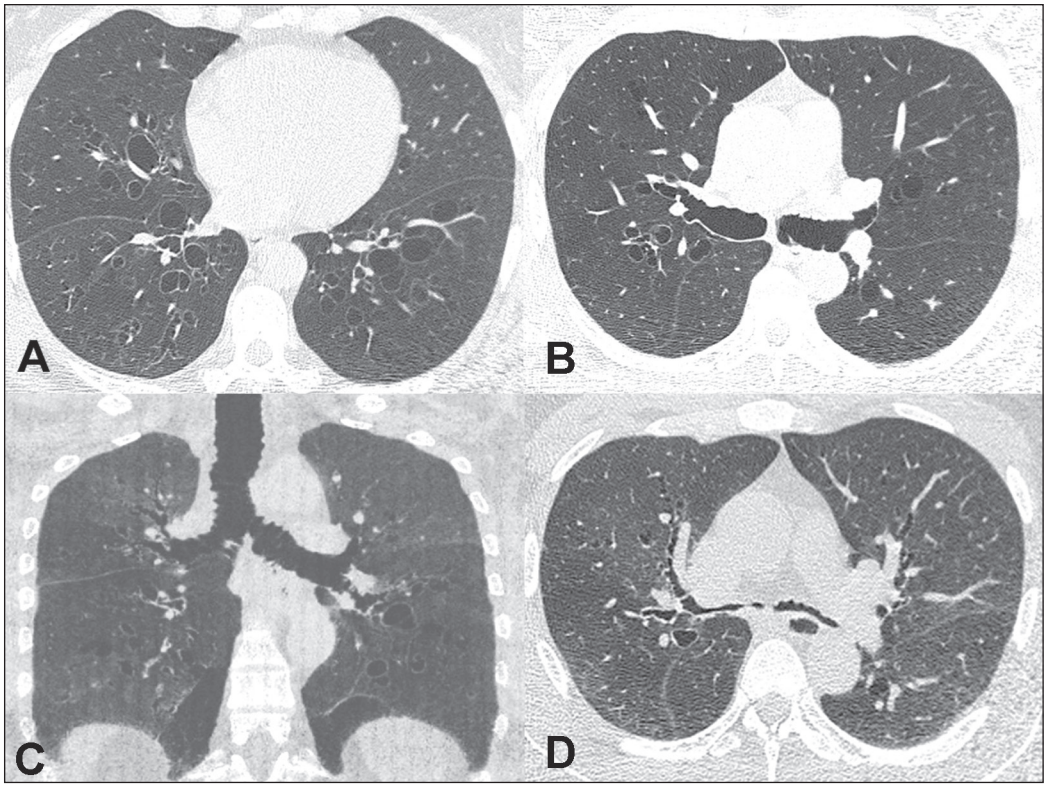

Figure 1. A,B: CT scan of the chest, acquired during inspiration, showing bilateral bronchiectasis, together with marked dilatation of the main bronchi. **C:** Coronal reconstruction showing, in addition to the bronchiectasis, dilatation of the main bronchi and the trachea. **D:** Slice acquired during expiration, showing near-total collapse of the bronchial tree.

expiration[7,8]. The treatment is generally supportive, including respiratory therapy, appropriate antibiotic therapy for the recurrent infections, and smoking cessation[9,10].

In conclusion, Mounier-Kuhn syndrome should be considered in patients who present with bronchiectasis accompanied by abnormal dilatation of the trachea and the main bronchi. The preferred diagnostic imaging method is CT of the chest.

REFERENCES

1. Togni Filho PH, Casagrande JLM, Lederman HM. Utility of the inspiratory phase in high-resolution computed tomography evaluations of pediatric patients with bronchiolitis obliterans after allogeneic bone marrow transplant: reducing patient radiation exposure. Radiol Bras. 2017;50:90–6.
2. Duarte ML, Brito MCB, Traple FAL, et al. Plasmacytoma of the trachea: a surprising diagnosis. Radiol Bras. 2017;50:66–7.
3. Torres PPTS, Rabahi M, Pinto AS, et al. Primary tracheobronchial amyloidosis. Radiol Bras. 2017;50:267–8.
4. Brito MCB, Ota MK, Leitão Filho FSS, et al. Radiologist agreement on the quantification of bronchiectasis by high-resolution computed tomography Radiol Bras. 2017;50:26–31.
5. Woodring JH, Howard RS 2nd, Rehm SR. Congenital tracheobronchomegaly (Mounier-Kuhn syndrome): a report of 10 cases and review of the literature. J Thorac Imaging. 1991;6:1–10.
6. Menon B, Aggarwal B, Iqbal A. Mounier-Kuhn syndrome: report of 8 cases of tracheobronchomegaly with associated complications. South Med J. 2008;101:83–7.
7. Shin MS, Jackson RM, Ho KJ. Tracheobronchomegaly (Mounier-Kuhn syndrome): CT diagnosis. AJR Am J Roentgenol. 1988;150:777–9.
8. Marchiori E, Sousa AS Jr, Zanetti G, et al. Mounier-Kuhn syndrome: the role of bronchiectasis in clinical presentation. Ann Thorac Med. 2012;7:51.
9. Collard P, Freitag L, Reynaert MS, et al. Respiratory failure due to tracheobronchomalacia. Thorax. 1996;51:224–6.
10. Falconer M, Collins DR, Feeney J, et al. Mounier-Kuhn syndrome in an older patient. Age Ageing. 2008;37:115–6.

Rômulo Florêncio Tristão Santos[1,a], Tiago Kojun Tibana[1,b], Isa Félix Adôrno[1,c], Edson Marchiori[2,d], Thiago Franchi Nunes[1,e]

1. Universidade Federal de Mato Grosso do Sul (UFMS), Campo Grande, MS, Brazil. 2. Universidade Federal do Rio de Janeiro (UFRJ), Rio de Janeiro, RJ, Brazil.

Correspondence: Dr. Edson Marchiori. Rua Thomaz Cameron, 438, Valparaiso. Petrópolis, RJ, Brazil, 25685-120. Email: edmarchiori@gmail.com.

a. 0000-0002-8679-7369; b. 0000-0001-5930-1383; c. 0000-0002-2106-1211; d. 0000-0001-8797-7380; e. 0000-0003-0006-3725.

Received 19 September 2017. Accepted after revision 9 October 2017.

http://dx.doi.org/10.1590/0100-3984.2017.0167

Gorham-Stout syndrome: the radiologic-pathologic correlation as a diagnostic pathway when bone is vanishing

Dear Editor,

A 34-year-old previously healthy man presented with a 12-month history of progressive polyarthralgia and edema of the hips, right ankle, and intercostal spaces. He reported no history of trauma. Conventional radiography revealed several mixed lesions (predominantly osteolytic lesions) in the pelvic ring, proximal femur, distal femur, distal tibia, both tali, and lumbar vertebral bodies, as well as unconsolidated fractures of the costal arches, with no periosteal reaction or associated soft tissue changes (Figure 1). The initial hypotheses of multifocal osteolysis were secondary osteolytic conditions such as infection, cancer (primary or metastatic), inflammatory disorders, and endocrine disorders. The results of laboratory tests (complete blood count, protein profiles, parathyroid hormone level, ionic calcium level, and phosphate level) were normal, as were those

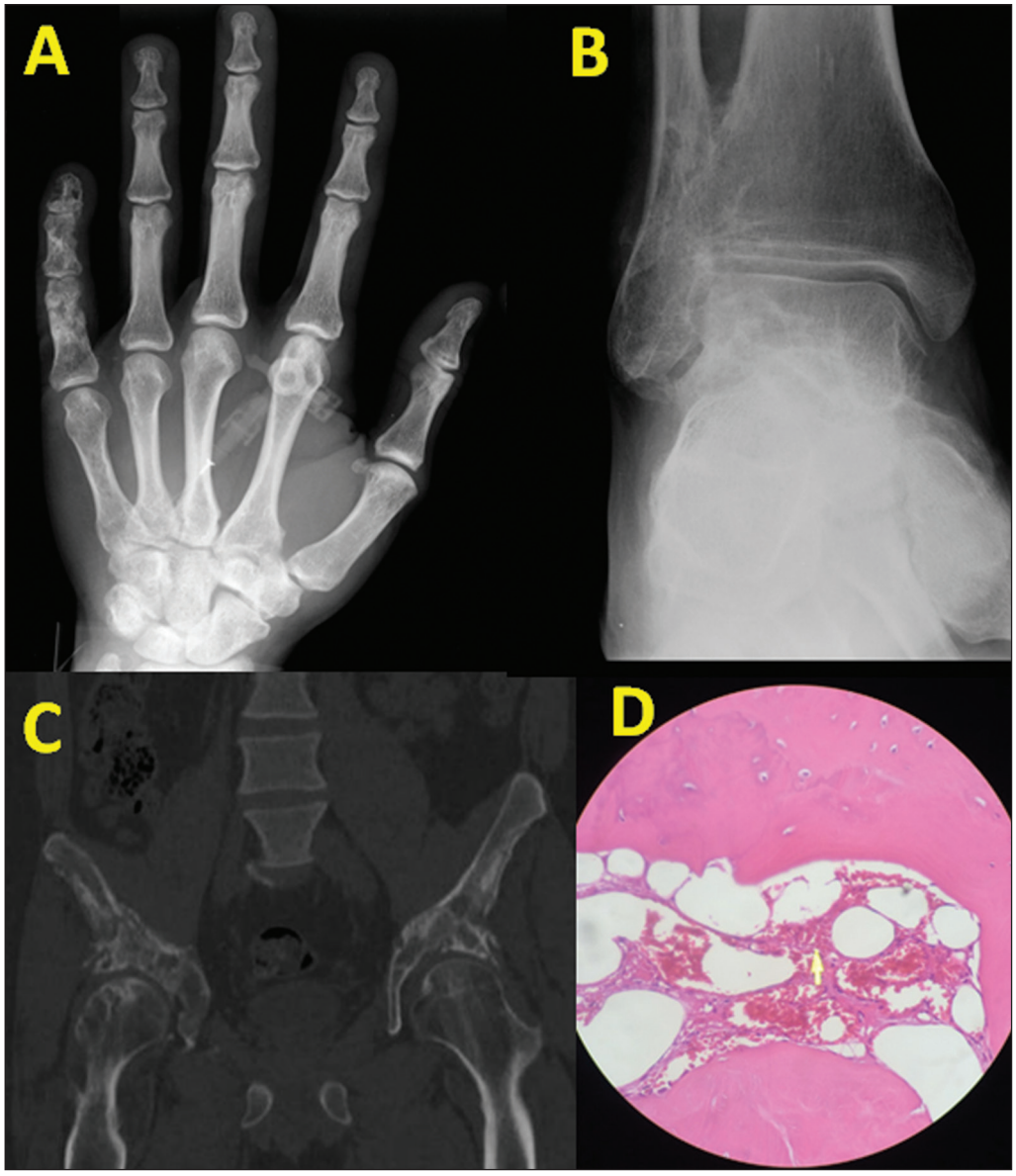

Figure 1. A: X-ray of the right hand, demonstrating permeative osteolytic lesions in the middle and distal phalanges of the fifth finger. A similar pattern is seen on computed tomography: lesions of the left malleolus and fibular metadiaphysis **(B)**; and lesions affecting the iliac and femur **(C)**. **D:** Histology of the lesion biopsied in the right iliac, showing cavernous proliferation of intraosseous capillaries throughout the preserved bone tissue (hematoxylin-eosin staining).

of computed tomography staging. After the primary hypothesis of multifocal osteolysis had been excluded, the radiological hypothesis of Gorham-Stout syndrome was proposed. The patient was submitted to surgical biopsy of the right iliac bone, and the histopathology demonstrated spongy bone marrow tissue, the bone marrow having been replaced by fibrovascular tissue with innumerable capillary and cavernous vessels, cortical bone resorption, and reactive immature bone, consistent with massive osteolysis (Figure 1).

Initially described as "missing bone disease with intraosseous vascular alterations"[1,2], the monocentric form of osteolysis of a bone or contiguous area of bone, with a predilection for the axial skeleton, was subsequently dubbed Gorham-Stout syndrome. The process is usually monostotic, although there have been a few reports of cases in which it was polyostotic[3]. In decreasing order of frequency, it affects the scapula, proximal end of the humerus, femur, rib, iliac bone, ischium, and sacrum[3]. It is currently known by a variety of names, including massive osteolysis, idiopathic osteolysis, vanishing bone disease, disappearing bone disease, phantom bone disease, spontaneous bone absorption, progressive bone atrophy, bone hemangiomatosis, and lymphangiomatosis of bone.

Gorham-Stout syndrome primarily affects children and young adults, the affected individuals presenting nonspecific complaints such as pain and joint edema. The pathological mechanisms proposed to date are multifactorial, with no known triggering factor. The histopathological hallmark is the replacement of normal bone by fibrous tissue with aggressive, expansile, non-neoplastic proliferation of capillary or cavernous blood vessels[3–7], a process that can culminate in the replacement of the entire bone by fibrous tissue[7]. Histopathologically and radiologically, the syndrome initially presents as foci of rarefaction in the bone marrow and subcortical bone, with slow, irregular progression that can result in effacement of the diaphysis of the bones, narrowing of the involved ends, and, in some cases, the complete disappearance of the bone. Pathological fractures that do not consolidate are common and are characteristic of the syndrome[1,8]. The pathological-radiological diagnosis proposed by Heffez et al.[6] is based on specific criteria, including positive biopsy findings for angiomatous tissue and an osteolytic pattern

on X-rays, as well as negativity for hereditary, metabolic, neoplastic, immunologic and infectious etiologies.

In summary, although rare, the diagnosis of Gorham-Stout syndrome should be considered in cases of focal or multifocal osteolysis in previously healthy young individuals who test negative for inflammatory, infectious, metabolic, and neoplastic conditions.

REFERENCES

1. El-Kouba G, Santos RA, Pilluski PC, et al. Síndrome de Gorham-Stout: "doença do osso fantasma". Rev Bras Ortop. 2010;45:618–22.
2. Gorham LW, Stout AP. Massive osteolysis (acute spontaneous absorption of bone, phantom bone, disappearing bone); its relation to hemangiomatosis. J Bone Joint Surg Am. 1955;37-A:985–1004.
3. Nikolaou VS, Chytas D, Korres D, et al. Vanishing bone disease (Gorham-Stout syndrome): a review of a rare entity. World J Orthop. 2014;5:694–8.
4. Collins J. Case 92: Gorham syndrome. Radiology. 2006;238:1066–9.
5. Hardegger F, Simpson LA, Segmueller G. The syndrome of idiopathic osteolysis. Classification, review, and case report. J Bone Joint Surg Br. 1985;67:89–93.
6. Heffez L, Doku HC, Carter BL, et al. Perspectives on massive osteolysis. Report of a case and review of the literature. Oral Surg Oral Med Oral Pathol. 1983;55:331–43.
7. Patel DV. Gorham's disease or massive osteolysis. Clin Med Res. 2005; 3:65–74.
8. Choma ND, Biscotti CV, Bauer TW, et al. Gorham's syndrome: a case report and review of the literature. Am J Med. 1987;83:1151–6.

Rubens Gabriel Feijó Andrade[1,a], Ana Paula Vieira Fernandes Benites Sperb[2,b], Bruno Hochhegger[2,3,c], Osvaldo Andre Serafini[1,d], Simone Gianella Valduga[1,2,e]

1. Hospital São Lucas – Pontifícia Universidade Católica do Rio Grande do Sul (PUCRS), Porto Alegre, RS, Brazil. 2. Centro Clínico – Pontifícia Universidade Católica do Rio Grande do Sul (PUCRS), Porto Alegre, RS, Brazil. 3. Universidade Federal de Ciências da Saúde de Porto Alegre (UFCSPA), Porto Alegre, RS, Brazil.

Correspondence: Dr. Rubens Feijó Andrade. PUCRS, Centro Clínico – Radiologia. Avenida Ipiranga, 6690, Petrópolis. Porto Alegre, RS, Brazil, 90610-000. Email: rubensfeijoandrade@gmail.com.

a. https://orcid.org/0000-0003-1459-8667; b. https://orcid.org/0000-0001-5998-0240; c. https://orcid.org/0000-0003-1984-4636; d. https://orcid.org/0000-0002-9303-7237; e. https://orcid.org/0000-0001-9167-7284.

Received 23 August 2017. Accepted after revision 16 October 2017.

http://dx.doi.org/10.1590/0100-3984.2017.0145

Intestinal obstruction by a phytobezoar in a patient with a history of gastroplasty

Dear Editor,

We report the case of a 28-year-old female patient with a one-year history of gastroplasty who was suffering from abdominal pain one day after eating a large amount of jackfruit. The physical exam revealed diffuse pain on palpation, positive abrupt decompression and absence of bowel sounds. Computed tomography (CT) of the abdomen and pelvis showed signs of intestinal obstruction. Intraoperatively, intraluminal content, consistent with a phytobezoar (a jackfruit "bolus"), was observed impacting the distal anastomosis of the gastric bypass (Figure 1).

A bezoar is a mass of exogenous undigested material that accumulates in the gastrointestinal tract, usually in the stomach or ileus, and causes intestinal obstruction[1]. Bezoars are associated with predisposing factors such as poor mastication, psychiatric disorders, and impaired gastric motility.

Bezoars are classified, according to their composition, as phytobezoars (composed of vegetable fibers), lactobezoars (com-

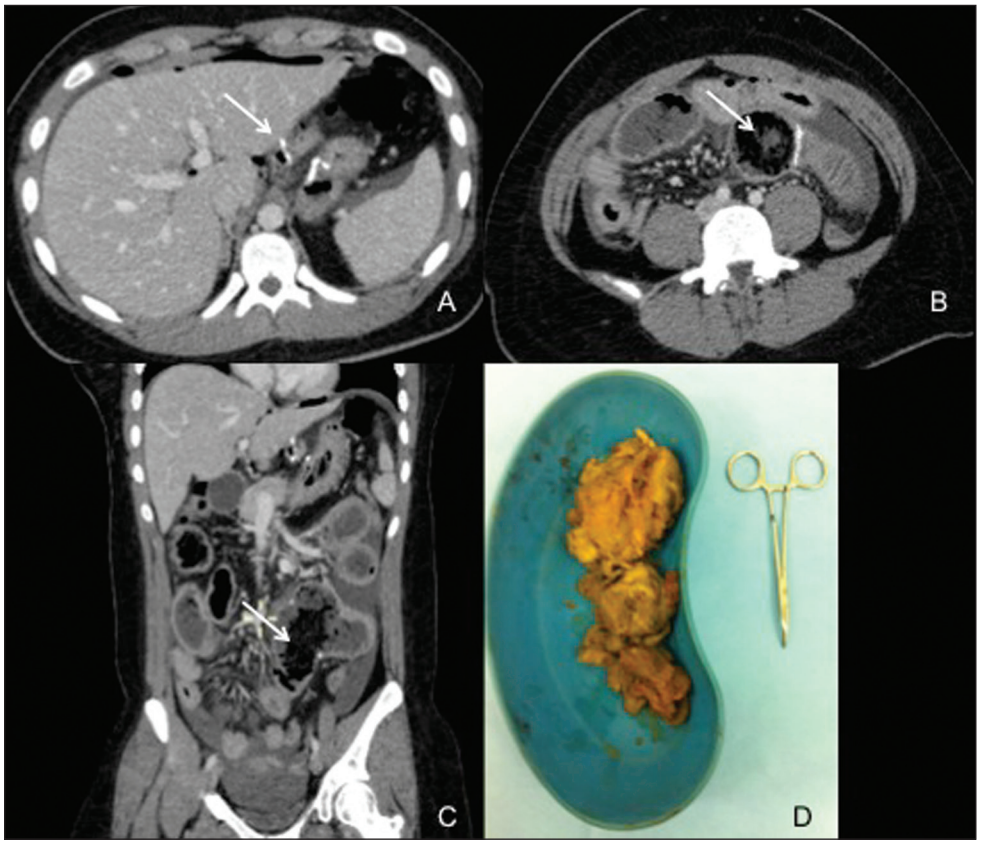

Figure 1. CT of the abdomen and pelvis, with intravenous contrast. **A:** Axial sequence showing signs of gastroplasty (arrow) and a small amount of perisplenic fluid. **B:** Axial sequence showing distension of the jejunal loop related to enteric anastomosis, highlighting the accumulation of material with low-grade intraluminal attenuation, corresponding to a phytobezoar (arrow). **C:** Coronal reconstruction confirming the signs of intestinal obstruction and again showing significant distention of the small intestine loop that participates in the enteric anastomosis, containing an accumulation of material with low attenuation (phytobezoar, arrow). **D:** Surgical specimen. Material removed from within the jejunal loop related to the surgical anastomosis, characterized by a bolus of undigested agglomerated vegetable fiber (a jackfruit phytobezoar).