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Serous papillary cystadenofibroma of vulva: A histopathological surprise

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ABSTRACT

INTRODUCTION: Serous Papillary Cystadenofibromas (SPCAF) of the vulva is rare.**CASE REPORT:** We report a case of a 45-year-old female who presented with a painless slow growing mass in the genital region. Examination revealed a 10 × 8 cm swelling from the vulva. USG was suggestive of a complex cystic lesion and MRI showed a low signal intensity lesion on T2W image. She underwent wide local excision and the histopathology was suggestive of a SPCAF.**DISCUSSION:** Vulval tumors rare- account for 4 % of female genital tract tumors. Mainstay of treatment in cases of SPCAF is wide local excision. Histopathology confirms the diagnosis and is used to rule out malignant transformation.**CONCLUSION:** These represent uncommon tumors with high degree of heterogeneity which becomes a major challenge and systematic evaluation is crucial for clinical decision-making and patient management.© 2021 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Cystadenofibroma (CAF) of the female genital tract is a rare, slow growing benign epithelial neoplasm of unknown etiology. It usually occurs as a combination of solid and cystic component. This case report has been reported in line with the SCARE 2018 guidelines [1].

2. Case report

A 45-year-old female, with no prior medical history, presented with a history of swelling in the genital region for the last 3 years. It was slowly increasing in size, with no associated bleeding or white discharge or mass per vagina. Bowel and bladder habits were regular, and no history of gastrointestinal or gynecological malignancies in the family. She was para 3 with tubectomy done 15 years back, with regular menstrual cycles and gave no history of use of hormone replacement therapy. On physical examination, vital were stable and systemic examination was normal. Local examination revealed a 10 × 8 cm pedunculated swelling, soft in consistency, arising from the right genital fold - vulva (Fig. 1). Per speculum examination showed normal cervix and no discharge. Per vaginal examination was normal, with no adnexal masses. Blood investigations were within normal limits. Fine Needle Aspiration Cytology (FNAC) was not done as perineal hernia was considered

**Fig. 1.** Local Examination – soft, pedunculated swelling arising from the vulva.

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as a differential diagnosis. Soft tissue Ultrasonogram showed a heterogeneous complex cyst, with septations and internal echoes

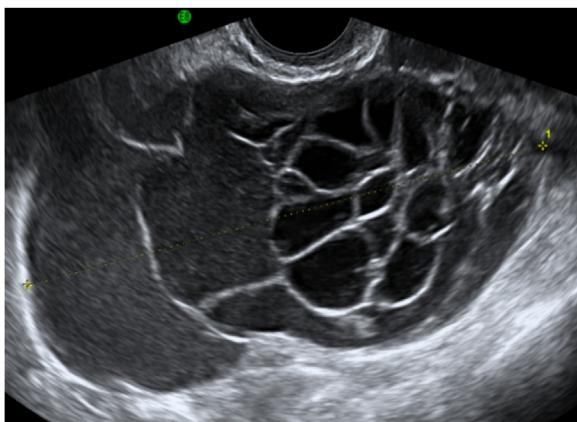


Fig. 2. USG - Encapsulated heterogeneous complex cyst, with septations and internal echoes.



Fig. 3. MRI - low signal intensity on T2W images suggestive of a CAF.

which was encapsulated with no vascularity (Fig. 2). Magnetic Resonance Imaging (MRI) on T2W image showed low signal, well defined multiloculated cystic swelling with rest of abdomen & pelvic structures-normal (Fig. 3). With the provisional diagnosis of a soft tissue tumour, she underwent wide-local excision under general anaesthesia. The postoperative period was uneventful and the patient was discharged on the 3rd postoperative day. On follow up, histopathology report showed blunt papillae with prominent stromal component and no evidence of necrosis, haemorrhage or increased mitosis with clear margins – suggestive of Serous Papillary Cystadenofibroma (Fig. 4). Patient is currently on regular follow up and is doing well.

3. Discussion

Vulval tumors rare- account for 4 % of female genital tract tumors [2]. WHO Classifies it as (a) Epithelial, (b) Neuroectoder-

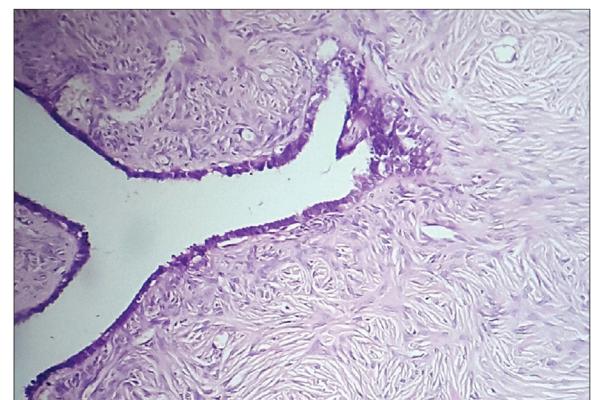


Fig. 4. Histopathology – blunt papillae with prominent stromal component.

mal, (c) Soft tissue tumors, (d) Melanocytic, (e) Germ cell tumors, (f) Lymphoid & Myeloid and (g) Metastatic tumors [3]. Soft tissue tumors are further classified as (1) Benign, (2) Malignant and (3) Others, of which Serous Papillary Cystadenofibromas (SPCAF) of the vulva is rare, accounting for 0.2 %. CAFs were first diagnosed by Iwanow in 1909 [4]. It is common in middle aged (16–56 years) women. Most women are asymptomatic, and in majority of the cases it is an incidental finding or they present as a slow growing mass, as was in our case. Pre-operative diagnosis is missed due to its rarity. The etiology of these tumors is unknown but most occur spontaneously. They present as painless, well circumscribed slow growing swelling. On ultrasonogram, it is seen as a complex cyst with solid components, often misdiagnosed as malignant tumor. It is an uncommon benign neoplasm containing epithelial and fibrous stromal components. A characteristic appearance is seen on MRI – due to the presence of fibrous component, a low signal intensity on T2W images. This helps differentiate it from malignant tumors [5]. Histopathologically, CAFs have more prominent stromal component than cystadenomas and tend to form more prominent blunt papillae that can extend into the lumen of the cyst or outward from its wall. Mainstay of treatment remains surgical with wide local excision. Histopathology is used to confirm diagnosis and to rule out malignant transformation. Chemotherapy has been tried and is controversial. Due to its rarity, there are no well-defined guidelines. Post-operatively, Cisplatin is being tried for complete clearance [4].

4. Conclusion

SPCAF is a tumor with high degree of heterogeneity which becomes a major challenge and systematic evaluation is crucial for clinical decision-making and patient management. MRI is highly specific and gives origin and internal characterization of the tumor, but histopathology confirms diagnosis.

Declaration of Competing Interest

The authors report no declarations of interest.

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None.

Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

A.P. Roshini – Design, Selection and recruitment of study subject, Data collection and monitoring of data, Interpretation of data, Drafting final report, maintaining patients file and master file of project.

Audi Pandarinath – Concept, Design, Selection and recruitment of study subject, Data collection and monitoring of data, Interpretation of data, Drafting final report.

Mervyn Corriea – Design, Selection and recruitment of study subject, Data collection and monitoring of data, Interpretation of data, Drafting final report.

Registration of research studies

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Dr. Pandarinath Audi.

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References

- [1] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical Case REport (SCARE) guidelines, *Int. J. Surg.* (60) (2018) 132–136.
- [2] M.B. Reedy, W.E. Richards, F. Ueland, K. Uy, E.Y. Lee, C. Bryant, J.R. van Nagell Jr., Ovarian tumors: a case report and literature review, *Gynecol. Oncol.* 75 (1999) 293–297.
- [3] R.E. Scully, Vulval tumors. A review, *Am. J. Pathol.* 87 (1977) 686–720.
- [4] Y. Gurbuz, S.K. Ozkara, Immunohistochemical profile of serous papillary cystadenofibroma of the fallopian tube: a clue of paramesonephric origin, *Appl. Immunohistochem. Mol. Morphol.* 11 (2003) 153–155.
- [5] D.C. Jung, S.H. Kim, MR imaging findings of ovarian cystadenofibroma and cystadenocarcinofibroma: clues for differential diagnosis, *Korean J. Radiol.* 7 (2006) 199–204.

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