Case Report

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Pathology Section

Malignant Nodular Hidradenoma- A Distinctly Rare and Diagnostically Challenging Sweat Gland Tumor

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ABSTRACT

Malignant Nodular Hidradenoma otherwise designated as Hidradenocarcinoma is a malignant sweat gland tumor which mandates attention due to its rarity and its increased potential for local destruction and metastasis. Though it was first reported in 1954, only 70 cases have been documented till date as per review of literature. The tumor possesses distinctive histological and immunohistochemical features with definitive criteria designed for detection of malignancy. This case report describes the occurrence of malignant nodular hidradenoma in a 40 year old male on the dorsum of the left foot.

Keywords: Eccrine sweat gland tumor, Hidradenocarcinoma, Malignant nodular hidradenoma

CASE SUMMARY

A 40 year old male presented to the surgical clinic with a painless, progressively enlarging nodular swelling over the left foot which was present since two and a half years [Table/Fig-1]. His past history revealed a similar swelling three years back at the same site which was excised in another surgical centre, details of which were not available with the patient. The swelling recurred six months after the previous surgery at the operated site and gradually progressed to the present size.

On local examination, a solitary, bosselated swelling with variable consistency which measured 6 x 4 cms was palpated on the dorsum of the left foot. It was soft to cystic in consistency .The swelling was fixed to skin but free from the underlying structures. There was no inguinal lymphadenopathy. No abnormalities were detected on thorough clinical examination. Provisionally, a primary malignant neoplasm was suspected by the clinician and the patient was posted for wide local excision. Per- operative observation revealed that, the mass was not adherent to the extensor tendons [Table/Fig-2]. A complete wide excision with a 2 cms clear margin of healthy adjacent tissue was performed and the wound was closed with split skin graft. The resected mass submitted for histopathological examination.

HISTOPATHOLOGICAL FINDINGS

Gross Examination- Specimen consisted of a single, skin covered, irregular, nodular mass measuring 4.5 cms x3 cms.

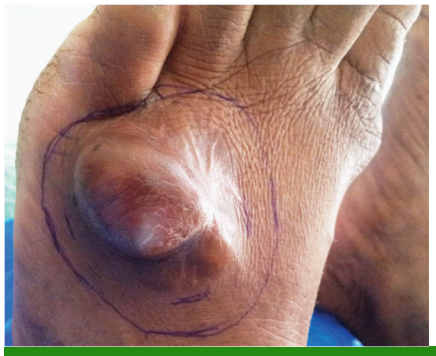
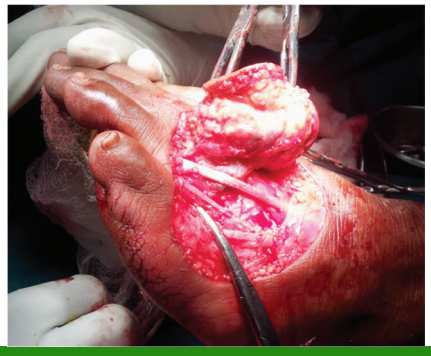
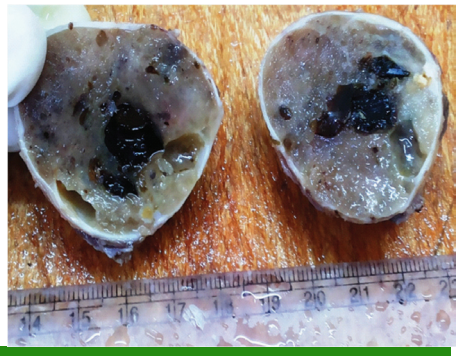
Cut Section- Showed solid and cystic grey brown to grey white areas admixed with hemorrhagic and mucoid areas [Table/Fig-3]. Multiple bits were submitted from these areas and all the surgical margins for processing.

Microscopic Examination- Showed epidermis with basket weave type of hyperkeratosis, papillomatosis and spongiosis. The dermis showed an asymmetrical, poorly circumscribed tumor composed of epithelial cell lobules with solid sheets of cells as well as tubular lumina of varying sizes lined by columnar cells [Table/Fig-4]. Some of the tubules showed cystic diatation with dense, intraluminal eosinophilic secretions [Table/Fig-5]. The tumor cells were round, polyhedral to fusiform with pale, eosinophilic cytoplasm and round to elongated, vesicular nuclei [Table/Fig-6]. Some nuclei were irregular and hyperchromatic [Table/Fig-7]. Occasional atypical mitoses (1-2/HPF) were seen. Focal areas in the tumor showed keratin pearl formations. The tumor nodules were separated by eosinophilic, hyalinised stroma. Vascular channels adjacent to the tumor and capsule showed invasion [Table/Fig-8]. Large areas of necrosis within the tumor and comedo patterns of necrosis were evident [Table/Fig-9]. All the surgical margins were free from tumor invasion.

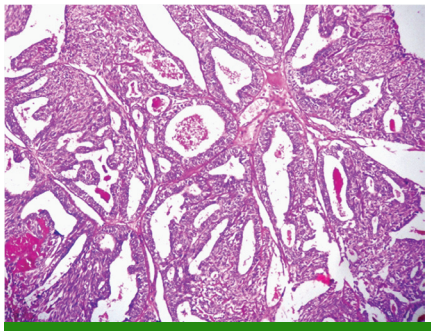
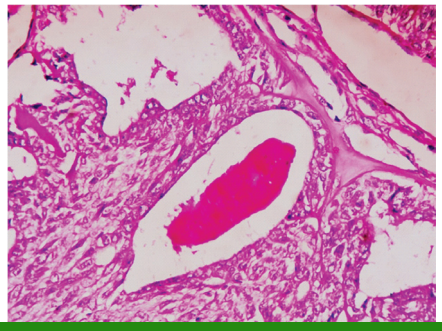
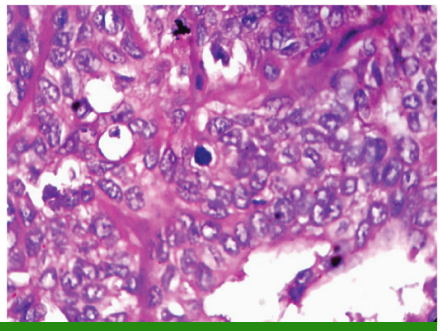
Immunohistochemistry showed positivity for epithelial membrane antigen. Based on these findings, the final diagnosis of malignant nodular hidradenoma was arrived at.

DISCUSSION

Malignant nodular hidradenoma or hidradenocarcinoma is an extremely rare malignant adnexal tumor. Though its benign counterpart is frequently encountered, very few case reports and case series have been documented as per review of literature. In a case series of 120 sweat gland tumors, which

[Table/Fig-1]: Clinical picture of the lesion
[Table/Fig-2]: Per- operative photograph- Extensor tendons free from the mass
[Table/Fig-3]: Cut section of the resected mass showing predominantly solid, partly hemorrhagic and cystic areas containing mucoid material

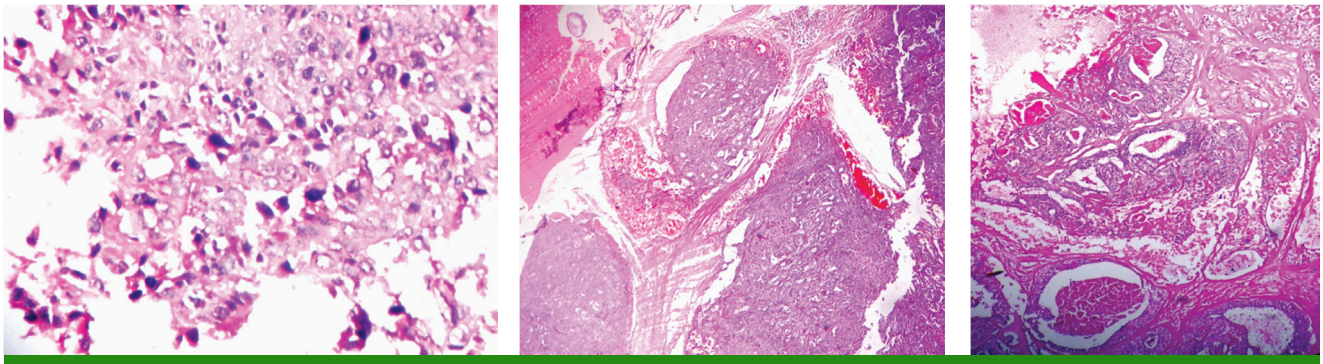
[Table/Fig-4]: Tumor lobules composed of solid sheets and tubules lined by tumor cells, (H&E, ×100)
[Table/Fig-5]: Intraluminal dense eosinophilic secretion, (H&E, ×400)
[Table/Fig-6]: Round, polyhedral to fusiform tumor cells with pale eosinophlic cytoplasm and round to elongated vesicular nuclei, (H&E, ×1000)

is the largest series till date, Berg and McDivitt reported 17 hidradenocarcinomas [1]. Many a times, this tumor has been mistaken for clear cell hidradenocarcinoma, [2] malignant acrospiroma [3] or clear cell eccrine carcinoma [4]. Majority of the studies on hidradenocarcinomas indicate an aggressive clinical course with repeated local recurrences. The paucity of cases documented in literature coupled with the limited details available on follow up of this tumor add up to the hindrances in diagnosis and management. Nevertheless, a precise diagnosis and differentiation from its benign counterpart are two essential factors which clearly influence treatment options for the neoplasm. The histological criteria outlined and accepted for diagnosis of malignant nodular hidradenoma include lack of circumscription, an infiltrative growth pattern, mitoses, nuclear pleomorphism, necrosis, vascular invasion, perineural invasion and deep extension [5]. Tumors with one or two of these features have been designated as atypical hidradenomas while, tumors with more than three of these features have been graded as malignant hidradenomas in the earlier studies [6]. Large zones of necrosis especially of comedo type as seen in the present case is a characteristic feature of malignant hidradenoma [6]. Hyalinized stroma, ductal and epidermoid elements as noticed in the present case were also observed in the other case studies in majority of the malignant hidradenomas [6].

Nodular hidradenoma, also known as eccrine hidradenoma or clear cell hidradenoma is a benign tumor arising from the eccrine sweat glands [7]. Clinically, it presents as a solitary, well circumscribed, encapsulated, intradermal nodule [8]. However, it does not possess any distinctive clinical features to help differentiate it from other sweat gland tumors. Definitive subtyping is possible only on histpathological evaluation [9]. Some of the hidradenomas show predominance of clear cells and are hence designated as clear cell hidradenomas. Since the solid component was predominant in our case the designation of nodular hidradenoma seemed more appropriate.

Malignant Nodular Hidradenoma which is synonymous with nodular hidradenocarcinoma was earlier reported as clear cell carcinoma by Keasby and Hadley in 1954 [10]. Malignant nodular hidradenoma is distinctly rare and the diagnosis as atypical or malignant can often be challenging as in the present case, since it is morphologically similar to the benign counterpart. The histologic criteria which helped in arriving at the diagnosis of malignancy in the present case included asymmetry of the tumor, poor circumscription, infiltrative growth pattern, presence of predominantly solid sheets of tumor cells, nuclear atypia, mitotic activity, large islands of necrosis with focal comedo patterns and vascular invasion.

Management involves wide excision of the lesion as was done in the present case. Keeping in view that it had recurred, the



[Table/Fig-7]: Irregular tumor cells with hyperchromatic nuclei, (H&E, ×400)
[Table/Fig-8]: Vascular and capsular invasion, (H&E, ×100)
[Table/Fig-9]: Large areas of necrosis and comedo patterns, (H&E, ×400)

patient was referred to a higher centre for further management and hence lost to follow- up [6].

CONCLUSION

Malignant nodular hidradenoma has a potential for local recurrenceand a tendency to metastasize. The aggressive nature and course of the tumor demands prompt recognition of the histologic features for categorization as malignant in order to institute appropriate management.

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