

Osteosarcoma of long bone metastatic to the pancreas-an unusual site of metastasis- A case report

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ABSTRACT

Background: Osteosarcoma is the most common malignant bone tumor in children and adolescents and possesses a high potential for metastasis. The most common sites of metastases are the lungs, pleurae, and bone. Unusual sites of metastasis that have been reported include pericardium, kidney, adrenal glands, brain, breast, liver, peritoneum, muscle, subcutaneous tissue, non-regional lymph nodes, heart, dura, diaphragm, stomach, duodenum, and pancreas. Pancreatic metastasis is exceptionally unusual. **Case Summary:** We have reported a case of pancreatic metastasis from osteosarcoma right radius in a 19 years old girl. She was diagnosed to have osteosarcoma of lower end of right radius, treated with chemotherapy followed by diaphyseal resection of radius with centralization of ulna and wrist arthrodesis. After about one and half year of disease free period, she developed pulmonary metastases & underwent bilateral pulmonary metastasectomy followed by chemotherapy. She remained disease free for three years when she presented with features of obstructive jaundice. CECT abdomen and chest showed an ill-marginated poorly enhancing hypodense mass lesion of size 3.5cm in the head of pancreas with essentially normal chest scans. USG & EUS guided FNAC were inconclusive. EUS showed hypoechoic mass lesion in the head of pancreas infiltrating portal vein-SMV-splenic vein confluence. In view of indefinite tissue diagnosis and radiologically resectable pancreatic lesion, exploratory laparotomy was done which showed a hard lobulated mass in head & neck of pancreas completely encasing SMA and MCA making it unresectable. Incisional biopsy showed metastasis of high grade spindle cell sarcoma: Osteosarcoma. **Conclusion:** Although extremely rare, osteosarcoma metastasis should be included in the differential diagnosis of pancreatic mass lesions, particularly in patients with a primary tumor.

Keywords: metastasis, osteosarcoma, pancreas

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Conflict Of Interest: Nil**INTRODUCTION**

Osteosarcoma is the commonest malignant bone tumor in children and adolescents and possesses a high potential for metastasis. The most common sites of metastases are lungs, pleurae, and bone. Unusual sites of metastasis that have been reported include pericardium, kidney, adrenal glands, brain, breast, liver, peritoneum, muscle, subcutaneous tissue, non-regional lymph nodes, heart, dura, diaphragm, stomach, duodenum, and pancreas. Pancreatic metastasis is exceptionally unusual. We report a rare case of pancreatic metastasis from osteosarcoma right radius in a 13 years old girl.

CASE REPORT

A 13 year old girl diagnosed to have non-metastatic osteosarcoma of lower end of right radius in May 2007. She was treated with neo-adjuvant chemotherapy (ifosfamide, adriamycin and cisplatin) followed by diaphyseal resection of radius with centralization of ulna and wrist arthrodesis in Dec

2007. Histopathology showed fibroblastic osteosarcoma with 80 % tumor necrosis. She completed her adjuvant chemotherapy in April 2008.

In Jan 2009, she presented with soft tissue recurrence locally with no evidence of distant metastasis which was managed with above elbow amputation. Postoperative chemotherapy was delayed due to surgical site infection. She underwent re-evaluation for metastasis before starting adjuvant chemotherapy which revealed bilateral lower zone multiple pulmonary nodules in April 2009. She was treated with six cycles of salvage chemotherapy (gemcitabine + venorelbine) till Nov 2009 and pulmonary metastasectomy on 31th Dec 2009 followed by chemotherapy.

She remained asymptomatic for about three years. In Nov 2012, she presented with features of obstructive jaundice. CECT abdomen showed a poorly enhancing hypodense mass lesion in the head and neck of pancreas with poorly defined fat planes with

SMV (Figure 1). CECT chest was essentially normal. EUS showed hypoechoic mass lesion of size 48x45mm in the head of pancreas

infiltrating portal vein-SMV-splenic vein confluence. USG guided FNAC & EUS - FNAC were inconclusive.



Figure 1: Computed tomographic scan of a 19 year old girl with history of osteosarcoma of right radius and recurrent lung metastases, showing ill-margined, poorly enhancing, hypodense lesion measuring approximately 33x37x32mm (APxMLxCC) in head and neck of pancreas encasing and compressing terminal part of CBD with upstream biliary dilatation.

In view of no tissue diagnosis, single site of disease and radiologically borderline resectable pancreatic lesion, exploratory laparotomy was done on 07/01/13 with intent to do Fortner's procedure (pancreaticoduodenectomy with vascular resection &

reconstruction). Per operatively she had large hard lobulated mass in head & neck of pancreas completely encasing SMA making it unresectable. Biopsy showed metastasis of high grade osteosarcoma (Figure 2). She was treated with CBD stenting and

palliative chemotherapy with high dose Methotrexate but she succumbed

to death 9 months after diagnosis of pancreatic metastasis.

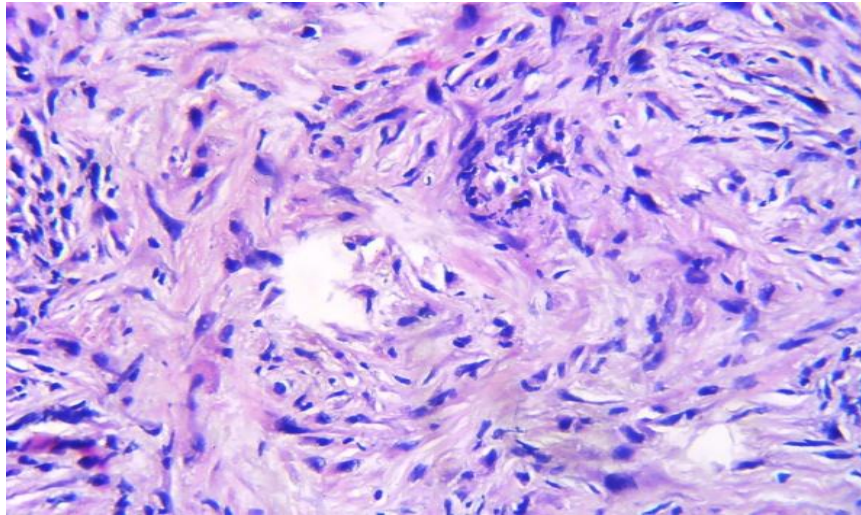


Figure 2: Histological section of pancreatic mass lesion showing metastasis of high grade spindle cell sarcoma with extensive collagen formation, scanty osteoid and focal necrosis.

DISCUSSION

Osteosarcoma is a deadly form of musculoskeletal cancer of childhood that most commonly causes patients to die of pulmonary metastatic disease. Surgical resection of pulmonary metastases combined with adjuvant chemotherapy has resulted in overall survival at 3 year of 30-50 % compared to < 10 % with no surgical intervention. Aggressive attempts at salvage after relapse can cause metastases in other extra pulmonary sites, such as the spine, brain, and

heart. In concordance with above statement our patient developed pancreatic metastasis following second line of chemotherapy.

Local recurrence is believed to represent an inherent biologic aggressiveness and a tendency to metastasize with even poorer prognosis. Despite having poor histologic response to NACT, early local recurrence and pulmonary metastasis, our patient fared well to metastasectomy and chemotherapy and

remained disease free for about 3 years.

Osteosarcoma metastatic to pancreas is exceptionally rare with only five such reports¹⁻⁵ in English literature briefed in the table. Four out of these including our case shared common feature of pancreatic metastasis with pulmonary metastasis. In contrast to pulmonary metastasectomy which results in long

term survival, there are no similar data for outcome following resection of osteosarcomatous pancreatic metastasis. Overall survival in such patient remained poor in all case reports with the range of 5-16 months. However, with surgical resection of solitary pancreatic metastasis, outcome may be better as shown by Aarvold et al² and Konstantinos L. et al³.

Table: Salient points from previous reports of pancreatic metastasis from osteosarcoma

S.N.	Reference/report	Primary site of OGS	Associated Lung metastasis	Prior chemotherapy	Treatment offered for pancreatic metastasis	Overall survival after diagnosis of Pancreatic mets
1	A. Aarvold (2007)	Femur	Present	Received	Whipple's procedure	Not reported (DFS>11 m)
2	Serhat Avcu (2009)	Thoracic vertebrae	Nil	Nil	Palliative chemotherapy	Not reported
3	Konstantinos L. (2010)	Fibula	Present	Received	Whipple's procedure	16 months
4	A S Khan (2011)	Maxilla	Present	Received	Not described	Not reported

5	M. Giovannini (2012)	Maxilla	present	Received	Palliative chemotherapy	5 months
-	Our Case (2013)	Radius	Present	Received	Attempted resection–Pall. Chemotherapy	9 months

EUS-FNAB is a reliable method for diagnosis of pancreatic lesions. Such an approach should be considered before any therapeutic decision is made, notably pancreatectomy, in patients with pancreatic mass and history of primary tumor^{4, 5}. In our case EUS-FNAB could not produce definite tissue diagnosis because of poor tissue yield, it definitely helped to define vascular invasion by tumor mass.

Although localized metastases to the pancreas are rare, many case reports and small studies have shown favorable outcomes for pancreatic metastasectomy notably for RCC, colorectal cancer, melanoma and sarcoma with best outcome for RCC. Criteria for selection of patients for pancreatic metastasectomy should include: primary cancer type that is

associated with good outcome, control of primary cancer site, demonstration of isolated metastases and patient fitness to tolerate pancreatectomy. Histological diagnosis is not necessary in order to proceed with surgery but it may indicate the outcome⁶.

CONCLUSION:

Secondary tumors of the pancreas ought to be considered in both the clinical, radiological, and pathological differential diagnosis of pancreatic lesions. Surgical resection may be considered in good surgical risk patient with isolated pancreatic lesion.

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