Recurrent Hidradenocarcinoma in the Abdominal Wall with no Distant Metastasis: A Case Report and Literature Review

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Abstract

Background: Hidradenocarcinoma is a rare, aggressive tumor with a high rate of local recurrence and distant metastasis. There are no current management guidelines, and surgery has been used to control the primary disease. Even with wide surgical excision, the course of the tumor is unpredictable, and the prognosis for survival is poor. The most common location is the scalp, followed by the extremities, and rarely the trunk.

Objective: To present a rare recurrent hidradenocarcinoma in the abdominal wall with no distant metastasis.

Case presentation: We present a rare case of recurrent hidradenocarcinoma on the abdominal wall in a 56-year-old male with no known distant metastasis treated with wide surgical excision.

Conclusion: This case represents the second reported case of hidradenocarcinoma involving the abdominal wall in the literature. Wide surgical excisions of the tumor with wide safe margins carry excellent outcomes on 5-year follow-up.

Keywords: Recurrent hidradenocarcinoma; Abdominal wall hidradenocarcinoma; Distant metastasis; Case report.

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https://djm.uodiyala.edu.iq/index.php/djm

Received: 22 November 2024 Accepted: 28 February 2025 Published: 25 April 2025

Introduction

Hidradenocarcinoma is a rare, aggressive, malignant adnexal tumor of the sweat glands (1). It accounts for approximately 6% of all malignant eccrine tumors and less than 0.001% of all tumors (2). It has a slight female predominance (with a male-to-female ratio of 4 to 6) and tends to occur in the fifth to seventh decade of life (3, 4). It was first reported as clear-cell eccrine carcinoma by Keasbey and Hadley in 1954 (5), and it is also known in the literature as clear-cell hidradenocarcinoma, solid-cystic adenocarcinoma, malignant clear-cell hidradenoma, malignant clear-cell myoepithelioma, malignant acrospiroma, and clear-cell eccrine carcinoma (6). They are mainly presented as isolated, hard, asymptomatic intradermal erythematous or violaceous nodules on the head, neck, trunk, limbs, or mouth (1). However, involvement of other body sites like the trunk and extremities occurs on rare occasions (6). Rehman et al. reported the first case of hidradenocarcinoma on the left abdominal wall in a 78-year-old woman. However, no similar cases are reported in the literature (7). Here, we present a rare recurrent hidradenocarcinoma in the

abdominal wall with no distant metastases.

Case presentation

A 56-year-old male presented to the Surgical Department of Al-Ramadi Teaching Hospital, Ramadi City, Iraq, with a gradually growing, painless swelling in the right lower aspect of the anterior abdomen that caused him no discomfort (Figure 1A). The mass started as a small lump and had been growing for the past 6 months before presentation. The patient denied any history of abdominal trauma. On physical examination, the mass measured 5 x 6 cm, solid, firm in consistency with irregular borders. The skin had mild redness over the swelling and the neighboring skin, it was fixed over the mass, but as a whole the mass was mobile to the underlying muscular layer. There were no associated systemic symptoms. Complete blood count and blood chemistries back with came no abnormalities. Differential diagnoses included; sebaceous cyst, lipoma, hemangioma, or an inflamed lymph node. It was excised under local anesthesia and was sent for histopathological evaluation. The patient, however, didn't bring the result of histopathology to the surgeon's attention. Eight months later, the patient comes back to the Surgical Department, complaining of a recurrent mass in the exact location, this time growing around the previous surgical scar. The dimensions were 5 x 4 cm on measurement. The mass looked similar to the previous presentation, having firm skin, irregular borders, mobile, but the skin was a little darker, elevated, reddishblue, and nodular (Figure 1B). There were no associated systemic symptoms and no palpable lymph nodes in any part of the body. The erythrocyte sedimentation rate returned elevated (58 mm/hour), but all other blood chemistries were within the normal ranges. The previous histopathological report, 8 months earlier, confirmed the diagnosis of hidradenocarcinoma (a malignant tumor of the adnexa). The diagnosis was confirmed as a recurrence of the previously

excised hidradenocarcinoma of the trunk. The mass was excised under general anesthesia, leaving safe margins all around the tumor. The excised mass measured 18 x 11 x 7 cm (Figure 2A), including safe margins: upper and lower excisional margins about 1.5 cm from the mass, both lateral excisional margins about 4-5 cm from the mass, and deep excisional margin about 1 cm from the mass. The tumor itself measured 7 x 5 cm and was encapsulated. Grossly, there were visible necrotic and hemorrhagic foci (Figure 2B). The histopathological examination showed that skin and subcutaneous tissue were infiltrated by deep dermal well-defined tumor masses and nodules composed of atypical large epithelial cells with infrequent mitoses arranged in solid cords and sheets, associated with extensive multifocal necrosis and hemorrhages filling some cystic cavities. The features were consistent with recurrent malignant nonmelanocytic skin adnexal tumor/solid and cystic eccrine hidradenocarcinoma-high-grade. All lateral, surrounding excisional safe margins were tumor-free (Figure 3). A positron emission tomography (PET) scan showed no distant metastasis. As such, according to the tumor, node, and metastasis (TNM) classification, the tumor stage was stage II. Since no lymph nodes were detected and after consultation with the Oncology Department, no other adjuvant therapy was suggested. The patient was advised to follow-up monthly for recurrence. After 5 years of follow-up, the patient was free of recurrence or distant metastasis. The patient gave his consent for the publication of the case with its related images.



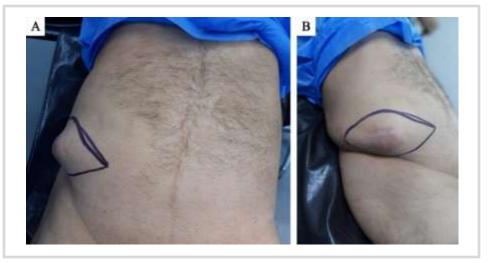


Figure 1. Pictures of the mass before the surgery. A) Anterior view of the abdomen. B) Lateral view of the abdomen.

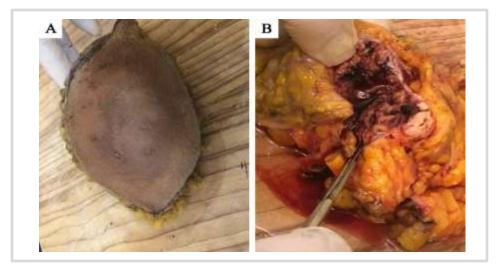


Figure 2. A) Picture of the mass from the outside after excision. B) Visible foci of necrosis and hemorrhages within the mass.

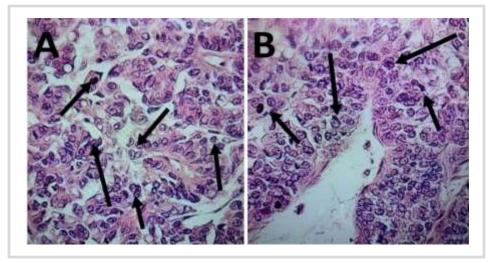


Figure 3. Variable sizes ducts lined by malignant epithelial cells with strong positive for keratin immunohistochemistry as shown by black arrows [Magnification x10 (A) and x40 (B)].

Discussion

Hidradenocarcinoma is an aggressive malignant tumor with a high tendency of local and distant metastasis (8, 9). It arises de novo but can also arise from nodular hidradenoma of the adnexa. The histological study is of prime importance in distinguishing between benign and malignant forms, which shows increased mitotic activity, local extension, widespread necrosis, and angiolymphatic invasion (10,11). The malignant form is grossly larger, more nodular, and asymmetric (12). Hidradenocarcinoma is further divided into eccrine and apocrine subtypes (Table 1).

Table 1. The characteristics of eccrine and apocrine types of hidradenocarcinoma.

Category	Eccrine Hidradenocarcinoma	Apocrine Hidradenocarcinoma	Supp.
			Ref.
Gland	Found throughout the body,	Found only in specific regions	(10)
Location	especially on palms, soles, and	such as axillae, anogenital region,	
	axillae.	abdomen & chest.	
Histological	Contains poroid cells with	Contains clear, squamoid, poroid	(11)
Characteristics	atypical features.	cells.	
Differentiation	Generally considered eccrine	Considered apocrine type when	(13)
	type if only poroid cells are	clear and squamoid cells are also	
	present.	observed.	

Mixed histological features suggest apocrine differentiation, making strict distinction unnecessary (13). This explains the tumor's ability to arise all over the body, even where normally apocrine/eccrine glands are not found (14). Most cases are asymptomatic; they rarely present with symptoms, and when they do, it is primarily due to metastasis. Local symptoms are rarely seen and include bleeding or ulceration. For that, most cases are presented in the late stages. This requires an evaluation of the patient nodal spread (seen in 39%) and visceral metastasis (seen in 28%) at the time of presentation. Indeed, hidradenocarcinomas have an aggressive nature, a high rate of recurrence (50% despite surgery), and metastasis (60% of patients in 2 years). As for the prognosis, although earlier studies had discussed poor prognosis, with an estimated 30% survival for five years yet most of those studies involved small sampling power (15); a recent study by Gao et al. examined 289 cases in 18 years-time and revealed that the hidradenocarcinomas have a good prognosis and the tumer size was the most

important variable that defines overall survival and cancer free period which was reported to be 60.21% and 90.52% respectivley (1). The biopsy is the main way to confirm the case. Imaging studies (Ultrasonography, CT, PET scan) are pivotal in excluding other differential diagnoses, guiding biopsy, excluding metastasis, and following up cases for prognostic value. Currently, the mainstay of treatment is wide surgical excision despite the absence of a clear guideline. The usefulness of lymph node dissection is controversial because it has not clearly shown. Similarly, adjuvant chemotherapy and radiotherapy use has not yet established (15). Other therapeutic been approaches are currently being researched, including PI3K/Akt/mTOR pathway inhibitors, targeted therapy, EGFR inhibitors, and hormonal therapy. However, there is no concrete evidence to implement them due to the lack of sufficient patients owing to the tumor's rarity (3). What is unique about this case is that it represented the second reported case of hidradenocarcinoma involving the abdominal wall without a history

that significantly contributed to the successful completion of this research.

in 2021 was at the site of previous trauma (7). The recurrence was short despite the absence of local and distant metastasis at both consultation times. The cases were well documented and followed for a long time (5 years). It underscores the success of early and aggressive surgical approaches and emphasizes the role of close

of prior trauma. The first case by Rehman et al.

Conclusion

Wide surgical excision with a safe margin was a satisfactory treatment for hidradenocarcinoma.

follow-up, even for such aggressive tumors.

Recommendations

Owing to the rarity and aggressive behavior of hidradenocarcinoma, particularly in uncommon sites such as the abdominal wall, surgeons should remain aware of its clinical similarity to benign skin or soft tissue swellings. Early detection, accompanied by thorough histopathological examination, is of utmost importance. It is essential for future surgeons to adopt a proactive approach to suspicious or recurrent skin lesions, favoring wide local excision and collaboration within a multidisciplinary team. Long-term follow-up is critical for the early identification of local recurrence or distant metastasis. This case underscores the importance of including hidradenocarcinoma in the differential diagnosis and highlights the value of applying sound surgical oncologic principles in its management.

Source of funding: No source of funding.

Ethical clearance: The study was approved by the Ethical Approval Committee of the University of Anbar, Anbar, Iraq (Reference number 39 on 22-8-2024). Informed consent was obtained from the patient for publication of the case with its related images.

Conflict of interest: None.

Acknowledgments:

The authors would like to express their sincere gratitude and deep appreciation to the University of Anbar for its generous support and for providing the essential resources and facilities

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تكرار الإصابة بسرطان الغدد العرقية في جدار البطن دون وجود نقائل بعيدة: تقرير حالة ومراجعة أدبية الإصابة بسرطان العانى، ٢ رائد العانى، ٣ وسن نورى، ٤ إسراء مهدى السودانى

الملخص

الخلفية: يُعد سرطان الغدد اللمفاوية ورمًا نادرًا وعدوانيًا، يتميز بمعدل تكرار موضعي مرتفع وانتشار ورم خبيث بعيد. لا توجد إرشادات علاجية حالية، وقد استُخدمت الجراحة للسيطرة على المرض الأساسي. حتى مع الاستئصال الجراحي الواسع، لا يُمكن التنبؤ بمسار الورم، وتوقعات البقاء على قيد الحياة ضعيفة. الموقع الأكثر شيوعًا هو فروة الرأس، تليها الأطراف، ونادرًا ما يكون الجذع.

الأهداف: عرض حالة نادرة من سرطان الغدد اللمفاوية المتكرر في جدار البطن دون انتشار ورم خبيث بعيد.

عرض الحالة: نُقدم حالة نادرة من سرطان الغدد اللمفاوية المتكرر في جدار البطن لدى رجل يبلغ من العمر ٥٦ عامًا، ولم يُكتشف لديه أي انتشار ورم خبيث بعيد، وقد عولج باستنصال جراحي واسع.

الاستنتاج: تُمثل هذه الحالة الثانية المُبلغ عنها لسرطان الغدد اللمفاوية في جدار البطن في الدراسات العلمية. يُحقق الاستئصال الجراحي الواسع للورم مع هوامش أمان واسعة نتائج ممتازة بعد متابعة لمدة ٥ سنوات.

الكلمات المفتاحية: سرطان الغدد الليمفاوية المتكرر؛ سرطان الغدد الليمفاوية في جدار البطن؛ النقائل البعيدة؛ تقرير حالة.

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تاريخ الاستلام: ٢٢ تشرين الثاني ٢٠٢٤

تاریخ القبول: ۲۸ شباط ۲۰۲۰

تاریخ النشر: ۲۰۲۰ نیسان ۲۰۲۰

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' قسم العلوم الأساسية - كلية الطب - جامعة ابن سينا للعلوم الطبية والصيدلانية – بغداد - العراق.