CASE REPORT

Isolated implant metastasis in chest wall due to seeding of transpleurally placed PTBD catheter tract in a case of hilar cholangiocarcinoma

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SUMMARY

Percutaneous transhepatic biliary drainage (PTBD) catheter site metastasis in cases of cholangiocarcinoma is reported sporadically. But it is unusual to see left-sided tumour metastasising to the right PTBD catheter site. Metastasis, in general, has a poor prognosis, but recurrence along the catheter tract in the absence of other systemic diseases can be a different scenario altogether. To date, there is no consensus on the management of this form of metastasis. But carefully selected patients can benefit from aggressive surgical resection. We report a case of a young patient with isolated chest wall metastasis 1 year after resection of left-sided hilar cholangiocarcinoma. The metastasis was resected and, on pathological analysis, was confirmed to be due to implantation of malignant cells along the tract of the PTBD catheter placed via a transpleural route.

BACKGROUND

Since 1973, percutaneous transhepatic biliary drainage (PTBD) has been widely practised for malignant biliary obstruction with diagnostic, therapeutic and palliative intent.^{1,2} While minor complications such as pain (20%), catheter slippage (18%) and catheter block (27%) are common, more serious complications such as bleeding, flaring of cholangitis, biliary peritonitis and sepsis can occur in 7%–10% of cases.^{3,4} Moreover, there has been a growing concern for catheter tract metastases

Figure 2 Axial cut contrast-enhanced CT image of the primary tumour seen as hypodense soft tissue at the primary confluence (white arrow) with bilobar intrahepatic biliary radical dilatation.

which occurs in 0.6%–6% of cases.⁵ Although many surgeons condemn the percutaneous route in favour of endoscopic drainage,⁶ the modality is still of immense value in high and complex biliary obstruction where the endoscopic route can fail.

CASE PRESENTATION

A 41-year-old man presented to our clinic with a painful swelling on his right lower chest wall. On examination, a 5×5 cm hard tender nodule was palpable in the right anterolateral chest wall over the sixth rib and adjacent intercostal spaces (figure 1). The lump was fixed to the chest wall. The overlying skin was freely movable but showed a scar which corroborated with a previous PTBD insertion.

On reviewing his medical records, it was found that he is a follow-up case for type-IIIB hilar cholangiocarcinoma. One year earlier, he had developed painless progressive cholestatic jaundice associated with loss of weight and appetite. His maximum preoperative serum bilirubin and alkaline phosphatase were 16 mg/dL and 425 IU/L, respectively. Cross-sectional imaging revealed bilobar intrahepatic biliary radical dilatation (IHBRD) with a heterogeneous soft tissue mass at the primary confluence (figure 2). The patient developed cholangitis during preoperative evaluation. Repeated endoscopic drainage attempts failed to negotiate the tight hilar stricture. Interventional radiology opinion was sought in desperation as cholangitis

Figure 1 Clinical (A), preoperative (B) and intraoperative (C) images of the chest wall metastasis (black arrow) and its relation with ribs marked with a marker during surgery.

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Figure 3 Follow-up positron emission CT scan showing intensely FDG-avid right chest wall lesion in relation to the sixth rib abutting segment VII of the right lobe of the liver.

worsened. An 8 French PTBD catheter was placed via a transpleural approach into the right ductal system and advanced into the common bile duct. The left duct was drained simultaneously via an epigastric route. On recovery from cholangitis, he underwent left hepatectomy with Roux-en-Y hepaticojejunostomy. Drainage catheters were removed during surgery. The histopathology report showed left ductal cholangiocarcinoma with clear surgical margins. The harvested lymph nodes harboured no metastasis. Following postoperative recovery, he received 45 Gy external beam radiation and three cycles of gemcitabine-based and oxaliplatin-based chemotherapy.

INVESTIGATIONS

A follow-up positron emission CT (PECT) after completion of adjuvant therapy revealed an intensely fluoro deoxy glucose (FDG)-avid (maximum standardised uptake value (SUV_{max}) 10.80) lesion in the subcapsular region of segment VII of the liver extending through the sixth intercostal space into the subcutaneous plane associated with a cystic lesion (figure 3). Clinically, patient was asymptomatic and no lump was palpable. Image-guided Fine needle aspiration cytology (FNAC) was also inconclusive and patient was closely followed up until the lesion became clinically apparent in 2-month time.

A contrast-enhanced CT scan of the abdomen and chest was done at the present follow-up which revealed a heterogeneous soft tissue lesion (3.4×3.3×1.8 cm) in the right anterolateral chest wall extending through the sixth intercostal space and causing indentation on segment VII of the liver (figure 4). A true-cut biopsy confirmed it to be metastatic adenocarcinoma. As there was no clinical or radiological evidence of intra-abdominal and systemic dissemination and the recurrence was exactly at the old PTBD catheter site, a diagnosis of isolated implant metastasis of the catheter tract was made in a multidisciplinary meeting involving surgeons, radiologists and a radiotherapist. A decision of surgical resection via a direct transpleural core down approach was made.

Figure 4 Axial contrast-enhanced CT image showing a heterogeneous cystic lesion in the right anterolateral chest wall.

TREATMENT

During surgery the metastasis was resected en-bloc from the skin down to the liver surface with segments of the sixth and seventh rib, intercostal muscle, parietal pleura and disc of the diaphragm keeping a 2 cm circumferential margin (figures 5 and 6). The diaphragmatic defect was closed using a PTFE patch. No attempt was made to reconstruct the rib cage defect. Soft tissue and skin flaps were mobilised and apposed primarily after placing a chest tube. The patient made an uneventful recovery and was discharged in a satisfactory condition.

OUTCOME AND FOLLOW-UP

Pathological analysis of the resected specimen revealed a cystic nodule (2×2×1.5 cm) in the subcutaneous plane filled with greenish jelly. It had a wall thickness of 0.1 cm. Microscopic examination revealed a cyst wall lined by multilayered pleomorphic tumour cells with vesicular to hyperchromatic nuclei and moderate amount of cytoplasm (figure 7). The surrounding stroma showed desmoplastic reaction with myxoid degeneration and foamy macrophages. The epidermis and dermis were free but the underlying soft tissue, skeletal muscle and diaphragm were invaded by tumour cells. The resected wedge of the liver only showed features of chronic macrovesicular steatohepatitis. Tumour cells showed membranous positivity for CK 7 (figure 7).

Figure 5 Intraoperative image showing en-bloc metastatectomy incorporating skin, soft tissue, rib segments, disc of diaphragm and part of the liver.

Figure 6 En-bloc metastatectomy specimen comprising skin, subcutaneous tissue, rib segments, intercostal muscles, diaphragm and liver tissue.

confirming our previous postulation of catheter tract implant metastasis.

Presently, the patient is asymptomatic and under close follow-up. After six postoperative weeks, a follow-up PECT (figure 8) was done which showed inflammatory changes at the surgical site (SUVmax 5.1). Serum carbohydrate antigen (CA) 19.9 level was 5.0 IU/mL. He was started on oral capecitabine therapy on completion of which PECT and CA 19.9 level will be repeated.

DISCUSSION

Metastasis of biliary and pancreatic tumours along the PTBD catheter tract is a unique phenomenon and less commonly reported than port site metastasis following laparoscopic removal of incidental gall bladder cancers. While the approximate incidence of port site metastasis is around 10%,⁷ catheter

Figure 7 Photomicrograph of the specimen showing (A) irregular glands lined by moderately pleomorphic tumour cells with vesicular chromatin and prominent nucleoli in the deep dermis and (B) a typical neutrophil rich fibroinflammatory stroma (H&E 200 $\times$). (C) Immunostaining of tumour cells showing strong diffuse membranous positivity for CK 7 (200 $\times$).

Figure 8 Positron emission CT image after six postoperative weeks showing no residual disease except inflammatory changes at the surgical site.

tract recurrence is seen in only 0.6%–6% of cases.⁵ The precise mechanism of catheter tract metastasis is unclear but appears analogous to the mechanism for port site metastasis. While venturi effect of pneumoperitoneum along with its immunity lowering capability promotes the seeding of malignant cells at port site, inflammation initiated by surgical trauma favours their proliferation. Given the observation that catheter tract recurrence is more likely with long duration catheters, multiple catheters, repeated manipulations or exchanges and that more than one-third of malignant biliary obstructions are associated with positive bile cytology,⁸ it is postulated that malignant cells in the bile stick to the catheter, puncture the needle or guide wire during manipulation and disseminate along the tract.

In 1982, Oleaga *et al*⁹ reported the first case of cutaneous metastasis in hilar cholangiocarcinoma. Since then, similar cases have been reported anecdotally in the medical literature.^{2 4 5 8 10–16} In a retrospective review of PTBD tract metastasis by Sakata *et al*,⁵ the incidence of catheter tract metastasis was found to be 4.5%. A pooled analysis of all reported cases until 2005 (n=10) found that the median time to detection was 14 months postinsertion, 60% were isolated and 40% had well differentiated tumour histology.⁵ A similar review by Rosich-Medina *et al*¹⁶ subsequently showed that the mean age of patients was 68 years and mean time to detection was 9 months with a male predominance. Based on their 30 years of experience (1977–2007) of treating 445 patients with cholangiocarcinoma with preoperative PTBD, Takahashi *et al*⁶ reported 25 catheter tract recurrences in 23 patients (two patients with two metastases each; incidence 5.2%). These were predominantly metachronous in nature (82.6%), occurring within first 1.5 years of surgery. Prolonged catheterisation, multiple catheters or manipulations, papillary histology and well-differentiated tumours were found to significantly predispose to catheter tract seeding. In 2015, Liu *et al*¹⁷ reviewed the English literature and found 30 reported cases of cutaneous metastasis in hilar cholangiocarcinoma. Half of

these cases occurred at the PTBD catheter site and two-thirds were solitary. Three-fourth of them occurred in men.

In the absence of consensus, some observers considered catheter tract recurrence to be incurable and have taken a palliative approach while others have contemplated aggressive surgical resection.⁶ Takahashi *et al*⁶ advocate en-bloc resection of isolated metastasis along with excision of the catheter tract, which may require liver resection. In their opinion, all PTBD catheter tracts should be extirpated during curative surgery of primary tumours. Shimizu *et al*,¹⁴ Uesaka *et al*¹⁸ and Kondo *et al*¹⁹ tried ethanol injection into the tract with mixed results. Overall survival after detection has been reported to be as poor as 4 months, with male sex, solitary lesions and recurrence within the first year having worst outcomes.^{6,17} However, the patient's age and the number and size of the metastasis did not seem to affect the prognosis. Sakata *et al*⁵ observed that 80% of patients with isolated PTBD tract metastasis survived more than a year following resection. The median survival of six patients undergoing resection was 22.5 months (range 9–65 months) with two surviving beyond 5 years and three surviving more than 2 years.⁵ In a review by Rosich-Medina *et al*,¹⁶ postexcision median survival was 10.5 months (n=4; range 8–18 months).

Learning points

- The small but sizeable risk of catheter tract seeding should be borne in mind while using percutaneous transhepatic biliary drainage (PTBD) for biliary decompression in malignant biliary obstruction.
- Complete extirpation or chemical ablation of such catheter tracts during curative surgery should be strongly considered to reduce the risk of subsequent recurrence.
- Carefully selected patients of catheter tract recurrence with no other forms of systemic metastasis can benefit from aggressive surgical treatment.

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