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Estrogen receptor positive, progesterone receptor negative, leiomyoma of the areola of a male patient

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Abstract

Leiomyoma of the nipple and areola is a rare subtype of genital leiomyoma. The etiology of the tumor is not well understood. However, sex hormones like estrogen and progesterone have been implicated in the tumorigenesis. Hereby, we report a 47-year-old man with an estrogen receptor positive, progesterone receptor negative, leiomyoma of the areola.

Keywords: areola, estrogen, leiomyoma, male, progesterone, receptor

Introduction

Cutaneous leiomyomas are rare, benign, smooth muscle tumors, which are classified into three subtypes including piloleiomyomas, angioleiomyomas, and genital leiomyomas. These are derived from scrotal, vulvar, nipple, or areola smooth muscle [1, 2]. Leiomyoma of the nipple and areola is the least common subtype of genital leiomyoma [1, 3]. Its exact cause still remains unknown. However, trauma and hormonal drugs like oral contraceptives and tamoxifen have been implicated in the development of nipple leiomyoma [1, 2, 4]. Recently, it has been proposed that sex hormones like estrogen and progesterone may play an important role in the tumorigenesis [1, 2].

Case Synopsis

A 47-year-old man presented with an asymptomatic

lump on his left breast for further clinical evaluation. The patient stated that the lesion had been present for two years and that the size of the lesion increased slowly within this period. The patient did not recall any skin trauma or injury to the affected area. The past medical history included a left suboccipital craniectomy in the United Kingdom seven years prior. However, the patient could not state the underlying intracranial disorder. Therefore, a cranial magnetic resonance imaging (MRI) was performed. The cranial MRI of the patient showed a postoperative cystic encephalomalacia in the left cerebellum and retrocerebellar cystic lesion measuring 68x30mm in size (arachnoid cyst). Physical examination of the patient revealed a skin colored, well defined, firm nodule measuring 6mm in



Figure 1. The tumor presenting as a skin-colored nodule on the left areola (black arrow).

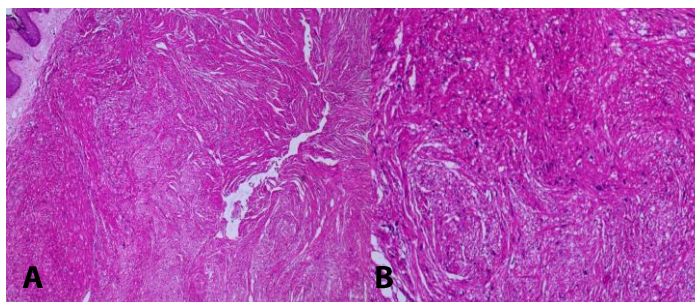


Figure 2. A) Intersecting bundles of smooth muscle fibers in the dermis. H&E, 40x. **B)** There were no nuclear atypia, mitotic activity or necrosis. H&E 100x.

diameter. The lesion was located on the left areola, at the medial side of the nipple (**Figure 1**). Ultrasonography showed a well circumscribed, hypoechoic, subcutaneous 6x6mm mass with posterior acoustic shadowing on the areola of the left breast.

In order to reach a definitive diagnosis, an excisional biopsy was performed. The histopathological evaluation of the specimen revealed an ill-defined tumor in the dermis composed of intersecting bundles of smooth muscle fibers. Nuclear atypia, mitotic activity, and necrosis were not observed (**Figure 2**). The immunohistochemical analysis of the tumor showed smooth muscle actin expression. Moreover, Masson trichrome stain confirmed the positivity for smooth muscle fibers. In addition, the spindle cells were positive for estrogen receptor (**Figure 3**). The estrogen receptor positivity was observed in 60% of the tumor nuclei. However, the spindle cells were negative for progesterone receptors.

The diagnosis of leiomyoma of the areola was made based on clinical and histological findings.

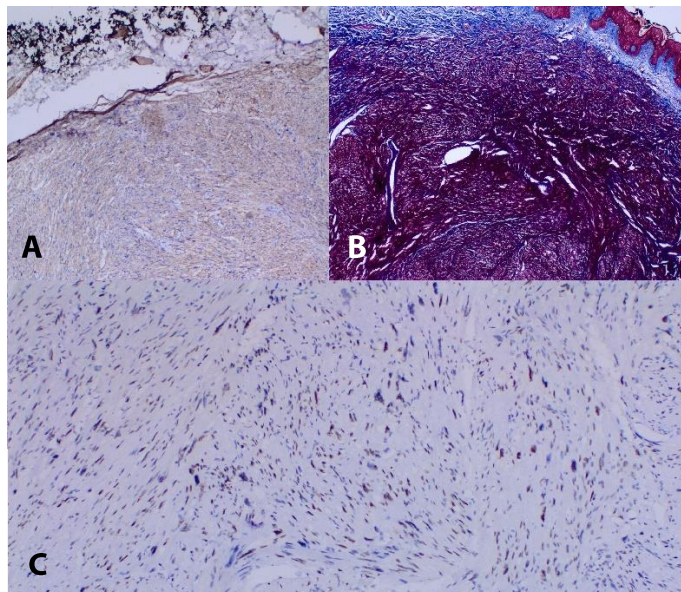


Figure 3. A) The spindle cells expressed smooth muscle actin immunohistochemically, 40x. **B)** Red staining of the cytoplasm on Masson's trichrome stain was observed (Masson's trichrome, 40x). **C)** Positivity for estrogen receptors in 60% of tumor nuclei (Estrogen receptor immunostaining, 100x).

Laboratory tests including urinalysis, thyroid-stimulating hormone, iron, folate, vitamin B12, carcinoembryonic antigen, free prostate specific antigen, cancer antigens (CA) 15-3, CA 19-9, and CA 125 were all within normal limits. Hepatitis B surface antigen, hepatitis C antibody and anti-human immunodeficiency virus antibody were all negative. Complete blood count and biochemistry panel were within normal limits except slightly elevated levels of serum eosinophil percentage 6.9% (range: 0.5-6), total bilirubin 1.53 mg/dL (range: 0.2-1.2), and direct bilirubin 0.45 mg/dL (range: 0-0.3). The abdominal ultrasound examination revealed mild hepatic steatosis.

Table 1. The estrogen, progesterone and androgen receptor data for leiomyomas of the nipple and scrotum.

Study (Year)	Patients/ Tumor	Receptors		
		Estrogen	Progesterone	Androgen
Ramos Rodriguez et al. (2017) [3]	One male/ nipple	+	+	
Nakamura et al. (2012), [6] Suárez-Peñaranda et al. (2007), [7]	One male/ nipple	-	-	
	One male/ areola	-	-	-
	Two males/ scrotum	-	-	+
Chaudhary et al. (2004), [5]	Two females/ nipple	+	+	

Case Discussion

Ramos Rodriguez et al. reported estrogen and progesterone receptor-positive bilateral nipple leiomyoma in a man who had a medical history of prostate cancer, bilateral gynecomastia, and erectile dysfunction. They suggested the possible role of hormonal dysregulation in the development of bilateral nipple leiomyomas [3]. Chaudhary et al. investigated the estrogen and progesterone receptor status of the specimens obtained from two female patients with nipple leiomyoma and ten patients with breast carcinoma. Normal subareolar muscle fibers of patients treated with mastectomy for breast carcinoma showed positive estrogen and progesterone receptor on immunohistochemistry. Similarly, the two cases with nipple leiomyoma showed immunoreactivity for estrogen and progesterone receptors [5].

In contrast, Nakamura et al. [6] reported an estrogen and progesterone receptor-negative leiomyoma on the left nipple of a man who had spironolactone-

induced gynecomastia. Nakamura et al. expected the tumor to be estrogen and progesterone receptor positive as the patient had gynecomastia. However, sex hormone receptor stains were negative [6]. Suárez-Peñaranda et al. examined the immunohistochemical findings of three cases of male genital leiomyomas (two in the scrotum and one in the areola). Estrogen and progesterone receptors were negative in all cases whereas androgen receptors were expressed in the scrotal tumors. Moreover, androgen receptor immunostaining was also negative in the nipple [7]. The estrogen, progesterone, and androgen receptor data for leiomyoma of the nipple is summarized in **Table 1**.

Conclusion

These results mentioned above, made the role of sex hormones suspicious in the etiology of leiomyoma. To the best of our knowledge, this is the first report describing an estrogen receptor-positive and progesterone receptor-negative leiomyoma of a man's areola.

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