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Metastatic Intraosseous Adenoid Cystic Carcinoma of Mandible - A Rare Central Salivary Gland Neoplasm

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CASE HISTORY

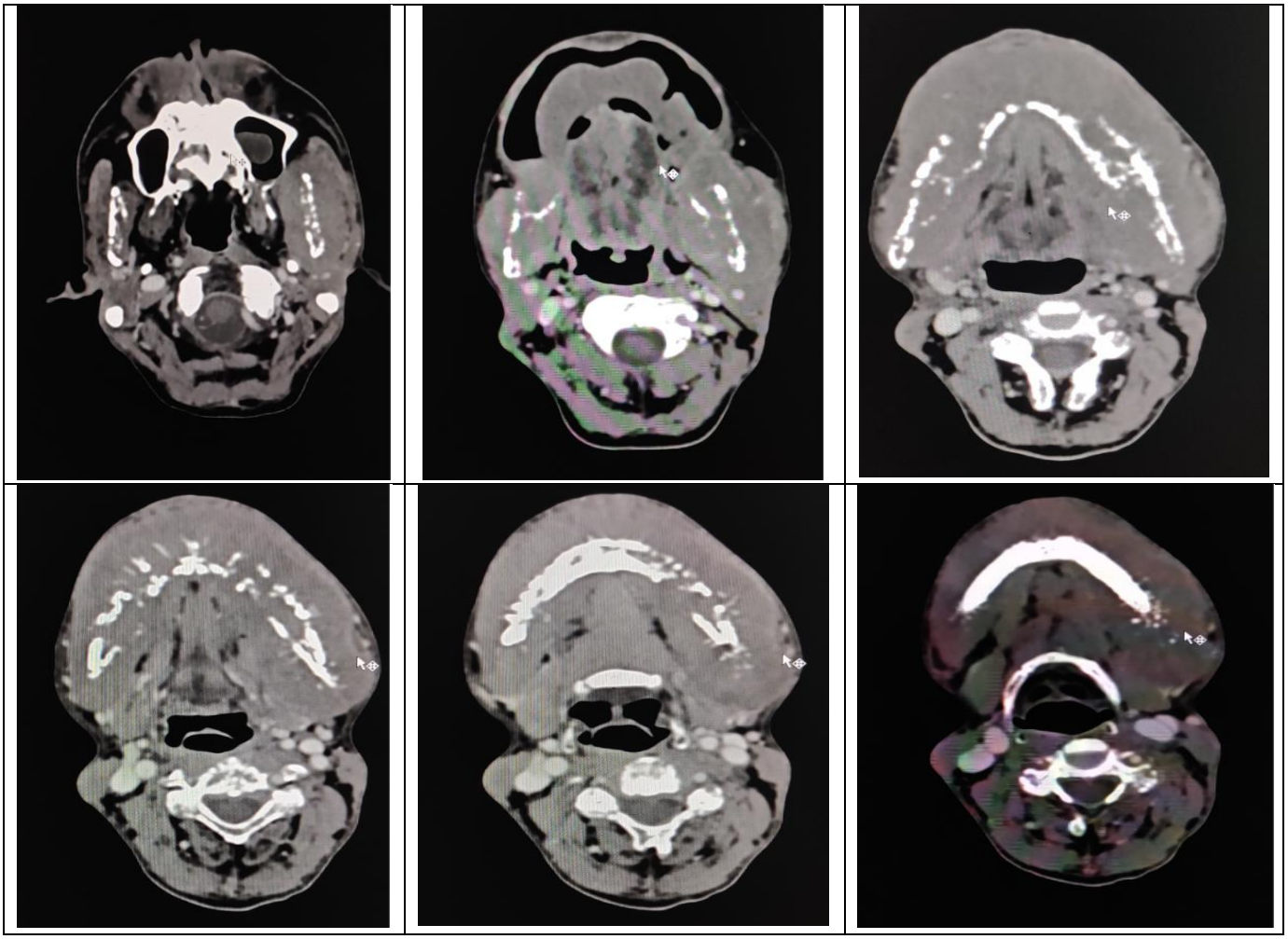
A 40 year old male subject came to MNJ Institute of Oncology and Regional Cancer Centre, Hyderabad with complaints of left lower molar tooth ache for 7 months and swelling of jaw, which gradually increased in size over 2 months.

The patient is unable to open his mouth completely with restriction of jaw movements.

History of weight loss.

He is a chronic smoker for 20 years and takes alcohol occasionally.

Patient underwent contrast enhanced CT of head, neck and abdomen



Figures 1A-1F: Serial axial sections of contrast enhanced CT of head and neck shows heterogeneously enhancing soft tissue density lesion in the mandible involving the body and rami with lysis and resorption of both outer and inner cortex. Laterally, the lesion is abutting, displacing masseter muscles with loss of fat planes.

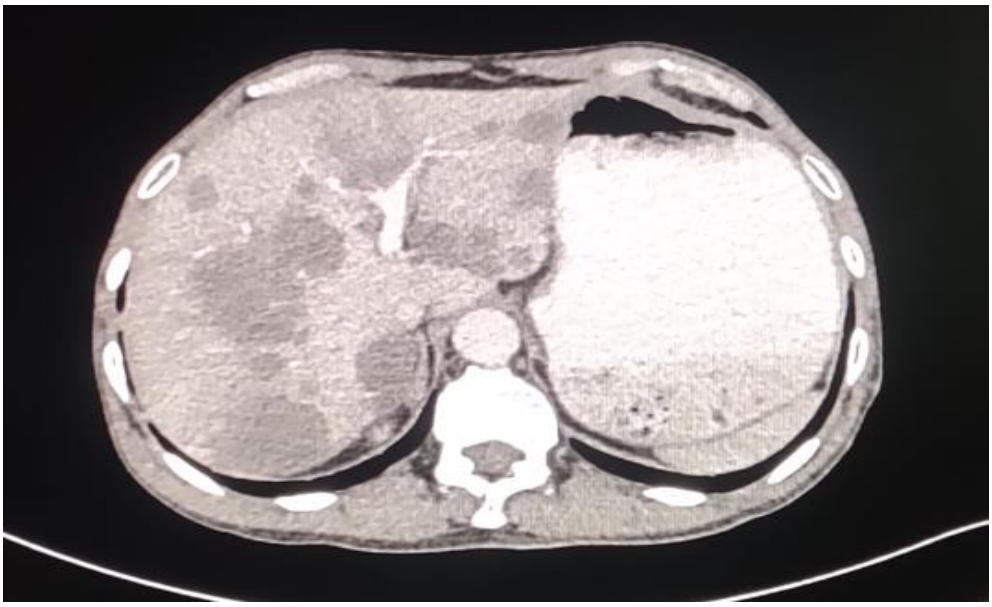


Figure 2: Axial section of CECT abdomen shows multiple non-enhancing hypodense lesions in both lobes of liver, suggestive of metastases

Subsequently, FDG PET-CT was done. It shows intense FDG concentration with lytic destruction of the entire mandibular arch, bilateral rami and condyles with surrounding tissue stranding.

- Intense FDG uptake noted in sternum, C3, C4, C5, C6, C7 and D3 vertebral bodies.
- Moderately increased FDG uptake noted in periphery of multiple hypodense lesions in both lobes of liver suggestive of necrotic liver metastases.
- Focal increased tracer uptake noted in right level 2 cervical lymphnodes.

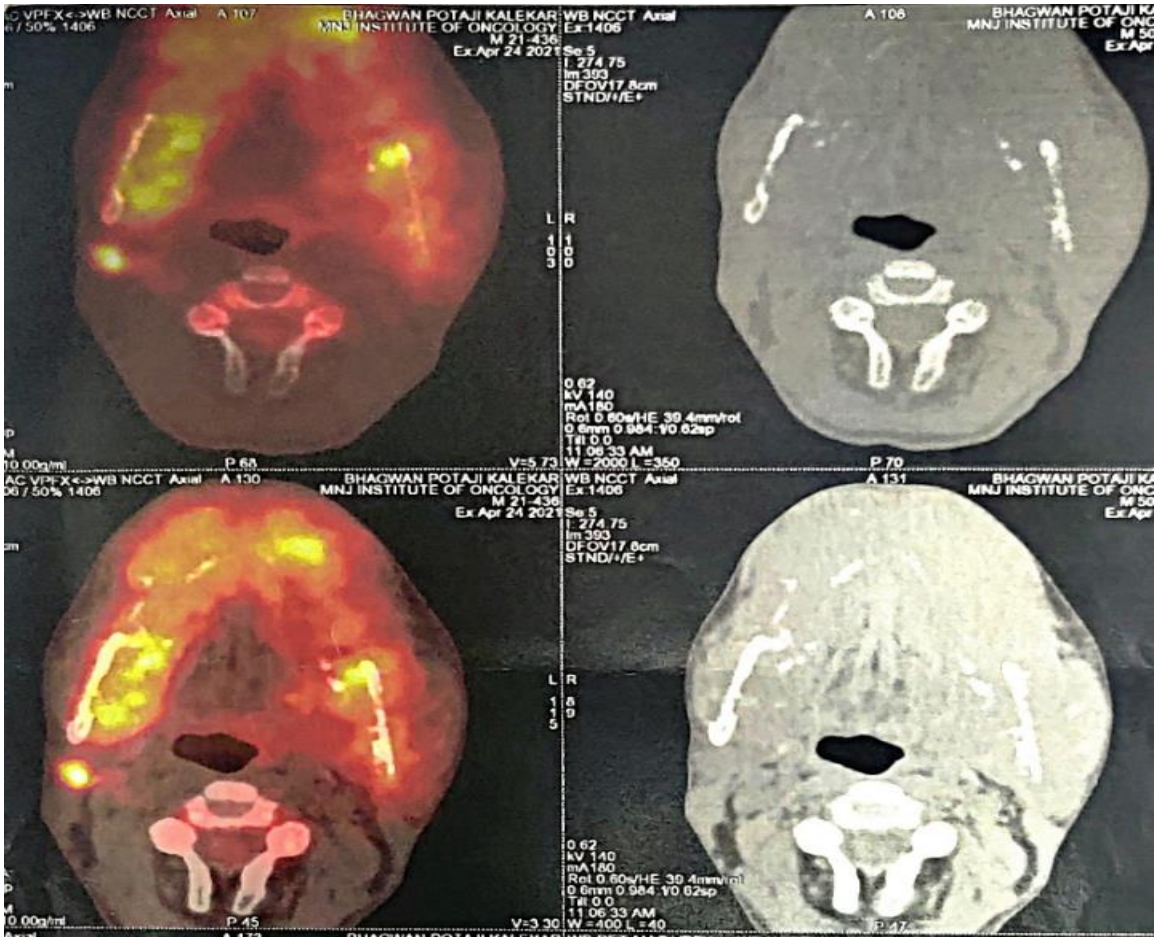
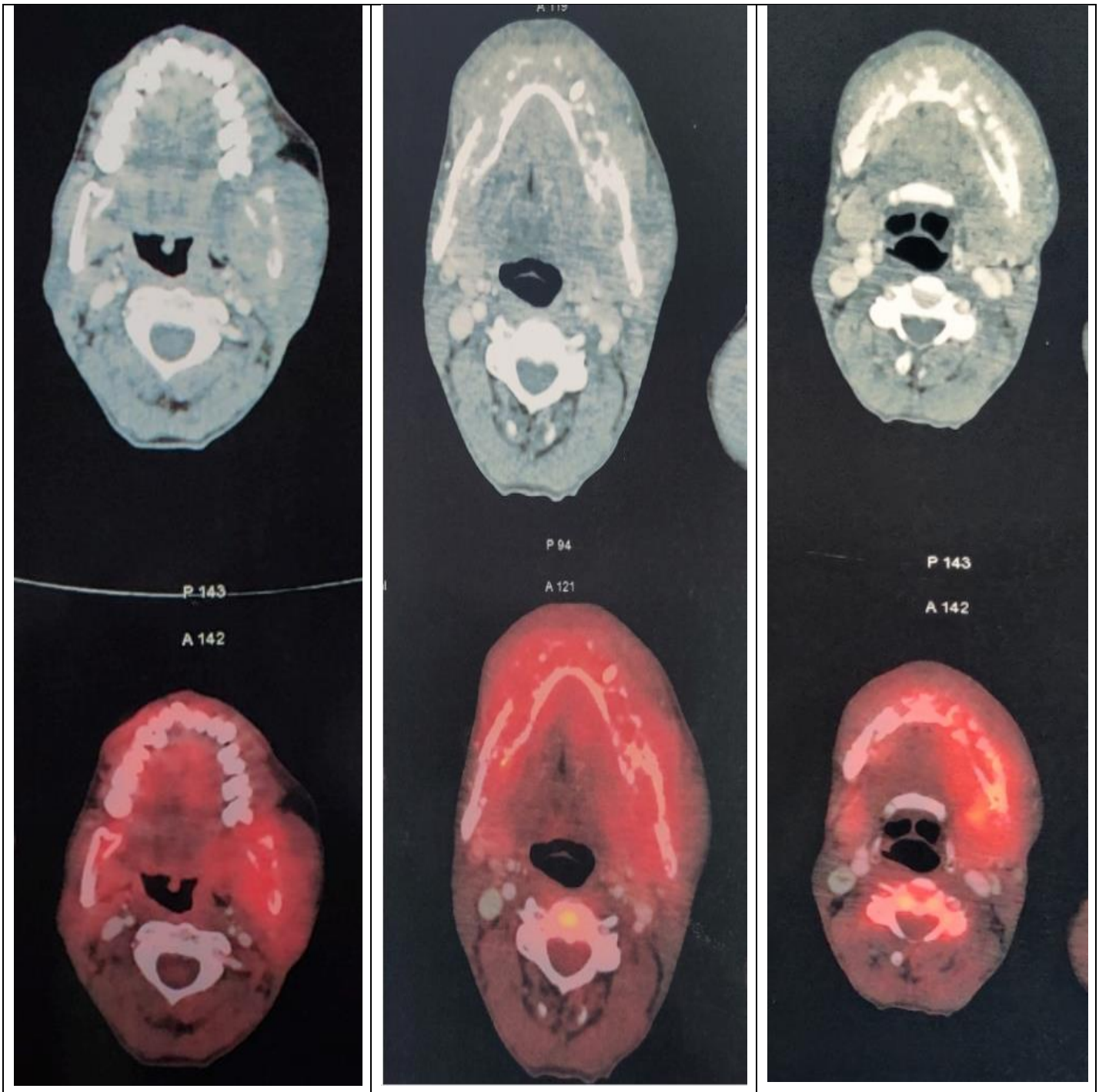


Figure 3: FDG PET-CT showing intense FDG uptake with lytic destruction entire mandibular arch bilateral rami and condyles



Figures 4A to 4C: Serial axial FDG PET-CT sections showing increased FDG uptake with lytic destruction of mandibular arch, bilateral rami and condyles. Normal physiological uptake in bilateral salivary glands

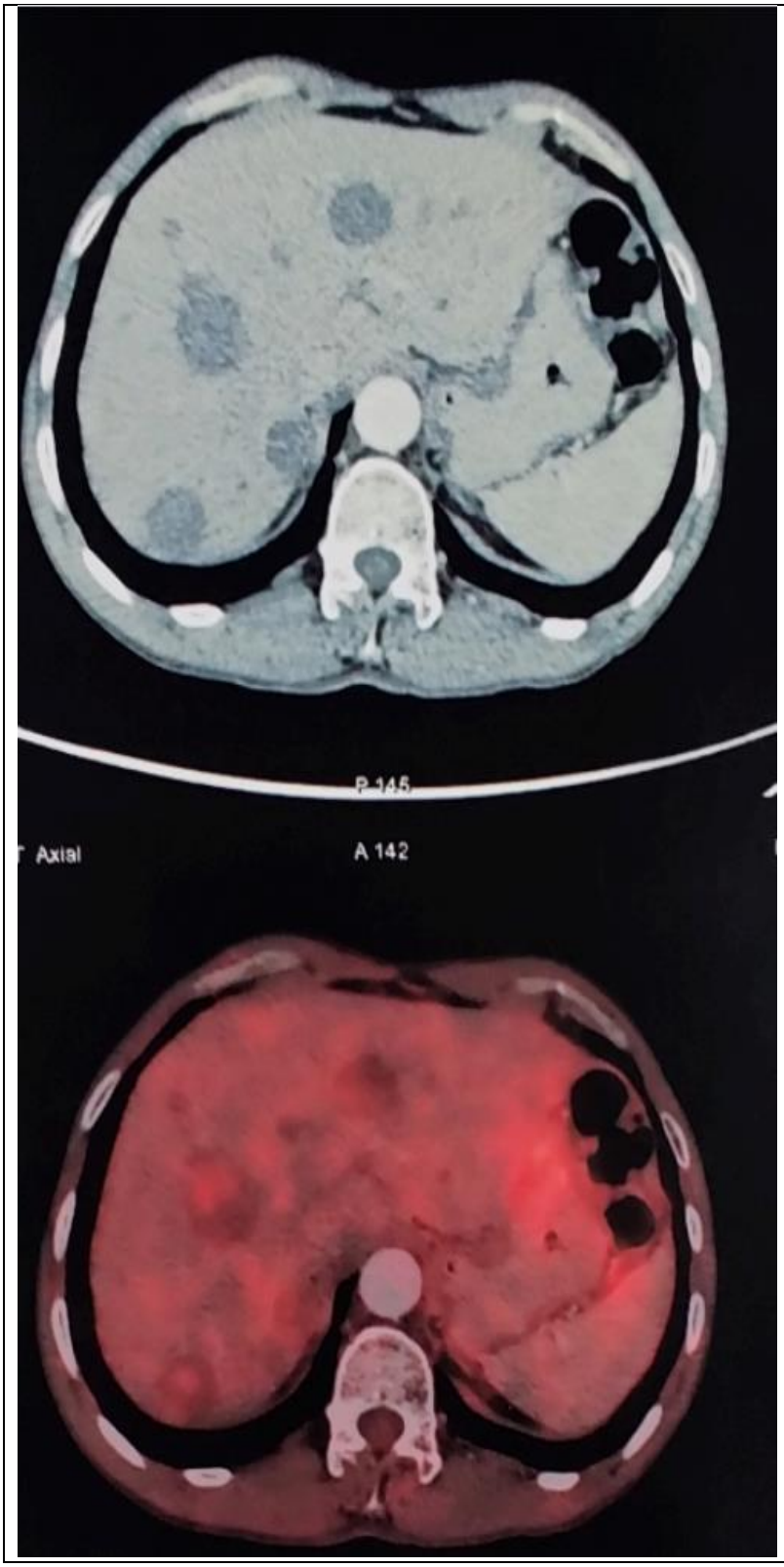


Figure 5:
Axial FDG PET-CT section showing multiple hypodense lesions with increased tracer uptake in both lobes of liver.

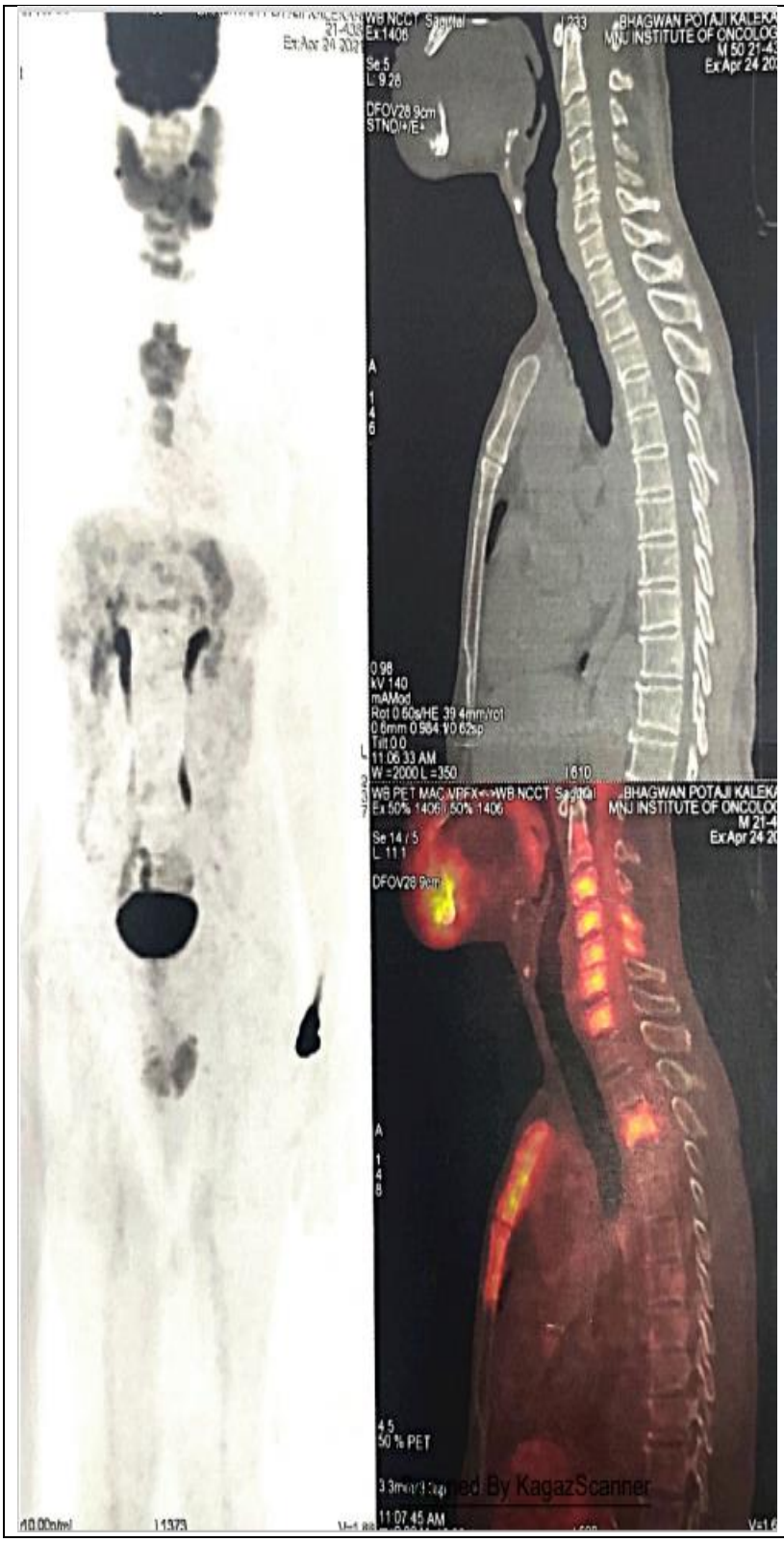


Figure 6:
Sagittal FDG PET-CT section showing increased tracer uptake in sternum, C2, C3, C4, C5, C6, C7 and D3 vertebral bodies.

- Provisional diagnosis:
  1. Osteosarcoma of mandible.
  2. Malignant ameloblastoma.
  3. Adenoid cystic carcinoma of mandible.

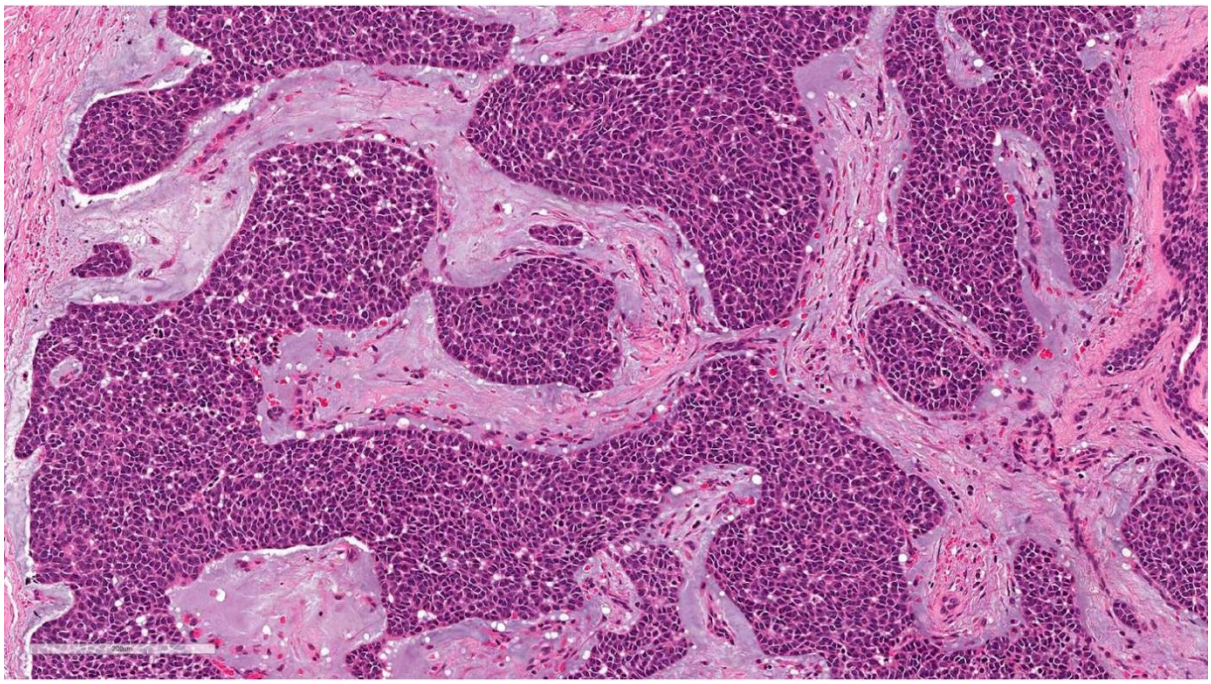
PATHOLOGICAL DISCUSSION

Histopathological examination showed multiple cores of tumour tissue with cells in sheets, nests composed of large round to polygonal cells, with scant to moderate pale eosinophilic vacuolated cytoplasm, large round dark nucleus with moderate nuclear atypia.

Foci of tiny cystic spaces surrounded by tumour tissue with areas of hyalinization were seen. Intervening stroma showed fibro collagen, areas of haemorrhage and tiny foci of bony trabeculae. Features suggestive of poorly differentiated Adenoid cystic carcinoma.

Immunohistochemistry revealed positive for Vimentin and Pan Cytokeratin (PCK).

Findings consistent with Adenoid Cystic Carcinoma of Mandible.



CASE DISCUSSION:

- ACC is a malignant salivary gland tumour, first described by Billroth as "Cylindroma".[1] It predominantly affects the minor salivary glands and accounts for 6-10 percent of all salivary gland tumours.[2] Rarely, it occurs centrally(intraosseous) causing lytic destruction of bone and affects mandible more commonly than maxilla.[2,3,4]
- Until now, only 48 cases of this rare central intraosseous tumour have been reported in the literature.
- Numerous theories have been proposed to explain the origin of central malignant salivary gland tumours: 1) Entrapment of ectopic salivary gland tissue in jaws. 2) Neoplastic transformation of odontogenic cyst epithelium. 3) Neoplastic transformation of sinus epithelium.[5]
- Central salivary gland neoplasms can occur at any age with no gender predilection. Mandible is the most common site but they seldom occur in maxilla with typical primary presentations being pain, swelling and rarely paraesthesia and numbness.[5,6]
- Batsakis proposed the criteria for diagnosis of primary intraosseous salivary gland neoplasms[4], which are: 1) Radiographic evidence of osteolysis. 2) Presence of intact cortical planes. 3) Presence of intact mucous membrane overlying the lesion. 4) Absence of primary tumour within major or minor salivary glands. 5) Histological confirmation of the typical architecture and morphological features of salivary gland tumour.
- Brookstone and Huvos proposed a staging system for central salivary gland malignancies based on the destruction of bone and cortex[5]- Stage 1: lesions with intact cortical plates and no bony expansion; Stage 2: lesions with intact cortical plate but bony expansion; Stage 3: lesions with cortical perforation or nodal disease.
- Though spread to regional lymph nodes is rare, distant metastasis to lungs and bones is common.[2,7]
- Solid, tubular and cribriform variants are the common histological patterns of ACC. Solid variants are high grade lesions with high recurrence rates.

RADIOLOGICAL FEATURES

- CT is useful in determining tumour diameter, extent of bone destruction, lymph node status and metastases to other organs.
- In cases of ACC of palate, CT is helpful in determining greater palatine foramen(GPF) enlargement, pterygopalatine fossa involvement, and foramen rotundum and cavernous sinus involvement, all of which are indicative of perineural spread of the tumour[8,9]. Studies on CT image analysis showed that patients with ACC in the palate with intact mucosa are more likely than non-ACC patients to show signs of perineural spread.[9,10]
- FDG-PET is complementary and sometimes superior to anatomical imaging modalities for staging of malignant central salivary gland tumours.

High grade tumours tend to be hypermetabolic than low and intermediate grade malignancies.[11]

TREATMENT

Surgery is the best treatment for ACC. However complete resection is difficult due to vascular and perineural infiltration[12]. Postoperative radiotherapy is needed to reduce local and regional recurrence.[13]

FINAL DIAGNOSIS

Adenoidcystic Carcinoma of Mandible with Liver and Bone Metastases.

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