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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Kelser de Souza Kock, PhD, Physiotherapist, Professor, Department of Physiotherapy/Medicine, University of South of Santa Catarina, Tubarão 88700000, SC, Brazil. kelserkock@yahoo.com.br

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CASE REPORT

Malignant form of hidroacanthoma simplex: A case report

Yi-Fei Yang, Rong Wang, Hui Xu, Wei-Guo Long, Xiao-Hui Zhao, Yu-Mei Li

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Yi-Fei Yang, Rong Wang, Hui Xu, Yu-Mei Li, Department of Dermatology, The Affiliated Hospital of Jiangsu University, Zhenjiang 212001, Jiangsu Province, China

Wei-Guo Long, Xiao-Hui Zhao, Department of Pathology, The Affiliated Hospital of Jiangsu University, Zhenjiang 212001, Jiangsu Province, China

Corresponding author: Yu-Mei Li, Doctor, MD, PhD, Chief Doctor, Department of Dermatology, The Affiliated Hospital of Jiangsu University, No. 438 Jiefanglu Road, Zhenjiang 212001, Jiangsu Province, China. l.yumei@aliyun.com

Abstract

BACKGROUND

This paper presents a case of malignant hidroacanthoma simplex (HAS) and review the literature of previous cases to summarize the histopathological and immunohistochemical features and display the dermoscopic features of malignant HAS.

CASE SUMMARY

We present an 88-year-old Asian female with malignant HAS. The diagnosis was made according to the histopathological and immunohistochemical results after biopsy. Previous case reports of malignant HAS were retrieved from PubMed to characterize the histopathological and immunohistochemical features. We also display the dermoscopic features of malignant HAS that have not been reported.

CONCLUSION

Our findings demonstrate that prompt surgical treatment is an effective strategy for malignant HAS. Histopathology and immunohistochemistry are valuable diagnostic tools. This is the first case report to display the dermoscopic features of malignant HAS, and we speculate that dermoscopy may contribute to the diagnosis of malignant HAS.

Key Words: Malignant hidroacanthoma simplex; Dermoscopy; Immunohistochemistry; Histopathology; Diagnosis; Case report

Core Tip: Malignant hidroacanthoma simplex (HAS) is a clinically uncommon malignant cutaneous tumour with only a few case reports. Herein, we present an additional case of malignant HAS. By combining this case with a literature review of previous cases retrieved in PubMed, we summarize the histopathological and immunohistochemical features of malignant HAS and found that timely surgical operation is an effective treatment. Furthermore, we first display dermoscopic features of malignant HAS and speculate that dermoscopy may be a valuable tool for the early diagnosis of malignant HAS.

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INTRODUCTION

Hidroacanthoma simplex (HAS), a rare tumour arising from terminal sweat ducts, was initially characterized in 1956[1]. As an intraepidermal variant of eccrine poroma, the vast majority of HAS cases are benign[2,3]. Malignant transformation is an infrequent occurrence and no dermoscopic features of malignant HAS have yet been reported. Here, we present a case of malignant HAS and review the literature of previous cases to summarize the histopathological and immunohistochemical features and display the dermoscopic features of malignant HAS.

CASE PRESENTATION

Chief complaints

An 88-year-old Asian female presented to the Department of Dermatology with a complaint of a cutaneous mass on the right thigh for 30 mo.

History of present illness

The lesion first appeared as a slightly elevated papule 30 mo prior and then gradually enlarged to become a brown-coloured verrucous lump.

History of past illness

The patient had a history of hypertension for more than 10 years and the patient had undergone surgery for squamous cell carcinoma of the tongue 21 years prior.

Personal and family history

The patient denied any family history of autoimmune disease, malignant tumours or other genetic conditions.

Physical examination

The vital signs of the patient were as follows: body temperature, 36.8°C; heart rate, 82 beats per min; blood pressure, 130/80 mmHg; and respiratory rate, 19 breaths per min. Furthermore, a well-demarcated tumour (3.0 cm × 2.8 cm) associated with exulceration was notted on the right thigh (Figure 1A), and no palpable lymph nodes were detected.

Laboratory examinations

The glomerular filtration rate was 59 mL/min; uric acid was 456 μmol/L; triglyceride was 2.74 mmol/L; and total cholesterol was 6.30 mmol/L. Routine examination of the patient's stools, blood and urine did not indicate any abnormalities.

Imaging examinations

Dermoscopy revealed hyperkeratosis with fine scales, dotted vessels and linear telangiectasis in the papilla, a white to red structureless area, and exulceration (Figure 1B and C).

MULTIDISCIPLINARY EXPERT CONSULTATION

Haematoxylin-eosin staining was used for histopathological examination, and immunohistochemistry was performed according to the instructions of the Maxvision 2 HRP-Polymer anti-Mouse/Rabbit Immunohistochemistry (IHC) Kit. Histopathological examination revealed irregularly thickened epidermis with hyperkeratosis and parakeratosis, papillary formation in local areas, a multinodular pattern of tumour nests, and widened, blunted epithelial feet within the epidermis (Figure 2A). There was moderate or abundant cytoplasm, which was slightly less stained than the surrounding

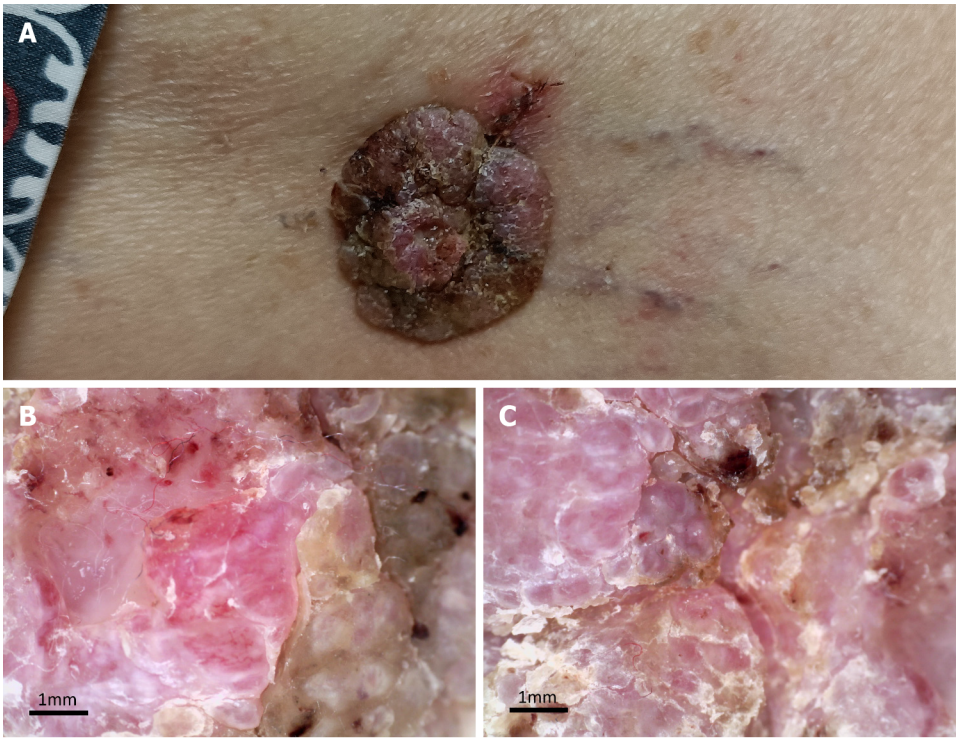

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Figure 1 Clinical image and dermoscopic examination of the tumour. A: Clinical picture; B and C: Dermoscopic images of the lesion. (B) ×50, (C) ×50.

residual squamous epithelium (Figure 2B). The neoplastic cells exhibited pleomorphism with nuclear atypia and mitotic figures, and scattered dyskeratotic cells were observed within the epidermis (Figure 2C). No invasive growth was observed. Immunohistochemical staining was positive for cytokeratin 5/6 and epithelial membrane antigen (Figure 2D and E). Carcinoembryonic antigen expression was absent in neoplastic cells, but it highlighted the presence of ductal structures (Figure 2F).

FINAL DIAGNOSIS

The final diagnosis was established as malignant hidroacanthoma simplex.

TREATMENT

Radical resection and flap transplantation were performed under general anaesthesia. Vacuum sealing drainage was used to promote wound recovery after surgery.

OUTCOME AND FOLLOW-UP

There was no recurrence in the six-month postoperative follow-up.

DISCUSSION

HAS is a rare form of the four subtypes of eccrine poroma (EP) and seldom undergoes malignant transformation. We searched PubMed using the keyword ‘malignant hidroacanthoma simplex’ and reviewed 10 case reports of malignant HAS (Tables 1 and 2)[2-11]. Malignant HAS primarily affects the extremities, and the majority of patients are over 70 years old. Although malignant HAS has the potential to regionally and distantly metastasize[10], prompt surgery, including moth micrographic surgery[11] has been demonstrated as an effective treatment strategy with no instances of recurrence.

Malignant HAS lacks specific clinical manifestations and usually presents as pigmented wart-like lumps. Malignant HAS is often mistaken for other cutaneous neoplasms such as Bowen’s disease (BD) or seborrheic keratosis (SK).

Table 1 Summary of 10 malignant hidroacanthoma simplex cases

Case	Ref.	Age	Gender	Duration (yr)	Location	Size (cm)	Color	Ulceration	Clinical feature	Recurrence
1	Sun Kim *et al*[2], 2012	69	F	1	Suprapubic area	2 × 2	Pigmented	(-)	Verrucous nodule	No Recurrence
2	Lee *et al*[3], 2006	71	F	NR	Right knee	1.8 × 2.0	Pigmented	(-)	Hyperkeratotic tumor	NR
3	Ishida *et al* [10], 2009	72	M	3	Right thigh	1.7 × 1.2	Brown to black	(-)	Flat plaque	Liver and bone metastases before surgery
4	Bardach *et al* [4], 1978	70	F	15-20	Right leg	Not mentioned	Red	(-)	Irregularly shaped and crusted	NR
5	Yang *et al* [11], 2022	80	M	5	Left foot	1.2 × 0.8	Black	(-)	Elevated nodule	No Recurrence
6	Kohli *et al* [5], 2015	79	M	NR	Scalp	0.6 × 0.5	Pink	(+)	Papule	No Recurrence
7	Piqué *et al* [6], 1995	73	F	15	Right leg	3 × 3	Pigmented	(-)	Verrucous lesion	NR
8	Ansai *et al* [7], 1994	75	M	2	Right ankle	2.5 × 3.3	Pigmented	(-)	Verrucous plaque	No Recurrence
9	Lee *et al*[8], 2000	67	F	16	Right thigh	5 × 7	Pigmented	(-)	Verrucous lesion	NR
10	Takano *et al* [9], 1989	74	F	NR	Left thigh	1.5 × 1.5	Light brown	(+)	Elevated nodule	Died of cardiac and respiratory failure

NR: Not reported.

Table 2 Histopathological and immunohistochemical features of 10 cases

Case	Intraepidermal nests	Sharp delineation	Hyperkeratosis or acanthotic epidermis	Invasive growth	Nuclear and cytoplasmic pleomorphisms	Mitotic figure	Ductal structure	Immunohistochemistry
1	(+)	(-)	(+)	(+)	(-)	(+)	(+)	EMA (+), CK10 (+), CK14(+)
2	(+)	(+)	(-)	(-)	(-)	(+)	(-)	EMA (+), CEA (+)
3	(+)	(+)	(+)	(+)	(+)	(+)	(+)	NR
4	(+)	(+)	(+)	(-)	(-)	(-)	(+)	NR
5	(+)	(+)	(+)	(-)	(+)	(+)	(+)	CK5/6 (+), P63 (+), CEA (-) CK7 (-)
6	(+)	(+)	(+)	(-)	(+)	(+)	(+)	CytokeratinAE1/3 (+), EMA (+), CEA (+), CK7 (+)
7	(+)	(+)	(+)	(-)	(+)	(+)	(+)	CAM5.2 (+), CEA (+), S100 (+), EMA (+)
8	(+)	(+)	(+)	(+)	(+)	(+)	(-)	NR
9	(+)	(+)	(+)	(+)	(+)	(-)	(-)	EMA (+), CEA (+), S100 (+)
10	(+)	(+)	(-)	(-)	(+)	(-)	(-)	NR

EMA: Epithelial membrane antigen; CEA: Carcinoembryonic antigen; CK10: Cytokeratin10; CK14: Cytokeratin 14; CK5/6: Cytokeratin 5/6; CK7: Cytokeratin 7; CK19: Cytokeratin 19; P63: Transformation-related protein 63; CAM5.2: Cytokeratin CAM5.2; S100: S100 proteins; NR: Not reported; (+): Positive; (-): Negative.

Histopathology is an indispensable tool for diagnosing malignant HAS. Through the analysis of 10 cases, we aimed to identify the pathological features of malignant hidroacanthoma simplex: (1) Tumour nests are well-demarcated, and the epidermis often exhibits irregular acanthosis; (2) Most tumour cells are characterized by vacuolated nuclei and small nucleoli; (3) Some tumour nests show invasive growth, whereas neoplastic cells exhibit nuclear and cytoplasmic

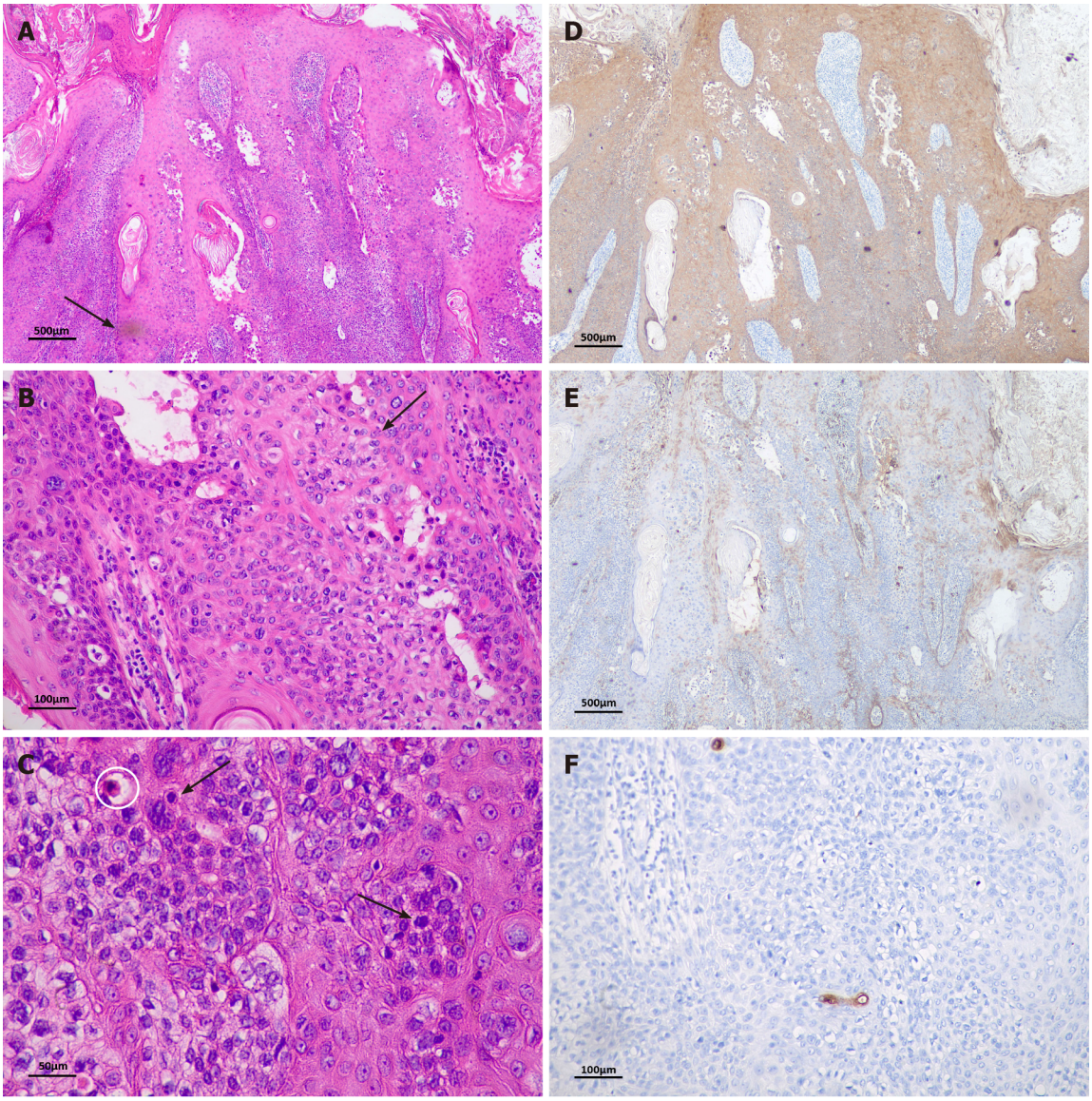

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Figure 2 Histopathological and immunohistochemical analysis. A-C: Haematoxylin and eosin staining of resected specimen (A) ×40, (B) ×200, (C) ×400. A: Blunted epithelial feet (white arrow); B: Neoplastic cells were less stained than the surrounding epithelium (black arrow); C: Dyskeratotic cells (white circle) and mitotic figures (black arrows); D: Immunohistochemical staining showed positivity for CK5/6 (×40); E: EMA (×40); F: CEA was only expressed in ductal structures ×200.

pleomorphisms and mitotic figures; and (4) Ductal differentiation can be observed. IHC has emerged as a powerful diagnostic examination. As a subtype of EP, HAS can arise from the ductal part of either the large or small sweat glands. Previous research has revealed that the majority of EP tumour cells express cytokeratin 5 (CK5) and cytokeratin 14, and squamous epithelial-like sections express cytokeratin 1 and cytokeratin 10, whereas ductal areas express cytokeratin 77 and cytokeratin 6[12]. Epithelial membrane antigen (EMA) has been reported to be positive in the cytoplasm of neoplastic cells of HAS and negative in SK and BD[13]. CEA was found to highlight the ductal structures and intracytoplasmic lumina[3]. In this case, IHC staining revealed positivity for CK5/6 and EMA, and CEA was only expressed in ductal structures.

Furthermore, dermoscopy, an emerging dermatological examination tool, may also be helpful for differential diagnosis. Glomerular vessels and surface scales exhibit high sensitivity and specificity in BD[14,15]. Milia-like cysts and cerebriform appearance are considered highly sensitive to SK[16,17]. Shiiya *et al*[18] proposed that fine scales arranged orbicularly, scattered fine black dots or globules and the absence of glomerular vessels could aid in the precise diagnosis of HAS. However, no dermoscopic features of malignant HAS have yet been documented. In this study, besides fine scales and hyperkeratosis, our dermoscopic images showed that linear telangiectasis were also exhibited in the papilla, which has not been reported before. Therefore, we speculate that the appearance of telangiectasis may contribute to the differential diagnosis of malignant HAS and HAS.

CONCLUSION

Although malignant HAS is a malignant adnexal adenoma, prompt surgical resection can achieve good therapeutic results. As a clinically uncommon tumour, malignant HAS is often misdiagnosed as BD or SK. Precise diagnosis depends on histopathological examination, and immunohistochemical analysis is also valuable. Furthermore, we are the first to display dermoscopic features of malignant HAS and found linear telangiectasis that had not been reported in studies of HAS. Therefore, we speculate that telangiectasis appearance may contribute to the differential diagnosis of malignant and benign forms of HAS and that dermoscopy may be a valuable tool for the early diagnosis of malignant HAS.

FOOTNOTES

Author contributions: Yang YF contributed to manuscript writing and editing, Wang R contributed to data collection; Long WG and Zhao XH contributed to data analysis; Xu H and Li YM contributed to conceptualization and supervision; all authors have read and approved the final manuscript.

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ORCID number: Yi-Fei Yang 0009-0006-1935-9744; Rong Wang 0009-0001-8256-8649; Hui xu 0000-0001-6913-6043; Yu-Mei Li 0000-0002-3050-8652.

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