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Fibroadenoma as a rare cause of perianal pain: A review of literature

Sonia Morales Artero, Camilo Castellón, Elena Larraz, Federico Álvarez, Montserrat Calvo, Mar De Castro

ABSTRACT

Introduction: Fibroadenomas are the most frequent breast tumors in women. However, their location in the anogenital region is extremely rare. Case Report: We present a case of a perianal fibroadenoma in a young woman. It was necessary the surgical treatment to reach a definitive diagnosis. Conclusion: It's important to highlight the glandular mammary-like tissue present in the perianal region as the origin of different lesions, in order to make an adequate differential diagnosis.

Keywords: Apocrine, Fibroadenoma, Perianal pain

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INTRODUCTION

Fibroadenoma (FA) is the most common breast tumor in women. Several extramammary locations have been described (gallbladder, prostate, face and eyelid) [1], while the anogenital region (perineal, perianal and vulvar) a very rare one. This tumor was traditionally considered to originate in ectopic breast tissue, as other breast tumors do; however, after van der Putte's publication in 1991 [2], it is accepted as normal to find glandular mammary-like tissue in the anogenital region, with response to humoral stimulation owing to the transformation of eccrine glands into apocrine glands.

CASE REPORT

A 34-year-old woman with a history of polycystic ovary syndrome, reporting a one year long perianal tumor with progressive growth, with menstrual pain not responding to oral contraceptives prescribed for dysmenorrhea. She has no fever, suppuration or rectal bleeding. At rectal examination, next to the anal verge, located at 9h in genupectoral position, a 5 cm soft elastic tumor is palpated, showing no inflammatory signs. A percutaneous puncture is done, obtaining no pus. Endoanal ecography is normal. Pelvic magnetic resonance (Figure 1) shows a solid non-specific ovoid mass in the subcutaneous tissue, measuring 67 mm in its longer axis, ruling out lipoma or endometriosic implant, but consistent with an inflammatory or neoplastic mass; no abnormal lymph nodes are identified.

Under spinal anesthesia, complete excision of the lesion is achieved with security margins. The patient is discharged on the first postoperative day and no early complications occur. Macroscopically, a nodular non encapsulated well limited lesion is identified, measuring 6x4.5x1 cm, pink colour (Figure 2). Microscopically, a

fibroepithelial neoformation, with a predominant stromal component made of fibroblastic type cells, displayed over a collagen matrix. The epithelial component is organized in ducts and acini with a visible and free lumen (pericanalicular pattern), and no endoluminal necrosis or calcification is identified. Signs of apocrine differentiation can be seen on the luminal epithelium in some ducts. There are no signs suggesting malignancy. Immunohistochemistry reveals activity for humoral receptors (estrogens and progesterone) on the luminal epithelium. According to the pathologic findings, a fibroadenoma with apocrine differentiation is diagnosed (Figure 3).

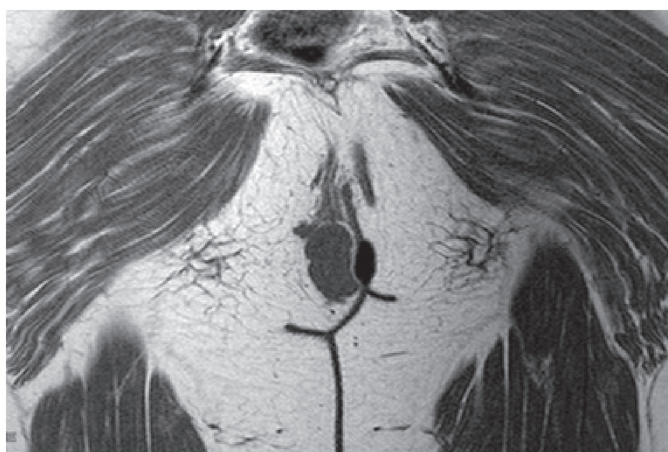

Figure 1: Preoperative Magnetic Resonance Image: a solid non-specific ovoid mass in the subcutaneous tissue, measuring 67 mm in its longer axis.

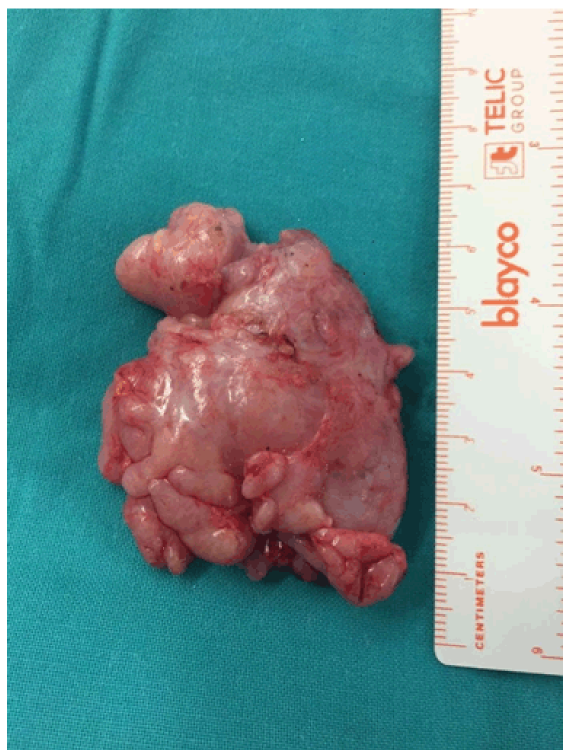

Figure 2: Resected nodular lesion measuring 6x4.5x1 cm and pink colour.

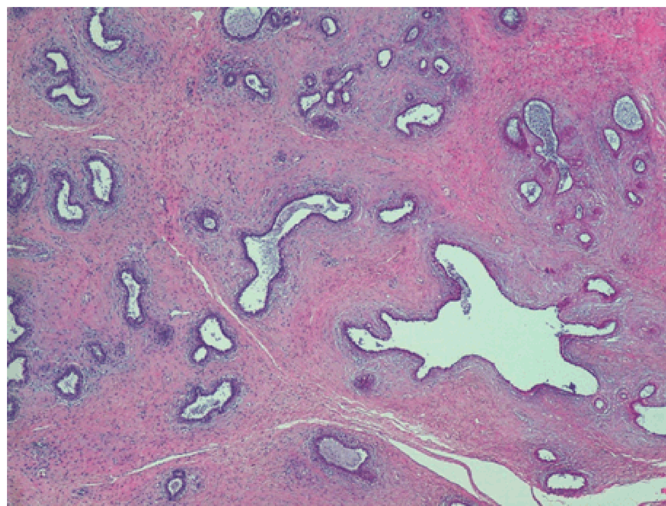

Figure 3: Benign fibroepithelial neoformation, with a predominant stromal component made of fibroblastic type cells, displayed over a collagen matrix.

DISCUSSION

Anogenital FA is more frequent in women in the fourth decade of life [3]. However, perianal location is an exception. A thorough review of literature has only found 13 cases of perianal FA in women (Table 1). All the lesions have been unique, except for one case in which three simultaneous lesions were found [4]. Most of the cases presented in women of fertile age, with a median age at diagnosis of 42,7 years (27–85 years) and a median size of 3.9 cm (1.5–10 cm). In one case, the diagnosis was made incidentally on pathology after perianal fistulectomy [5]; in another case, the lesion showed a progressive growth during pregnancy [6]. Half of the lesions found are pediculated and 42.8% of them were accompanied by perianal pain or discomfort. Our case was the only one that showed pain directly related with menstruation. Time of evolution has been widely variable, from three weeks to 17 years.

Preoperative diagnosis is not easy; differential diagnosis must be made between hemorrhoids, perianal abscess, endometriosis, primary anorectal tumors and other malignant lesions of the anogenital region (Paget's disease and invasive carcinoma) [1, 6–10]. Magnetic resonance and puncture for cytology may be helpful [5]. However, surgical treatment is mandatory to reach final pathologic diagnosis. This lesions are usually well limited, microscopically identical to breast FA, with glandular structures surrounded by a low cellularity, scarcely mitotic stroma [3, 5]. Most of them have estrogen and progesterone receptors. Phyllodes tumor (PT), which is more irregular and with more stromal cellularity, must be differentiated [3, 9, 10]. There are three types of PT in the breast, depending on the identification of stromal atypia: benign, low grade and high grade [3, 10]. High grade PT of the anogenital region has not been described up to now, although lesions with pathological findings of FA and PT

Table 1: Features of perianal fibroadenomas described in literature

Author (year)	Age	Maximum size (cm)	Estrogen receptors	Progesterone receptors	Symptoms	With pedicle	Time of evolution
Assor (1977)	37	4	ND	ND	No	Yes	10 yrs
Donati (1996)	27	4	ND	ND	No	Yes	17 yrs
Solomon (2006)	65	2,1	+	+	Yes	Yes (*)	3 months
Choi (2007)	30	4,5	+	+	Yes	Yes(*)	3 weeks
Ahmed (2007)	36	5,2	+	+	No	Yes (*)	ND
Vella (2008)	46	3	+	+	No	No	3 months
Doganavsargil (2008)	42	4	+	+	Yes	No	1 yrs
Charfi (2009)	85	10	+	+	ND	Yes	ND
Ekci (2010)	45	2	-	+	No (**)	No	ND
Kazakov (2010)	46	1,5	ND	ND	ND	No	ND
	42	2	ND	ND	ND	No	2 yrs
Grube-Pagola (2012)	28	5	+	+	Yes	Yes	4 yrs
Duncan (2016)	36	2,1	+	ND	Yes	No	ND
Morales (2018)	34	6	+	+	Yes	No	1 yr

ND: not described; (*): clinical presentation as hemorrhoids; (**): clinical presentation as perianal fistula.

on the same specimen have been reported [9]. Prognosis after surgical excision is excellent and no recurrences have been found in cases of FA without simultaneous PT.

CONCLUSION

In conclusion, the presence of mammary-like glands on the anogenital region has to be taken into account whenever a perianal tumor is found in women; and differential diagnosis between other more common perianal lesions must include a benign or malignant lesion of mammary tissue.

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Author Contributions

Sonia Morales Artero – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Camilo Castellon – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Elena Larraz – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Federico Álvarez – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Montserrat Calvo – Substantial contributions to conception and design, Acquisition of data, Analysis

and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Mar De Castro – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

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

Conflict of Interest
Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

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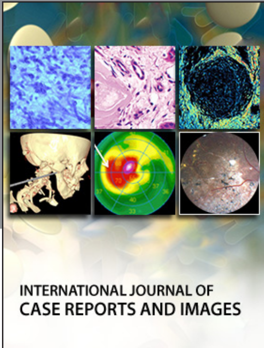

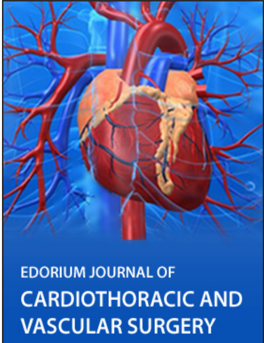

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