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Solitary Metastatic Brain Lesion Prior to Locoregional Recurrence in Hilar Cholangiocarcinoma: An Extremely Rare Case Report

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Abstract

Background

Cholangiocarcinoma is an aggressive malignancy originating from the biliary tract epithelium, is a type of cancer that is difficult to treat and surgical is the only potential curative treatment, although it fails to reduce the recurrence rate. The recurrence rate following primary curative resection is currently at the level of 50–75%. The most common metastatic sites from this malignancy include regional lymph nodes, liver, lungs, and peritoneum. Brain metastases originating from biliary tract cancers, particularly hilar cholangiocarcinoma, are extremely rare events and no standardized therapeutic protocol has been established.

Methods

Patient concerns: A 67-year-old male patient who was diagnosed with hilar cholangiocarcinoma earlier, underwent extended left hepatectomy with caudate lobe resection, common bile duct resection, and hepaticojejunostomy in June 2024 and followed by adjuvant chemotherapy with Gemcitabine and carboplatin, which was completed on October 22, 2024. Routine follow-ups indicated no local recurrence at the surgical site. In mid-2026, the patient presented with a recent history of severe,

recurrent headaches. There were no prominent systemic symptoms such as jaundice or weight loss at the time of presentation. **Diagnosis:** MRI (Magnetic resonance imaging) of the brain revealed a 25 × 24 × 19 mm well defined lobulated heterogeneously enhancing lesion in posterior tectal plate and cerebral aqueduct on right side with non-enhancing necrotic area within. PET-CT (Positron emission tomography - computed tomography) Show FDG (Fluorodeoxyglucose)-avid peripherally enhancing, centrally hypodense lesion in the midbrain and periaqueductal white matter and right half of floor of fourth ventricle and right inferior cerebellar vermis with perifocal edema, measuring 20×20×25 mm. **Intervention and outcomes:** The patient was therefore planned for hippocampal-avoidance whole-brain radiation therapy with simultaneous integrated boost to the metastatic lesion.

Conclusion

Our case highlights an extremely rare presentation of isolated brain metastasis from hilar cholangiocarcinoma occurring approximately two years after curative surgery and adjuvant chemotherapy, despite complete systemic disease control. The combination of controlled primary disease, absence of extracranial metastasis and management using

contemporary hippocampal-avoidance whole Brain Radiation therapy (WBRT) with simultaneous integrated boost (SIB) distinguishes this case from many previously reported cases. It reinforces the importance of maintaining a high index of suspicion for CNS metastasis in patients

presenting with new neurological symptoms after treatment for cholangiocarcinoma and highlights the value of multidisciplinary management using advanced radiation techniques for unresectable lesions.

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