

Primary pure squamous cell carcinoma of the duodenum: a case report and review of literature

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Abstract

Primary pure squamous cell carcinomas (SCC) of the duodenum are very uncommon. To the best of our knowledge, only a few cases of SCC of the duodenum exist in the reported literature. Here, we report a case of SCC of the duodenum in northern China. A 45-year-old Chinese Han male patient presented with abdominal pain and weight loss. CT, endoscopic examinations, X-rays, and immunohistochemical markers were used to confirm this rare diagnosis of SCC. We performed a pancreaticoduodenectomy with a curative intention. However, histological examination revealed SCC of the duodenum. Postoperative chemotherapy was started after surgery. To the best of our knowledge, pancreaticoduodenectomy is the preferred form of treatment for carcinoma of the duodenum. This is supplemented with chemotherapy, which can further prolong survival.

Key words: squamous cell carcinoma; duodenal primary tumor

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The duodenum is a unique location for primary squamous cell carcinoma (SCC). Although this tumor was first reported in 1940 in the English language literature [1], there is still little information on this subject with very few articles and case reports. So far, fewer than 10 cases of primary SCC of the duodenum have been reported. Here, we present a case of a 45-year-old male with primary pure SCC of the duodenum.

Case presentation

A 45-year-old Chinese Han man complained of pain in the right upper quadrant and right interscapular region, weight loss of 5 kg, and melena associated with an episode of fever. At the same time, a history of abdominal fullness, discomfort, and jaundice was presented. No evidence of any relevant disease in the family history was found.

Physical examination through abdominal palpation revealed no mass. However, barium swallow exploration confirmed the duodenal mass and fistula (Fig. 1). Abdominal ultrasound scan confirmed a good

hemodynamic state with an epigastric mass measuring 7.5–8 cm, fixed. Abdominal CT scan highlighted a duodenal tumor with encroachment on the duodenal and infiltration of the surroundings (Fig. 2). Endoscopic examination also showed a circumferential tumor reducing the lumen in the junction of the bulb and descending duodenum.

A cephalic duodenopancreatectomy (Whipple procedure) and resection of the hepatic flexure was performed (Fig. 3). The postoperative recovery of the patient was quick and uneventful. Histological examination showed a primary duodenal SCC moderately differentiated (G2). Keratinization was also observed (Fig. 4). To confirm the diagnosis, additional immunohistochemical staining analyses (HCK (+), CK 5/6 (+), and Ki-67 40% (+)) of the duodenal lesion were performed (Fig. 4).

Since patient histology involved SCC, adjuvant treatments of oxaliplatin infusions were elected in combination with oral capecitabine. Patient experienced no complications and is currently following surveillance.

Discussion

SCC of the duodenum is exceedingly rare and only occasional case reports are seen in the medical literature. Our review of the literature (English) revealed only 9 cases of pure SCC of the duodenum (Table 1) [1-7]. Therefore, little information is available as only a few cases present with this diagnosis. Most of the SCC of the duodenum are metastatic tumors from other solid organs such as the cervix, breast, lung, pancreas, and stomach [8].

Since non-specific clinical presentation of SCC can occur, endoscopic examinations are preferred and precise biopsies are recommended in symptomatic patients. CT, ultrasound examinations, X-rays, and immunohistochemical markers were used to confirm this rare diagnosis of primary SCC of the duodenum. This carcinoma is usually localized to the second part of the duodenum [3]. In this case, the imaging investigations did not reveal tumors in other locations and the patient did not present with jaundice. The pathogenesis of SCC of the duodenum is still uncertain. Barnhill and colleagues reported an interesting duodenal tumor with tripartite differentiation into adenocarcinoma, SCC, and neuroendocrine carcinoma. In their case, they speculated that the tumor had arisen from pluripotent duodenal stem cells capable of differentiating into multiple cell types [9]. The tumor in this case may also have arisen from

such pluripotent duodenal stem cells. Amjad A reported a patient with Lynch syndrome who was diagnosed with a SCC. This patient's duodenal SCC showed loss of MSH2 and MSH6 [5], which suggested a pathogenic role for the MSH2 and MSH6 germline mutation in this tumor.

Diagnosis of SCC of the duodenum depended on CT scans, endoscopic examination, etc. Histopathological examination is the most reliable way to distinguish between the primary and metastatic tumors of the gastrointestinal tract. Pathogenetic differences between metastatic and primary tumors also help clinicians in differential diagnosis [8]. Although no other elements were recognized, malignant squamous cells were positive for keratinization

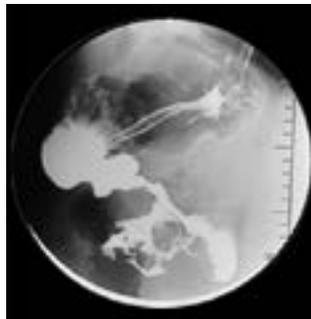


Fig. 1 Barium swallow exploration showing duodenal fistula and widening of the duodenal frame

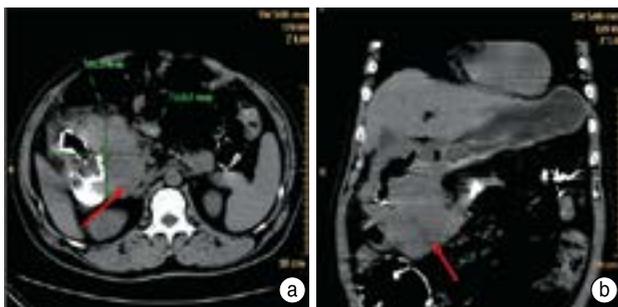


Fig. 2 CT scan. (a) Inhomogeneous duodenal mass (sagittal section); (b) Duodenal tumor (transversal section)

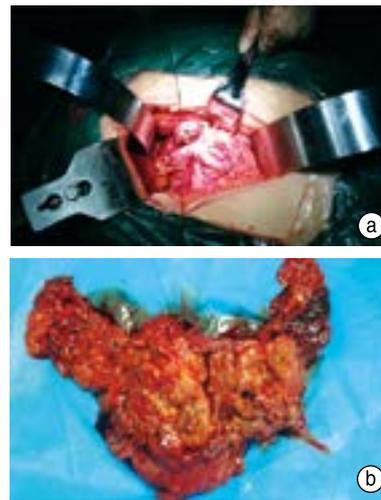


Fig. 3 (a) Intraoperative aspect showing operatory field; (b) Macroscopic view of the mass

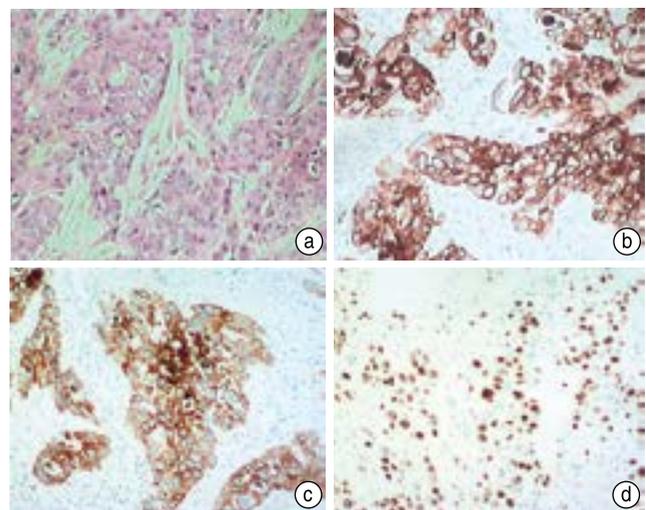


Fig. 4 (a) Pure squamous cell carcinoma of the duodenum with keratinization (HE staining × 200); Immunohistochemistry positive for CK5/6. Photomicrograph of CK5/6 (b), HCK (c), and Ki67(d) staining shows diffused staining of the tumor cells

Table 1 Review of clinical characteristics of patients from published case series of duodenal squamous cell carcinoma

Ref#	Time	Author	Age (years)	Gender	Sites	Treatment	Outcome
1	1986	Friedman E	61	Male	Transverse duodenum	Partial duodenectomy	NR
2	2006	von Delius S	75	Female	Duodenal bulb	NR	NR
3	2009	Terada T	75	Male	Descending part of duodenum	Chemotherapy + radiation	Died 17 months latter
3	2009	Terada T	58	Female	Descending part of duodenum	Chemotherapy + radiation	Died 21 months later
3	2009	Terada T	54	Male	Descending part of duodenum	Surgery	
4	2012	Diffaa A	60	Female	Transverse duodenum	Palliative chemotherapy	Died 1 month later
5	2014	Amjad A	58	Male	Duodenum	Surgery (distal gastrectomy and BillrothII gastrojejunostomy) Chemotherapy (5-fluorouracil+cisplatin)	Surveillance
6	2014	Graur F	47	Female	Descending part of duodenum	Duodenopancreatectomy	NR
7	2015	Battal M	39	Male	Transverse duodenum	Surgery	Surveillance

and intercellular bridges. Immunohistochemistry may also be useful in making the right diagnosis [10]. Similar to this method, CK7, CK20, and TTF-1 levels have also been widely used to distinguish pulmonary carcinomas from gastrointestinal carcinomas [8]. CK5, p63, and p16 were previously selected as immunohistochemical markers to successfully diagnose SCC of the cervix that had metastasized to the duodenum [11]. Distinguishing between the two requires extensive evaluation, including patient clinical history evaluation, histological examination, immunohistochemical analysis, and possibly microarray data analysis.

Conclusion

The optimal treatment and prognosis of SCC of the duodenum is elusive because of the rarity of this disease. Endoscopic and radiological evaluations can prove insufficient to distinguish between benign and malignant tumors. Thus, extensive surgery may be required. In our patient, the tumor was localized to the duodenum with negative margins of resection. Resected lymph nodes were also negative. The metastatic work-up did not show any other primary focus of the disease. Follow-up needs to be more frequent to detect possible early recurrences or distal metastases.

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Ethics approval and consent to participate

The patient described in this case report agreed to the information being used for publication.

Conflicts of interest

The authors indicated no potential conflicts of interest.

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