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Characteristics of Mandibular Ameloblastoma and Postoperative Complication Influencing Factors in Cipto Mangunkusumo General Hospital during January 2008 – December 2012

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Abstract

Introduction. Ameloblastoma is a quite rare case but a common odontogenic tumor found, about 11% of all odontogenic tumors. The tumor is locally aggressive odontogenic one with a tendency to have recurrence and may cause severe facial deformity and dysfunction if not treated properly. The slow growing nature of this tumor usually lead to a delay in diagnostic. Recurrence rate of ameloblastoma reported as 15–25% after radical treatment and 75–90% after conservative treatment. This study aimed to find out the characteristics and influencing factors that contributed to postoperative complication.

Method. Those diagnosed as ameloblastoma who underwent total mandibulectomy, hemi-mandibulectomy, segmental mandibulectomy, and subtotal mandibulectomy as the first surgery followed by reconstruction using with reconstruction plate or bone graft in dr. Cipto Mangunkusumo general hospital in during January 2008 – December 2012 were reviewed descriptively using cross sectional retrospective study.

Results. Twenty-three subjects managed in the oncology surgery division, Department of Surgery, dr. Cipto Mangunkusumo general hospital during such a period. There were 7 males and 9 females aged in ranged of 20–55-year-old. The majority complained painless swelling (9 subjects, 39.1%) for less than 2 years (12 subjects, 52.2%). The most factor found to be related was tooth extraction (8 subjects, 34.8%), following removal of teeth cyst (6 subjects, 26.1%). The x-ray finding of panoramic view was multilocular (19 subjects, 82.6%) and the most surgical procedure preceded was partial resection of hemi-mandibulectomy (17 subjects, 73.9%). The most histopathological finding was follicular type (8 subjects (34.8%). Morbidity rate was 21.7%, no mortality. The most complication found was plate exposed (3 subjects, 13.08%). Median length of stay was 9 days (ranged of 7–26 days). There was no recurrence found in this study for 1-year postoperative follow-up. No significance relation between characteristics and complication.

Conclusion. The postoperative recurrence rate of mandibular ameloblastoma might be be minimized by a wide excision beyond safety margin.

Keywords: mandibular ameloblastoma, wide excision, safety margin, complication

Introduction

Ameloblastoma, though quite rare, is the most odontogenic tumor found which is 1% of all tumors in the head and neck region and approximately 11% of all odontogenic tumors.1 Ameloblastoma of the jaws is the most encountered odontogenic tumor in Asia and Africa, and referred to the second most odontogenic tumor found in North and South America.2 The age group predilection peaks are in the 20s and 30s, with the average age between 30 and 40 years; and the majority of cases occur in the 30 to 60 years age group.3 It is a locally aggressive tumor that – if neglected – may reach an enormous size and cause severe facial disfigurement, as well as functional impairment.4 Twenty percent of tumor may be found in the upper jaw, predominantly in the canines or molar region. In the mandible, 70% found in the molar region or the ascending ramus, 20% in the premolar region and 10% in the anterior part.5 Ameloblastoma found to be equal in frequency in both genders.5 It often presents as a slow growing mass, painless swelling, leading to expansion of the cortical bone, perforation of the lingual and/or buccal plates and infiltration of surrounding soft tissue. There is often delay in the diagnosis because of its slow-growing nature.6 Thorough understanding of its clinicopathological behavior is essential to avoid recurrence associated with inadequately treated disease.4

Currently, wide resection and immediate reconstruction is the treatment of choice in most cases of mandibular ameloblastoma.5 Complication rate after ameloblastoma surgery is surgical site infection 13.3–26.3%, plate exposure 5.9–46.15%.7,8 Recurrence rates of ameloblastoma are reported as high as 15–25% after radical treatment and 75–90% after conservative treatment.5 Any delayed diagnosis and management contributes to morbidity of severe facial disfigurement and functional impairment.

This study was addressed to review the characteristics and related factors contributed to postoperative complication. Information of these characteristics may escort surgeon in diagnosis and minimize postsurgical complication.

Method

In this retrospective study, it found 23 subjects diagnosed as ameloblastoma and managed during January 2008 – December 2012. There were 7 males and 16 females. X-ray (i.e. orthopantomography) were done in all subjects, purposed to find out
the accurate size and of tumor despite the tumor extension. Histological diagnosis and classification were based on the criteria defined by the World Health Organization (WHO) of histological classification. The methods of surgical procedure consisted of mandibular segmental resection, mandibular partial resection and mandibular total resection with immediate reconstruction using non-vascularized bone graft and reconstruction plate. The resection preceded beyond the safety margin for at least 2 cm of normal bone. Any information with respect to age, gender, clinical duration, risk factor, site, size, clinical presentation, radiographic features, histopathologic findings, management, postoperative complication (surgical site infection, plate exposure and recurrence) and length of hospital stay (LOS) had been verified. The impact of resection on the recurrence in 1–year follow-up period was reviewed.

Results

All subjects treated in this study preceded for the first surgical intervention, there were no secondary one. The frequency was higher in females (16 subjects, 69.6%) than males (7 subjects, 30.4%) with male and female ratio of 1:2.3. The mean age was 33.65 years old (ranged of 20–55 years old). Out of 23 subjects, 9 subjects (39.1%) occurred in the second decades of life. Bony hard swelling was the most clinical symptom found (9 subjects, 39.1%). Additional symptoms found were pain (5 subjects, 21.7%), displacement of teeth or tooth mobility (4 subjects, 17.4%), ulceration (4 subjects, 17.4%) and disturbed mastication (1 subject, 4.3%). There were 8 subjects (34.8%) associated with teeth extraction of third molars. The median duration of symptoms before presentation was 2 years (ranged of 4 months to 17 years).

The size of tumor was in ranged of 3 cm to 20 cm in diameter with average of 11.09 cm. Radiographically, 19 subjects (82.6%) were multilocular and 4 subjects (17.4%) were unilocular; mostly located in the body of mandible (15 subjects, 65.2%).

Histopathological findings consist of 8 subjects (82.65%) were follicular, 4 subjects (17.4%) were plexiform, 4 subjects (17.4%) were unicystic and the rest were mixed type (7 subjects, 30.4%). There was no subject of peripheral or desmoplastic ameloblastoma found in this study.

All the subjects were treated surgically with wide margin and immediate bony reconstruction with non-vascularized bone graft and reconstruction plate. Partial resection was preceded in 17 subjects (73.9%), segmental resection in 4 subjects (8.3%) and total resection in 2 subjects (8.7%). Complications occurred in 5 subjects (21.7%); developed surgical site infection in two subjects (8.7%), plate exposure in 3 subjects (13.08%) requiring plate removal. There is no recurrence within 1–year follow-up in this study.

The median length of hospital stay was 9 days (ranged of 7–26 days). Those with LOS ≤14 days were 21 subjects (91.1%), while as only 2 subjects (8.7%) with LOS >14 days as there were postoperative complication (surgical site infection) which managed by conservative wound care, keeping oral hygiene and prompt antibiotic as dictated by the results of the culture and sensitivity test.

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| Table 1. Subjects characteristics | Frequency | Percenta |
|----------------------------------|-----------|----------|
| **Gender**                       |           |          |
| Males                            | 7         | 30.4     |
| Females                          | 16        | 69.6     |
| **Age**                          |           |          |
| 20–29 years                      | 9         | 39.1     |
| 30–39 years                      | 7         | 30.4     |
| 40–49 years                      | 4         | 17.4     |
| ≥50 years                        | 3         | 13.0     |
| **Mean of age**                  | 33.65 years ± 10.50 |
| **Clinical features**            |           |          |
| Swelling                         | 9         | 39.1     |
| Pain                             | 5         | 21.7     |
| Mobility tooth                   | 4         | 17.4     |
| Ulceration                       | 4         | 17.4     |
| Disturbed mastication            | 1         | 4.3      |
| **Onset**                        |           |          |
| ≤2 years                         | 12        | 52.2     |
| >2 years                         | 11        | 47.8     |
| **Median of onset**              | 2 (0.4–17) years |
| **Risk factor**                  |           |          |
| Tooth extraction                 | 8         | 34.8     |
| Cyst removal history             | 6         | 26.1     |
| None                             | 9         | 39.1     |
| **Size**                         |           |          |
| <5 cm                            | 4         | 17.4     |
| 5–13 cm                          | 12        | 52.2     |
| >13 cm                           | 7         | 30.4     |
| **Mean of tumor size**           | 11.09 ± 5.73 cm in diameter |
| **Radiographic features**        |           |          |
| Multilocular                     | 19        | 82.6     |
| Unilocular                       | 4         | 17.4     |

| Table 1. Subjects characteristics (cont.) | Frequency | Percenta |
|-------------------------------------------|-----------|----------|
| **Sites**                                 |           |          |
| Corpus                                    | 15        | 65.2     |
| Angle                                     | 4         | 17.4     |
| Ramus                                     | 3         | 13       |
| Synphysial                                 | 1         | 4.3      |
| **Surgical intervention**                 |           |          |
| Partial mandibulectomy                    | 17        | 73.9     |
| Segmental resection                       | 4         | 17.4     |
| Total resection                           | 2         | 8.7      |
| **Histopathologic types**                 |           |          |
| Multi–cystic                              | 19        | 82.6     |
| - Follicular                               | 8         | 34.8     |
| - Plexiform                                | 4         | 17.4     |
| - Mixed                                    | 7         | 30.4     |
| Desmoplastic                              | 0         | 0        |
| Uni–cystic                                 | 4         | 17.4     |
| Extra-osseous/Peripheral                   | 0         | 0        |
| **Postoperative complication**            |           |          |
| Yes                                        | 5         | 21.7     |
| - Surgical site infection                  | 2         | 8.7      |
| - Plate exposure                           | 3         | 13.08    |
| - Recurrence                               | 0         | 0        |
| No                                         | 18        | 78.3     |
| Length of stay                            |           |          |
| ≤14 days                                   | 21        | 91.3     |
| >14 days                                   | 2         | 8.7      |
| **Median of length of stay**              | 9 (7–26) days |
symptoms in this study disclosed that this seems to be common in many developing countries. Factors like ignorance, poverty, poor communication and inadequate access between rural basic health facilities and the tertiary centers may contribute to late presentation. Analysis of clinical signs and symptoms in this study disclosed that the most common presenting complaint was slowly enlarging, painless swelling (39.1%), and this was found like other studies. The age distribution in this study (which was 33.65 years old) was found lower than those reported but, consistent with those reported by Krishnapillai and Nagata et al. Dodge, however, noted that ameloblastoma tend to occur at a younger age in developing countries and attributed this to the accelerated aging process due to poor nutrition and health care.

Ameloblastoma is thought to be occurred in equal between males and females. However, in the study it showed that females were predominant. Somehow, it should be noted that in a study, a review addressed only to those in the mandible. Rather than maxilla, it realized that ameloblastoma frequently occurs in the mandible; predominantly in the body and posterior mandible as it found in the recent study.

Previous reports regarding imaging for diagnostic purpose highlighted plain x-ray photo denotes lytic and radiolucent lesion depicted as unilocular, multi-cystic or soap-bubble appearance. In

| Table 2. Subject characteristics and postoperative complications |
|---------------------------------------------------------------|
| Patient’s characteristics | Postoperative complications (21.7%) | Description |
|---------------------------|-----------------------------------|-------------|
| Genders                   | Frequency | %     | Frequency | %     | Fisher’s Exact Test |
| Males                     | 1         | 14.3  | 6         | 85.7  | p = 0.508          |
| Females                   | 4         | 25.0  | 12        | 75.0  | p = 0.586          |
| Ages                      | Frequency | %     | Frequency | %     | Fisher’s Exact Test |
| 20–29 years               | 3         | 20.0  | 12        | 80.0  | p = 0.508          |
| 30–39 years               | 2         | 25.0  | 6         | 75.0  | p = 0.586          |
| Clinical features         | Frequency | %     | Frequency | %     |
| Swelling                  | 2         | 22.2  | 7         | 77.8  |
| Pain                      | 0         | 0.0   | 5         | 100.0 |
| Mobility tooth            | 2         | 50.0  | 2         | 50.0  |
| Ulceration                | 1         | 100   | 0         | 0.0   |
| Disturbed mastication     | 0         | 0.0   | 4         | 100.0 |
| Onset                     | Frequency | %     | Frequency | %     | Fisher’s Exact Test |
| ≤2 years                  | 2         | 16.7  | 10        | 83.3  | p = 0.455          |
| >2 years                  | 3         | 27.3  | 8         | 72.7  | p = 0.586          |
| Risk factors              | Frequency | %     | Frequency | %     | Fisher’s Exact Test |
| Yes                       | 2         | 25.0  | 6         | 75.0  | p = 0.586          |
| No                        | 3         | 20.0  | 12        | 80.0  | p = 0.586          |
| Size                      | Frequency | %     | Frequency | %     | Fisher’s Exact Test |
| ≤10cm                     | 1         | 9.1   | 10        | 90.9  | p = 0.185          |
| >10 cm                    | 4         | 33.3  | 8         | 66.7  | p = 0.021          |
| Radiographic appearance   | Frequency | %     | Frequency | %     |
| Unilocular                | 5         | 26.3  | 14        | 73.7  |
| Multilocular              | 0         | 0.0   | 4         | 100.0 |
| Site                      | Frequency | %     | Frequency | %     | Kolmogorov Smirnov |
| Corpus                    | 3         | 20.0  | 12        | 80.0  | p = 0.586          |
| Angle                     | 1         | 25.0  | 3         | 75.0  | p = 0.214          |
| Ramus                     | 0         | 0.0   | 3         | 100.0 |
| Symphysial                | 1         | 100.0 | 0         | 0.0   |
| Surgical intervention     | Frequency | %     | Frequency | %     |
| Partial resection         | 4         | 23.5  | 13        | 76.5  |
| Segmental resection       | 0         | 0.0   | 4         | 100.0 |
| Total resection           | 1         | 50.0  | 1         | 50.0  |
| Histopathologic type      | Frequency | %     | Frequency | %     |
| Follicular                | 2         | 25.0  | 6         | 75.0  |
| Plexiform                 | 1         | 25.0  | 3         | 75.0  |
| Uni-cystic                | 0         | 0.0   | 4         | 100.0 |
| Mixed                     | 2         | 28.6  | 5         | 71.4  |

Discussion

Even though ameloblastoma is a benign tumor, the nature of this odontogenic neoplasm tumor of epithelial origin is locally invasive. To date, the pathogenesis of this tumor remains unclear. It presents frequently as asymptomatic intraoral swelling but, in some cases, may be associated with a variety of symptoms including pain, paresthesia and loose teeth. This tumor occasionally found incidentally on routine dental x-rays. If untreated, ameloblastoma can result in severe facial deformity and they are associated with extensive local bone erosion and destruction. The average duration from onset to presentation of two years in this study indicated that most of the cases of ameloblastoma presented late for definitive treatment.

This seems to be common in many developing countries. Factors like ignorance, poverty, poor communication and inadequate access between rural basic health facilities and the tertiary centers may contribute to late presentation. Analysis of clinical signs and symptoms in this study disclosed that the most common presenting
the study, multicellular radiolucency feature refers to the most radiographic finding (82.6%), like other studies. World Health Organization (2005) classify benign ameloblastoma into 1) solid or multi-cystic, 2) extra-osseous/peripheral, 3) desmoplastic, and 4) unicystic. In this study, out of 23 subjects, 8 (34.8%) were of follicular type, 4 subjects (17.4%) were of plexiform type, 7 subjects (30.4%) were of mixed type and 4 subjects (17.4%) were unicystic type. This finding is similar to those reported by Bukhari, Moraes and Giradili et al.

Current recommendations of treatment are segmental resection, with at least a 1–2 cm margin followed by immediate bony reconstruction. Radiotherapy is not indicated, since the lesions referred to radioresistant. Recurrence of ameloblastoma is related to inadequate surgical treatment, incomplete removal of the tumor or the procedure of enucleation and curettage is employed. Postoperative follow-up is very important because more than 50% recurrences occurs within five years following treatment. There are reports reveal the recurrence time is between 1 and 15 years, whereas 2–5 years refers to the most common. In this study, all subjects treated by surgical resection with safety margin of 2 cm and it was found no recurrence within 1–year follow-up.

Postoperative complications noted in the study were intra–oral plate exposure, which is found in 3 subjects and surgical site infection, which is found in two subjects. Intra–oral plate exposure occurred in between 1 and 3 months following surgery requiring plate removal with no further reconstruction. Blackwell et al reported 3 subjects (21.4%) with hardware extrusion through the skin in cheek area. It was assumed as the impact of soft tissue contracture within dead space medial to reconstruction plate. Another study of Onoda et al showed postoperatively a reconstruction plate exposed by 3 months in one out of 17 subjects (5.9%). The exposed plate removed 11 months after reconstruction surgery and followed by mandible reconstruction using vascularized bone graft and soft tissue transfer. Surgical site infection may be treated conservatively through wound care, oral care hygiene and the use of therapeutic antibiotic in accordance with bacterial culture and sensitivity test. Length of hospital stay in the study ranged of 7 to 26 days (median 9 days) while as study of Bianchi et al reported LOS ranged of 6 to 16 days (mean, 8.6 days). LOS more than 14 days in the study found in 2 subjects (8.7%) due to postoperative complication i.e. surgical site infection as above described. In attempt to find out correlation between subject’s characteristic with postoperative complications it found there was no correlation. Sample size might be the issue (table 2).

**Conclusion**

The postoperative complication in management of mandibular ameloblastoma might be minimized. Recurrence might be minimized through wide excision involving safety margin. Wound infection might be avoided with good preoperative preparation. Plate exposure might be avoided by recognizing meticulous dissection and a good management of dead space.

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