The presence of extra nipples (polythelia) and breasts (polymastia) is not uncommon. Although often found within the milk line, running from the axilla to the groin or pubic region, there are descriptions in which they occur elsewhere, in atypical localizations such as vulva and torso. The authors report a case of ectopic breast fibroadenoma in the pubic region, which was surgically excised.

INTRODUCTION

The presence of extra nipples (polythelia) and breasts (polymastia) is not uncommon. The genesis of this breast tissue lies in quiescent primordial cells that, although occur more often along “Hughes lines” (mammary lines or “milk lines”) supernumerary, it can also have an ectopic location. There are rare, unusual locations, usually referred as “mammae erraticae”: buttock, back of neck, face, flank, upper arm, hip, shoulders and midline of the back and chest. It can occur in males, but it is far much more common in females. Wherever it occurs, this ectopic breast tissue can undergo the same pathologic changes as normally positioned breasts, such as fibroadenomas and carcinomas.

Although polythelia is congenital in origin, polymastia and ectopic mammary tissue may not appear until enhanced by sex hormones during specific periods of female life, such as puberty, pregnancy and puerperium (if breast feeding the child). This ectopic breast tissue can undergo the same pathologic changes as normally positioned breasts. Cases of ectopic breasts with benign cystic changes, benign tumors (adenomas and fibroadenomas) and carcinomas are documented.

The ectopic tissue can be surgically excised if symptomatic or if it represents a cosmetic problem. In cases of carcinoma, wide surgical excisions are recommended. The clinical workup and appropriate therapies need to be finetuned in cases of ectopic breast tissue.

In this article, we present a case report of an ectopic breast fibroadenoma which was at the pubic region.

CASE REPORT

A 37 years old female who had an elongated mass, 10 x 5 centimeters of diameter, smooth and elastic in consistency, localized in the pubic region. This mass had been noticed for the first time about 6 years ago and had been growing since then. The only additional complaint was local pruritus.

She had a female phenotype. The pubic hair distribution and secondary sexual characters were normal, so was the gynecological exam.

She was a schizophrenic patient under neuroleptic medication (flufenazine and clozapine) and had smoking habits (20 - 30 cigarettes per day).

In her gynecological history, our patient had menarche at 12 years old, regular menstrual cycles of 30 days cycle and 6 days duration. Regarding her obstetric history, she had two voluntary pregnancy interruptions, free of intercurrences, and underwent bilateral tubal ligation at 21 years old.

Her mother revealed that there was no one else in the family with similar masses nor any mass suspicious of being supernumerary breast tissue.

Several complementary examinations were done: analytic
blood tests, including hormone determinations, which had all normal results, and ultrasonographic exam of the urinary system, which was also normal.

The excision of the lesion was done under general anesthesia. There were no complications in the post-operative period. The anatomical-pathological diagnosis was of breast fibroadenoma.

**Figure 1**
Figure 1: Pubic mass with pubic hair around it

**Figure 3**
Figure 3: Pubic mass after trichotomy

**Figure 2**
Figure 2: Pubic mass relation with external genitalia

**Figure 4**
Figure 4: Excision of pubic mass
Ectopic Breast Fibroadenoma

**Figure 5**
Figure 5: Mammary fibroadenoma

**Figure 6**
Figure 6: Mammary fibroadenoma

**Figure 7**
Figure 7: Mammary fibroadenoma

**Figure 8**
Figure 8: Mammary fibroadenoma
DISCUSSION

Fertility goddesses of the ancients often had multiple breasts. The goddess Artemis of Ephesus, which cult was fertility and wild orgies, had a plethora of breasts arranged in neat rows on her torso.

For centuries there was the belief that polymastia was synonymous of abundant fertility and femininity.

In the 19th-century medical literature more fertility, including higher incidence of plural births, were attributed to females who had supernumerary breasts.

The full spectrum of extra parts - from nipples to fully formed breasts - is wide-ranging. Polymastia may be seen as a normally shaped female breast with nipple and areola, or as attenuated forms - breast tissue with nipple lacking areola, glandular tissue with areola but without nipple, or only ectopic breast tissue with neither areola nor nipple. Supernumerary nipples are often miniature compared to normally placed nipples.
The incidence of polymastia and polythelia is not well known and although it seems to occur in 1 - 2% of the general population, it has been reported to be as high as 6%. It is more common in females than in males.

The genesis of supernumerary breast lies in hidden, quiescent primordial tissue along the Hugh lines, also known as mammary lines or milk lines.

By the fourth - fifth weeks of normal embryonic development, two surface thickenings occur along the sides of the embryo, at an equal distance from midline, extending from the axilla to the groin or pubic region. In the subsequent 2 - 3 months, these thickenings become more pronounced as mammary ridges - the future site of breasts. Normally most of these ridges - all except two, one in each side, which will be the future breasts - regress. However, the quiescent mammary tissue can blossom in the right hormonal milieu. That is why these lesions frequently appear in the puberty, pregnancy and puerperium.

Most cases of supernumerary mammary tissue are described as masses localized within the milk line. The axilla is by far the most frequent location, and is seldom under valorized because it is mistaken for an enlarged lymph node. Actually, any mass localized along the mammary ridges should always heighten the hypothesis of mammary tissue.

Although rare, unusual locations have also been reported in the literature as “mammae erraticae”: the buttock, back of the neck, face, flank, upper arm, hip, shoulders and midline of the back and chest.

The ectopic breast tissue has the same physiology and can be the origin of the same pathology as normally positioned breasts.

There are in the literature descriptions of ectopic breasts with benign cystic changes, benign tumors (adenomas and fibroadenomas) and carcinomas. Although rare, there are case reports of mammary carcinoma of the vulva, for instance.

Fortunately this ectopic tissue often represents only a cosmetic problem. In this context, it can be surgically excised, and also whenever there are symptoms such as tenderness or local discomfort related with milk secretion. In cases of malignant mass, wide surgical excisions are recommended, with appropriate follow-up treatment.

Polythelia is linked with several abnormalities of the urinary tract. Such abnormalities include supernumerary kidneys, failure of renal formation and renal carcinoma. The presence of extra nipples in children should always heighten the clinical suspicion of possible renal and urinary tract anomalies.

Although not as frequent as the previous association, the cardiovascular - congenital heart anomalies, high blood pressure and conduction or rhythm disturbances, central nervous and gastrointestinal systems can be linked with polythelia, and so can chromosomal abnormalities and genetic syndromes, too. An association with testicular tumors has also been cited.

Although commonly polymastia and polythelia occur sporadically, familial cases have been reported, even across generations, in which siblings are affected.

Our patient had an attenuated forms of breast tissue, with neither areola nor nipple, which probably underwent a pathological process and became a fibroadenoma over a period of several years (16 years perhaps). Because she is a schizophrenic patient, no one can be sure for how long she had really had this mass. The authors believe that it had appeared in her first pregnancy and had not been noticed by her because of the small size then and the possibility of being obscured by the pubic hair. She had no urinary tract anomalies, which is consistent with this attenuated form, lacking nipple, since only polythelia is linked with that kind of anomalies.

CONCLUSIONS

Ectopic breast tissue can undergo the same pathologic changes as normally positioned breasts. Cases of ectopic breast with benign cystic changes, benign tumors (adenomas and fibroadenomas) and carcinomas are documented, which can present a challenge for both the clinician and the anatomical-pathologist in making the correct diagnosis.

Further research is needed to more deeply understand this phenomenon and its associations and to refine appropriate clinical workup in such cases.

References

Ectopic Breast Fibroadenoma

Obstet Invest. 2004; 58(3):151-4. (ISSN: 0378-7346)


Author Information

Carla Lilaia
Gynecology Resident, Gynecology Department, Hospital Distrital de Faro

Eunice Capela
Specialist in Gynecology and Obstetrics, Hospitalar Assistant, Gynecology Department, Hospital Distrital de Faro

Jesús Siles Cadilla
Specialist in Anatomo-Pathology, Hospitalar Assistant, Pathology Department, Hospital Distrital de Faro

Beatriz Cabrita
Gynecology and Obstetrics Department Director, Gynecology Department, Hospital Distrital de Faro