it may be considered only as adjuvant therapy in selected cases, such as recurrence, perineural, vascular, or lymphatic invasion, or deep involvement of muscle and bone.[1]

Conventional surgical excision is associated with high recurrence rates due to the difficulty in clinically defining tumor margins and the limited evaluation of surgical margins. In contrast, Mohs micrographic surgery allows near-complete assessment of both lateral and deep margins, resulting in lower recurrence rates and greater preservation of healthy tissue, an aspect that is particularly important for lesions located in the head and neck region.[1]

(A) Initial clinical lesion

(B) Surgical defect following four stages of Mohs micrographic surgery.

(C) Immediate surgical reconstruction.

(D) Scar outcome during postoperative follow-up.

In our case, the patient required four stages of Mohs micrographic surgery to achieve tumor-free margins, highlighting the characteristic infiltrative pattern of microcystic adnexal carcinoma. This finding is consistent with reports in the literature describing a frequent discrepancy between the apparent clinical size and the true extent of tumor infiltration.

CONCLUSION

Microcystic adnexal carcinoma is a rare, locally aggressive adnexal malignancy that is frequently underdiagnosed because of its clinical and histopathological similarity to other cutaneous neoplasms, particularly morpheaform basal cell carcinoma. Early recognition and appropriate clinicopathological correlation are essential for establishing an accurate diagnosis and ensuring timely treatment. Furthermore, obtaining sufficiently deep biopsy specimens is crucial for identifying the characteristic infiltrative pattern of this neoplasm. Mohs micrographic surgery remains the treatment of choice because of the frequent subclinical extension of the tumor and its lower recurrence rates.

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