Soft fibroma of the nipple: case report

Fibroma mole no mamilo: relato de caso

Fibroma blando del pezón: reporte de un caso

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ABSTRACT

Objective: To report the case of a 20-year-old female patient with an unusual presentation of soft nipple fibroma as well as its surgical excision with confirmation of the diagnosis by histopathology, also reviewing all cases of soft nipple fibroma so far published. **Case details:** Female, 20 years old, with a pedunculated, soft and painless lesion on the left nipple with insidious growth since she was six years old. Tangential surgical excision and histopathological examination were performed with findings of irregular acanthosis, hyperkeratosis, papillomatosis, dermis with connective tissue with loose collagen fibers and numerous capillaries with discrete mononuclear infiltrate, evidencing the diagnosis of soft fibroma. **Final considerations:** Soft nipple fibroma is a rare benign lesion that usually evolves well after surgical treatment. To date, there are few publications of soft fibroma with this location. Even though it is a benign skin neoplasm, soft fibroma can cause local pain due to trauma in the region, blisters due to local friction and decreased self-esteem when related to aesthetic issues and personal acceptance.

Keywords: Fibroma, Nipples, Polyps.

RESUMO

Objetivo: Relatar o caso de uma paciente de 20 anos de idade com apresentação incomum de fibroma mole no mamilo assim como sua excisão cirúrgica com a confirmação do diagnóstico pela histopatologia, revisando também todos os casos já publicados até o momento de fibroma mole no mamilo. **Detalhamento do caso:** Mulher, 20 anos de idade, com lesão pedunculada, amolecida e indolor no mamilo esquerdo com crescimento insidioso desde seus seis anos de idade. Realizado excisão cirúrgica tangencial e exame histopatológico com achados de acantose irregular, hiperqueratose, papilomatose, derme com tecido conectivo com fibras colágenas frouxas e numerosos capilares com discreto infiltrado mononuclear, evidenciando o diagnóstico de fibroma mole. **Considerações finais:** O fibroma mole no mamilo é uma lesão benigna rara que geralmente evolui bem após tratamento cirúrgico. Até o momento existem poucas publicações de fibroma mole com esta localização. Mesmo sendo uma neoplasia benigna da pele, o fibroma mole pode acarretar dor local devido a traumas na região, bolhas por atrito local e diminuição da autoestima quando relacionado à questões estéticas e à aceitação pessoal.

Palavras-chave: Fibroma, Mamilos, Pólipos.

RESUMEN

Objetivo: Reportar el caso de una paciente de sexo femenino de 20 años de edad con una presentación inusual de fibroma blando del pezón, así como su extirpación quirúrgica con confirmación del diagnóstico por histopatología, revisando además todos los casos de fibroma blando del pezón publicados hasta el momento.

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Detalle del caso: Femenina, 20 años, con lesión pedunculada, blanda e indolora en pezón izquierdo de crecimiento insidioso desde los 6 años. Se realizó escisión quirúrgica tangencial y examen histopatológico con hallazgos de acantosis irregular, hiperqueratosis, papilomatosis, dermis con tejido conectivo con fibras colágenas sueltas y numerosos capilares con discreto infiltrado mononuclear, evidenciando el diagnóstico de fibroma blando. **Consideraciones finales:** El fibroma blando del pezón es una lesión benigna rara que suele evolucionar bien después del tratamiento quirúrgico. Hasta la fecha existen pocas publicaciones de fibroma blando con esta localización. Aunque es una neoplasia cutánea benigna, el fibroma blando puede causar dolor local por traumatismo en la región, ampollas por roce local y disminución de la autoestima cuando se relaciona con cuestiones estéticas y de aceptación personal.

Palabras clave: Fibroma, Pezones, Pólipos.

INTRODUCTION

The skin is the largest organ in the human body, an important barrier between our organism and the external environment. In addition to being able to arouse sensations, it expresses several of significant signs and symptoms, which are of fundamental aid in the identification of many diseases. The diseases that affect the skin can be restricted to it or sometimes the skin expresses signs and symptoms as the first manifestation, or late manifestation of systemic diseases. The breast and nipple form a unique functional unit of the skin with significant emotional impact. The evaluation and management of dermatologic diseases in this area requires proper sensitivity and a suitable environment for physical examination (MINISTÉRIO DA SAÚDE, 1992),

Anatomically, the female breast is composed of lobes: milk-producing glands; ducts: small tubes that carry milk from the lobes to the nipple and stroma: adipose tissue and connective tissue that surrounds the ducts and lobes in addition to blood vessels and lymphatic vessels. The mammary gland is a paired organ, which is located on the anterior and superior wall of the chest and is supported by the pectoralis major muscle; extends from the second to sixth rib in the vertical plane and from the sternum to the anterior axillary line in the horizontal plane (WALDMAN RA, et al., 2019),

Soft fibroma, also called acrochordon or fibroepithelial polyp, is a very common benign fibrohistiocytic mesoderm-derived proliferation of the skin found in approximately 50% of individuals predominantly in adults and elderly people. Usually it does not cause any symptoms and varies in size between 2 and 10 mm in diameter (SOCIEDADE BRASILEIRA DE DERMATOLOGIA, 2016).

Different areas can be affected, however it most presents itself in the axilla, around 48% of the cases, followed by the neck and inguinal region. Moreover, soft fibromas can increase in diameter over time, which may happen according to the person's biological conditions (BANIK R and LUBACH B, 1987).

The cause of the appearance of soft fibroma is still not very well defined, however, it may be associated with family history of soft fibromas and chronic diseases such as obesity, dyslipidemia, diabetes *mellitus* and hypothyroidism. Human papillomaviruses 6 and 11 have also been incriminated in their development. (AKPINAR F and DERVIS E, 2012). Although soft fibroma were once thought to be associated with colonic polyps, more recent studies have not confirmed this relationship (GOULD BE, et al., 1998).

Most of the time soft fibroma presents itself as a pedunculated papule with a variably sized stalk, cauliflower-like surface, softened consistency and skin-colored to slightly hyperpigmented. The diagnosis of soft fibroma of the nipple is very rare. Even though their clinical characteristics are usually markable, the unusual shapes, size, and locations may cause confusion, hence the histopathological analysis is essential. Histologically, soft fibroma is a polypoid lesion with loose to dense collagenous stroma and thin-walled blood vessels (ARORA BK, 2019).

Despite being related to aesthetic issues, soft fibroma can cause discomfort or bleeding when rubbing against clothing, for instance, or even during hair removal, interfering with the patient's routine. Surgical excision is the treatment of choice. In most cases, an imperceptible scar is left, unless the patient has a tendency to develop a keloid scar (ELDER DE, et al., 2007).

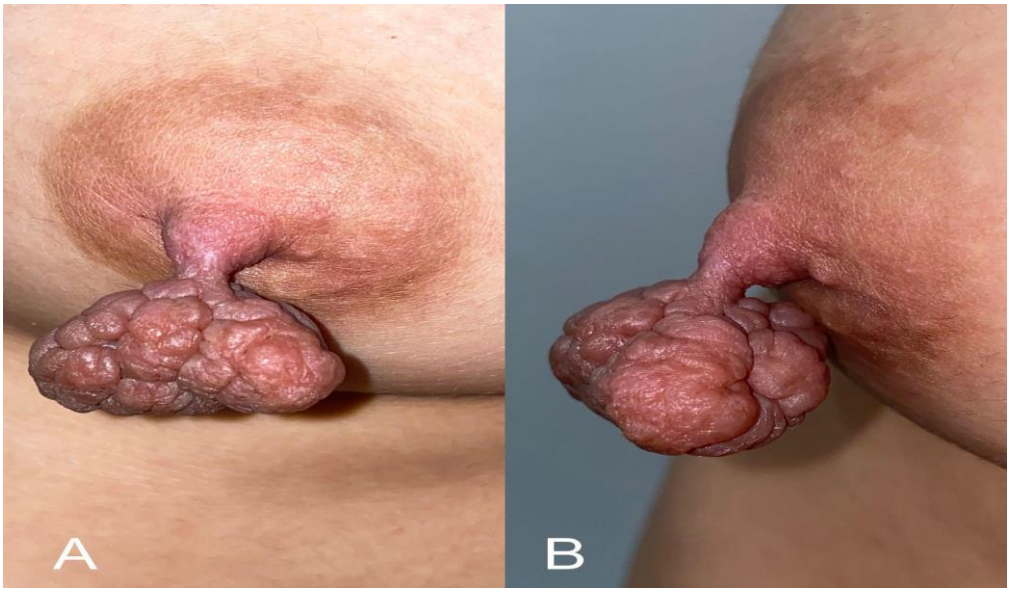
This paper aims to report a case study of a 20-year-old female patient who presented soft fibroma of the nipple since childhood, as well as the surgical treatment of the lesion, histopathological analysis, two-year follow-up after surgery and revision of literature of all similar cases so far published.

DETAILS OF THE CASE

This is a case report reviewed and approved by the Research Ethics Committee (CEP) (number 5.702.636 and CAAE 63987522.0.0000.5519), with the patient's agreement through the signing of the Free and Informed Assent Term (TCLE) and the Free and Informed Consent and Image Use Authorization Term.

A 20-year-old woman presented with a pedunculated, exophytic, papillomatous, normochromic, soft nodule on the left nipple (**Figure 1**). The lesion was approximately 3 cm in its long axis with no bleeding, discharge, or ulcers on the surface. The growth was insidious and progressive from the age of six years. The rest of the skin was normal.

Figure 1 - Left nipple.



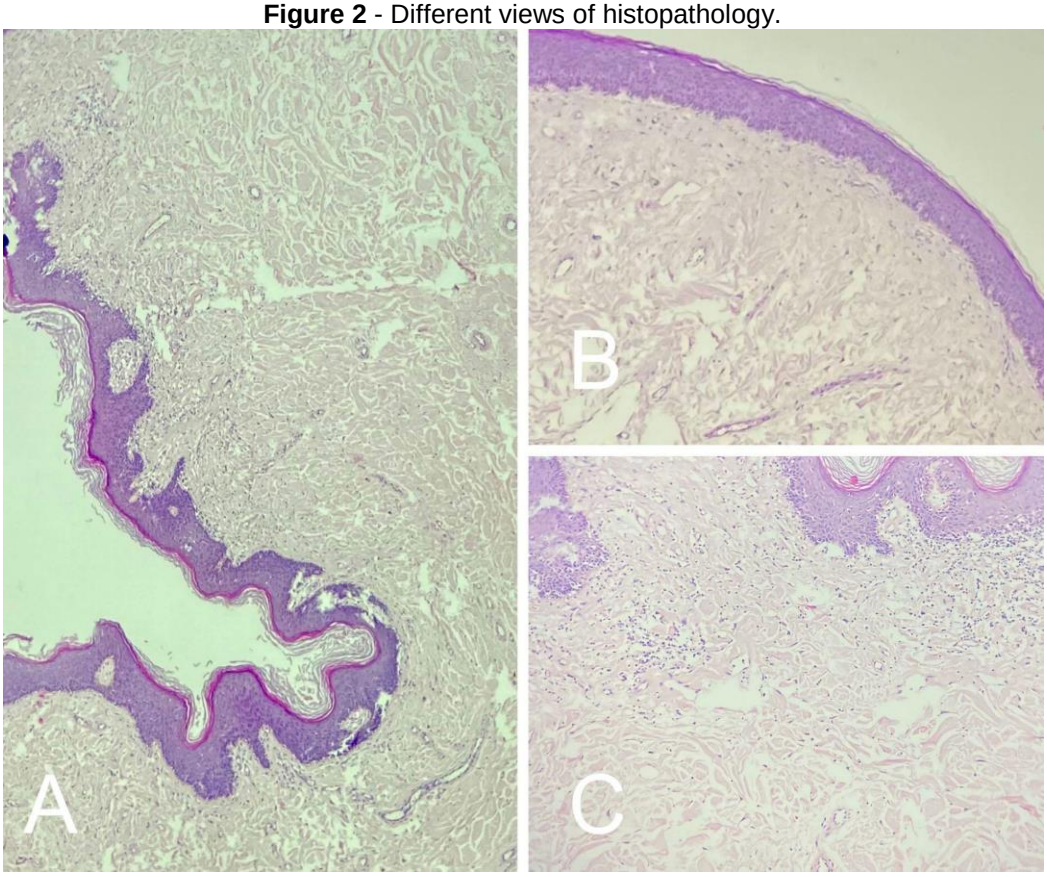
Subtitle: (A) Frontal view. (B) Lateral view.
Source: Assis GG, et al., 2022.

The patient complained about the aesthetic appearance of the lesion and that she had started getting concerned due to its increasing size. The patient denied any pain, itching, fluid leakage, trauma, previous surgery, local inflammation or other symptoms. She also denied use of medication or any personal or family history of breast cancer. The patient was healthy with no history of metabolic or endocrinologic diseases.

There were no other clinical findings in the areola-nipple complex, breast nodules or lymph node enlargement. Breast ultrasound demonstrated a pedunculated lesion on the left nipple associated with normal and regular skin, normal areola-nipple complex, normal homogeneous hypoechoic subcutaneous fat, normal heterogeneous glandular parenchyma, preserved retromammary fascia and no suggestive signs of malignancy.

The lesion was surgically treated with local anesthesia (2% lidocaine and epinephrine) followed by tangential excision (shaving) and primary closure with simple nylon 5.0 stitches. The stitches were removed after ten days. There were no complications after the surgery. Histopathological evaluation showed regular acanthosis, hyperkeratosis, papillomatosis and dermis with loose collagen fibers, numerous capillaries and mild dispersed mononuclear infiltrate (**Figure 2**).

Figure 2 - Different views of histopathology.



Subtitle: (A) (B) and (C): Hematoxylin and eosin.
Source: Assis GG, et al., 2022.

The clinical and histopathological findings of this patient are compatible with soft fibroma of the nipple. The patient evolved with an inconspicuous scar after two years of follow-up (**Figure 3**). The patient was satisfied with the aesthetic outcome.

Figure 3 - Postoperative.

Subtitle: (A) Immediate postoperative. (B) Follow-up after two years.
Source: Assis GG, et al., 2022.

DISCUSSION

Soft fibroma is a benign fibrohistiocytic proliferation that usually presents itself as a pedunculated, soft, skin-colored papule or nodule, affecting mainly the neck, axilla and groin (WALLS SP, et al., 2022). It may be associated with obesity, dyslipidemia, diabetes mellitus and hypothyroidism (AKPINAR F and DERVIS E, 2012).

Cases of soft fibroma of the nipple are rare. The first scientific description of soft fibroma of the nipple happened in 1993 in Japan with the case report of a 33-year-old woman (HIGAKY Y, et al., 1993). To date, 18 cases of soft fibroma of the nipple have been described (WALLS SP, et al., 2022; HIGAKY Y, et al., 1993; COSTA AL, et al., 2009; SEO BF and JUNG S, 2012; SHABAAN AM, et al., 2013; BELLI AK, et al., 2013; VAGHOLKAR K, et al., 2013; NAKAI N, et al., 2013; YOU HS, et al., 2015; KIM S, et al., 2018; IM B, et al., 2018; ARORA BK, 2019; YUCUMÁ D, et al., 2020; VERMA SB, 2022; HUGHES EJ, et al., 2022; KIM JG, et al., 2018)

Soft fibroma affects men and women equally, a significant sex preference could not be found (BANIK R and LUBACH B, 1987). However, almost all reported cases of soft fibroma of the nipple are in female patients (WALLS SP, et al., 2022; HIGAKY Y, et al., 1993; COSTA AL, et al., 2009; SEO BF and JUNG S, 2012; SHABAAN AM, et al., 2013; BELLI AK, et al., 2013; VAGHOLKAR K, et al., 2013; NAKAI N, et al., 2013; YOU HS, et al., 2015; KIM S, et al., 2018; IM B, et al., 2018; ARORA BK, 2019; YUCUMÁ D, et al., 2020; VERMA SB, 2022; HUGHES EJ, et al., 2022). Only one male case was published (KIM JG, et al., 2018).

The risk of getting soft fibroma increases with age. They usually begin in the second decade, with an increase in frequency up to the fifth decade; older than this age range, there is no further growth (BANIK R and LUBACH B, 1987). The age of the patients with soft fibroma of the nipple varies from two to 62 years, with most of them older than 22 years. Two reported cases occurred in pediatric patients (COSTA AL, et al., 2019; HUGHES EJ, et al., 2022). The lesion of our patient appeared at the age of six, which is earlier than it is generally reported.

The dimensions of the soft fibroma usually vary between 2 and 10 mm in diameter. Soft fibroma rarely grows more than 5 cm. Most of the reported cases of soft fibroma of the nipple were smaller than 5 cm. The size of the reported cases varied from 0.4 cm to 12 cm. The largest soft nipple fibroma described so far had dimensions of 12 x 7 cm with three years of growth in a 40-year-old female (ARORA BK, 2019).

Most patients did not have any symptoms when seeking medical care; two cases presented blisters below the soft fibroma region possibly caused by local friction (HIGAKY Y, et al., 1993; NAKAI N, et al., 2013); one patient arrived for initial care due to rupture and bleeding from her nipple lesion, which was also correlated with the possibility of trauma (IM B, et al, 2018); other patient reported that the interest in treatment existed after her marriage and that she was looking for aesthetic improvement (VERMA SB, 2022). The benignity of this type of cutaneous proliferation can be reinforced by the large proportion of asymptomatic cases, with complications being more related to socio-environmental factors such as local trauma and personal acceptance.

Soft fibroma may be associated with obesity, dyslipidemia, diabetes *mellitus* and hypothyroidism. Our patient was healthy with no comorbidities (AKPINAR F and DERVIS E, 2012).

The histopathology of soft fibroma reveals, in most cases, acanthosis, hyperkeratosis, papillomatosis and hypocellular stroma with loose collagen fibers, dilated capillaries and absence of skin adnexal structures (**Board 1**).

Board 1 - Data collection of previously described cases of soft fibroma of the nipple.

YEAR	AUTHORS	AGE	SEX	LARGEST AXIS DIMENSION	HISTOPATHOLOGICAL FINDINGS
1993	HIGAKI Y, et al.	33	F	0.6 cm	Loose fibrous tissues with dilated vessels. A subepidermal blister overlying the tumor. The lower part of the thinned epidermis which formed the roof of the blister showed degeneration. Presence of erythrocytes around the capillaries. Cell infiltration composed mainly of lymphocytes and neutrophils.
2009	COSTA AL, et al.	2	F	< 1 cm	Hyperplastic epidermis surrounding a core of fibrovascular tissue with dense collagen fibers without obvious sebaceous glands or sweat glands. No smooth muscle bundles or lactiferous ducts were found.
2012	SEO BF and JUNG S	43	F	1 cm	Proliferation of fibroblasts between abundant collagen bundles and scattered vessels was revealed, with a lining of epidermis.
2013	VAGHOLKAR K, et al.	23	F	-	Spindle shaped cells with benign cellular characteristics and thin walled vasculature.
2013	SHAABAN AM, et al.	45	F	4 cm	Hypocellular lesion. Proliferation in the stroma of rather bland spindle and stellate-shaped cells.
2013	BELLI AK, et al.	38	F	5.2 cm	Lesion had a core of fibrovascular tissue with dense collagen fibers surrounded by a hyperplastic epidermis.
2013	NAKAI N, et al.	51	F	0.4 cm	Loose fibrous tissues with congested dilated capillaries. A subepidermal blister, wide-spread and complete degeneration of epidermal cells, many neutrophils in the blister close to the fibrous tissues, and moderate numbers of erythrocytes in the fibrous tissues.
2013	NAKAI N, et al.	29	F	0.4 cm	The papule consisted of loose fibrous tissues with congested dilated capillaries. Intraepidermal blisters containing moderate numbers of neutrophils, focal epidermal necrosis and degeneration of epidermal cells, and many extravasated erythrocytes in the fibrous tissues.
2015	YOU HS, et al.	51	F	2.4 cm	Papillomatosis, hyperkeratosis, and regular acanthosis of the epidermis with fibrocollagenous tissue in the dermis.

YEAR	AUTHORS	AGE	SEX	LARGEST AXIS DIMENSION	HISTOPATHOLOGICAL FINDINGS
2018	KIM JG, et al.	34	M	-	Mild papillomatosis and acanthosis with increased collagen fibers and dilated capillaries.
2018	IM B, et al.	51	F	0.5 cm	Polypoid lesion with mild acanthosis of an epidermal lining and loosely arranged fibrocollagenous stroma.
2018	IM B, et al.	32	F	1.7 cm	Furrowed mass with loosely arranged collagen fibers with mild hyperkeratosis, papillomatosis, and acanthosis of the epidermis.
2018	KIM S, et al.	41	F	2.2 cm	Hyperplastic epidermis surrounding a core of fibrovascular tissue with dense collagen fibers.
2019	ARORA BK	40	F	12 cm	Stratified epithelium with underlying subepithelial composed of loose collagenous tissue.
2020	YUCUMÁ D, et al.	22	F	3.5 cm	Polypoidal lesion covered by squamous epithelium with fibrocollagenous hypocellular stroma and small vessels in the stroma.
2022	WALLS SP, et al.	62	F	3.1 cm	Extensive fibrovascular tissue with dense collagen fibers surrounded by a hyperplastic epidermis.
2022	VERMA SB	23	F	6 cm	Areas of atrophic epidermis and acanthosis. Keratin whorls, strands, and other debris in the interlobular. Core of the polyp is filled with longitudinally oriented collagen bundles and elongated blood vessels in the center.
2022	HUGHES EJ, et al.	14	F	1 cm	Polypoid lesion covered by papillomatous epidermis, collagenous stroma with absence of skin adnexal structures, smooth muscle bundles of nipple seen at base only.

Subtitle: F: Female. M: Male.
Source: Assis GG, et al., 2022; extracted data from Higaky Y, et al., 1993; Costa AL, et al., 2019; Seo BF and Jung S, 2012; Vagholkar K, et al., 2013; Shabaan AM, et al., 2013; Belli AK, et al., 2013; Nakai N, et al., 2013; You HS, et al., 2015; Kim JG, et al., 2018; Im B, et al., 2018; Kim S, et al., 2018; Arora BK, 2019; YucumÁ D, et al., 2020; Walls SP, et al., 2022; Verma SB, 2022; Hughes EJ, et al., 2022

The epidermis of the majority of the cases of soft fibroma of the nipple was intact and hyperplastic with regular acanthosis and papillomatosis. In some cases, however, there were areas of epidermal atrophy, focal or wide-spread epidermal necrosis, degeneration of epidermal cells and subepidermal or intraepidermal blisters containing neutrophils

The dermis of soft fibroma of the nipple usually shows loosely arranged and hypocellular fibrocollagenous stroma with spindle shaped cells and dilated thin-walled scattered vessels. Presence of erythrocytes and even inflammatory infiltration composed mainly of lymphocytes and neutrophils is occasionally reported. Sebaceous glands, sweat glands, smooth muscle bundles and lactiferous ducts are generally absent. Smooth muscle bundles of nipple may be seen at base of the lesion.

The differential diagnosis of soft fibroma of the nipple includes neurofibroma, adenoma, papilloma, epidermal nevus and common wart (HUGHES EJ, et al., 2022).

Neurofibroma is a rare type of benign tumor of neuroectodermal origin and it is most common on the trunk, extremities, head and neck. It may present multiple or isolated lesions and is often associated with neurofibromatosis. Clinically, it presents itself as a dome-shaped or pedunculated papule, normochromic, which has the pathognomonic sign of “buttonhole”, which is its sinking during palpation (NASRI S, et al., 2021). In the histopathological evaluation, they are extraneural dermal tumors, slightly eosinophilic and circumscribed without a capsule. The cells are thin spindle-shaped with elongated and wavy nuclei that are regularly separated by fine collagen fibers with a wavy appearance. The stroma is myxoid and mast cells may be present (PRIETO VG and REED RJ, 2015).

Nipple adenoma is a rare benign neoplasm of the greater lactiferous ducts, it presents itself as a palpable nodule, which may have ulcerations and papillary discharge (ANSARI MS, et al., 2020). In histopathology, it has dilated tubular structures at the dermo-epidermal junction and throughout the dermis, which are lined by a peripheral layer of cuboid cells and a columnar layer in the ductal lumen region, where occasionally there is decapitation secretion (BEER T, et al., 2015).

Nipple papillomatosis is characterized by a benign growth of numerous papillomas arising from the lactiferous duct nipple epithelium also known as florid nipple papillomatosis. It mainly occurs in women between the ages of 35 and 55, but it can occur at any age. It occasionally affects men. Nipple papillomatosis presents itself as a palpable mass or swelling on the surface of the nipple (DENNIS MA, et al., 2000). Histologically, it shows nipple papillomatosis and proliferating ductal structures that extend to the sinus stroma, lined by a double layer of epithelium. Other histological features include the presence of cobble rim filled cysts and small apical cell snouts. (ELDER DE, et al., 2007)

Epidermal nevus is another possible differential diagnosis. Although epidermal nevus typically appear as plaques consisting of closely set or coalescing, skin-colored or brown, verrucous papules; they can also have unusual presentations, such as a pedunculated lesion. Histologically, it is characterized by hyperkeratosis with broad papillomatosis and acanthosis. Sometimes epidermolytic hyperkeratosis is present within the lesion. (SIM JH, et al., 2010)

Common wart is another important diagnosis to add to the list of possibilities. Warts are benign epidermal proliferation that are caused by human papillomaviruses (HPV). Common wart is usually associated with 1, 2 and 4 (XU X, et al., 2015). Warts can be found isolated or in groups and appear as a firm and painless hyperkeratotic papule with pinpoint blackdots most common on fingers, dorsal hands, elbows and knees. Histological analysis shows acanthosis, papillomatosis, hyperkeratosis, vertical layers of parakeratotic cells and foci of clustered keratohyalin granules. (XU X, et al., 2015).

Surgical excision is indicated for soft fibroma when there are cosmetic complaints, pain, bleeding, pruritus, ulcers, progressive growth or unclear diagnosis. Tangential excision (shaving) with scalpel or scissors is the treatment of choice for soft fibrome. Electrosurgery is an option in lesions with thin rods, but may be associated with increased risk of post-inflammatory hyperpigmentation. Small lesions of soft fibrome may be treated surgically without local anesthesia.

All reported cases of soft fibroma of the nipple were successfully treated with surgery. However, none of the papers showed an analysis of the surgical follow-up. In this work, though, such analysis is presented with excellent cosmetic outcomes after a two years follow-up.

Soft fibroma of the nipple is a benign proliferation that can cause significant psychological distress, embarrassment in the presence of partners and difficulty in self-acceptance. Ambulatory surgery is a safe and effective treatment that is usually associated with great cosmetic outcome and improvement of the patient's quality of life. Histological analysis of soft fibroma is essential especially in atypical locations such as the nipple.

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