CASE REPORT

Adenosquamous Carcinoma of the Duodenum: a Rare Entity

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Abstract Adenosquamous carcinomas (ASC) of the duodenum are extremely rare neoplasms. They have been reported throughout gastrointestinal tract, including the gastroesophageal junction and the anal canal. Only a few cases of ASC of the small bowel and duodenum have been reported in the literature. Here in we report a case ASC of the second part of the duodenum in a 78-year-old man.

Keywords Adenosquamous · Carcinoma · Duodenum

Introduction

Malignant neoplasms arising from the small bowel are relatively rare, accounting for only 2 % of all GI cancers. There are several subtypes among these small intestinal cancers, including adenocarcinoma (40–50 %), carcinoid (30 %), lymphoma (14 %), and sarcoma (11 %). Primary duodenal adenocarcinoma accounts for less than 0.5 % of all gastrointestinal cancers. [1] Gastrointestinal carcinomas with squamous differentiation are adenosquamous carcinoma, adenoacanthoma, and squamous cell carcinoma. [2] ASCs are rare tumours of the bowel

containing both malignant glandular and squamous components. They have been mostly reported in the gastroesophageal junction and the anal canal where glandular and squamous epithelium are normally juxtaposed. [1, 2] They have also been reported in other parts of the gut as oesophagus, stomach and colon. A very few cases of small intestinal ASCs and that to mostly in the jejunum and ileum adenosquamous carcinomas have been reported in the literature [3–6]. We report a case of ASC of the second part of the duodenum in a 78-year-old man.

Case Report

A gentleman aged 78-years presented with passage of black coloured stools. The general physical and systemic examination was unremarkable. Upper gastrointestinal endoscopy revealed a haemorrhagic ulcerative lesion in the second part of the duodenum, Fig. 1. The biopsy from the growth suggested adenocarcinoma with squamoid differentiation, Fig. 2. Contrast enhanced computerised tomography of the whole abdomen and pelvis suggested an ill defined lesion involving the second part of duodenum, predominantly along its medial wall. It measures approximately 3.5 x 2.5 cms and 4 cms in axial and coronal dimensions respectively. Growth was bulging into the duodenal lumen without proximal gastric dilatation and extends into the pancreaticoduodenal groove with loss of fat plane with the uncinate process, suggesting infiltration. Mild prominence of proximal biliary and pancreatic ducts was also noted. Gall bladder exhibits wall calcifications and sludge/calculus in neck. Fat planes with superior mesenteric vein, superior

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mesenteric artery and portal vein are well maintained. No lymphadenopathy or evidence of metastatic disease was seen, Fig. 3. Laboratory investigations including complete blood counts, renal and liver function tests and CA –19-9 were within normal limits. After preoperative preparation he underwent classical pancreaticoduodenectomy and intraoperatively a mass of approximately 3 x 3 cms was found in the second part of duodenum with no evidence of any distant metastasis. Pancreaticoduodenectomy with pancreaticojejunostomy, hepaticojejunostomy and gastrojejunostomy was done. The pathologic diagnosis was adenosquamous carcinoma of the perampullary duodenum, with invasion into the distal common bile duct, pancreas and peripancreatic soft tissue. The tumour comprised of biphasic components, including poorly differentiated adenocarcinoma and moderately differentiated squamous cell carcinoma. The two components were mixed together without a clear border (Fig. 4a). The poorly differentiated adenocarcinoma was composed of signet ring cells containing intracytoplasmic mucin, and the moderately differentiated squamous cell carcinoma composed of keratinized polygonal cells arranged in nests (Fig. 4b). Immunohistochemically, the tumor cells were diffusely positive for cytokeratin 7, and the squamous part was reactive for cytokeratin 5, cytokeratin 6 and p63. The special stain of d-PAS demonstrated the intracytoplasmic mucin in the signet ring cells. The immunophenotype was also compatible with adenosquamous carcinoma. Patient is doing fine at 3 months follow up.

Discussion

Small intestine cancers are relatively uncommon amongst of all the gastrointestinal cancers. The most common types include the adenocarcinoma, lymphoma, carcinoid and sarcoma. ASCs are the extremely rare tumours of the small intestine. They have been reported in various organs, including

Fig. 2 Endoscopic biopsy from the ulcerative growth suggested adenocarcinoma with squamoid differentiation. (H&E X 400)

lung, esophagus, stomach, gallbladder, common bile duct, pancreas, and colorectum. [5–9] Only a few cases, as the present case have been reported to arise from the duodenum. [4, 5] As per the WHO definition, ASC should consist of both adenocarcinoma and squamous cell carcinoma components in different quantity. There may be a transitional area between these two components, or these two components may be separated completely. In the present case, there was no clear demarcation between the adenocarcinoma and squamous cell carcinoma components. As to the origin of these tumors, it has not been clarified yet. Several theories have been proposed to explain the histopathogenesis of both glandular and squamous epithelia in the same tumour. Different hypothesis are that the tumor may be a result of overgrowth from or inclusion of adjoining different epithelium, stimulation of undifferentiated basal cells of the intestine into glandular and squamous epithelia, metaplasia due to chronic irritation and squamous differentiation in a pre-existing adenocarcinoma. [10] The clinical presentation of the adenosquamous tumors can be

Fig. 1 Upper gastrointestinal endoscopy revealed a haemorrhagic ulcerative lesion in the second part of the duodenum

Fig. 3 CECT whole abdomen and pelvis: Ill defined lesion involving the second part of duodenum, predominantly along its medial wall measuring 3.5x2.5x4 cms. Growth is bulging into the duodenal lumen without proximal gastric dilatation and extends into the pancreaticoduodenal groove with loss of fat plane with the uncinate process, suggesting

infiltration. Mild prominence of proximal biliary and pancreatic ducts noted. Gall bladder exhibits wall calcifications and sludge/calculus in neck. Fat planes with superior mesenteric vein, superior mesenteric artery and portal vein are well maintained. No evidence lymphadenopathy or evidence of metastatic disease

varied depending upon the area of origin. Our patient presented with passage of black coloured stools. Diagnosis duodenal ASC established with upper gastrointestinal endoscopy with biopsy, and contrast enhanced computerised tomography of abdomen. The treatment of choice is the surgical resection. Due to limited number of these cases, there is no effective neoadjuvant or adjuvant therapy documented in the literature. On the basis of pre-operative findings, our patient underwent

classical pancreaticoduodenectomy. Pancreatic invasion and lymphovascular emboli were noted additionally in final histopathology, which are the poor prognostic features. In comparison with adenocarcinoma, ASC of the gall bladder and colorectum has shorter doubling time and hence poorer prognosis. [11] The true prognosis of this tumour in duodenum remains to be investigated after more number of cases are reported. We have reported an extremely rare case of duodenal

Fig. 4 **a** Two components mixed together without a clear border. (H&E X 400) **b** Poorly differentiated adenocarcinoma composed of signet ring cells containing intracytoplasmic mucin, and the moderately differentiated squamous cell carcinoma composed of keratinized polygonal cells arranged in nests (H&E X 400)

ASC. The etiopathogenesis, radiological findings, and clinical presentation of the tumour are of interest to clinicians.

Conflict of Interest Nil

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Author Contributions

	Author 1	Author 2
Concepts	✓	✓
Design	✓	✓
Definition of intellectual content	✓	✓
Literature search	✓	✓
Clinical studies	✓	✓
Experimental studies		
Data acquisition	✓	✓
Manuscript preparation	✓	✓
Manuscript editing		✓
Manuscript review	✓	
Guarantor		✓

Compliance with Ethical Standards

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References

1. Kummar S, Ciesielski TE, Fogarasi MC (2002) Management of small bowel adenocarcinoma. *Oncology (Williston Park)* 16: 1364–1369
2. de la Cruz A, de la Cruz E, Sanchez MJ, Ortiz S, Lobato A, Merino E (1993) Adenosquamous carcinoma of the duodenum: an immunohistochemical study. *Pathol Res Pract* 189:481–485
3. Friedman E, Kwan MR, Cummins L (1986) Squamous cell carcinoma of the transverse duodenum. *Gastrointest Endosc* 32:99–101
4. Tocchi A, Mazzoni G, Puma F, Miccini M, Cassini D, Bettelli E, et al. (2003) Adenocarcinoma of the third and fourth portions of the duodenum: results of surgical treatment. *Arch Surg* 138:80–85
5. Mori M, Iwashita A, Enjoji M (1986) Adenosquamous carcinoma of the stomach: a clinicopathologic analysis of 28 cases. *Cancer* 57: 333–339
6. Ercolani G, Nagino M, Sano T, Kamiya J, Kondo S, Kanai M, et al. (1999) Advanced adenosquamous carcinoma of the gallbladder with biliobiliary fistula: an uncommon case treated by hepatopancreatoduodenectomy. *Hepato-Gastroenterology* 46: 1650–1654
7. Kim KW, Kim SH, Kim MA, Lee JM, Park HS, Kim JY, et al. (2009) Adenosquamous carcinoma of the extrahepatic bile duct:

- clinicopathologic and radiologic features. *Abdom Imaging* 34:217–224
8. Lu BC, Wang C, Yu JH, Shen ZH, Yang JH (2014) A huge adenosquamous carcinoma of the pancreas with sarcomatoid change: an unusual case report. *World J Gastroenterol* 20: 16381–16386
 9. Cagir B, Nagy MW, Topham A, Rakinic J, Fry RD (1999) Adenosquamous carcinoma of the colon, rectum, and anus: epidemiology, distribution, and survival characteristics. *Dis Colon Rectum* 42:258–263
 10. Hsueh C, Chen ML, Liao CY, Huang YC, Ho SY, Lee KW, et al. (2011) Adenosquamous carcinoma of the duodenal third portion. *J Radiol Sci* 36:119–123
 11. Charbit A, Malaise P, Tubiana M (1971) Relation between the pathological nature and the growth rate of human tumors. *Eur J Cancer* 7:307–315