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Oral presentation

Intrahepatic cholangiocellular carcinoma associated with von Meyenburg complexes: case report and review of literature

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Introduction

Intrahepatic cholangiocarcinoma (ICC) arising in bile duct hamartoma (BDH), also referred to as "von Meyenburg complexes", is very rarely seen. Here, we report a case of an ICC in direct association with BDH. The case is discussed and the literature reviewed.

Case report

A 73-year-old obese male patient (BMI 46 kg/m^2) was admitted to hospital due to cardiac failure and septicaemia. Tracheotomy and assisted artificial respiration were performed due to cardiorespiratory failure. A subacute myocardial infarction was diagnosed and chronic pneumonia suspected. Additionally, on ultrasonography, a hepatic mass was revealed. After death, an autopsy was performed and approved the clinical diagnosis. The liver displayed a nodular surface and signs of chronic congestion. There was a scirrhous, gray-white tumor of the right liver lobe (4 × 4 cm). Macroscopically, a primary tumor of the colon and pancreas was excluded. On histology, surprisingly, multiple dilated bile duct, some containing bile plugs, were found. The tumor itself showed small glandular units lined by cuboidal cells with marked atypia. The neoplastic glands were embedded in a dense desmoplastic stroma and invaded the liver parenchyma. Twice a direct transition of normal cuboid bile duct epithelium of the BDH in a neoplastic tubular epithelium was seen. The tumor displayed immunoreactivity for CK7.

Conclusion

Occasionally, ICC was reported in association with multiple BDHs. In this case a clear transition of non-neoplastic bile duct epithelium to the neoplastic invasive glands could be demonstrated. Also, this rare case underlined the importance of autopsy, as the presence of multiple BDHs in this case, as well as the tumor entity, was clinically not known.