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RARE CASE OF IMMUNOHISTOCHEMICALLY PROVEN ABRIKOSSOFF TUMOR LOCATED IN THE VULVA AND THIGH

Dermatology

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ABSTRACT

A 27-year-old female presented to the dermatology department with complaints of an asymptomatic single hard skin-coloured growth over the left side of labia majora, with a similar lesion over the inner aspect of right thigh, ongoing since 3 years. An excisional biopsy and immunohistochemical study concluded a diagnosis of Benign Granular Cell Tumor (Abrikossoff tumor). Most of the cases of Abrikossoff tumor in literature have been found either in the oral cavity or the gastrointestinal tract. We present this case for its rare presentation in the genital area.

KEYWORDS

Abrikossoff tumor, Granular Cell Tumor, benign, vulva and thigh, immunohistochemically positive[S100].

INTRODUCTION:

Abrikossoff tumor or Granular Cell Tumor is a rare tumor consisting of cells with characteristic granular cytoplasm. These cells are of neural or nerve sheath in origin. It was first described by a Russian Pathologist, Aleksei Ivanovich Abrikossoff in 1926[1,2]. It occurs in both sexes, with a female predominance in the 4th to 6th decades[3]. Though it commonly occurs in head and neck region, it can occur at any site in the body. It is predominantly benign, with a 2% likelihood of malignancy and metastatic potential.

CASE REPORT:

A 27-year-old female came with complaints of a single raised skin-coloured growth over left side of labia majora since 36 months, spontaneous in onset and gradually progressive in size. She developed another single hard mass beneath the skin over inner aspect of right thigh since 18 months. There was no itching or pain or any discharge from the lesions. There was no history of any congenital abnormalities in the patient or immediate family. Past, personal, and medical histories revealed no significant findings.

General physical and systemic examinations were unremarkable. Cutaneous examination showed solitary skin-coloured nodule, 3x2 cm in size over left side of labia majora. It was non-tender and firm-to-hard in consistency (Fig.1). Another solitary nontender nodule measuring 2x3 cm, which was firm-to-hard in consistency with overlying hyperpigmentation was present over inner aspect of right thigh. There was no associated local lymphadenopathy.

Blood tests including serology for infectious diseases were normal. Excisional biopsies of both lesions were performed under saddle block anesthesia by the Hospital's Gynaecology team.

Histopathology study showed mildly hyperplastic epidermis and dermis showed spindle to rounded cells in sheets, fascicles and vague storiform pattern (Fig.2a). Cells showed vesicular, oval to elongated nuclei and small inconspicuous nucleoli. Cytoplasm is abundant and granular with few pustulo-ovoid bodies (Fig.2b).

Thigh mass showed similar morphology as the vulval mass. No evidence of Grenz zone, nuclear atypia, increased mitosis or necrosis were seen. On further immunohistochemical staining, the tumor cells showed diffuse strong granular cytoplasmic positivity for S100, confirming the diagnosis of Granular Cell Tumour (Fig.3).

The patient was monitored over a one-year follow-up period, during which no recurrence was observed. She is currently in the third trimester of pregnancy.

DISCUSSION:

Granular Cell Tumors (GCTs) are rare neoplasms of neural or nerve sheath origin, predominantly benign but with a 2% risk of malignancy and metastasis. GCTs have been recognized for nearly a century, first described by Abrikossoff in 1926, who originally postulated a myogenic origin and named it Granular Cell Myoblastoma [1,2]. GCTs may be associated with syndromes such as Noonan syndrome, Neurofibromatosis Type 1, and LEOPARD syndrome, often presenting with multiple lesions that complicate diagnosis and treatment.[3].

GCTs can occur anywhere in the body, with two-thirds in the head and neck region. [4]. Other various locations include the skin, oral cavity, gastrointestinal tract, breast, and respiratory tract. They also occur in rarer sites like the vulva, accounting for 3–16% of cases[3]. In the vulva, the labia majora is the most frequent site of occurrence [3]. Differential diagnoses include Bartholin's duct cysts, sebaceous cysts, lipomas, and even malignant entities like melanoma [3].

GCTs can occur at any age but are most common between 30 and 50 years[5]. Familial predisposition is suspected when multiple benign or malignant lesions occur in family members[5].

Histologically, GCTs are classified as benign, atypical, or malignant, based on six criteria established by Fanburg-Smith et al. in 1998. It includes necrosis, spindling, vesicular nuclei, increased mitotic activity, high nuclear to cytoplasmic ratio, and pleomorphism [6]. Immunohistochemical staining is essential for diagnosis, with markers such as S100, CD68, and neuron-specific enolase (ENO2) commonly found in GCTs [3]. Furthermore, Ki-67 immunostaining values higher than 10% can aid in the histological classification of malignant cases [6]. Prior to treatment, a thorough history, physical exam, and imaging (chest X-ray, abdominal ultrasound, pelvic MRI) are essential to rule out multicentric lesions and metastases.[6].

Benign GCTs are treated with local excision with clear margins. Mohs micrographic surgery may be used for ill-defined or cosmetically sensitive areas [3]. Malignant GCTs are rare but carry a poor prognosis, with 60% mortality within three years. Poor outcomes are linked to infiltrative margins, high Ki-67, and recurrence within 2 years[6].

For patients with malignant GCTs, wide local excision with negative margins is critical to reduce recurrence. Lymphadenectomy and vulvectomy are sometimes necessary for extensive cases, though the role of adjuvant chemotherapy or radiotherapy remains controversial [6]. Studies suggest that recurrence rates are lower with clear surgical margins [7,8]. Even after conservative management, GCTs can rarely recur [7].

CONCLUSION:

Granular cell tumor needs to be kept in differential diagnosis of vulval growths. Gold standard for diagnosis of GCT is immunohistochemistry. Follow-up is essential for monitoring recurrence or metastasis, especially in high-risk cases [8].

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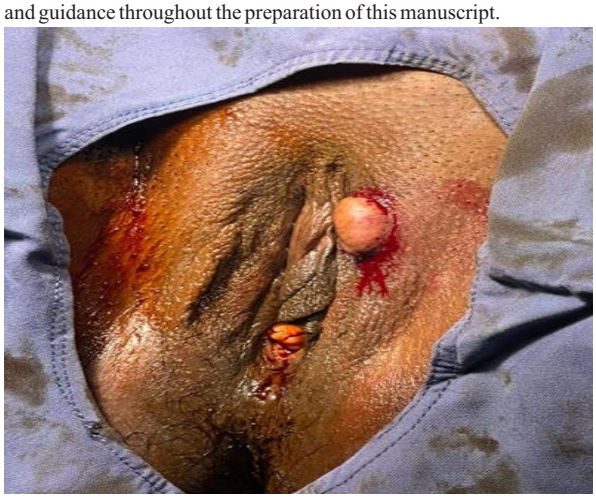

Figure1: Skin-coloured Nodule, 3x2 cm In Size Over Left Side Of Labia Majora

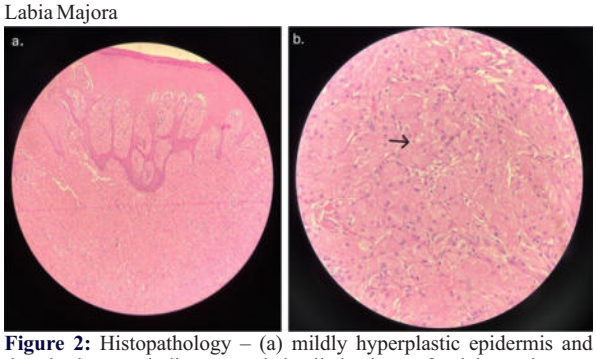

Figure 2: Histopathology – (a) mildly hyperplastic epidermis and dermis shows spindle to rounded cells in sheets, fascicles and vague storiform pattern - Under 10x. (b) Cytoplasm is abundant and granular with few pustulo-ovoid bodies (Black arrow) -Under 40x.

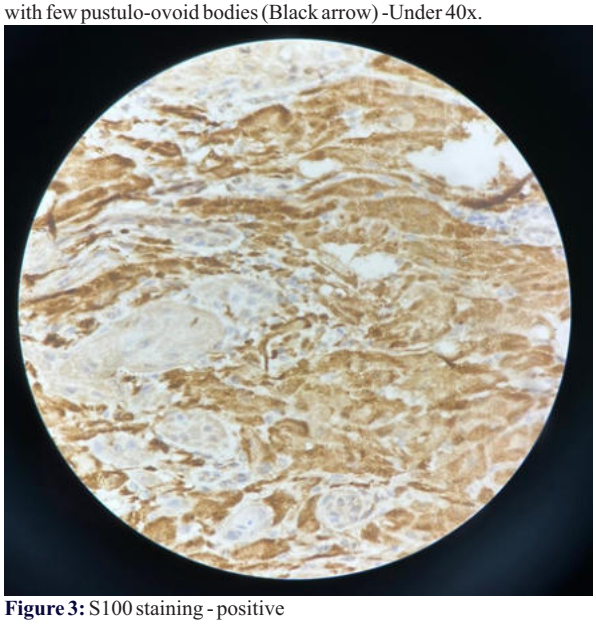

Figure 3: S100 staining - positive

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