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Hemoperitoneum due to traumatic rupture of a duodenal angiomyolipoma: An unusual way of revealing a rare tumor

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Abstract:*Angiomyolipomas (AML) are mixed mesenchymal tumors. Composed of different proportions of adipose tissue, smooth epithelioid muscle cells and abnormal blood vessels. Tumors most often benign, they sit especially on the level of the kidneys and often associated with tuberous sclerosis (TSC). Extrarenal localization is much rarer, at the level of the duodenum, it remains exceptional. We report the case of an asymptomatic duodenal angiomyolipoma, revealed by a hemoperitoneum after post-traumatic rupture in a 65-year-old woman, who had not previously presented any manifestation of (TSC). Operated as part of the emergency for hemoperitoneum of great abundance, a complete tumor resection was performed. An anatomopathological examination of the operating room confirmed the diagnosis of duodenal AML. No tumor recurrence was observed thereafter.*

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I. Introduction

Angiomyolipomas are described as hamartomas composed of smooth muscles, thick-walled blood vessels and adipose tissue [1]. Belonging to the family of peri-vascular epithelioid cell tumors (PE Comes) [2]. These are relatively rare tumors which occur sporadically in 80% of cases or sometimes as part of a tuberous sclerosis complex (60% to 80% of TSC have AML) [3,4,5]. the most frequently affected sites are the kidneys and the liver (90% of cases) [6,7], with an incidence of renal damage in 0.3 - 3% [8]. In the gastrointestinal tract and apart from the hepatic injury, other localizations have been reported, particularly those concerning the duodenum, appendix, pancreas and stomach [6-9]. AML of the duodenum is rare, three cases have been reported in the literature [10,11,12]. The treatment of AML is based on complete surgical excision. Even if they are considered to be benign tumors, they can be invasive and recurrent. In this case report, we present the observation of an asymptomatic duodenal AML revealed by a hemoperitoneum by post-traumatic tumor rupture in a 65-year-old woman admitted to the emergency ward in a hypovolemic shock table requiring resuscitation measures and an emergency laparotomy.

II. Clinical case:

This is a about 65-year-old women patient with no history of disease and no manifestations of tuberous sclerosis, who consulted for acute abdominal pain following a fall in height with reception on the right flank. The patient did not report any previous functional signs (vomiting, hematemesis, melena, early satiety, etc.) The clinical examination on admission found a patient with an altered general condition, skin and mucous pallor, cooling of the extremities, tachycardia, tension arterial at 60 / 30mmHg and dyspnea in favor of hypovolemic shock. The abdomen was generally tender and tense. Biologically there was anemia at 7.2 g / dl with coagulation disorder TP equal to 40% but without associated thrombocytopenia. After the implementation of resuscitation measures, the abdominal computed tomography had revealed an intraperitoneal effusion of average abundance of hematic density with integrity of the large arterial trunks, associated with a tissue formation of 60x30 mm para duodenale ruptured of probable angiomyolipomatous nature (Figure1).

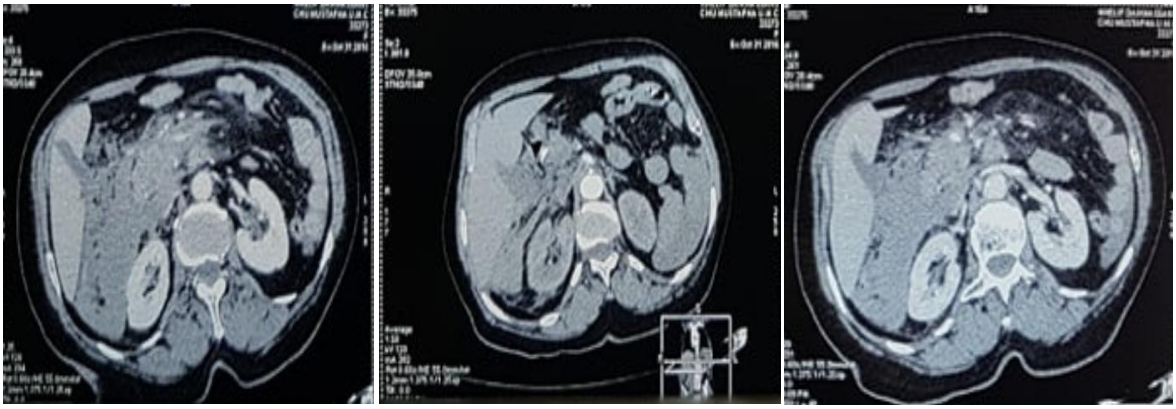

Figure 1: *Hemorrhagic appearance in scanography imagery*

The laparotomy performed had confirmed the abundant hemoperitoneum and the presence of a 75 mm long axis exoduminal duodenal (D1) tumor formation, ruptured and hemorrhagic. We proceed to a complete excision of the mass with hemostasis and contact drainage. The follow-up operations were simple with resumption of feeding on D2 and removal of the drains on D5. The patient was discharged on D10 postoperatively. The pathology examination confirmed the diagnosis of duodenal angiomyolipoma. Follow-up up to 42 months remains satisfactory and no recurrence has been observed.

III. Discussion:

AML is defined histologically as benign mesenchymal hamartomas. Described for the first time in 1951 by Morgan in the kidneys [4], other localizations have been reported since, notably in the mediastinum, the retroperitoneum, the abdominal wall, the heart, the lungs, the stomach, the spleen, bones, transverse colon, vagina, upper lip, nasal cavity, spinal cord, parotid glands and skin [1, 13,14]. Duodenal involvement is exceptional, it is often expressed clinically by satiety, melena, hematemesis, anemia, general weakness, vomiting and abdominal pain [10, 11,12]. In our observation, no previous symptomatology was reported by the patient, the diagnosis having been made in a traumatic context.

The preoperative diagnosis of extrarenal AML is not easy, however, computed tomography and magnetic resonance imaging could help with the diagnosis, by describing lesions with a fatty contingent with abnormal vascularization [10, 15]. Our observation seems to correlate with data from the literature. The presumed CT diagnosis of duodenal AML was confirmed by the anatomopathological study of the operating room. Complete surgical resection is the treatment of choice for tumors larger than 4 cm in diameter [10]. Inadequate resection could be the cause of recurrence [10,16]. We report the case of a duodenal AML ruptured in the peritoneal cavity whose postoperative follow-up at 48 months finds no radiologically detectable tumor recurrence (Figure 2)

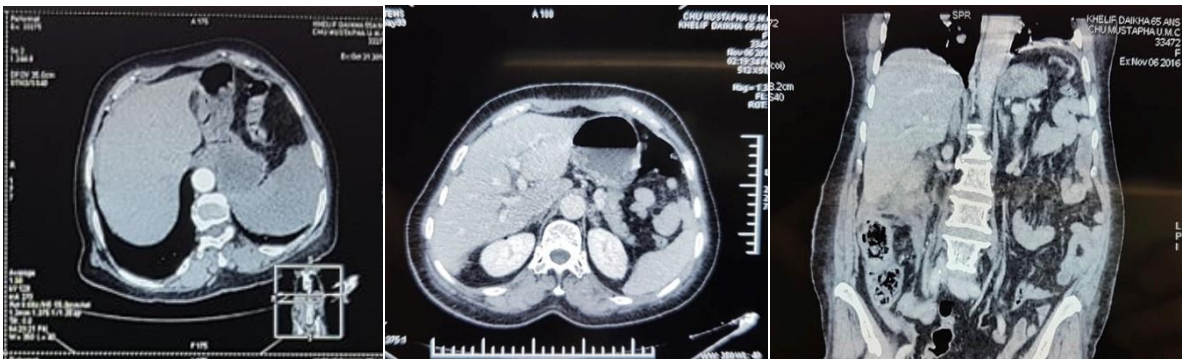

Figure 2: *CT scan after 48 months*

Histologically, AML is made up of different proportions of vascular tissue, smooth muscle and fat quotas. However, a large component of epithelioid cells could include AML in the group of PEComes (Tumors of peri-vascular epithelioid cells) [2]. No malignancy criterion has been observed to date on digestive AML, positive immunoreactivity for α-SMA and HMB-45 is typical of AML and can be used to eliminate differential diagnoses, such as angiolipomas, angio -leiomyomas, liposarcomas and leiomyosarcomas [14,16]. Duodenal LMA has demonstrated focal or zero immunoreactivity for HMB-45 in spindle cells [10]. Toye and Czarnecki [11] described the first case of duodenal AML in a 60-year-old woman with anemia, satiety and a duodenal

mass of 36 × 36 mm. De Padua [12] reported another case in a 66-year-old man with asthenia, severe anemia, melena and a duodenal mass of 40 × 40 mm. Wang Y [10], presented the case of a young 22-year-old patient suffering from early satiety, intermittent abdominal pain, vomiting and several duodenal polypoid formations. Unlike the three cases described, our 65-year-old patient reported no symptoms, LMA presented as an incidentaloma revealed by a post traumatic hemorrhagic rupture. In the majority of cases, AML are sporadic (80%), sometimes (20%), they are associated with TSC or lymphangioleiomyomatose [4,17], none of the four published cases was associated with sclerosis tuberose. To date, no therapy has proven its worth apart from surgical tumor resections [18] for which no recurrence has been observed.

IV. Conclusion

Duodenal angiomyolipoma is a rare benign tumor, which can be the cause of intraperitoneal bleeding by post-traumatic rupture causing hypovolemic shock, which can compromise life prognosis. To date duodenal angiomyolipoma is considered a benign tumor and complete surgical excision remains the treatment of choice. However, a better understanding of this pathology by studying new cases remains essential in order to better understand the clinical pathological aspects and the evolutionary potential of these tumors

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K. HAIL, et. al. "Hemoperitoneum due to traumatic rupture of a duodenal angiomyolipoma: An unusual way of revealing a rare tumor." *IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)*, 19(6), 2020, pp. 21-23.