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A clitoral epidermal inclusion cyst in a postmenopausal woman: a case report

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ABSTRACT

Background. Clitoral cysts are rare conditions typically observed in the paediatric population, manifesting as either benign or malign entities, and may be congenital or acquired. The most prevalent forms are epidermal inclusion cysts, often associated with trauma, particularly female genital mutilation, although their aetiology can sometimes remain unclear.
This study aims to present an atypical case of a clitoral cyst occurring in an adult, post-menopausal woman, without any evident risk factor.
Case presentation. A 56-year-old post-menopause patient was referred to the emergency department with a 4 cm spontaneous swelling in the clitoral area. The patient denied any history of genital trauma and reported a gradual increase in size over the past two years, accompanied by discomfort and dyspareunia.
Upon clinical examination, ultrasound revealed a unilocular, hypoechoic, non-vascularized cystic structure. Subsequently, surgical excision was performed, and pathologic examination confirmed it as an epidermal inclusion cyst.
To date, the literature reports only 14 cases of spontaneous clitoral epidermal inclusion cyst in post-puberal and adult patients, highlighting their exceedingly rare occurrence in adult women without a history of trauma.
Conclusions. Despite their benign and rare nature, epidermal inclusion clitoral cysts may induce significant discomfort, potentially leading to sexual dysfunction if left untreated. Therefore, it is imperative for clinicians to recognize them, considering the like hood of their occurrence and growth even post-menopausal women. Addressing this condition through proper counselling and prompt surgical intervention is crucial to ensure a satisfactory quality of life for affected patients.

INTRODUCTION

Epidermal inclusion cysts are benign, spontaneous, cutaneous cysts that are usually found on the face, neck, back or scalp, and less commonly on the vulvar area, though they can occur in various locations. They are caused by the implantation of epidermal elements into the dermis layer of the

skin and are usually filled with laminated keratinous material [1]. Epidermoid cysts of the clitoris are a very rare cause of acquired non-hormonal clitoromegaly.

Their occurrence usually follows a traumatic event, which is commonly represented by the female genital mutilation practice in case the cyst is located in the genital area. These cysts are extremely uncommon without prior history of genital surgery, infibulation, circumcision, or genital piercing [2].

The diagnosis of epidermoid cysts is usually done clinically and is based on the appearance of a soft, freely moveable cyst, often with a visible central punctum [1]. Radiologic imaging may be helpful to corroborate the clinical diagnosis; ultrasound evaluation shows these cysts as unilocular, hypoechoic, non-vascularized masses [3], and magnetic resonance imaging assessment could be useful, especially for determining the surgical excision strategy, given that such cysts have well-defined masses with high T2 signal background [3].

Since their degeneration into malignant squamous cell carcinoma is very rare, epidermal inclusion cysts generally do not require treatment but follow-up only [4]. They often stop growing after reaching a size of 1-5 cm in diameter [5]. However, if their growth causes severe discomfort and pain, a surgical approach should be considered as the first-line treatment [5].

While existing literature predominantly describes rare cases of spontaneous clitoral epidermal inclusion cysts, mostly in the paediatric population, this study presents a case of a large clitoral epidermal inclusion cyst in a post-menopausal woman without a history of genital trauma. Additionally, a review of all reported cases of spontaneous clitoral epidermal inclusion cysts in post-pubertal women was performed, contributing to the understanding of this rare condition.

CASE PRESENTATION

A 56-year-old woman referred to the gynaecological outpatient clinic for a routine check. She reported no history of genital trauma or mutilation and had two vaginal deliveries. Apart from hypertension, she was in good health. The patient complained of a painless clitoral mass that had been progressively growing over the past two years, causing dyspareunia and psychological distress during sexual intercourses. There were no urinary complaints.

Pelvic examination revealed a mobile, soft, non-tender, well-defined clitoral mass measuring approximately 4 cm, located above the urethra. The clitoral hood could not be retracted for visualization. No visible punctum or skin anomalies were noted (**Figure 1**). Results of the general physical examination were normal and there were no other signs of hyperandrogenism.

An ultrasound scan characterized the mass as a 37 × 23 mm non-vascularized anechoic cyst. Ultrasound examinations of the uterus, adnexal regions, and bladder were normal. Lab investigations were within normal limits. Discussions regarding the pros and cons of a wait and see approach took place, considering the patient's discomfort, and an elective surgery for the mass excision was organized with the patient's consent.

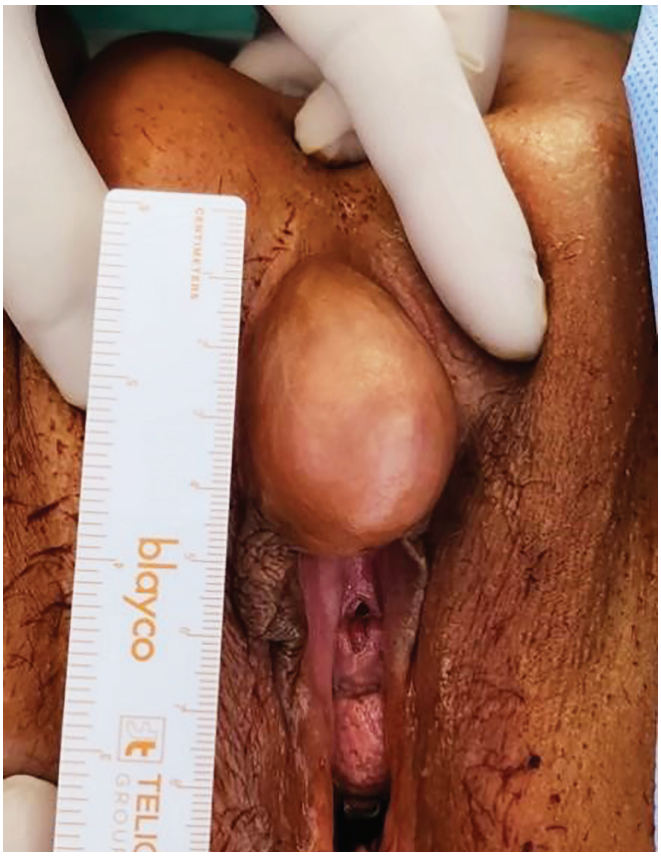

Figure 1. *Clitoral cyst before surgery.*

The mass was excided under spinal anaesthesia by both a gynaecologist and a plastic surgeon, ensuring a collaborative and comprehensive approach. They performed a longitudinal incision, and the cyst was removed with combination of sharp and

blunt dissection, paying attention to avoiding the neurovascular bundle areas of the clitoris. The incision site was sutured with separate 3-0 absorbable sutures (**Figure 2**), chosen for their appropriate dissolution rate and to minimize the need for suture removal in this sensitive area. The patient was discharged on the same day with anti-inflammatory medication and a wound-healing topic treatment, aimed at ensuring optimal recovery and minimizing discomfort. The post-operative period was completely uneventful.

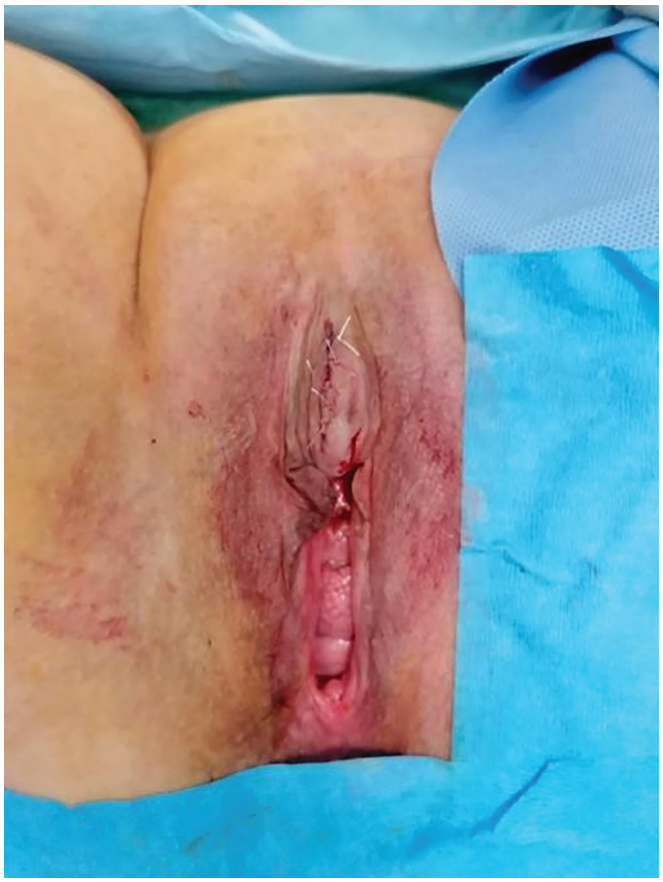

Figure 2. *Inspection of vulvar region following surgery.*

The histopathology report revealed a 4 cm cystic formation, with thin walls, lined with stratified keratinizing epithelium. These findings were instrumental in confirming the diagnosis of an epidermal inclusion cyst, guiding further management decisions.

A 6-week follow-up visit was scheduled, during which it was noted that the lesion was completely remodelled and adherent, without clinical signs of recurrence (**Figure 3**). Sensibility was preserved, and the patient referred a positive and satisfying return to her sexual life.

A signed informed consent for reporting and publishing this case was obtained from the patient.

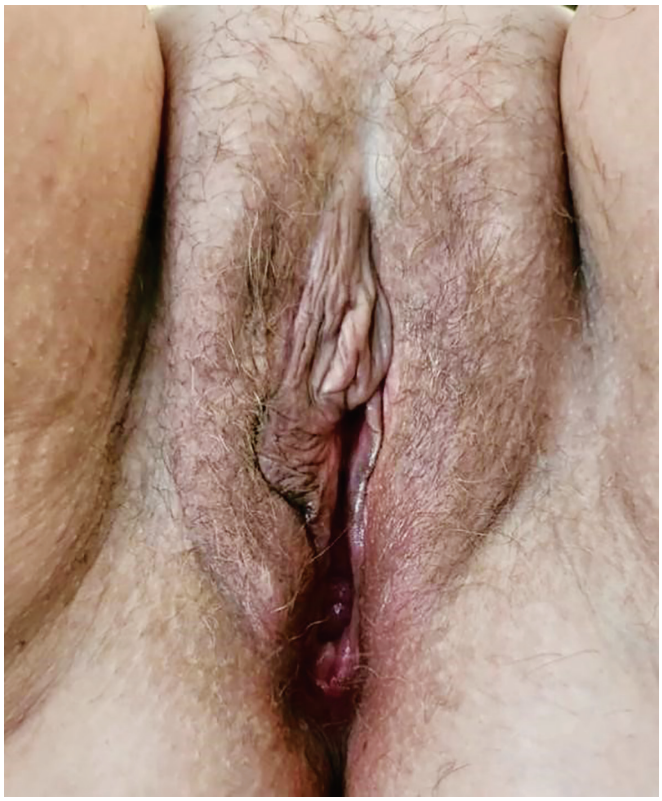

Figure 3. *Inspection of vulvar region at 6-week follow-up.*

DISCUSSION

When conducting a comprehensive search across major databases including PubMed, Medline, and Google Scholar, using key terms such as "clitoral", "clitoris", "epidermal cyst" and "epidermoid cyst", a total of 41 results were identified. Focusing on case reports and meta-analyses in English that involved adult and post-pubertal patients without history of genital trauma or surgery, 14 cases from 1999 until nowadays [3-14] were reviewed, in addition to the current case under analysis (**Table 1**). Among the reviewed cases, one occurred 18 months after caesarean-section delivery [6], and another during pregnancy [9]. In the latter case, the patient described a gradual and painless clitoral enlargement during pregnancy, with spontaneous regression within two months post-delivery. Intriguingly, five months later, the clitoral enlargement recurred and persisted during her second pregnancy.

Additionally, a case involving a 17-year-old patient highlighted a progressive regrowth of the

cyst following vaginal delivery. Another unique case involved a 15-year-old patient experiencing a spontaneous recurrence seven years after iatrogenic drainage [8].

In the reviewed cases (**Table 1**), the mean age at the time of diagnosis was 27.5 ± 11.1 years old, with a mean time of growth of 21 ± 20.4 months. The mean cyst size was 4.8 ± 1.6 cm. Noteworthy findings include sexual-related symptoms reported by most adult patients, with dyspareunia observed in 25% and anorgasmia in 8.3%. Additionally, two patients presented urinary symptoms such as urinary frequency and voiding difficulty (16.6%), and a 47 years-old woman reported difficulties in walking due to the large cyst size (8.3%). Psychological distress was reported in 33% of the cases.

All patients underwent surgical excision using various techniques such as longitudinal incision, supra-meatal inverted-U-incision, and curvilinear incision, with no discernible clinical difference in post-operative recovery and patients' satisfaction. There has been only one case of post-operative complication described – a secondary infection

Table 1. *Reported cases of adult-onset spontaneous epidermoid cyst of the clitoris.*

	Reference	Age (years)	Time of growth (months)	Size (cm)	Clinical presentation	Treatment	Follow-up (months)	
1	Current case	56	24	4	Dyspareunia and psychological distress	Surgical excision (longitudinal incision)	3	Well-being, no recurrence
2	Fux-Otta *et al*. [4], 2022	16	12	4	Emotional discomfort	Surgical excision	12	Sexual satisfaction, discomfort resolution, well-being
3	Prasad *et al*. [5], 2022	43	36	6	Dyspareunia	Surgical excision (curvilinear incision)	3	Asymptomatic, sexual discomfort resolution, no recurrence
4	Oluwaferanmi *et al*. [6], 2022	31, 18 months postpartum	19	8	ND	Surgical excision	ND	Secondary infection, successfully treated with antibiotics
5	Mahmoudnejad *et al*. [7], 2021	45	6	5	Asymptomatic	Surgical excision (supra-meatal inverted-U-incision)	6	Sexual and tactile sensation preserved, no recurrence
		35	24	3	Dyspareunia and difficulties in voiding		12	Asymptomatic, no recurrence
		25	24	5	Urinary symptoms (urinary frequency and difficulties in voiding)		48	Asymptomatic, sexual well-being
6	Angela M. DiCarlo-Meacham *et al*. [8], 2020	15, recurred at 22	84	4	Anorgasmia	Surgical excision (transverse incision)	7	Orgasm regained
7	Hughes *et al*. [9], 2013	31, pregnant	5 postpartum	7	Psychological distress	Surgical excision	2	Asymptomatic, no recurrence
9	Johnson *et al*. [3], 2010	15	24	5	Asymptomatic, social anxiety	Surgical excision	3	Anatomy restoration, sexual and tactile sensation preserved, no recurrence
10	Paulus *et al*. [10], 2010	47	12	4	Pain	Surgical excision	ND	ND
11	Anderson-Mueller *et al*. [11], 2009	17	Spontaneous discharge, recurrence	6	Pain	Surgical excision	ND	Asymptomatic
12	Fedele *et al*. [12], 2007	22	10	7	Asymptomatic	Surgical excision (vertical incision)	ND	Anatomic restoration, preservation of sensibility
13	Linck *et al*. [13], 2002	28	ND	3	ND	Surgical excision	ND	ND
14	Schmidt *et al*. [14], 1999	16	8-10	2	ND	Surgical excision	18	No recurrence

Table 2. *Causes of clitoromegaly.*

Congenital	Acquired	
	Hormonal	Non hormonal
CAH	Polycystic ovary syndrome	Neurofibromatosis
Maternal hyperandrogenism:	Virilizing ovarian tumours	Vulvar tumours, primary or metastatic
Luteoma	Leydig-cell tumour	Haematomas
Hyperreaction luteinalis	Granulosa cell tumour	Inguinal hernias
Virilizing tumours	Thecomas	Abscesses
Exogenous androgens intake	Sex-cord stromal tumours	Diverticula
	Virilizing adrenal tumours	Endometrioma
	Adrenocortical carcinoma	
	Adrenocortical adenoma	
	Synthetic androgen exposure	

on the 2nd post-operative day, successfully treated with antibiotics

Follow-up was reported in 66,7% of cases (10/15 patients). Women attended follow-up visits from three months to four years after the surgery (median 9 months), with all reporting resolution of the symptoms. Anatomical restoration, preserved sexual function, and maintained tactile sensation were consistently observed. Importantly, no cases of cyst recurrence were reported after surgery.

Conditions affecting the female external genitalia encompass vaginal (hymenial), para-urethral, and clitoral cysts. Among these, clitoral conditions include a spectrum of benign, malignant, and rarely metastatic lesions. Clitoromegaly is a rare condition that can be either congenital or acquired and can be attributable to hormonal or non-hormonal causes (**Table 2**).

The most common form of congenital clitoromegaly is associated with congenital adrenal hyperplasia (CAH), a condition that includes a group of genetic disorders affecting the adrenal glands and resulting in an increased production of adrenal-derived androgens. The excessive production of testosterone leads to various clinical signs and symptoms, including genitals' virilization, among which clitoromegaly is one of the most typical forms. More commonly, adult patients with ambiguous genitalia are found to have late-onset CAH, and present typical clinical signs such as hirsutism, irregular menses, or acne [15].

Acquired clitoromegaly is unusual and can be due to either hormonal or non-hormonal causes.

Hormonal causes of clitoromegaly often involve virilizing tumours, including Leydig-cell tumours, sex-cord stromal tumours, granulosa cell tumours or thecomas. Additional hormonal contributors encompass polycystic ovary syndrome and Cushing syndrome, and specific conditions observed during pregnancy, such as luteoma, hyperreaction luteinalis, and placental aromatase deficiency [9].

Non-hormonal causes, though rarer, encompass a spectrum of conditions. Non-tumoral forms include abscesses, diverticula, inguinal hernias, post-traumatic hematomas, or endometriomas. Tumoral forms range from benign entities like leiomyomas, pseudo-lymphomas, fibromas, angiokeratomas, haemangiomas, hemangiopericytomas, granular cell tumours, or neurofibromas, to malignancies such as endodermal sinus tumours, sarcomas, schwannomas, lymphomas, rhabdomyosarcomas, epithelioid haemangioendotheliomas or secondary metastasis (**Table 3**).

Acquired non-hormonal clitoromegaly is a rare phenomenon, and withing this spectrum, the occurrence of epidermoid cyst in the clitoris is even more uncommon. Typically associated with genital mutilation in certain ethnic communities in Africa and West Asia, these cysts also manifest spontaneously without prior history of infibulation or piercing [16]. The evaluation of clitoromegaly poses a diagnostic challenge for clinicians due to the rarity and diverse aetiologies of this condition. Achieving an accurate differential diagnosis becomes imperative for guiding appropriate and effective treatment strategies.

Table 3. *Vulvar tumors.*

Benign	Malign
Leiomyoma	Endodermal sinus tumour
Pseudo lymphoma	Sarcoma (liposarcoma)
Fibroma	Schwannoma
Angiokeratoma	Lymphoma
Haemangioma	Rhabdomyosarcoma
Hemangiopericytoma	Epithelioid haemangioendothelioma
Granular tumour cell	
Neurofibroma	Metastasis

CONCLUSIONS

While clitoral cysts are exceedingly rare, they play a crucial role in the differential diagnosis of clitoral hypertrophy. The evaluation of adult-onset clitoromegaly demands a comprehensive approach, involving meticulous physical examination, hormonal testing, and imaging studies. Surgical removal emerges as the preferred treatment for clitoral cyst due to the rarity of spontaneous regression and the potential recurrence after the procedure.
Studies have shown that timely diagnosis and treatment play a pivotal role in preventing negative outcomes, particularly in not-yet sexually active patients, while also contributing to the restoration of sexual well-being in those who have initiated sexual activity [17, 18].

COMPLIANCE WITH ETHICAL STANDARDS

Authors' contribution

F.S.: Writing – review & editing. A.L.: Conceptualization, writing – original draft. A.C, A.T., L.T.: Writing – review & editing. V.R.: Conceptualization, writing – review & editing.

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N/A.

Disclosure of interests

The authors declare that they have no conflict of interests.

Ethical approval

N/A.

Informed consent

A signed informed consent for reporting and publishing this case was obtained from the patient.

Data sharing

Data are available under reasonable request to the corresponding author.

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