Case Report

An Unusual Presentation of a Nodular Hidradenoma: A Case Report

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Abstract

Hidradenoma also called nodulocystic hidradenoma, Clear cell hidradenoma or acrospiroma is a rare benign epithelial tumor mostly occurring in adult females. It usually appears as a single, slow enlarging, well-circumscribed, freely movable, firm, and non-tender cutaneous lesion commonly seen on the scalp, face, anterior trunk, and proximal limbs. Less commonly seen in the distal limb. An 18-year-old female patient visited the Outdoor Patient Department (OPD) of the hospital of the National Institute of Unani Medicine (NIUM), Bangalore, Karnataka, India; with chief complaints of swelling in the right lower leg for 3 months. This swelling was insidious in onset, and gradually enlarged from a small papule to golf ball size, the shape was nearly hemispherical. The patient consulted a general practitioner where FNAC was done, there was serous discharge from the swelling. It was a case of nodular hidradenoma having an unusual presentation as black colour with brownish hue having an unusual larger size, ulcerated surface with serous discharge at the right lower limb. Wide surgical excision with appropriate margins to prevent local recurrence was done. After excision collagen dressing done on weekly basis for 5-6 weeks. Routine follow-up 3, 6 and 12 months later showed no signs of recurrence. Nodular hidradenoma is usually located over the head, neck, and trunk. In our case, the lesion was present in the lower limb and the presence of an unusually large tumor mass, leading to serous discharge, makes our case even more interesting.

Keywords: Nodular hidradenoma; Case report; Serous discharge

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Introduction

Hidradenoma is also called Nodulocystic hidradenoma, Clear cell hidradenoma or Acrospiroma.[1] Nodular hidradenoma is a rare benign epithelial tumor. Previously thought to have eccrine differentiation. But now it is accepted that it may have eccrine or apocrine differentiation.[2,3] It usually appears as a single, slow enlarging, well-circumscribed, freely movable, firm and non-tender cutaneous lesion.[4] Most commonly appear on the scalp, face, anterior trunk, and proximal limbs.[5] They are usually asymptomatic, present in middle-aged and elderly females.[6] Its size ranges from 5 to 30 mm. The skin above the tumor may be smooth, thickened, atrophic or ulcerated, and may have a skin-coloured, red, brown or blue appearance. Serous or haemorrhagic discharge may occur from some tumors.[3,6] Unusual presentation are painful or rapid increasing neoplasm, childhood onset, and larger size.[7] Chances of recurrence, conversion in malignancy, and metastasis are also common with this tumor.[8] Use of fine needle aspiration cytology (FNAC) for the diagnosis of skin lesion has been described in the literature; however, the number is still limited because there are very few case report that describe its cytological findings.[9]

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Case summary

An 18-year-old female Hindu patient from Tamil Nadu, India, with no significant past medical or family history, approached the outpatient department (OPD) of surgery of National Institute of Unani Medicine (NIUM) Hospital, Bangalore, India, on September 21, 2022. She presented with the chief complaints of a dark swelling in the right lower leg from 3 months (Figure 1). This swelling was insidious in onset, gradually enlarged from a small papule to golf ball size, the shape was nearly hemispherical. The patient consulted a general practitioner where FNAC was done. Physical examination revealed a well-circumscribed, hemispherical, firm, partially mobile, non-tender dermal nodule, measuring 5cm×5cm×2.5cm on the right lateral aspect of the right leg near the knee joint. The skin over the nodule was cracked (ulcerated) but taut with a brownish hue (appearance) (Figure 1). There was also clear serous discharge from the swelling. No regional lymphadenopathy was found, and no systemic abnormality was detected.

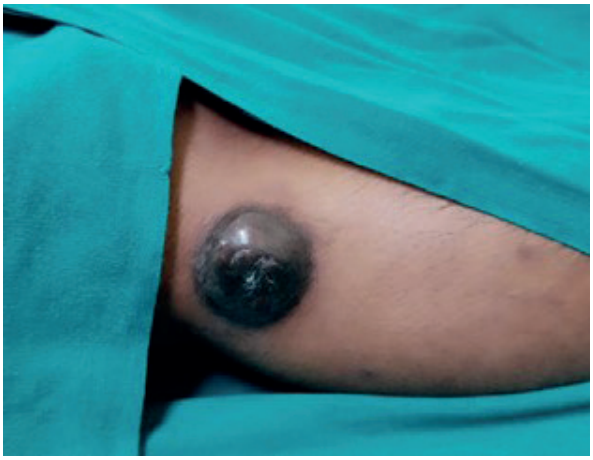

Figure 1: Patient at the time of presentation at Surgery OPD

The patient was admitted on September 21, 2022. Investigations were done for the final diagnosis. On September 23, 2022, a surgical intervention (wide surgical excision with appropriate margins) was performed; after excision, collagen dressing was done on a weekly basis for 5-6 weeks. To reach the final diagnosis, the patient was thoroughly investigated. Hematological investigations were nearly in the normal range. Plain radiographs of the right leg in anteroposterior and lateral view show no involvement of underlying bone structure. Radiographic impression reveals well-defined soft tissue density with air lucencies in the lateral aspect of the right knee joints. Hence, provisional diagnosis may be either an infected sebaceous cyst or soft tissue tumor. FNAC was done, where the cytological findings show highly cellular cells arranged in clusters and follicles. The cells show moderate to severe pleomorphism. Few spindle cells and cytoplasmic vacuolation were noted. It revealed a highly vascular and cellular tumor.

Biopsy was done in view of pleomorphism to rule out sarcoma or intermediate-grade vascular tumor. Excisional biopsy with a 1cm clear margin of nodule under local anesthesia was performed and the sample was sent for histopathological examination. On gross examination, single skin covered with tissue fragment measuring 7.5cm×5cm×2.5 cm with external surface shows a nodule with ulcerated surface measuring 4.5cm×4cm×2cm. The cut surface of the nodule shows a solid cystic tumor measuring 4cm×2.5cm×4cm. The surface of the tumor shows whitish areas and pultaceous material and friable papillary excrescences.

Histological examination revealed tissue fragments lined by stratified squamous epithelium exhibiting hyperplasia with areas of ulceration and underlying tumor having features of nodular hidradenoma. Stroma is oedematous and shows mixed inflammatory cell infiltration. Section studied from both long axis and short axis margins are free of tumor. A deep margin is formed by the tumor (Figure 2). Based on these results, wide surgical excision with appropriate margins to prevent local recurrence was done (Figure 3). After excision collagen dressing was done on weekly basis for 5-6 weeks after that, dressing with betadine ointment was done. Routine follow-up was done on 3-, 6-, and 12-months following surgery showed no signs of recurrence.

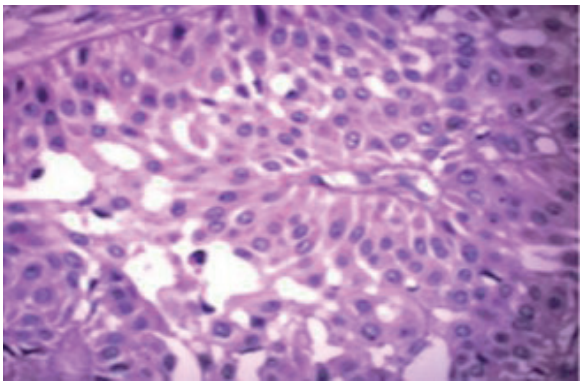

Figure 2: Histopathological examination showing mixed inflammatory cell infiltration.

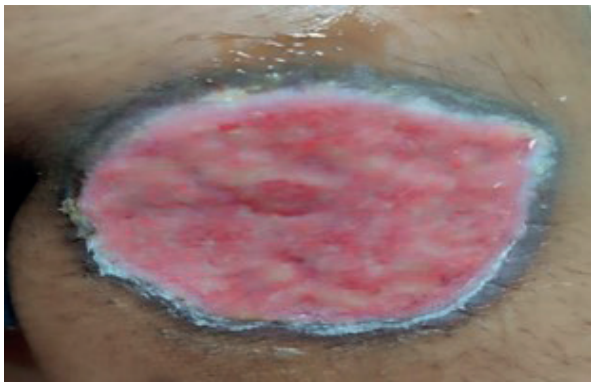

Figure 3: Postoperative view of the lesion

Discussion

The first case of nodular hidradenoma was recorded in 1949. It is an uncommon benign adnexal tumor[8,10], characterized by solitary, well-defined, encapsulated, progressive increasing, asymptomatic, firm nodule of size 5-30 mm.[3,4] Nodular hidradenoma has various synonyms because of its dissimilar histomorphological patterns like clear cell hidradenoma, solid cystic hidradenoma, clear cell acrospiroma, and eccrine acrospiroma. It usually appears overhead, anterior surface of the trunk, and rarely over extremities. As it is an asymptomatic and slowly enlarging tumor, a recent increase in size may indicate any trauma, sudden hemorrhage, or malignant transformation.[10] As per Hernandez-Perez and Cestoni- Parducci review literature hidradenoma has female predominance, with a mean age at presentation of 37.2 years. And common site of occurrence was head (30.3%) then upper limb (25.8%) and trunk (20.2%). The cause of these lesions may be trauma or it may occur spontaneously. Patients may complain of pain on pressure, pruritus, or a burning sensation in the lesion.[4]

Cytological characteristics of nodular hidradenoma are rarely described in the literature. Therefore, cytologists may face difficulty in their diagnosis. The cytological findings are high cellularity, an admixture of two kinds of cells one is eosinophilic or polygonal cells and another one is clear cells, a three-dimensional arrangement of cells with rounded, rosette, and tubular-like structure and background having hyaline material.[9,11] In our case cytological findings were not as similar as described in the literature. As Fine needle aspiration cytology is not enough to make the diagnosis of nodular hidradenoma. For both diagnosis and treatment, an excisional biopsy is required.[12] Our final diagnosis of nodular hidradenoma was confirmed by a histopathological test. The histological feature of nodular hidradenoma is well-circumscribed, unencapsulated but sometimes encapsulated as in our case, having solid cystic components in the dermis. There is a Grenz zone between the tumor and the dermis. Eosinophilic/polyhedral and clear cell form solid areas while cystic areas may contain eosinophilic homogenous material.[4,11]

Malignant transformation is also reported in the literature but the exact incidence rate remains unknown. In the case of malignant transformation histological features are characterized by nuclear atypia, necrosis, and abnormal mitosis.[2,8] Wide surgical excision with the negative marking is the treatment of choice to minimize the risk of recurrence.[12]

Patient's perspective

With the outcome of the surgery, the patient was satisfied. Previously she was very uncomfortable with the discharge of the swelling but after surgery and regular dressing the wound healed and there was no sign of local recurrence after 12 months of surgery.

Conflict of interest: None declared.

Informed consent: Patient's informed consent was taken.

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Authors' contributions: MF and SSA performed the surgery; MAJA did the follow-up of the patient; and RM prepared the case report. All the authors drafted, edited and approved the final version of this case report.

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