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Vaterian Ampulloma a Rare Digestive Tumor: Two Cases Report and Review of Literature

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Abstract

In Burkina Faso, the diagnosis of vaterian ampullama is infrequent and late due to the rarity of the tumor but also the absence of lateroscopes during upper digestive endoscopies. The objective of this work was to report two cases of vaterian ampluloma, and their evolution under treatment, in our context. We report two cases of unusual discovery of a Vaterian ampulloma complicated by cholestasis by bile duct obstruction in two patients at the University Hospital of Bogodogo. Their management is difficult in our context due to late diagnosis and the absence of certain interventional endoscopy procedures.

Keywords

Adenomyoma, Ampulla of Vater, Cholestasis Syndrome, Ouagadougou

1. Introduction

Ampullary tumors are rare and represent only 0.6 to 0.8% of all digestive cancers [1]. Adenomyoma of the ampulla of Vater is a rare and benign lesion of the bili-opancreatic junction, discovered in 70% of cases at the degenerated stage [2]. This type of tumor is usually diagnosed between the ages of 50 and 70, with a slight male predominance [1]. Very often asymptomatic for a long time, the diagnosis is frequently fortuitous on the basis of a cephalic duodeno-pancreatectomy [3]. Sometimes it can cause cholestatic jaundice and cause discussion of lithiasis of the main bile duct [4]. Abdominal computed tomography (CT) and Magnetic Resonance Imaging (MRI) can be used to suggest the diagnosis of biductal dilation of the bile ducts and the Wirsung duct with an obstacle of the vascular site, while

endoscopy ultrasound is used to assess locoregional invasion and obtain a pre-therapeutic map [5]. Upper gastrointestinal endoscopy makes a substantial contribution to this diagnosis through the performance of biopsies with anatomical pathological studies. However, the fear of cholestatic jaundice with dilation of the intra- and extra-hepatic bile ducts is to eliminate cancer of the bilio-digestive crossroads. We report two cases of unusual discovery of a valve ampulloma complicated by cholestasis by obstruction of the main bile duct in two patients at the University Hospital of Bogodogo.

2. Observation 1

This was a 48-year-old patient with a cardiovascular risk factor of high blood pressure on amlodipine for 03 years and type 2 diabetes followed by hypoglycemic sulfonamide for 03 years. She was admitted for conjunctival jaundice associated with dark urine and vomiting that had been evolving for three weeks. At the interrogation, no history of biliary disease was found. The physical examination revealed pain in the right hypochondrium, a clinical cholestasis syndrome with flamboyant jaundice but without pruritus, a syndrome of alteration of the general condition made up of anorexia, asthenia and thinness (BMI = 17 Kg/m^2), as well as a fever of 38.7˚C. Upper digestive fibroscopy noted erosive antral gastropathy and ulcerative lesions of the papilla that could suggest a Vaterian ampuloma. On abdominal CT scan, there was dilation of the intra- and extra-hepatic bile ducts without formal visualization of a mass of the head of the pancreas (**Figure 1**). The bili-MRI found a suspicious tissue mass of the head of the pancreas responsible for a significant dilation of the intra- and extra-hepatic bile ducts and moderate dilation of the main pancreatic duct formerly known as the Wirsung duct (**Figure 2**). On abdominal CT scan, there was dilation of the intra- and extra-hepatic bile ducts without formal visualization of a mass of the head of the pancreas (**Figure 1**). The bili-MRI found a suspicious tissue mass of the head of the pancreas responsible for a significant dilation of the intra- and extra-hepatic bile ducts and

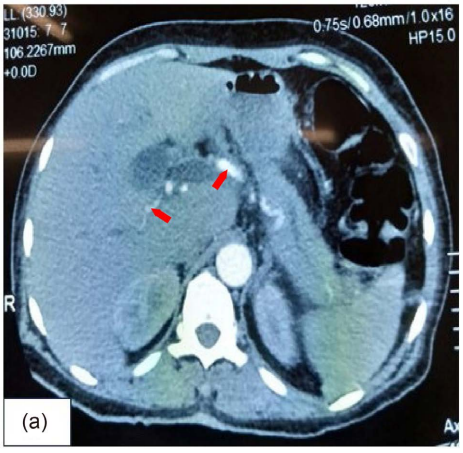

Figure 1. Injected abdominal computed tomography showing dilation of the intra- and extra-hepatic bile ducts (patient number 1).

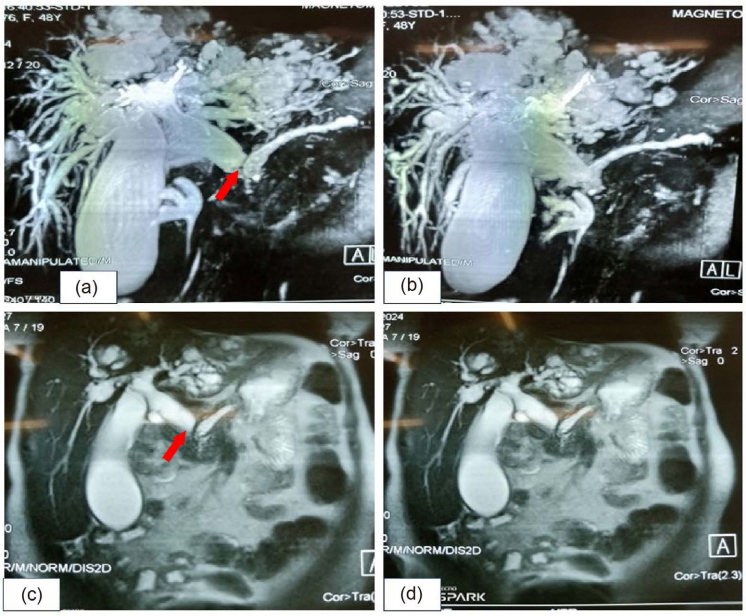

Figure 2. Bili-MRI showing a tissue mass of the biliodigestive junction responsible for a significant dilation of the intra- and extra-hepatic bile ducts and a moderate dilation of the main pancreatic duct suggestive of a Vater's ampulloma (patient number 1).

moderate dilation of the main pancreatic duct formerly known as the Wirsung duct (**Figure 2**).

On the biological level, there was hyperleukocytosis (21.6 × 109/L) predominantly polynuclear neutrophils, hypochromic microcytic anaemia at 8g/dL, hepatic cytolysis (AST = 60 IU/L; ALT = 90 IU/L) and biological cholestasis (total bilirubin = 264 µmol/L; direct bilirubin at 256 µmol/L), the prothrombin level was 60%. Angiocholitis has been suspected in the presence of Charcot's triad (onset of pain, then fever and then jaundice) and persistent pain beyond 6 hours. The diagnosis of a vaterian ampulloma complicated by cholestasis and angiocholitis was retained. A double antibiotic therapy combining third-generation cephalosporins with imidazoles allowed an amendment of the infectious syndrome after 07 days. The bilio-digestive bypass was indicated but could not be performed due to the patient's financial difficulties and poor nutritional status (BMI = 16 Kg/m2) exposing the patient to the risk of postoperative complications. The patient was then discharged from the hospital at the request of the family and lost to follow-up.

3. Observation 2

An 85-year-old patient was admitted for hyperthermia that had been evolving for two weeks. She had high blood pressure discovered for 5 years without follow-up without any other particular pathological history. The physical examination found: a syndrome of alteration of the general condition consisting of asthenia, anorexia and unquantified weight loss, a clinical cholestasis syndrome consisting of jaundice, colored urine and discolored stools, a pulmonary condensation syn-

drome with a wet cough and bilateral crackling rattles as well as hyperthermia at 38.5˚C. At the paraclinical level, chest X-ray made it possible to objectify bilateral opacities at the two pulmonary bases, suggesting bilateral basal lung disease, and the haemogram revealed hyperleukocytosis (13.6 × 109/L) with a predominance of neutrophils (11 × 109/L) and hypochromic microcytic anaemia at 9 g/dL. The other laboratory tests noted conjugated bilirubin cholestasis (direct bilirubin = 230 µmol/L and total bilirubin = 300 µmol/L), moderate hepatic cytolysis (AST= 240 IU/L, ALT=154 IU/L), prothrombin level was 55%, blood glucose (4 mmol/L) and normal serum creatinine. The complete blood ionogram also found no abnormalities. Magnetic Resonance Imaging of the bile ducts (Bili-MRI) revealed a suspicious nodular signal thickening of the vasterial ampulla measured at 14x13 mm with bile duct and Wirsung ectasia (**Figure 3**). The pancreas and gallbladder were normal in appearance without a mass or vesicular suldge syndrome. There was no vascular thrombosis, no intra-abdominal lymphadenopathy, no intraperitoneal effusion. We have retained the diagnosis of a vaterian blister complicated by cholestasis by obstruction on the biliary and pancreatic tracts associated with community-acquired pneumonia. Treatment with cholestyramine and amoxicillin + clavulanic acid was instituted. The evolution was unfavorable towards the patient's death after one week due to multiple organ failure.

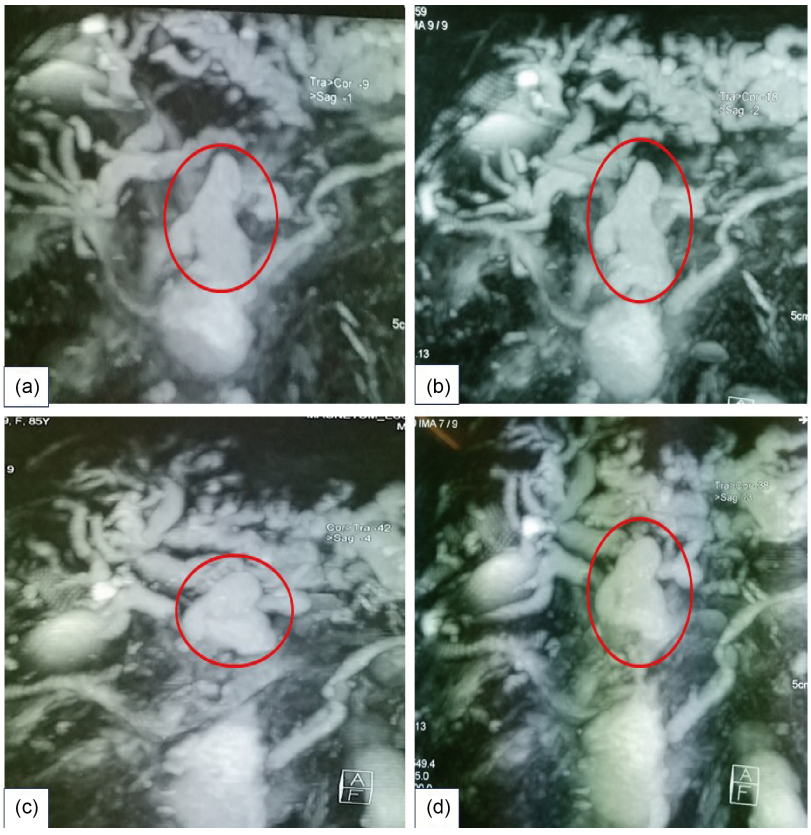

Figure 3. Bili-MRI showing a suspicious nodular signal thickening of the vaterian ampulla measured at 14 × 13 mm with bile duct ectasia and the main pancreatic duct (Wirsung) suggestive of a vaterian ampulloma (patient number 2).

4. Discussion

The Vaterian ampulloma was described for the first time in 1913, by Bravet, as a neoformation at the level of the ampulla of Vater. Since then, several other locations of this tumor have been described on the hepatobiliary and intestinal tracts. Vaterian ampullomas have been described in the majority of cases in the gallbladder, but rare cases are reported elsewhere in the gastrointestinal tract, including the stomach, small intestine, bile ducts and ampullary region [6]. Localization at the level of the main bile duct appears rare, but causes major clinical disturbances such as chronic cholestasis with all its consequences. In addition, this pathology, although benign, is a stress for both the patient and the medical profession, since the fear remains cancer and the differential diagnoses are then made with cholangiocarcinoma and malignant tumors of the biliopancreatic junction [2].

Although rare, the vaterial ampulloma can be revealed by a clinical and biological cholestasis picture in the event of obstruction of the main bile duct, as is the case in our two patients. In these symptomatic forms, jaundice dominates the clinical picture. Other non-specific clinical signs, such as asthenia, dyspepsia, and weight loss, were our patients' complaints. Indeed, in the study conducted by Laurent P et al, jaundice was the most frequent sign, observed in 30 cases (66.7%): 14 cases in the form of naked jaundice (31.1%) while in 16 cases was associated with abdominal pain (35.5%) [2]. The diagnosis may also be incidental during an endoscopy indicated for other reasons. Indeed, the condition can remain asymptomatic for a long time, so diagnoses are made late in our context, often with an alteration in the general condition that does not allow for adequate management. In our context, although upper gastrointestinal endoscopy is available in large cities throughout the country, the absence of a lateroscope makes it difficult to make a detailed assessment of the lesions of the ampulla of Vater. Indeed, duodenoscopy (high-definition lateroscope essential) with multiple biopsies generally allows the precise location and diameter of the lesion, describes the endoluminal or endoampullary development, the degree of stenosis, the presence of ulceration. However, in axial vision, some lesions could be visualized in one of our patients but no biopsy was possible.

Ampullomas are frequently encountered in patients with familial adenomatous polyposis. For these patients, the prevalence of ampullary lesions is estimated to be between 3 and 12% depending on the series [1]. Familial adenomatous polyposis is a genetic disease with autosomal inheritance that is rare in our context but which predisposes to the occurrence of ampulloma. Our patients should have benefited from subsequent colonoscopies to look for these contributing factors.

Sometimes, imaging does not make it possible to decide on budding forms with adenoma and in stenosing forms with cholangiocarcinoma or cancers of the head of the pancreas. Biopsies are of interest here in the confirmation of diagnosis, but biopsy sites are not unanimously accepted by the authors. Hammarstrom et al. propose endoscopic sphincterotomy and multiple biopsies for lesions in the ampullary region, while other authors perform endoscopic ultrasound to allow for

guided and deep biopsy swabs and to assess the locoregional extent of the lesion [6]-[8]. In our context, in the absence of endoscopic ultrasound and interventional endoscopy such as sphincterotomy to facilitate biopsies, especially in ampullary forms, surgery remains the best way both to remove the obstacle and to make a histological study. However, the surgery was not performed on patient number 1 due to the lack of financial means and poor nutritional status (BMI = 16 Kg/m^2). The second patient died before surgical management.

Histologically, ampullary adenomas present two histological subtypes depending on their anatomical position in relation to the ampulla of Vater: the intestinal subtype developed on the duodenal adenoma and the pancreato-biliary subtype developed on the ductal side [1]. The prognosis of the intestinal type of ampullary tumor is much better than that of the excreto-biliary type [9]. When the histological appearance does not easily point to a specific type, it is recommended to carry out an immunohistochemical study: an immunohistological profile cytokeratin 7+/cytokeratin 20− is in favor of a pancreatobiliary origin. A cytokeratin 7−/cytokeratin 20+ immunohistological profile is in favor of an intestinal origin. The KRAS mutation is found in about half of cases and could be a prognostic element [10] [11].

In Burkina Faso, histology and immunohistochemistry are available in the capital, when biopsies are performed. However, the absence of histological examination in our patients should not lead to the rejection of the diagnosis, although it constitutes a major limitation to our two observations. Imaging allowed us to establish a presumptive diagnosis by highlighting an outgrowth of the ampulla of Vater sometimes associated with a dilation of the biliopancreatic tract (**Figures 1-3**). Obtaining histological confirmation post-surgical or post-mortem could provide confirmation of the diagnosis, however, for cultural reasons, autopsies are rarely performed in our context. The management of vaterian ampulomas is surgical and consists of a bilio-digestive diversion, a surgical ampullectomy or a cephalic duedeno-pancreatectomy. The placement of an endoluminal stent to prevent the risk of biliopancreatic junction obstruction is now well established and strongly recommended [12]. However, there are no specific recommendations for performing endoscopic ampullectomy, and some technical details may vary from team to team [13]. Although rare, both complications such as acute pancreatitis and hemorrhage are common and should be postponed surgery if in doubt.

The management of the vaterian ampulloma is often based on a presumptive diagnosis and requires close multidisciplinary collaboration involving hepato-gastroenterologists, radiologists, surgeons, anaesthetists, etc. In general, the prognosis for this condition is poor in Burkina Faso.

5. Conclusion

The discovery of a Vater's ampulloma is a rare but not exceptional eventuality. Despite progress in the field of endoscopic and morphological explorations, histology remains the examination that provides the definitive diagnosis. Their man-

agement is difficult in our context because of the late diagnosis and the inadequacy of the technical platform for adequate patient care.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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