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CASE REPORT

Unilateral Galactocele in a 2-Year-Old Boy: the Role of GATA-3

Javier Arredondo Montero[1,2] · Mónica Bronte Anaut[3] · Lidia Ayuso González[1] · Sara Hernández-Martín[1] · Yerani Ruiz de Azúa-Ciria[4]

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Abstract
Galactoceles are benign lesions formed at the expense of breast glandular tissue. Its occurrence in boys is exceptional, with isolated reports in the scientific literature. We present the case of a 2-year-old boy who debuted with a unilateral breast enlargement of 18 months of evolution, with no other associated symptoms. Initial hormonal study showed no alterations. Ultrasonography showed the presence of a homogeneous 5-cm retroareolar cyst. Surgical excision was indicated. During the procedure, abundant milk drained from the lesion. The histopathological study, supported by GATA-3 immunohistochemical staining, confirmed the diagnosis of galactocele. The range of possible differential diagnoses for unilateral or bilateral breast enlargement in boys is wide and includes neoplastic, vascular, and hormonal etiologies. Galactocele, which is an exceptional entity in this group, should be considered. Immunohistochemical techniques such as GATA-3, characteristic of the breast epithelium, can contribute to the diagnosis.

Keywords Galactocele · Pediatric · Male · Unilateral · GATA3 · Immunohistochemical · Diagnosis

We present the case of a 2-year-old boy with no previous medical history who debuted with a right breast enlargement at 7 months of age. He had no associated symptoms. Clinical examination showed normal external male genitalia (Tanner stage 1) and a right unilateral breast enlargement (Fig. 1, above). An extended hormonal study was performed at that time, showing the following values: total testosterone: 0.02 ng/mL (normal range: 2.4–8.7 ng/mL), antimullerian hormone: 23 ng/mL (normal range 0.77–14.5 ng/mL), androstenedione: < 0.3 µg/mL (normal range 0.6–3.7 µg/mL), follitropin: 0.8 IU/L (normal range 0.95–11, 95 IU/L), lutropin: 0.11 IU/L (normal range 0.57–12.07 IU/L), inhibin B: 148 pg/mL, IGF-BP3: 3390 µg/mL, choriogonadotropin: < 1.2 mU/mL, 17-OH-progesterone alpha: 0.1 µg/mL, and estradiol: < 10 pg/mL. Due to a sample processing error, serum prolactin could not be determined. An ultrasound study was requested, which showed a right retroareolar suprafascial cystic lesion, with a major axis of 5 cm (Fig. 1, bottom).

Surgical excision was indicated. An inferior periareolar approach was performed. A large cyst was identified with an adequate cleavage plane with respect to the surrounding tissues. During excision, the cyst was opened and abundant milk drained from the inside. A complete macroscopic excision of the lesion was achieved.

Histological study of the lesion showed a cyst lined by columnar secretory epithelium with dilated ductal structures (Figs. 2 and 3). The epithelium was diffusely positive for GATA-3 (Fig. 4), which favored the diagnosis of galactocele. In the rest of the paraffin-embedded sections the presence of terminal lobular units was not observed, only branching ducts of large and medium caliber. Postoperative evolution

Javier Arredondo Montero and Mónica Bronte Anaut contributed equally to this work.

All authors of the manuscript declare that it is an original contribution, not previously published.

✉ Javier Arredondo Montero
Javier.montero.arredondo@gmail.com;
jarredondom@alumni.unav.es

1 Pediatric Surgery Department, Hospital Universitario de Navarra, Calle Irunlarrea 3, 31008 Pamplona, Navarra, Spain

2 School of Medicine, University of Navarra, Pamplona, Navarra, Spain

3 Pathology Department, Hospital Universitario de Araba, Vitoria, Spain

4 Pathology Department, Hospital Universitario de Navarra, Pamplona, Navarra, Spain

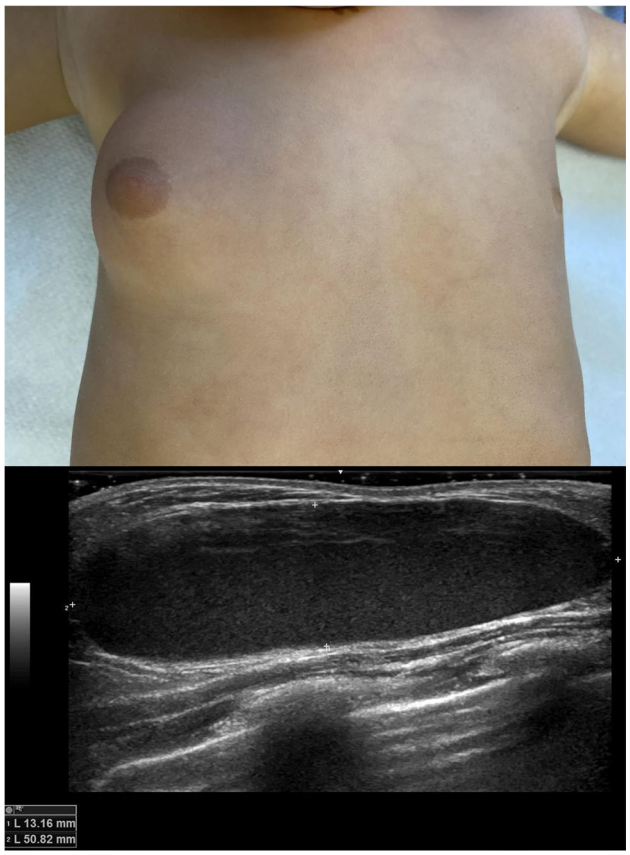

Fig. 1 Above: clinical photograph of the patient showing the enlargement of the right breast, without other findings. Below: ultrasound showing the presence of a cystic retroareolar suprafascial cystic tumor, without signs of complication

was favorable. The patient is currently asymptomatic, without relapse and under follow-up.

Clinically, the diagnosis of pediatric breast enlargement encompasses several possible diagnoses, including maternal estrogenic influence in the first weeks of life, gynecomastia, neoplasms, lymphatic malformations, and infrequent entities such as galactocele [1]. Histologically, the absence of pseudoangiomatous stromal hyperplasia (very distinctive of gynecomastia), the absence of vascular spaces or areas of proliferating cells and the absence of fibrosis would favor the diagnosis of galactocele. However, in borderline cases, immunohistochemical techniques can be used to corroborate the diagnosis. This is, to the best of our knowledge, the first case in which a specific immunohistochemical marker of breast epithelium, GATA-3, has been used to confirm the diagnosis of galactocele.

Galactoceles are benign lesions formed at the expense of mammary glandular tissue, which usually present secretion of milk. In relation to its etiology and formation, there are different hypotheses, such as prolactin stimulation, ductal obstruction, and local trauma.

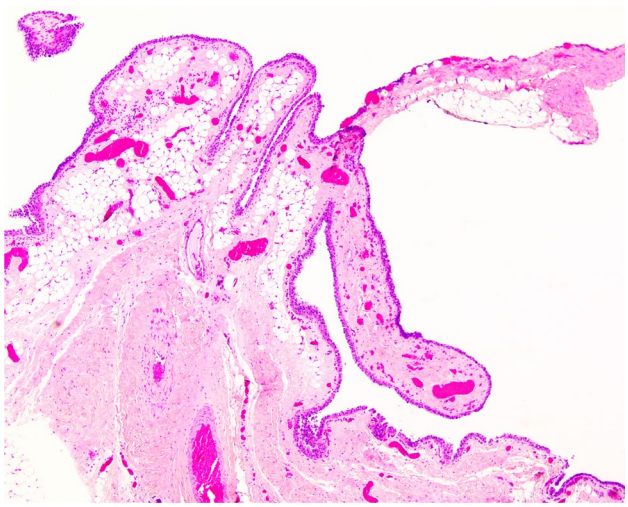

Fig. 2 Photomicrograph (Hematoxylin and Eosin, ×4). Dilated ductal structures lined by columnar secretory and apocrine-type epithelium. No lobular development is seen

The origin of galactoceles may be in the neonatal period. In relation to its natural course, they may not resorb but continue to expand due to the variation of morphological (formation of acini) and functional (secretary epithelium) maturation stages in breast development during the first 2 years of life [2].

These lesions are more frequent in girls. To the best of our knowledge, there are only few isolated reports in boys to date. They are usually asymptomatic, although patients may experience local discomfort, nipple discharge or infection. Galactoceles can be unilateral or bilateral [3]. Isolated clinical associations have been established with hypothyroidism, panhypopituitarism, and hyperprolactinemia [3–6].

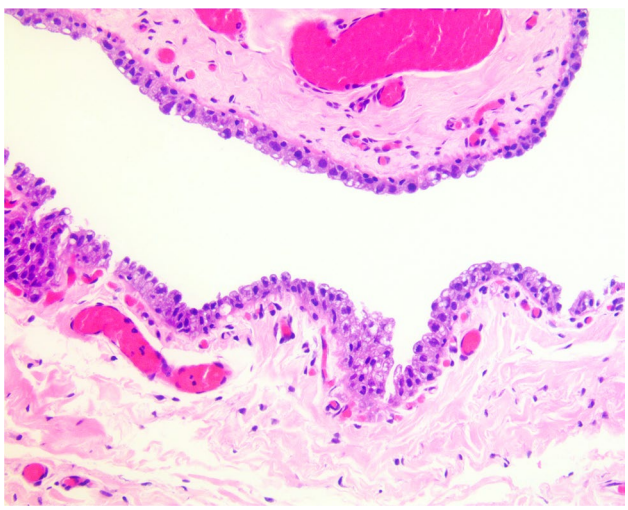

Fig. 3 Photomicrograph (Hematoxylin and Eosin, ×20). Detail of the columnar secretory and apocrine-type epithelium. No lobular development is seen

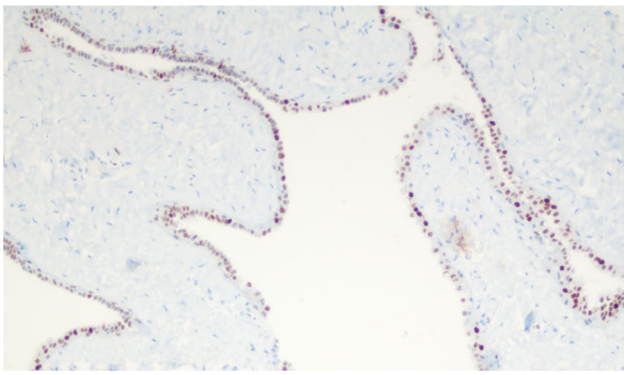

Fig. 4 Photomicrograph (GATA-3, immunohistochemical staining, ×20). The epithelium is diffusely positive for GATA-3

Ultrasonographically, galactoceles are characterized as well-demarcated suprafascial cystic lesions. However, they can sometimes present as complex masses.

Histopathological diagnosis of galactoceles is usually not necessary in certain situations, such as in adult females in the breastfeeding period. A compatible ultrasound study and milk collection by fine needle puncture are considered diagnostic, although in the case of atypical presentations, such as the one reported here, diagnostic confirmation can be achieved through histological examination. Immunohistochemical positivity for GATA-3, characteristic of mammary glandular tissue, constitutes an additional confirmatory data.

Regarding the treatment, needle aspiration is usually suggested as one of the first therapeutic lines, exeresis being reserved for refractory or recurrent cases. Nevertheless, in pediatric population, the evidence is limited, and there is no consensus on the ideal treatment. Recently, some authors have questioned whether it is necessary to treat if galactoceles are small and asymptomatic [7].

In conclusion, the range of possible differential diagnoses for unilateral or bilateral breast enlargement in pediatrics is wide, including neoplastic, vascular, and hormonal etiologies among others. The clinical course, hormonal study and radiological findings help to establish an adequate diagnostic suspicion. Galactocele in young boys is a poorly characterized entity, and evidence in the literature on the optimal diagnostic-therapeutic approach is limited. In doubtful cases, histopathological study remains a useful tool. Targeted immunohistochemical stains, such as GATA-3, can contribute to the diagnosis. The report of cases such as the one presented here could help to better understand the etiopathogenesis, diagnosis, and management of this disease.

Author Contribution Javier Arredondo Montero and Mónica Bronte Anaut: conceptualization, literature search, study design, resources, data extraction, data curation, data interpretation, writing—original draft, and writing—review and editing.

Yerani Ruiz de Azúa: resources, data curation, data interpretation, supervision, validation, and writing—review and editing.

Lidia Ayuso González and Sara Hernández-Martín: resources, supervision, validation, and writing—review and editing.

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Data availability All clinical and histopathologic material of this case is included in the present document.

Declarations

Consent to Participate Prior to the submission of this article, verbal and written informed consent was obtained from the legal guardians of the patient whose clinical photographs are included in this publication. The patients' medical records were accessed in accordance with the specific hospital regulations applicable to this type of case.

Conflict of Interest The authors declare no competing interests.

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