

Clinical Letter

Chest Wall CT-Fistulography for Surgical Planning in a Patient With Chronic Granulomatous Disease

To the Director,

Chronic granulomatous disease (CGD) is a rare immunodeficiency caused by a defect in the nicotinamide adenine dinucleotide phosphate (NADPH) oxidase enzyme complex that predisposes to recurrent bacterial and fungal infections from an early age.^{1,2} Most infections are caused by bacteria (*Staphylococcus*, *Klebsiella*, *Serratia*, etc.), but fungal and mycobacterial infections may also occur. The most affected organs are the lungs, skin, lymph nodes, and the musculoskeletal system.¹

We present the case of a 31-year-old male with X-linked CGD, on chronic antibiotic treatment, with a long-standing medical history of infections (pulmonary and soft-tissue) and recurrent thoracic and abdominal fistulas, who presented complaining of inflammation of the chest wall. The patient had undergone multiple surgical interventions in the past, including a right middle lobectomy for pulmonary aspergillosis and surgical debridement for soft-tissue infections. On physical examination, two small fistulous orifices through which fluid material was draining (Fig. 1A) were found in the anterior chest wall, with doubts about the extent in depth of the inflammatory changes. A contrast-enhanced

thoracic computed tomography (CT) confirmed the existence of inflammatory changes in the anterior chest wall (Fig. 1B, C) but ruled out the presence of pulmonary consolidations or pleural fluid collections.

The decision was made to perform a CT-fistulography using iso-osmolar iodinated contrast (iodixanol 320 mg/mL, diluted to 25% with saline) through the two cutaneous fistulous openings, due to its excellent anatomical and contrast resolution. CT-fistulography revealed a subcutaneous communication between the two orifices, ruled out involvement of deeper tissues (Fig. 1D-F) and aided the thoracic and plastic surgery teams in planning the resection of the fistulous tract and the reconstruction of the anterior chest wall (Fig. 1G).

CGD is a genetically predisposed disease (X-linked or autosomal recessive) caused by a defect in the NADPH oxidase enzyme complex, affecting phagocytosis and resulting in the inability to eliminate catalase-positive pathogens such as *Staphylococcus*, *Mycobacteria*, *Serratia*, *Klebsiella*, *Pseudomonas*, and fungi. Consequently, recurrent infections lead to the formation of inflammatory granulomas. Pulmonary involvement is the most frequent manifestation, including pneumonia or lung abscesses that can extend to the chest wall. It can also manifest as lymphadenopathy, liver abscesses, soft tissue infections, osteomyelitis, brain abscesses, gastrointestinal infections, and organomegaly.^{1,2} Occasionally, thoracic surgery is necessary to treat aggressive pulmonary infections,

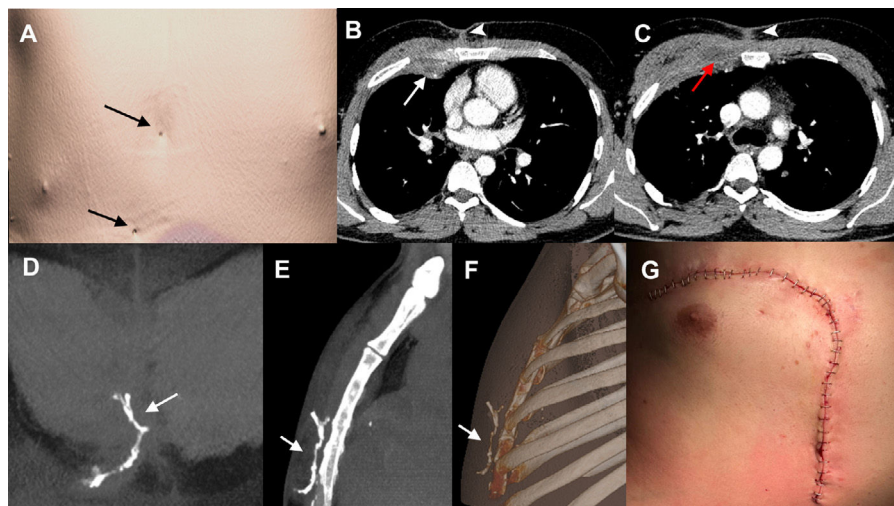


Fig. 1. (A) 3D CT reconstruction image of the patient's body surface. Two cutaneous openings are clearly visible in the anterior thoracic wall (arrows). (B, C) Axial contrast-enhanced thoracic CT images (mediastinal window) at two different levels show inflammatory changes in the anterior chest wall: a collection in the right anterior pectoral muscles (red arrow), a small extrapleural collection adjacent to the right internal mammary vessels (white arrow), and several linear fistulous tracts extending toward the skin surface (arrowheads). (D-F) Thoracic CT reconstruction images (D, coronal maximum intensity projection [MIP]; E, sagittal MIP; F, sagittal 3D) after the administration of diluted iodinated contrast through two skin fistulous openings on the anterior chest wall demonstrate communication between the two openings and the absence of deep soft tissue extension (arrows). (G) Post-surgical image of the anterior chest wall after the resection of the fistulous tract and the utilization of a vascularized muscle flap.

<https://doi.org/10.1016/j.arbres.2024.09.013>

0300-2896/© 2024 SEPAR. Published by Elsevier España, S.L.U. All rights are reserved, including those for text and data mining, AI training, and similar technologies.

which are associated with greater complications and poorer prognosis.³

CGD patients require prolonged antibiotic treatments and prophylactic antimicrobial therapy. Hematopoietic stem cell transplantation is the curative treatment for this disease, which has improved the prognosis of these patients.^{1,2}

CT-fistulography is a technique used for the diagnosis of abdominal wall and perianal fistulas in some patients and, to our knowledge, has not been used in the chest wall.⁴ In CT-fistulography, acquisition of isovolumetric data sets after the instillation of contrast into the fistula delineates the tract and its components, allows the acquisition of images with great anatomical detail in different spatial planes, and can help surgeons plan the surgical intervention.

We believe that this case is interesting because CT-fistulography ruled out communication between the cutaneous-subcutaneous inflammatory process and the extrapleural fat, pleura, and lung parenchyma. This facilitated appropriate therapeutic planning by demonstrating a communication between the two skin fistulous openings. Meticulous surgical planning is essential in these patients, given their higher risk of complications. We have found no previous descriptions in the scientific literature of CT-fistulography of the chest wall.

Artificial Intelligence Involvement

None of the material has been partially or totally produced with the help of any artificial intelligence tool.

Funding of the Study

This study did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflicts of Interest

The authors declare not to have any conflicts of interest that may be considered to influence directly or indirectly the content of the manuscript.

References

1. Lee M, Lee MS, Lee JS, Ko SY, Jeong SY. Spectrum of imaging findings of chronic granulomatous disease: a single center experience. *Diagn Interv Radiol.* 2017;23:472-7.
2. Yu H-H, Yang Y-H, Chiang B-L. Chronic granulomatous disease: a comprehensive review. *Clin Rev Allergy Immunol.* 2021;61:101-13.
3. Feingold PL, Quadri HS, Steinberg SM, Malech HL, Gallin JI, Zerbe CS, et al. Thoracic surgery in chronic granulomatous disease: a 25-year single-institution experience. *J Clin Immunol.* 2016;36:677-83.
4. Tonolini M, Magistrelli P. Enterocutaneous fistulas: a primer for radiologists with emphasis on CT and MRI. *Insights Imaging.* 2017;8:537-48.

Abel González-Huete^{a,*}, Luis Gorospe-Sarasúa^a,
Gemma María Muñoz-Molina^b

^a *Department of Radiology, Hospital Universitario Ramón y Cajal, Madrid, Spain*

^b *Department of Thoracic Surgery, Hospital Universitario Ramón y Cajal, Madrid, Spain*

* Corresponding author.

E-mail address: abelgonzalezhuete@gmail.com
(A. González-Huete).