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Clear cell hidradenoma of the hand: A case report in an 83-year old patient

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ABSTRACT

INTRODUCTION: Clear cell hidradenoma (CCH) is a superficial adnexal tumor of the sweat glands. It generally appears on the trunk or scalp and is uncommon on the upper and lower limbs; it is extremely rare on the hand. CCH tend to be benign, with low malignancy risk. Treatment is based on complete surgical excision. We report a rare case of a CCH of the palm of the hand in an 83-year old patient.
PRESENTATION OF CASE: An 83-year old male patient presented with a small mass on the palmar surface of his left hand, which was progressively increasing over 5 years. The tumor was surgically excised after sonography and sent for histologic examination, based on which diagnosis of CCH was made. Three months after surgery, the patient had no recurrence and was symptom free.
DISCUSSION: CCH is a rare tumor of the distal extremities and to the best of our knowledge, only one case of this tumor on the hand has been reported. Our case represents a rare CCH located at the palm of the hand, which was successfully surgical excised without recurrence. Therefore, CCH needs to be considered in the differential diagnosis when encountering masses on the distal extremities. Hidradenocarcinoma is the malignant variant that arises from the same cells.
CONCLUSION: We report the second case of CCH on the palmar surface of the hand. Treatment of choice is surgical excision, followed by histological analysis and close follow-up for recurrence.

1. Introduction

Clear cell hidradenoma (CCH) is a rare adnexal tumor of the apical sweat gland [1]. This tumor has been called by different names in literature over the years, including clear cell myoepithelioma, solid cystic hidradenoma and eccrine acrospiroma [2]. More recently, some authors [3,4] reported two different types of nodular hidradenoma: CCH that arises with apocrine differentiation and poroid hidradenoma which has eccrine differentiation. Hidradenocarcinomas (HAC) are the malignant equivalent of these tumors, which tend to appear de novo, and not as progression of CCH.

CCH generally present as cystic nodules, between one and three cm in diameter, usually located on the trunk or scalp. It can occasionally present on the limbs, but very rarely on the hands or feet. CCH are more common in women, usually between the 4th and 8th decade of life, but have been described in all ages [5]. They tend to be painless and can appear with blue or red discoloration of the overlying skin, with possible ulceration with fluid discharge [6]. They are generally benign, with a low risk of malignancy. However, the lesion may recur after surgical excision with a reported incidence of 10% [7]. Treatment usually consists of complete surgical excision and histologic examination for definitive diagnosis [5,8].

We present a rare case of a clear cell hidradenoma located on the palmar surface of the hand in an 83-year old man.

2. Case presentation

This paper is reported in line with the SCARE 2018 criteria [9].

An 83-year old male patient was admitted to the outpatient orthopedic clinic of our hospital for the presence of a small mass on the palmar surface of his left hand. The mass had been slowly but steadily increasing in size over the previous five years, and in the last few months caused some discomfort during daily activities to the patient. He referred no previous trauma, injury nor chronic irritation to the hand. History was noncontributory other than for hypertension and surgery to repair an abdominal aortic aneurism seven years earlier. On the clinical exam, there was a 1.5 cm mass, with a slight blue discoloration of the overlying skin, at the intersection of the proximal palmar crease and the third metacarpal bone. The mass was of elastic consistence, non-mobile, non-pulsating,

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and slightly painful to palpation. Hand and wrist had full active range of motion, and there was no presence of lymphadenopathy.

Sonography confirmed the presence of a solid/cystic neoplasm located in the subcutaneous tissue of the palm of the hand, therefore surgical excisional biopsy was planned. Excision of the mass was performed, by the senior author, in the operating room. A tourniquet was placed at the arm and after preparing the surgical field, the skin was carefully incised over the mass using a V-shaped incision. The mass was adhered to the dermal layer with epidermal connection. Through careful dissection, which was very difficult, it was fully removed, placing particular attention to not damage the skin, which appeared very thin over the mass. The tumor was circular, nodular, soft with a greyish coloration, around 1.2 cm in diameter (Fig. 1). The wound was thoroughly irrigated and closed with simple non-continuous nonabsorbable 3.0 stiches.

The histological examination showed a well-circumscribed nodule, with smooth borders, constituted by lobules of basophilic and clear cells separated by homogeneous collagenous material. There was no cytological atypia, mitotic figures or necrosis. Based on these finding, clear cell nodular hidradenoma was diagnosed (Fig. 2). At three months follow up, the patient did not have recurrence and did not report any symptoms (Fig. 3), and was satisfied with the result.

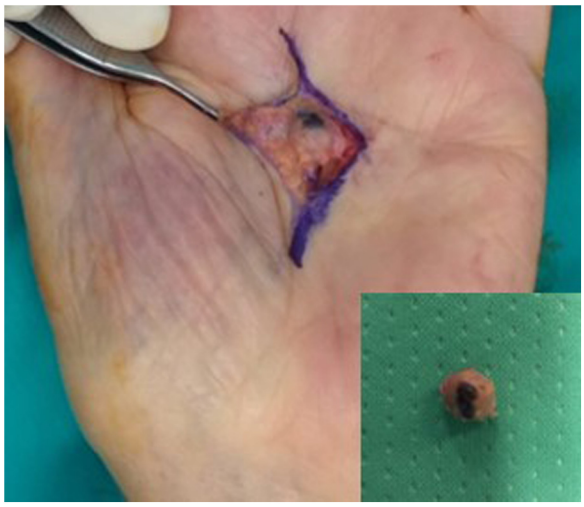

Fig. 1. Surgical view of the mass during and after accurate dissection. It was round, 1.2 cm in diameter, with a greyish coloration and nodular appearance. A small area with blueish coloration can be seen, which was the one underlying the skin.

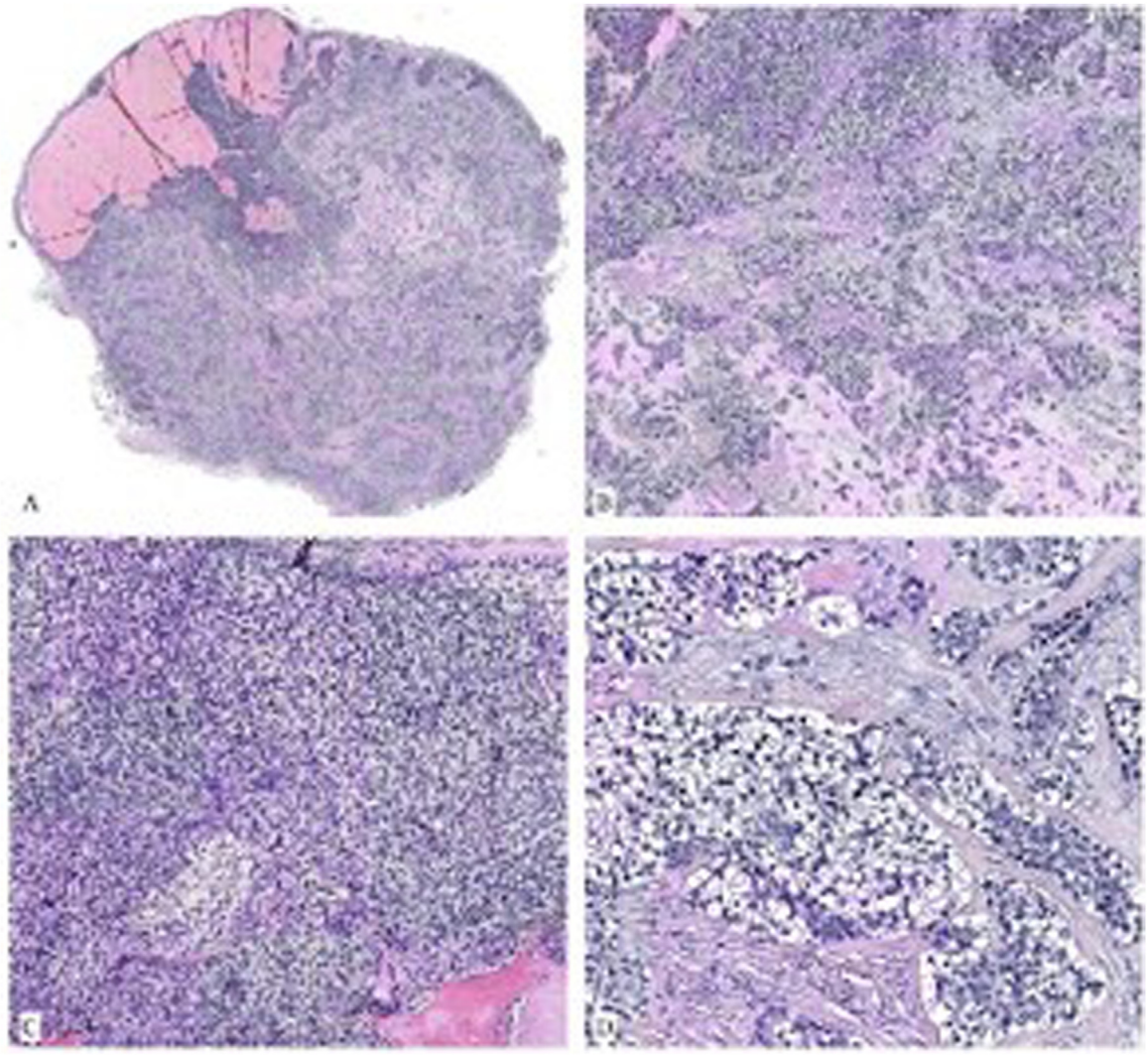

Fig. 2. Histological aspect of the mass. Well-circumscribed nodule, with smooth borders, constituted by lobules of basophilic and clear cells separated by homogeneous collagenous material. On the top there is a cystic dilated space full of sweat (A). At higher magnification, it is possible to appreciate that the two cell populations intermingle each other. Cytological atypia, mitotic figures or necrosis are absent (B, C, D).

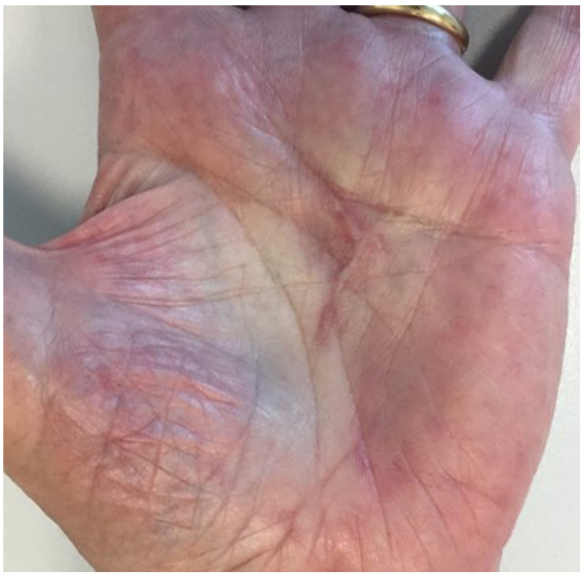

Fig. 3. Clinical aspect of the palm of the operated hand three months after surgery. The V-shaped scar is present without any recurrence.

3. Discussion

Clear cell hidradenoma is a rare benign tumor of the apical sweat glands. The most common location is the trunk and scalp, while it is rare on the upper and lower limbs and extremely rare on the distal extremities (hand or foot). After review of literature, one previous case report of this presentation has been described. Shin et al. [10] described a 51-year old woman that presented with a nodule in the thenar area of her left palm. After histological examination of the mass, they diagnosed the lesion as a CCH, but apocrine differentiation was not observed. They concluded that the tumor might have originated from the acrosyringium of the eccrine gland.

Few cases of CCH located at the level of the limbs were reported. Three of them were observed at the upper limb during skeletal growth and they had three different location: arm, forearm and wrist [7,11,12]. Two of them had a successful surgical excision without recurrence [7,11], while the third, observed in a 7-month old boy and located at wrist, was left untreated and spontaneously regressed in three months. Other three cases of CCH have been described on the foot, two located at the third and fourth toe [2,4] and the other at the lateral part of the foot [13]. All these cases were surgically excised and only one case recurred, shortly after the initial surgery; this patient underwent a second surgical excision, and no further recurrence was observed [13]. All the reported cases, located at the upper and lower limbs, were diagnosed by histological examination performed after surgical excision. In only one case, in which the diagnosis was made with a punch biopsy, CCH regressed spontaneously after three months.

In our case, the mass increased in size slowly but steadily and caused some discomfort to the patient, therefore we opted to perform a surgical excision to resolve the patient's symptoms and make a definitive diagnosis. We believe that the only observed case of tumor regression was related to the age of the patient, therefore a "wait and watch" approach may be considered exclusively in very young patients. The differential diagnosis is often very complex and an incisional, excisional or needle biopsy is required to make a definitive diagnosis. Since these tumors are usually small in size, an excisional biopsy is preferred. In fact, in no cases an incisional biopsy was described and a needle biopsy has been used only in the youngest patient, a seven-month old, who was too young to undergo excisional biopsy. CCH recurrences are usually uncommon, however a routine follow-up is recommended as they are possible.

Regarding the surgical procedure, we believe that in these cases the skin should be excised, but we did not perform a skin excision because we did not know the final diagnosis. In our case, the surgical procedure was difficult because the mass was adhered to the dermal layer with epidermal connection.

Regarding the surgical procedure, we believe that in these cases the overlying skin should be excised, but at the time we did not perform a skin excision because we did not know the final diagnosis and did not believe it was necessary. The surgical procedure was difficult because the mass was adhered to the dermal layer with epidermal connection, but through careful dissection the mass was removed as a whole and it did not appear to invade the surrounding tissues.

Hidradenocarcinoma (HAC) is the malignant variant of CCH which are also very rare on the hand; most of them seem to appear de novo. Jinnah et al. [14] described a case of HAC in a 56-year-old female, who presented a 3.5 cm mass on the right palm, which underwent fine needle aspiration followed by complete surgical excision. Chan et al. [15] described another HAC of the middle finger in a 70-year old man, which they ended up treating with amputation. Other authors [16] described a case of HAC on the thumb of a 70-year old female patient with had "unusual histological features of in situ malignant changes", which suggests a progression of CCH to HAC.

4. Conclusions

We report the second case of CCH on the palmar surface of the hand. CCH represent a rare benign tumor originating from the apical sweat glands. The surgeon needs to keep this type of tumor amongst the differential diagnosis when encountering small subcutaneous masses. Treatment of choice is surgical excision, followed by histological analysis and close clinical follow-up over the next three to six months to look for recurrences. Surgical treatment should involve complete and careful removal of the mass from the surrounding tissue when this is possible. Wide excisional margins in delicate areas such as the hand are not necessary if there is confidence that the mass has been completely removed.

Declaration of Competing Interest

We certify that no benefits in any form have been received or will be received from a commercial party related to the subject of this article.

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Ethical approval

The study was notified to the ethical committee of our hospital; it does not need a specific ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Kristian Efremov: conceptualization, writing, original draft
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Lidio Petrungaro: investigation
Fernando De Maio: writing, review & editing
Monia Di Prete: histological analysis
Amedeo Ferlosio: histological analysis
Ilaria Tresoldi: investigation
Pasquale Farsetti: supervision

Registration of research studies

N/A.

Guarantor

Prof. Pasquale Farsetti.

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