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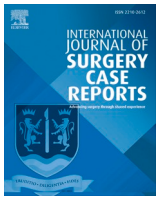

Case report

Nodular hidradenocarcinoma of the forehead presenting as a lipoma: A case report with a review of literature

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ABSTRACT

Introduction: Hidradenocarcinoma is an uncommon slow-growing malignant tumor that originates from sweat glands, it is most seen in the head and neck, and present typically as hard asymptomatic nodule.
Diagnosis is confirmed by histopathology and immunohistochemistry, and wide local excision is the mainstay of the treatment.
In our study, we discuss the approach and management of this rare disease, with focusing on the importance of histopathology in diagnosing such cases.
Presentation of case: We present a very rare case of a 58-year-old female who presented with painless slow growing forehead swelling for 2 years, with clinical feature suggestive of lipoma. The patient underwent surgical excision, and post-operative histopathological examination of the specimen was consistent with nodular hidradenocarcinoma.
Discussion: Hidradenocarcinoma is a very rare malignant tumor that was first reported in 1954. This tumor was reported to be more common in males, and the incidence increases with age. The origin of hidradenocarcinoma is mostly de novo. The definitive diagnosis requires histopathological examination, and wide local excision is the mainstay of the treatment.
Conclusion: Hidradenocarcinoma is a very rare sweat gland malignancy, and the diagnosis can be challenging. In our study, we emphasize the importance of raising the awareness among the surgical oncology society regarding this rare tumor, and we discuss the approach and management of such an uncommon disease, with focusing on the importance of histopathology in diagnosing such cases.

1. Introduction

Hidradenocarcinoma is an uncommon slow-growing malignant tumor that originates from sweat glands, representing only 6 % of malignant eccrine tumors and seen in 1:13,000 skin biopsies, it accounts for less than 0.001 % of all tumors [1–3].

Hidraadenocarcinoma is most seen in the head, neck (especially on the face), trunk, limbs, and oral cavity and present typically as hard asymptomatic nodules [2,3].

Diagnosis is confirmed based on histopathology and immunohistochemistry, and wide local excision is the mainstay of the treatment [3].

This rare tumor carries a high risk of local recurrence and distant metastasis, most commonly to regional lymph nodes and distant viscera and carries a poor prognosis [1–3].

We present a very rare case of a A 58-year-old female who presented to our clinic in a Military hospital with painless slow growing forehead swelling for 2 years, with clinical features suggestive of lipoma. The patient underwent surgical excision, and post-operative histopathological examination of the specimen was consistent with nodular hidradenocarcinoma.

This work has been reported in line with the SCARE 2020 criteria [4].

2. Presentation of case

A 58-year-old Saudi house wife female patient known case of ischemic heart disease, atrial fibrillation, mitral stenosis, cerebral vascular accident, osteoporosis, and disc prolapse presented to our clinic with painless slow growing forehead swelling for 2 years, associated with serous discharge. No history of trauma or bleeding, no personal or

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family history of malignancies. Past surgical and psychosocial history was unremarkable. Patient was on Bisoprolol, Apixiban, Lasix and Omeprazole.

Local examination revealed a solitary skin colored, firm, non-tender, non-fluctuant, round, mobile mass of size 4 × 4 cm which was located just above the medial one third of the left eyebrow. The overlying skin was smooth with no erythema, discharge, or ulceration, no palpable lymph nodes.

Rest of the physical examination was unremarkable. Appearance and clinical feature were initially suggestive of lipoma.

Laboratory results were unremarkable.

The patient underwent surgical excision by the surgical team under local anesthesia as she was high risk for general anesthesia, after optimizing her blood pressure, glucose readings, cardiac and anticoagulant status. Post-operative she did well and was discharged home on the next day after giving her wound care instructions and follow up appointment.

Histopathological examination of the specimen showed a well circumscribed nodule measuring 2 × 1 cm and an ill-circumscribed lobulated infiltrating mass extending to the dermis, formed of variable dimension nodules, composed of sheets of basophilic cells with focal glandular-like structures, the basaloid cells showed marked atypia, distinct cell membranes, and prominent nucleoli. Some cells have clear vacuolated spaces, there were foci of squamous differentiation and duct formation (Fig. 1).

These features were consistent with Nodular Hidradenocarcinoma. The margins were irregular and the tumor reaching margins which means re-excision is needed. Post-operative staging CT scan showed 3 suspicious pulmonary nodules measuring 9 mm, 9 mm and 6 mm.

The patient was then referred to Plastic Surgery Department for further assessment and management.

A multidisciplinary meeting was held and recommended no further surgical intervention with regular follow-up every 6-months, as the patient is high risk for surgery when considering her comorbidities, she is also asymptomatic with no evidence of local disease recurrence at 6 months follow-up post-surgery. Intervention adherence and tolerability is irrelevant.

3. Discussion

Hidradenocarcinoma is a very rare malignant tumor that was first reported by Keasby and Hadley in 1954, it is also known as malignant clear cell hidradenoma, clear cell hidradenocarcinoma, and malignant acrospiroma [2].

Hidradenocarcinoma is reported to be more common in males, and the incidence increases with age. A recent study conducted by Gao et al. included 289 patients has concluded that the majority of these patients were males, accounting for 58.8 %, and showed that the mean age was 62.5 years, ranging between 15 and 89 years, with 60.6 % of patients were 60 years or older [2].

The origin of hidradenocarcinoma is mostly de novo, however, some of the cases result from malignant transformations of benign hidradenoma, with some similarities in clinical manifestations, therefore, the distinction is important to establish in the accordance of histopathology identification of the tumor, and adequate excision of these tumors is crucial [5].

This rare tumor usually presents as a solitary hard nodule mostly seen in the head and neck region, accounting for 41.9 % of the cases [2].

Identifying the origin of this uncommon tumor might be challenging; considering the presence of clinical, cytological, and histological similarities with other tumors, therefore, the definitive diagnosis depends on histopathological characteristics of this rare tumor, in addition to immunohistochemistry [3]. Investigating the lymph nodes to determine its involvement and the disease extent is unfavorable if it wasn't identified clinically as lymphadenopathy, and it showed no difference or benefit in the overall survival, particularly at advanced stages [6].

Singh et al. [7] reported a case of hidradenocarcinoma of the eyelid

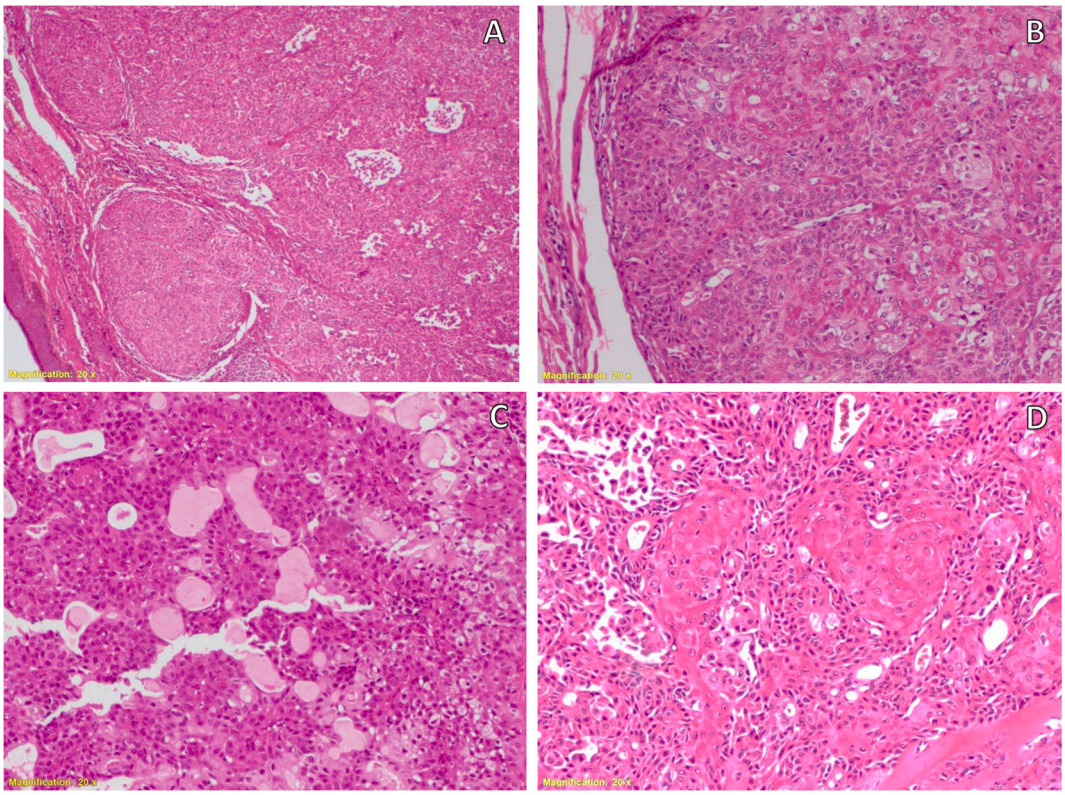

Fig. 1. (A) Nodular dermally based tumor, (B): the tumor cells are epithelioid, basaloid and may often show clear cell changes. (C): Tubular Lumina are lined by columnar secretory cells. (D): foci of squamous differentiation and duct formation.

that presented as a mass causing excessive tearing, ptosis and ectropion. This tumor was diagnosed initially as meibomian gland carcinoma, and the definitive diagnosis of hidradenocarcinoma was made only after surgical excision and histopathological examination of the specimen.

Aggressive growth was reported in the literature, in a patient who presented with focal seizures and scalp swelling, it was managed by near-total decompression and adjuvant radiotherapy, and the post-operative histopathology confirmed the diagnosis of scalp hidradenocarcinoma with brain extension [8].

Our patient presented with a forehead swelling that was consistent initially with lipoma, however, the pattern of growth along with the CT scan findings was suspicious, and the final diagnosis of hidradenocarcinoma was made after the histopathological confirmation.

Wide local excision is the mainstay of the treatment, with at least 3 cm margins. In addition, Mohs micrographic surgery has shown excellent control [3]. Tolkachjov et al. conducted a study from 1993 to 2013, that is the largest reported series of HAC treated with MMS included 10 patients with long-term follow-up (86.06 months) and concluded that none of those patients had recurrence, metastasis, or disease-related mortality [9].

Radiotherapy has conflicting results, in which it was effective in some cases, in contrast, it showed resistance in others. It can be considered in cases of impossible surgical intervention and is preferred to be given in high doses (50Gy–70Gy) [3].

The efficiency of adjuvant chemotherapy has not been demonstrated, examples include 5-fluorouracil and capecitabine, while Targeted therapy like Trastuzumab has shown to be effective in stabilizing metastatic hidradenocarcinoma [3].

Moreover, Electrochemotherapy (ECT) is a recent treatment option that has an excellent outcome when combined with RT, as it also allows for the preservation of the cosmesis and function of the surrounding tissues [3].

Kyrgias G et al. reported a case of temporal hidradenocarcinoma that was treated successfully with concomitant electrochemotherapy and radiotherapy with no evidence of disease recurrence or metastasis [10]. In addition, Obermann et al. reported the first case of successful treatment with anti-PD-1 in a metastatic cutaneous hidradenocarcinoma of the flank to the lung, bone and brain [11].

Our patient underwent surgical excision under local anesthesia successfully, the wound was closed primarily without the need for subsequent flap transfer, with no evidence of disease recurrence at 6-months follow-up.

Long term follow-up of patients diagnosed with hidradenocarcinoma is advised, as some reported cases recognized an association of hidradenocarcinoma with second primary malignancies development like rectal cancer, non-epithelial skin cancer and mesothelioma, therefore, early screening tests may be considered [12].

Hidradenocarcinoma is well known to recur and metastasize in 50 % and 60 % respectively, and it carries a poor prognosis with a 5-year survival rate of 30 %. Fatal scalp hidradenocarcinoma was reported in a patient who died of cavernous sinus thrombosis after tumor recurrence and loss of follow-up [13].

4. Conclusion

Hidradenocarcinoma is a very rare sweat gland malignancy. The diagnosis of Hidradenocarcinoma can be challenging due to rarity of this disease, as well as similarity in manifestation with other conditions, and determining the origin of the tumor requires histopathological confirmation. In our study, we emphasize the importance of raising the awareness among the surgical oncology society regarding this rare tumor, and we discuss the approach and management of such an uncommon disease, with focusing on the importance of histopathology in diagnosing such cases.

Patient's perspective

Our patient was satisfied regarding the treatment she received, and attended her follow-ups regularly with no evidence of disease recurrence or metastasis 6 months postoperatively.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Ethical approval

Ethical approval has been exempted by our institution because this is a case report and no new studies or new techniques were carried out.

Author contribution

asem H. Alshareef: Idea, Supervision, writing the manuscript, reviewing the final manuscript, correspondence.

Raghad A. Ghazzawi: Data collection, writing the manuscript, submission, editing

Ghofran E. Sheikh: Writing the manuscript.

Research registration

Not applicable.

Guarantor

The guarantors for this case report are Basem AlShareef, Raghad Ghazzawi, Ghofran Sheikh.

Declaration of competing interest

This manuscript has never been published or presented elsewhere in part or in entirety and is not under consideration by another journal. We have read and understood your journal's policies, and we believe that neither the manuscript nor the study violates any of these. There is no other conflict of interest to be disclosed.

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