



A RARE CASE REPORT ON PLEXIFORM AMELOBLASTOMA

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Abstract : Plexiform ameloblastoma is a rare, benign but locally aggressive odontogenic tumor arising from the remnants of dental epithelium. It predominantly affects the posterior mandible and is most commonly observed in middle-aged adults. Treatment typically involves surgical resection with clear margins, as conservative approaches are associated with higher recurrence risks. A 29 year old female patient admitted with complaints of swelling over the right side of face about 7 months backs, patient noticed a facial disfigurement over right side of face. Patient was clinically diagnosed with Plexiform Ameloblastoma and undergone treatment with IV Antibiotics and other supportive measures.

IndexTerms - Plexiform ameloblastoma, odontogenic tumor, posterior mandible

1. INTRODUCTION

Plexiform ameloblastoma is a rare histological variant of ameloblastoma, a benign odontogenic tumor arising from remnants of the dental epithelium. It is characterized by interconnected cords or strands of epithelial cells in a loose, vascular stroma. Most commonly affecting the mandible, it presents as a slow-growing, painless swelling that can cause facial asymmetry and tooth displacement^[1]. Despite their slow-growing nature, their local invasiveness necessitates timely intervention to avoid complications like facial deformities and functional impairments. This variant is characterized histologically by a plexiform arrangement of epithelial cells in a loose, vascular stroma, distinguishing it from other ameloblastoma subtypes^[2]. It most often occurs in adults between 30 and 50 years of age and slightly favors males. Despite its benign nature, it is locally aggressive and requires appropriate surgical management to prevent recurrence^[3].

The tumor commonly presents as a painless swelling in the jaw, often accompanied by facial asymmetry, tooth mobility, root resorption, and occasionally, difficulty chewing^[4]. The exact cause is unknown, it is thought to arise from remnants of the dental lamina, enamel organ, or basal cells of the oral epithelium. Genetic mutations such as in the MAPK pathway have been implicated. Radiographically, it appears as a multilocular or unilocular radiolucency with a soap-bubble or honeycomb pattern^[4]. Diagnosis is confirmed by histopathology showing a plexiform arrangement of epithelial cells in a loose, vascular stroma^[5]. Management involves surgical excision, ranging from conservative enucleation to aggressive resection, depending on lesion size and location. Long-term follow-up is essential to monitor for recurrence^[6]. Ameloblastomas account for about 1% of all jaw tumors, with the plexiform variant being less common. It predominantly affects adults aged 30–50 years, with a slight male predilection^[7].

2. CASE REPORT

A 29 years old female patient reported with chief complaint of swelling over the right side of face about 7 months back and she had noticed a facial disfigurement over right side of her face and was taken to nearby hospital where all the primary investigation and biopsy was done. The histopathology report revealed plexiform type of Ameloblastoma. She was conscious, oriented and Afebrile. On examination, chest was clear, she was able to move all limbs and GI was soft, non-tender. During the admission time all vitals were stable. On monitoring the laboratory reports, CRP (D1- 43.3mg/L. D4-15mg/L) was elevated and HB (D3- 11.3gm/dL, D4-10.8gm/dL) were declined.

On extra oral examination a diffuse swelling of approximately 4x4 cm noted over right side of face extending from Zygomatic arch to lower border of mandible superoinferiorly and extending from the nasolabial fold to tragus of ear In the anteroposterior

dimension. The swelling was firm in consistency, non tender, non compressible and non reducible. The skin over swelling was intact. The lower border of mandible was intact.

On intra oral examination there was a firm bony swelling noted over right side of mandible with expansion of alveolar bone. Vestibular obliteration noted over right buccal vestibule. No associated paraesthesia or mobility of tooth noted.

CT Findings reveals a large expansile mixed solid cystic heterogeneously enhancing lesion seen in the right hemimandible associated with thinning and erosion of mandibular cortex.

Patient was undergone the surgical procedure of Enucleation with peripheral osteotomy and chemical cauterization under GA. It was under aseptic condition painting and draping were done, LA with infiltration given irt lower right buccal vestibule, Crevicular incision given from 47 to 42, Anterior releasing incision given irt 43 and posterior releasing incision given extending to upper buccal vestibule irt 18, Buccal and lingual flap elevated, extraction of 47,46,44,45 done, Enucleation and extraction of 48 done, Buccal and lingual flap elevated, extraction of 47,46,44,45 done, Enucleation and extraction of 48 done, Peripheral osteotomy and chemical cauterization done using Carnoy's solution, Medicated gauze placed in bony cavity and closure done with 3.0 vicryl and 3.0 silk.

The condition was managed with INJ. AUGMENTIN (AMOXICILLIN+POTTASIIUM CLAVULANATE, IV, 1.2gm, Q12H), INJ. METROGYL (METRONIDAZOLE, IV, 500mg, Q8H), INJ. PACTIV (PARACETAMOL, 1gm, IV, Q8H), INJ. DEXA (DEXAMETHASONE, 8mg, IV, BD), INJ. PANTOP (PANTOPRAZOLE, 40mg, IV, BD), INJ. MECOBALAMINE (METHYL COBALAMINE, 1000 IU, IV, Q24H), METROGYL DG GEL (METRONIDAZOLE, L/A, TDS), SYP. IBUGESIC PLUS (IBUPROFEN, 5ml, RT, TID), SYP. ZINCOVIT (MULTIVITAMIN, 5ml, RT, OD), T. CHYMORAL FORTE (TRYPSIN+CHYMOTRYPSIN, RT, TDS), PROHANCE HP (PROTIEN SUPPLEMENT, RT, OD). On 6th day patient was symptomatically better and discharged with T. AUGMENTIN (AMOXICILLIN+POTTASIIUM CLAVULANATE, 625mg, P/O, 1-0-1), T. DICLORAN A (DICLOFENAC +PARACETAMOL, 50mg/325mg, P/O, 1-0-1), T. CHYMORAL FORTE (TRYPSIN+CHYMOTRYPSIN, P/O, 1-0-1), T. MEDROL (METHYL PREDNISOLONE, 4mg, P/O, 1-0-0), T. RENERVE PLUS (MULTIVITAMIN, P/O, 1-0-0), T. PANTOP (PANTOPRAZOLE, 40mg, P/O, 1-0-0), T. COBADEX CZS (MULTIVITAMIN, P/O, 0-1-0). METROGYL DG GEL (METRONIDAZOLE, L/A) for 1 week.

3. DISCUSSION

Plexiform ameloblastoma is a rare, benign odontogenic tumor, typically affecting the mandible. It is characterized by a plexiform pattern of interconnected cords of epithelial cells in a vascular stroma.

Despite being benign, it is locally aggressive and has a high recurrence rate if not adequately treated. Symptoms include painless jaw swelling, facial asymmetry, tooth displacement, and root resorption. It often grows slowly, leading to delayed diagnosis^[8]. Causes are linked to dental epithelial remnants, genetic mutations and developmental anomalies during tooth formation. A history of previous ameloblastoma increases the risk of recurrence, particularly if the tumor was not completely excised. Chronic irritation or trauma to the jaw and developmental anomalies during tooth formation, such as issues with the dental lamina or enamel organ, can also contribute to the tumor's formation^[9]. While these factors may increase the likelihood of plexiform ameloblastoma, the exact cause remains largely unknown. Diagnosis involves clinical examination, radiographic imaging (typically multilocular radiolucency), and histopathology, which reveals the characteristic plexiform structure^[10]. Treatment is primarily surgical, with conservative approaches (enucleation, curettage) for smaller lesions and aggressive resection for larger or recurrent cases to minimize recurrence risk^[11].

Ajinkya Varkhede, JV Tupkari *et al* presented a case report on Plexiform Ameloblastoma. A case of 12 year old girl had been reported to the Department of Oral Pathology and Microbiology with complaint of swelling on left side of face since last 2 years. The medical history was unremarkable. Clinical examination revealed diffuse, smooth –surfaced, hard swelling on left side of face. It extends from the zygomatic region to the inferior border of mandible superoinferiorly, and from the corner of mouth to the angle of mandible anteroposteriorly. The swelling extends from distal of first molar posteriorly and results in obliteration of buccal vestibule. Panoramic radiography showed a large multilocular radiolucent area occupying the left mandible from the first molar tooth to the neck of condylar process and the coronoid process including the left ascending ramus area. The base of the mandible and the anterior border of the ramus was damaged and thinned. The histopathological processing of the tumor revealed a plexiform ameloblastoma predominantly composed of epithelium arranged as a tangled network of anastomosing strands. Under general anesthesia and nasoendotracheal intubation and all aseptic precautions, tumor mass was exposed buccally and lingually. After extraction of lower first premolar, osteotomy cut was placed and completed buccally and lingually. Thus tumor mass was removed along with bone margin of 1.5 cm. Microvascular free fibula graft was harvested from right leg along with peroneal artery and vessels. Graft dimension was 14cmX3.5cmX3cm. Hemostasis was achieved, vacuum drain was secured and closure was done in layers. Antibiotics, analgesics and anti-inflammatory drugs were given postoperatively. Both wounds healed unevenly and sutures were removed on 7th postoperative day. Patient has been kept under periodic follow up since then. No recurrence had been reported till date^[12].

Paramjit Kajla, Jeevan Lata *et al* presented a case report on Plexiform Ameloblastoma. A case of 43-year-old female patient reported in our department with persistent painless slow-growing swelling involving the left body of the mandible region for the past 5–6 months. There was no history of any pain, pus discharge, paresthesia, or anesthesia in the affected jaw. Histopathological features revealed both follicular as well as plexiform ameloblastoma which adds to unique case till reported in literature. Ameloblastoma was known for its high recurrence rate if excision was incomplete and same is true with hybrid ameloblastoma. Therefore, the treatment of choice was surgical excision with wide-free margins. Bachmann and Linfesty reported a solid/multicystic type ameloblastoma that showed a variety of histologic types, with plexiform and follicular predominating and was treated with partial resection of the mandible. The present case was treated with segmental resection followed by

reconstruction of deformity with reconstruction plate fixed with locking screws under general anesthesia. She made uneventful recovery during her follow-up visits and is still under follow-up ^[13].

In our case, following diagnosis through an intraoral examination and CT scan, the patient underwent enucleation with peripheral osteotomy and chemical cauterization. Postoperatively, IV antibiotics and supportive measures were administered, and the patient was later discharged with oral antibiotics and vitamin supplements.

4. CONCLUSION

Plexiform ameloblastoma is a rare odontogenic tumor that requires timely diagnosis and appropriate management to prevent complications and recurrence. Its locally aggressive nature necessitates surgical resection with clear margins to achieve optimal outcomes. Early recognition of clinical signs, such as facial swelling or disfigurement, and confirmation through imaging and histopathological evaluation are crucial. Multidisciplinary care, including surgical intervention and supportive measures, plays a vital role in ensuring patient recovery and minimizing recurrence risks. Ongoing follow-up is essential to monitor for any signs of recurrence and to maintain long-term oral and facial function.

5. REFERENCE

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