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Review Article

Retrospective Diagnosis of a Stromal Tumor of a Giant Meckel's Diverticulum Revealed by an Abdominal Mass.

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Abstract

Retrospective Diagnosis of a Stromal Tumor Arising from a Giant Meckel's Diverticulum Revealed by an Abdominal Mass

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract, with an incidence ranging from 0.3% to 4% in the general population. Gastrointestinal stromal tumors (GISTs) are rare, and only a few cases of GISTs originating from MD have been reported. We report the case of a male patient in whom a stromal tumor arising from MD was diagnosed retrospectively, following the histopathological examination of a large ileal mass that was incidentally discovered as an abdominal mass.

Keywords: *Meckel's diverticulum; GIST; Stromal tumor.*

Introduction

Meckel's diverticulum (MD) represents the most common congenital anomaly of the digestive tract, with an estimated prevalence of 0.3% to 4% in the general population [1,2]. Gastrointestinal stromal tumors (GISTs) are uncommon, and their development from MD has only been described in rare cases [4,5]. Here, we report the case of a patient diagnosed with a DM stromal tumor retrospectively, following pathological analysis of a large ileal mass discovered incidentally during the exploration of an abdominal mass.

Observation

MFM, 75 years old, father of ten children, known to have a prostate adenoma, was hospitalized in January 2010 for an abdominal mass, associated with diffuse pain evolving for three years, without deterioration of the general condition. On clinical examination, the general condition was preserved. There was a mass of approximately 16 cm in diameter, periumbilical, extending from the right flank to the right iliac fossa, firm, painless, mobile in relation to the superficial plane and to respiration. There was no jaundice, ascites, or collateral venous circulation. The clinical examination was otherwise unremarkable, with the exception of

prostatic hypertrophy on digital rectal examination. The plain abdomen revealed a central water opacity. Ultrasound revealed a median, supra- and para-umbilical right tissue mass measuring 17 x 7.5 cm, hypoechoic, homogeneous, with a digestive appearance (small or transverse colon) (FIG. 1). Computed tomography (CT) showed an intraperitoneal formation measuring 18 x 10 cm in close contact with the small bowel loops, with a thickened wall and fluid-aerated content, without satellite lymphadenopathy (FIG. 2.3 4.5.6). A hepatic subcapsular effusion at the level of segment VI was noted, as well as multiple right basal atelectases and a slight pleural effusion. Prostatic hypertrophy was also observed.

The patient underwent surgery. Surgical exploration revealed an abscessed mass, vascularized at the expense of the last ileal loops, with agglutination of the loops and invasion of the anterior abdominal wall (FIG 7, 8, 9, 10). The postoperative course was marked by anemia (6 g/dL) on the 3rd day, requiring a transfusion of 3 CG.

The anatomopathological examination of the surgical specimen showed a proliferation of epithelioid cells, with a mitotic index of 1 mitosis per 50 fields. Immunohistochemistry revealed a strong positivity of the tumor cells for CD117, while the anti-CD34, anti-desmin, anti-HHF35 and anti-AE1/AE3 markers were negative (Fig 11,12,13,14,15).

Treatment with imatinib (Glivec®) at a daily dose of 400 mg was initiated, with good clinical and biological tolerance.

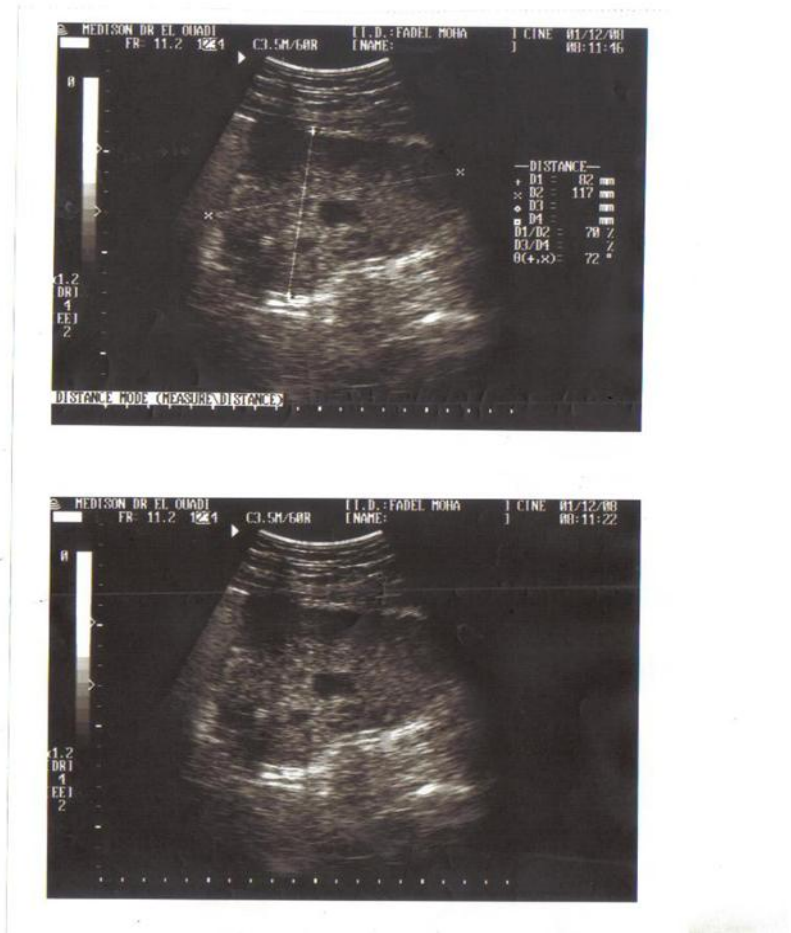

Fig. 1 ; Abdominal ultrasound showing a mass large, multi-lobed, ileal tissue mass

FIG 2, 3, 4, 5, 6: CT sections showing a large tissue mass with polylobed ileal contours.

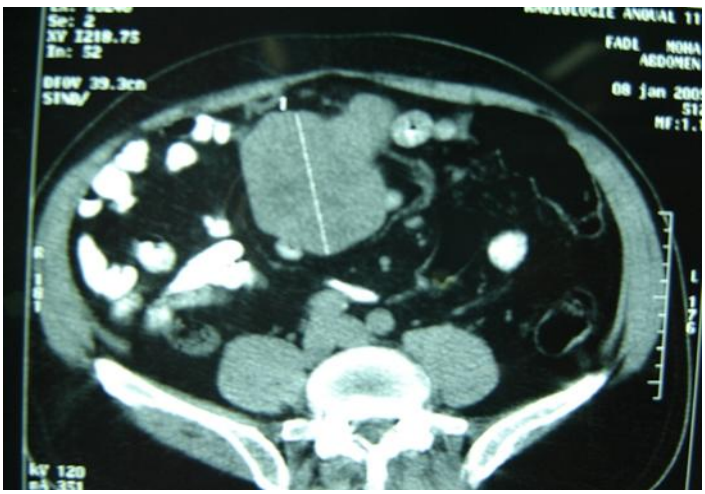

Fig 2

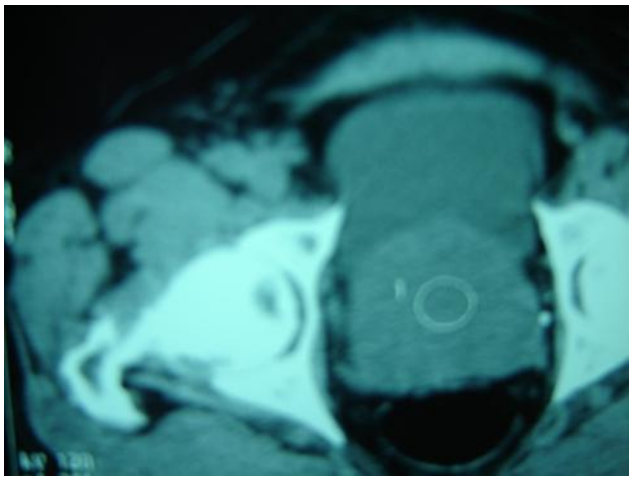

Fig 3

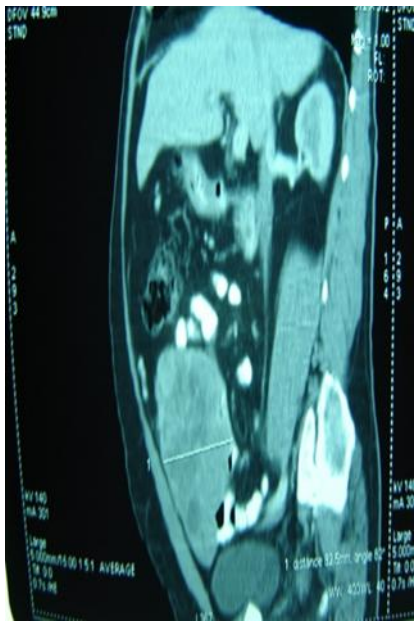

Fig 4

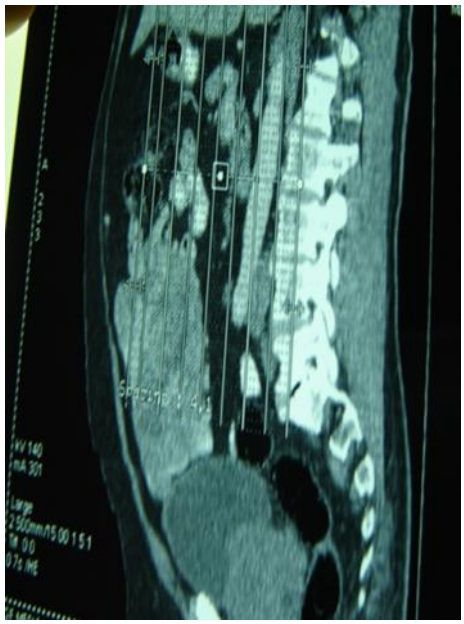

Fig 5

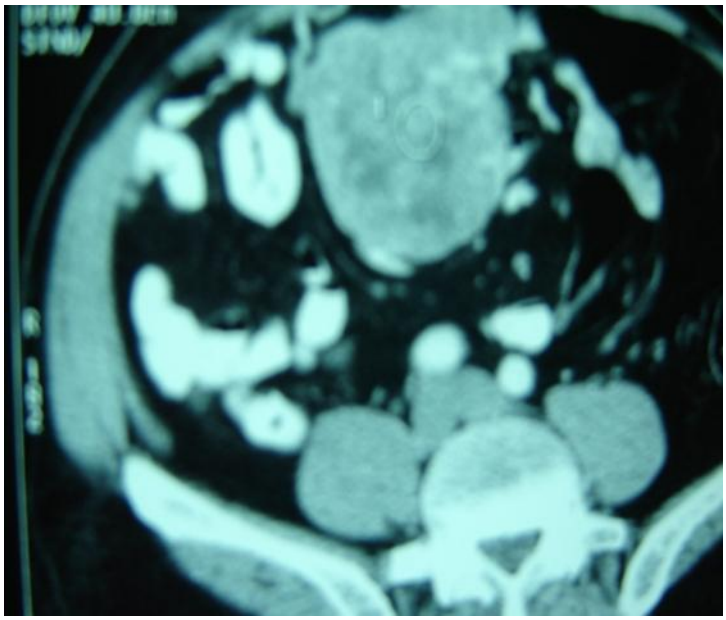

Fig 6

Fig 7, 8, 9, 10: surgical specimen showing the ileal mass

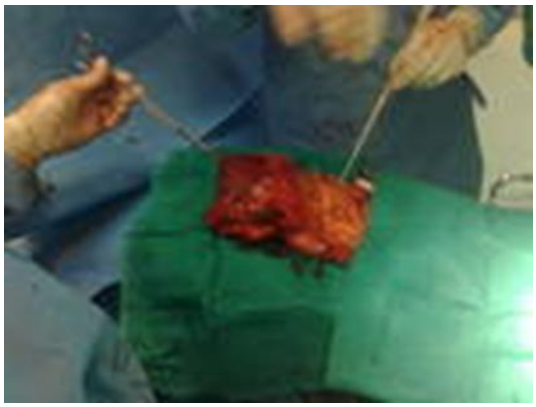

Fig 7

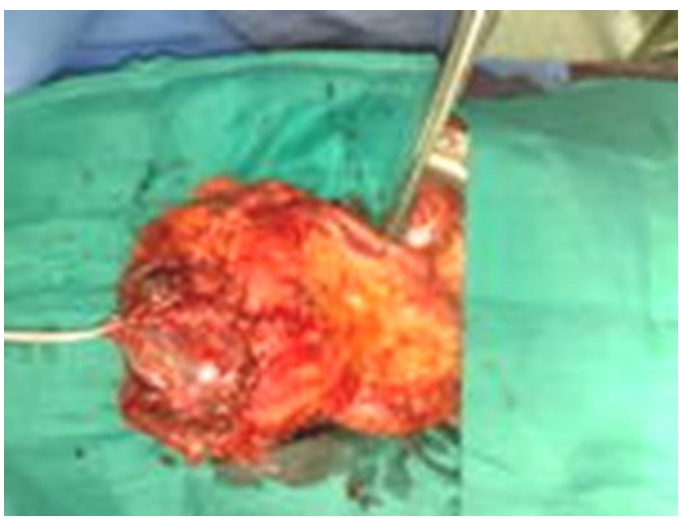

Fig 8

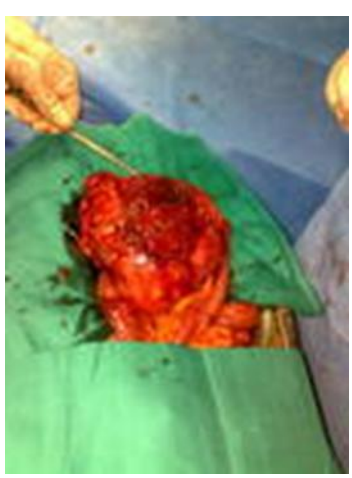

Fig 9

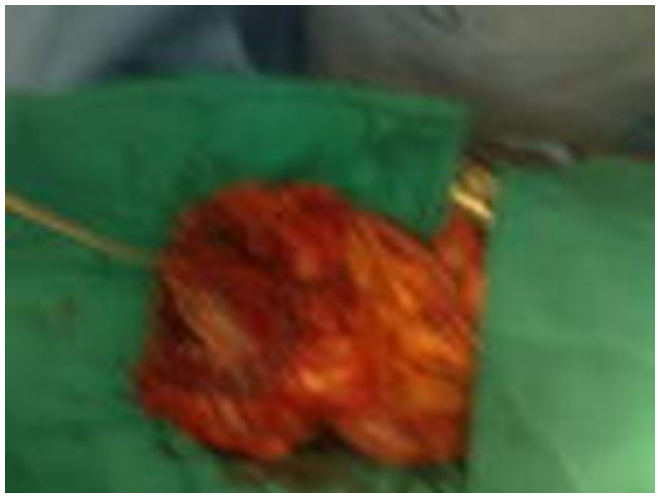

Fig 10

Fig 11,12,13,14,15,16,17,18 : The anatomopathological examination of the surgical specimen showed a proliferation of epithelioid cells, with a mitotic index of 1 mitosis per 50 fields.

Immunohistochemistry revealed strong positivity of tumor cells for CD117, while anti-CD34, anti-desmin, anti-HHF35 and anti-AE1/AE3 markers were negative.

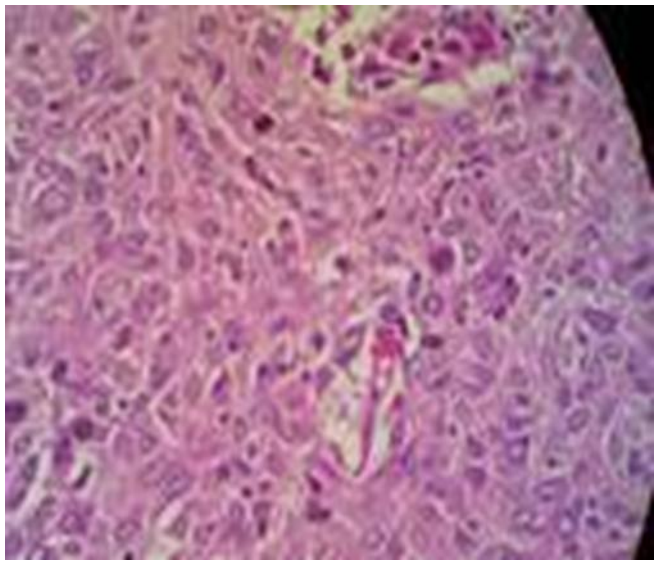

Fig 11: HE GX40 coloring

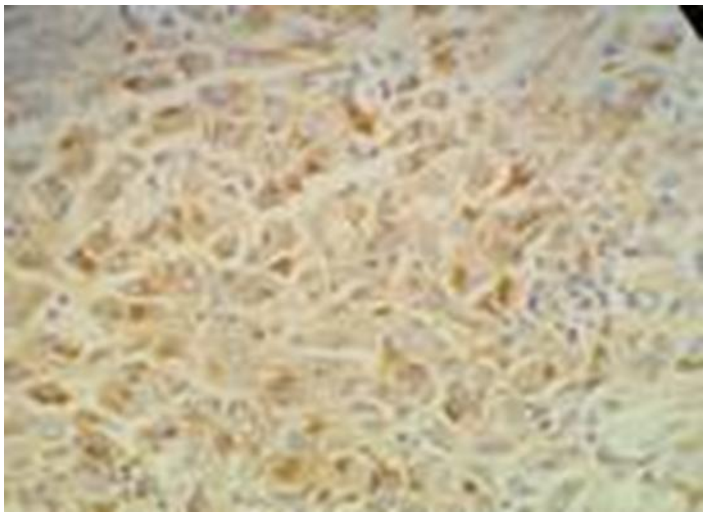

Fig 12

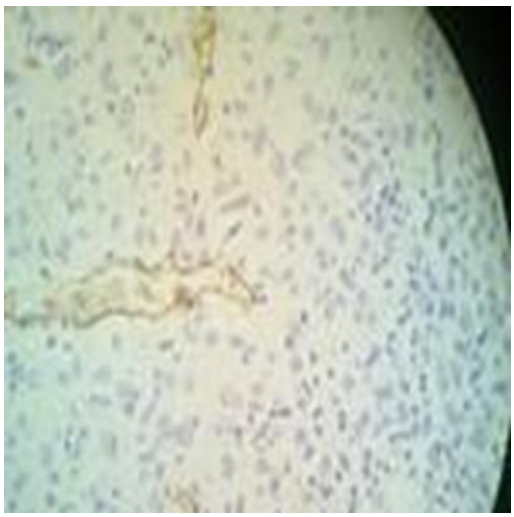

Fig 13

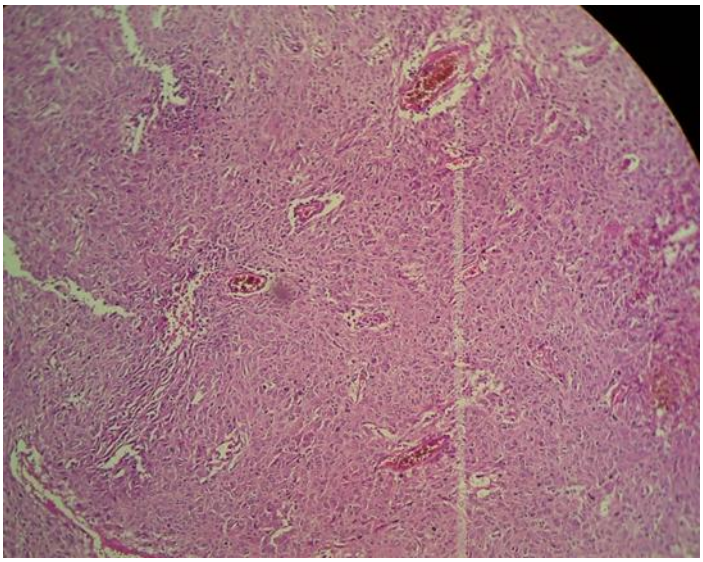

Fig 14

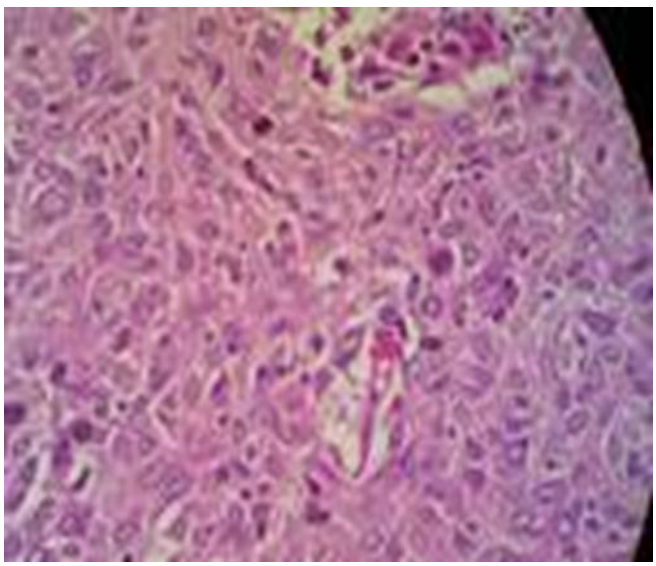

Fig 15

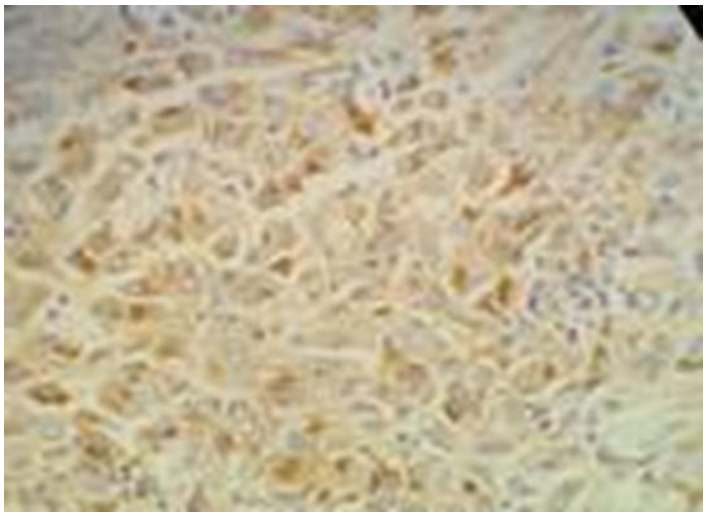

Fig 16: Immunohistochemistry (IHC): expression of antiCD117 antibody (C KIT) by tumor cells (GX40)

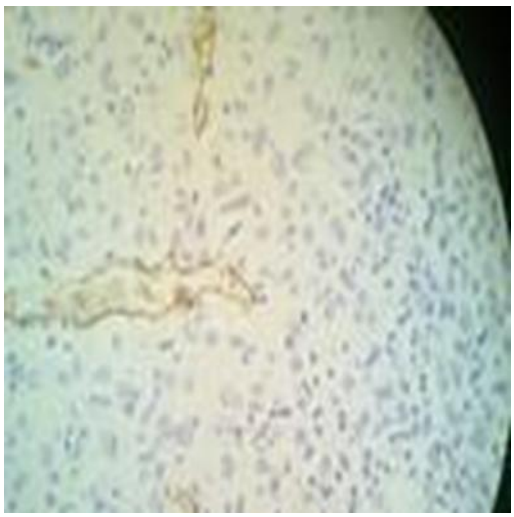

Fig 17: Absence of tumor cell labeling by anti-CD34(GX40)

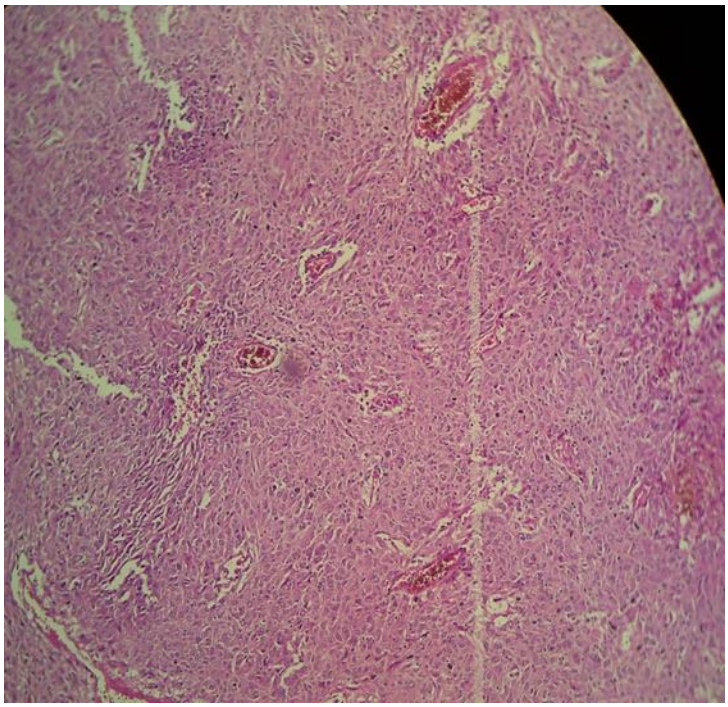

Fig 18: Coloration HE GX 20 ;proliferation tumorale

Discussion

Meckel's diverticulum (MD) is the most common congenital anomaly of the digestive tract, with an incidence in the general population ranging from 0.3 to 4% [1, 2, 6].

Primary tumors arising from a DM are rare, representing approximately 1% of diagnosed DMs. Among them, only a minority are malignant[7,8]. Carcinoid tumors are the most common, representing 33 to 44% of all DM tumors[9 ,10,11]. Leiomyosarcomas and adenocarcinomas are less common, representing 18 to 25% and 12 to 16% of cases respectively[9,12,13,14]. In the English-language literature, about sixty leiomyosarcomas or fibrosarcomas of DM have been reported, but only two observations of GIST confirmed by positive CD117 immunostaining have been published [15].

The "giant" appearance of Meckel's diverticulum is rare, observed in only 0.5% of cases [2]. In our patient, it was a giant DM measuring approximately 15 × 10 cm.

Although GISTs are rare, their annual incidence in France is approximately 900 new cases [1]. They represent 0.1 to 1% of gastrointestinal cancers. The term "GIST" was first used in 1983 by Mazur and Clark [11]. These connective tissue tumors are nevertheless the most common tumors of the digestive tract. They can

develop throughout the digestive tract, with a predilection for the stomach (approximately 60% of cases) and the small intestine (20 to 30%). They are more rarely located in the colon (10%), the rectum or the perianal region (< 5%), and exceptionally in the esophagus, mesentery or appendix (< 1%) [1, 11]. The DM therefore constitutes an exceptional site for the development of mesenchymal tumors, particularly GISTs [16].

DM GISTs present clinically with hemorrhage, perforation, appendiceal syndrome, or abdominal pain related to pelvic tumor syndrome. Most DM tumors are discovered incidentally intraoperatively during ileal exploration [14]. However, the prevalence of DM GISTs diagnosed in this way is unknown [1, 14, 16]. It is exceptional for Meckel's diverticulum to be palpable [2], but in our patient, a palpable mass was present.

Selective arteriography of the superior mesenteric artery allows visualization of an omphalomesenteric artery in 50% of cases [2,14,17]. It was not performed in our patient. 99mTc-pertechnetate scintigraphy remains the most effective method for detecting gastric mucosal heterotopia, with a diagnostic success rate approaching 80% according to some series [2,14].

The unprepared abdomen has very limited diagnostic value. It may reveal water opacity, as was the case in our patient[14]. It may also show tumor calcifications, especially in the case of leiomyosarcomas, and anfractuous lucency [7,18].

Abdominal ultrasound and computed tomography (CT) usually show a large, polylobed, homogeneous or heterogeneous tissue mass, as observed in our patient [7].

The differential diagnosis with other tumors expressing c-Kit (small cell lung carcinoma, adenoid cystic carcinoma, mastocytosis, melanoma, testicular seminomas) does not generally arise, because the clinical context and the morphological appearance are different [2,6]. Only a digestive location of a melanoma can pose a real diagnostic problem [6]. The morphological appearance can then be close to TSD. A strong positivity to PS100 (weakly expressed or absent in GIST) points towards the diagnosis of melanoma [6]. Treatment is based above all on complete surgical excision of the tumor [14]. Lymph node dissection is not systematic [9,14]. The 5-year survival rate is approximately 40%, but incomplete surgery reduces this rate to less than 10 % [1,14]. The recurrence rate is approximately 40%, occurring within a median of 2 years. This risk is mainly linked to the size of the tumor and its mitotic index [1].

Radiotherapy and chemotherapy are not very effective. A new molecule, STI 571 (Glivec®), a tyrosine kinase inhibitor, has demonstrated efficacy in recurrent aggressive tumors with metastatic locations [2, 6].

Conclusion

This observation illustrates the progress made in recent years in the diagnosis of GISTs, as well as the possibility, although rare, of their development from a Meckel diverticulum. In our case, the diagnosis was made intraoperatively and confirmed by pathological analysis of the surgical specimen. In light of the literature and reported cases, the surgeon must always bear in mind the possibility that a Meckel diverticulum is the site of a stromal tumor, the diagnosis of which is based on histological examination.

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