CASE REPORTS

A rare cause of axillary swelling

Uma causa rara de tumefação axilar

Mafalda Moreira¹, Sónia Pinto Pereira¹, Ana Sofia Marinho², Berta Bonet², Sandra Teixeira¹

ABSTRACT

Axillary swelling is a common condition in pediatric age. The most common diagnosis is lymph node swelling, but it can also be caused by soft tissue tumors, vascular lesions, or inflammation of sweat glands. In rare cases, it can be due to ectopic breast tissue (EBT).

A 14-year-old female presented with right axillary pain with one year of evolution and swelling for the past two months, gradually increasing in size, especially during menstruation. Sonography revealed EBT in both axillae, and further study showed duplication of the excretory system in both kidneys. The tissue on the right axilla was excised.

EBT should be considered in the differential diagnosis of axillary swelling in adolescents and young females. When the diagnosis is established, the presence of associated urologic anomalies should be investigated. Clinicians should be aware that EBT can undergo the same pathological changes as normally located breast tissue. Surgical removal should be considered for cosmetic and prophylactic treatment.

Keywords: axillary swelling; ectopic breast tissue; surgical excision; urologic anomaly

RESUMO

A tumefação axilar é um problema comum em idade pediátrica. O diagnóstico mais frequente é linfadenopatia, mas tumores dos tecidos moles, lesões vasculares, ou hidradenite são também diagnósticos possíveis. Em casos raros, a condição pode ser causada por tecido mamário ectópico (TME).

Uma adolescente de 14 anos foi avaliada por dor na região axilar direita durante o último ano e tumefação nos últimos dois meses, com aumento gradual de tamanho, especialmente durante a menstruação. A ecografia evidenciou TME em ambas as axilas e o estudo complementar revelou duplicação do sistema excretor renal bilateralmente. O tecido axilar direito foi excisado.

O TME deve ser considerado no diagnóstico diferencial de tumefações axilares em adolescentes e jovens adultas. Perante confirmação do diagnóstico, devem ser investigadas malformações nefro-urológicas associadas. Os clínicos devem ter presente que o TME pode estar sujeito às mesmas transformações que o tecido mamário corretamente localizado. A remoção cirúrgica deve ser equacionada por razões estéticas e profiláticas.

Palavras-chave: alteração urológica; excisão cirúrgica; tecido mamário ectópico; tumefação axilar

Department of Pediatrics, Centro Hospitalar do Tâmega e Sousa. 4560-136 Guilhufe – Penafiel, Portugal. mafalda.moreira@campus.ul.pt; sonia_1995@live.com; 71025@chts.min-saude.pt

Department of Pediatric Surgery, Centro Materno-Infantil do Norte, Centro Hospitalar Universitário do Porto. 4050-651 Porto, Portugal. anasofia.marinho.sm@gmail.com; bonet.berta@gmail.com

INTRODUCTION

Axillary swelling is a usual cause of concern among children and adolescents and their parents, frequently requiring medical observation. The most common diagnosis is lymph node swelling, caused by lymphadenitis or lymphoma. Rarely, it can also be caused by soft tissue tumors (as rhabdomyosarcoma, lipoma, or fibroma), vascular lesions (as angiodysplasia or aneurysms), or inflammation of the sweat glands,¹⁻⁵ and by ectopic breast tissue (EBT), an undervalued diagnosis. EBT usually develops as a slowly growing mass and is most commonly identified in young women, with a few cases in the literature reported in adolescents.^{1,6}

CASE DESCRIPTION

A healthy 14-year-old girl presented in consultation with pain in the right axilla with one year of evolution and swelling for the past two

months, gradually increasing in size, especially during menstruation. She had the menarche at the age of ten years and used to razorshave the axillae. The girl denied other associated symptoms and had no relevant family medical history. On examination, a 4 x 4-cm swelling without inflammatory signs, poorly marginated, freely mobile, and slightly painful on palpation was noted (Figure 1). No changes were identified in both breasts or left axilla. The initial differential diagnoses were hidradenitis and lymphadenitis. Further study through sonography of the region detected the presence of mammary tissue in both axillae (Figure 2) and allowed to establish the diagnosis of ectopic breast tissue (EBT). Given this diagnosis, an abdominal ultrasound was performed to search for associated nephron-urologic anomalies, which revealed a parenchymal septum dividing the two sinusal regions in both kidneys, compatible with duplication of the excretory systems. Due to associated pain and cosmetic reasons, excision of the ectopic tissue of the right axilla was performed, followed by macroscopic and histological confirmation of the diagnosis of EBT. The girl maintains regular clinical surveillance of the left axilla.



Figure 1 - Slight swelling in right axilla

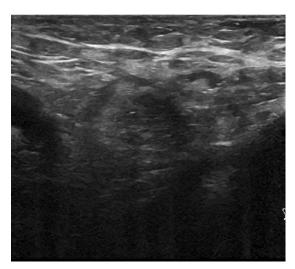


Figure 2 - Sonography revealing mammary tissue in axilla

DISCUSSION/CONCLUSIONS

EBT, or accessory breast tissue, is an uncommon entity affecting 2–6% of females and 1–3% of males, 1,4,5,7-10 more frequently diagnosed in Asian people (prevalence of 5% in the Japanese vs. 0.6% in the Caucasian population). 1,3,9,10 It presents bilaterally in two-thirds of cases and in the axillae in 70% (other possible locations are the vulva and groin). 1,3-5,7,11,13 The condition usually occurs sporadically, but a hereditary predisposition has been reported. 1,5,13,14 Because EBT is under the same hormonal influence as normal breast tissue, it usually appears during periods of hormonal stimulation, as

menstruation, pregnancy, and lactation. ^{1-3,5,8,14,15} In the present case, the condition developed in a girl younger than usually reported.

EBT results from an error during embryonic development. During the 6th week, the mammary ridges originate pairs of mammary glands, all of which involute around the third month, except one. ^{1,2,5,8,14,15} When more than one pair remains, the condition is named polymastia (presence of accessory glandular tissue) or polythelia (if supernumerary nipples are present). ^{1,4,5,13-15} In 1915, Kajava devised a classification system of polymastia that is still used nowadays, in which Class I concerns complete breast, with glandular tissue, nipple, and areola; Class II only glandular tissue and nipple, with no areola; Class III only glandular tissue and areola, with no nipple; Class IV only

glandular tissue; Class V only nipple and areola, with no glandular tissue; Class VI only the nipple (also designated as polythelia); Class VII only the areola; and Class VIII only a patch of hair.^{12,14} The present case falls into class IV.

Due to concomitant development of the mammary structure and genitourinary system, the association of EBT and urological anomalies such as supernumerary kidneys, failure of renal development, renal adenocarcinoma, hydronephrosis, and ureteric stenosis has been reported.^{3-5,8} For this reason, abdominal sonography should be performed as soon as the diagnosis is confirmed. In this case, this exam revealed a duplication of the excretory system in both kidneys.

The clinical presentation is variable. The most common symptoms are swelling of the affected region with associated pain, discomfort, and restriction of movements, which are exacerbated during menstruation and pregnancy.^{2-4,8,12,13,15} Some women also complain of cosmetic problems, which may lead to psychological disturbances, especially in adolescence.^{8,9,14} Physical examination should include the contralateral breast since the condition is bilateral in most cases.^{1,3,7}

Sonographic examination is key for diagnosis, allowing the identification of a hypoechoic septate tissue similar to orthotopic mammary tissue. In addition, proper mass characterization is also relevant and should include shape, borders, internal components, vascularity, and relationship with adjacent tissues. ^{5,10} Magnetic resonance imaging can be required in some cases and typically shows an ill-defined subcutaneous mass. ^{2,3,6,8,14}

Some authors consider surgical removal as the treatment of choice, either for esthetic reasons or prophylactically to prevent the development of potential complications, like fibroadenoma or carcinoma. 1,3-5,8,10,13 However, potential surgical complications, such as contour irregularity, seroma, or recurrence, should also be weighted in the treatment decision. 4,12 In the present case, due to pain and cosmetic concerns, the option was to remove the ectopic tissue on the right axilla. In addition, the patient was kept on regular medical surveillance and advised to perform self-examination of the left axilla.

With this case, the authors intend to emphasize the importance of considering EBT in the differential diagnosis of axillary swelling in adolescents. Despite being a rare condition, the pathological and psychological problems associated with the condition require investigation and monitoring.

TAKE-HOME MESSAGES

The diagnosis of EBT (ectopic breast tissue) should be considered in adolescents and young women presenting with axillary swelling, particularly in those with menstrual cycle changes.

Abdominal ultrasound should be performed at diagnosis due to the acknowledged association between EBT and urologic anomalies.

Similarly to normally located breast tissue, EBT can undergo pathological changes, such as inflammation, fibrosis, fibroadenoma,

and carcinoma, requiring continuous surveillance.

Some authors argue in favor of surgical removal for cosmetic and prophylactic reasons.

AUTHORSHIP

Mafalda Moreira - Conceptualization; Investigation; Project Administration; Visualization; Writing - original draft; Writing - review & editing

Sónia Pinto Pereira - Validation; Writing - review & editing
Ana Sofia Marinho - Validation; Writing - review & editing
Berta Bonet - Validation; Writing - review & editing
Sandra Teixeira. Investigation; Supervision; Validation; Writing - review & editing

REFERENCES

- Ritter L, Sorge I, Till H, Hirsch W. Accessory Breast Tissue (Mamma Aberrata) as a Rare Differential Diagnosis of Soft Tissue Swelling in the Axilla. Fortschr Röntgenstr 2013; 185: 74-5.
- Laor T, Collins MH, Emery KH, Donnely LF, Bove KE, Ballard ET. MRI appearance of accessory breast tissue: a diagnostic consideration for an axillary mass in a peripubertal or pubertal girl. Am J Roentgenol 2004; 183 (6):1779-81.
- Teixeira IP, Azzopardi C, Chowdhury R, Zsolt O, Castro M, Teh
 J. Ectopic breast tissue presenting as an enlarging abdominal mass. Radiology Case Reports 2020; 15: 733–40.
- Singal R, Mehta SK, Bala J, Zaman M, Mittal A, Gupta G, et al. A Study of Evaluation and Management of Rare Congenital Breast Diseases. Journal of Clinical and Diagnostic Research 2016; 10(10): PC18-24.
- Surd A, Mironescu A, Gocan H. Fibroadenoma in axillary supernumerary breast in a 17 years old girl: case report. Journal of Pediatric and Adolescent Gynecology 2016.
- 6. Navarro O. Soft Tissue Masses in Children. Radiologic Clinics of North Americ 2011; 49(6): 1235–59.
- 7. Burdick AE, Thomas KA, Welsh E, Powell J, Elgart GW. Axillary polymastia. Journal of the American Academy of Dermatology 2003; 49(6): 1154-6.
- 8. Kogut M, Bider M, Enk A, Hassel JC. Axillary Accessory Breast Tissue Case Report and Review of Literature. Journal der Deutschen Dermatologischen Gesellschaft 2014.
- Seifert F, Rudelius M, Ring J, Gutermuth J, Andres C. Bilateral axillary ectopic breast tissue. The Lancet 2012; 380: 835.
- 10. Gentile P, Izzo V, Cervelli V. Fibroadenoma in the Bilateral Accessory Axillary Breast. Aesth Plast Surg 2010; 34:657–9.
- 11. Guray M, Sahin AA. Benign breast diseases: classification, diagnosis and management. Oncologist 2006; 11(5):425-49.
- 12. Patel P, Ibrahim A, Zhang J, Nguyen JT, Lin SJ, Lee BT. Accessory

- Breast Tissue. Eplasty 2012; 12: ic5.
- 13. Alghamdi H, Abdelhadi M. Accessory Breasts: When to Excise? The Breast Journal 2005; 11(2): 155–7.
- Amaranathan A, Balaguruswamy K, Bhat RV, Bora MK. AN Ectopic Breast Tissue Presenting with Fibroadenoma in Axilla. Case Reports in Surgery 2013.
- Waldman RA, Finch J, Grant-Kels JM, Whitaker-Worth D. Skin diseases of the breast and nipple - Benign and malignant tumors. J Am Acad Dermatol 2019; 80: 1467-81.

CORRESPONDENCE TO

Mafalda Moreira
Department of Pediatrics
Centro Hospitalar do Tâmega e Sousa
Avenida do Hospital Padre Américo, Nº 210
4560-136 Guilhufe – Penafiel
Email: mafalda.moreira@campus.ul.pt

Received for publication: 19.08.2020 Accepted in revised form: 30.04.2021