



Sporadic Nonampullary Duodenal Adenoma: A Surgical Case Report and Literature Review

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Abstract

Background: Sporadic nonampullary duodenal adenoma is a rare lesion and is almost exclusively an incidental finding on endoscopy. Its potential for malignant transformation is not negligible; therefore, resection should always be considered. Some techniques for the removal of these lesions have been proposed based on preoperative staging; all of these techniques require extremely qualified expertise because of the lesion site acting as a limiting factor for the resection technique and the duodenum's complex, peculiar anatomy.

Case Summary: We present a case of asymptomatic 61-year-old woman with an endoscopic finding of a sporadic nonampullary duodenal adenoma and high-grade dysplasia. Staging ruled out metastatic disease, but endoscopic ultrasound revealed the involvement of deep duodenal layers. Endoscopic resection was contraindicated due to the site and depth of the lesion. Further, the enhanced recovery after surgery multimodal protocol was applied to the patient. Therefore, the lesion was surgically resected through pancreatoduodenectomy with the preservation of pylorus and pancreatic-gastric anastomosis. The patient progressed with no complications, related or unrelated to the surgical site, and transfusion of blood derivatives was not required. On 3rd day after surgery, the abdominal drain was removed. The patient was discharged on 11th day after surgery with prescribed bland oral diet, no abdominal pain, and normal bowel movements. Histopathological examination of the surgical specimen confirmed high-grade dysplasia without adenocarcinoma, despite invasion of the mucosa, and lesion-free surgical margins.

Conclusion: Sporadic nonampullary duodenal adenoma should be managed by a multidisciplinary team in centers with experience in duodenal lesion resection.

Keywords: Adenoma; Case report; Duodenal neoplasm; Duodenum; Pancreatoduodenectomy

Core Tip

Sporadic duodenal nonampullary adenoma is a rare lesion with potential for malignant transformation. Its conduct is defined by size and cellular differentiation. We present a rare case in a female patient, in which the lesion was small; however, it presented high-degree dysplasia and deep-layer involvement in Endoscopic Ultrasound (EUS), with no possibility of endoscopic resection. A duodenopancreatectomy was performed, and the histopathological examination showed invasion of the mucosa, i.e., EUS staging overestimated the depth. In a multidisciplinary discussion, endoscopists decided not to resect the lesion based on its location; therefore, the conduct could not be different in this case.

Introduction

Sporadic nonampullary duodenal adenoma is a rare lesion and is usually an incidental finding on endoscopy [1,2]. It mostly affects individuals in their sixth and seventh decades of life, and the second duodenal segment is the most common site of the lesion [3]. Adenomas may be sessile structures, slightly more elevated than the normal mucosa [4], and their incidence is the lowest among duodenal polyps [1,4]. Although there are some uncertainties about their natural evolution, they are considered premalignant lesions [5]. According to the Vienna classification, these lesions are divided into adenomas with low-, moderate-, and high-grade dysplasia [5], each requiring specific treatment [5]. Herein, we report of the case of a duodenal adenoma that was surgically resected at a single center with vast experience in upper digestive tract surgery.

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Case Presentation

An asymptomatic 61-year-old woman was referred to the department of upper digestive tract surgery at Hospital da Cidade, in Salvador, State of Bahia, Brazil. She had previously undergone an elective upper digestive endoscopy, in which a sessile elevated lesion with a depressed central area and 1.5-cm greater diameter was detected in the second duodenal segment (Figure 1a, 1b). A biopsy revealed a tubular adenoma with high-grade dysplasia. Colonoscopy was subsequently performed but showed no abnormalities. Staging by magnetic resonance imaging showed no changes in the duodenal wall or abdominal cavity (Figure 2). An Endoscopic Ultrasound (EUS) was performed (Figure 3), which revealed a cleavage plane with the duodenal papilla and involvement of the submucosa of the duodenum, with possible invasion of the muscularis propria.

Multidisciplinary expert consultation

Multidisciplinary discussion is routinely conducted at the authors' digestive surgery department. It involves surgeons, endoscopists, gastroenterologists, oncologists, and radiologists. In this case report, endoscopists contraindicated endoscopic resection, based on EUS results. The surgical team contraindicated Laparoscopic-Endoscopic Cooperative Surgery (LECS) as the site was unfavorable for the use of a stapler.

Final diagnosis

Histological analysis of the surgical specimen (Figures 5-7) showed an ulcerated adenoma with high-grade dysplasia, invasion of

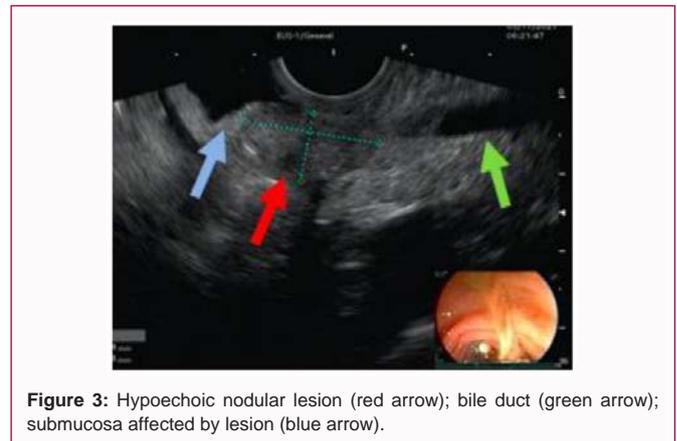


Figure 3: Hypoechoic nodular lesion (red arrow); bile duct (green arrow); submucosa affected by lesion (blue arrow).



Figure 1: A: Sessile elevated lesion, with a depressed central area; destructuring of crypts and the loss of the vascular pattern in the depressed area detected by Narrow-Band Imaging (NBI) (B: Duodenal papilla).

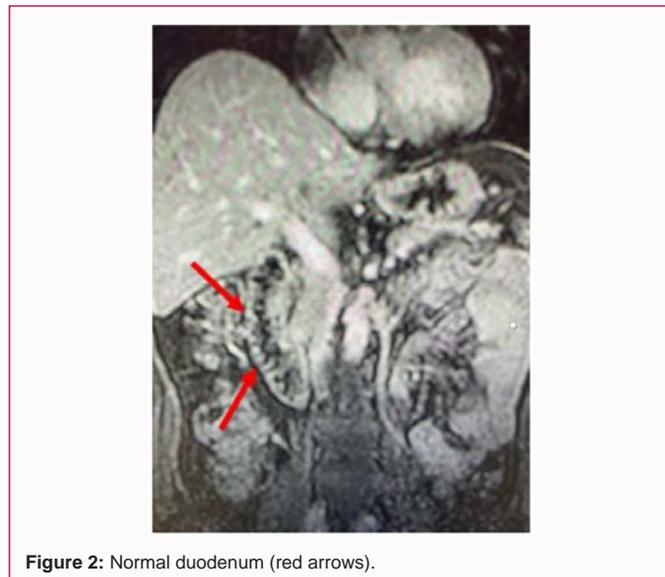


Figure 2: Normal duodenum (red arrows).

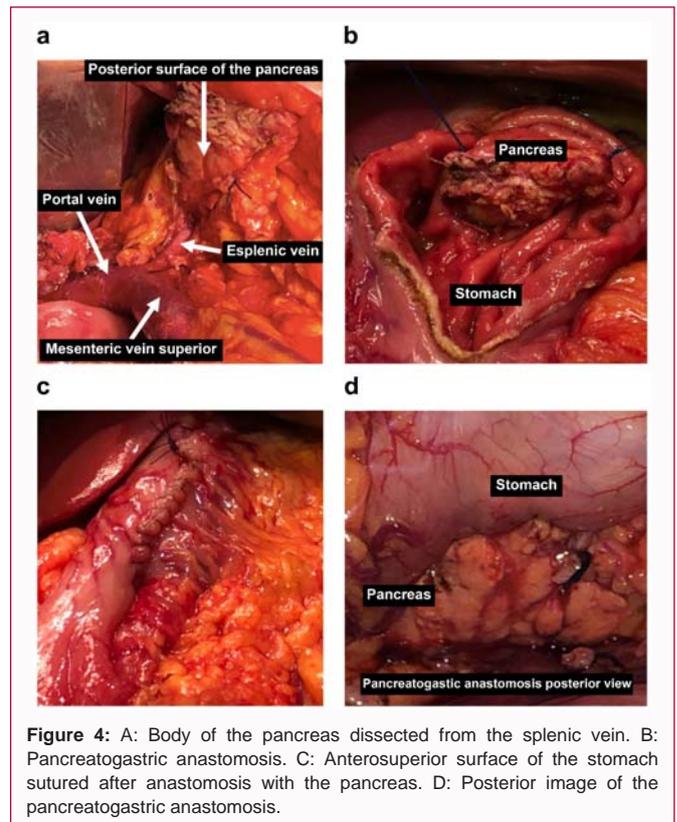


Figure 4: A: Body of the pancreas dissected from the splenic vein. B: Pancreatogastric anastomosis. C: Anterosuperior surface of the stomach sutured after anastomosis with the pancreas. D: Posterior image of the pancreatogastric anastomosis.

the mucosa, and negative surgical margins. There was no readmission.

Treatment

The patient underwent pancreatoduodenectomy with preservation of the pylorus. The enhanced recovery after surgery protocol was used in the surgery, with physical therapy for muscle tonus and nutritional immunomodulating supplementation for 7 days before the surgery. Surgery duration was 210 min, and blood derivatives were not used. A single-loop reconstruction of the digestive tract was performed with lateroterminal choledochojejunal and duodenojejunal anastomoses. For the pancreas, pancreaticogastric anastomosis was performed because of the small diameter of the Wirsung duct (Figure 4). A Trelumina (Freka) feeding tube was placed during the surgery. A suction drain was placed below the anastomoses.

Outcome and follow-up

The patient stayed in the intensive care unit for 24 h, did not receive any opioids, and walked on the 1st Postoperative Day (POD)

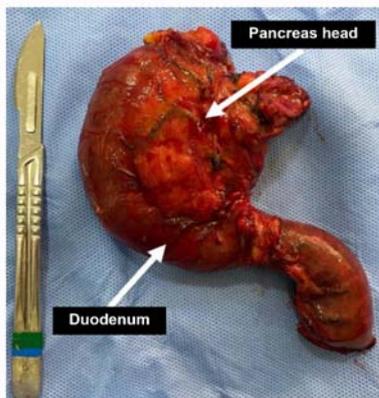


Figure 5: Surgical piece from duodenopancreatectomy.



Figure 6: Sporadic nonampullary duodenal adenoma with central ulceration (blue arrow).

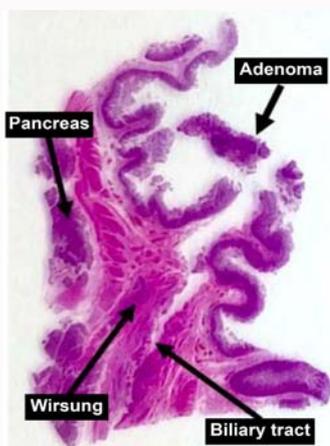


Figure 7: Histological analysis showing an adenoma with high-degree dysplasia.

and started chewing gum. The abdominal drain was removed on the 3rd POD; the drain amylase level was 100 U/L. The patient had grade-1 gastroparesis before the 7th POD. Enteral feeding through Trelumina tube was started on the 3rd POD, and the caloric needs were met on the seventh POD. The patient was discharged on the 11th POD on a soft oral diet, without abdominal pain, and with normal bowel movements. She is followed up on every six months in an outpatient setting and has a good quality of life.

Discussion

Incidence

Sporadic polyps of the duodenum are found in only 5% of

Table 1: Multivariate analysis results of the factors predictive of progression to adenoma in duodenal adenocarcinoma.

	Odds ratio	95% CI	P value
Lesion with high-grade dysplasia	11.07	1.99–61.52	0.006
Lesion \geq 2 cm	8.9	1.42–55.85	0.02

Okada et al. [5]. CI: Confidence Interval.

patients who undergo upper digestive endoscopy [6]. The incidence of sporadic adenoma varied between 0.03% and 6.9% in previous retrospective studies [4,7,8]. Our patient did not have any complaints, and the discovery of the sporadic adenoma was an incidental finding during a check-up, which is consistent with the findings reported by other studies.

Risk of malignant transformation of sporadic nonampullary duodenal adenoma

Okada et al. [5] evaluated a series of 46 patients with sporadic nonampullary duodenal adenoma; of them, 43 had low-grade dysplasia and 3 had high-grade dysplasia. Patients were followed up for >6 months, without treatment and based on biopsy histology and endoscopic observation. Of the 43 patients with low-grade dysplasia, 9 (20.9%) progressed to high-grade dysplasia and 2 to *in situ* carcinoma. Of these 11 patients, 9 underwent lesion resection, and the frequency of adenocarcinoma was high (54.5%). One of the three patients in the high-degree dysplasia group had the lesion removed, which was positive for noninvasive carcinoma, whereas the lesions of the two other patients remained unchanged. A multivariate analysis (Table 1) revealed that lesions with high-grade dysplasia in the first biopsy and diameter of \geq 2 cm could predict the progression to adenocarcinoma. In the present case, the lesion was 1.5 cm in diameter, but high-grade dysplasia was detected in the first biopsy.

Treatment

Conservative: The association of adenoma and carcinoma is relevant, and its incidence varies between 30% and 85% in retrospective studies [9-12]. Therefore, these lesions should be removed by either endoscopy or surgery, depending on the local expertise. Conservative treatment is an exception and may be discussed on a case-by-case basis for patients with low-grade dysplasia for whom resection involves high risk.

Endoscopic resection: In the last decades, with the advent of Endoscopic Submucosal Dissection (ESD), developed in East Asian countries, there have been significant advances in less invasive methods of esophagus, stomach, and colon lesion removal [13]. The complication rates of duodenal lesion removal by this method (even superficial lesions, i.e., mucosal or submucosal) are high [13]. The most frequently reported adverse events are duodenal perforation (incidence rate: 9% to 39%), which can occur during the procedure or later in the first 24 h after dissection and is associated with severe peritonitis [13,14], and bleeding (incidence rate: 18%) [13]. Purely endoscopic methods performed in the duodenum surgery are limited by the lesion size and limited to cases without submucosal invasion.

Association of laparoscopy with endoscopy: Laparoscopic-endoscopic cooperative surgery: LECS is feasible at only specialized centers. The initial indication is similar to that of ESD for the removal of these lesions [14], but it is contraindicated in lesions located near the ampulla of Vater or in cases of submucosal invasion [14]. However, more studies are needed to confirm the safety of this method [14]. Our surgery department has good experience and results with LECS for subepithelial lesions of the stomach. In cases

of duodenal lesions, regardless of the method used, multidisciplinary discussion is fundamental for selecting the best approach. Duodenal LECS, in particular, should be restricted to lesions contralateral to the duodenal papilla and exophytic lesions to minimize the risk of duodenal stenosis due to clamping. In other words, duodenal LECS is an exclusion procedure.

Surgical resection: The endoscopic removal of duodenal lesions has the following limitations: Size of the lesion, site of the lesion, depth of the lesion, and expertise of the endoscopist. In the present case, the lesion was of 1.5 cm, adjacent to the duodenal papilla, and with high-grade dysplasia (Figure 1b). Preoperative staging by EUS showed involvement of the submucosa (Figure 2) and probably of the muscularis propria as well. Surgical resection was thus indicated: Pancreatoduodenectomy with preservation of the pylorus. Histology of the surgical specimen showed invasion of the mucosa, but this information is retrospective; still, it would not have changed our approach had it been known to us at the time. The anatomic location of the lesion (Figure 1b) contraindicated the removal of the lesion by endoscopy or LECS. However, it is important to bear in mind that staging by EUS may overestimate the depth of invasion, in addition to being operator-dependent. In cases with favorable anatomic locations, we suggest performing a resection by ESD or LECS, and subsequently analyzing the specimen to decide whether additional surgical procedures are needed. A review of the literature showed that morbidity associated with this type of surgery varied between 30% and 60% [15-27]. In our department, we stratify the adverse events based on the Clavien–Dindo classification [28]. At our institution, 31 of the 155 patients (20%) had major Clavien–Dindo complications (IV–V) [28]. These quality indicators are essential for the safe indication of a major surgical procedure and for informing patients and relatives about its risks and benefits. Our patient experienced one adverse event: Grade-1 (Clavien–Dindo II) gastroparesis. She had been using a Trelumina feeding tube since the surgery, which allowed aspirating the gastric content and maintaining the flow of enteral feeding, thus minimizing bacterial translocation.

Conclusion

Sporadic nonampullary duodenal adenoma is a lesion with malignant transformation potential, and its treatment should be managed by a multidisciplinary team at centers having experience in the resection of duodenal lesions.

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