CASE REPORT

Digital lesion leading to diagnosis of cholangiocarcinoma

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SUMMARY

Cholangiocarcinomas (CCA) are uncommon malignancies that present late and have a poor prognosis. The patients may remain undiagnosed for many years because of non-specific presentation. CCA metastasises commonly to liver and adjacent lymph nodes. It can rarely metastasise to bone, lung, colon, pancreas, adrenal glands and spleen. Multiple treatment options are available including surgery, chemotherapy, radiotherapy and photodynamic therapy. The tumours have high rate of recurrence and most patients require palliative care. Our patient is a middle-aged man who presented with pain in right ring finger, workup of which revealed digital metastasis from underlying cholangiocarcinoma.

BACKGROUND

Cholangiocarcinomas refer to cancers arising from the epithelium of the intrahepatic, perihilar or distal extrahepatic biliary ducts.¹ Intrahepatic cholangiocarcinomas originate from small peripheral intrahepatic ductules or they may arise from large intrahepatic ducts proximal to the bifurcation of the right and left hepatic ducts.² Intrahepatic cholangiocarcinoma represents less than 10% of all cholangiocarcinoma cases.^{3 4} Clinical

Figure 2 Coronal T2-weighted MRI of the liver revealed a 3.1×3.9×4.6 cm hyperintense mass in inferior segment of right lobe of liver with ascites and splenomegaly.

presentation of CCA is non-specific. Patients with extrahepatic CCA tend to present with symptoms of obstructive jaundice or complicating cholangitis. Patients with intrahepatic masses may present with abdominal pain, malaise, night sweats, weight loss and anorexia.^{5 6} Here, we report an unusual case of intrahepatic CCA in which the patient presented with painful digital metastasis, biopsy and amputation of which led to diagnosis of CCA.

CASE PRESENTATION

This is a middle-aged patient, with progressive swelling of right ring finger, specifically the middle phalanx with inability to flex the proximal interphalangeal joint. The patient had a long-standing history of hepatitis C and cirrhosis of liver. Physical examination revealed a vitally stable, alert and oriented, afebrile male with a swollen and tender middle phalanx of the right ring finger. Patient was unable to bend the proximal interphalangeal joint.

INVESTIGATIONS

Laboratory analysis was significant for mild transaminitis, normal bilirubin, alpha-fetoprotein and carcinoembryonic antigen (CEA) with an elevated CA 19-9 at 57 ng/mL. X-ray of right hand (**figure 1**) revealed a destructive lesion of the distal, proximal and middle phalanx of the right ring finger. MRI of right hand revealed destruction of proximal and

Figure 1 X-ray of hand revealed osteolytic lesion in the proximal portion of middle

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Figure 3 Coronal CT sections revealed osteolytic lesion in 10th rib.

mid portion of middle phalanx and partial destruction of distal end of proximal phalanx of ring finger associated with gadolinium contrast enhancement of the mass. Amputation of the right ring finger was performed. Pathology revealed a moderate to poorly differentiated metastatic adenocarcinoma. Cytokeratin (CK) 7, Cancer Antigen (CA) 19–9, pancytokeratin and S-100 immunostaining was positive. Hepar-1, CDX2, polyclonal CEA, alpha-fetoprotein, CK 20, thyroid transcription factor 1, renal cell carcinoma and prostate-specific antigen were negative. Mucicarmine stain was focally and weakly positive. Though below mentioned differential diagnosis were considered, the histomorphology and results of special stains favoured cholangiocarcinoma. Neuroendocrine differentiation was indicated by positive S-100 staining. MRI of the liver ([figure 2](#)) was

Figure 4 Sagittal CT scan revealed compression fracture of T11 thoracic vertebra.

performed which revealed a 3.1×3.9×4.6 cm hyperintense mass in the inferior segment of right lobe of liver with ascites and moderate splenomegaly. The liver mass revealed heterogeneous enhancement with gadolinium infusion. Bone scan was suggestive of multiple bone metastasis including ribs and vertebrae ([figure 3](#) and [figure 4](#)).

DIFFERENTIAL DIAGNOSIS

Differential diagnoses based on immunostains included hepatocellular carcinoma, intrahepatic and extrahepatic cholangiocarcinoma, lung adenocarcinoma, colonic adenocarcinoma, synovial sarcoma monophasic and carcinoma of unknown primary.

TREATMENT

Chemotherapy with gemcitabine and carboplatin was administered with some stabilisation of disease.

OUTCOME AND FOLLOW-UP

Subsequently, the patient developed massive ascites and became cachectic, leading to hospice referral and death ultimately 6 months later.

DISCUSSION

After lung and liver, the skeletal system is the third most frequent site of metastasis. Almost 80% of bony metastatic disease is accounted for by lung (47%), renal (13%) and breast (12%) malignancies.⁷ Axial skeletal system, which includes the spine, pelvis, skull and proximal femur are the most common sites of osseous metastasis. Metastasis to hands and feet, termed acral metastasis is a rare occurrence (0.007% to 3% of bony metastasis), especially as the initial presentation of an underlying malignancy. In cases of unilateral hand metastasis, higher incidence is noted in the dominant hand, probably aided by increased blood circulation. The middle finger (29%) and thumb (27%) account for most cases. Metastasis to hands is more common than metastasis to feet. Acral metastasis are more common in men.⁸

High index of clinical suspicion is required to suspect and recognise acral metastasis. Besides the prognosis being poor, the diagnosis may be delayed due to the unusual presentation.⁹ Median survival of patients with osseous metastasis is short, usually 6 months' duration.¹⁰ However, recent developments in targeted and immunotherapy will most likely improve survival prognosis. The presenting feature of acral metastasis is usually a swollen, erythematous, warm and tender digit. Differential diagnosis includes osteomyelitis,¹¹ inflammatory or non-inflammatory arthritis and cardiac embolisation to digits.

Acral metastasis, not confirmed radiographically, was clinically diagnosed in 1906, when a patient with breast cancer presented with metacarpal metastasis.¹² In 1907, the first radiographically confirmed case of acral metastasis was reported. Since then few other case reports have been published in literature.

Learning points

- ▶ Cholangiocarcinomas are rare malignancies that can metastasise to bone.
- ▶ In our patient, the diagnosis was made from the unusual site of digital metastasis.
- ▶ Surgical resection is the only potentially curative treatment available.
- ▶ For patients who are unable to undergo surgery, chemotherapy is given to improve progression-free survival.

Treatment involves symptomatic management and pain control. Amputation of digit and radiotherapy have been used for pain relief. Systemic treatment of primary malignancy is initiated in feasible cases.^{13 14}

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Competing interests None declared.

Patient consent Detail has been removed from this case description/these case descriptions to ensure anonymity. The editors and reviewers have seen the detailed information available and are satisfied that the information backs up the case the authors are making.

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