

Solitary rib metastasis from primary nasopharyngeal carcinoma: A rare clinical presentation

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Abstract

We report a case of a middle age male who presented to our tertiary care university hospital with the complaints of nasal obstruction and decrease hearing. The CT scan of head and neck exhibited a mass in nasopharynx and enlarged bilateral cervical lymph nodes. Biopsy from nasopharynx confirmed the lesion as poorly differentiated non-keratinizing squamous cell carcinoma and staged as cT2N2M0. He received neoadjuvant chemotherapy. Subsequently, he underwent chemo radiation therapy. He represented with left chest wall pain. Imaging confirmed isolated lesion on left sided 6th rib. Rib lesion was resected followed by radiation therapy to surgical bed and systemic treatment. The patient remained disease free for 4.5 years. Later, his disease relapsed, and he died of systemic disease progression. To the best of the author's knowledge, only few cases have been reported with isolated rib metastasis from nasopharyngeal carcinoma and this is the first case in which metastasectomy was considered.

Keywords: Nasopharyngeal carcinoma, Radiation Therapy, Rare presentation, Rib metastasis, Metastasectomy.

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Introduction

¹Worldwide nasopharyngeal carcinoma (NPC) is a common malignancy among head and neck cancers. In Pakistan, the incidence is not very high. It is ranked as 27th most diagnosed malignancy.¹

The typical distant sites are bone, lung, and liver. Bone metastasis is usually multifocal with predilection for spine and pelvis bones.² NPC with bony metastasis involving solitary rib is an uncommon presentation. We report a case of an NPC patient who presented with locoregional advanced disease. After initial management, he developed a single skeletal metastasis in left sided 6th rib. The management of NPC with skeletal oligo metastatic disease has not been well studied. Therefore, it was very challenging to decide metastasectomy and radiation therapy. Considering the rarity of this distinctive presentation and lack of evidence for management, we report this case to share our experience.

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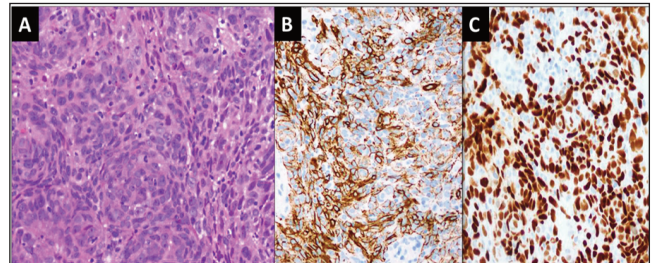


Figure-1: [A] Tumour cells are arranged in sheets and exhibit prominent nucleoli and frequent mitoses. The sprinkling of mature lymphocytes among tumour cells is also seen. (H&E stain, 400x magnification). Tumour cells demonstrate positive expression for [B] Cytokeratin 5/6 and [C] p63 IHC stains.

Case Report

A 48 years male presented to us with few months history of nasal obstruction and decreased hearing in June 2014 at Aga Khan University Hospital, Karachi, Pakistan. Nasopharyngoscope showed a protruding mass in the nasopharynx. Biopsy of the mass confirmed this lesion as poorly differentiated non-keratinizing squamous cell carcinoma with positive immunohistochemical markers, Cytokeratin AE1/AE3, Cytokeratin 5/6 and p63 (Figure-1) while EBV markers were not performed. The CT scan showed a mass in the right lateral wall of nasopharynx along with bilateral enlarged cervical lymph nodes while negative for distant metastasis. He received two cycles of neoadjuvant cisplatin and 5-fluorouracil (5-FU). Subsequently, he underwent concurrent chemo radiation therapy (CCRT) by conformal radiation therapy (3DCRT) technique with a dose of 70 Grays (Gy). He was on three monthly follow up.

After two years in July 2016, he returned with pain in left chest wall region. The CT scan confirmed isolated lesion on left sided 6th rib (Figure-2), and the PET/CT scan confirmed the rib lesion as the only site of distant metastasis. CT guided biopsy of the rib lesion was consistent with a nasopharyngeal metastasis. The case was discussed in MDT and the consensus for treatment strategy involved metastasectomy and surgical bed radiation followed by chemotherapy. Therefore, resection of rib lesion was performed followed by localised radiation (30 Gy/ 10 sessions) to the surgical bed. The patient received three cycles of cisplatin and 5-FU after local treatment. He

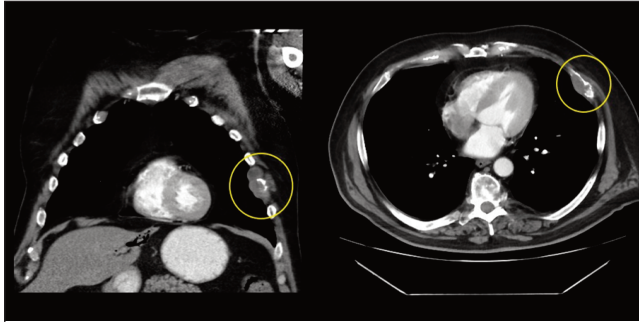


Figure-2: CT scan chest with contrast showing a soft tissue mass involving left sided sixth rib extending into 5th and 6th intercostal space (Coronal & Axial view).

continued on close follow up and remained disease free for 4.5 years. Later on the disease relapsed in lungs and liver. He died of systemic disease progression after one year. The consent has been taken from next of kin for publication of the case.

The purpose of reporting this case is because he presented at two following timelines 1) In 2016: At two years post curative treatment, he developed single metastasis in the ribs and the treatment was offered with curative intent rather than palliative treatment in oligometastatic stage IV disease. He remained disease free for 4.5 years after aggressive treatment. 2) In 2020: He developed systemic disease involving distant organs.

Single bony metastasis is a rare occurrence in nasopharyngeal carcinoma and we, the authors strongly feel that this case report would be beneficial for readers especially for those who treat nasopharyngeal carcinoma and deal with oligometastatic disease. Any oncological treatment deserves long term follow-up which is 7 years in this case in which two different metastatic events occurred.

Discussion

Nasopharyngeal cancer is a specific entity, unlike other head and neck cancers NPC has a unique pattern of distant metastasis. Bone, lung, and liver are common sites for distant metastases. To find the pattern of skeletal metastases, Sham JS et al. reported in their analysis that the axial skeleton is the most common site, frequently occurring in the spine and pelvis. Lumbar spine (28.4%) is the first region of involvement, followed by dorsal spine (27%), sacrum and pelvis (16.3%), femur (9.9%), and rib and sternum (7.8%).²

The isolated rib metastasis, in the absence of locoregional recurrence, is a rare presentation. Francesca De Felice et al., reported a case of metastatic NPC who initially presented with locally advanced disease and was treated with concurrent chemotherapy and radiation therapy. The

patient presented back with solitary metastases in the 10th rib. He was being managed with palliative chemotherapy and localised radiation to rib for local control.³ M. Paul et al. reported another case of a teenager boy who presented with locoregional advanced disease along with synchronous chest wall single metastasis. He was being managed with neoadjuvant chemotherapy followed by 35 fractions of radiation to face and neck region. Till last follow up after three years of diagnosis, he was still in a symptom-free and relapse-free state.⁴

Many studies have identified stage IV NPC disease as a heterogeneous group in which duration of metastatic free survival varies considerably, ranging from months to years.⁵ Good prognostic factors i.e. young age, isolated bony or lung metastasis, response to chemotherapy and loco regional disease control are all associated with long-term survival.⁶ Therefore, it is recommended to treat these patients aggressively with multi-modality therapy.⁵⁻⁷

NPC is a highly chemotherapy sensitive cancer. The platinum based chemotherapy regimen has the highest response rates.⁸ Although chemotherapy plays a major role, but the significance of the combined modality treatment is undeniable. In a recently published article, the author stressed over management of oligo-metastatic disease in curative arena.⁹ The role of surgical resection in skeletal metastasis from NPC has not been studied well. However, surgical resection in pulmonary oligo metastases has proved superior to chemotherapy alone. Chung et al. reported 2-year survival rate of 80% in 12 patients who had surgical resection and only 24.1% in 65 patients who had chemotherapy alone.¹⁰ Similarly Xun Cao et al. advocate the role of combined modality treatment and consider surgical resection in patients having good prognostic factors i.e. age less than 45 years and disease-free interval (DFI) greater than 1 year.⁶

We are fully aware of the fact that our patient presented in September 2014 and was treated with curative intent and he was kept on regular oncological follow up, which is an international standard. Our plan was to monitor him until five years after treatment.

Our patient was young with DFI of two years, therefore he was a suitable candidate for aggressive local treatment of the metastatic site. He remained alive for 7.5 years after the first presentation and disease free for 4.5 years after treatment for oligo-metastatic disease.

The clinical presentation with solitary rib metastases from NPC is considered a rare entity. Few case reports have been published in the literature with a solitary rib metastases from NPC in adults. In addition to that, the role of combined

modalities i.e. metastasectomy and radiation has never been reported previously for bony metastasis.

Conclusion

The clinical presentation with solitary rib metastases from NPC is considered a rare entity and there is no defined best treatment strategy to manage solitary rib metastasis. We believe that few patients with favourable factors and controlled loco-regional disease can get benefit from aggressive localized treatment with surgery and radiation followed by chemotherapy.

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