Joint Bone Spine 86 (2019) 263–264

Available online at
ScienceDirect
www.sciencedirect.com

Elsevier Masson France
EM|consulte
www.em-consulte.com/en

Letter to the Editor

Breast involvement in granulomatosis with polyangiitis

ARTICLE INFO

Keywords:
Granulomatosis with polyangiitis
Wegener's granulomatosis
Breast involvement

Granulomatosis with polyangiitis (GPA) or Wegener's granulomatosis is a multisystem disorder characterized by necrotizing granulomatous inflammation and vasculitis of small vessels, which can affect any organ system. The most common sites of involvement are the upper and lower respiratory tracts and kidneys. Breast involvement is very rare [1–3]. We recently diagnosed a 25-year-old man with GPA on the basis of a 2-week history of fatigue, fever, weight loss, a persistent palatal ulcer, sinonasal involvement confirmed by computed tomography, oligoarthritis affecting wrists and knees, bilateral pleural effusion, renal involvement (asymptomatic hematuria, granular casts, subnephrotic proteinuria, and hypertension, with normal serum creatinine), and vasculitic skin lesions involving the lower extremities, accompanied by focal necrosis and ulcerations. The biopsy of the edge of one of the cutaneous ulcers confirmed the presence of necrotizing vasculitis, and the proteinase 3 (PR3-ANCA) antibodies were positive at a titer of 907 (positive > 7).

On physical examination, this patient presented palpable painless masses in the right breast. Mammography showed several solid nodules, with ill-defined margins, with the largest located in the right superior external quadrant (Fig. 1A and B). Ultrasonography confirmed the presence of various hyperechoic solid lesions, some of them with anechoic areas of necrosis (BI-RADS category 3; Fig. 1C). An ultrasound-guided needle core biopsy of the largest nodule was performed, and histopathological examination revealed neutrophilic vasculitis with fibrinoid necrosis, without atypical cells, consistent with breast involvement of GPA (Fig. 1D). The patient was treated with prednisone (1 mg/kg/day) and rituximab (1,000 mg on days 1 and 15), with complete disappearance of the breast nodules in the next 2 weeks.

Breast involvement by GPA is very infrequent, with few cases described in the literature. Most cases present in women between the third and seventh decades, are unilateral, and are usually accompanied by the other characteristic systemic manifestations of the disease [1]. However, some cases have been published in which uni- or bilateral breast involvement was the presenting symptom of GPA, with the other suggestive manifestations of the disease not appearing for several months or years [1–3]. Some of these cases mimicked a breast carcinoma [1,3,4] or they presented as recurrent breast abscesses [1,5] or extensive ulcero-necrotizing lesions [6,7]. It is important to highlight that in many of these cases, ANCA were negative at the initial stages of the disease (it may take up to 5–7 years to become positive) [1–7].

In conclusion, although rare, our case and other published reports provide evidence that GPA can also affect the breast and this may be its presenting manifestation. This possibility must be taken into account in the differential diagnosis of granulomatous mastitis, necrosis of the breast, as well as of breast tumors [1–3]. It should be noted that, rarely, it can precede the classic manifestations of the disease by several months or years.

Data Availability

The authors confirm that all data underlying the findings are fully available without restriction. All relevant data are within the paper.

Ethics approval

In accordance with the guidelines of our institutional ethics committee (Clinical Research Ethics Committee of Bellvitge University Hospital-IDIBELL), formal approval for this study was not required. The local ethics committee agreed that the findings in this report were based on normal clinical practice and were therefore suitable for dissemination. This study was conducted in accordance with the principles of the Declaration of Helsinki and the International Conference for Harmonization.

Funding

None. This study is not a part of corporate sponsored research.

Contribution of authors

All authors had access to the data and played a role in writing this manuscript.

Disclosure of interest

The authors declare that they have no competing interest.

https://doi.org/10.1016/j.jbspin.2018.05.004

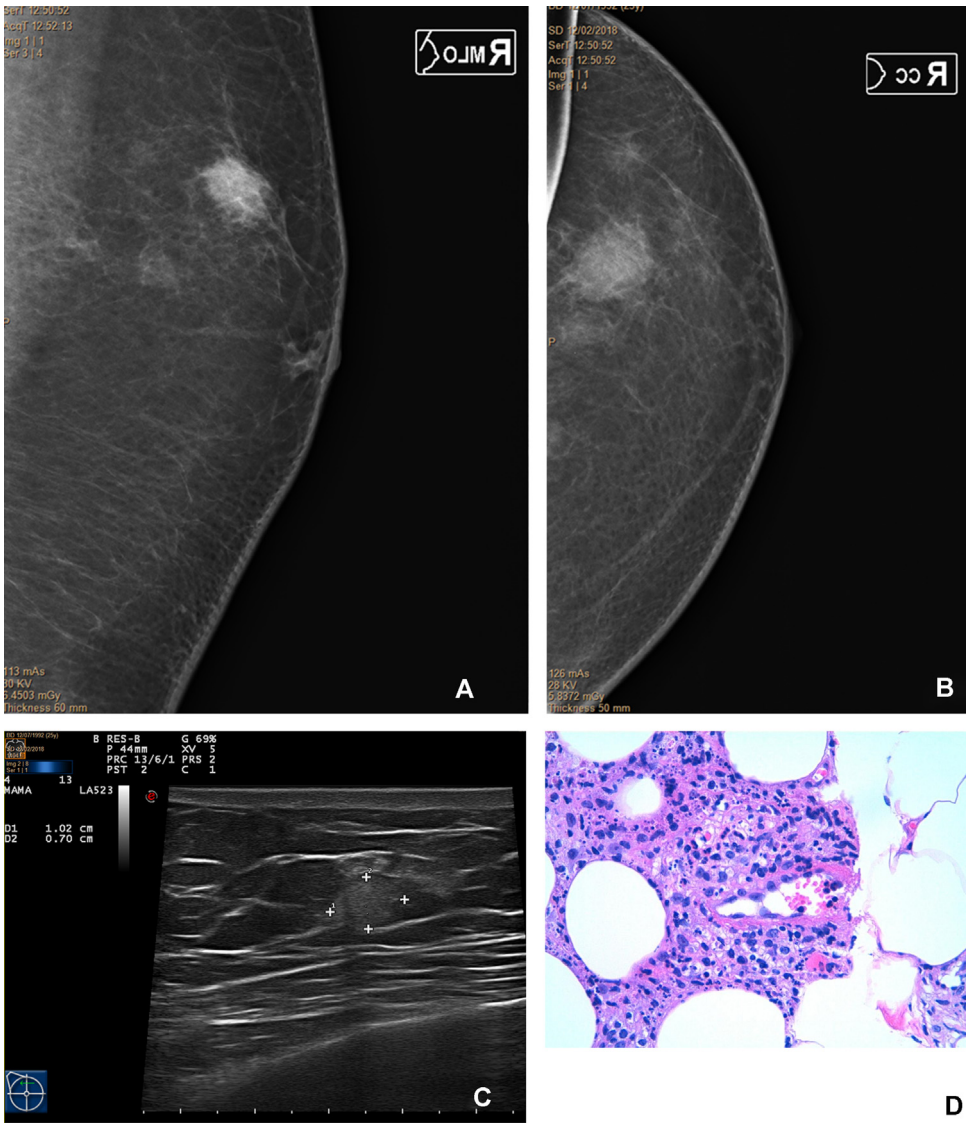

Fig. 1. Right mediolateral oblique (MLO) and craniocaudal (CC) mammograms (Fig. 1A and 1B, respectively) demonstrate several solid nodules with ill-defined margins, with the largest located in the right superior external quadrant. US image of the largest nodule (Fig. 1C) shows a hyperechoic solid lesion of 1.02 × 0.7 cm. The results of its biopsy showed changes of panniculitis, an inflammatory infiltrate composed mainly of polymorphonuclear cells, and foci of neutrophilic vasculitis with fibrinoid necrosis (Fig. 1D).

References

[1] Allende DS, Booth CN. Wegener's granulomatosis of the breast: a rare entity with daily clinical relevance. Ann Diagn Pathol 2009;13:351–7.
[2] Stappaerts I, Colpaert C, Verbraecken J, et al. Granulomatous mastitis as presenting sign of Wegener's granulomatosis. Acta Clin Belg 1999;54:207–10.
[3] Mengoli MC, Ragazzi M, Lococo F, et al. Breast granulomatosis with polyangiitis mimicking breast cancer. Pathologica 2017;109:405–7.
[4] Ene N, Bélénotti P, Benyamine A, et al. Granulomatosis with polyangiitis (previously Wegener's granulomatosis) mimicking malingering. Rev Med Interne 2014;35:540–2.
[5] Comas AG, Diana CA, Crespo CC, et al. Wegener's granulomatosis presented as recurrent breast abscess. Breast J 2010;16:82–4.
[6] Gallais Sérézal I, Jennische K. Image gallery: symmetrical necrosis of the breast as an inaugural manifestation of granulomatosis with polyangiitis (Wegener granulomatosis). Br J Dermatol 2016;175:e132.
[7] Daguzan M, Martinez-Mena C, Hermans P. A case of breast necrosis. Rev Med Brux 2012;33:112–5.

Judit Lluch [a]
Laia Montserrat Pérez-Tapia [b]
Maria del Rosario Taco-Sánchez [c]
Javier Narváez [a,*]

[a] *Departments of Rheumatology, Hospital Universitario de Bellvitge-IDIBELL, 08907 Barcelona, Spain*
[b] *Radiology Department, Hospital Universitario de Bellvitge-IDIBELL, 08907 Barcelona, Spain*
[c] *Pathology Department, Hospital Universitario de Bellvitge-IDIBELL, 08907 Barcelona, Spain*

* Corresponding author. Department of Rheumatology (Planta 10-2), Hospital Universitario de Bellvitge, Feixa Llarga, s/n, Hospitalet de Llobregat, 08907 Barcelona, Spain.
E-mail address: fjnarvaez@bellvitgehospital.cat (J. Narváez)

Accepted 16 May 2018
Available online 22 May 2018