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Bile Duct Resection for Eosinophilic Cholangitis

Running head: Surgery for Eosinophilic Cholangitis

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Introduction

Eosinophilic cholangitis (EC) has been labeled a 'sheep in wolves clothing'(1) and 'malignant masquerade'(2) for its benign impersonation of more serious pathologies. The term characterizes a transmural, eosinophil predominant inflammation of the bile ducts (3) within the spectrum of eosinophilic cholangiopathy which also accounts for eosinophilic cholecystitis, the analogous inflammation of the gallbladder (4-6).

We present the case of a 51-year-old female with presumed cholangiocarcinoma causing biliary obstruction undergoing bile duct resection and Roux-en-Y hepaticojejunostomy reconstruction for eosinophilic cholangitis as one of only 30 reported cases in the literature since 1985 to highlight the rarity and diagnostic vigilance required by clinicians.

Case report

A 51-year-old female presented to the Emergency Department with jaundice, right upper quadrant and epigastric pain radiating to the back. This was associated with 12 kilograms of weight loss over a 12-month period. There was no nausea, vomiting, diarrhoea or temporal association with food. There was no personal or family history of malignancy. Clinical examination revealed jaundice and tender epigastric and right upper quadrant regions to deep palpation.

Biochemistry on admission was significant for an eosinophilia of 0.9 x 10^9/L (reference 0.0 – 0.4 x 10^9/L) and a mixed intrahepatic and cholestatic picture of liver function tests; total bilirubin 33µmol/L (reference < 18µmol/L), ALP 902 U/L (reference 35 – 105 U/L), GGT 1697 U/L (reference < 44 U/L), ALT 971 U/L (reference < 33 U/L). An elevated CA 19.9 of 50 kU/L (reference < 37 kU/L) was also recorded on admission. Additional serum levels of lipase, C-reactive protein (CRP), leukocytes, anti-nuclear antibodies, neutrophil cytoplasmic antibodies and IgG subclasses were all within normal limits. Faecal microscopy and

culture were negative for *Campylobacter, Shigella, Strongyloides* and *Toxoplasma sp*.

Abdominal ultrasound demonstrated echogenic material within the gallbladder, cystic and common bile ducts suggestive of biliary obstruction. A triple phase, contrast-enhanced computer tomography (CT) qualified the previous findings in depicting an enhancing mass within the common hepatic duct (CHD) extending to the proximal common bile duct (CBD) with evidence of intra- and extrahepatic biliary duct dilatation (see Figure 1). Magnetic resonance cholangiopancreatography (MRCP) revealed a transition point 1mm below the biliary confluence and a 17mm focal stricture in the bile duct near the insertion of the cystic duct. On coronal True-FISP sequence there was the appearance of a circumferential mass lesion consistent with the low T1, moderately high T2 signal, 17mm mass in the hilum correlating with the stricture location (see Figure 2). Staging positron emission tomography (PET) scan demonstrated focal FDG-uptake corresponding to the contrast-enhancing lesion on CT with no uptake suggestive of locoregional or disseminated disease (see Figure 3). This

radiological evidence was in keeping with the clinical concern for a primary cholangiocarcinoma.

The patient underwent negative staging laparoscopy before proceeding to open exploration. The CBD was dilated proximally and thickened to the CHD with fibrotic tissue. The resected extrahepatic bile duct margin was negative for malignancy on frozen section thus an extra-hepatic bile duct excision with Roux-en-Y reconstruction was completed.

Formal histology of the bile duct was suggestive of a stenosing inflammatory process in keeping with eosinophilic cholangitis. Similar transmural inflammatory features were also evident in sections from the cystic duct and proximal gallbladder.

The patient had an uncomplicated recovery and was discharged day five post-operatively without adjunctive corticosteroid therapy. At two-week follow-up the patient was pain-free with normal biochemistry. An MRCP 8 months post-operatively showed normal intra-hepatic bile

ducts without evidence of disease recurrence. At 18 month follow-up the patient remains well with normal biochemistry.

Discussion

The current case illustrates the need for improved diagnostic rationale for eosinophilic cholangitis given its relatively benign aetiology contrasted with radical management. Presently the overwhelming opinion favours resection in fear of a missed cholangiocarcinoma diagnosis (7). There is a paucity of evidence to support sole clinical and radiological diagnoses however, with Spy-Glass cholangioscopy there is scope for further investigation of indeterminate bile duct lesions with direct visualisation and tissue sampling (8). In 1985 Butler et al. (9) proposed a diagnostic trial of corticosteroids, which has been trialed and reported in a number of subsequent cases (10). Currently there remains no guideline for dosage, timing or measure of response for corticosteroids and as such may be viewed as a higher risk strategy compared to surgery due to the diagnostic uncertainty. Certainly, in the case presented, with clinical features and cardinal radiological findings suggestive of malignancy, there was less harm in treating for the suspected poorest prognosis.

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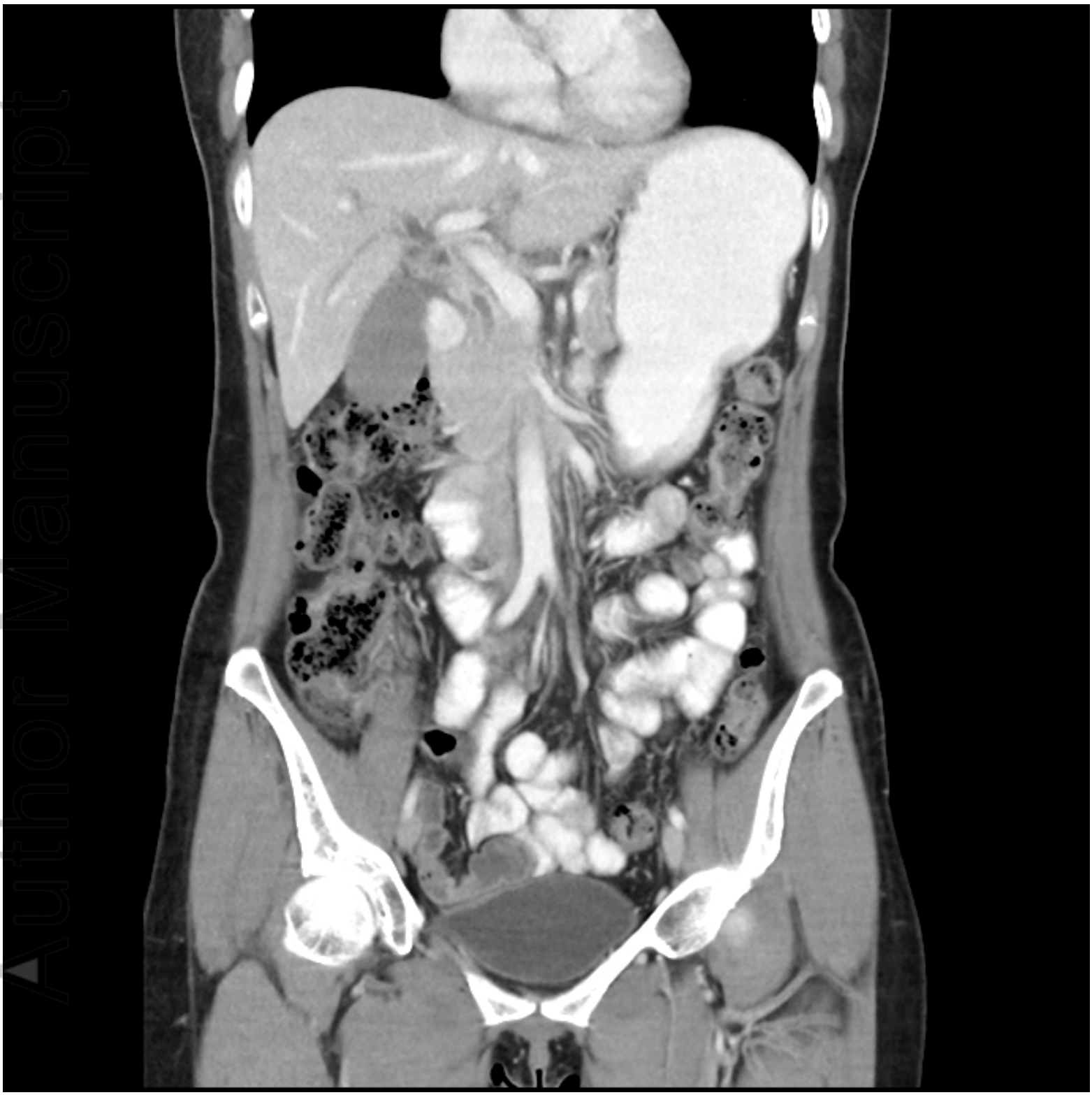
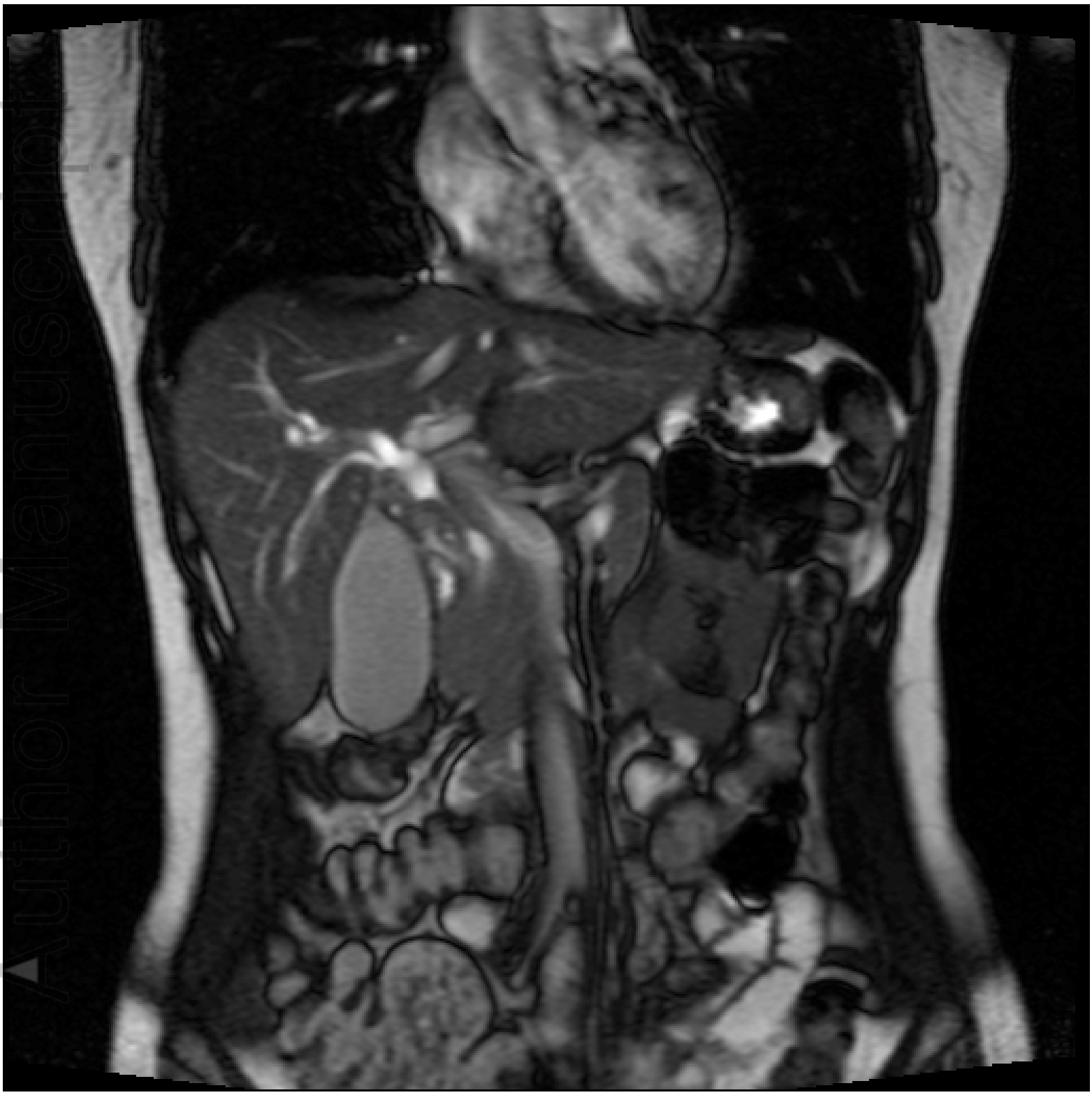


Figure_1.png



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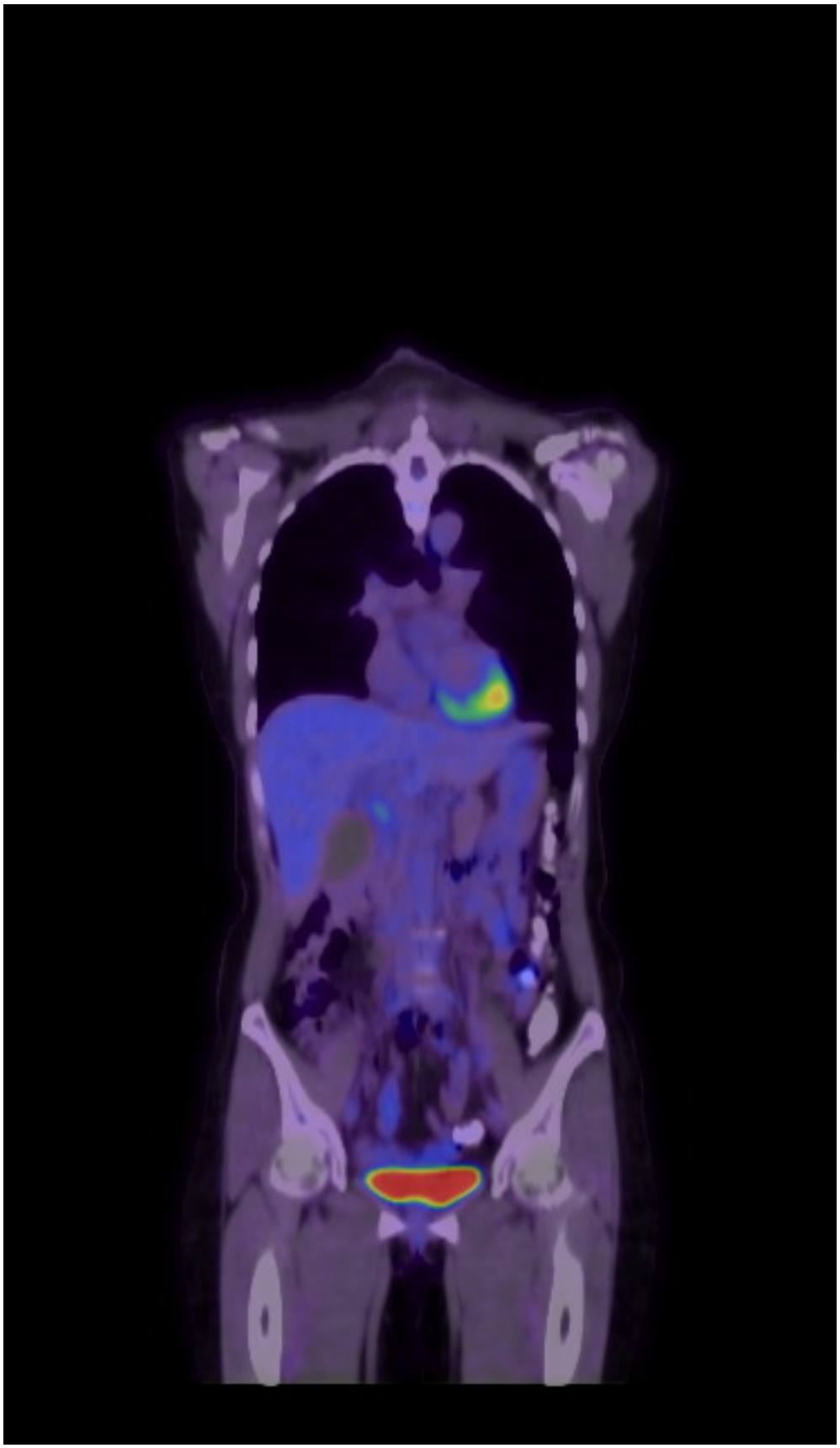

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