The Aesthetic and Oncological Challenges in the Management of an Atypical Nodular Hidradenoma of the Pinna

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Abstract Nodular hidradenomas are benign adenomatous tumours of sweat gland origin, which commonly arise from the skin of the head, neck and extremities. Some of these benign appearing lesions exhibit aggressive clinical behaviour with rapid growth, pigmentation and ulceration. These tumors are designated as “atypical nodular hidradenomas” and are known to histologically possess some focal atypical features i.e. nuclear pleomorphism, focal necrosis or prominent mitotic activity. Nodular hidradenomas involving the pinna are rare and to the best of our knowledge there have been no prior reports in the English language literature. We describe a unique case of a 45-year-old woman with an atypical nodular hidradenoma of the left pinna and discuss the aesthetic and the oncological issues pertaining to the reconstruction of the pinna.

Keywords Nodular hidradenoma · Atypical nodular hidradenoma · Malignant hidradenoma

Introduction

Nodular hidradenomas are benign adenomatous tumours of sweat gland origin, which commonly arise from the skin of head, neck and the extremities [1]. The malignant counterpart of hidradenoma (malignant hidradenoma or hidradenocarcinoma)

is an extraordinarily rare tumour. Rendering a diagnosis of hidradenocarcinoma can be challenging to the pathologists as some of these lesions do not possess the well-established criteria of malignancy [2]. Certain benign-appearing hidradenomas show focal atypical features and these tumors have been designated as atypical hidradenomas [3]. Herein we report a case of a 45-year-old woman with an atypical nodular hidradenoma of the pinna and discuss the aesthetic and the oncological issues pertaining to the reconstruction of the pinna. Nodular hidradenomas involving the pinna are rare and to the best of our knowledge there have been no prior reports in the English language literature.

Case Report

A 45-year-old lady with no comorbid illnesses presented to our centre with an ulcerated nodule involving her left pinna. It began as a pea sized nodule in the antihelix region twenty years prior, an increase in the size of the nodule with associated ulceration over a period of four months prompted her to seek further treatment with us. Her general examination and examination of the other systems were normal. Clinical examination revealed an ulcerated nodule 4×3 cm along the anthelical fold of the left pinna. It was seen to superiorly extend up to the scaphoid fossa and inferiorly up to the level of the anti-tragus. The medial extent was up to the level of the concha-symba. The external auditory canal was however free (Fig. 1). There was no significant parotid or cervical adenopathy. A CT scan of the head and neck region confirmed the origin and extent of the swelling (Fig. 2). The patient was taken up for surgery, which entailed a wide local excision after a wedge biopsy from the ulcer suggested a possibility of carcinoma of skin adnexal origin. The pinna was advanced to achieve a primary closure of the helix and the antihelix after a frozen section confirmation of tumor free margins. The tissue in the region of the acoustic

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Fig. 1 **a** Clinical picture at presentation showing the nodular ulcerated lesion in the left pinna
b CT scan reconstructed image showing the extent of the lesion in the left pinna

meatus was left to heal by secondary intention. The final histopathology showed a $3 \times 2.5 \times 1.5$ cm tumour, showing solid nests of tumor cells separated by fibrous stroma. The cells were small and round with moderate cytoplasm and vesicular nuclei and occasional mitotic figures. The tumor lacked necrosis and did not demonstrate either angiolymphatic or perineural invasion. Although the tumor showed an infiltrative pattern the underlying cartilage was uninvolved, the histological features suggested a diagnosis of atypical nodular hidradenoma (Fig. 3a–d). The patient is on follow up with a reasonable cosmetic and functional outcome (Fig. 4). A delayed definitive reconstruction of the pinna is being planned for the patient.

Discussion

Nodular hidradenoma was first described by Liu in 1949, who termed it as eccrine acrospiroma. It is also known by several other terminologies i.e. clear cell hidradenoma, solid cystic hidradenoma, clear cell myoepithelioma, eccrine sweat gland adenoma and large cell hidradenoma among others [4, 5].

Nodular hidradenomas can occur at any stage of life but are usually known to occur between the fourth and the eighth decades of life and are more commonly in females. They are usually seen as asymptomatic well defined, slowly growing tumours in the dermal layer of the skin [6]. Some of the benign appearing adenomas can show aggressive clinical behaviour

with rapid growth, pigmentation and ulceration. They are designated as “atypical hidradenomas” and are known to histologically possess some focal atypical features which may include nuclear pleomorphism, focal necrosis, or prominent mitotic activity. These features are more pronounced in cases of malignant transformation and are histologically appreciated by nuclear atypia, increased number of mitotic figures, infiltrative pattern, angiolymphatic or perineural invasion with areas of necrosis [2].

Routine haematoxylin and eosin staining is usually sufficient for a diagnosis of nodular hidradenoma and its variants, immunohistochemical analysis is not considered mandatory. It has however been suggested by some authors that atypical hidradenomas have a significantly different immune-profile from its malignant counterpart showing a lower Ki-67 and PHH3 expression and a significantly different EGFR expression [3].

Wide excision with clear margins and appropriate reconstruction is the treatment of choice for all atypical nodular and malignant nodular hidradenomas [7, 8]. Reconstruction of the ear is a challenge considering its the high ratio of skin coverage to cartilage, inconsistent blood supply and complex three-dimensional anatomy with its many curves, peaks and valleys. The ear has got an aesthetic significance apart from having a functional significance as it forms a supporting structure for spectacles and ear rings.

Defects of the middle one-third of the pinna are the most difficult to reconstruct. A wide array of flaps, grafts and primary

Fig. 2 **a** Intraoperative photograph following wide excision of the tumor
b Specimen photograph

Fig. 3 **a** H&EX10 Tumor showing solid nests of tumor cells separated by fibrous stroma **b** H&E X 40 Tumor showing infiltrative growth pattern. However underlying cartilage is not infiltrated. Solid islands of cells are separated by dense fibrous stroma **c** H&E X 40: Tumor showing polygonal to small basaloid cells with dark stained nuclei. Arrow points to a mitotic figure **d** IHC Ki 67 $\times 20$ Proliferative activity is very high (50 %)

closure techniques have been used in the reconstruction of the pinna; the choice of reconstruction is largely determined by the size and location of the defect [9]. Smaller defects of the middle of third of the pinna may be closed primarily by converting the defect into a wedge as was done in our patient. The disadvantage of this approach is that it has a direct impact on vertical height of the pinna. Healing by secondary intention has been described as a valuable tool as was also used in our patient. Tubed flaps provide cosmetically unacceptable results due to the lack of a

cartilaginous framework for support and are generally not recommended for the middle one third defects. The larger defects are ideally reconstructed using a complex two-staged composite flap using autogenous cartilage and full-thickness retroauricular skin grafts. A “delayed reconstruction” as a method of care has been suggested for the skin cancers of the pinna. This is believed to allow the surgeon time to choose the best option for reconstruction and also allows the patient better choices following tumor extirpation with significantly reduced costs of care [10].

Atypical nodular hidradenomas, due to its unique histology and immune-profile is believed to be having an increased tendency for local recurrences, but without distant metastasis. The clinical significance of the above finding is however unclear [11].

In conclusion our report highlights the unique clinico-pathological features of atypical nodular hidradenomas of the pinna and emphasizes the need for of complete local excision with clear margins. Reconstruction of the defects of the middle one third of the pinna, especially those involving the cartilaginous framework is a major technical challenge and the management options needs to be individualized.

Fig. 4 Post operative clinical photograph two months following reconstruction of the pinna

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