

# Recurrent Ameloblastoma of the Mandible: Two Cases Report

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## Abstract

Six cases of ameloblastoma were treated by enucleation and peripheral osteotomy during 1992-2001. Histologically, desmoplastic reaction, epithelial cells with severe fibrous, and keratin formation in the acanthomatous follicles were seen. The disorder was in the mandible in five cases, and in the maxilla in one case. Two cases were male and four were female, and the age at surgery was between 20 to 28 years. For all cases, a uniform surgical protocol was applied. The lesions were removed primarily by enucleation with peripheral osteotomy. There was recurrence in two cases. An en bloc resection was done for the first case and a radical resection with titanium bone plate reconstruction for the second case. The patients did not have any problem, bone grafting being recommended for the first case as soon as possible. Based on our knowledge, the procedure was successful in approximately 70% of cases, but more radical surgery methods may be recommended in the initial surgery.

**Keywords:** Ameloblastoma; Recurrent; Treatment; Iran

## Case Presentation

A 26-year-old woman referred due to a slow-growing, painful mass on the right side of the mandible beginning six months before her referral. Clinical exams revealed an expansive lesion occupying the right angle of the mandible. The findings on primary panoramic view showed a multilocular radiolucency with involvement of the neurovascular bundle and the distal root of the second molar (Figure 1). The lesion was associated with an erupted third molar, pushed inferio-posteriorly. An incisional biopsy was taken, and histopathologic report showed follicular ameloblastoma. Cyst enucleation and peripheral osteotomy were performed, neurovascular bundle retracted gently, and the underlying bone removed. The panoramic view five years after operation showed recurrence (Figure 2).

After 9 years, the patient was admitted due to re-

currence very late in the course of the disease. The lesion was extended from the entire ramus to the canine area. The intraoral view demonstrated the involved area and cortical expansion (Figure 3). An intraoral hemimandibulectomy with a cuff of normal tissue and preservation of the head of the condyle was done. The panorex view taken 13 months later revealed the resected area and bone plate reconstruction. Although the condylar head had been displaced downward and forward, the mouth opening was within normal limits without deviation (Figure 4). A bone graft was suggested after the first post-operative year.

## Discussion

Ameloblastoma is a benign odontogenic tumour of epithelial origin without induction in the connective tissue.<sup>1</sup> Multicystic ameloblastoma mainly affects adult patients between the third and seventh decades of life, frequently in the posterior region of the mandible.<sup>2</sup> Ameloblastoma, a benign tumor of odontogenic type, represents 10% of all the tumors of the jaw bone.<sup>3</sup> Thirty-nine (54.9%) out of the 71 subjects were males,

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**Figure 1:** Panoramic view before initial treatment



**Figure 2:** Panoramic view showed recurrence

and 32 (45.1%) were females. Sixty-two (87.3%) out of the 71 ameloblastomas were located in the mandible. Swelling was the most common symptom and was experienced by 27 (38.0%) patients.<sup>4</sup> Presenting symptoms included swelling (n=182; 98.9%), pain (n=64; 36.0%), mobile teeth/history of extraction (n=104; 57.5%), purulent discharge (n=39; 21.7%), and paresthesia (n=10; 5.6%).<sup>5</sup> Histopathologically, it occurs in six patterns: plexiform, follicular, acanthomatous, granular cell, basal cell, and desmoplastic type.<sup>6</sup> The most common histologic pattern was plexiform, rather than follicular or acanthomatous.<sup>4</sup> Follicular ameloblastoma was the most common histological type (50 cases, 64.9%), followed by plexiform ameloblastoma (10 cases, 13.0%). Four (5.2%) cases of desmoplastic and 3 (3.9%) cases of acanthomatous ameloblastoma were seen while the basal cell variant accounted for 2 (2.6%) cases. Only 1 case of the unicystic type was seen.<sup>7</sup> The histopathological study on the surgical specimens revealed that 50% of the lesions were unicystic, 45% were multicystic, and the other 5% of carcinomatous ameloblastic patterns.<sup>8</sup> Cardinal radiographic criteria include the density of the lesion and the location with regard to the adjacent tooth structures within the jaw.<sup>9</sup> The correlation between recurrence and the treatment method or histopathological type was significant. Additional criteria in the evaluation of jaw lesions were demarcation, morphological



**Figure 3:** Intraoral view of Recurrence



**Figure 4:** Resected area with safe margin

characteristics, cortical involvement, periosteal and soft tissue changes.<sup>10</sup> Radiographically, 42 (59.2%) out of the 71 tumors were unilocular with a well-demarcated border. Of the remaining 29 cases, 14 were multilocular, 2 were of soap-bubble shape, and 13 were unknown in appearance.<sup>4</sup> Two-dimensional imaging allowed a better observation than 3D imaging of the deep structures, whereas 3D imaging was superior in visualizing the morphological changes of the compromised bones and the spatial relationship between the tumors and surrounding structures.<sup>11</sup> The results suggested that MR imaging was the best imaging method for visualization of the tumours, followed by contrast enhanced CT.<sup>12</sup> The first radiographic sign of recurrence was scalloping of the sclerotic margin of the regenerated bone.<sup>13</sup> Good results can be achieved in the treatment of ameloblastoma in children using conservative surgery. In the event of recurrence, a second surgery can be successful. Patients' compliance and careful follow-up are important.<sup>14</sup> The intraluminal subtype of unicystic ameloblastoma may do well with enucleation, but the intramural subtype may not. Since these cannot be identified preoperatively, more aggressive treatment is recommended, including peripheral ostectomy or enucleation with subsequent treatment of the surrounding bone with

liquid nitrogen, Carnoy's solution, or similar physico-chemical modalities. The peripheral ameloblastoma has a different origin and responds to local excision.<sup>15</sup> Surgical options include segmental resection, en bloc ostectomy, simple curettage, and excision with peripheral ostectomy.<sup>16</sup> Four treatment modalities for unicystic ameloblastomas were identified. The recurrence rates were 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by application of Carnoy's solution and 18% for marsupialization with/without other treatments in a second phase.<sup>17</sup> Preservation of the inferior alveolar nerve may be possible in the management of the unicystic type of ameloblastoma. However, a more radical approach is necessary for treatment of multicystic or solid tumors, especially those exhibiting a follicular pattern.<sup>18</sup> A conservative approach concerning the inferior alveolar nerve is suggested. Removal of excess perimandibular soft tissue is not indicated. The overlying attached mucosal surface should, however, be excised together with the underlying bone.<sup>19</sup> Inducible nitric oxide synthase expression and VEGF expression may be closely related to the angiogenesis and invasive biological behavior of ameloblastomas.<sup>20</sup> The increased TGF-beta1 expression in tumors with a high risk of recurrence could be explained, using the fact that although TGF-beta acts as a potent tumor suppressor in the early stages of tumor progression, later it seems to enhance the invasive phenotype of the tumor.<sup>21</sup> Ameloblastomas carry a certain risk of developing local recurrences depending on the histology and type of surgical treatment. Long-term follow-up should be arranged.<sup>22</sup> The development of new prototyping systems provides accurate 3D biomodels on which surgery can be simulated, especially in

cases of ameloblastoma in which the safety margin is important for treatment success.<sup>23</sup> Marsupialization was performed in 31 cystic ameloblastomas before surgery, and the effective rate of marsupialization was 74.2%. Recurrence was observed in 7.1% (3/42) after radical surgery and in 33.3% (12/36) in conservative treatments. Relatively higher tendencies of recurrence were observed in the multicystic type and follicular and/or plexiform pattern tumors.<sup>24</sup> A resection with safety margin is the best method to treat most proven ameloblastomas, and conservative treatment is reasonable for patients in their first decade or with unicystic or plexiform ameloblastoma.<sup>10</sup> In this study, it was confirmed that ameloblastoma cells had a major role in decreased bone formation by secreting sFRP-2 in cell culture model. Although sFRP-2 has a great effect on tumor progression, inhibition of sFRP-2's anti-bone formation activity and cell proliferative activity may reduce the invasive property of ameloblastoma and possibility of recurrence rate.<sup>25</sup>

Our results showed that conservative approaches such as simple enucleation and curettage at primary surgery produce the most recurrences. In contrast, more radical procedures with good visibility and adequate access could increase the cure rate. A close follow up is stressed.

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