

Cyodiagnosis of primary intraosseous squamous cell carcinoma of left mandible with unusual finding

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Abstract

Primary intraosseous carcinoma of mandible originating as a de-novo is a rare neoplastic entity. Its origin thought to be from previous odontogenic cyst or de novo rather than pre formed epithelia lesion. It presumed to be originated from residues of odontogenic epithelium. Fine needle aspiration cytology (FNAC) diagnosis of Primary intraosseous carcinoma (PIOC) is rare. Primary intraosseous carcinoma of squamous cell origin was very infrequent and only few cases are reported in available literature.

The role of this case report to highlight the importance of fine needle aspiration cytology for rapid and proper diagnosing such rare lesions of mandible with its unusual clinical presentation as discharging sinus.

Keywords: Cytology, Squamous cell carcinoma, Mandible, Intraosseous carcinoma.

Introduction

Primary intraosseous carcinoma (PIOC) of mandible is a rare neoplastic lesion with disputed origin and defined as squamous cell carcinoma of de novo origin from residues of odontogenic epithelium for jaw. It is a squamous cell carcinoma arising primarily from the mandible with no origin from oral mucosa.⁽¹⁾ It was first described by Loos in 1913 and named as intraalveolar epidermoid carcinoma by Wills in 1948.⁽²⁾ In 1972 Pinborg, finally coined the term primary intraosseous carcinoma (PIOC).⁽³⁾

As per WHO the primary intraosseous carcinoma was defined as "A squamous cell carcinoma arising within the jaw, having no initial connection with the oral mucosa and presumably developing from residues of the odontogenic epithelium. Upto two-thirds of PIOC is malignant transformation within odontogenic cysts, while PIOC arising denovo is relatively rare. In the available English literature from various databases we found only 35 case of primary intraosseous carcinoma.⁽⁵⁾ Due to its rarity and unusual clinical presentation, we here in report a case of PIOC of left mandible presented as a discharging sinus diagnosed on FNAC OPD procedure.

Case Report

A 45 year old male presented to Surgery OPD of our hospital with chief complaint of swelling of the left lower jaw and discharging sinus since 5 months. Patient visited to OPD due to pain at left jaw. Personal and past history was not contributory except history of tobacco chewing was present since last 20 years. On local examination his oral mucosa was intact, no local growth or ulcer is noted. A hard, regular, immobile jaw swelling along with a discharging sinus was noted on left side. There was discharge of necrotic foul smelling pus like material oozing intermittently through the site.

Clinically suspected as odontogenic carcinoma of mandible and FNAC was advised to rule out malignancy. Pan-oramic radiograph demonstrated an ill-defined radiolucent lesion on anterior aspect of mandible. Dislocated teeth was also noted. On gross examination, it was 4x3 cms, tender, hard swelling present on facial aspect of left mandible. Overlying skin was normal with no detectable lymphadenopathy at head and neck.

A 22 guage needle was introduced and aspirate was taken under all aseptic precautions. It was scant hemorrhagic aspirate. Smears are made and stained with H&E and Leishman stain.

Cytology: Multiple cellular smears show sheets, clusters and predominantly scattered single polygonal neoplastic cells(Fig. 1). Individual tumor cells were polygonal shape having pleomorphic hyperchromatic to vesicular nuclei and occasional nucleoli (Fig. 2). Cytoplasm moderate to abundant with eosinophilia. Background shows numerous inflammatory cells predominantly keratinous debris.(Fig. 2).

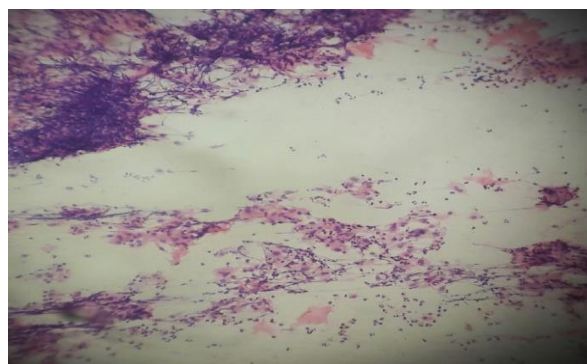


Fig. 1: Photomicrograph showing sheets of neoplastic squamous cell with inflammatory and keratinous background. (H&E, x100)

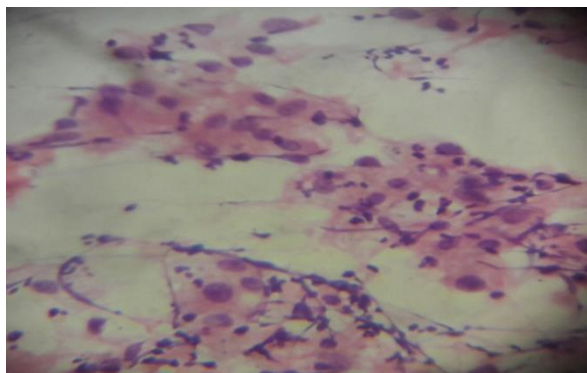


Fig. 2: Individual tumor cells with pleomorphic hyperchromatic with occasional nucleoli and abundant keratinous cytoplasm s/o Squamous cell carcinoma. (H&E, x400)

Based on microscopic findings and radiologic view final diagnosis of PIOC was offered. Left hemimandibulectomy was advised.

Discussion

Mandible is the most common site for occurrence of primary intraosseous carcinoma. PIOC of squamous cell origin was infrequent in occurrence and only handful of cases was reported in the available literature. It occurs mainly in the men. Ratio of occurrence in men: women is 2.2:1.⁽⁴⁾ Age ranges from 4-90 with mean age of 57 years.⁽⁴⁾ Diagnosis of primary intraosseous squamous cell carcinoma is confirmed only when metastasis from distant sites is ruled out. Diagnostic aids such as chest X ray, abdomen echo and bone scan can be helpful for general evaluation.

Presumably the site of origin of PIOC of squamous origin is residues of odontogenic epithelium⁽⁶⁾ hence previously called as odontogenic carcinoma.⁽⁶⁾ There are several classifications but Waldron and Mustubel's⁽⁷⁾ is widely accepted and frequently cited according to which PIOC may have different origins.

Type-1: PIOC ex. Odontogenic cyst

Type-2a: Malignant ameloblastoma

Type-2b: Ameloblastic carcinoma arising denovo, ex ameloblastoma or ex odontogenic cyst.

Type-3: PIOC arising denovo

a. Keratinizing type

b. Non keratinizing type

Type-4: Intraosseous mucoepidermoid carcinoma.

Primary intraosseous carcinoma of mandible must have the following criteria as--Histological evidence of squamous cell carcinoma, Absence of ulcer formation on the overlying mucosa and, Absence of a distant primary tumor at the time of diagnosis and at least 6 months during the follow up period.

In our case patient, we did not find any oral lesion or ulcer. On X ray view it was purely within the posterior mandible and microscopic examination revealed neoplastic squamous cells preliminary

diagnosis of primary PIOC was rendered, thus fulfilled all the criteria of PIOC of squamous origin.

In an analysis proposed by Thomas et al in 2001, there were totally 35 cases of PIOC arising denovo reported in the literature since 1964.⁽⁵⁾ The clinical features were non-specific while pain, swelling and sometimes sensory disturbances presented in most of the patients. Radiological diagnosis of PIOC could not possible due to its differential diagnosis and great variation though it may be one of the most effective methods for early detection of the bony lesions.⁽⁶⁾ Computed tomography also acts as useful evaluation tool to show the extent of lesion within jaw bone and soft tissue involvement. Lymph node don't have any prognostic value in PIOC. Due to its intraosseous nature, prognosis was very hard to predict. In our case, no lymph node was involved. Surgical treatment of choice was Commando's operation (hemimandibulectomy) which was done in our case and we confirmed the primary intraosseous squamous cell carcinoma of left mandible.

We are presenting this case for its rarity, unusual clinical presentation as discharging sinus and most importantly diagnosed on cytology. The FNAC has an extra edge in such lesions due to its simple, easy, rapid and less invasive and cost effective nature.

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