

Nevertheless, several studies indicate a higher risk of lower intestinal perforation in patients treated with tocilizumab,³⁻⁵ with a described incidence of close to 2/1,000 patients/year in whom at least one dose of tocilizumab has been administered,⁶ especially at 8 mg/kg versus 4 mg/kg.⁷ Similarly, the risk of intestinal perforation is higher compared to other disease-modulating biological drugs.^{3,4,8,9} These perforations appear in the first 12 months after treatment, do not increase over time,^{5,9} and are always more frequent in patients who have received corticosteroids.^{5,6} A possible explanation could be the role of IL-6³ in the intestinal barrier function, as well as a lower intensity in the immediate inflammatory response.⁵ Finally, it should be noted that mortality after perforation in these cases can reach 46%,³ a fact that would significantly worsen the already uncertain prognosis of patients with COVID-19 infection.¹⁰

REFERENCES

- Kim J, Thomsen T, Sell N, Goldsmith AJ. American Journal of Emergency Medicine Abdominal and testicular pain: an atypical presentation of COVID-19. *Am J Emerg Med*. 2020. <http://dx.doi.org/10.1016/j.ajem.2020.03.052>.
- Kremer JM, Blanco R, Brzosko M, Burgos-vargas R, Halland A, Vernon E, et al. Tocilizumab inhibits structural joint damage in rheumatoid arthritis patients with inadequate responses to methotrexate results from the double-blind treatment phase of a randomized placebo-controlled trial of tocilizumab safety and prevention of structur. *Arthritis Rheum*. 2011;63:609-21.
- Strangfeld A, Richter A, Siegmund B, Herzer P, Rockwitz K, Demary W, et al. Risk for lower intestinal perforations in patients with rheumatoid arthritis treated with tocilizumab in comparison to treatment with other biologic or conventional synthetic DMARDs. *Ann Rheum Dis*. 2017;76:504-10.
- Xie F, Yun H, Bernatsky S, Curtis JR. Risk for gastrointestinal perforation among rheumatoid arthritis patients receiving tofacitinib, tocilizumab, or other biologics. *Arthritis Rheumatol*. 2016;68:2612-7.
- Yamamoto K, Goto H, Hirao K, Nakajima A, Origasa H. Longterm safety of tocilizumab: results from 3 years of followup postmarketing surveillance of 5573 patients with rheumatoid arthritis in Japan. *J Rheumatol*. 2015;42:1368-75.
- Schiff MH, Kremer JM, Jahreis A, Vernon E, Isaacs JD, Vollenhoven RF Van. Integrated safety in tocilizumab clinical trials. *Arthritis Res Ther*. 2011;13:R141.
- Emery P, Keystone E, Tony HP, Cantagrel A, Vollenhoven R Van, Sanchez A, et al. IL-6 receptor inhibition with tocilizumab improves treatment outcomes in patients with rheumatoid arthritis refractory to anti-tumour necrosis factor biologicals: results from a 24-week multicentre randomised placebo-controlled trial. *Ann Rheum Dis*. 2008;67:1516-23.
- Sepriano A, Kerschbaumer A, Smolen JS, Van Der Heijde D, Dougados M, van Vollenhoven R, et al. Safety of synthetic and biological DMARDs?: a systematic literature review informing the 2019 update of the EULAR recommendations for the management of rheumatoid arthritis. *Ann Rheum Dis*. 2020. <http://dx.doi.org/10.1136/annrheumdis-2019-216653>.
- Monemi S, Berber E, Sarsour K, Wang J. Incidence of gastrointestinal perforations in patients with rheumatoid arthritis treated with tocilizumab from clinical trial, postmarketing, and real-world data sources. *Rheumatol Ther*. 2016;3:337-52.
- Aminian A, Safari S, Razeghian-Jahromi A, Ghorbani M, Delaney CP. COVID-19 outbreak and surgical practice: unexpected fatality in perioperative period. *Ann Surg*. 2020. <http://dx.doi.org/10.1097/SLA.0000000000003925>.

Paula González Guardiola^{a,*}, Jose Ángel Díez Ares^{a,b},
Nuria Peris Tomás^{a,b}, Juan Carlos Sebastián Tomás^a,
Sergio Navarro Martínez^a

^aHospital Doctor Peset, Valencia, Spain

^bUnidad de Cirugía Bariátrica, Hospital Doctor Peset, Valencia, Spain

*Corresponding author.

E-mail address: p.gonzalez.g@gmail.com

(P. González Guardiola).

2173-5077/

© 2020 AEC. Published by Elsevier España, S.L.U. All rights reserved.

Scientific letter

Retrorectal broncogenic cyst[☆]

Quiste broncogénico retrorrectal



Bronchogenic cysts develop between the third and seventh weeks of intrauterine life and are due to abnormalities in the embryological development of the primitive intestine and the

tracheobronchial tree. Their typical location is mediastinal, while retroperitoneal cysts are very infrequent and the retrorectal location is exceptional.¹

[☆] Please cite this article as: García-Romera Á, Tuñón-Féquant C, Díaz-Pérez B, Bravo-Gutiérrez A, Alarcó-Hernández A. Quiste broncogénico retrorrectal. *Cir Esp*. 2021;99:157-159.

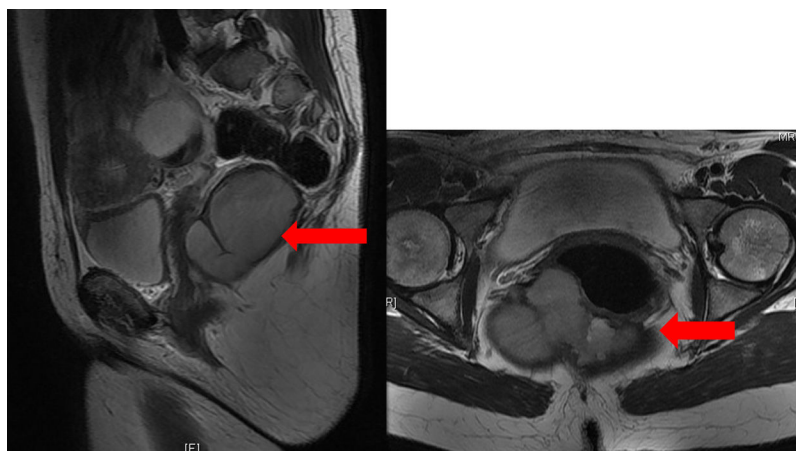


Fig. 1 – Sagittal and axial projections of the cystic mass (marked with an arrow), and their relationship with the rectum and the levator ani muscles.

We present the case of a 32-year-old patient who was admitted for hypogastric abdominal pain and right iliac fossa pain over the past 6 months, whose intensity had increased in the last month. There was no associated fever or any other symptoms. Upon examination, the patient presented discomfort in the hypogastrium and right iliac fossa, with no peritoneal irritation or palpated masses.

Lab work showed no findings of interest. Abdominal tomography revealed a right pararectal septated cystic mass measuring 10×6 cm. In view of the findings, the patient was admitted to hospital and the study was completed with an MRI, which showed a multiloculated presacral cystic mass measuring $10.3 \times 7.8 \times 5.7$ cm in diameter, suggestive of retrorectal cystic hamartoma or tailgut cyst (Fig. 1).

Given the possible complication of this lesion, the possibility of degeneration and the patient's symptoms, laparoscopic surgery was performed in which a large cystic mass was observed that extended retrorectally from the mid-rectum to the levator ani muscles without depending on them. We conducted a difficult dissection of the entire lesion, during which an incidental lesion of the rectum measuring 1.5 cm occurred, which was sutured endoanally. Finally, the cystic lesion was removed and a protective ileostomy was performed.

The anatomic pathology study revealed a cavitory lesion lined by respiratory-type epithelium (cylindrical pseudostratified ciliated with goblet cells), which was compatible with a bronchogenic cyst.

The patient was discharged on the sixth postoperative day and is currently awaiting stoma closure.

Bronchogenic cysts are caused by an abnormal development of the ventral diverticulum of the tracheobronchial tree and are usually located in the lung parenchyma or mediastinum.²

When they are totally separated from the tracheobronchial tree, they can migrate to atypical locations, specifically to the retroperitoneum in 0.03%² of cases. At this level, the peripancreatic location is the most frequent (28% of cases³), while the retrorectal location is very unusual.

The incidence of bronchogenic cysts is similar in both sexes. Mean patient age is around 40 years, and mean cyst size is 6 cm.⁴

Most cases are diagnosed incidentally.¹ However, these cysts can bleed, perforate, compress adjacent structures due to their growth, and even become malignant.⁵

Tomography and magnetic resonance imaging will be the fundamental tests used for diagnosis, but if these lesions are found at the biliary or pancreatic level, it is advisable to carry out endoscopic ultrasound and fine-needle aspiration of the lesion.³

The differential diagnosis should include a variety of entities, such as cystic hamartomas, sacrococcygeal teratoma, anterior sacral meningocele, rectal duplication cysts, and rectal leiomyosarcomas.⁶

Treatment is surgical to control symptoms and avoid the risk of malignization.³ The recommended surgical approaches are sacrococcygeal for lesions located below S4, transanal in those smaller than 4 cm, or abdominal in large lesions and above S3.⁷ Although the first 2 are the most popular, the laparoscopic abdominal approach has also been described.^{3,8}

Histological analysis will show cartilage and bronchial glands with seromucosal content.⁹

In the present case, the initial imaging diagnosis was cystic hamartoma, a very rare benign tumor. To avoid the risk of dissemination in the case of malignant disease, we decided not to perform ultrasound-guided needle aspiration of the lesion. Although it is true that the laparoscopic approach has not been widely described in the literature, we chose this as it was a large lesion located above the levator ani muscles in a young patient in whom we did not want to alter the perineal muscles.

We believe that information about a rare disease, in an exceptional location, with risk of malignization, and the experience of its laparoscopic treatment is important for colorectal surgeons.

REFERENCES

1. Pasquer A, Djeudji F, Hervieu V, Rabeyrin M, Barth X. A rare retrorectal presentation of a bronchogenic cyst: a case report. *Int J Surg Case Rep.* 2016;24:112-4. <http://dx.doi.org/10.1016/j.ijscr.2016.05.028>.

2. Brient C, Muller C, Cassagneau P, Taieb D, Sebag F, Henry JR, et al. Kyste bronchogénique rétroperitoneal. *J Chir Visc*. 2012;149:361–3.
3. Runge T, Blank A, Schäfer SC, Candinas D, Gloor B, Angst E. A retroperitoneal bronchogenic cyst mimicking a pancreatic or adrenal mass. *Case Rep Gastroenterol*. 2013;7:428–32. <http://dx.doi.org/10.1159/000355879>.
4. Dua KS, Vijayapal AS, Kengis J, Shidham VB. Ciliated foregut cyst of the pancreas: preoperative diagnosis using endoscopic ultrasound guided fine needle aspiration cytology—a case report with a review of the literature. *Cytojournal*. 2009;6:22. <http://dx.doi.org/10.4103/1742-6413.56362>.
5. Goh BK, Chan HS, Wong WK. A rare case of “giant” right sided retroperitoneal bronchogenic cyst. *Dig Dis Sci*. 2004;49:1491–2.
6. Cai Y, Guo Z, Cai Q, Dai S, Gao W, Niu Y, et al. Bronchogenic cysts in retroperitoneal region. *Abdom Imaging*. 2013;38:211–4. <http://dx.doi.org/10.1159/000355879>.
7. Pidala MJ, Eisenstat TE, Rubin RJ, Salvati EP. Presacral cysts: transrectal excision in select patients. *Am Surg*. 1999;65:112–5.
8. Díaz Nieto R, Naranjo Torres A, Gomez Alvarez M, Ruiz Rabelo JF, Pérez Manrique MC, Ciria Bru R, et al. Intraabdominal bronchogenic cyst. *J Gastrointest Surg*. 2010;14:756–8. <http://dx.doi.org/10.1007/s11605-009-0932-5>.
9. Woon CS, Pambuccian SE, Lai R, Jessurun J, Woon CS, Pambuccian SE, et al. Ciliated foregut cyst of pancreas: cytologic findings on endoscopic ultrasound-guided fine-needle aspiration. *Diagn Cytopathol*. 2007;35:433–8. <http://dx.doi.org/10.1002/dc.20659>.

Ángel García-Romera*, Carlota Tuñón-Féquant,
Beatriz Díaz-Pérez, Alberto Bravo-Gutiérrez,
Antonio Alarcó-Hernández

Servicio de Cirugía General y del Aparato Digestivo, Hospital
Universitario de Canarias, La Laguna, Santa Cruz de Tenerife, Spain

*Corresponding author.

E-mail address: garcia.romera1988@gmail.com
(Á. García-Romera).

2173-5077/

© 2020 AEC. Published by Elsevier España, S.L.U. All rights reserved.

Neuroblastic tumors in adults[☆]

Tumores neuroblásticos en el adulto



Neuroblastic tumors are neoplasms of the sympathetic nervous system originating from primitive sympathetic adrenergic cells formed in the neural crest during embryonic development. They are almost exclusively found in children; their presentation in adults is very rare and associated with a poor prognosis. The location of these tumors is usually intra-abdominal, generally adrenal, and their treatment is surgical whenever possible¹. We present the case of a neuroblastic tumor diagnosed in an adult.

A 22-year-old male patient with no relevant personal history underwent an abdominal ultrasound to study mild, nonspecific, self-limited low back pain, as the origin could not be identified as mechanical and/or urinary. As an incidental finding during this ultrasound, a large right retroperitoneal mass was observed. On examination, a mass was palpable that occupied the entire right hemiabdomen, accompanied by mild local discomfort. The patient did not present obstructive intestinal or urological symptoms, fever, or other symptoms. Lab work demonstrated a slight increase in C-reactive protein (CRP) (1.40 mg/dL) and a sedimentation

rate of 22 mm/h; the remaining parameters were normal. After hospital admittance for further study, a computed tomography (CT) scan revealed a voluminous mass occupying the right hypochondrium and flank. The mass measured 15 × 20 × 29 cm and appeared to have a retroperitoneal origin (the adrenal gland was an initial possibility), with no evidence of lymphadenopathies or metastatic thoracoabdominal extension (Fig. 1). Catecholamine/metanephrine levels in urine were normal.

At this time, we decided to perform diagnostic-therapeutic surgery. A bilateral subcostal laparotomy revealed a large mass measuring 35 × 20 cm that appeared to originate in the right adrenal gland. It was resected, after identification, ligation and division of the right inferior adrenal artery and vein (Fig. 1b). The histopathological study of the specimen revealed a poorly differentiated intermixed/nodular ganglioneuroblastoma of adrenal origin, with R1 resection margin. Subsequently, the patient was discharged three days after the procedure, with outpatient follow-ups by the Surgery and Oncology Services. On the postoperative ¹²³I-MIBG scinti-

[☆] Please cite this article as: Baeza Murcia M, Valero Navarro G, Flores Pastor B, García Marín JA, Aguayo Albasini JL. Tumores neuroblásticos en el adulto. *Cir Esp*. 2021;99:159–161.