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CASE REPORT

A VERY RARE CASE OF SKIN ADNEXAL NEOPLASM: HIDRADENOMA

Sunil Kumar[1], Basavaraj Badadal[2], M. B. Patil[3], Ramakanth Baloorkar[4], Dayanand Biradar[5]

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ABSTRACT: DEFINITION: Hidradenoma is a form of benign adnexal neoplasm that is a close relative of poroma, but is characterized by cells with ample cytoplasm[1-4]. Here we present a very rare and interesting case of a 35 year old female patient who presented to surgical opd with the complaints of swelling in front of the middle of the neck since two years. Swelling was not associated with any other complaints like pain, difficulty in swallowing, difficulty in speaking and difficulty in pronunciation of words. With adequate pre-operative preparation and normal routine blood investigations, patient was posted for surgery under monitored anesthesia care {ie.MAC}. A wide local excision was performed and specimen was sent for histopathological examination. HPR revealed an eccrine clear cell hidradenoma of neck. On follow-up for 2 year there has been no recurrence.
KEYWORDS: Hidradenoma, Skin adnexal neoplasm.

INTRODUCTION: Hidradenoma is a benign tumor, which usually presents as a solitary, skin-colored lesion and occurs more commonly in females[5]. Hidradenoma may have variable histomorphological patterns reflected by the various terms used to describe this entity: nodular hidradenoma, eccrine acrospiroma, solid-cystic hidradenoma, clear cell hidradenoma, and clear cell acrospiroma. In fact, some tumours have epidermal attachment, and occasionally may also have features overlapping with those of typical poromas.

Clear cell change and/or squamous metaplasia may be prominent. However, squamoid change does not seem to denote a worse prognosis. The lesion is also characterized by its pushy, but well-circumscribed, peripheral border. Nodular hidradenoma should be fully excised, as malignant transformation may be present in other areas of the lesion. Furthermore, hidradenoma has a recurrence rate of approximately 12% if not fully excised, especially in lesions with irregular peripheral margins.[6]

CASE REPORT: A 35 year old female patient presented to our surgical opd with complaints of swelling in front of the middle of the neck since two years. Swelling was not associated with any other complaints like pain, difficulty in swallowing, difficulty in speaking and difficulty in pronunciation of words.

On examination there was a solitary swelling horizontally measuring 3cm*2cm, swelling was horizontally oval in shape. Skin over the swelling was normal ie.no dilated or engorged veins & there were no scar marks over the swelling. Surrounding skin was hyper pigmented but non-erythematous. Swelling was having well-defined edges & margins, surface was smooth.

Swelling was firm to hard in consistency. Swelling was not moving with deglutition. Swelling was freely mobile both in cranio-caudal and horizontal directions. With adequate pre-operative preparation and normal routine blood investigations, patient was posted for surgery under monitored anesthesia care {i.e. MAC}. A wide local excision was performed and specimen was sent for

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histopathological examination. HPR revealed an eccrine clear cell hidradenoma of neck. Since it was an outpatient procedure patient was discharged 4 hours after surgery.

Patient was put on Co-Amoxiclav group of antibiotics with pain killers and antacids and seratiopeptidase for one week. Sutures were removed after 8 days. On follow up for 2 year there has been no recurrence.

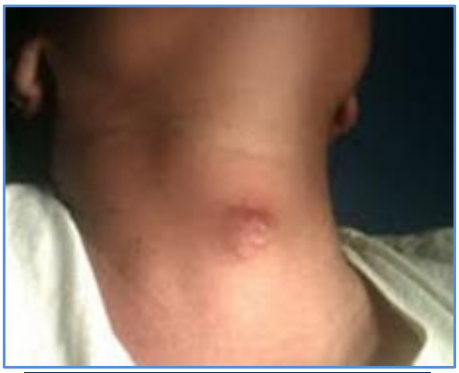

Fig. 1: Clinical Photograph

Fig. 2: Intra-Operative Photograph

Fig. 3: After skin closure

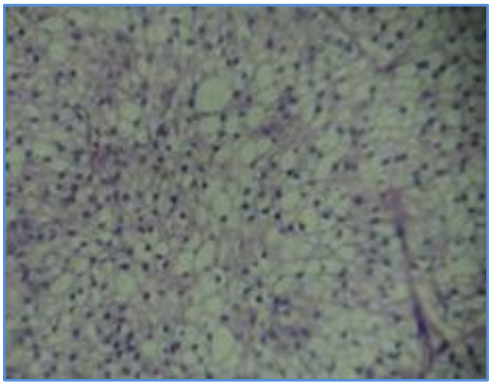

Fig. 4A: Histopathology showing Squamous metaplasia and small, monomorphous and polyhedral cells

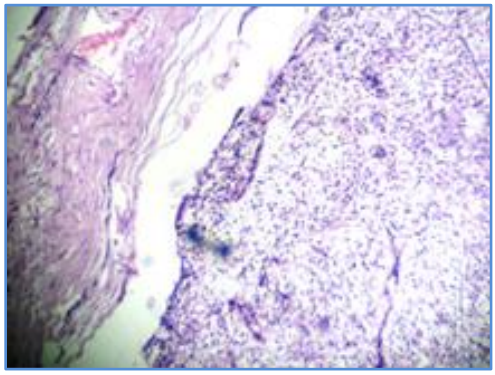

Fig. 4B: Histopathology showing pushy, well-circumscribed, peripheral border

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DISCUSSION: The Classification of cutaneous sweat gland adnexal lesions is presented in Table 1.[1-4]

Origin	Benign	Malignant
Eccrine& apocrine (mixed origin)	Hidrocytoma Apocrine/eccrine nevus Tubulopapillary hidradenoma[7-9] Chondroid syringoma[10]	Malignant tumor of the skin (has eccrine /apocrine & mesenchymal components)
Eccrine	Poroma Hidradenoma Spiradenoma Cylindroma Syringometaplasia Syringoma Syringofibroadenoma[11]	Porocarcinoma Hidradenocarcinoma Spiradenocarcinoma Malignant cylindroma Syringoid carcinoma Microcystic adnexal carcinoma Mucinous carcinoma Adenoid cystic carcinoma Aggressive digital papillary adenocarcinoma
Apocrine	SCAP[12] Hidradenoma papilliferum[13,14]	Syringocystadenocarcinoma Apocrine carcinoma Extramammary Paget's disease[15]

Table 1

SCAP: SyringoCystAdenocarcinoma Papilliferum[12]

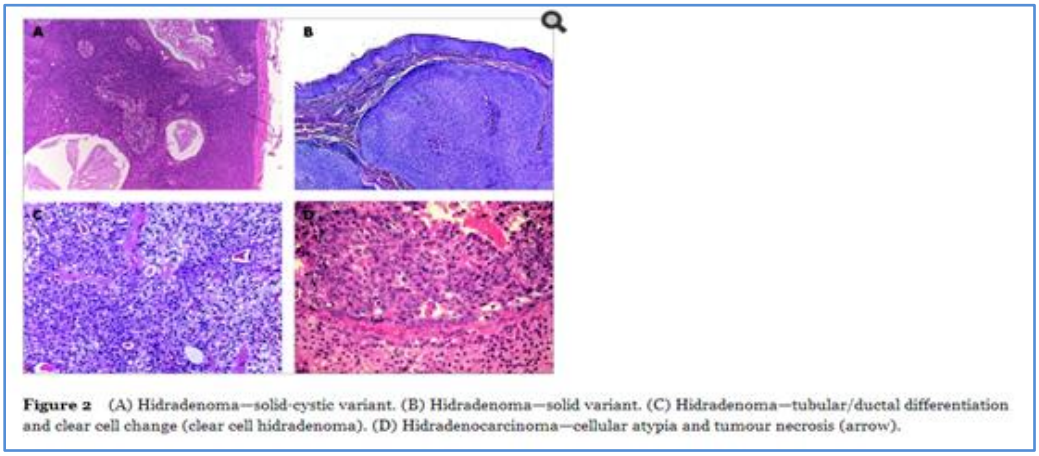

Figure 2 (A) Hidradenoma—solid-cystic variant. (B) Hidradenoma—solid variant. (C) Hidradenoma—tubular/ductal differentiation and clear cell change (clear cell hidradenoma). (D) Hidradenocarcinoma—cellular atypia and tumour necrosis (arrow).

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HISTOLOGICAL TYPES OF HIDRADENOMA: Most cases of hidradenocarcinoma arise de novo. In some cases the tumor may also arise in pre-existing hidradenoma[6]. Hidradenocarcinoma is also called by different names such as malignant nodular/clear cell hidradenoma, malignant clear cell acrospiroma, clear cell eccrine carcinoma or primary mucoepidermoid cutaneous carcinoma. Histologically, it is a multinodular solid malignant neoplasm, showing ductal structures and intracytoplasmic tubular vacuoles, with areas of tumor necrosis. The tumor cells have similar morphology as those of nodular hidradenoma, but may also show cytonuclear atypia and increased mitotic activity. Apocrine differentiation is commonly seen.

Evidence of nodular hidradenoma remnants may be quite oftenly seen. An infiltrative growth pattern is not seen usually, and the carcinoma is distinguished from benign hidradenoma by the presence of brisk mitotic activity and cellular pleomorphism. The tumor cells stain positively for LMWK, and the ductal structures/luminal surfaces are highlighted by EMA and CEA. Even though these rare tumours do not always behave aggressively, they may have an aggressive course with metastasis and/or local recurrence. The primary treatment is wide local excision with or without lymph node dissection.[16,17]

In some cases clear cell hidradenoma and hidradenocarcinoma may occasionally mimic metastatic clear cell carcinomas including thyroid, lung or renal cell carcinomas. However, the first two are usually distinguished by their positivity to thyroid transcription factor-1 (TTF-1), and the latter by its prominent vascularity, and the presence of hemorrhage and focal granular necrosis within the lesion[18]. Renal cell carcinoma also expresses both EMA and CD10.

Composite/mixed adnexal Tumours: Cutaneous adnexal tumours may sometime display a varied composition with a mixture of eccrine, apocrine, sebaceous and pilar differentiation.[19-22] The diagnosis of these mixed lesions relies on histological evaluation, and most of the times they are classified according to the predominant morphological component. If no component is predominant, a different terminology is used to describe these lesions, including “combined adnexal tumours of the skin”[19], “benign adnexal tumor with multi-directional differentiation”[22], “benign adnexal tumor of mixed lineage”[2,5] and “composite adnexal tumours of the skin”[23].

ABBREVIATIONS:

CEA - Carcinoembryonic antigen.
EMA - Epithelial membrane antigen.
EMPD - Extramammary Paget’s disease.
LMWK - Low molecular weight keratin.
SCAP - Syringocyastadenoma papilliferum.
TTF- 1 -Thyroid transcription factor-1.

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AUTHORS:

1. Sunil Kumar
2. Basavaraj Badadal
3. M. B. Patil
4. Ramakanth Baloorkar
5. Dayanand Biradar

PARTICULARS OF CONTRIBUTORS:

1. Post Graduate, Department of General Surgery, BLDEU's Shri B.M. Patil Medical College Hospital & Research Centre, Bijapur.
2. Assistant Professor, Department of General Surgery, BLDEU's Shri B.M. Patil Medical College Hospital & Research Centre, Bijapur.
3. Professor & Unit Head, Department of General Surgery, BLDEU's Shri B.M. Patil Medical College Hospital & Research Centre, Bijapur.
4. Associate Professor, Department of General Surgery, BLDEU's Shri B.M. Patil Medical College Hospital & Research Centre, Bijapur.
5. Assistant Professor, Department of General Surgery, BLDEU's Shri B.M. Patil Medical College Hospital & Research Centre, Bijapur.

NAME ADDRESS EMAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Sunil Kumar,
Final Year Post Graduate,
Department of General Surgery,
BLDEU's Shri B.M. Patil Medical College Hospital & Research Centre,
Bangaramma Sajjan Campus,
Bijapur-586103.
Email: dkdrsunil0@gmail.com

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