Gluomangiomyoma of the clitoris: A case report and literature review

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Abstract
Glomus tumor is an uncommon, benign, soft-tissue lesion in which the cells recapitulate the structure of the normal modified smooth muscle cells of the glomus body. Glomus tumors usually occur in tissues that normally contain glomus bodies; only rarely can they develop in sites where glomus bodies are normally sparse or absent. There are three subtypes of glomus tumor, with glomangiomyoma being the rarest. No more than 10 cases of glomus tumor in female genitalia have previously been reported, involving the vulva, vaginal area, periurethral area and clitoris. A clitoral glomangiomyoma is extremely rare. This is a case report of a glomangiomyoma in the clitoral area. Published reports of glomus tumor in the female external genitalia are reviewed.
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1. Introduction

Glomus tumors are benign soft-tissue neoplasms arising from the specialized epithelioid cells of the glomus body. The glomus body has a role in thermoregulation and is most commonly located in the subungal region and lateral aspects of the digits [1]. Glomus tumors are relatively rare and account for less than 2% of soft-tissue tumors in the extremities. Histologically, glomus tumors are divided into three subtypes—solid glomus tumor, glomangioma or glomangiomyoma—differing in the relative proportions of the three components, namely epithelioid tumor cells, vascular elements, and smooth-muscle component. Most glomus tumors are less than 2 cm in diameter [1]. In this report, the clinical, radiological, and pathological features of a rare case of a clitoral glomangiomyoma are described.

2. Case Presentation

A 36-year-old Caucasian woman was seen in the gynecologic oncology clinic for an uncomfortable 1 cm nodule under the clitoris. This was noticed to be gradually increasing in size over a 12-month period. There was a reported history of post-coital ache at the clitoris lasting 24 h but no dyspareunia. There was no history of genital trauma, infections, or similar previous lesions. The only medical history was a cervical intraepithelial neoplasia 3, which was treated with a large loop excision of the transformation zone. There was no relevant family history. The patient had a normal adult hair pattern and body mass index of 37 kg/m². On pelvic examination, a 1-cm firm tender mass adjacent to the glans clitoris on the right with smooth overlying skin was noted (Fig. 1). No pigmentation or ulceration of the skin was seen. Magnetic resonance imaging (MRI) of the pelvis with contrast was done to characterize the lesion. It confirmed a homogeneously solid clitoral lesion measuring 14 × 10 × 13 mm (Fig. 2). It was moderately T2 hyperintense with homogeneous enhancement following contrast. There was diffusion restriction suggestive of marked cellularity. There was no lymphadenopathy or invasion of adjacent structures, but malignancy could not be excluded.

A surgical excision was carried out given the patient's symptoms and the potential for malignancy. A lateral approach to the fold of the clitoris medial to the pudendal cleft was taken to avoid the neurovascular bundle. The soft-tissue mass was then separated with sharp and blunt dissection from its capsule and the surrounding tissue. The mass was excised completely and removed intact with minimal blood loss and the overlying tissue closed with 2 layers of absorbable sutures. The patient was discharged home on the same day of surgery. A 14-mm firm mass with smooth outer surface was excised with narrow but clear margins, given the potential to compromise patient's sexual function.
3. Pathological Findings

Macroscopic examination revealed a firm, circumscribed mass measuring 15 × 14 × 12 mm with white cut surface, which was processed in its entirety. The histopathology showed a well circumscribed tumor with multiple slit-like, almost pericytomatous vessels. The vessels were separated by cells with mostly round uniform nuclei merging with spindle cells with fusiform nuclei and cytoplasmic eosinophilia (Fig. 3). The morphology of the tumor was between glomus cells and smooth muscle cells. The immunohistochemistry showed positive staining for desmin, caldesmon and smooth muscle actin and negative for cytokeratin and S100, in keeping with a glomangiomyoma (Fig. 4).

4. Discussion

Glomus tumors usually present as lesions which measure less than 2 cm, and comprise less than 2% of soft-tissue tumors in the extremities. The typical presentation of glomus tumors is as a painful nodule in the dermis or subcutis of the distal extremities, especially the tips and nail beds of digits. Glomus tumors in other anatomical sites where glomus bodies are sparse or even absent, including the viscera, have also been reported [2]. There are isolated cases reported in the female genital tract, including in the cervix, uterus, ovary, vulva, vaginal area [3], periurethral area [1,4] and clitoris [1,5]. Usually, dermal glomus tumors present with symptoms of pain; however, the patient in this case presented with only minor discomfort and a post-coital ache.

The distribution and proportion of the three main components of the glomus tumor (vessels, glomus cells and spindle cells) may vary considerably and this determines their subtypes.

The list of differential diagnoses for clitoral masses is extensive and includes the more commonly seen lesions of squamous cell carcinoma, neurofibroma [6], epidermoid cyst [7], peri-clitoral abscess [8].
hemangioma [9] and leiomyoma [10]. Clitoral epidermal inclusion cysts have a distinctive cystic appearance on MRI like Bartholin gland and Skene gland cysts. Peri-clitoral abscess is often hypointense on T1WI (weighted imaging), heterogeneously hyper-intense on T2 - WI, with thick rim enhancement. Similar to the previously reported MRI appearance of glomus tumor at the more common location of the subungual region, a solid and well-defined ovoid mass in this case was seen with high T2 signal [11]. The high T2 signal can sometimes be misleading and suggest a cystic nature to the lesion, but the homogenous enhancement on the post-contrast sequence confirms a solid rather than cystic lesion.

Surgical excision of clitoral masses presents challenges due to bleeding risk, as well as cosmetic and sexual function concerns. An eccentric, semilunar incision was performed lateral to the clitoral hood, allowing access to the mass while avoiding the innervation and blood supply, similar to the method previously described [5]. Local excision was considered adequate and no adjuvant therapy was required in this case. The patient was followed up with clinical observation and was well 5 months after the surgery.

5. Conclusion

In conclusion, glomangiomyoma located in the clitoris is a rare benign condition, and should be included in the differential diagnosis of clitoral smooth tumors. Surgical planning involving the clitoris is complicated due to the risk of functional impairment. MRI provides assistance in differential diagnosis and planning treatment; however, histopathology is needed for definitive diagnosis.

Contributors

Huan Xie contributed to data curation, resources, and writing the original draft.
Sameera Ansar contributed to drafting the manuscript, and review and editing.
Lyndal Anderson contributed to conceptualization, validation, and review and editing.
Samir Saidi contributed to conceptualization, and review and editing.
Yu Xuan Kitzing contributed to investigation, and review and editing.
Sunaina Anand contributed to conceptualization, and review and editing.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Provenance and Peer Review

This case report was peer reviewed.

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