A Rare Case Of Pectoral Hidradenocarcinoma And Brief Review

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Abstract

Sweat gland tumours are in general uncommon and particular the malignant variety. Hidradenocarcinoma is one malignant variant with exceedingly low incidence with some studies reporting an incidence of 0.05%[1]. Although rare, these tumours have a significant metastatic potential and given the relative paucity of published data with only just over 50 case reports and small case series, there is little information available on the natural history and appropriate management for this disease[2]. Perianal, scalp, abdominal and distal extremity lesions have been reported; however, there are few reported cases of hidradenocarcinomas on the chest and pectoral region. We report below a rare case of hidradenocarcinoma in a male presenting as a cystic lesion on the left pectoral area. We provide a short review on hidradenocarcinomas and treatments currently applied.

CASE REPORT

A 52-year-old man presented to his local general practitioner (GP) on the 13 th of August 2010 for review of a left pectoral lump. He reported that the lump had been present for at least two months and clinically was consistent with a simple cyst. An attempt was made by the GP to excise the lesion; however, during the procedure it was noted that the lesion extended deep and there were a number of surrounding vessels suggesting increased vascularity. As a result, a biopsy was taken and histopathology indicated an appendageal tumour with both cystic and solid components. The tumour cells were relatively uniform and showed features of eccrine differentiation. They contained both duct-like structures as well as foci of squamous differentiation. Of further note was that groups and columns of tumour cells showed a locally infiltrative growth pattern in the surrounding sclerotic stroma. These features were consistent with a low-grade cystic and solid hidradenocarcinoma. He was subsequently referred to the Toowoomba Base Hospital (Queensland, Australia) for review in a surgical clinic for definitive management.

His past medical history included ischaemic heart disease (IHD) with previous myocardial infarction (MI) and unstable angina (UA). He had a history of hypertension, chronic obstructive pulmonary disease (COPD), obesity, gout, osteoarthritis and depression.

On the 13 th of September 2010 he underwent an elective wide local excision (WLE) of the left pectoral hidradenocarcinoma with flap repair under local anaesthetic due to a number of comorbidities. The procedure was conducted as a day case and he was successfully discharged home. The results of the histopathology confirmed residual low-grade hidradenocarcinoma with adequate excision margin and evidence of lymphovascular invasion. His recovery was complicated when he represented on the 20 th of September 2010 with a local wound infection. Clinically the previous excision site appeared erythematous with a small surrounding area of cellulitis. There was a small pustular discharge from the wound. Clinically there was no suggestion of local collection. Wound swabs grew both Staphylococcus aureus and Klebsiella oxytoca and he was subsequently treated with intravenous antibiotics both flucloxacillin and gentamicin for seven days. Clinically the wound improved and there was no dehiscence. He was discharged on the 27 th of September.

His case was discussed at a multidisciplinary meeting conducted at the Toowoomba Base Hospital on the 23 rd of October 2010 and the decision made was to consider radiation oncology referral. He was reviewed in the radiation oncology clinic and underwent adjuvant radiotherapy of 45 gray (Gy) in 15 fractions which was completed on the 13 th of December 2010. He has since been reviewed in the

surgical outpatient clinic with complete healing of the surgical wound and no evidence of local recurrence.

DISCUSSION

Sweat gland tumours are exceedingly rare with estimates suggesting they account for as little as 0.005% of all skin tumours[3]. Furthermore, when considering the sweat gland carcinomas, some studies in the United States have reported incidences around 0.05%[1]. The first reported case of sweat gland tumour dates back to 1865; however, it was not until the 1940s and later 1950s that interest had sparked the development of a classification system[3]. Following on, the largest case series of malignant sweat gland tumours was provided by Berg and McDivitt in 1968 with a total of 101 cases included and today still remains the largest case series published[4]. There still, however, remains a significant deficit in the form of a reliable classification system and set of guidelines for management of these tumours, particularly when considering the malignant variants such as hidradenocarcinoma. This is somewhat a reflection of the rarity and thus relative paucity of published data beyond a hand full of case series and reports.

Hidradenocarcinoma (also referred to as malignant nodular/clear cell hidroadenoma) is a malignant intradermal tumour of the sweat gland with a reported incidence in the United States of less than 0.05%[1]. Collectively this malignant tumour accounts for up to 6% of malignant eccrine tumours and represents the malignant equivalent of the hidradenoma[1,5]. Typically the tumour originates in the ductal or secretary part of the sweat gland and presents clinically as an asymptomatic solitary skin lesion measuring approximately 1-5cm[1,6]. Characteristically it is often covered by a pink, purple of blue section of skin which may be intact or ulcerated. Generally it is slow-growing but may undergo a rapid phase of growth in a short period of time[1,6]. Clinically there are no unique distinguishable features and patients are asymptomatic with any obvious influence from the primary lesion other than pain, discomfort, bleeding upon physical contact, or ulceration[1]. At an unknown point in time, the tumour transitions into an aggressive form with expansion to regional or distant metastatic sites, most commonly lymph nodes[1].

Histologically there are currently no standardised guidelines for diagnosis. It is often difficult to differentiate hidradenocarcinoma from the benign equivalent hidradenoma. The two types share the same composition of clear cells, squamoid cells, oncocytoid cells and transitional elements; however, the distinguishing features generally exist in the fact that hidradenocarcinoma usually shows features cytoarchitecturally consistent with malignant tumour including asymmetry, infiltrative growth, intravascular invasion, necrosis, cellular and nuclear pleomorphism and atypical mitotic figures, to name a few[5]. There are times, however, this distinction is difficult. There has been some suggestion from the limited pathological case series that immunohistochemical studies may assist in helping make this distinction. Ko et al. (2006) showed that hidradenocarcinoma is strongly positive for ki67 and p53 along with a number of other immunohistochemical stains and it is hoped that these will play a significant role in future diagnostic studies[7].

The location of the primary lesion of hidradenocarcinoma is another area of interest which is still poorly understood. We presented here a case of left pectoral region hidradenocarcinoma of which very few cases are present in the literature. Hidradenocarcinoma is a sweat gland tumour arising from eccrine sweat glands which open directly onto the skin, not apocrine glands which are specialised sweat glands that arise in association with hair follicles[3]. The distinction is important in that eccrine glands can exist literally anywhere on the skin surface whereas apocrine glands are concentrated in hair-rich environments such as the axilla, groin, perineum and eyelids[3]. Thus based on this distribution, hidradenocarcinomas can potentially occur anywhere and there have been reported cases including the scalp, dura of the brain and neck as well as abdomen, chest, breast and limbs[8,9,10]. From what little evidence and cohort studies are available it appears there is no difference in behaviour of these tumours based on location. In one case series of 55 patients, most lesions (48%) occurred in the head and neck region, whilst other studies have reported a greater preponderance in the genital skin and perineum[3]. Furthermore, it appears that a history of previous radiation therapy to the region with the primary lesion may be a risk factor for the development of this malignant tumour[3]. Age especially may be a risk factor with peak incidence suggested as between 50-70 years of age[3]. What is clear, however, is that further studies particularly focusing on the molecular basis of these sweat gland tumours need to be conducted to help predict location and whether this impacts on the natural history of the disease.

Disease recurrence is an area of great concern regarding hidradenocarcinomas and the primary impetus behind further research into understanding these sweat gland malignancies.

Recurrence rates of 50-60% have been reported, some within the first 2 years of primary treatment[6,8]. As a result, prognosis is poor with 5-year disease-free survival estimated as less than 30% and even higher in the available smaller case series[6]. Metastatic disease spreads initially from primary lesion to regional lymph nodes and extensively haematogenously to periesophageal, peribronchial, periaortic and retroperitoneal lymph nodes and to bones, vertebrae, ribs, pelvis, lung parenchyma and pleura[6]. As a result, further studies are required to help reduce the mortality and morbidity of this disease.

Current treatment philosophies rely mainly on individual centre experience based on relatively small case series. It is still unclear what the best method for treatment is. Most clinicians conclude that the primary treatment of choice is wide local excision with regional lymph node dissection if required[1,6]. Wong et al. (1994) suggested a 2cm margin for primary disease and local recurrence be used and more recently a 3-5cm wide local excision has been used. These are purely experiential and no large prospective trials are available regarding outcomes including local recurrence and survival based on these suggestions. Radiotherapy has been described in a select number of cases mainly dependent on histopathological classification[1,6]. Moderately or poorly differentiated tumours likely display higher rates of local recurrence and metastasis and thus current practice is to consider radiotherapy in these select cases[6]. The choice was made in the case presented above for radiotherapy based on lymphovascular invasion and concern for recurrence on regional spread. Chemotherapy and sentinel lymph node biopsy is still an area of controversy; however, early reports suggest a role of sentinel biopsy for detecting subclinical metastasis[1,6].

Ultimately, it is apparent that much work needs to be conducted into better understanding hidradenocarcinoma and, for that matter, malignant sweat gland tumours. Unfortunately, given the rarity of these tumours, it is difficult to amass large trials with sufficient numbers to generate a significant result. Histopathological classification is also an area which needs revision and certainly a consensus needs to be approached sooner than later to ensure a strict guideline for diagnosis is reached. This will hopefully help reduce false negative diagnosis. Finally, current treatment guidelines should be adopted regarding wide local excision; however, regarding adjuvant therapy, discussion at multidisciplinary meetings should become the norm such that a consensus is reached regarding whether adjuvant therapy should be adopted, given no current studies implicating a benefit.

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