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Development of adenocarcinoma at the remnant intrapancreatic bile duct 16 years after resection of the choledochal cyst

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Introduction : One of the main reasons to resect choledochal cyst (CC) is prevention of malignant transformation. After CC resection, the remnant intrapancreatic bile is no longer exposed to the enzymatic insult from pancreatic juice activated by the bile, thus the risk of malignant transformation disappears theoretically.

Methods : We present a case showing development of adenocarcinoma at the remnant CC 16 years after CC resection.

Results : This 43 year-old female patient underwent resection of type I CC 16 years before, with leaving a small remnant CC portion because it was located deep within the pancreas close to the anomalous union of pancreatobiliary duct (AUPBD). A small polypoid lesion was identified within the residual cyst, which was proven to be chronic inflammation on endoscopic retrograde cholangiopancreatography (ERCP) biopsy. Four years later, intrahepatic ducts were dilated due to anastomotic stricture of the hepaticojejunostomy, which was resolved after repeated balloon dilatation. On follow-up examination at postoperative 16 years, a mass within the pancreas head was identified, which was proven to be adenocarcinoma on biopsy. Thus pylorus-preserving pancreatoduodenectomy was performed. Pathologic examination showed a 3.5 cm-sized moderately differentiated adenocarcinoma arising from the remnant CC, with no lymph node metastasis. The patient recovered uneventfully and currently undergoes adjuvant chemotherapy.

Conclusions : This case indicates that remnant CC can undergo malignant transformation although pancreatic juice is no longer activated within the remnant cyst. Thus complete resection of CC coupled with very long-term follow-up is emphasized, especially in the era where laparoscopic resection is attempted.

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