A Case of Desmoplastic Ameloblastoma of the Anterior Maxilla

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CASE REPORT

A 16-year-old female patient reported to department of Oral Medicine and Radiology of Sardar Patel Postgraduate Institute of Dental and Medical Sciences Lucknow, with a chief complaint of swelling on the right side of the face since 2 years. Patient noticed that swelling started gradually 2 years back and attained to the present size not associated with pain and other symptoms. Her past medical and dental history were noncontributory. General physical examination revealed a moderately built and nourished individual of normal gait with vital signs within the normal limits.

Extraoral examination revealed a diffused swelling in the right anterior side of the face, involving the body of maxilla. Swelling measuring 2 × 3 cm causing deviation of upper lip, obliteration of nasolabial fold (Fig. 1). Swelling was bony hard, nontender, no localized raise of temperature noticed, nonfluctuant, noncompressible, fixed to the underlying body structures.

Intraoral examination revealed a well-circumscribed, nontender swelling present in the right labial vestibule measuring 2 × 3 cm, with a smooth surface extending anteriorly upto the labial vestibule of 11, posteriorly upto the distal aspect of 13. Extrusion of 11 and displacement of 12, expansion of labial cortex seen. Grade 1 mobility was elicited in 11. Electrical and thermal vitality tests were positive for 11, 12, 13, 14 and 21 (Fig. 2).

Aspiration was done and was not significant. A provisional diagnosis of a benign lesion of bony origin involving the right maxilla was made, and differential diagnosis of adenomatoid odontogenic tumor, fibrous dysplasia, odontogenic myxoma, ameloblastoma were considered.

IOPA and occlusal radiograph revealed a mixed radiopaque/radiolucent lesion giving a honeycomb pattern (Fig. 3). OPG revealed a honeycomb pattern extending from the distal aspect of the root of 11 upto the mesial aspect of the root of 13, showed displacement of 12, 13 and extrusion of 11. No evidence of root resorption seen. PNS view revealed slight haziness of the right maxillary sinus (Fig. 4).
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An excisional biopsy was advised. Surgical excision with partial maxillectomy of the right maxilla and extraction of 11, 12, 13 under general anesthesia was done.

The subsequent histopathology revealed a peripheral ameloblast like cells with cystic degeneration at the center, with desmoplastic connective tissue stroma, suggestive of desmoplastic variant of ameloblastoma (Fig. 5).

Final diagnosis of desmoplastic ameloblastoma of the right anterior maxilla was made. On subsequent follow-up for 8 months no recurrence of the lesion was reported.

DISCUSSION

Desmoplastic ameloblastoma is an unusual variant of ameloblastoma, which has a low occurrence rate and is characterized...
by marked stromal desmoplasia. It comprises of 4 to 13% of all ameloblastomas and average age is 17 to 72 years with the mean age of 41 years. The present case was being reported in a 16-year-old female in the anterior maxillary region in contrast to lesion reported by Tadahiko Kawai et al in the molar premolar region of left mandible in a 56 years old male. Maxillary lesions are more insidious than mandibular tumors owing to its proximity to the vital structures and the maxillary sinus.

Radiographically desmoplastic ameloblastoma may show either a multilocular mixed radiolucent/radiopaque appearance or multifocal appearance of minute flecks of bone similar to that seen in benign fibro-osseous lesions. Case reports has been cited in with a unilocular lesion with well-defined borders involving ramus of mandible and a radiolucent area of ill-defined outline in the body of mandible.

A benign but locally invasive variant of the intraosseous infiltrative, ameloblastoma consisting of proliferating irregularly shaped islands and narrow cords of odontogenic epithelium of varying sizes embedded in a desmoplastic connective tissue stroma. The histopathological feature of the present case characterized as containing desmoplastic areas in association with typical areas of follicular ameloblastoma. Another case of a “hybrid” type of ameloblastoma has been described with an ameloblastic tumor cells with dense collagenous stroma and the tumor cells showed basal cell variants of ameloblastoma.

CONCLUSION

The desmoplastic ameloblastoma is a histological variant of ameloblastoma. In the present case, the tumor occurred in a 16-year-old female in the upper anterior maxilla, which was treated by partial maxillectomy with extraction of the upper right anteriors. The entire maxilla possesses a thin cortical plate that offers a little resistance to the tumor, thereby enhancing its rapid spread into the adjacent vital structures. In our case, the lesion was well-demarcated with no extension into the adjacent structures. The behavior and the eventual prognosis of desmoplastic ameloblastomas still cannot be predicted with certainty. Many more detailed reports on the desmoplastic ameloblastoma, including long follow ups are needed for proper assessment of the treatment modalities.

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