classification, based on the type of fibrillar component in amyloid deposits, there are innumerable subtypes. In the vast majority of cases, light-chain amyloid fibrils and serum amyloid A are identified[1].

In the thoracic compartment, amyloidosis typically affects the heart but can also involve the pulmonary parenchyma, pleura, lymph node chains, tracheobronchial tree, and other sites[1,2]. Pulmonary involvement is rare, reported as tracheobronchial, diffuse/alveolar-septal, or nodular manifestations, the first being the most common[2–4].

The tracheobronchial manifestation of amyloidosis is characterized by the deposition of amyloid material in the trachea and main bronchi, resulting in thickening of the walls, narrowing of the lumina, and consequent airway obstruction, as well as consolidations, atelectasis, pulmonary hyperinflation, and bronchiectasis[3].

Clinically, amyloidosis-related tracheobronchial impairment can be asymptomatic or can manifest as dyspnea, wheezing, hemoptysis, cough, or recurrent pneumonia[4,5]. The symptoms can be similar to those of bronchial diseases that are more common, including bronchial asthma[5].

Chest CT has been shown to be the imaging exam of choice for the evaluation of thoracic diseases[6–9], as well as for that of diseases of the tracheobronchial tree[10–12]. In individuals with amyloidosis, a CT scan can reveal smooth or irregular/nodular thickening of the tracheal wall and bronchi, which can be accompanied by calcified nodules in the submucosa[4]. The differential diagnoses of diffuse tracheobronchial diseases include vasculitis (Wegener's granulomatosis), tracheobronchial papillomatosis, infectious involvement (rhinoscleroma, caused by infection with Klebsiella rhinoscleromatis), tracheopathia osteochondroplastica, and relapsing polychondritis[13]. Unlike tracheal involvement in tracheopathia osteochondroplastica or relapsing polychondritis, tracheobronchial amyloidosis involves the posterior membranous wall of the trachea[4,13].

In individuals with amyloidosis, bronchoscopy usually shows thickening of the walls of the trachea and bronchi, with flat, multifocal, grayish-yellow plaques in the trachea and bronchi. In rare cases, amyloid pseudotumors can be seen[5,13]. Histopathological findings of the disease include amyloid thickening of the submucosa, in nodular masses or laminae, showing apple-green birefringence after staining with Congo red[14]. There is also a reduction in the number of submucosal glands, together with calcifications and foci of bone metaplasia in the upper airways[14].

In patients suspected of having bronchial asthma who present with atypical symptoms and respond poorly to clinical treatment, various differential diagnoses should be considered[15]. The patient in question was initially diagnosed with asthma but did not respond to treatment, and the definitive diagnosis of primary tracheobronchial amyloidosis was made after a directed follow-up assessment. We can conclude that, albeit rare, tracheobronchial amyloidosis should be considered in such patients.

REFERENCES

1. Czeyda-Pommersheim F, Hwang M, Chen SS, et al. Amyloidosis: modern cross-sectional imaging. Radiographics. 2015;35:1381–92.
2. Marchiori E, Souza Jr AS, Ferreira A, et al. Amiloidose pulmonar: aspectos na tomografia computadorizada. Radiol Bras. 2003;36:89–94.
3. Lee AY, Godwin JD, Pipavath SN. Case 182: pulmonary amyloidosis. Radiology. 2012;263:929–32.
4. Ngo AV, Walker CM, Chung JH, et al. Tumors and tumorlike conditions of the large airways. AJR Am J Roentgenol. 2013;201:301–13.
5. Serraj M, Kamaoui I, Znati K, et al. Pseudotumoral tracheobronchial amyloidosis mimicking asthma: a case report. J Med Case Rep. 2012;6:40.
6. Francisco FAF, Rodrigues RS, Barreto MM, et al. Can chest high-resolution computed tomography findings diagnose pulmonary alveolar microlithiasis? Radiol Bras. 2015;48:205–10.
7. Batista MN, Barreto MM, Cavaguti RF, et al. Pulmonary artery sarcoma mimicking chronic pulmonary thromboembolism. Radiol Bras. 2015; 48:333–4.
8. Torres PPTS, Moreira MAR, Silva DGST, et al. High-resolution computed tomography and histopathological findings in hypersensitivity pneumonitis: a pictorial essay. Radiol Bras. 2016;49:112–6.
9. Mogami R, Goldenberg T, Marca PGC, et al. Pulmonary infection caused by *Mycobacterium kansasii*: findings on computed tomography of the chest. Radiol Bras. 2016;49:209–13.
10. Ribeiro GMR, Natal MRC, Silva EF, et al. Tracheobronchopathia osteochondroplastica: computed tomography, bronchoscopy and histopathological findings. Radiol Bras. 2016;49:56–7.
11. Barbosa BC, Amorim VB, Ribeiro LFM, et al. Tuberculosis: tracheal involvement. Radiol Bras. 2016;49:410–1.
12. Barbosa AGJ, Penha D, Zanetti G, et al. Foreign body in the bronchus of a child: the importance of making the correct diagnosis. Radiol Bras. 2016;49:340–2.
13. Prince JS, Duhamel DR, Levin DL, et al. Nonneoplastic lesions of the tracheobronchial wall: radiologic findings with bronchoscopic correlation. Radiographics. 2002;22 Spec No:S215–30.
14. Kurtz KA, Kirby PA. Pathologic quiz case: a 49-year-old man with chronic cough and a left lung hilar mass. Tracheobronchial amyloidosis. Arch Pathol Lab Med. 2003;127:e420–2.
15. Tilles SA. Differential diagnosis of adult asthma. Med Clin North Am. 2006;90:61–76.

Pedro Paulo Teixeira e Silva Torres[1], Matheus Rabahi[2], Sebastião Alves Pinto[3], Karla Cristina de Morais Arantes Curado[4], Marcelo Fouad Rabahi[3]

1. Multimagem Diagnósticos, Goiânia, GO, Brazil. 2. Pontifícia Universidade Católica de Goiás (PUC Goiás), Goiânia, GO, Brazil. 3. Universidade Federal de Goiás (UFG), Goiânia, GO, Brazil. 4. Hospital e Maternidade Jardim América, Goiânia, GO, Brazil. Mailing address: Dr. Pedro Paulo Teixeira e Silva Torres. Rua 9, nº 326, Residencial Amaury Menezes, ap. 1502, Setor Oeste. Goiânia, GO, Brazil, 74110-100. E-mail: pedroptstorres@yahoo.com.br.

http://dx.doi.org/10.1590/0100-3984.2015.0177

Xanthogranulomatous cystitis in a child

Dear Editor,

A seven-year-old female patient with acute appendicitis underwent an emergency appendectomy. During the procedure, as incidental findings, a bulky bladder and a probable collection adhered to the wall were observed. Cystoscopy revealed an enlarged bladder with diffuse thickening of its walls. Subsequently, computed tomography of the abdomen showed a well-defined, hypointense collection, with cystic attenuation, with regular contours, showing no enhancement and in contact with the right lateral wall of the bladder (Figure 1). An investigation of pathological antecedents revealed that the patient had experienced recurrent episodes of cystitis in the last year. The decision was made to perform laparoscopic surgery, during which a small communicating orifice was identified (between the lesion and the interior of the bladder) and partial cystectomy was performed. Histopathological analysis demonstrated fibroadipose tissue exhibiting a xanthogranulomatous reaction (characterized by the presence of xanthomatous macrophages), together with a giant-cell reaction, cholesterol crystals, and mild chronic inflammatory infiltrate. A similar macrophage reaction was observed in the lymph node (Figure 2). In view of those findings, the main diagnostic hypothesis was xanthogranulomatous cystitis.

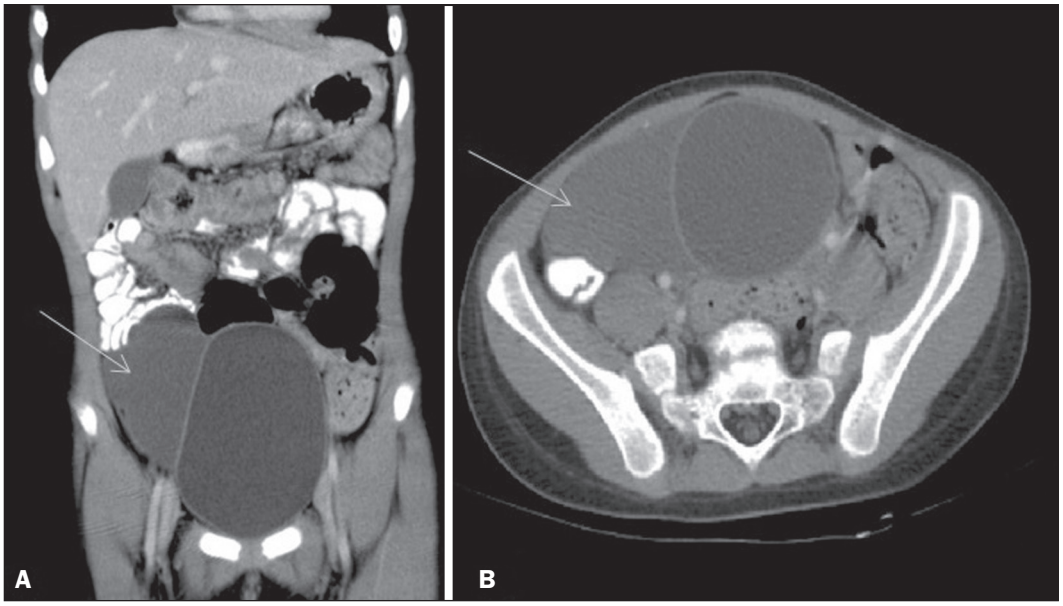

Figure 1. Coronal (**A**) and axial (**B**) reconstructions of contrast-enhanced computed tomography of the abdomen, showing a well-defined, hypointense collection with regular contours, with cystic attenuation, showing no enhancement and in contact with the right lateral wall of the bladder.

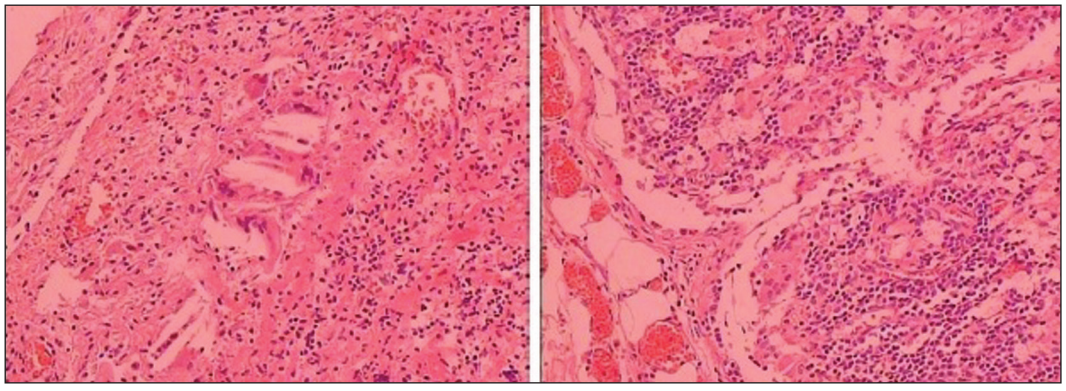

Figure 2. Histopathological section showing fibroadipose tissue with a xanthogranulomatous reaction (characterized by the presence of xanthomatous macrophages), a giant-cell reaction, cholesterol crystals, and mild chronic inflammatory infiltrate. A similar macrophage reaction can be seen in the lymph node.

Xanthogranulomatous cystitis is a rare chronic inflammatory disease, only approximately 30 cases having been documented in the literature. It has a benign course and its origin remains obscure. However, previous reports have suggested possible associations with a remnant of the urachus, chronic infection, malignant bladder tumor, and immune disorders[1]. The clinical symptoms are non-specific and therefore do not facilitate the differential diagnosis with other diseases of the bladder. The most common forms of presentation are irritative urinary symptoms, a palpable mass in the abdomen, and hematuria[2–4]. Among the cases published in the literature, that the mean age at onset is approximately 46 years, with no gender predominance, and the preferential location is in the dome of the bladder[1,4,5]. However, the case presented here was in a seven-year-old (pediatric) patient, in whom the lesion was located in the right lateral wall, thus ruling out any association with the urachus.

In individuals with xanthogranulomatous cystitis, conservative treatment is not effective. Such individuals require surgical resection by partial cystectomy, which is currently the gold standard treatment fore the disease[2–5]. Xanthogranulomatous lesions can occur at sites other than the bladder, typically the kidneys or, less frequently, the gall bladder, pancreas, appendix, colon, ovary, endometrium, and brain, usually mimicking malignancy[2–4].

Xanthogranulomatous cystitis is an extremely rare disease and continues to be the subject of many studies, because little is known about its true cause and behavior over the long term. This case highlights the importance of recognizing an unusual lesion that can present in individuals of any age and can impede the final diagnosis.

REFERENCES

1. Yamamoto S, Yoshida K, Tsumura K, et al. Xanthogranulomatous cystitis treated by transurethral resection. Urol Case Rep. 2015;3:143–5.
2. Izquierdo García FM, García Diez F, Miguelez Simon A, et al. Cistitis xantogranulomatosa: presentación de un caso. Arch Esp Urol. 2001;54:263–5.
3. Hayashi N, Wada T, Kiyota H, et al. Xanthogranulomatous cystitis. Int J Urol. 2003;10:498–500.
4. Wang Y, Han XC, Zheng LQ, et al. Xanthogranulomatous cystitis imitating bladder neoplasm: a case report and review of literature. Int J Clin Exp Pathol. 2014;7:8255–8.
5. Ekici S, Ekici I, Ruacan S, et al. Xanthogranulomatous cystitis: a challenging imitator of bladder cancer. ScientificWorldJournal. 2010;10:1169–73.

Roberto César Teixeira Dantas[1], Ivo Lima Viana[1], Camila Soares Moreira de Sousa[1], Breno Braga Bastos[2], Carla Lorena Vasques Mendes de Miranda[1]

1. Medimagem, Teresina, PI, Brazil. 2. UDI 24 horas, Teresina, PI, Brazil. Mailing address: Dra. Camila Soares Moreira de Sousa. Medimagem – Radiologia. Rua Paissandu, 1862, Centro. Teresina, PI, Brazil, 64001-120. E-mail: camilasoares__@hotmail.com.

http://dx.doi.org/10.1590/0100-3984.2015.0225