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LETTER TO THE EDITOR

Colon hemangiolymphangioma—a rare case of subepithelial polyp

F. Castro-Poças · Luís Lobo · Teresina Amaro · José Soares · Miguel Mascarenhas Saraiva

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Dear Editor:

The majority of colon polyps have an epithelial nature. There is a small subset of them that are submucosal proliferations of mesenchymal cells, such as lipomas, leiomyomas, schwannomas, gastrointestinal stromal tumours and vascular lesions [1]. These last ones are extremely rare when responsible for polypoid lesions of the colon [2].

Clinical case

An asymptomatic 64-year-old man without a relevant clinical history was referred for endoscopic colorectal cancer screening.

In the colonoscopy, a polypoid lesion was identified, which was pediculated and measuring about 25 mm at the proximal transverse colon. The lesion was covered by normal-appearing mucosa.

We performed a miniprobe endoscopic ultrasonography, 12 MHz, which showed a polypoid morphology lesion, which heterogeneous and cystic, with various anechoic cavities separated by different thickness septa, being a part of them incomplete. The lesion was well delimited with regular boarders and located in the submucosa, with interface preservation with the adjacent layers. The size of the lesion was 27.1 by 20 mm.

The patient required a definitive diagnosis. The endoscopic resection was performed with a hot snare. There were no complications.

The patient is well with no additional colorectal lesions 2 years after the polyp's resection.

The macroscopic examination of the excised specimen showed a circumscribed round polyp, which is 25 mm in diameter, of soft consistency and covered by normal-appearing mucosa. The histological examination showed a lesion with capillary and lymphatic-type blood vessels in mucosa and submucosa; positivity for CD31 and D2-40 was expressed in the endothelial cells of lymphatic vessels. A definitive diagnosis of colon hemangiolymphangioma was made.

Discussion

It is well known that the vast majority of polyp lesions have epithelial nature, rarely having a subepithelial mesenchymal cells one [1].

Excluding certain vascular lesions, such as angioectasias, the real vascular tumours, such as haemangiomas, are rare in the digestive tract [3]. Among vascular tumours, hemangiolymphangiomas are a rare entity. Recently Andra-Luca and Montgmory published the first description of hemangiolymphangiomas in the digestive tract [2]. During a period of 22 years, they identified 27 patients with vascular tumours, being 12 cases of hemangiolymphangiomas, eight located in the small bowel and four in the colon.

F. Castro-Poças · M. M. Saraiva
Department of Gastroenterology, ManoPh-CUF Institute, Porto, Portugal

L. Lobo · J. Soares
Department of Gastroenterology, Pedro Hispano Hospital, Matosinhos, Portugal

T. Amaro
Department of Pathology, Pedro Hispano Hospital, Matosinhos, Portugal

F. Castro-Poças (✉)
R. Prof. Egas Moniz, n.° 1868, 4510-119 Gondomar, Portugal
e-mail: castro.pocas@sapo.pt

Hemangiolymphangiomas consist of a proliferation [3] or a network of vascular spaces of varied nature (veins, venules, arteries, arterioles, capillaries or lymphatic) liming by a benign endothelial lining with intervening connective tissue stroma and forming a tumour. Therefore the hemangiolymphangiomas show haemangioma and lymphangioma characteristics with an anastomosis network composed by blood and lymphatic vessels [4].

In our case, the identification of this lesion was an incidental finding which is in agreement with the few reported cases in the literature, where in the majority of cases, the lesions are associated with non-specific signs or symptoms and rarely anaemia or gastrointestinal bleeding [2].

In the series published by Andra-Luca, of the five resected cases, one was performed by surgery and four by endoscopy. The authors call our attention to the fact that we showed a considered eventual follow-up of the lesions, due to the risk of bleeding or recurrent subsequent lesions.

The colonoscopy performed in our patient 2 years after the lesion's resection was normal.

According to our knowledge, this was the first case of hemangiolymphangioma characterised by endoscopic ultrasonography. As we know, this technique has acquired a main role in the characterisation and clinical orientation in the subepithelial lesions of the digestive tract.

Naturally we need more cases of hemangiolymphangioma to try to define its ultrasonographic semiology, which can help in the clinical orientation, namely in the asymptomatic cases. On the one hand, when clarifying what the digestive wall layers involved by the lesion, it can help to decide between endoscopic or surgical resection. On the other hand, when contributing in a decisive way to different diagnosis possibilities, it can identify lesions that do not require any therapeutic approach or follow-up, such as lipoma.

In our case, the ultrasonographic information that the lesion was located in the submucosa was fundamental for choosing endoscopic resection and not surgical.

In spite of the colon polyps with subepithelial nature being rare, particularly vascular tumours, we can speculate that in the future, we will find more cases, since colonoscopy was recognised as the gold standard screening procedure for colorectal cancer.

In summary, we have reported a rare case of submucosal hemangiolymphangioma of the colon. Maybe it was also the first description, in the English literature, of a hemangiolymphangioma by endoscopic ultrasonography. These lesions may be treated successfully by standard polypectomy techniques.

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