Radiographic Evaluation of Odontogenic Keratocyst: A 14-year Retrospective Study

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Abstract—Odontogenic keratocyst is regarded as a relatively common developmental odontogenic lesion representing 12% to 14% of all odontogenic cyst of the jaw. The lesion is of important interest for its potentially destructive behaviour and relatively high recurrence rate from 21.1% to as high as 35.4% in patients associated with Nevoid basal cell carcinoma syndrome. This study aims to determine the radiographic characteristic of odontogenic keratocyst (OKC) using conventional radiographs and cone beam computed tomography (CBCT) images. Patients histopathologically diagnosed as OKC from 2003 to 2016 by Oral and Maxillofacial Pathology Department were retrospectively reviewed. Radiographs of these cases from the archives of the Department of Oral and Maxillofacial Radiology, Faculty of Dentistry Mahidol University were retrieved. Assessment of the location, shape, border, cortication, locularity, relationship of lesion to embedded tooth, displacement of adjacent tooth, root resorption and bony expansion of the lesion were conducted. Radiographs of 89 patients with the mean age of 31.5 years old were analysed. A total of 100 OKCs were studied. The most common location was at the ramus of mandible followed by posterior maxilla. Most cases presented as a well-defined unilocular radiolucency with smooth and corticated border. The lesions were in association with embedded tooth in 42 lesions. The lesions might relate to an embedded tooth by surrounding an entire tooth, attached to the CEJ level or extending to part of root. Bony expansion and teeth displacement could be found and root resorption were not common. These features facilitate in guiding the clinicians to formulate the differential diagnosis.

Keywords—odontogenic keratocyst, keratocystic odontogenic tumor, radiographic features

I. INTRODUCTION

Odontogenic keratocyst (OKC) was first introduced by Philipsen in 1956 and was formerly known as keratocystic odontogenic tumor [1]. This lesion has been recently reassigned as a cystic lesion in the 4th edition of the World Health Organization Classification of Head and Neck Tumours [2]. OKC is defined as “an odontogenic cyst characterized by a thin, regular lining of parakeratinized stratified squamous epithelium with palisading hyperchromatic basal cells” [3]. General consensus concurred that OKC derived from the remnants of dental lamina although some researchers suggested the possibility of this lesion arising from extension of basal cells of overlying the oral epithelium [4]. OKC has been gaining particular interest due to its aggressive nature and recurrence potential particularly in lesions occurring in the tooth bearing area [5, 6].

As the third most common cyst of the jaw, OKC represents 10% to 20% of odontogenic cyst affecting a wide age group but mainly prevalence in the second and third decade of life [4, 5, 7]. This lesion is slightly more predominant in male and most commonly found in the posterior region of the mandible [6, 8]. The majority of OKC cases occurred as a solitary lesion. Multiple lesions are frequently associated with nevoid basal cell carcinoma syndrome (NBCCS) [4, 9]. Clinically, most patients presented with an asymptomatic slow growing swelling and less common with pain and discharge on the affected site [5, 6, 10].

The aim of this study is to determine the radiographic characteristic of all OKC cases using conventional radiographs and cone beam computed tomography (CBCT) images.

II. MATERIAL AND METHODS

Cases histopathologically diagnosed as OKC from 2003 to 2016 by Oral and Maxillofacial Pathology Department, Mahidol University were retrospectively reviewed. Radiographs of these cases from the archives of the Department of Oral and Maxillofacial Radiology, Faculty of Dentistry Mahidol University were retrieved. There was a total of 100 OKC in 89 cases, of which six cases exhibited multiple lesions. Demographic data such as age and gender were collected while radiographic features were analysed. Cases were excluded if severe artefacts were found on the radiographs that could interfere with radiographic interpretation.

The location of the cyst was established by dividing the maxilla into left and right sides with two anatomic regions on each side which are the anterior (from midline to distal surface of canine) or posterior (from mesial of canine to maxillary...
Lesions in the mandible were divided into left and right sides and further dividing into three segments; anterior (from midline to the distal surface of canine), posterior (from mesial of premolar to the angle of mandible) and ramus (extending from the angle to the sigmoid notch).

Assessment of the shape, border, cortication, locularity, relationship of lesion to embedded tooth, displacement of adjacent tooth, root resorption and bony expansion of the lesion were conducted. The lesions were classified as unilocular or multilocular depending on the presence of at least one septum separating the lesion.

Descriptive statistics were used to analyze the radiographic features of OKC in order to determine the frequency and percentage.

III. RESULTS AND DISCUSSION

A. Patients Demographic

This study consists of total number of 89 cases with 43 (48.3%) of the patients were males and 46 (51.7%) were females. The ratio of male to female patients were 1:1.07 showing almost equal gender predilection. The age of patients at the time of presentation ranged from 10 to 87 years old, with the mean age of 31.5 years. Patients in the second and third decades were most commonly affected (54 out of 89, 60.7%).

B. Radiographic Features

Anatomic locations

From 100 lesions, 59 OKCs were identified in the mandible and 41 presented in the maxilla as shown in Table 1. The ramus of the mandible was found to be the most commonly affected region in this series (32 lesions) followed by the posterior maxilla (29 lesions). Five lesions in maxilla and five lesions in mandible crossing the midline were observed.

| Location     | Right | Left | Crossing midline | Total number of lesions |
|--------------|-------|------|------------------|------------------------|
| Anterior maxilla | 7     | 0    | 5                | 12                     |
| Posterior maxilla | 15    | 14   | 0                | 29                     |
| Anterior mandible | 2     | 0    | 5                | 7                      |
| Posterior mandible | 12    | 8    | 0                | 20                     |
| Ramus         | 20    | 12   | 0                | 32                     |
| Total         | 56    | 34   | 10               | 100                    |

Radigraphic appearance

Eighty-three OKCs were presented as unilocular and 17 lesions were multilocular. The unilocular:multilocular ratio of all OKCs was 4.8:1. Those lesions affecting the maxilla were predominantly unilocular lesions (36 out of 41 lesions). Most of the multilocular OKCs were found in the posterior mandible (11/17 multilocular lesions). One multilocular OKC located at the anterior maxilla, two at posterior maxilla and three lesions at the anterior mandible. Fifty-nine lesions presented with a smooth border while 41 OKCs exhibited scalloped border.

Association with embedded tooth

Forty-two OKCs were associated with an embedded tooth. Unerupted third molar was the most embedded tooth associated with the lesion (37/42; 88%). Among the cases associated with the embedded tooth, the lesion attached to the tooth at the cementoenamel junction (CEJ) in 17/42 (40.4%), and OKCs enveloping the entire tooth in 16/42 (38.1%) and beyond the CEJ in 9/42 (21.4%).

Effect on surrounding structures

Bony expansion in the buccal, lingual or palatal aspect was detected in 45 lesions from a total number of 70 OKCs that were justifiable for the assessment of cortical enlargement. The majority of OKCs associated with bony expansion were located at the posterior maxilla and ramus of the mandible (33/45 lesion; 73%). Teeth displacement were found in 30 OKCs while root resorption was noted in six OKCs, mostly involving lesions in the mandible.

Cone beam computed tomography evaluation

A total number of 30 cases incorporated CBCT imaging, involving 18 lesions in the maxilla and 12 OKCs in the mandible. Seventeen out of 18 lesions in the upper jaw involved the posterior aspect of the maxilla while nine out of twelve mandibular OKCs displayed lesion extension to the ramus region. According to the three-dimensional assessment, 25 out of 30 lesions portrayed bony expansion. In exception of one lesion located in the anterior segment of the maxilla, all lesions raised the floor of the affected maxillary sinus and invade into the maxillary antrum. From the examination of the mandibular lesion, three OKCs exhibit inferior displacement of the mandibular canal and five lesions eroded the cortical border of the mandibular canal. In both jaws, nine OKCs displayed cortical perforation in multiple planes. Interestingly upon CBCT images observation, one maxillary OKC was discovered to contain a few irregular shaped calcified foci, while one mandibular lesion appeared to exhibit a lamellar periosteal bone formation.

Multiple lesions associated with nevoid basal cell carcinoma syndrome patients

The present study includes five patients associated with NBCCS presented with multiple OKCs ranging from two to four lesions located in multiple quadrants. Fifteen OKCs
were found and the most lesions were located at the posterior region of the maxilla and mandible (12/15 lesions). Twelve lesions presented with a smooth and unilocular appearance while only three lesions showed a scalloped and multilocular features.

In this study, the age ranged from 10 to 87 years old similar to previous report [6]. The mean age recorded in our population was 31.5 years old, in consistent with previous studies [8, 11]. The highest frequency of cases affected patients in the second and third decades in agreement with existing researches [7, 12]. In this study, OKC showed no gender predominance with the ratio of 1:1.07 in contrast to previous researches that reported a male predominance [13, 14].

Radiographically, in consistent with previous studies, OKC was found in a higher number in the mandible than the maxilla [13, 16]. In the present study, 83% of lesions were unilocular while 17% were multilocular with the ratio of 4.8:1, almost similar to a previous study [16]. Forty one percent of OKCs presented with scalloped border involving all 17 multilocular lesions. Although most OKCs presented as a completely radiolucent lesion, this study identified one lesion containing radiopaque foci and one OKC with high density areas within the maxillary sinus which were detected on CBCT images. This might be dystrophic calcification that occurred within a long establish cyst [17].

In this study, 42% of OKCs associated with embedded tooth which is a higher occurrence compared to those reported by Chirapathomsakul et al that noted a slightly lower percentage of 31.3%. The majority of the involved teeth were the third molars, comprising 37 out of 42 (88%) of all lesions which is supported by previous studies [8, 10, 18, 19]. In this study, these 42 lesions were further classified according to the lesion attachments, 40.4% of OKCs were found attached to the CEJ, 38.1% covering part of the root and 21.4% enveloping the entire tooth. OKC may be mistaken as a dentigerous cyst (DC) if it envelope the crown of unerupted tooth. However, in DC the lesion most frequently found attached at the CEJ level [16]. Hence, if a radiolucent lesion is found to envelope an entire unerupted tooth or attach at the level below the CEJ, it may have a higher chance to be an OKC.

The characteristic growth pattern of OKC along the confinement of the cortical boundary until it reaches a considerably large size to cause a noticeable buccal expansion is well recognized [17]. However, in this study, more than half of OKCs presented bony expansion particularly at the posterior maxilla and ramus of the mandible and this is in consistent with a previous study [8]. In a study [11], OKC caused more displacement of impacted third molar in mesio-distal compared to ameloblastoma and DC. In this study, 30% of OKCs found to cause tooth displacement. Only six OKC displayed root resorption in this series which is slightly higher than other reports [6, 10]. CBCT images were prescribed for 30 lesions in the present studies. All lesions affecting the posterior maxilla raised the cortical floor of the maxillary sinus and further invaded into the affected sinus similar to a finding in a study by Gumusok et al [20]. On CBCT images, displacement and erosion of the cortex of inferior alveolar canal can be found in consistent with a study reported by Gamba et al [21]. Two OKCs involving the maxillary sinus displayed high density areas that were undetected in panoramic radiograph. These high-density areas seen on CBCT images might be the desquamated keratin content that occasionally increase the radiographic attenuation in CT images [22].

OKC may appear as the first presentation of NBCCS [23]. According to this study, five cases showed multiple OKCs and associated with NBCCS from the total of 89 cases (5.6%). This showed higher occurrence than a previous study [5]. In this study, the syndromic patients age ranged from 10 to 37 years involving three males and two females with the number of lesions varied from two to four lesions. We found that 10 out of 15 OKCs (67%) located in the maxilla in contrast to a finding by Khaliq et al that recorded 77% of syndromic lesions located in the mandible. Regarding the periphery and locularity, 80% of OKCs in syndromic cases displayed smooth border with unilocular presentation in consistent with existing study [23].

IV. CONCLUSION

Within the limitation of this retrospective study, the radiographic features of OKC is in consistent with the previous reports as they were predominantly found at the posterior segment of the mandible and presented as a well-defined unilocular radiolucent lesion. OKC may appear with a smooth or scalloped border and may associate with embedded tooth, particularly the impacted third molar. Upon examination of the relation of OKC with unerupted tooth, attachment at the CEJ and encapsulate the entire tooth were almost equally found. The least presentation showed the lesion attached to the part of root below CEJ. Bony expansion and teeth displacement might be common, while root resorption was considered a rare occurrence.

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