Anterior mandibular swelling – A Case Report

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Abstract:
Predilection of lesions to occur at certain specific sites is of great aid in arriving at a logical diagnosis. However tendency of lesions to appear at particular site does not follow a rule book. Enigmatic lesions like ameloblastomas have varied presentation. Here is an unusual case report of a patient who presented to us with an anterior mandibular swelling. Although clinical and radiographic features were suggestive of central giant cell granuloma, histopathological diagnosis was of ameloblastic carcinoma. Ameloblastomas are considered to be benign lesions; however, some can be reclassified as malignant when metastases occur or present with a very aggressive behavior. A detailed deliberation of differential diagnosis of anterior mandibular swellings is also done.

Key words: ameloblastoma, anterior, carcinoma, giant cell granuloma, mandible

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Introduction:

Predilection of lesions to occur at certain specific sites is of great aid in arriving at a logical diagnosis. Location may also point to the general nature of the disease. For example location below the inferior alveolar canal usually suggests that the problem is not odontogenic, location above the inferior alveolar canal certainly includes odontogenic lesions although other conditions may also be seen (1).

Some conditions tend to occur in a single anatomic site while others have a greater tendency to occur at a particular site though may also appear in other regions. Common lesions affecting the anterior mandible include the median mandibular cyst, central giant cell granuloma, calcifying odontogenic cyst, desmoplastic ameloblastoma, lesions of hyperparathyroidism and rarely aneurysmal bone cyst. However tendency of lesions to appear at particular sites do not follow a rule of the book. The present case illustrates that enigmatic lesions like ameloblastomas show varied nature and a broader perspective of differential diagnosis should be considered.

Case report:

A 60 year old female patient presented to the Department of Oral medicine and Radiology of KLE Society's institute of dental sciences, Bangalore with swelling of the lower front region of the face since four months. The swelling was reported to be insidious in onset and progressively increased in size. The patient gave a history of trauma to the lower jaw and spine six months back, for which she was bedridden for six months. It was reported that the lower front teeth became mobile following the accident. There was no history of discharge from the swelling or any pain associated with the swelling. Patient complained of paresthesia of the lower lip and lower one – third of the face. Medical and family histories were non- contributory.

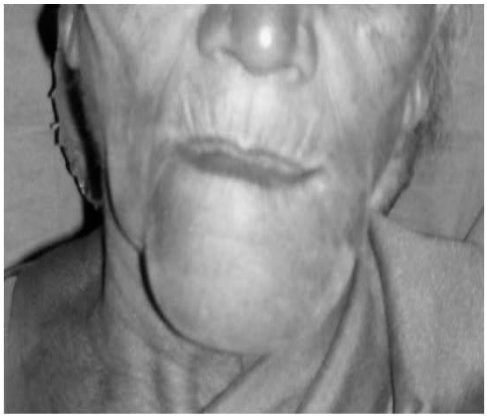

Figure 1: Diffuse swelling of the anterior region of the mandible

A diffuse swelling of the anterior region of the mandible extending below the chin with complete obliteration of the mento- labial sulcus was noted on extraoral examination. [Fig 1]. Severely concave profile was observed due to anterior mandibular swelling . The overlying skin was slightly stretched out. The swelling was firm and non tender on palpation. There was no local elevation of temperature. Both right and left submandibular lymph nodes were palpable, mobile, firm and non tender.

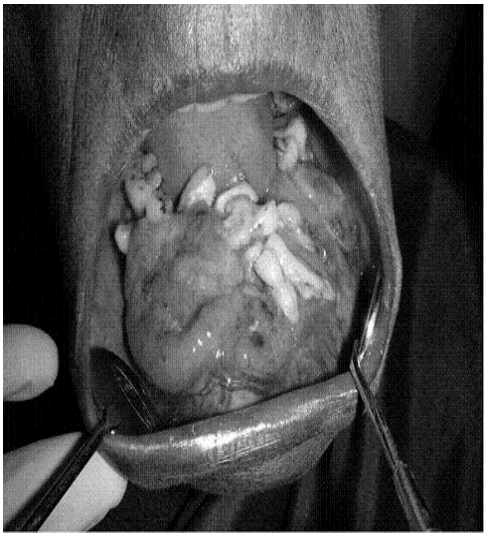

Figure 2: Intraorally, large diffuse irregular growth with displaced teeth on the surface of growth.

Intraoral examination revealed a large diffuse irregular growth (fig 2), measuring about 6cm X 5 cm crossing the midline, extending from the left lower second premolar to the right lower second premolar; the teeth were displaced out of their sockets and had migrated to the surface of the growth. The surface of the growth was bosselated, erythematous nonulcerated with prominent blood vessels. There was complete obliteration of the lower labial vestibule, but lingual sulcus was not obliterated and lingual cortex was palpable. The growth was nontender, mostly firm in consistency with softening in few areas. There was no detectable thrill or bruit. The lower first molars were grade III mobile on both sides and upper right second molar and lower left second molar was missing following extraction. Rest of the teeth component showed attrition and severe gingival recession.

Radiographic investigations were done. Mandibular occlusal radiograph showed a large ill-defined radiolucency, with nonsclerotic borders, severe expansion of labial cortical plate [fig 3]. Within the radiolucency thin wispy incomplete septae could be visible. The teeth were displaced out of the socket giving a floating teeth appearance. The lingual cortex was intact. Lateral cephalogram revealed ballooning out and perforation of the buccal cortex in the anterior region of mandible, triangular crenations at the margins, and incomplete septae within the radiolucency. Slight root resorption was seen in the mandibular anterior teeth. Orthopantomogram showed a large ill defined radiolucency with complete loss of normal trabecular architecture extending from the lower left second premolar to the opposite side second premolar [fig 4]. Severe thinning and scalloping of the inferior border of mandible was present. Chest radiograph appeared normal.

CT mandible picture revealed enhancing expansive osteolytic lesion involving the mid-body of mandible with irregular non-enhancing central area suggestive of central area of necrosis. Serum calcium, phosphorus and alkaline phosphatase were within normal limits [calcium- 9.5mg/dl, phosphorus – 3.5mg/dl, and Alkaline phosphatase – 198U/L].

Based on the history and clinical and radiological examination it was provisionally diagnosed as central giant cell granuloma. Calcifying epithelial odontogenic cyst, brown tumor of hyperparathyroidism, ameloblastoma and aneurysmal bone cyst were considered as differential diagnoses before the lesion was surgically excised en mass.

Differential diagnosis

A number of conditions affecting the jaws may present with an expansile lesion of the anterior mandible. Central giant cell granuloma warrants consideration as a first possibility. Central giant cell granuloma occurs most commonly in the mandible than maxilla, mostly occurs anterior to the first molars and often cross the midline. It occurs in all age groups but is more common in the young and there's a slight female predilection. Trauma is often associated with it. Clinical behavior ranges from a slow growing asymptomatic swelling to an aggressive lesion which manifests with pain and displacement of teeth. Radiographically the lesion may initially occur as a solitary cyst like radiolucency and as it grows larger, it may develop a multilocular radiolucency. In mature lesions internally there are few wispy septae which may be attached to the marginal crenations (1, 2, 3). In our

case except for the age all the findings were in favour of central giant cell granuloma.

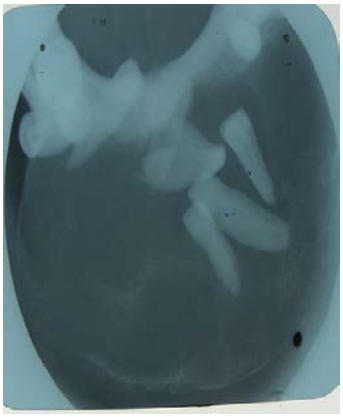

Figure 3: Mandibular occlusal radiograph showed a large ill-defined radiolucency, presence of thin wispy incomplete septae within the radiolucency

Calcifying odontogenic cyst was considered as one of the differential diagnosis. It has a wide age distribution with neoplastic variants mostly occurring in older age. Clinically the lesion usually appears as a slow growing painless swelling. It has a nearly equal distribution between jaws. Most of it occurs anterior to first molars, especially associated with cuspids and incisors. In most cases it causes expansion of bone and may destroy the cortical plate along with displacement of teeth. Radiographically appears as unilocular well – defined radiolucency, occasionally may be multilocular. Often radioopaque structures are seen within the radiolucency (1, 2, 3).

Ameloblastomas also primarily affect the posterior mandible except desmoplastic variant in patients from 20 to 60 years of age. In our case except for a wide age range of occurrence and severe buccal cortical plate expansion with resorption/ displacement of adjacent teeth, none of the features favoured the typical presentation of ameloblastoma such as a well-defined multilocular radiolucency with soap bubble or honey comb appearance or sometimes unilocular radiolucency, often associated with severe buccal as well lingual cortical expansion (1, 2, 3).

Brown tumor of hyperparathyroidism may also affect the anterior mandible. It occurs mostly in old age with a female predilection. There is generalized involvement of maxilla and mandible. Patient complains of polyuria, polydipsia, abdominal pain, muscular and joint pain and emotion instability. It is often associated with sudden drifting of teeth and spacing of teeth. Radiographically it appears as multiple unilocular or multilocular radiolucencies in jaws with altered trabecular pattern, there is loss of lamina dura. However in our case blood investigations revealed normal levels of serum parathormone and calcium. These findings essentially excluded the possibility of hyperparathyroidism (1, 2, 3).

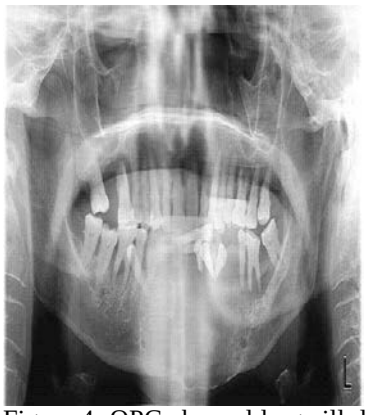

Figure 4: OPG showed large ill defined radiolucency with complete loss of normal trabecular architecture, severe thinning and scalloping of the inferior border of mandible

Aneurysmal bone cyst was also considered as another differential diagnosis. It mostly occurs in the first three decades with most developing in the 2nd decade. Equal to slight female predilection is noticed. Mandibular predominance is noted with majority occurring in premolar-molar area. Prior history of trauma is often present. Progressive swelling over the area of bone involvement is a common finding. On occasion migration and resorption of involved teeth may be present. Radiographically appears as eccentrically located ballooned out multilocular radiolucency with marked cortical expansion and thinning. Except the positive history of trauma, and ballooning out of the cortex none of features in our case matched with general manifestation of aneurysmal bone cyst (1, 2, 3).

Histopathological Diagnosis

Microscopic examination showed numerous interlacing strands of epithelial cells of odontogenic origin. Very small areas of stromal cysts were seen with capillaries and some areas showed perivascular hyalinization. Interlacing epithelial chords of cells showed hyperchromatism and reverse polarity of nucleus at many sites. Nuclear and cellular pleomorphism was also noticed [fig 5]. Another section revealed cells with vesicular nucleus and mitotic figures. Though the architecture of some

areas was that of a plexiform ameloblastoma, but the malignant cytology of other areas warranted the designation of ameloblastic carcinoma or carcinoma ex ameloblastoma.

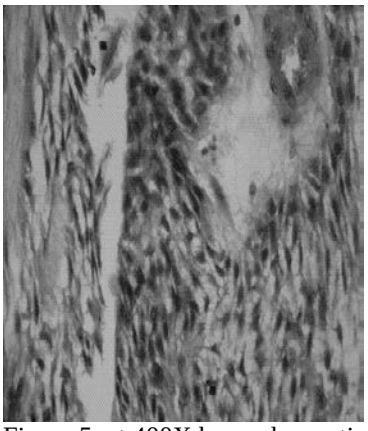

Figure 5: at 400X hyperchromatic dysplastic cells showing cellular and nuclear pleomorphism

Discussion

Ameloblastic carcinoma was first described by Robinson (4). The term 'ameloblastic carcinoma' refers to any ameloblastoma in which there is histological evidence of malignancy in the primary tumour or the recurrent tumour, regardless of whether it has metastasized while malignant ameloblastoma refers to those ameloblastomas that metastasize yet reveal an apparent 'benign' or typical histologic appearance in both primary and the metastatic lesions (5). Ameloblastic carcinoma is a rare lesion, however compared to malignant ameloblastoma it seems to be more common [in 2:1 ratio] (6). It occurs in a wide range of age groups, but the mean age of 30.1 years is in agreement with that reported for ameloblastomas. There is no apparent gender predilection. The most commonly involved area is the posterior portion of the mandible (5). Most common clinical signs and symptoms include swelling, pain, trismus, and dysphonia and mental nerve parasthesia. Clinically, these carcinomas are more aggressive than most typical ameloblastomas. Perforation of the cortical plate, extension into surrounding soft tissue, numerous recurrent lesions and metastasis, usually to cervical lymph nodes, can be associated with ameloblastic carcinomas (5). In cases of maxillary ameloblastic carcinoma the most frequent feature is a mass in the cheek, and/or anesthesia of the infraorbital nerve and occasionally fistula in the palate (6). In rare cases there may be malignancy associated hypercalcaemia (7).

Radiographically, these tumours present as an ill-defined destructive radiolucency containing occasional focal calcifications (4). Though the radiographic feature of our case was suggestive of an aggressive osteolytic lesion but the presence of triangular crenations at the margins and involvement of anterior mandible was more mimicking of central giant cell granuloma.

Histopathologically the primary tumour or the metastatic deposits shows microscopic pattern of ameloblastoma in addition to cytologic features of malignancy. These include an increased nuclear to cytoplasmic ratio, nuclear hyperchromatism, and the presence of mitoses. Necrosis in tumor islands and areas of dystrophic calcification may be present (8).

A single definitive diagnostic criterion for ameloblastic carcinoma is elusive. The constellation of discussed histologic features suggests a malignancy and warrants a careful review of submitted tissue (9).

The term ameloblastic carcinoma can be applied to our case, which showed focal histologic evidence of malignant disease including cytologic atypia and mitoses with indisputable features of classic plexiform ameloblastoma.

Adequate surgical resection is currently the most appropriate modality for the treatment of ameloblastic carcinoma with radiotherapy being used for metastatic disease not amenable to surgical resection. Chemotherapy as primary treatment does not appear indicated (10). Metastasic disease in the lungs and regional lymph nodes has been described in the presurgical and follow- up period. The prognosis of ameloblastic carcinoma is evaluated on the basis of possible relapse and metastasis (4). After 8 months of regular follow up, the patient remains free of locoregional disease.

Conclusion

The lesion in the present case deviates from the usual plexiform variant of ameloblastoma in terms of locus, presentations, although it still meets the histologic criteria. Clinical behavior of ameloblastoma is quite variable and difficult to predict. It shows a spectrum of histology and biologic behavior from benignity at one end to malignancy at the other. The present case illustrates the malignant portion of this enigmatic lesion. It highlights the importance of histopathologic examination from multiple sites of the lesion to rule out malignant transformation at any of the sites, for an accurate diagnosis. We advocate that ameloblastoma should also be considered in the differential diagnosis of swellings of anterior

mandible even though it has a marked propensity to occur in the posterior mandibular area.

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