Aggressive Digital Papillary Adenocarcinoma in a Young Female—a Rare Presentation

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Received: 20 October 2010 / Accepted: 27 December 2010 / Published online: 22 January 2011
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Abstract A 20 year old female presented with a recurrent soft tissue swelling over the medial aspect of proximal phalanx of left little finger. It had recurred one year after excision. There was no lymphadenopathy or bony involvement. Previous histopathology reports were not available. After excision histopathological diagnosis was aggressive digital papillary adenocarcinoma. Later Ray's amputation of little finger was planned. Aggressive digital papillary adenocarcinomas are rare sweat gland tumors which occur on hands, fingers, and toes. They have high local recurrence rate and may metastasize to lungs and lymph nodes.

Keywords Aggressive digital papillary adenocarcinoma · Recurrence · Eccrine gland tumor

Introduction

Aggressive digital papillary adenocarcinomas are rare adnexal tumors with high local recurrence and variable biological behavior. They characteristically occur on hands, fingers, and toes. Median age of occurrence is 52 years. It is rare in females with male: female ratio of 9: 1 [1].

Aggressive digital papillary adenoma [ADPA] and aggressive digital papillary adenocarcinoma [ADPACa]

form a morphological spectrum. It is advisable to regard them as potentially malignant and the designation adenocarcinoma is the preferred term, since histological features are not predictive of the outcome [2, 3].

Case Report

A 20 year old female presented with history of recurrent soft tissue swelling on medial aspect of the proximal phalanx of the left little finger. It extended upto the web space and measured 4×3×2 cm. The overlying skin was normal (Fig. 1). There was no lymphadenopathy or bony involvement. It was excised 1 year back and had recurred over a period of 1 year. Imaging studies were not done and previous histopathological reports were not available. Excision under anaesthesia was done (Fig. 2).

Histopathological examination showed reticular dermis replaced by the tumor tissue. Tumor showed varied growth patterns, such as comedo, cribriform, solid, papillary, cystic and glandular. The individual tumor cells had vesicular to hyperchromatic nuclei with moderate pleomorphism. Mitotic rate was 2–3 / HPF. There were areas of hemorrhage, necrosis and desmoplasia. It was reported as aggressive digital papillary adenocarcinoma (Figs. 3, 4 and 5). Later Ray's amputation of the little finger was planned. Post amputation follow up did not show any recurrence.

Discussion

Aggressive digital papillary tumors are rare sweat gland neoplasms, first described by Helwig E.B in 1984 [2] and later by Kao G.F. in 1987 [4]. They characteristically occur

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Fig. 1 Recurrent soft tissue swelling over the proximal phalanx of the left little finger on the medial aspect, extending upto the web space

on the hands, fingers and toes. These tumors were so named because of their potential for aggressive local growth, resulting in high recurrence rate and their predominant digital location, commonest being the volar aspect of digital tip between the nail bed and distal interphalangeal joint [4]. They occur predominantly in men with mean age of 52 years. Their average size is 1.7 cm. They may be present for 2 months to 15 years with a mean of 29 months. Pain is a frequent presenting complaint. They may present as a simple nail bed infection [6].

When these tumors were first described, they were divided into APDA and APDACa. Subsequent follow-up of lesions designated as adenomas found a metastatic potential of 14%. Thus all of these tumors are more correctly classified as carcinomas with no benign counterpart. They are locally aggressive with a 50% local recurrence rate; 14% of tumors metastasize [7].

Fig. 2 Operative site after excision of aggressive digital papillary adenocarcinoma

Fig. 3 Section (4 x) showing tumour tissue arranged in solid, comedo and cribriform pattern surrounded by cystic space and haemorrhage

They are divided into APDA and APDACa only on histological ground based on glandular differentiation, necrosis, cytologic atypia, mitotic rate and invasion of soft tissues, bone and blood vessels [4]. The histologic features closely resemble a metastatic infiltrating duct cell carcinoma of the breast. Metastatic disease most commonly affects the lungs and lymph nodes. Sentinel lymph node biopsy in addition to local treatment has been performed in rare instances, but its value has not been assessed in larger prospective trials as yet [8].

Sixty-seven cases of aggressive digital papillary adenoma and adenocarcinoma were studied according to their clinical characteristics and histologic features. Fifty of these were originally diagnosed as adenoma and 17 as adenocarcinoma. Follow up on 45 (67%) of the patients was obtained. None of the clinical or histologic parameters

Fig. 4 Section (10 X) shows cystic space lined by papillary structures and solid areas of tumor tissue in the reticular dermis

Fig. 5 Section (40 X) from the tumor exhibiting solid, cribriform and comedo pattern separated by thick fibrous connective tissue

studied were found to be predictive of recurrence or metastasis, indicating that the originally proposed criteria for distinguishing between benign (adenoma) and malignant (adenocarcinoma) do not predict biologic behavior. When primary tumors were treated by subsequent re-excision or amputation, only one recurred (5%). When not so treated, 11 recurred (50%) regardless of the original diagnosis ($p < 0.05$). Metastasis occurred in six (14%) cases and in three cases led to the death of the patient. Pulmonary metastases were observed in five cases. No effective treatment for widespread metastatic disease has yet been developed [5].

The surgical management of APDACA should include excision of the tumor on first hand to prove the diagnosis. Amputation of the toe / finger should be done to prevent recurrence and also to prevent metastases.

In our case, the patient was a young female. The site of the lesion and histological features were classical of APDACA. She had recurrence, but no metastasis. We

planned Ray's amputation of the little finger of the hand. Post amputation follow up did not show any recurrence.

Conclusion

Aggressive digital papillary adenocarcinomas are rare sweat gland neoplasms, which are rare in females. They characteristically occur on the hands, fingers and toes. They have potential for aggressive local growth, resulting in high recurrence rate. Metastases can occur to the lungs and lymph nodes. The surgical management should include excision of the tumor and amputation of the toe/finger to prevent recurrence and metastases.

References

1. Chi CC, Kuo TT, Wang SH (2007) Aggressive digital papillary adenocarcinoma: a silent malignancy masquerading as acquired digital fibrokeratoma. *Am J Clin Dermatol* 8(4):243–245
2. Helwig EB (1984) Eccrine acrospiroma. *J Cutan Pathol* 11:415–420. doi:10.1111/j.1600-0560.1984.tb00398.x
3. Jih DM, Elenitsas R, Vittorio CC (2001) Aggressive digital papillary adenocarcinoma: a case report and review of literature. *Am J Dermatopathol* 23:154–157
4. Kao GF, Helwig EB, Graham JH (1987) Aggressive digital papillary adenoma and adenocarcinoma: a Clinicopathological study of 57 patients with histochemical, immune histochemical and ultra structural study. *J Cutan Pathol* 14:129–146
5. Duke WH, Sherrod TT, Lupton GP (2000) Aggressive digital papillary adenocarcinoma. *Am J Surg Pathol* 24:775–784
6. Gorva AD, Mohil R, Srinivasan MS (2005) Aggressive digital papillary adenocarcinoma presenting as a paronychia of the finger. *J Hand Surg J Br Soc Surg Hand* 30(5):534. doi:10.1016/j.jhsb.2005.01.011
7. Keramidas EG, Miller G, Revelos K, Kitsanta P, Page RE (2006) Aggressive digital papillary adenoma-adenocarcinoma. *Scand J Plast Reconstr Surg Hand Surg* 40(3):189–92. doi:10.1080/02844310600763741
8. Matsysik TS, Port M, Black JR (1990) Aggressive digital papillary adenoma: a case report. *Cutis* 46:125–127