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Original Research Paper

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RADIOTHERAPY IN HIDRADENOCARCINOMA – A REVIEW OF TREATMENT STRATEGIES FOR THIS RARE MALIGNANCY

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ABSTRACT Hidradenocarcinoma is a rare and aggressive malignancy originating from sweat glands, presenting significant challenges in management. With limited available data and no well-established treatment guidelines, the optimal approach to treating hidradenocarcinoma remains undefined. This case report presents a patient with hidradenocarcinoma who underwent surgical resection followed by adjuvant radiation. The 34-year-old female patient experienced recurrent hidradenocarcinoma in the forearm, resulting in wide local excision and subsequent histopathological confirmation. Following surgery, adjuvant radiotherapy was administered to the tumour bed. Despite initial local control, the patient developed axillary lymph node metastasis, leading to axillary lymph node dissection and additional adjuvant radiotherapy. The patient demonstrated good local control without significant morbidity during the one year follow up period. This case highlights the successful use of radiotherapy for local control in hidradenocarcinoma and emphasizes the need for multidisciplinary approaches in managing this aggressive tumour. while further research is required to define the role of radiotherapy in metastatic hidradenocarcinoma, this case contributes to the existing literature and underscores the importance of collaborative efforts in optimizing treatment strategies for this rare malignancy.

KEYWORDS : radiotherapy, hidradenocarcinoma, rare tumour, surgery

INTRODUCTION

Hidradenocarcinoma is an extremely rare and aggressive malignant tumor originating from the sweat glands, characterized by a high risk of both local recurrence and distant metastasis. It comprises less than 0.001% of all tumors and typically manifests as a slow-growing, asymptomatic skin lesion[1]. The tumor commonly arises de novo, with a predilection for the head and neck region, rarely occurs on the distal extremities[2]. The average age of onset is around 50 years[3].

Given its rarity, there is limited body of data available on hidradenocarcinoma, resulting in an absence of well-established treatment guidelines. Surgical intervention represents the primary treatment modality for hidradenocarcinoma[4]. However, the post-surgical recurrence rate is alarmingly high at 50%, necessitating the consideration of adjuvant treatments like radiotherapy to improve local control[5]. The role of external beam radiotherapy as an adjuvant therapy has been a subject of controversy. Due to its scarcity of cases and the lack of comprehensive studies, the optimal approach to utilizing EBRT in hidradenocarcinoma remains uncertain. The challenge lies in determining the appropriate dose, fractionation schedule and target volume for radiotherapy. Despite these challenges, there is recognized need for adjuvant therapies to mitigate the risk of local recurrence. This case report presents a patient with hidradenocarcinoma who received radiotherapy as an adjuvant treatment following surgical resection.

Case Report

A 34-year-old female patient presented with a recurrent swelling on the medial aspect of her right forearm near the elbow joint. The patient had a history of similar complaints and had previously undergone wide local excision four times in a span of two years, with the most recent surgery in November 2016, resulting in a diagnosis of recurrent hidradenocarcinoma. After a symptom-free period of five years, the patient experienced a recurrence and sought further evaluation at our hospital. On examination, a well-defined, mobile, non-tender swelling measuring 5x4cm was identified over the ventral aspect of the right forearm, located 1cm distal to the elbow joint. Further evaluation using MRI of the proximal forearm revealed an ill-defined T2 hyperintense lesion measuring 5.3x2.2x1.3cm in the subcutaneous plane of the flexor aspect, extending into the underlying muscles, suggesting a malignant lesion (as shown in fig 1). The patient underwent wide local excision of the lesion, and the post operative histopathological examination confirmed recurrent hidradenocarcinoma involving the inferior surgical margin while the remaining margins were free. Following this the patient received adjuvant external beam radiotherapy to the tumour bed, administering a dose of 66Gy in 33 fractions, with 2Gy per fraction and five fractions per week using conventional **2D radiotherapy using cobalt-60**. The patient responded well to the treatment and remained symptom-free for six months, after which she reported right axillary swelling. A positron emission tomography-computed tomography scan revealed a necrotic right axillary lymph node measuring 1.3x1.2cm with necrosis and an SUV max of 6.3. Subsequently, the patient underwent right axillary lymph node dissection, which confirmed a malignant adnexal tumour indicative of recurrent hidradenocarcinoma. Histopathology specimen showed two out of nine lymph nodes with metastatic carcinoma with extranodal extension and lymphovascular invasion. As a result, the patient received adjuvant radiotherapy to the axilla, receiving a dose of 60Gy in 30 fractions via **Intensity modulated radiotherapy (IMRT), 2Gy per** fraction for 5 fractions a week. The patient tolerated the treatment well and has been on follow-up for one year, demonstrating food local control without significant morbidity.

DISCUSSION

Hidradenocarcinoma, an uncommon and aggressive tumor, poses a significant challenge in terms of management. Typically occurring in individuals aged between the fifth and seventh decade, with a slight female predilection, this rare malignancy predominantly affects the head and neck region and less frequently the extremities, trunk, abdomen, digits, elbows, and scalp. While hidradenocarcinoma can arise de novo, it may also develop from preexisting hidradenoma[6]. Clinically, it manifests as a solitary, subcutaneous, firm nodule, or erythematous plaque with telangiectasia, often

expanding gradually in a circumferential manner and leading to pain, discomfort, ulceration or bleeding[7]. Nodal involvement and visceral metastasis are observed in 39% and 28% of patients respectively[8]. The diagnosis of hidradenocarcinoma is confirmed through post operative histopathological examination, revealing two distinct cell types: dark spindle cells with eosinophilic cytoplasm and larger clear cells exhibiting atypical mitotic figures and nuclear pleomorphism. Immunohistochemistry shows positive expression of epithelial membrane antigen (EMA), carcinoembryonic antigen (CEA) and S100 protein and strong positivity for ki67 and p53[9]. Hidradenocarcinoma's occurrence in the elbow region is extremely rare, further complicating its effective management.

Wide local excision is considered the cornerstone of treatment for hidradenocarcinoma, while the necessity of sentinel lymph node biopsy remains a topic of debate[10]. In cases without distant metastasis, clinically involved lymph node regions should undergo dissection and irradiation, whereas clinically uninvolved primary lymph node regions may undergo either dissection or irradiation[4]. The local recurrence rate following surgery ranges from 10% to 50% and the five year post surgical survival rate is less than 30%.[11] Given the aggressive nature of the disease, adjuvant therapy is recommended to enhance local control. However, the role of radiotherapy and chemotherapy in hidradenocarcinoma is not well defined in the literature. Radiotherapy is selectively employed in patients with high-risk features, such as positive margins, lymph node involvement, perineural and lymphovascular invasion, deep infiltration, or poor differentiation[12]. The prescribed radiotherapy dose typically ranges from 66Gy to 70Gy to the tumor bed and 50Gy to the regional lymphatics, with newer technologies enabling improved tolerance[13]. Notable, a study by Yavel et. Al described the successful treatment of a scalp tumor with Mohs surgery followed by radiotherapy of 50Gy in 25 fractions delivered to the right posterior hemi scalp and postauricular and occipital lymph nodes, resulting in disease-free status after 12 months[2]. Additionally, Lalya et al. reported a case of hidradenocarcinoma in the parotid region, which recurred post-surgery but achieved good local control following high dose radiation of 66Gy to the tumour bed and 50Gy to the regional lymphatics[4].

The literature review revealed numerous case reports, with the majority focusing on the head and neck region, while only a limited number involved the extremities. Surgical excision emerged as the primary treatment approach across almost all cases, while adjuvant radiotherapy was utilized in a few instances. The utilization of chemotherapy or hormonal therapy was minimal and yielded diverse outcomes in the limited number of cases where they were employed (Table 1).

The presented case highlights an unusual instance of hidradenocarcinoma occurring in a less common age group and location. Our patient initially presented with a swelling on the forearm, which was eventually diagnosed as hidradenocarcinoma. Following standard protocols, wide local excision was performed on the lesion. However, postoperative evaluation revealed malignant cells in the inferior margin, indicating a high risk of recurrence. As a result, the patient received adjuvant radiation therapy to the post operative site, administering a dose of 66Gy in 33fractions. Despite this treatment six months later, axillary lymph node metastasis developed, necessitating axillary lymph node dissection and additional adjuvant radiotherapy in view of extranodal extension and multiple nodes positive. The patient has been consistently monitored and one year later follow-up examinations showed good local control without significant complications. It is important to note that the optimal treatment approach for hidradenocarcinoma remains undefined due to limited available data and the scarcity of cases. In this case we employed radiotherapy along with surgery to achieve good local control of the post operative site as well as to manage the metastatic lymph node. However, the role of radiation in the metastatic setting still requires further evaluation.

This case is significant due to the rarity and unusual presentation of hidradenocarcinoma. This contributes to the medical literature and enhances our knowledge base regarding hidradenocarcinoma. Furthermore, this case has important clinical implications. It highlights the challenges faced in managing hidradenocarcinoma and the need for multidisciplinary treatment approaches. The successful local control achieved through interventions such as wide local excision, radiation therapy and lymph node dissection demonstrate the potential effectiveness of this treatment strategy. It also highlights that radiotherapy can be employed with the aim of achieving local control in metastatic hidradenocarcinoma. Further research and clinical studies will help establish more specific guidelines and treatment recommendations, ultimately improving patient outcomes and providing more emphasis on the utilization of adjuvant radiotherapy in this challenging scenario.

CONCLUSION

In summary, hidradenocarcinoma is an exceptionally rare and aggressive tumor, presenting significant challenges in the management. Due to its scarcity of cases and limited data, the optimal treatment approach remains uncertain. This case underscores the necessity of a multidisciplinary treatment approach and highlights the potential efficacy of radiation therapy post surgery in achieving local control. Further research is imperative to develop specific guidelines and enhance patient outcomes particularly concerning the importance of radiotherapy in managing primary and metastatic hidradenocarcinoma.

Ethical Consideration

Ethical approval was not necessary for the preparation of this article.

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Nil.

Conflict Of Interest

The authors had no conflicts of interest to declare.

Table 1: Review Of Literature

Sl no	Author	Title	Sample	Year	Site	Treatment	Radiotherapy dose
1	Miller et al	Management of metastatic apocrine hidradenocarcinoma with chemotherapy and radiation	1	2015	axilla	Chemotherapy (3 cycles carboplatin followed by tamoxifen) followed by radiation therapy	50.4Gy in 28 fractions
2	Giannelli et al	Complete response in a patient with gynaecological hidradenocarcinoma treated with exclusive external beam radiotherapy and	1	2017	vulva	External beam radiotherapy followed by brachytherapy	60.2Gy in 28 fraction Simultaneous integrated boost followed by

		brachytherapy: a case report.					28Gy in 4 fraction with brachytherapy
3	Khan et al	Hidradenocarcinoma: Five Years of Local and Systemic Control of a Rare Sweat Gland Neoplasm with Nodal Metastasis	1	2018	Scalp	Wide local excision with right neck dissection followed by concurrent chemoradiotherapy(cisplatin)	60Gy in 30Fraction
4	Rafols et al	An Unusual Case of Lower Extremity Clear Cell Hidradenocarcinoma.	1	2020	thigh	Wide local excision followed by adjuvant radiotherapy	64Gy in 32fraction to the tumor bed and 50Gy in 25Fr to the lymph nodes
5	Rehman et al	Hidradenocarcinoma of the Abdominal Wall Treated with Wide Surgical Excision and Adjuvant Radiotherapy.	1	2021	Abdominal wall	Surgical excision followed by adjuvant radiotherapy	60Gy in 30Fraction

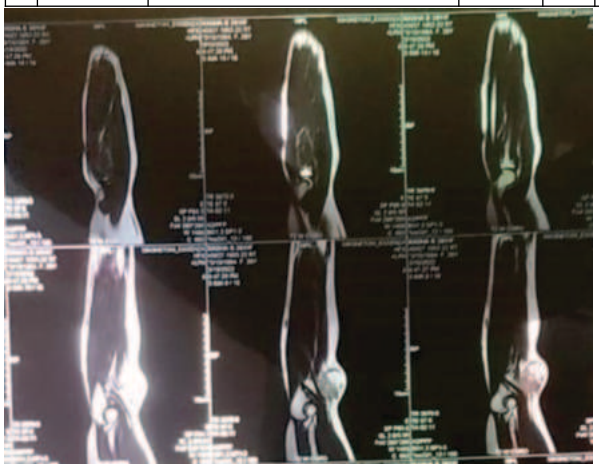

Figure 1 – MRI of right forearm

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