Study of the clinico-pathological patterns of odontogenic tumors  
(1994-2002)  

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**Abstract**

Odontogenic tumors were believed to be common in Africa. Their relative frequency varied from 8.6% to 30% of all oral tumors. In Sudan, however, these tumors constituted 8.6%.

Majority of odontogenic tumors were defined as true neoplasms while the nature of the rest remained uncertain. These lesions showed histological features, which mimic some stages of odontogenesis with varied inductive effect and biological behavior. This study described the clinico-pathologic patterns of these tumors in the Sudan.

**General objectives:** were to enhance knowledge on the nature of these tumors.

**Specific objectives:** were to study the clinical features and revision of their histological characteristics.

**Materials:** The records of (n=151) patients with histologically confirmed odontogenic tumors during the period 1994 –2002, were retrieved from various institutions. The institutions were Khartoum Teaching Dental Hospital, Omdurman Teaching Hospital, Medical Corp Hospital, and Police Central Hospital. Information about age, sex, site of tumor, symptoms, signs and their duration were obtained.

Paraffin blocks of the same patients were retrieved from the National Health Laboratory and El Zahrawi Laboratory. Sections (5 µm thick) from these blocks were prepared and stained by Hematoxylin and Eosin for re-evaluation.

**Methods:** The clinical data collected were analyzed and assessed. Furthermore, the previous diagnosis on each of these sections was revised. The revision of both the clinical and histological data was according to the
Histological Typing of Odontogenic Tumors, WHO (9).

**Results:** Of all tumors ameloblastoma was the most frequent odontogenic tumor (97 cases, 64.7%), followed by odontogenic myxoma (12 cases, 8%), odontogenic fibroma (11 cases, 7.3%), ameloblastic fibroma (8 cases, 5.3%), and adenomatoid odontogenic tumor (7 cases, 4.7%). Most ameloblastomas affected male (male: female, 1.7:1) and occurred more commonly in the mandible (55 cases, 87.3%). The age mean and range were 35.6 and 14 - 80 years, respectively. The clinical features were similar to other African series. The most common histological type of ameloblastoma was the conventional (solid) type (35 cases, 79.5%), followed by unicystic ameloblastoma (9 cases, 20.5%), and desmoplastic ameloblastoma (one case, 2.3%). The most common histological pattern in the conventional type was the follicular (20 cases, 58.9%), followed by plexiform (7 cases, 20.6%), mixed (5 cases, 14.7%), granular cell type (2 cases, 5.9%), and basal cell type (one case, 2.5%). However, no acanthomatous type was noted. The two cases (10%) of recurrent ameloblastoma were found to be follicular type, while recurrence of plexiform type was not observed.

Odontogenic myxoma showed mandibular preponderance with no segmental predilection. The mean age and range were 22.2 and 11 – 45 years, respectively. Unlike reports from other parts of the World, there was a pronounced male predilection (male: female, 5:1). Under the microscope odontogenic myxoma showed predominant myxoid tissue but some have unusually high amount of collagen fibres and calcifications.

Odontogenic fibroma showed female predilection (7 cases, 63.6%). The mean age and range were 30 and 13 – 80 years, respectively. Both proliferative odontogenic epithelium and collagenous fibrous tissue showed variation in quality and quantity. However, adenomatoid odontogenic tumor
showed female predilection and affected older age group than those reported from other African countries. This is probably due to the nature of this tumor, which is rather slower in its growths as well as symptom presentation.

The only squamous odontogenic tumor in this study occupied the posterior maxilla and showed extensive invasion of the maxillary and palatal tissues and the deeper intracranial area. This feature was contrary to the benign biological behavior of this tumor.

**Conclusion:** This study documented the relative frequency and the clinico-pathological patterns of (n=151) of odontogenic tumors during the period 1994 –2002. The material was retrieved from the following settings Khartoum Teaching Dental Hospital, Omdurman Teaching Hospital, Medical Corp Hospital, and Police Central Hospital, the National Health Laboratory, and El Zahrawi Laboratory. The data was assessed and reclassified according to the Histological Typing of Odontogenic Tumors, WHO (Kramer et al., 1992).

Statistical analysis showed that ameloblastoma was by far the most frequent tumor followed by odontogenic myxoma, odontogenic fibroma, ameloblastic fibroma, and adenomatoid odontogenic tumor. The odontogenic myxoma and adenomatoid odontogenic tumor showed unique clinico-pathological patterns that probably peculiar to Sudanese patients. A further study in the tumorigenesis of these lesions is important.
ملخص البحث

تُستَهَدَ 60% 6.8% من الأنسجة المحيطة بالإنسة. 8% من الأنسجة الخارجية. 3% من الأنسجة الخارجية. و 30% من الأنسجة الخارجية. 8.6% من الأنسجة الخارجية. 30% من الأنسجة الخارجية. 8.6% من الأنسجة الخارجية.

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Chapter One

1.1. Introduction

Broca was the first to classify odontogenic tumors according to the stage of the development of the tooth. He designated the term odontomes for such lesions \(^{(1)}\). Malassez and Bland-Sutton classified these tumors according to the tissue of origin \(^{(2)}\) and later, Gabell modified the classification by including the non-neoplastic cysts of dental origin. Gabell classified the lesions into three main groups of odontomes, epithelial odontomes, composite odontomes, and connective tissue odontomes which arisen from the dental epithelium, dental epithelium and dental mesenchyme, and dental mesenchyme only respectively \(^{(3)}\).

Thoma and Goldman (1946) proposed a classification based on the structure and inductive effect exerted by one tissue upon another. They divided odontogenic tumors into 3 main groups \(^{(4)}\).

The American Academy of Oral Pathology modified, similarly, these classifications and divided the tumors into 3 groups \(^{(5)}\). Pindborg introduced a classification based on the inductive changes produced by dental tissues on each others, leading to omitting some lesions and including others \(^{(6)}\).
The WHO classification of odontogenic tumors in 1971 was dependent on the structural characteristics of these lesions. They divided these lesions into 5 groups (7). However, Reichart and his colleagues (1983) proposed a classification of odontogenic tumors using four histogenetic cell groups (epithelial, ectomesenchyme, neuroectodermal, and mesenchyme), depending on the biological behavior of these tumors (8).

The latest WHO classification on odontogenic tumors (1992) was based on the tumor behavior and the degree of inductive interaction. They further divided the lesions according to the types of odontogenic tissues involved. They divided the benign lesions into 3 groups. These were odontogenic epithelium without odontogenic ectomesenchyme, odontogenic epithelium with odontogenic ectomesenchyme, and odontogenic ectomesenchyme with or without included odontogenic epithelium (9). Over the years there have been attempts to produce a logical classification, however, uncertainties remained because of the complexity of the tissues and the rarity of some of these lesions (9).

Tumors of odontogenic origins were lesions that derived from odontogenic epithelium or ectomesenchyme or both; that was part of the tooth or tooth–forming apparatus (10). However, some of these lesions had been defined as true neoplasms while the nature of the remainder was still
uncertain \(^{(11)}\). A universally accepted histological characterization of odontogenic tumors in an orderly manner would be helpful to the clinicians as well as to the pathologists \(^{(12)}\). It appeared that such a unified concept was not achievable. Several histological classifications, however, had been proposed.

Early reports from Africa odontogenic tumors were suggested to be common particularly ameloblastoma \(^{(13-17)}\). Kov-J and Laing (1966) studied the tumors of the jaws in Ghana; of the 70 jaw tumors 22 cases (31.4\%) were tumors of odontogenic origin \(^{(18)}\). While, reports from Nigeria showed that the relative frequency of odontogenic tumors range between 15\% to 30\% of all tumors of the mouth and jaws \(^{(15,17)}\).

On the other hand in Zimbabwe the relative frequency of odontogenic tumors was 8.6\% of all orofacial lesions \(^{(19)}\). While in Egypt, odontogenic tumors were found to be 16.9\% and 20\% of all oral tumors in adult and children, respectively \(^{(20)}\). In Uganda the relative frequency of odontogenic tumors ranged between 21.8\% to 37.2\% \(^{(21)}\). In Tanzania, odontogenic tumors comprised about 12.2\% of all oral tumors and tumor like conditions \(^{(22)}\).

Odontogenic tumors were similarly common in Asia. In China, odontogenic tumors accounted for 71.9\% of the tumors of the jawbones \(^{(23)}\).
In Japan they constituted 13% of all oral tumors\textsuperscript{(24)}.

In South America, odontogenic tumors were not common and showed variable pattern of occurrence. In Brazil, odontogenic tumors constituted 2.4% of all oral and maxillary lesions\textsuperscript{(25)}, in Chile accounting for 1.29% of all oral neoplasms\textsuperscript{(26)}, and in Mexico accounting for 3.7% of the oral cavity tumors\textsuperscript{(27)}. In North America, odontogenic tumors were rare in general, and comprised about 1.3% and 1.11% of all oral biopsy specimens in USA and Canada, respectively\textsuperscript{(28,29)}.

Early reports from the Sudan showed that odontogenic tumors constituted 12.9% of all oral neoplasms\textsuperscript{(30)}. Recently, the relative frequency of odontogenic tumors was found to be 8.6% of all oral neoplasms\textsuperscript{(31)}. Odontogenic tumors showed varieties of histological and biological behavior. Establishment of a good knowledge on the nature of these lesions is important and necessary.
1.2. Literature Review

1.2.1. Ameloblastoma:

Broca was the first to report the tumor and he considered it as an overgrowth of the dental germ (1). Falkson in 1879 was the first who gave a detailed description of ameloblastoma and he proposed a theory of enamel organ origin (32). Several terms has been given to the tumor since its first description. There were up to fifty synonyms in English language literature. The most common names were adamantine epithelioma, adamantinoma, adamantinoblastoma, epithelial odontome, and multilocular cyst. Churchill and Ivy introduced the term ameloblastoma in 1930 (33).

Robinson reviewed the literature in 1937, in which he surveyed three hundred and seventy nine cases from the literature (34).

On the other hand Small and Waldron reviewed the world literature, and collected 1000 reported cases of ameloblastoma, including that of the
Robinson’s series. They found that the mean age was 38.9 years. Of the 925 reported cases 752 cases (81.3%) were located in the mandible and 173 cases (18.7%) were found in the maxilla. Seventy percent of these cases were seen in the molar-ramus area, 20% occurred in the premolar area, and 10% occurred in the symphyseal area. In the maxilla the molar area was the most affected region. In Sweden, Larsson and Almeren (1978) reviewed the Swedish Cancer Registry during 1958-1971; about 49 cases of ameloblastomas were found. These cases were revised according to the classification criteria of odontogenic tumors of WHO 1971. Ameloblastoma constituted 3.2 % of all the 969 bone tumors and 43.7 % of all the 71 skull and face tumors during the same period. The males were more affected than females with a ratio of (1.4:1). The age range and mean were 2 – 93 and 50.5 years, respectively.

The mandible was found to be the preferable site and constituted about 74 % of the cases while only 8 cases (26 %) were seen in the maxilla. Microscopically, re-examined ameloblastomas showed follicular, plexiform and acanthomatous type in addition to a mixture of the three types. Moreover, three cases were cystic ameloblastoma and six cases were arising in the wall of a cyst.

It was found that ameloblastoma at peripheral location of the lesion
grew in trabecular and cellular pattern rather than follicular or cystic pattern. Moreover in peripheral location squamous metaplasia was frequently seen. They found difficulty in re-confirming the malignant ameloblastoma because of the lack of the well defined histological criteria \(^{(36)}\).

In Poland, Stypulkowska and his colleagues (1998) studied 164 cases of odontogenic tumors seen between 1956 and 1996. Ameloblastoma was found to be the most common odontogenic tumor accounted for 36.6 % of all cases \(^{(37)}\).

In USA, Regezi and his associates studied 706 cases of odontogenic tumors during the period 1934 – 1975. Ameloblastoma was found to be the second frequent odontogenic neoplasm after odontoma and comprised about 11% of the cases \(^{(28)}\). On the other hand Yamane and his colleagues (1984) in New York studied 48 cases of non-inductive epithelial odontogenic tumors retrieved from files of New York University Dental Center from 1964 to 1982. He showed that 38 cases (79%) were ameloblastoma and 7 cases (18 %) were pseudoglandular variant of ameloblastoma. While, one case (2.6%) was a peripheral ameloblastoma and another case (2.6 %) was a unicystic type. The age range and mean were 18-81 years and 51 years, respectively. The average age for plexiform type was 36 years and hence 15 years younger. There was a slight female predilection with male to female ratio
The mandible was the preferable site and 82% were seen there, while 13% were seen in the maxilla. The posterior molar region was the preferable site for both jaws. Six cases (15.8%) were located in the anterior jaw and a case (2.6%) was bilateral. The authors realized that the pseudoglandular variant occurred frequently in older patients and in the posterior region.\(^{(38)}\)

Reichert and his colleagues (1995) reviewed the world literature for ameloblastoma. They analyzed 3677 cases of ameloblastoma retrieved from the literature during 1960-1993. The median age worldwide was found to be 35 years with age ranged from 4-92 years. There was a slight male predilection (53%). The mean age at presentation for blacks, Caucasians, and Asians was 28.7, 39.9, and 41.2 years, respectively. It was also found that the average age was 27.7 and 39.1 years for developing countries and industrialized countries, respectively. The mandible was frequently affected than maxilla and was involved 5 times as frequent as the maxilla. The anterior area was more affected in black than other races. It was also found that the average duration was 2.3 years.\(^{(39)}\)

In USA, Waldron and his colleagues studied 116 cases of ameloblastoma. They showed that there was a slight male predilection (1.2:1). The age range and mean were 15 to 92 years and 43.8 years, respectively.
The peak incidence was in the third and fourth decades of life, with another slight peak at the 7th decades of life. The mandible was the preferable site (88 %). The molar – ascending ramus area was the most common site and comprised about 61 % of the cases. Thirteen percent were seen in the premolar area, while the anterior area was the least affected area constituting about 11 % of the cases. In the maxilla, the anterior region was the most common site (50 %), followed by the posterior region (43 %), and the premolar area (7%)\(^{(40)}\). In Canada, Daley and his associates found that ameloblastoma was the second frequent odontogenic tumor after odontoma and constituted about 13.52 % of the cases\(^{(29)}\).

In Mexico, Mosqueda and his associates studied 349 cases of odontogenic tumor during 1960– 1996. Ameloblastoma was the second frequent odontogenic tumor after odontoma and accounted for 34.6 % of all cases\(^{(27)}\). However, in Chile, Ochsenius and his colleagues reviewed the records of 28,041 specimens from 1975 to 2000 in the Oral Pathology Referral Institute. They found that 362 cases (1.29 %) were found to be odontogenic tumors. Ameloblastoma was the second frequent odontogenic tumor (20.4%), preceded by odontoma. The clinical presentation was found to be similar to those reported in the North American series but different from those found in Asian and African series\(^{(26)}\).
In Brazil, Santos and his associates retrieved 127 cases of odontogenic tumors from 5,289 oral and maxillary lesions diagnosed at the Division of Oral pathology; Federal University of Rio Grande do Norte during the period 1970–1999. It was found that ameloblastoma was the second frequent odontogenic tumor after odontoma, accounting for 30.7 % of all odontogenic neoplasms \(^{(25)}\). Martins studied 45 cases of odontogenic tumor which constituted 0.78 % of all oral cavity benign tumors during 1980-1997 at the stomatology and Head and Neck services of Heliopolis Hospital, Sao Paulo, Brazil. Ameloblastoma constituted 64.4% (29 cases) of all odontogenic tumors. He found that the mandible was the preferable site (93.1%). There was no racial nor gender predilection. The peak incidence was in the age group 20 to 30 years old \(^{(41)}\).

In Asia ameloblastoma was the commonest odontogenic tumor. Gunhan and his colleagues studied 409 cases of odontogenic tumors in Turkish population. Ameloblastoma was found to be the most common odontogenic tumor and constituting 36.5% of these lesions. The mandibular molar region was found to be the commonest site \(^{(42)}\). In China, Wu and his coworkers retrieved 204,683 specimens from University Department of Pathology, during 1963-1982. They found that 114 cases were tumor of the jawbones, and 82 cases were odontogenic tumors. Ameloblastoma was also
found to be the most common odontogenic tumor accounted for 62% of the cases \(^{(23)}\). In another study in China, Lu and his associates studied 759 cases of odontogenic tumors from the records of West China University of Medical Sciences during the period 1952–1994. Ameloblastoma was found to be the most common odontogenic tumor constituted 58.6% of all cases studied \(^{(43)}\).

In Malaysia, Siar and his colleagues studied ameloblastoma during 1967 and 1991 in Institute for Medical Research, Kuala Lumpur. It was found that there was a male predilection \((1.14:1)\), and 72% of the cases were in the second, third and fourth decades of life. The mean age was 30.8 years, and the mandible was also the preferable site \((93\%)\). It was also found that about 337 cases \((86.4\%)\) were conventional ameloblastoma, 49 cases \((12.6\%)\) were unicystic ameloblastoma and 4 cases \((1.03\%)\) were peripheral ameloblastoma \(^{(44)}\).

In India, 122 primary tumors of the jaw during 17-year-period were retrieved from records of the department of pathology at Baranas Hindu University, Varanasi. Ameloblastoma was found to be the most common odontogenic tumor and accounted for 46% of all odontogenic neoplasms. On the other hand ameloblastoma was similarly found to be the most common odontogenic neoplasm in Indian children and accounted for 30.8%
of all odontogenic neoplasms. The age ranged between 10-12 years old. The duration of the lesion varied from 1.5 to 6 months. However, all pediatric ameloblastomas were found to be solid type \(^{(45)}\).

In Japan, five hundred and three specimens were collected, of which 80% were benign neoplasms, 13% were odontogenic tumors. Ameloblastoma was also found to be the commonest odontogenic neoplasm accounted for 53% followed by odontoma (33.3\%) \(^{(24)}\). In another study in Japan, Utsumi studied 21,702 specimens from which 41 cases (22.65\%) were odontogenic tumors. Ameloblastoma was found to be the most common odontogenic neoplasm \(^{(46)}\). On the other hand Tanaka studied the oral neoplasm in Japanese children and he retrieved and analyzed 105 records of which odontoma was the most common odontogenic tumor accounting for 42.4 \% followed by ameloblastoma (33.3 \%) \(^{(47)}\).

In Korea, Kim and Jang (2001) studied 71 intrasosseous ameloblastomas. The age range and mean were 11-70 and 30.4 years, respectively. There was a male predilection (54.9 \%). The mandible was the preferable site and accounted for 78.3 \% of cases. The most common radiographic feature was unilocular radiolucency with well-demarcated border that accounted for 59.2\% of all cases. Of these 19.7\% showed multilocular radiolucency and 2.8 \% showed soap-bubble appearance, in
particular\textsuperscript{(48)}.

In Africa, early reports suggested that odontogenic tumors were not uncommon particularly ameloblastoma \textsuperscript{(13)}. In Ghana, Kovi-J (1966) reviewed and analyzed 7945 histologic specimens and 1159 postmortem examination. Odontogenic tumors accounted for 31.4%. Ameloblastoma was found to be the most common odontogenic tumor, accounting for 91\% (20 cases). It was also found that the peak incidence of ameloblastoma was at the age group 30-40 years\textsuperscript{(18)}.

In Nigeria, several reports concerning odontogenic tumors were documented. Mosadomi (1975) in a series of 29 cases of odontogenic tumors excluding cysts, fibromas and myxomas, found that ameloblastoma was the commonest odontogenic tumor constituting 65\%. Moreover, he showed that there was no gender predilection. The mandible was the most favorable site and all cases of ameloblastoma were seen there. The peak incidence was seen in the third decade of life with 47.4 \% of the series were reported there, and the mean age was 30 years. He concluded that the high incidence of jaw tumors was unjustified and he thought it could be due to harvesting phenomenon \textsuperscript{(15)}.

Daramola and his associates (1975) studied ameloblastoma in Nigerian children under the age of 18 years. He reviewed 70 patients of
which 16 patients (25.7%) were children under 18 years of age. The male-female ratio was (1.7:1) with significant male predilection. The age range and mean were 5-17 and 13.4 years, respectively. The average duration of the tumor was one year and 8 months. The mandible was the preferable site accounting for 97%, of which 46.7% were located in the symphyseal region of the mandible. About 31.25% were associated with unerupted teeth and one case (6.25%) was seen in the maxilla at the premaxillary region.

On the other hand Adekeye (1980) surveyed about 109 cases of ameloblastoma in Nigerian patient during the period 1970 to 1978. All patients were seen and treated at the maxillofacial unit of the Ahmadu Bello University Hospital, Kaduna, Nigeria. It was shown that there was a male predilection with male to female ratio was 1.7:1. The age mean and range were 30.5 and 9-70 years, respectively. About 67.9 % were seen in the 3rd and 4th decades of life. The mandible was the preferable site (99.1%) and 55% were seen in the horizontal ramus that represented the common site in the mandible. The duration of the neoplasm ranged from one month to 20 years with mean duration 4.2 years. The common presenting feature was swelling, although pain was seen in 8.3 % of the cases. About 34.9% of cases showed erosion of cortical bone with penetration of periosteum and invasion of adjacent soft tissues. However, 2.8% of cases were associated
with ulceration and 2.8% were associated with unerupted teeth. Radiologically, most of the cases showed multilocular radiolucency; honeycomb or soap bubble appearance was the most radiographic presentation accounting for 89.9% of the cases. The remaining 10.1% were unilocular radiolucency\(^{(50)}\).

Although Mosadomi (1975) raised suspicion of the previous results concerning the high prevalence of ameloblastoma in blacks (Mosadomi, 1975), Sawyer and his colleagues (1985) carried a study comparing the Nigerian population with those of American blacks and whites. The data was collected from two sources, from Lagos, Nigeria and from Richmond, Virginia (USA). From Nigeria, they analyzed files of the biopsy service at Lagos University Teaching Hospital during 1969–1979 and 871 records were collected. While, from USA, files biopsies of Virginia Commonwealth University were reviewed and 29037 black and white patients were analyzed of which 5095 were black, 23942 were white, during period 1957 – 1979. In Nigeria, they analyzed 871 records and about 46 cases (5.3%) were ameloblastoma. While in America, they analyzed 29037 patient records showed that only 30 cases (0.1%) were ameloblastoma. Of the 5095 American Black patient records reviewed only 17 cases (0.33%) were ameloblastoma. While, reviewing 23942 American White patient records
showed that 10 cases (0.04 %) were ameloblastoma. The age range and mean of Nigerian patient were 9-60 and 31.8 years, respectively, while, the age range and mean of the American Black were 11-69 and 39.4 years, respectively. On the other hand the age range and mean of the American White were 17-87 and 44.2 years, respectively.

In Nigeria, about 61.9 % of the cases were seen in the first three decades of life and 66.7 % were seen between 21-50 years of age. In the American blacks 43.8% were seen in the fifth decade of life and 49.9 % were seen between 21-50 years of age. On the other hand 60% of American white patients were seen in the fifth decade of life and also about 60% were seen between 21-50 yrs of age. In Nigeria, the males were more affected (52.2%) while in American Blacks females were more affected (58.8%). On the other hand American white males were more affected and accounted for 60% of the cases. The mandible was the preferable site for both populations; however, the posterior segment of the horizontal ramus of the mandible was frequently involved in American Black accounted for 47.1%. While in Nigerian patients the anterior segment was more affected and accounted for 39.1 % of the cases. On the other hand no segmental predilection was found in American Whites. Only one case in Nigeria patients was seen in the maxilla, another case was seen in extraosseous location, and only one case
below the age of 10.

In Nigerian patients the size of the lesion was larger with longer duration while American patients showed smaller size with shorter duration. It was concluded that there was statistically significant difference in pattern of occurrence between Nigerian population and American Blacks. It was found that the occurrence rate was higher in American Blacks than American Whites\(^{(16)}\).

Taiwo and his associates in 1990 reviewed 203 orofacial swelling of Nigerian children below 16 years of age, during period January 1978-31 December 1988 (11-year period). They found that males were frequently affected than females with 70% were seen in males. It was also found that 10% of the orofacial swellings were odontogenic tumors while 20% were non-odontogenic. Ameloblastoma constituted 5% of all orofacial swelling and 50% of all odontogenic tumors. The mandible was the preferable site and 90% of ameloblastoma were seen there. The peak in incidence was in age group 11-16 years in which 90% cases were seen\(^{(51)}\).

More recently, Arotiba and his associates in 1997 reviewed 415 cases of oral and jaw tumors from the records of cancer registry of University College Hospital, Ibadan, of which 128 cases were odontogenic tumors during the period period 1980-1994. These cases were retrieved from
department of oral and maxillofacial surgery, the histopathology record books of the department of oral pathology and the. Ameloblastoma was the most common odontogenic tumor comprised 58% of all odontogenic neoplasms and 18.3% of all oral neoplasms. There was a male preponderance with male to female ratio 1.5:1. the age range and mean were 8-72 years and 33 years, respectively. The peak incidence was in the third decade (30%). The mandible was also the preferable site accounted for 91% of cases, while maxillary lesions constituted 9%. The horizontal ramus was similarly the preferable site (80%). The duration range and median were 1 month to 15 years and 21 months, respectively. Radiographically, multilocular radiolucency was the most common picture accounted for 90%, while unilocular radiolucency was seen in 10% of the cases (17).

In South Africa, Shear and Singh (1978) tried to find a correlation between the ameloblastoma and the dentigerous cyst in Black and White population. They surveyed the Witwatersrand area at which the population at risk was 1567280 blacks and 974390 whites. They also surveyed all pathology registries in the region during the period 1965-1974. About 42 cases of ameloblastoma were found of which 36 cases (85.7%) were in Blacks and 3 cases (7.14%) in Whites. The Black males were frequently affected with male to female ratio was (1.8:1). While White females were
more affected with male to female ratio (1:2).

The highest incidence of ameloblastoma was found to be 5.1 per million per year in age group 40-49 years for Black male, and 3.33 per million per year in age group 30-39 years for black female. While the highest incidence for White male was 1.15 per million per year at age group 20-29 yrs and 1.82 per million per year for White female at age group 40-49 yrs. It was also found that the average annual incidence for Black male for all ages was 2.41 per million per year and 2.14 per million per year for Black female. While the average annual incidence for White male for all ages was 0.21 per million per year and that for White female was 0.41 per million per year. Therefore, the incidence of ameloblastoma was higher in Black than White particularly males. While, the incidence of dentigerous cyst was very much higher in Whites than in Blacks.

They concluded that ameloblastomas and dentigerous cysts were unrelated lesions, and hence ameloblastoma did not arise in a pre-existing dentigerous cyst. Considering the etiological factors they suggested environmental factors for development of ameloblastoma such as carcinogens that might be present in diet habits of Black\(^{(52)}\).

In Zimbabwe, Chidzonga and his colleagues (1996) surveyed 1723 orofacial biopsies, during a 10-year period, at the Department of Surgery,
Medical School, University of Zimbabwe, Harare. About 148 cases of odontogenic tumors were found and hence odontogenic tumors were not uncommon in Zimbabwe constituted about 8.6 % of all orofacial swelling. On the other hand ameloblastoma was found to be the most common odontogenic tumor constituted 6.8 % of all orofacial swelling and 79.1 % of all odontogenic tumors \(^{(53)}\).

In Tanzania, Simon and his colleagues (1998) reviewed odontogenic tumors collected from four referral centers during 1982-1997. It was found that odontogenic tumors constituted about 12.2 % of all oral tumors and tumor-like conditions. Ameloblastoma was the most common odontogenic tumor constituting 73.7% of all odontogenic tumors. About 50% of the patients were presented after 3 or more years \(^{(22)}\).

In Egypt, El Sharkawy (1982) studied the oral tumors in children and compared the results with those of adult. He reviewed 238 cases at the department of oral surgery, Faculty of Oral and Dental Medicine and Department of Pediatric Surgery, Faculty of Medicine at Cairo University during 1974-1979. He found that about 55 cases were pediatric lesions affecting children below 15 years of age while 183 cases were lesions affected adult. Concerning the adult patients 145 cases (79.2 %) were benign tumors and 38 cases (20.8 %) were malignant tumors. While in Egyptian
children below 15 years of age, 90% of oral tumors were benign and 9.1% were malignant tumors. In adult 31 cases (16.9%) were odontogenic tumors while in Egyptian children 11 cases (20%) were odontogenic tumors from all orofacial tumors. On the other hand ameloblastoma was found to be the most common odontogenic tumor, while in children ameloblastoma was not observed below the age of 15 years. Therefore, odontogenic tumors were rare in children and constituted about 26.2%, while 73.8% of odontogenic tumors were seen in adult (22).

In Ethiopia, Christos studied 21 cases of ameloblastoma. There was a male predilection with male to female ratio 2:1. The mean age was 26 years, although lesions were reported in children. The duration range and mean were 3 months to 12 and 4 years, respectively. The mandible was the preferable site constituted about 56% of cases (54).

Early reports from Sudan showed that odontogenic tumors were common. Hickey-B (1958) studied retrospectively 1,337 specimens received during 1935 to 1954 in the Stack Laboratories for Medical Research. Ameloblastoma constituted 12.9% of all oral tumors (30). While, El-Abdin and Ruprecht (1989) studied 7 Sudanese patients clinically diagnosed as odontogenic cyst, on revision showed histological features of unicystic ameloblastoma. The male to female ratio was 1:2. The age ranged between
15 to 56 years \(^{55}\). Later, Idris and his associates (1995) studied the epidemiology of oral neoplasms in Sudan. They showed that odontogenic tumors were common in Sudan and constituted about 8.6% of oral neoplasms. Ameloblastoma was the most common odontogenic tumor comprised about 88.3% of all odontogenic tumors, and about 7.5% of all oral neoplasms. Most cases of odontogenic tumors were referred from West and South Sudan \(^{31}\).

### 1.2.1.1. Histology of ameloblastoma:

In Sweden, Larsson-A and Almen-H \(1978\) reviewed all ameloblastomas reported to the Swedish Cancer Registry during the period 1958-1971. They found that sections showed predominantly follicular, plexiform or acanthomatous as well as a mixture of the three types. Three cases (9.7%) were unicystic ameloblastoma. They realized that the tumor frequently grew peripherally in a trabecular and cellular manner rather than in a follicular or cystic pattern. Also at the periphery of the tumor ameloblastoma showed squamous metaplasia. Of these re-examined sections the plexiform pattern was the most common pattern constituting 45.16% (14 cases), followed by the follicular pattern 12.9% (4 cases). About three cases showed mixed follicular and plexiform pattern while 4 cases were acanthomatous type. However, three cases were cystic ameloblastoma \(^{36}\).
On the other hand Anneroth and Hansen (1982) studied the variation of keratinization in odontogenic tumors. They described 2 cases of acanthomatous ameloblastoma, and showed that it could be confused with other keratinizing odontogenic tumors. They also showed that acanthomatous ameloblastoma can be differentiated by the presence of squamous metaplasia, with sometimes keratin formation, and the presence of polarized preameloblast-like columnar or cuboidal cells on the periphery of the epithelial odontogenic components, a widened basement membrane and absence of pleomorphism (56).

Sapp and Jesvold (1983) studied hyaline deposits in odontogenic tumors; and found that 40.3% of the cases were showing these hyaline deposits. While 38.2% cases of ameloblastoma were showing these hyaline deposits. The deposits were found predominantly in plexiform type and in cystic areas of other types. The deposits were located in the area of the basement membrane, close to the palisaded columnar epithelial cells with their nuclei away from the basement membrane. In cystic areas, appeared as a thickened basement membrane within which occasionally focal areas of calcification were seen. They concluded that these hyaline materials were not specific to any one lesion; however, they were frequently found in number of epithelium-containing odontogenic tumors. Therefore, the
presence of these hyaline deposits was not diagnostic for any entity lesion (57).

Yamane and his colleagues (1984) studied 48 cases of non-inductive epithelial odontogenic tumors retrieved from files of New York University Dental Center. They found that about 38 cases (79%) were ameloblastoma of which 7 cases (18%) were pseudoglandular variant of ameloblastoma. The follicular pattern showed granular cell changes, squamous metaplasia, or cystic degeneration or combination of these patterns in the center of the follicles. They realized that the plexiform pattern was similar to dental lamina stage of tooth development. They showed a rare pattern called pseudoglandular ameloblastoma, which composed of gland-like structures. This pseudoglandular variant constituted 19% (7 cases) of the ameloblastoma cases studied. They showed that about 10 cases (20%) of pseudoglandular variant occurred in combination with other variants. They also showed a new variant (glycogen-rich ameloblastoma). This variant was formed of peripheral columnar cells, intermediate cuboidal and central stellate reticulum cells. The peripheral and intermediate cells showed vacuolated clear cytoplasm containing glycogen and glycoprotein vacuoles similar to that of mucoepidermoid tumor. They attributed the pseudoglandular pattern to two mechanisms. The first mechanism where the
epithelial cell proliferate into parallel rows enclosing the connective tissue which undergone liquefactive degeneration. The second mechanism where that the peripheral cells differentiate into stellate reticulum cells forming elongated enamel follicle-like structures with cystic degeneration of the central stellate reticulum cells forming microcysts. They concluded that the histogenesis of ameloblastoma was similar to salivary gland tumors since both originated from pluripotential cells of the embryonic epithelium. Hence ameloblastoma recapitulated different stages of teeth development\(^{(38)}\).

Waldron and El-Mofty in 1987 studied 116 ameloblastomas from the Department of Oral Pathology, Washington University School of Dental Medicine. Histologically, they classified ameloblastoma in to follicular and plexiform or mixed patterns. They observed that the follicular pattern was the most common (45%), followed by plexiform pattern (12%), the mixed follicular-plexiform pattern (9%), basal cell variant 3%, and the granular cell variant that was the least common pattern (1%). They noted some degree of squamous metaplasia but not enough to group them as acanthomatous variant. The basal cell type formed of anastomosing strands or islands of basaloid epithelial cells in a mature fibrous connective tissue stroma. The peripheral cells showed palisading arrangement but without reversed nuclear polarity. The central epithelial cells showed spindle cells
with foci of squamous metaplasia\textsuperscript{40}.

WHO (1971) described ameloblastoma histologically in two patterns: follicular and plexiform. However, both patterns were frequently seen in the same tumor. The follicular pattern formed of discrete islands of epithelia resembled enamel organ. These follicles were composed of a central mass of polyhedral cells resembling stellate reticulum. A layer of ameloblast-like cells, which were cuboidal or columnar, lined the follicle. Cystic degeneration of stellate reticulum-like cells was frequently seen. On the other hand, the plexiform pattern was formed of interconnecting epithelium, which lined by a layer of cuboidal or columnar ameloblast-like cells surrounding stellate reticulum-like cells. Stromal degeneration was also commonly seen. The follicular pattern might show extensive squamous metaplasia or the granular cell subtypes. The connective tissue in the follicular pattern might undergo extensive hyalinization. The authors described the basaloid pattern, which referred to as basal cell type of ameloblastoma\textsuperscript{7}. While, in the revised edition WHO (1992) described a rare variant referred to as keratoameloblastoma showed extensive keratinization. While the papilliferous keratoameloblastoma was formed of microcysts lined by parakeratinized epithelium containing keratin or non-keratinized with papilliferous pattern. On the other hand they divided the
unicystic ameloblastoma into 3 histological patterns (9).

Reichart and his colleagues in 1995 reviewed the world literature for ameloblastoma. They reviewed 3677 cases, covering the period 1960-1993. They showed that about 74.9% were classic ameloblastoma, 15.7% were unicystic ameloblastoma, and 9.4% were peripheral ameloblastoma. They found that 15.5% were of mixed histological appearances. The follicular pattern was the most common histological pattern in ameloblastoma (33.9%), followed by the plexiform pattern (30.2%), the acanthomatous type (11.3%), the granular cell type (3.5%), and the desmoplastic type (1.4%). Similarly basal cell type was seen in 1.4% of the cases. While the papilliferous keratoameloblastoma was rare and constituted 0.1% of the cases (39).

In China, Wu and his associates (1985) surveyed 204,583 specimens from University Department of pathology, Hong Kong during period 1963-1982. They found that 114 tumors were affected the jawbones and 51 cases (62%) were ameloblastoma. Microscopically, these tumors displayed a direct connection of the tumor epithelium with the oral mucosal epithelium and showed a better prognosis. On the other hand basaloid pattern showed more aggressive biological behavior. They also observed that the neoplastic epithelial cells infiltrated the normal bone surrounding the tumor mass (23).
In Malaysia, Siar and Ng in 1993 reviewed about 401 cases of ameloblastoma from the Division of Stomatology, Institute for Medical Research, Kuala Lumpur during the period 1967-1991. They found that 337 cases (86.4%) were conventional ameloblastoma, 49 cases (12.6%) were unicystic ameloblastoma, and 4 cases (1.0%) were peripheral ameloblastoma. The plexiform pattern was similarly the most common type constituting 34.2%, followed by the follicular pattern constituting 16.5%. While, 17.7% were showing a mixture of both follicular and plexiform patterns (44). In Korea, Kim and Jang (2001) studied 71 cases of ameloblastoma. The most common histologic pattern was plexiform and 16 cases had developed in a cyst (48).

In Africa, Kovi reviewed 7,945 histological specimens and 1,159 postmortem examinations at Korle Bu Hospital, Accra, Ghana from January 1962 through September 1965. He found that four cases (20%) were follicular type, 4 cases (20%) were plexiform type, 3 cases (15%) were designated as stellate type, and 2 cases (10%) were primitive type. Keratinization in area of stellate reticulum cells were observed in 2 cases (10%), while acanthomatous type accounted for 3 cases (15%). Only one case (5%) was shown to be granular cell type with large polyhedral cells containing granular eosinophilic cytoplasm. He described a case designated
as siderotic ameloblastoma, which histologically showed epithelial strands in a loose fibrous stroma. The peripheral and stellate cells were containing brownish granules when stained with hematoxylin-eosin stain. Gomori’s iron reaction was strongly positive in the cases and the authors assumed that the iron-containing pigment granules were taken up by phagocytosis of the broken down red blood cells or due to metabolic derangement (18).

In Nigeria, Mosadomi (1975) reviewed 200 benign and malignant neoplasms of the oral cavity. He found that the most common type was the follicular type 13 cases (68.4%), followed by plexiform type 3 cases (15.8%), and mixed type 15.8% (3 cases). Follicular pattern showed cystic degeneration in 12 cases (63.2%) and acanthomatous changes in 6 cases (31.5%). On the other hand the plexiform pattern showed stromal degeneration in 2 cases (10.5%) and one case (5.3%) was plexiform type showed haemangioameloblastoma. The based cell type was only one case (5.3%). However, one case (5.3%) showed atypia and focal dyskeratosis with loosely cellular and myxomatous stroma and without evidence of metastasis was reported (15).

Sawyer and his associates (1985) studied two populations, data from Nigeria at the Department of oral pathology and Biology, Lagos University Teaching Hospital during 1969-1979. And data form USA, Department of
Oral Pathology, School of Dentistry, Medical College of Virginia Commonwealth University. Microscopically, the follicular pattern was the most common in Nigerian patients (39.1%), followed by plexiform (19.6%), acanthomatous variant (13.0%), the mixed pattern (10.9%), the granular cell type (10.9%), and the based cell type was (6.5%). On the other hand the follicular type was similarly the most common pattern in American Blacks (35.4%), followd by the acanthomatous type (29.4%), the plexiform pattern (17.6%), and the mixed type (17.6%). However, no basal cell or granular cell types were reported in American Black in this series. In the American whites the follicular type was very common (50%), followed by plexiform type (20%), the acanthomatous (20%), and the mixed type (10%). Similarly no basal cell or granular cell types were recorded among the American white in this series. Recently, Namin and his colleagues studied the role of human papilloma virus as a possible etiologic factor of ameloblastoma and they found that the virus was significantly detected in fifty blocks of ameloblastoma compared to control group.

Regnani and his colleagues established a comparative study for data collected from Dental Hospital and Medical Hospital and they observed the bias in data coming from Medical Hospital. In Medical Hospital odontomas were underestimated while ameloblastomas were overestimated due to the
reference pattern of these lesions. While in Dental Hospital odontomas were frequently seen by dentists and treated without initial diagnostic biopsy\(^{59}\).

Philipsen and his associates in 2001 reported a case of desmoplastic ameloblastoma and reviewed the world literature and found 100 cases. They found that the relative frequency varied between 4-13% of all ameloblastomas. The age range and mean were 17-72 and 42.9 years, respectively. They noticed equal gender and jaw distributions, although 5.4% of cases were found in the molar mandibular area. The histological features were typical to that previously described in WHO classification\(^{(9,60)}\).

1.2.1.2. **Unicystic ameloblastoma**

There were several terms proposed for unicystic ameloblastoma, including mural ameloblastoma, ameloblastoma associated with odontogenic cyst, cystogenic ameloblastoma.

Robinson (1977) was the first to describe the term unicystic ameloblastoma to a distinct entity. He studied 20 cases of unicystic ameloblastoma, retrieved from the Diagnostic Pathology Services of University Hospital and the School of Dentistry, University of Alabama in Birmingham. He described a criterion to define ameloblastic epithelium. The first criterion was that the basal cells were columnar cells with hyperchromatic nuclei and the overlying cells were loose cells with absence of cohesiveness. The
second criterion was the presence of the down growths of the ameloblastic epithelia into the connective tissue wall. The third criterion was the presence of follicles similar to enamel organ in the connective tissue wall. Lastly, presence of intraluminal nodules formed of anastomosing cords and islands of epithelium.

These cases were divided into two groups. The first group was similar to dentigerous cyst while the second group was similar clinically and radiographically to residual or primordial cysts. The dentigerous group was all associated with unerupted third molar. In addition to that all cases in this group were seen in the mandible. In this group 10 cases (71.4 %) were seen in patients below 20 years old, while 12 cases (85.7 %) were seen in patients below 29 years of age. Only 2 cases (14.3 %) were seen in age group 40-49 years.

The recurrence rate for the first group was 25 % (8 cases) observed for a period from 5-14 years, all the cases were treated by enucleation. Two of the recurrent cases were free of the disease 4 years postrecurrence clinically and radiographically. The age range for the second group was 12-79 years and the recurrence rate was 33.3 % observed for a period from 4-13 years.

The author concluded that the criteria for ameloblastic epithelium were similar to that of Vickers and Gorlin (61). He also showed that the
average age was not in coincidence with that of solid ameloblastoma. The average age was found to be 27.7 years for both groups. He speculated that the lesion could arise from a pre-existing cyst or began de novo. He also concluded that unicystic ameloblastoma showed less aggressive biologic behavior than solid ameloblastoma. He finally recommended enucleation for treatment of unicystic ameloblastoma (62).

Gardner (1981) was the first to present the term plexiform unicystic ameloblastoma. This lesion did not fulfil the Vickers and Gorlin’s criteria for unicystic ameloblastoma (58). He studied 19 cases what he called plexiform unicystic ameloblastoma under the light microscope. He compared these lesions with unicystic ameloblastoma and plexiform ameloblastoma according to the Vickers and Gorlin's criteria (63).

He found that the age range for 19 cases was 12-58 yrs, and 8 cases (42.1%) were between 16-20 yrs. There was a male predilection with male to female ratio (1.7:1). Thirteen cases (68.4 %) were clinically diagnosed as dentigerous cyst. The mandible was the preferable site and constituted 94.7 % (18 cases) of the cases. The follow-up of three patients revealed that one lesion recurred 3½ years following curettage. Microscopically, these lesions showed network of anastomosing strands of stratified squamous epithelium in which the basal cells were not obvious. These lesions didn’t fulfill
Vickers and Gorlin’s criteria for ameloblastoma. The central cells were squamous with prominent cytoplasmic processes and these cells were widely separated by intercellular spaces. There was variable sizes of nodular epithelium proliferating into the lumen of the cyst. The connective tissue was delicate and contained numerous capillaries with scanty chronic inflammatory infiltrate. He also found that 47.4% (9 cases) were associated histologically with ameloblastoma ranging from prominent characteristics to only a small focus of ameloblastoma. He also observed that the epithelial proliferation fused with that of ameloblastoma in 2 cases.

He concluded that plexiform unicystic ameloblastoma although did not fulfil Vickers and Gorlin's criteria, it should be considered as an ameloblastoma. He recommended serial section for the specimens to disclose any associated characteristic ameloblastoma. However, he did not reach a conclusion concerning progression of plexiform unicystic ameloblastoma in to ameloblastoma. He considered that the biological behavior of this plexiform unicystic ameloblastoma could be similar to that of unicystic ameloblastoma with similar age distribution.

He divided the plexiform unicystic ameloblastoma into 2 groups. The first groups showed proliferation into the cystic lumen without the involvement of the surrounding connective tissue wall. This group treated by
enucleation with regular follow-up. While the second group in which the proliferating epithelial invaded the connective tissue, should be treated by marginal resection and regular follow-up. The lesion was treated by enucleation under general anesthesia with no postoperative complication. Follow-up for 3 years revealed that the patient was free of the disease. He finally introduced the term plexiform unicystic ameloblastoma to that lesion previously known as pseudoameloblastomatous change in the wall of a cyst or as ameloblastoma-like tissue\(^60\).

On the other hand McMillan (1981) studied five cases of ameloblastoma assumed to arise from the walls of dentigerous cysts. He had given certain evidence supporting his concept. He concluded that all dentigerous cysts should be examined histologically with care. He also pointed out that all his cases were of younger age patient with average age 14.8 years that differ from that of solid ameloblastoma. Hence, he suggested that ameloblastoma could arise in the wall of dentigerous cyst since dentigerous cysts were similarly seen in children and adolescents. All the cases showed some degree of inflammatory response which frequently seen in dentigerous cyst and not in uncomplicated ameloblastoma. Therefore, these lesions were more probably dentigerous cyst in which ameloblastoma developed. Also, he pointed out that the reduced enamel epithelium was
continueing with the cystic epithelial lining and hence this supported the concept that these lesions were similarly dentigerous cysts. He also showed that the lining epithelial of the cyst was continuous with that of ameloblastomatous epithelium.

Hence, he concluded that the cystic epithelium undergone ameloblastomatous transformation. He also showed that these cases fulfilled the Vickers and Gorlin’s criteria for ameloblastoma \(^{61}\). Follow-up for periods ranging from 1 to 7 years showed no recurrence. He considered these lesions as less aggressive variety of the neoplasm \(^{64}\).

In 1983 Gardner attempted to find the relationship of plexiform unicystic ameloblastoma to the conventional ameloblastoma. He studied 10 cases of plexiform unicystic ameloblastoma which showed concomitantly areas of other patterns of ameloblastoma. The age range was 12 to 23 yrs with average age 18.2 years. Also, all the cases were seen in the mandible and associated with unerupted tooth which is frequently mandibular third molar. The ratio of male to female was (1.5:1). One case showed unicystic ameloblastoma lined by columnar and hyperchromatic cells with islands of conventional ameloblastoma in the connective tissue wall.

A recurrent lesion showed plexiform unicystic ameloblastoma fulfilling the criteria of Vickers and Gorlin \(^{58}\). While a second case of
plexiform unicystic ameloblastoma when recurred showed conventional ameloblastoma. He concluded that the plexiform unicystic ameloblastoma was probably a histologically undifferentiated variant of conventional ameloblastoma. Of the nine cases reviewed only one case showed recurrence (65).

In another study Gardner and Corio in 1984 examined 46 cases of plexiform unicystic ameloblastoma that exhibited no areas of other histological patterns of ameloblastoma. Their main aim was to determine the biologic behavior of these lesions. They divided plexiform unicystic ameloblastoma into 2 groups. Those resembled dentigerous cyst radiographically called dentigerous group, and those that were not associated with unerupted teeth called non-dentigerous group.

All cases were seen in the mandible and the posterior region of the mandible was the preferable site. Of the unerupted teeth associated with the plexiform unicystic ameloblastoma the third molar was more frequently involved 71.1% (27 cases). There was a male predilection with male to female ratio (1.9:1). The mean age was 17.6 and 20.8 years for dentigerous group and non-dentigerous group, respectively. The mean age for all the cases was 17.9 years. The peak incidence was in the age group 10-19 years. Of the 28 cases treated by enucleation or curettage the recurrence rate
was 10.7% (3 cases) which was less than that of the conventional ameloblastoma (55% to 90%) when treated by curettage.

The author concluded that the plexiform unicystic ameloblastoma, therefore, should be treated by enucleation rather than marginal or segmental resection. He also recommended treatment by marginal resection or segmental resection when the proliferating epithelial cells infiltrated the periphery of the connective tissue wall of the cyst, and concluded that the biologic behavior of the plexiform unicystic ameloblastoma was similar to that of other unicystic ameloblastoma \(^{(66)}\). Unicystic ameloblastoma can be mistaken clinically for a globulo-maxillary cyst and Van Wyk and his colleagues (1986) presented such a case \(^{(67)}\).

In 1987 Waldron and El-Mofty studied 116 ameloblastoma from the Department of Oral Pathology, Washington University School of Dental Medicine. They found that 12 cases were unicystic ameloblastoma comprising about 11% of all cases of ameloblastoma seen. The male were more affected with male to female ratio (1.75:1). The age range and mean were 17-36 and 22 years, respectively. All cases were located in the mandible, 9 cases (75%) were seen in the posterior mandible, 7 cases (58.3%) were in the third molar area, and 4 cases (33.3%) were associated with unerupted mandibular third molar. Histologically, all lesions showed
fibrous connective tissue lined with polarized columnar or cuboidal epithelial cells which showed reversed nuclear polarity and cytoplasmic vacuolization close to the basement membrane. Some cases showed epithelial projection in the fibrous tissue wall with islands of follicular ameloblastoma. 3 cases (25%) showed intraluminal proliferation with feature identical to that of plexiform unicystic ameloblastoma. 

On the other hand Ackerman and his colleagues in 1988 presented a histological classification of unicystic ameloblastoma. They studied 57 cases of unicystic ameloblastoma retrieved over a 30-year period from the Department of Oral Pathology, South African Institute for Medical Research, Johannesburg. There was a male predilection with male to female ratio was (1.3:1). Also there was black preponderance (51cases, 89.5%). The mandible was the preferable site and constituted 52 cases (91.2%), and the mean age was 23.8 years, which was younger than that of the multicystic type.

They classified unicystic ameloblastoma into 3 groups: group-1 (42%) the cyst lined by variable non-descriptive epithelium, group-2 (9%) was formed of cystic lesion with intraluminal plexiform proliferation of epithelium, and group-3 (49%) showed cystic lesion with invasion of epithelium into the fibrous cyst wall in either follicular or plexiform
patterns. They recommended enucleation for group-1 and -2 lesions while for group 3 they recommended more aggressive surgery similar to that of conventional ameloblastoma. However, they disagreed with the concept that unicystic ameloblastoma originated from pre-existing odontogenic cysts\(^{(68)}\).

Punnia and Moorthy in 1989 presented a case of unicystic ameloblastoma in a 29-year-old Caucasian woman, which was previously treated 21 years ago with mandibular swelling associated with unerupted first and second mandibular molars. The lesion at that time had been misdiagnosed as dentigerous cyst.

Later, she was presented with multilocular radiolucency at the same site. Biopsy showed follicular ameloblastoma. However, re-examination of previous biopsy showed unicystic ameloblastoma. While biopsy taken from the recurrence showed follicular ameloblastoma.

They recommended enucleation with cryotherapy for the remnants or removing small thickness of the surrounding bone of the cavity. He also recommended excision of the periosteum when there was cortical bone erosion. He recommended long-term follow-up\(^{(69)}\).

On the other hand Philipsen and Reichart (1998) reviewed 193 published cases of unicystic ameloblastoma. They found that the unilocular pattern was more common than the multilocular radiographic pattern. The
mean age of the dentigerous variant (16.5 yrs) was 20 years younger than that of the non-dentigerous variant which was age 35.2 years. There was a male predilection for the dentigerous variant with male to female ratio (1.5:1). There was a female predilection for the non-dentigerous variant with male to female ratio was (1:1.8). The mandible was the preferable site, and 50-80% of the cases were associated with tooth impaction and the mandibular third molar was more frequently involved. The mean age for unilocular impaction-associated tumors was 22 years while for the multilocular tumors unrelated to an impacted tooth was 33 years. The authors recommended enucleation for unicystic ameloblastoma, while the other types that showing intramural growths should be treated radically similar to solid ameloblastoma (70).

In China, Li and his associates (2000) studied the clinicopathological features and biologic behavior of 33 cases of unicystic ameloblastoma retrieved from the Department of Oral Pathology, Peking University School of Stomatology, Beijing. They found that unicystic ameloblastoma accounted for 19% of all cases of ameloblastoma seen during a 15-year period. They found that the male to female ratio was (1.75:1), and the age range and mean were 8-60 and 25.3 years, respectively. The peak incidence was in the second and third decades, and the mandible was the preferable
site (90.9%).

Histologically, they found that 24.2% of the lesions were simple cystic lesion, 30.3% were showing intraluminal nodules and 45.5% showed infiltration of the cyst capsule with tumor islands. The recurrence rate was 35% for 29 cases followed for more than four years. They also found that the recurrence rate was 35.7% for those types showing invasion of the cyst fibrous capsule, and the recurrence rate was 6.7% for other types. They concluded that the long-term follow-up was mandatory. They insisted on the high recurrence rate for those lesions showing invasions of the cyst capsule as well as maxillary lesions (71).

In USA, Ord and his colleagues (2002) studied ameloblastoma in children at the Department of Oral and Maxillofacial Surgery, University of Maryland. They found that 9 cases (81.8%) were unicystic ameloblastoma. The average age was 15.5 years. They reviewed the literature and found that 76.5% of ameloblastoma in the West were unicystic ameloblastoma, while in African children 19.5% were unicystic ameloblastoma. In African children, it was more common in the mandibular symphysis (44.2%), in Western children only 5.8% were located in the symphyseal area. They showed that the recurrence rate for 20 children reviewed was 40%. The authors concluded that unicystic ameloblastoma was commonly seen in
children. They also recommended more aggressive surgery for unicystic ameloblastoma showing mural invasion \(^{(72)}\).

### 1.2.1.3. Peripheral ameloblastoma

Peripheral ameloblastoma had been given various terms such as mucosal, extra-medullary, extraosseous and soft tissue ameloblastoma. Peripheral ameloblastoma was the commonest peripheral odontogenic tumor.

Gardner defined peripheral ameloblastoma as an odontogenic tumor with histologic characteristics of an intra-osseous ameloblastoma but occurring solely in the soft tissues covering the tooth-bearing parts of the jaws. However, peripheral ameloblastoma arises from the surface epithelium or from residues of the dental lamina lying outside the bone \(^{(73)}\).

Gardner studied 21 cases of peripheral ameloblastoma of which 5 were reported as basal cell carcinoma of the gingiva. He found that the age ranged between 23-82 years and with peak incidence in the fourth and the fifth decades. There was a male predilection with a male to female ratio of (1.6:1), and the mandible was the preferable site in 13 cases (61.9%).

Histologically, some cases showed mixed pattern while others showed follicular and even acanthomatous patterns well as basal cell types. However, the acanthomatous pattern was the predominant pattern seen in 17
cases. One case showed basaloid pattern with keratin pearls and acanthomatous area while another case showed calcifications within the acanthomatous areas. Eight cases showed continuity of the tumor cells with the surface epithelium, while five cases showed a band of connective tissue between the tumor cells and the surface epithelium. One case showed ghost cells and multinucleated giant cells that had phagocytosed ghost cells. There were lymphocytic infiltrate associated with epithelial islands seen in 13 cases, a feature that was absent in intra-osseous ameloblastoma but was seen in the basal cell carcinoma of the skin.

The authors postulated two sources from which peripheral ameloblastoma originated. Those showed continuity with the surface epithelial were arisen from the surface epithelium, while those which showed no direct connection with surface epithelium were arisen from the rests of Serres, remnants of the dental lamina within the gingiva. He also suggested multifocal origin in some cases showed continuity at considerable length of epithelium. He concluded that excision of the lesion down to the periosteum would be curative for peripheral ameloblastoma. He also insisted on the pathologist examination of the margins and suggested periodical re-examination of the surgical site. He finally concluded that peripheral ameloblastoma and basal cell carcinoma of the gingiva represented the same
Buchner (1987) reviewed the English literature for the peripheral epithelial odontogenic tumors. He analyzed the data concerning the clinical and histological features of 48 well-documented cases. He found that about 26 well-documented cases of peripheral ameloblastoma occurring in the gingiva and the alveolar mucosa. He also found 4 cases of peripheral ameloblastoma in extra-gingival locations and 6 cases that were reported as basal cell carcinoma of the gingiva and the alveolar mucosa. He excluded the extra-gingival peripheral ameloblastoma because their origin was controversial. He found that the age ranged from 23-92 years with a mean age of 52.3 years. There was a peak incidence in the fifth and sixth decades (44%). There was a male predilection with male to female ratio (1.7:1).

The mandible was the preferable site in 59% of cases. The canine-premolar area was more frequently involved accounting for 57.9% (11 cases), and of these 9 cases (47.4%) were seen in the lingual gingiva. In the maxilla the molar-retromolar area was more frequently involved accounting for 76.9% (10 cases). Clinically, peripheral ameloblastoma was commonly seen as a painless, sessile, firm and exophytic growth. The surface of the lesion was smooth, although granular or pebbly-surface lesions were reported. The color of the lesion was usually pink but in some cases it was
red or even dark red.

The duration range from 1 month to 2 years with a mean duration of 1 year. The size ranged from 0.3 to 2 cm in diameter with a mean size 1.3 cm.

Radiographically, twenty nine cases showed no radiologic bone involvement while 2 cases showed cupping or saucerization of the underlying bone. Histologically, the acanthomatous type was the most commonly seen type. In 19 cases (70%) there was a direct continuity between the tumor and the surface epithelium, of which 7 cases (26%) showed multifocal continuity. On the other hand 8 cases (30%) showed a band of connective tissue between the tumor and the surface epithelium.

For the histogenesis of peripheral ameloblastoma the author considered two sources of origin, the remnants of dental lamina (rests of Serres) or the surface epithelium. He recommended surgical excision and close long-term follow-up for the peripheral ameloblastoma. He considered peripheral ameloblastoma as aggressive tumor that did not invade the bone. He also recommended pathological examination of the surgical margins.\(^{(74)}\)

Waldron (1987) studied 116 cases of ameloblastoma, of these 6 cases (5.17%) were diagnosed as peripheral ameloblastoma. He found that 4 of the 6 cases were located in the posterior portion of the mandibular gingival or
alveolar mucosa. One case was located in the posterior palatal mucosa. He found that all the cases were presented as sessile, soft tissue nodules covered by stratified squamous epithelium. They were small and ranged from 0.5-0.7 cm in diameter. Histologically, 3 cases showed follicular pattern and 2 cases showed plexiform pattern. While one case was predominantly showing basal cell type. He noted that 5 cases showed direct continuity with the oral epithelium (40).

1.2.2. Squamous odontogenic tumor:

Pullon and colleagues were the first to describe this lesion as specific entity in 1975. They reported six cases and introduced the term squamous odontogenic tumor. One of these cases showed multiple involvement of the four quadrants. Histologically, all cases showed scattered islands of stratified squamous epithelium in which basal cells were seen at the periphery. Mature collagenous connective tissue was noted in which squamous island were invested. Cystic degeneration and intraepithelial calcification were also noted. In four cases the maxillary cuspid area was involved. While in the mandibular lesions the posterior region was involved. The age range and mean were 11 to 42 and 25.8 years, respectively. There was gender predilection. One case showed recurrence following surgical treatment (75).

McNeill and colleagues (1980) reported a case of multiple
involvement of all four quadrants by squamous odontogenic tumor. Histologically, it showed fibrous connective tissue containing squamous epithelial islands with basal cell layer at the periphery. Vacuolated squamous cells were noted, which was coalesced forming cystic spaces. Laminated calcified deposits in the epithelial islands, occasional keratinization, pseudoepitheliomatous hyperplasia, and scattered inflammatory cells were noted.

They concluded that the tumor represented a hyperplastic proliferation of mature benign squamous islands or epithelial rests of Malassez. They suggested that there was an interaction between the epithelial islands and the connective tissue or there was an abnormal stimulus acted on both tissues. They considered the lesion as a fibrosquamous dysplasia. The treatment recommended was local and total excision with no evidence of recurrence after one year \(^{(76)}\).

On the other hand Goldblatt, L.I. (1982) and his colleagues reported 5 cases of squamous odontogenic tumor and reviewed the literature. They collected eleven acceptable cases from literature. They found that the age range and mean were 11 to 67 and 35.8 years, respectively. There were three cases which showed multiple site involvement. Fifty percent of the cases were located in the mandible and 37.5% were located in the maxilla. The
premolar-molar area was the commonest area involved in the mandible while the incisor-canine area was more frequently involved in the maxilla.

Histologically, the tumor was formed of epithelial islands of variable shapes. These islands were not surrounded by columnar palisaded polarized epithelium but with uniform squamous epithelial cells. The laminated calcified materials were seen in the connective tissue or within the epithelial islands. Intraepithelial eosinophilic crystalloidal structures were seen in some cases and they suggested that these structures represented prekeratin or glycoprotein materials. The authors suggested that the tumor more probably arisen from the epithelial rests of Malassez, from the surface mucosa, or from the epithelial rests of Serres (77).

Norris and his colleagues (1984) reported a case of squamous odontogenic tumor that showed malignant transformation into low-grade epidermoid carcinoma (78).

Baden and his coworkers (1993) reported three cases of squamous odontogenic tumor including the first completely extraosseous case. The first case showed histologically islands of squamous epithelium with intracellular keratin and microcysts. The stroma showed hyalinization and hyaline collars around the epithelial islands. A fibrous capsule surrounding the tumor was seen. Microcysts and occasional cuboidal peripheral cell were
The authors reviewed the literature and found 32 acceptable cases of squamous odontogenic tumor. There were twelve cases of mural squamous odontogenic tumor within the walls of the cysts. Analysis of the literature showed that the age range and mean were 11 to 74 and 40 years, respectively. There was a peak incidence in the third decade. The male to female ratio was (1.7:1). The mandible was more frequently affected especially the molar-premolar area. While in maxilla the anterior region was more commonly affected. The treatment was local excision and 8 cases treated by en bloc resection with recurrence rate 6.25%.

They suggested that the different presentation of the gingival lesions might be due to pluripotential properties of undifferentiation cells that persisted to adulthood. Therefore, the peripheral lesion derived from the overlying gingival epithelium (79).

Leider and his coworkers reported 3 cases of multicentric squamous odontogenic tumor in 3 black siblings. It was attributed to the familial history of these patients. Since they were seen at the teeth bearing area they suggested that the tumor originated from the rests of Malassez within the periodontal ligament. However, two cases showed direct connection with the surface epithelium, hence suggesting the possibility of more than one source
of this tumor \( ^{(80)} \).

Ide and colleagues (1999) reported the first case of intraosseous squamous cell carcinoma arising in association with a squamous odontogenic tumor in a 53-year-old man. Histologically, the lesion showed the typical feature of squamous odontogenic tumor while a few foci of epithelial islands showed atypical features of squamous cell carcinoma. Two months later the typical findings of intraosseous squamous cell carcinoma were noted. They considered this lesion as the malignant variant of squamous odontogenic tumor \( ^{(81)} \).

1.2.3. Calcifying epithelial odontogenic tumor

Franklin and Pindborg (1976) reviewed the world literature for calcifying epithelial odontogenic tumor and collected 113 cases. They showed that the tumor was rare in comparison with ameloblastoma. The ratio of ameloblastoma cases to calcifying epithelial odontogenic tumor was 17:1 in India, 17:1 in USA and 13:1 in England. The frequency of occurrence of the neoplasm in the Armed Forces Institute of Pathology was 0.17% to 1%. The age range and mean for intraosseous lesions in females were 11 to 78 and 40.26 years, while the age range and mean for males were 8 to 92 and 40.56 years, respectively. The mean age for all cases at diagnosis were 40.41 and 31.4 years for the extraosseous and intraosseous lesions,
respectively. The peak incidence was in the third decade. There was a female predilection with 2:3 male to female ratio for the extraosseous lesions. However, the intraosseous type showed an equal sex distribution.

Sixty eight percent of cases were located in the mandible and 32% were located in the maxilla. The extraosseous cases were located frequently in the anterior region. About fifty two percent of cases were associated with an unerupted tooth or teeth and 10% of cases had an equivocal history of a related tooth or teeth. There were predilections for the molar-premolar region for the intraosseous type.

Clinically, the common presenting feature was a painless swelling. Other presenting features included nasal stuffiness, epistaxis, headaches, and proptosis.

Radiographically, the neoplasm showed unilocular or multilocular radiolucency with well or poorly defined border. Diffuse radiopacities were seen within the radiolucency. The recurrence rate was 14%, which was lower than that of ameloblastoma. The treatment ranged from simple enucleation or curettage to hemimandibulectomy. The follow-up periods ranged from few months to over 30 years. The authors recommended marginal resection with a rim of apparently normal tissue as a successful treatment procedure.
Ai and associates (1982) studied nine cases of calcifying epithelial odontogenic tumors retrieved from Shanghai, China. There were seven intraosseous and 2 extraosseous lesions. Six cases were seen in the mandible and one in the maxilla. The two extraosseous tumors were located in the mandible. The presenting feature was swelling of the jaw with displacement of the involved teeth. The extraosseous tumors were seen as firm enlargement of the gingiva with ulcerated surface or as an epulis.

Radiographically, three cases showed monocystic radiolucencies and 4 cases revealed mutlicystic radiolucencies. Both features showed small radiopacities and embedded teeth. Microscopically, the authors described four different patterns of calcifying odontogenic tumor.

The frequency of occurrence of this tumor was 1.8%. The male to female ratio was (1:3.5). The age ranged from 20 to 64 years with average age of 34.2 years. The authors suggested that the eosinophilic homogeneous material was amyloidal. Furthermore, they supported the hypothesis that the tumor arises from the reduced enamel epithelium\(^{(83)}\).

Ng and his associates in 1996 reviewed the clinicopathological characteristics of 13 cases of calcifying epithelial odontogenic tumor in the Malaysian patients over a 29-year period. Of the thirteen cases there was only one peripheral lesion. There was a female predilection with female to
male ratio was (1.6:1). The age range and mean were 19-16 years and 31.8 years, respectively. The maxilla was the preferable site in 77% of cases. Most cases (66.7%) were diagnosed clinically as dentigerous cyst. One case showed extensive calcification and clear cell differentiation (84).

Recently, Philipsen and Reichart (2000) reviewed the world literature and found 181 published papers on calcifying epithelial odontogenic tumor. They found that 94% were intraosseous and 6% were extraosseous variants. Radiographically, the intraosseous type showed irregular, unilocular or multilocular radiolucent area with variable degree of radiopacity. Sixty percent were associated with an unerupted tooth or odontoma. The relative frequency of the neoplasm was 1.2%.

The mean age was 34.4 and 38.9 years for the extraosseous and the intraosseous types, respectively. Both variants showed equal sex distribution. The mandible was more involved than maxilla with a maxilla to mandible ratio of 1:2 and the commonest site was the premolar-molar region.

Histologically, the authors showed the typical histological picture of the tumor. However, there were histological variants which include cementum-like component, clear cell variants, tumor-containing langerhans' cells, combined epithelial odontogenic tumor, and tumor with myoepithelial
cells. The authors couldn’t reach a conclusion about the nature of the amyloid-like material. The authors suggested that the tumor originated from a complex system of dental lamina or their remnants.

For the treatment of the tumor the authors recommended enucleation with a margin of macroscopically normal tissue, and maxillary lesions should be treated more aggressively \(^{(85)}\).

Aviel and his associates (2000) studied the nature of the amyloid deposit in calcifying epithelial odontogenic tumor using histologic, immunohistochemical, and ultrastructural techniques. It was found that the amyloid deposit was negative for basement membrane components positive for all cytokeratin stains. They concluded that the amyloid deposit was derived from filamentous degeneration of keratin filaments produced by the tumor squamous epithelium. Furthermore, this degeneration was part of the developmental or the aging process of the tumor \(^{(86)}\).

1.2.3.1. Peripheral calcifying epithelial odontogenic tumor

The peripheral CEOT was extremely rare Takeda and his co-workers (1983) reported a case of peripheral calcifying epithelial odontogenic tumor. They presented a case in a 31-year-old Japanese woman, located on the right upper molar region. The lesion appeared as well-circumscribed, firm, and reddish mass. Clinically was diagnosed as gingival epulis. The lesion was
surgically excised with the adjacent bone.

Histologically, the lesion showed the typical histological features of the calcifying epithelial odontogenic tumor. However, they showed club-like elongation of the rete pegs of the oral epithelium. These elongations showed direct continuity with the tumor cells. There was no evidence of bony involvement.

They speculated that the tumor appeared to arise from the remnants of dental lamina. The other possible origin was the basal layer of the gingival epithelium, which was provoked by unknown stimuli (87).

Houston and his associates in 1997 reported 2 cases of the extraosseous calcifying epithelial odontogenic tumor. They reviewed the English language literature and found 9 well-documented cases. They showed that the age range and mean were 12 to 64 and 34.4 years, respectively. Six of the tumors involved female patients and five involved male patients. Seven cases were located in the mandible and four cases were seen in the maxilla. Histologically, six of the lesions showed a predominant clear cell component. No recurrences were evident after simple surgical excision (88).

1.2.4. Ameloblastic fibroma

Cahn and Blum in 1952 postulated that ameloblastic fibroma
developed first into ameloblastic fibro-odontoma, and eventually into complex odontoma \(^{(89)}\). However, Shafer in 1955 separated this lesion from ameloblastoma and considered the it as a separate entity with distinct clinical and histological features \(^{(90)}\).

Eversole and his colleagues (1971) proposed a histogenetic theory for the development of mixed odontogenic tumors. They postulated that individual mixed odontogenic tumor arises at related coincident stages of the normal odontogenesis. These tumors were completely dependent on the presence of differentiation factors, which were not produced by these tumors. Therefore, the individual tumor proliferated without further differentiation. They postulated that the inductive materials represented overproduction of basal lamina. They disagreed with the concept that ameloblastic fibroma progressed into odontoma \(^{(93)}\).

Trodahl (1972) studied 24 cases of ameloblastic fibroma retrieved from the Armed Forces Institute of Pathology. He found that the age range and mean were 18 months to 41 years and 15.5 years, respectively. There was no gender predilection. The posterior mandible was the preferable site (63%), and the most presenting feature was swelling.

Radiographically, the most frequent radiographic picture was multilocular radiolucency, and 75% of the cases were associated with
unerupted teeth. Most of the cases were treated with conservative surgical procedures and the recurrence rate was (43.5%). The author assumed that this high recurrence rate was possibly due to incomplete excision of these lesions. Microscopically, he found no connection between the histological picture and the clinical behavior \(^{(92)}\).

On the other hand Slootweg (1981) analyzed the interrelationship of the mixed odontogenic tumors. He collected thirty three cases of ameloblastic fibroma, ameloblastic fibro-odontoma, and odontomas. He reviewed the world literature for mixed odontogenic tumors. He found a male predilection for ameloblastic fibro-odontoma male (56 %) with a mean age of 8.1 years, affecting younger age group unlike ameloblastic fibroma and odontomas. The mandible was the preferable site for ameloblastic fibro-odontoma in 54% of cases. Furthermore, complex odontoma and compound odontoma were very commonly seen in the maxilla and usually in the anterior region. On the other hand ameloblastic fibroma was frequently seen in the posterior region of the mandible.

The author concluded that ameloblastic fibroma did not progress into ameloblastic fibro-odontoma and therefore, the concept that ameloblastic fibroma developed into ameloblastic fibro-odontoma was invalid. According to age, site, and gender distribution, ameloblastic fibro-odontoma was
considered as an immature complex odontoma. He suggested that compound odontoma represented an early alteration of the odontogenic tissues, which showed histogenetic and morphogenetic differentiation. While, ameloblastic fibroma showed altered development in later life, with no morphogenetic and little histogenetic differentiation\(^\text{(93)}\).

Van Wyk and van der Vyver (1983) studied the epithelial-connective tissue interface in an ameloblastic fibroma with dentinoid formation. They found that the epithelial-connective tissue interface showed 4 different morphological patterns, namely cellular stroma, cell-free zone, amorphous hyaline material zone, and dentinoid material zone. Ultrastructurally, these zones corresponded to different stages of tooth development. They assumed that epithelial differentiation was a prerequisite for dentinoid formation. They concluded that all features that were noted represented an abortive inductive feature\(^\text{(94)}\).

Gardner (1984) attempted to clarify the interrelationships of the mixed odontogenic tumors. He disagreed with the concept that ameloblastic fibroma developed into an odontoma. He gave two reasons for that, the first was that ameloblastic fibroma was usually found in older age group than odontoma. Secondly, longstanding and recurrent ameloblastic fibroma did not show any further maturation. He thought that some of these lesions that
had been diagnosed as ameloblastic fibroma were actually developing odontomes. He considered that the ameloblastic fibro-odontoma represented two entities. One lesion that represented the developing odontoma and the second lesion represented ameloblastic fibro-odontoma. He noted that the distinction between these two entities was only achievable by clinical means (95).

Tatemoto and his colleagues (1988) studied four cases of ameloblastic fibroma immunohistochemically. They observed two types of epithelial cells: undifferentiated odontogenic cells and common ameloblastoma cells. The undifferentiated cell was strongly positive with keratin and PAS-positive. While ameloblastoma cells were slightly keratin positive and PAS-negative. The fibroma cells were strongly positive for vimentin, slightly positive for keratin, and negative for desmin. They realized the co-expression of keratin and vimentin in fibroma cells (96).

However, Becker and his colleagues in 1992 also studied 4 cases of ameloblastic fibroma immunohistochemically. They showed strong distribution of collagen type VI over collagen type I. The undulin was not expressed in the ectomesenchymal tissues except around capillaries and in areas with high cellularity. They suggested that the epithelial cell islands invaded adjacent mesenchymal tissue leading to formation of
ectomesencymal tumor stroma that was different from the surrounding mesenchyme \(^{(97)}\).

The WHO recently defined ameloblastic fibroma as a neoplasm composed of proliferating odontogenic epithelium dispersed in a cellular ectomesencymal tissue that resemble the dental papilla, and with varying degrees of inductive changes and dental hard tissue formation \(^{(9)}\).

Yamamoto and his colleagues (1995) studied mixed odontogenic tumors immunohistochemically. They observed that all epithelial cells were positive for cytokeratin. Tenascin was positive in dental papilla-like mesenchymal tissue, and negative in myxomatous areas and connective tissue. Ameloblastic fibroma showed vimentin positive. They suggested that this tumor had developed at the early stage of tooth formation. While ameloblastic fibroma and odonto-ameloblastoma were both positive for proliferating nuclear cell antigen. They concluded that these two tumors showed higher proliferation potential among the mixed odontogenic tumors \(^{(98)}\).

Philipsen and his colleagues (1997) reviewed the world literature of mixed odontogenic tumors and odontomas. They added 134 cases of odontoma. They proposed a hypothesis regarding the pathogenesis and relationship between these lesions. They suggested two different lines from
which the tumor developed. The first line was the neoplastic one represented by the tumor ameloblastic fibroma and ameloblastic fibrodentinoma. While, the second line was the hamartomatous line that was called the developing complex odontoma. The second line was further divided into three groups. The first group represented ameloblastic fibroma and ameloblastic fibrodentinoma which represented the first stage in the developing complex odontoma line. The second group was the ameloblastic fibro-odontoma which represented the second stage in the developing complex odontoma line. While the third group was the fully mineralized complex odontoma that represented the end stage of the development. The authors suggested that compound odontoma represented a malformation. It could be developed as a hyperdonta or as a hyperactivity of dental lamina (99).

Martins and coworkers (2001) compared the proliferative activity of ameloblastic fibroma with that of odontogenic myxoma. They used argyrophilic nucleolar organizer regions technique. They showed that there was no statistically significant difference between the epithelial and ectomesenchymal components. However, the proliferative index of ameloblastic fibroma was higher than that of odontogenic myxoma (100).

Lopez and his colleagues (2003) reported two cases of ameloblastic fibroma. They considered ameloblastic fibro-odontoma as a variant of
ameloblastic fibroma but containing an odontoma. They recommended conservative surgical approach for treatment of these lesions \(^{(101)}\).

### 1.2.5. Ameloblastic fibro-odontoma:

Hooker (1967) was the first who separated the group of lesions previously known as ameloblastic odontoma into 2 separate lesions. The first lesion was designated as ameloblastic fibro-odontoma that represented a combination of ameloblastic fibroma and complex odontoma. The biological behavior of this lesion was similar to ameloblastic fibroma. The second lesion was designated ameloblastic odontoma that later called odontoameloblastoma. This lesion was a combination of ameloblastoma and complex odontoma. The biological behavior was similar to the classic ameloblastoma \(^{(102)}\).

Ameloblastic fibro-odontoma was defined by WHO as a lesion resembled ameloblastic fibroma but with inductive changes yielding dentine and enamel \(^{(9)}\).

AL-Sebaei and his associates (2001) reported a case of ameloblastic fibro-odontoma. They recommended conservative surgical procedure with long-term follow-up. They recommended the separation of mixed odontogenic tumors and considered them as separate entities \(^{(103)}\).

While, Matsuzaka and his colleagues (2001) reported an unusual
case of ameloblastic fibro-odontoma arising from calcifying odontogenic cyst in a 23-year-old male patient. The lesion was located in left mandibular molar region. Histologically, showed cystic lesion lined by varying thickness epithelium with numerous ghost cells. A thick wall of connective tissue was separating this lesion from the attached ameloblastic fibro-odontoma \(^{(104)}\).

Chang and his colleagues (2002) reported a case of ameloblastic fibro-odontoma in a 26-year-old woman. The lesion was located in the posterior mandible. Radiographically, it showed well-circumscribed radiolucency except for the posterior aspect. Scattered foci of calcifications were noted coronal to the impacted lower third molar. Histologically, it showed features of both ameloblastic fibroma and complex odontoma. Dentine and enamel matrices were noted. The treatment was surgical excision with curettage and removal of the involved tooth. No evidence of recurrence was noted after one-year period. They recommended conservative surgical approach since the lesion was encapsulated and with little tendency to recur \(^{(105)}\).

1.2.6. Adenomatoid odontogenic tumor:

Philipsen and Birn (1969) were the first who gave the term adenomatoid odontogenic tumor. They reviewed the literature and found about 73 cases and they added 3 cases of their own to reach 76 cases.
They found that the tumor was more common in the second decade of life. The age range and mean were 5 to 38 years and 18 years, respectively. There was slight female preponderance (53%). The maxilla was the preferable location in 63% of cases, particularly, the anterior area (94.1%). Thirty-six cases (75%) were associated with embedded teeth and 37 cases (77%) were diagnosed primarily as dentigerous cyst. While two of the three cases found in the posterior region of maxilla and mandible were clinically diagnosed as dentigerous cyst. The cuspid area was by far the most involved area in which (62.1%) of the embedded teeth were canine teeth of both jaws.

They concluded that any cystic jaw lesions should be examined histologically. They suggested that the tumor probably originated from the reduced enamel epithelium surrounding the crown or from the epithelium lining the cystic cavity. They proposed the term adenomatoid odontogenic tumor for such lesions.

They also suggested that the tumor should not be classified as an epithelial odontogenic tumor without inductive changes, because of the presence of calcified and semicalcified structures in the neoplasm\(^{(106)}\).

Giansanti and associates (1970) reviewed the early literature for adenomatoid odontogenic tumor. They reviewed 108 cases and added 3
cases of their own to reach 111 cases.

They found that the mean age was 17.8 years. There was a female predilection (64%). The maxilla was more involved (65%) than the mandible (52%) especially the anterior part of the maxilla. Seventy four percent were associated with unerupted teeth, nine cases (14.1%) were associated with more than a single unerupted tooth. The canine was the most involved tooth in (68%) of cases, particularly the upper jaw. The growth of the lesion was stated as slowly progressive with mean duration 12 months.

The microscopical features were typical for adenomatoid odontogenic tumor. The extra-osseous lesions were five cases in which three were located on the maxilla and one on the mandible. All were seen in the anterior region. There were four females and one male. Most lesions were treated conservatively with no recurrence (107).

Courtney and Kerr (1975) studied twenty cases of adenomatoid odontogenic tumor retrieved from the files of the University of Michigan Department of Oral Pathology, during years 1949 to 1974. They found that there was female predilection with male to female ratio (1:3). The age range and mean were 5-37 and 16.5 years, respectively. The maxilla was the preferable site (60%), and (90%) occurred at or anterior to the canine teeth. Forty percent were associated with an impacted tooth and seven cases (70%)
were located between the roots of normally erupted teeth. All were presented as gingival swellings labial to the maxillary central incisors.

Macrosocopically, they were thick-walled sacular structures containing straw-colored fluid. On palpation gritty sensation was noted. Microscopically, all were encapsulated with the intraluminal projection of proliferating epithelium. They realized that there were 3 forms of epithelial proliferation. The first was the solid nodular form which forming small nests or rosette-like structures. The cells were spindle to cuboidal with abundant cytoplasm and large oval vesicular nuclei and one or two prominent nucleoli. Amorphous eosinophilic material was present between the epithelial cells.

The second form was the duct-like spaces that lined by a single row of cuboidal or columnar cells with a foamy cytoplasm and oval vesicular nuclei. These duct-like structures contained fibrillar eosinophilic material.

The third pattern was the trabecular or cripriform pattern, which formed of interlacing strands of epithelium one or two cells thickness. The cells showed condensed uniform eosinophilic cytoplasm with small round to oval-shaped hyperchromatic nuclei. This pattern was noted at the periphery of the lesions. There were varying amounts of calcifications with central calcification of the eosinophilic droplet and dystrophic calcification in the
connective capsule were noted.

The authors used special stains and concluded that the eosinophilic droplet and the eosinophilic material in duct spaces were connective tissue product rather than epithelium products. They also suggested that the droplet eosinophilic material was probably a continued basement membrane formation due to the absence of mesenchymal cells and hence accumulated as an eosinophilic droplet. While dystrophic calcification occurred in degenerated tumor cells.

They concluded that the lesion a hamartoma of residual odontogenic epithelium and should be treated conservatively\(^{(108)}\).

On the other hand Omara (1979) presented a case of adenomatoid odontogenic tumor in a 20-years old Sudanese male patient who referred to Khartoum Dental Hospital with a swelling in the mandibular right side of four months duration.

Microscopically, it showed the typical histologic feature AOT. However, areas of calcification were shown close to the connective tissue stroma. Concentric layers of adjacent cells surrounded the eosinophilic hyaline material were noted that were similar to cementicles. The treatment was curettage and enucleation. Clinically, it might be mistaken for a cyst\(^{(109)}\).
In Nigeria, Ajagbe and associates (1985) reported 13 cases of adenomatoid odontogenic tumor during 21 years period. There was an increased ratio of males. Also the mandible was the preferable site. They concluded that these results might be peculiar to Nigerians\textsuperscript{(110)}.

Awange (1991) reviewed the literature on adenomatoid odontogenic tumor from Africa and found 33 cases had been reported. He analyzed the data and found that the tumor affected young patients (10-28 years) with a mean age of 16.2 years. Females were affected more than males with female to male ratio (3:2). Sixty five percent of cases were located in the maxilla, (92.3\%) were located in the anterior region of the jaws, and (66.7\%) of the cases were associated with unerupted teeth. Clinically, the lesion showed painless swelling. They concluded that the histopathological confirmation was necessary since incorrect diagnosis might lead to mutilating surgery\textsuperscript{(111)}.

Philipsen and his associates (1991) reviewed the world literature and analyzed the data concerning adenomatoid odontogenic tumor. They classified the tumor clinically into central and peripheral variants. While the central variant was further divided into follicular (associated with embedded tooth) and extrafollicular (no embedded tooth) types. The tumor was slow growing with few or no symptoms causing tooth displacement. All variants showed identical histologic features. The central variant was the most
common variant accounted for (97.2%) and of which (73%) were the follicular type.

The male to female ratio was (1:1.9). The mean age was 17 and 24 years for the follicular and the extrafollicular types, respectively. Follicular variant was associated with single embedded tooth in (93.2%) of cases. The maxillary permanent canines accounted for (41.7%) of all cases, and all four canines for (60.1%) of adenomatoid odontogenic tumor-associated embedded teeth. The tumor was ranking the fourth among the odontogenic tumors. Conservative surgical excision was the treatment of choice with no recurrences (112).

El-Labban (1992) studied the nature of the eosinophilic and laminated masses in the adenomatoid odontogenic tumor under the light and electron microscope. She showed that the eosinophilic masses were formed of 3 types of fibrils: the thin collagen, the electron dense fibrils, and the amyloid filaments. The eosinophilic deposits resulted from changes surrounding blood vessels with deposition of collagen or fine filaments arranged into layers around these vessels. In all these areas there was a peripheral layer of fine filaments perpendicular to the epithelial basal lamina resembled early dentin formation that was reticulin positive.

The laminated masses represented calcified amyloid with or without
areas of calcified degraded collagen, which were indistinguishable from amyloid. She concluded that the fibrils forming the eosinophilic masses were related to changes in blood vessels. She also showed that calcification involved mainly the amyloid component (113).

Arotiba and his colleagues in 1995 studied thirteen cases of adenomatoid odontogenic tumor at Ibadan, Nigeria. The lesion accounted for (12.1%) of odontogenic tumors in their series. The male to female ratio was (1.2:1). The peak incidence was in the second decade of life with mean age 23.2 years. No recurrence was reported (114).

Takeda and Shimono in 1996 presented a case of adenomatoid odontogenic tumor that showed extensive induction of a dysplastic form of tubular dentin with globular calcification. They concluded that this case supported the categorization of adenomatoid odontogenic tumor as an odontogenic tumor consisting of a disorderly mixture of odontogenic epithelium and odontogenic ectomesenchyme with or without dental hard tissue formation (115).

More recently Arotiba and his colleagues in 1997 analyzed 57 cases of adenomatoid odontogenic tumor in Nigeria. They found that about 98.3% were intraosseous and 75% of which were follicular type. There was a female predilection with male to female ratio (1:1.4). Seventy five percent of
cases were seen in the second decade of life. Patients with follicular variant were relatively younger (15.2 ± 5.6 years) than those with extrafollicular variants (20.9 ± 13.8 years). The maxilla was affected nearly twice as frequent as the mandible (1.8:1). The canine tooth was frequently embedded in the tumor (76.9%). The follicular variants were most frequently located in the maxilla (76.3%), while the extrafollicular tumors were more commonly found in the mandible (69.2%) (116).

Philipsen and Reichart (1999) reviewed the world literature on adenomatoid odontogenic tumor from 1991 to 1999. They found that there were three variants two of them were intraosseous (follicular and extrafollicular) and the third was a peripheral variant. The intraosseous variants accounted for 96% of which 71% were follicular variant. The intraosseous variants were more commonly found in the maxilla than in the mandible with ratio (2.1:1).

More than two thirds were diagnosed in the second decade of life and more than half of the cases occurred within the teens (13-19 years of age). The female to male ratio was (1.9:1). There was a marked female predilection among certain Asian populations (3:1). All the four canines were involved in (59%) of cases and the maxillary canines alone accounted for (40%). The authors suggested that it was a benign hamartomatous, slow
growing lesion. They suggested that the lesion was being driven from the complex system of dental laminae or its remnants. The eosinophilic deposits (tumor droplets) were most probably representing some form of enamel matrix \(^{(117)}\).

Engin and his associates in 2001 presented a case of adenomatoid odontogenic tumor with unusual clinical features. The patient was 9-year-old male with a mass located in the posterior part of the maxilla. The lesion was unusually large (3×4 cm in diameter) and had perforated the buccal plate. The embedded tooth was the first premolar unlike the more typical canine. There was an irregular root resorption on the distal surface of the left central incisor. The treatment was enucleation and curettage and follow-up for 3 years showed no recurrence \(^{(118)}\).

### 1.2.7. Calcifying odontogenic cyst:

Gorlin and his colleagues were the first who described the calcifying odontogenic cyst as a distinct lesion derived from odontogenic epithelial remnants in 1962. They considered the lesion as a counterpart of the dermal calcifying epithelioma of Malherbe \(^{(119)}\).

Freedman and his associates (1975) reviewed the world literature and collected 64 cases and added 6 new cases. They found that the age range and
mean were 7 to 82 and 38.4 years, respectively. The male to female ratio was 1:1.18. Thirty-five cases were located in maxilla, 34 in the mandible, and one within the parotid gland. Seventy five percent were located anterior to the first molar and seventy percent of the maxillary lesions were below the age of 41, while 79.5% of the mandibular lesions were older than 41. Seventy eight percent were intraosseous and 21.5% involved soft tissue, and 23.6% of the intraosseous type were associated with embedded teeth and 11% were associated with odontomas. The duration ranged from 2 days to 7 years with mean duration 11 months.

Histologically, 88.5% were cystic and 11.5% were solid tumors. The characteristic feature was the presence of ghost cells. However, melanin pigmentation was seen in 5 of the 70 cases. The authors suggested that the tumor arisen from well-differentiated ameloblasts that were capable of production of enamel matrix represented by swollen individual ghost cells and by sheets of fused ghost cells, and proposed the term cystic calcifying odontogenic tumor (120).

On the other hand Praetorius and his associates (1981) studied the nature of the calcifying odontogenic cyst. They divided it into two entities, a cyst and a neoplasm. The cyst was further divided into 3 variants. The first was a simple unilocular cyst with moderate mural proliferations of
epithelium and no or spare amounts of dentinoid material (dysplastic dentin). The second variant was a unilocular cyst, which produced compound or complex odontomas in its luminal part, more rarely might produce an intramurally growing ameloblastic fibroma. The third variant was unilocular cyst with extensive luminal and ameloblastoma-like proliferations of epithelium. The calcifying odontogenic cyst was divided into intraosseous and peripheral. The neoplastic variant showed ameloblastoma-like strands and islands of odontogenic epithelium growing infiltratively in a mature connective tissue. Varying amounts of dentinoid material was formed in contact with the odontogenic epithelium. They suggested the term dentinogenic ghost cell tumor for the neoplastic type\textsuperscript{(121)}.

McGowan and Browne in 1982 studied 12 cases of calcifying odontogenic cyst. They found that the age range and mean were 11 to 87 and 44.25 years, respectively. There was no sex and site predilection. All the lesions were located anterior to the first molars. There were 10 intraosseous and 2 extraosseous lesions.

Histologically, ten cases were cystic, 2 were solid tumors, 9 cases showed areas of mineralization, and 2 cases with complex composite odontomes. While seven cases showed randomly arranged sheets of mineralized matrix contained cellular inclusions, four cases showed
dentinoid material. All lesions showed entrapped ghost cells and epithelial layer with prominent basal layer of tall columnar cells lining the cysts with reversed polarity of their nuclei, and these were overlied by a layer resembled the stellate reticulum. Some ghost cells undergone dystrophic mineralization. On aspiration, the cyst fluid was found to be similar to the patient's serum.

The treatment was removal by enucleation, and 2 cases showed recurrence. The authors suggested that the term calcifying odontogenic cyst was not satisfactory since not all lesions were cystic and not all lesions were calcified \(^{(122)}\).

Nagao and his associates (1983) reviewed the Japanese literature and found 23 well-documented cases of calcifying odontogenic cyst. The lesion affected both sexes equally. The mean age was 21 years. Clinically, the lesion was presented as painless swelling with cortical expansion. The maxilla was three times more involved than the mandible. No reported case of the peripheral type. Radiographically, the lesions were unilocular radiolucency with foci of radiopacities. Fifty percent of the cases showed unerupted teeth and root resorption. The treatment was simple enucleation with no recurrence \(^{(123)}\).

Siar and Ng (1988) studied the histological variation of the lining
epithelium of the calcifying odontogenic cyst under the light microscope. They showed two distinct types. The first was the classical highly differentiated lining epithelium characterized by a basal layer of columnar cells with nuclear polarization overlied by several layers of stellate reticulum-like cells which contained variable amount of ghost cells. The second type was the undifferentiated lining epithelium of 2-8 cell thick, non-keratinized, parakeratinized or orthokeratinized and resembled reduced enamel epithelium. They observed variations within the highly differentiated epithelium such as spindle-cell change, epithelial pearl formation, and ghost cell dominant type. Ten cases out of the thirty cases were associated with odontoma, three with melanin pigment, and one with clear cells. They concluded that the lesion had arisen either from the remnants of the dental epithelium or secondarily from the lining epithelium of pre-existing lesions (124).

Takeda and associates in 1990 studied the satellite cysts and odontogenic epithelial islands in connective tissue wall of unilocular type of calcifying odontogenic cyst. They classified the lesion into simple unicystic, odontoma-producing, and ameloblastomatous proliferating type. While the satellite cysts were classified into simple cystic, odontoma-producing, and ameloblastomatous.
They found that the histologic types of satellite cysts did not coincide with those of the main cystic lesions in some cases. Odontogenic epithelial islands with or without proliferating features were found in all cases with satellite cysts. Melanin pigment and melanocytes were seen in an ameloblastomatous satellite cyst out of the three pigmented calcifying odontogenic cysts (125).

On the other hand Buchner and his colleagues in 1990 studied and analyzed 17 cases of central calcifying odontogenic cyst. They showed that the lesions were usually diagnosed in the second decade of life. Both jaws were equally involved. Thirty five percent were associated with odontomes and of them 35% were associated with unerupted teeth. Most lesions were unilocular cysts while one was multilocular and one was a mixed lesion (partially cystic and partially solid). They recommended surgical excision and long-term follow-up for the treatment of calcifying odontogenic cyst (126).

Hong and associates in 1991 reviewed ninty two cases of calcifying odontogenic cyst and studied their nature and classified them on the basis of their clinicopathologic features. They divided the lesion into cystic and neoplastic. The cystic type was divided into 4 variants: the non-proliferative that was characterized by a simple unicystic structure, the proliferative
variant that characterized by a cystic structure with multiple daughter cysts, extensive ghost cell formations, and marked tendency for calcification. The ameloblastomatous variant was characterized by ameloblastoma-like, cyst-lining epithelium with ghost cells and calcifications. The fourth type was the calcifying odontogenic cyst associated with odontoma. While, the neoplastic type was divided into 3 variants: ameloblastoma ex-calcifying odontogenic cyst, which showed unifocal and multifocal intraluminal and intramural ameloblastoma proliferating from the calcifying odontogenic cyst-lining epithelium, the peripheral epithelial odontogenic ghost cell tumor, which occurred in the gingiva and resembled peripheral ameloblastoma except for clustered ghost cells in the central portion of epithelial islands and the presence of juxtaepithelial dentinoid, and lastly the third variant was the central epithelial odontogenic ghost cell tumor, which showed ameloblastomatous or adenomatoid odontogenic tumor-like epithelial clusters with ghost cell formation and juxtaepithelial dentinoid. They suggested that the ghost cells might be the result of coagulative necrosis (127).

Ng and Siar in 1995 analyzed morphometrically the epithelial components and the dentinoid components in 30 non-neoplastic calcifying odontogenic cyst. They showed two main types: an odontoma-producing type and a non-odontoma-producing variant. Morphometrically, the
odontoma-producing variant had more luminal and mural dentinoid formation as well as a greater amount of luminal ghost cells than the non-odontoma-producing type. The odontoma-producing type occurred in younger patients (mean age 13.5 years) and showed even sex distribution, while the non-odontoma-producing type occurred in older patients (mean age 39.5 years) and showed a female predilection. Both types were more prevalent in the Chinese population and the maxilla was the preferable site for both types \(^{(128)}\).

Johnson and associates in 1997 studied 57 cases of calcifying odontogenic cyst. There was no sex predilection and the age range and mean were 7 to 83 and 49.8 years, respectively. The mandible was the preferable site and 34 cases were located there. Thirty-eight cases were central type and 17 cases were peripheral types. Two cases were found both centrally and peripherally. The most common clinical sign for the central type was jaw expansion. While the presenting feature for the peripheral type was a nodular growth on the gingiva. The ghost cells were non-reactive for cytokeratin. No recurrences were found after surgical removal \(^{(129)}\).

According to Toida (1998) there were two concepts concerning calcifying odontogenic cyst. The monistic concept was that all calcifying odontogenic cysts were neoplastic in nature although the majority were
cystic in architecture. The dualistic concept was that the calcifying odontogenic cyst contained two entities: a cyst and a neoplasm. However, Toida favored the later concept\textsuperscript{(130)}.

Li and Yu in 2003 studied 21 cases of intraosseous calcifying odontogenic cyst. According to the biologic behavior they classified these cases into 3 groups: Cyst, benign tumor, and malignant tumor. Sixteen cases were unicystic lesions with or without associated odontoma that showed peak incidence in the second decade of life and the maxilla was the preferable site (69%) especially the canine-premolar region (62.5%). There was no recurrence in this cystic group.

The second group was benign tumor type, which showed solid tumors consisting of ameloblastoma-like sheets of odontogenic epithelium and contained ghost cells as well as calcification foci and juxtaepithelial dentinoid material. The two patients showed this pattern experienced multiple recurrences following conservative surgery. The other two lesions showed typical areas of calcifying odontogenic tumor and other types of odontogenic tumors such as ameloblastoma or odontogenic fibromyxoma. All the lesions of this group were seen in the mandible. The third group was the malignant tumor, which arose from a previously benign calcifying odontogenic cyst. This group showed some features of calcifying
odontogenic cyst and prominent mitotic activity, nuclear and cytoplasmic pleomorphism, areas of tumor necrosis, and infiltrative growth. They suggested the term calcifying odontogenic cyst should be confined to the cystic group while other groups should be classified separately\(^{(131)}\).

### 1.2.8. Odontoma:

The term odontoma was first designated for all tumor of tooth origin\(^{(1)}\). However, later this term was confined to a specific type of odontogenic tumor. The WHO in 1971 classified odontoma into complex and compound odontomes\(^{(7)}\).

Budnick in 1976 reviewed the English literature and collected sixty-five cases of odontomes. He added eighty-four cases from Emory University School of Dentistry. He found that the mean age for odontomes was 14.8 years. Sixty seven percent of all odontomes were located in the maxilla. Sixty one percent of compound odontomes were located in the anterior maxilla, while fifty nine percent of the complex odontomes were located in the posterior region of the jaws. Fifty eight percent of odontomes occurred in males. These lesions were usually asymptomatic. Sixty one percent were associated with impacted teeth of which sixty eight percent of the total impactions were associated with compound odontoma.

The author noted the predilection of complex odontomes,
supernumerary teeth, as well as adenomatoid odontogenic tumor for the anterior maxilla. He suggested that the anterior maxilla was more prone to disturbed odontogenesis. However, he did not give an explanation to this preponderance\textsuperscript{(132)}.

Toretti and his colleagues in 1984 retrieved 167 cases of odontomes from Temple University School of Dentistry. Odontomes represented 0.5% of all biopsies received. Compound odontoma constituted 55.1%, while complex odontomes represented 44.9% of the total. Eighty-one cases (51.2%) were located in the maxilla, while 77 cases (48.8%) occurred in the mandible. There was an equal sex distribution. The peak incidence was in the second decade (50.9%)\textsuperscript{(133)}.

Kaugars and his colleagues in 1989 retrieved about 351 cases of odontomes from the Medical College of Virginia. Odontomes accounted for (0.65%) of all biopsies received. They found that the median age was 16 years and the peak incidence was in the second decade (53.6%). There was an equal sex distribution. There was a predilection for the anterior maxilla (33.9%) with an equal distribution between both jaws. Fifty eight percent of cases were located between the canines. There was progressive predominance in the molar region with the increasing age. Forty seven percent were associated with unerupted tooth. Sixty four percent of unerupted teeth were located in
the posterior mandible. The average age were 16.5 and 22 years for the compound and complex odontoma, respectively.

Histologically, twenty seven percent of the cases were associated with a dentigerous cyst, and 0.9% of the cases were associated with calcifying odontogenic cyst. No recurrence was observed for all these cases. They were unable to differentiate between compound and complex odontoma because of the lack of established criteria for diagnosis. They realized the predilection for anterior maxilla for both odontoma as well as supernumerary teeth. They concluded that odontomes might be a less differentiated form of a supernumerary tooth (134).

Katz in 1989 analyzed three hundred and ninety six odontomes. He found that the compound odontoma was the most common type constituted 70% of all the cases. Both types showed equal gender distribution. Compound odontomes were commonly located in the canine region of either jaw while, complex odontomes were commonly seen in the molar regions and were more frequently associated with unerupted teeth than compound odontomes. He observed a correlation between the site of an odontoma and the age at presentation (135).

In USA, Owens and his associates studied 104 cases of odontomes. They found that the age predilection in the age group 11-20 years. They also showed maxillary predilection. They found that (85%) were correctly diagnosed clinically. Compound odontoma was more common (64.4%) than complex odontoma (31%) (136).

In Malaysia, Ng and Siar studied 104 cases of odontomes. They realized equal distribution in both jaws. The mean age was 24.8 years with
no sex predilection. However, (81.9%) were correctly diagnosed clinically. They found also compound odontoma is more common (43.3%) than complex odontoma (35.5%) \(^{(137)}\).

In Japan, Miki and his associates studied 47 cases of odontomes. They found that there was a male predilection (57%). The mean age was 22 years with greater number of cases were asymptomatic (63.8%). There was a mandibular predilection (57.4%). Compound odontoma was more common (53.2%) than complex odontoma (46.8%). Histologically, ghost cells were noted in (23.4%) of cases, while odontogenic epithelium was found in (34.1%) of cases. Odontoma was associated with impacted tooth in (57.4%) of cases \(^{(138)}\).

1.2.9. **Odontogenic fibroma:**

Gardner, D.G. (1980) attempted to clarify the different terms designated for central odontogenic fibroma. He divided the tumor into 3 types namely, the hyperplastic dental follicle, the simple central odontogenic fibroma, and the WHO type odontogenic fibroma. He assumed certain criteria for distinction between these 3 types. The criteria included clinical, histological and radiographical characteristics of these lesions \(^{(139)}\).

Jones and his associates (1989) presented 2 unusual cases of central odontogenic fibroma. The first case showed cystic lesion at the ramus of the
mandible. Microscopically, the lesion showed sparsely cellular fibrous tissue with abundant mucoid matrix and scattered islands of inactive odontogenic epithelium. Also a cystic area, lined by non-keratinized stratified squamous epithelium with Rushton hyaline bodies was noted.

The second case presented with paraesthesia at the lower lip, and later, a swelling was discovered at the anterior area of the mandible. Radiographically, the case showed ground glass appearance, similar to fibrous dysplasia. Microscopically, it showed moderate fibrous tissue stroma with metaplastic bone and islands and cords of odontogenic epithelium. The lesion was diagnosed as an ossifying variant of an odontogenic fibroma. The lesion recurred 16 months after surgery\(^{(140)}\).

Handlers and his colleagues in 1991 reported 19 cases of central odontogenic fibroma. The maxilla was the preferable site especially anterior to the molars. There was pronounced female predilection. Occasionally, the lesion was associated with unerupted mandibular third molar. They concluded that the histological pattern did not alter the clinical behavior and, therefore, it was unnecessary to separate the tumor into two variants\(^{(141)}\). Odontogenic fibroma was defined by WHO as a fibroblastic neoplasm containing varying amounts of apparently inactive odontogenic epithelium\(^{(9)}\).
Allen and his associates in 1992 reported three cases of central odontogenic fibroma (WHO type) with giant cell granuloma-like features. All lesions occurred in women and all were located in the mandibular premolar-molar region. One case recurred after conservative excision. They considered the lesion as a unique presentation of a central odontogenic fibroma (WHO type) (142). Gardner could not attain a conclusion concerning odontogenic fibroma with giant cell reaction from central giant cell granulomas (143).

On the other hand Hirschberg and his associates in 1996 studied central odontogenic fibroma and hyperplastic dental follicle using picrosirius red and polarizing microscopy. They found that there was no difference between the polarization colours of thin fibers in both lesions. While, central odontogenic fibroma showed more green and greenish-yellow polarization in thick fibers. They concluded that the picrosirius red polarization method could be a useful procedure to differentiate between central odontogenic fibroma and hyperplastic dental follicle (144).

However, Gardner in 1996 revised the current knowledge concerning the central odontogenic fibroma. He considered the separation of this lesion into WHO and simple types remained valid. He suggested the term (complex central odontogenic fibroma) for WHO type. He found
difficulty in distinction between central simple odontogenic fibroma and desmoplastic fibroma \(^{(143)}\).

On the other hand Odell and his associates studied 8 cases of central odontogenic fibroma with features of central giant cell granuloma. These lesions occurred over a wide age range. Radiographically, the lesions showed unilocular or multilocular radiolucencies with cortical expansion and even perforation in one case. Histologically, the lesions showed zones of typical giant cell granuloma in a fibrous stroma containing islands and strands of epithelial cells. The epithelial islands showed duct-like spaces or hyaline basement membrane globules. Trabeculae of osteoid were frequently seen. Two cases recurred after curettage. They concluded that this type of lesion showed an increased risk of recurrence. They could not assign the lesion to a variant of either tumors, but clinically they were close to giant cell granuloma \(^{(145)}\).

Ide and his colleagues in 2002 reported two cases which showed hybrid features of central odontogenic fibroma and ameloblastoma. The lesions were seen in the mandible of a middle-aged female. Radiographically, they were well-demarcated radiolucencies. Clinically, they were close to odontogenic fibroma than ameloblastoma. The lesions were designated as central odontogenic fibroma, epithelium-rich variant \(^{(146)}\).
1.2.10. **Odontogenic myxoma:**

Bloodgood presented the first reported case of myxoma in 1920 (147). White and his associates in 1975 studied nine cases of odontogenic myxoma and found that the average age and range were 26.5 and 11 to 62 years, respectively. There was a female predilection with male to female ratio 1:1.25. The mandible was the preferable site (77.7%).

Ultrastructurally two types of connective tissue cells were found, pale cells and dark cells. The pale cells were considered as secretory cells. The authors postulated that the connective tissue cells were probably active secretory cells, secreting myxoid material. Furthermore, the collagen molecules secretion was probably disturbed (148).

While, Goldblatt in 1976 showed that odontogenic myxoma was composed of two functional types of cells, secretory (type 1) and non-secretory (type 2). Type 1 cells were similar to fibroblast while type 2 cells devoid of Golgi apparatus and scanty rough endoplasmic reticulum. He noticed incomplete collagen fibrillogenesis with its phagocytosis by tumor cells, abundant secretion of acid mucopolysaccharide matrix. He concluded that the tumor cells resemble fibroblast of dental papilla and hence the tumor was of odontogenic origin (149).

Gundlach and Schulz in 1977 reported nine cases of odontogenic
myxoma. Clinically, the age ranged between 8 to 70 years with a mean age equal 34 years. There was a pronounced female predilection with male to female ratio (1:3.5). The duration ranged between 2 weeks to 12 years with a mean 42 months. The frequent presenting features were pain or paraesthesia. All tumors were located in the mandible especially the posterior area and one case was peripheral type. Ultrastructurally, two cell types were seen. The well differentiated fibroblast that appeared spindle-shaped on longitudinal sections and stellate on cross-section. The second type was the so-called myxoblast with rounded shape. Histochemically, the mesenchymal cells showed positive reaction to alkaline phosphatase and negative for acid phosphatase, leucin-amino-peptidase, and Beta-glucuronidase\textsuperscript{150}.

Similarly, Hasleton and his associates in 1978 described an abundant acid mucopolysaccharide stroma among their case. Ultrastructurally, they showed fibroblasts and occasional myofibroblasts. However, odontogenic epithelium was not seen in their case. Therefore, they concluded that the odontogenic epithelium was not a significant component in the histogenesis of the tumor\textsuperscript{151}.

Lamberg and his associates in 1984 reported the first well-documented case of malignant myxoma. They presented a case of maxillary myxoma, which recurred 3 weeks after enucleation. The tumor protruded
from the sinus through a tooth socket forming oro-antral fistula and causing destruction of the wall of the sinus and invading mucosa of the palate.

Microscopically, the lesion showed plum-shaped pleomorphic cells with interconnecting processes merged in a loose myxoid intercellular matrix. There were numerous atypical mitotic figures. The nuclei were large ovoid with granular cytoplasm. There were neither odontogenic epithelial islands nor collagen fibres.

The lesion was treated by irradiation with no obvious result. Finally, the lesion was treated by hemimaxillectomy with reconstruction. They concluded that the lesion should be classified as an odontogenic myxosarcoma (152).

Slootweg and his coworkers in 1985 analyzed the glycosaminoglycans in the extracellular matrix of a jaw myxoma. The results were compared to glycosaminoglycans in the dental pulp and periodontal ligament. It was found that glycosaminoglycans formed about (1%) of the total weight of the tumor and (17%) of the dry weight. The hyaluronic acid formed (72.4%) of the glycosaminoglycan. They concluded that the tumor matrix showed higher glycosaminoglycan content than the normal dental tissues as well as higher hyaluronic acid fraction than the normal dental tissues (153).
Lambardi and associates in 1988 showed that odontogenic myxoma was of mesenchymal origin since it showed positive activity to vimentin and S-100 proteins (154). Similarly, Moshiri and coworkers (1992) showed that odontogenic myxoma was of myofibroblastic origin. They studied 3 cases immunohistochemically (155). However, Takahashi and his associates showed that the tumor was of a dual fibroblastic-histiocytic origin. They studied odontogenic myxoma immunohistochemically (156).

Schmidt-Westhausen and his associates in 1994 showed that the tumor cells of odontogenic myxoma secreted a defective fibrillar collagen's type I and III unlike that of normal developing teeth (157).

Kaffe and his associates in 1997 reviewed the literature and added two new cases of odontogenic myxoma with a total of 162 cases. Seventy five percent of cases were diagnosed in the 2nd to 4th decades of life. There was a female predilection with male to female ratio (1:1.5).

The mandible was the preferable sites in (66.7%) of cases. The predominant radiographic pattern was the multilocular seen in (55%) of cases while (36%) were unilocal and (9%) were not loculated. The large lesions tended to show multilocular pattern. However, five percent of the cases were associated with an unerupted tooth (158).

Jaeger and his colleagues in 2000 were the first who established a
cell line derived from a human odontogenic myxoma. Ultrastructurally the
cells of cell line were similar to the tumor cells. While the matrix of the cell
line was formed of irregular filaments after 60 days. They concluded that
this cell line would be a useful model for investigating the biology of the
odontogenic myxoma\(^{(159)}\).

On the other hand Pahl and his colleagues in 2000 reported a case of
malignant odontogenic myxosarcoma in a 53-year-old patient located in the
maxilla. The tumor recurred two times after maxillectomy leading to death
of the patient by infiltration to the cranial cavity. Microscopically, the lesion
showed features of low-grade malignant myxosarcoma with cellular areas,
prominent mitotic figures, and nuclear pleomorphism\(^{(160)}\). Odontogenic
myxoma incidentally showed unusual histological features, such as that
reported by Oygur and his colleagues in 2001 in which dispersed
osteocement-like spherules were seen\(^{(161)}\).

1.2.11. Benign cementoblastoma:

Norberg was the first to describe this neoplasm in 1930\(^{(162)}\). However, Pynn and his colleagues in 2001 described a case of benign
cementoblastoma in a 23-year-old female patient. The authors reviewed the
literature and found that the lesion was very rare. The presenting feature was
bony swelling with mild pain. They found that 75% of patients were under
the age of 30. The mandible was more frequently affected and usually associated with the permanent first molar or the second premolar. Rarely it could be associated with multiple teeth. Radiographically, the lesion was radiopaque with well-defined radiolucent halo\(^{(163)}\).

Brannon and his associates in 2002 collected and analysed forty four cases of benign cementoblastoma from the Armed Forces Institute of Pathology. The age mean and range were 20 and 8-44 years, respectively. The cases showed male and mandibular predilection. The first permanent mandibular molar was involved in 52.3 % of cases. Microscopically these lesions showed the typical histological features of benign cementoblastoma\(^{(164)}\).

### 1.2.12. Malignant odontogenic tumors:

Elzay in 1982 proposed a classification system of malignant odontogenic tumor, which was a modification of the WHO classification in 1971\(^{(7)}\). The system categorized primary intraosseous carcinoma in to those arising from odontogenic cyst or ameloblastoma and those arising de novo\(^{(165)}\). Slootweg and Muller in 1984 modified the above classification for malignant tumors resemble ameloblastoma depending on the presence of histological features of malignancy\(^{(166)}\).

However, WHO in the recent classification categorized malignant
odontogenic tumors into odontogenic carcinoma and odontogenic sarcomas, in which the latter was further classified into malignant ameloblastoma, primary intraosseous carcinoma, and other malignant epithelial tumors. The clear odontogenic carcinoma was classified as a benign epithelial odontogenic tumor of local invassiveness \(^{(9)}\).

Thomas and his colleagues in 2001 reviewed the literature for primary intraosseous carcinoma and found 35 well-documented cases. They found that the mean age was 52.3 years, with male predilection. The posterior mandible was the favourable site \(^{(167)}\).

Bregni and his colleagues in 2001 reported two cases of ameloblastic fibrosarcoma and review the literature and found 62 well-documented cases. They noted male and mandibular predilection in these cases. The age mean and range were 27.3 and 3 to 83 years, respectively. Thirty six percent of these cases arose in previously benign ameloblastic fibroma. The mean age of ameloblastic fibrosarcoma arising de novo was younger (22.9 years) than those arising in previously benign ameloblastic fibroma (33 years) \(^{(168)}\).

Verneuil and his colleagues in 2002 reported a case of malignant ameloblastoma that metastasized to the regional lymph nodes with the histological characteristics of classical ameloblastoma. The lesion histologically a multiphasic, malignant ameloblastoma with fibro-osseuos
induction \(^{(169)}\).

Ariyoshi and his colleagues in 2002 reported a case of clear cell odontogenic carcinoma that showed mitotic figures and mild cellular and nuclear atypism. They also noted the presence of ghost cells and uncalcified amorphous deposits which resemble dentin. Although the clinical data indicated a benign course the lesion showed recurrence following conservative surgical management \(^{(170)}\).

Taylor in 2003 found that 2.2\% of all odontogenic tumors were malignant neoplasms, mostly were odontogenic carcinoma (6 cases) and a case was ameloblastic fibrosarcoma. They reviewed the literature and found twenty cases of malignant ameloblastoma with age mean and range 44.5 and 15-84 years, respectively. Clear cell odontogenic carcinoma showed female predilection with age mean and range 56.5 and 17-89 years, respectively \(^{(171)}\).

Dhir and his colleagues in 2003 reported a case of maxillary ameloblastic carcinoma and they reviewed the literature and found 18 well-documented cases. They found a wide age range with a mean age of 53.5. The favourable site for metastasis was the lungs followed by cervical lymph nodes and spine. These lesion showed cytological features of malignancy \(^{(172)}\).
Kumar and his associates in 2003 reported a case of clear cell odontogenic carcinoma that metastasized to distant organs particularly spine and hip bones. Both primary and metastatic lesions showed the typical features of clear cell odontogenic carcinoma. They reviewed the literature and no such a case has been reported (173).

Kunkel and his associates in 2004 reported a case of ameloblastic fibrosarcoma that showed multiple local recurrences and within 21 months after surgery pulmonary metastasis was noted. Histopathological and immunohistochemical study confirmed the malignant nature in both cell populations and the lesion was re-diagnosed as odontogenic carcinosarcoma (174).
1.3 Objectives

**General objectives:**

To report on the nature of odontogenic tumors among Sudanese patients.

**Specific objectives:**

To study the clinical features and the histological characteristics of odontogenic tumors seen among Sudanese patients during the period 1994-2002.
Chapter Two

2. Materials and methods

**Materials:** Records of (n=151) patients with histologically confirmed odontogenic tumors during the period 1994–2002 inclusive were retrieved from various institutions. The institutions were Khartoum Teaching Dental Hospital, Omdurman Teaching Hospital, Medical Corp Hospital, and Police Central Hospital. Information including age, sex, site of tumor, symptoms, signs and their duration were obtained.

Paraffin blocks of the same patients were retrieved from the National Health Laboratory and El Zahrawi Laboratory. Sections (5 µm thick) from these blocks were prepared and stained by Hematoxylin and Eosin for re-evaluation.

**Methods:** The clinical data collected were analyzed and assessed, furthermore, the previous diagnosis on each of these sections was revised. The revision of both the clinical and histological data was according to the Histological Typing of Odontogenic Tumors, WHO (9).

**Study design:** Hospital based, retrospective, and descriptive study.

**Data analysis:** By application of the SPSS computer program data
was analysed statistically.

Chapter Three

3. Results

3.1. Clinical Result:

Odontogenic tumors:

Relative frequency: One hundred and fifty six cases of odontogenic tumors were retrieved and analyzed. The relative frequency of different odontogenic tumors was obtained, (Table 1).

Site: Out of ninety-seven cases where the site was specified, the posterior areas in both jaws were more commonly affected (73 cases, 75.2 %) than the anterior area (14 cases, 14.4 %), (Table 2). The mandible by far was more affected (75 cases, 77.3%) than maxilla (22 cases, 22.7 %). However, the posterior mandible was more affected (56 cases, 57.7 %) than the anterior mandible (11 cases, 11.3 %). In the maxilla, the posterior area was more frequently affected (17 cases, 17.5 %) than anterior area (3 cases, 3.1 %), (Table 2).

Sex: Males were more affected than females. Eighty-nine cases (59.7%) were males and sixty cases (40.3 %) were females, (Table 2).

Age: There was a peak incidence in the third decade of life. While,
fifty-four cases (68.6 %) were seen in the second, third, and fourth decades of life, *(Table 2).* Odontogenic tumors were rarely found in children and no case was reported below the age of nine years in this series, *(Table 2).* The age range and mean were 14 to 80 and 32.15 years, respectively (standard deviation=16.93).

*Duration:* The duration range was 10 years with a mean duration 1.81-year and a median 1-year. However 77.9 % of cases were presented during the first two years of the initiation of the lesions.

*Ameloblastoma:*

Relative frequency: Ameloblastoma was found to be the most common odontogenic tumor in this study. Ninety-seven cases were found and constituted 64.7 % of all odontogenic tumors, *(Table 1).*

*Site:* Out of sixty three where the site was specified, the mandible by far was more involved (55 cases, 87.3%) than maxilla (8 cases, 12.7 %). However, the posterior mandible was more affected (45 cases, 71 %) than the anterior mandible (5 cases, 7.9 %). In the maxilla, only the posterior area was affected (8 cases, 12.7 %) and no case was reported in the anterior region, *(Table 2).*

*Sex:* Males were more affected than females, 63%(58 cases) of cases were males, and 37 % (34 cases) were females, *(Table 2).*
Age: There was a peak incidence in the third decade of life accounting for 34.1% (29 cases). Majority of cases 63.5% (54 cases) were distributed in the second, third, and fourth decades of life. Ameloblastoma was not recorded in children below the age of 14 years, *(Table 2)*. The age range and mean were 14 to 80 and 35.62 years, respectively (standard deviation=16.33). *(Table 2)*

Duration: The duration range was 9 years with a mean duration 1.94-year and a median 1-year. Seventy-four cases (73.9% of cases) were seen during the first two years of the onset of the symptoms of the lesions.

*Squamous odontogenic tumor:*

Relative frequency: One case was reported in this series. The lesion constituted about 0.7% of all odontogenic tumors. *(Table 1)*.

Site: The lesion presented as a swelling at the right side of the maxilla involving the right side of the palate with intracranial extension.

Sex: a male patient.

Age: The lesion was seen in a 22-year old patient.

Duration: The duration of the lesion was 9 years.

*Calcifying epithelial odontogenic tumor (Pindborg tumor):*

Relative frequency: There were three cases seen in this study and it constituting 2% of all odontogenic tumors, *(Table 1).*
Site: Two cases were seen in the mandible and one case in the maxilla. In the mandible one case was located in the posterior area and the other case was peripheral lesion located in the anterior region. In the maxilla the case was located in posterior region.

Sex: Two cases were males and one case was a female, with male to female ratio 2:1.

Age: The age range and mean were 20 to 52 and 35.00 years, respectively (standard deviation 16.09). Two cases were seen in the third and fourth decades, while one case was seen in the sixth decade of life.

Duration: The mean and the median duration both were 2 years.

Ameloblastic fibroma:

Relative frequency: Eight cases were recorded in this study and the lesion constituted 5.3% of all odontogenic tumors, ranking the fourth common odontogenic tumor, (Table 1)

Site: In five cases the site was reported in the mandible. Three cases (60 %) were located in the posterior mandible, one case (20 %) in the anterior mandible, and one case (20 %) involved both anterior and posterior mandible, (Table 2).

Sex: There was a marked male predilection, about 7 cases (87.5 %) were seen in males and a case (12.5 %) was seen in female.
Age: The peak incidence was seen in the second decade of life, 5 cases (62.5%). The age range and mean were 11 to 40 and 20.38 years, respectively (standard deviation =12.74). (Table 2).

5. Duration: Five cases (66.7%) were seen during the first two years of the onset of the lesion. The duration mean and range of the six reported cases were 0.50 and 2 years, respectively. (Table 2).

Ameloblastic fibro-odontoma:

Relative frequency: Two cases were reported in this study, and constituted 1.3% of all odontogenic tumors. (Table 1).

Site: The two lesions were located in the molar area of the mandible.

Sex: Both lesions were seen in males.

Age: The mean age was 27 years.

Duration: The duration of the lesion was 3 months.

Adenomatoid odontogenic tumor:

Relative frequency: In this study 7 cases were reported and the lesion constituted 4.7% of all odontogenic tumor, ranking the fifth common odontogenic tumor. (Table 1).

Site: The maxilla was the preferable site where 7 cases (87.5%) located while one case (12.5%) found in the mandible. In the maxilla six cases (75%) were located in the canine region and three cases (37.5%) were
associated with unerupted maxillary canine.

**Sex:** There was a female predilection in four cases (57.1%) and three cases (42.9%) were seen in male.

**Age:** There was a peak incidence in the second decade of life where three cases (37.5%) were reported. However, six cases (75%) were found in the second, third, and fourth decades of life. The age range and mean were 16 to 54 and 30 years, respectively (standard deviation=15.57). *(Table 2)*

**Duration:** Six cases (85.7%) were seen during the first 2 years of the onset of the lesion. The mean duration was 1.14 years and median was 1.00 years. The duration ranged between 3 months to 3 years.

**Calcifying odontogenic cyst:**

*Relative frequency:* There were three cases in this study. The lesion constituted 2% of all odontogenic tumors. *(Table 1).*

*Site:* Two cases were seen in the maxilla and one in the mandible. A lesion was presented as a huge maxillary swelling, at the molar area extending to the border of eye and nose. The involved teeth were loose.

*Sex:* All cases were seen in females.

*Age:* The mean age was 26 years.

**Odontoma:**

*Relative frequency:* Odontoma was divided into complex and
compound types. Complex odontoma was the most common type of odontoma (4 cases, 80 %). While, one case (20 %) was reported as compound odontoma in this series. Both types constituted 3.4 % of all odontogenic tumors.

**Site:** For complex odontoma 3 cases were seen in the posterior mandible. While, in case of the compound odontomas the site was not specified.

**Sex:** For complex odontoma 2 were males, one was a female. While, in compound odontoma the patient was a female.

**Age:** The age range and mean for complex odontoma were 13 to 25 and 18.33 years, respectively (standard deviation=6.11). *(Table 2).* Two cases (66.7 %) were seen in the second decade of life.

**Duration:** Four cases (75 %) were seen during the first 3 years of the initiation of the lesion. The mean duration was 3 years and median was 2 years. The duration ranged between 2 months to 8 years.

**Odontogenic fibroma:**

*Relative frequency:* Eleven cases were reported in this study. The lesion constituted 7.3 % of all odontogenic tumors. It was the third frequent odontogenic tumor after ameloblastoma and myxoma in this study. *(Table 1).*
Site: The mandible was more frequently affected and 8 cases (62.5\%) were located there and 3 cases (37.5\%) were located in the maxilla. In the mandible, the anterior and posterior areas were equally involved. One case involved both anterior and posterior area of the mandible. In maxilla, the two cases (18.18\%) were located in the posterior area, and a case involved both anterior and posterior area of the maxilla, (Table 2).

Sex: There was a marked female predilection, and 7 cases (63.6\%) were females and 4 cases (36.3\%) were males.

Age: The age range and mean were 13 to 80 and 30 years, respectively (standard deviation=22.06), (Table 2). The peak incidence (5 cases, 45.45\%) was in the second decade of life. Nine cases (81.81\%) were seen in the second, third, and fourth decades of life.

Duration: The range was 7 years with duration mean and median were 1.92- and 1.00-year, respectively. Six cases (54.54\%) were seen during the first 2 years of the onset of the lesion.

Odontogenic myxoma:

Relative frequency: There were 12 cases reported in this study. The lesion constituted 8\% of all odontogenic tumors in this study. It was the second frequent odontogenic tumor in this series. (Table 1).

Site: in five cases where the site was specified site, the mandible was
more affected than maxilla (3:2). In each jaw the lesions were evenly distributed.

*Sex:* There was a pronounced male predilection, and ten cases (81.8 %) were males.

*Age:* The peak incidence was in the third decade of life (7 cases, 60 %). Eleven cases (90 %) were seen in the second and third decades of life. The age range and mean were 11 to 45 and 22.20 years, respectively (standard deviation=9.69), *(Table 2).*

*Duration:* The range was 10 years with mean and median duration of 1.78 and 1.00 year, respectively. Ten cases (88.9 %) were presented to the clinics during the first two years of the onset of the neoplasm.

**Benign cementoblastoma:**

*Relative frequency:* Only one case was reported in this series and constituted 0.7 % of all odontogenic tumors, *(Table 1).*

*Gender:* The case was seen in a female patient.

*Age:* She was in the seventh decade of life.

*Duration:* The duration was 4 months.

**Malignant odontogenic tumor:**

*Relative frequency:* Only one case was reported in series as ameloblastic carcinoma and constituted 0.7% of all odontogenic tumors.
*Gender:* The case was seen in a female patient.

*Age:* She was 60 years old.

*Duration:* The duration was 2 years.

*Site:* found in the left posterior mandible.
Table (1): Description of the relative frequency of odontogenic tumors.

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<th>Histopathologic type</th>
<th>No. of cases</th>
<th>Percentage(%)</th>
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</thead>
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<td><strong>Odontogenic epithelium</strong>:</td>
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<td></td>
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<tr>
<td>1- Ameloblastoma</td>
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<td>64.7</td>
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<tr>
<td>2- Squamous odontogenic tumor</td>
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<tr>
<td>3- Calcifying epithelial odontogenic tumor</td>
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</tr>
<tr>
<td>4- Clear cell odontogenic tumor</td>
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<td>0.0</td>
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<tr>
<td><strong>Odontogenic epithelium with odontogenic ectomesenchyme</strong>:</td>
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<td></td>
</tr>
<tr>
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<tr>
<td>2- Ameloblastic fibro-odontoma</td>
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<td>3- Odontoameloblastoma</td>
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<td><strong>Total</strong></td>
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<td><strong>100</strong></td>
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* without odontogenic ectomesenchyme.

** with or without dental hard tissue formation.

*** with or without included odontogenic epithelium.
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<tr>
<th>Description</th>
<th>OT</th>
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<th>CEOT</th>
<th>AF</th>
<th>AFO</th>
<th>AOT</th>
<th>COC</th>
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<td>7</td>
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<td>5</td>
<td>11</td>
<td>12</td>
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*Abbreviations, OT, odontogenic tumors; AB, ameloblastoma; SOT, squamous odontogenic tumor; CEOT, calcifying odontogenic tumor; AF, ameloblastic fibroma; AFO, ameloblastic fibro-odontoma, AOT, adenomatoid odontogenic tumor; COC, calcifying odontogenic tumor; ODO, odontoma; OFIB, odontogenic fibroma; OMYX; odontogenic myxoma; BC, benign cementoblastoma; OCA, odontogenic carcinoma.
3.2. **Histological results:**

**Ameloblastoma:**

Of the forty-four cases that was confirmed histologically and clinically as ameloblastoma the following distribution was found. The solid conventional type was the most common type constituted 79.5% (35 cases) of the cases, followed by unicystic ameloblastoma 20.5% (9 cases), and desmoplastic ameloblastoma (2.3%) (1 case).

The histomorphic distribution for the solid conventional ameloblastoma was as follow. The follicular pattern was the most common type 58.9% (20 cases), followed by the plexiform type 20.6% (7 cases), the mixed pattern 14.7% (5 cases), the granular cell type 5.9% (2 cases), and basal cell variant 2.5% (1 case).

Four cases had been misdiagnosed as ameloblastoma and on re-examination they were consistent with fibrous dysplasia, calcifying odontogenic cyst, odontogenic carcinoma, and basaloid squamous cell carcinoma (Fig. 9).

The mixed pattern displayed 2 cases (40%) of recurrent ameloblastoma. Both lesions were located in posterior area of the mandible. They were seen in adult (40 to 60 years old), one was a male while the other was a female. In the follicular pattern 2 cases with recurrences were reported (10%) while in plexiform pattern no recurrence was stated.
**Follicular ameloblastoma:**

Follicular ameloblastoma was formed of follicles that were lined by columnar ameloblast-like cells with reversed polarity. Flattened stratum intermedium-like cells were sometimes noted close to ameloblast-like cells. In the center of these follicles loosely arranged spindle and stellar cells resembled stellate reticulum cells were frequently seen. These follicles were embedded in a fibrous connective tissue with variable degree of collagenization. These follicles sometimes showed cystic degeneration or squamous metaplasia (Fig.1). Some follicles showed amorphous eosinophilic material in the area of stellate reticulum-like cells, (Fig.2). While some follicles showed small duct-like structures containing eosinophilic collagenous material with some fibroblast-like cells, (Fig.3). One lesion showed presence of a focus of granular cells (Fig.4). Some follicles showed subepithelial hyalinization (Fig.4). Some cases showed a direct continuity of the neoplasm with the oral epithelium (Fig.5), invasion of adjacent soft tissues (Fig.6).

**Plexiform ameloblastoma:**

Plexiform ameloblastoma showed two distinct patterns. The first pattern showed masses and anastomosing strands of ameloblast-like cells embedded in a fibrous CT. The fibrous connective tissue sometimes showed stromal degeneration (Fig.7). The strands were formed of double rows of ameloblast-like cells. The second pattern showed anastomosing
double rows strands. Stellate reticulum-like cells were noted between these strands, in which some showed cystic degeneration (Fig.8). Densely packed rounded cells were sometimes replacing stellate reticulum-like cells. However, duct-like structures containing eosinophilic hyaline materials were noted in the area of stellate reticulum-like cells. (Fig.8). Connective tissue stroma was usually scanty. Some lesions showed a direct continuity of the neoplasm with the oral epithelium, with gradual or abrupt transition in others (Fig.9).

**Mixed pattern:**

Some cases showed both the follicular and the plexiform patterns in the same lesion.

**Granular cell type:**

Two cases were diagnosed as granular cell type of ameloblastoma. One case involved the anterior region of the mandible while; the other case was located in the posterior area of the mandible. Both cases were females. Their age ranged between 25 to 45 years. These lesions showed large cells with eosinophilic granular cytoplasm usually located in the area of stellate reticulum-like cells, and sometimes at the periphery of some follicles (Fig.10).

**Basal cell type:**

This type showed solid masses and strands of basaloid deeply staining cells forming trabecular pattern lying in a delicate fibrous
connective tissue (*Fig. 12*).

**Unicystic ameloblastoma:**

Nine cases were confirmed clinically and histologically as unicystic ameloblastoma.

**Clinically:**

The mean age was 25.7 years. All cases were located in the mandible, 8 cases in posterior region and a case in the anterior region. There was a female predilection and 5 cases were females and four cases were males (1:1.25, male: female ratio). However, two of the cases were recurrent cases (recurrence rate = 22.2%).

**Histologically:**

Unicystic ameloblastoma was seen as a cystic lesion lined by stratified squamous epithelium in one part, while in some areas showed well polarized hyperchromatic columnar basal cells above which loose spindle and stellate reticulum-like cells were noted (1st type WHO 1992) (*Fig. 12*).

The lesion sometimes showed a nodular projection into the cystic cavity usually of plexiform type (2nd type WHO 1992) (plexiform unicystic ameloblastoma) (*Fig. 13*) and (*Fig. 14*).

The lesion also sometimes showed mural proliferation into the connective tissue, which could be plexiform or follicular pattern. The early invasion of CT by anastomosing strands was noted (*Fig. 15*). While,
deeper invasion by anastomosing plexiform strands was also noted in another patient. On the other hand follicular pattern invading CT was also noted (Fig. 16). However, in one case both mural and luminal proliferations was observed (Fig. 14).

The connective tissue adjacent to the epithelium showed hyalinization (Fig. 15).

**Desmoplastic ameloblastoma:**

One case was confirmed histologically as desmoplastic ameloblastoma. The lesion was seen in an 18-year old male patient located in the posterior area of the mandible.

Histologically, the lesion showed slender anastomosing strands and very few small follicles embedded in collagenized fibrous connective tissue (Fig. 17). Eosinophilic hyalinization was noted around some follicles. There was a focal aggregation of multinucleated giant cells (Fig. 18). Cuffing of some blood vessels by eosinophilic collagen was also noted. The connective tissue showed chronic inflammatory cell infiltrate in some areas (Fig. 18). A follicle was noted, which showed duct-like structures containing eosinophilic fibrillar material. A double layer of cuboidal basophilic cells was lining these ducts) (Fig. 19).
Squamous odontogenic tumor showed variable sizes of masses and strands of innocuous odontogenic epithelium embedded in highly collagenized fibrous connective tissue. The connective tissue showed eosinophilic hyalinization in certain areas (Fig.20). Spherules of cementum-like calcifications were seen scattered in the tumor; these calcifications were usually seen within epithelial islands (Fig.21). The lesion was lined by a thin fibrous connective tissue (Fig.20). Flattened squamous cells bounded the epithelial islands and occasionally cuboidal cells were noted. Some islands showed keratin production (Fig.21).

**Calcifying epithelial odontogenic tumor:**

The lesion was formed of solid sheets, islands, and strands of large polyhedral cells with deeply staining nuclei lying in a fibrous connective tissue. The cells sometimes were dense compact or loose and widely separated (Fig.41). Within the epithelial islands two types of calcifications were noted: small lamellated basophilic calcifications and eosinophilic amyloid-like globules. Small follicles of odontogenic epithelium were noted. The peripheral type showed loose widely separated deeply staining nuclei, without prominent intercellular bridges. These cells lying in a fibrous connective tissue with scattered inactive odontogenic epithelium. (Fig. 42).

**Ameloblastic Fibroma:**

This lesion was formed of odontogenic epithelium with delicate
strands, finger-like projections, follicles, or rosette. The epithelial cells were embedded in primitive ectomesenchymal tissue that formed of primitive fibroblast-like cells with oval nuclei and a delicate stroma (Fig. 22). Ameloblast-like cells with a distinctive basement membrane lined these follicles. The central area of these follicles was occupied by stellate reticulum-like cells, which sometimes showed cystic degeneration (Fig. 22).

The lesion sometimes showed lobular configuration in which the tumor was separated into smaller lobules by fibrous connective tissue that showed sometimes hyalinization (Fig. 23). The lesion was surrounded by fibrous connective tissue (Fig. 23).

Of the eight cases diagnosed as ameloblastic fibroma, two cases were excluded because they were found histologically consistent with odontogenic fibroma and and ameloblastic fibro-odontoma (Fig. 24, 25). Two cases had been previously misdiagnosed as ameloblastoma with hyalinization. On re-examination they were found to be ameloblastic fibroma (Fig. 23).

**Ameloblastic fibro-odontoma:**

Two cases were found to be ameloblastic fibro-odontoma, a case had been misdiagnosed as ameloblastic fibroma but on re-examination was found to be ameloblastic fibro-odontoma (Fig. 25). The lesion showed follicles and rosette of odontogenic epithelium embedded in
primitive ectomesenchymal tissue. Dysplastic dentine and cementum-like calcifications were noted (Fig.22).

**Calcifying odontogenic cyst:**

Two cases were added, which had been misdiagnosed as ameloblastoma and adenomatoid odontogenic tumors. The three cases were cystic lesions lined by epithelium of variable thickness. In one area the epithelium was 2 to 3 cell thick stratified squamous epithelium. While in other areas the epithelium was formed of well-palisaded basal layer overlaid by loose stellate reticulum-like cells. Nodules of epithelial proliferation into the cystic cavity were noted within which ghost cells were found (Fig.26). One case showed small number of ghost cells confined only to the epithelium, while the other two cases showed extensive ghost cell formation in epithelium and surrounding connective tissue (Fig.27). Some of these ghost cells showed extensive dystrophic calcification (Fig.26). In some areas proliferation of anastomosing strands into the surrounding tissue was noted (Fig.28). The connective tissue adjacent to the epithelium was delicate and showed subepithelial hyalinization in certain areas. However, cholesterol clefts were noted in the connective tissue. The solid variant showed multiple small follicles lined by ameloblast-like cells within which some ghost cells were noted, representing the neoplastic variant (Fig.27).

**Adenomatoid odontogenic tumor:**
A case that had been misdiagnosed as adenomatoid odontogenic tumor was found to be a calcifying odontogenic tumor (Fig. 28). Of these lesions examined a case was found to be solid while the rest of the lesions were cystic. The cystic lesions were lined by stratified squamous epithelium of variable thickness (Fig. 29). Some lesions showed epithelial proliferation into the connective tissue capsules in a form of strands and nodules. Some nodules were projecting into cystic cavity (Fig. 29). Some lesions showed multiple small cysts lined by stratified squamous epithelium. The nodules were formed of whorls of spindle and polyhedral cells. Some duct-like structures lined by ameloblastic-like cells were noted. These ducts containing fibrillar eosinophilic hyaline material. This material was also noted at the periphery of nodules. In this area the epithelium formed anastomosing strands of cuboidal cells, in which some of these cells become enclaved by the eosinophilic hyaline material. Another eosinophilic hyaline material was noted at juxtaposition of epithelial whorls. Basophilic dystrophic calcification was noted in the connective tissue capsule together with small follicles of odontogenic epithelium (Fig. 30). The solid variant was formed of epithelial nodules that was surrounded by fibrous connective tissue capsule. The nodules were formed of whorls of spindle to polyhedral cells with eosinophilic amyloid-like material at the juxtaposition. The cells inbetween the nodules were cuboidal with deep basophilic nuclei forming anastomosing
strands and solid masses. Some of these cells were enclaved by eosinophilic hyaline material. Columnar ameloblast-like cells lined duct-like structures, which containing fibrillar eosinophilic materials (Fig.31). Some basophilic calcifications were noted within some nodules.

**Odontogenic fibroma:**

Of the 17 cases initially diagnosed as odontogenic fibroma two cases were found to be ameloblastoma and cementifying/ossifying fibroma. Two more sections were excluded because they did not fulfill the WHO criteria of odontogenic fibroma. Moreover two cases had been misdiagnosed as peripheral odontogenic fibroma were found to be peripheral ossifying fibroma (fibrous epulis) and the another case was just chronically inflamed gingiva. Thus eleven cases were confirmed as central odontogenic fibroma. Odontogenic fibroma in this study was divided into two types: WHO type and simple type, according to Gardner’s classification (Gardner, 1980). A case was found to be simple type while the rest were found to be WHO type.

**Odontogenic fibroma (WHO type):**

The WHO type was showing variable histological pattern. We further divided the WHO type into 4 groups. Those, which formed of well-collagenized moderately cellular tissue with scattered small islands of odontogenic epithelium and calcification (Fig.32). Those moderately collagenized, moderately cellular tissue with very thin anastomosing
strands of odontogenic epithelium and showing calcification (Fig.33). Those moderately collagenized, low cellular tissue with very thin anastomosing strand and islands of odontogenic epithelium and absence of calcification (Fig.34), and highly collagenized, highly cellular fibrous tissue with thick anastomosing strands and large solid islands of odontogenic epithelium with absence of calcification (Fig.24).

**Odontogenic fibroma (Simple type):**

The lesion showed plump fibroblasts that more or less were situated parallel to each other and collagen fibres showed storiform and whorls patterns. Scanty odontogenic epithelium was noted. Blood vessels were also noted in the fibrous connective tissue (Fig.35).
**Odontogenic myxoma:**

All sections showed spindle or stellar cells with fibrillar processes in a mucoid material. Varied amount of collagen fibres was noted, from scanty to more collagenous tissue. Scattered islands and strands of inactive odontogenic epithelium that were sometimes surrounded by a zone of hyalinization were noted (Fig.37).

The lesion showed invasion of the bone trabeculae leading to irregular resorption of them. One lesion showed an area with large pleomorphic cells, abundant eosinophilic granular cytoplasm and occasionally mitotic figures. The area also showed large number of endothelium-lined blood vessels (Fig.38).

Some lesions showed invasion of the gingiva tissue and separated from the lamina propria of the oral epithelium by horizontally situated collagen fibres and moderate chronic inflammatory cell infiltration (Fig. 39).

Abnormal calcification was noted in some lesions, some showed basophilic cementum-like calcifications while others showed eosinophilic dysplastic bone or dentine (Fig. 37). One case which had been misdiagnosed as an ossifying/cementifying fibroma when on re-examination was found to be an odontogenic myxoma.

**Odontoma:**
Complex odontoma was formed of disordered dental tissues consisting of dentine, cementum, and pulp tissues. The lesion showed dentine and cementum, forming trabecular pattern, between these trabeculae pulp-like tissues was noted.

Compound odontoma showed dense masses of dental hard tissues forming small teeth-like structures. These structures were formed of enamel, and pulp-like tissue. A fibrous connective tissue capsule surrounded this calcified hard tissue (*Fig. 40*).

**Benign cementoblastoma:**

The lesion showed a sheet of cementum-like tissue forming trabecular pattern with some elongated cementum-like tissue. In between these trabeculae a delicate fibrous connective tissue with scattered cementoblasts was noted. A fibrous tissue capsule was noted surrounding the lesion.

**Malignant odontogenic tumors:**

The lesion was first misdiagnosed as conventional ameloblastoma, however, careful histological study showed features of cytological atypia. The lesion was formed of odontogenic epithelium forming follicles and strands resembling conventional ameloblastoma but hyperchromatic nuclei and pleomorphic cells were clearly observed. Mitotic figures as well as tumor necrosis were also noted. (*Figure 43* and *Figure 44*).
4. Discussion

This study documented for the first time the histopathological patterns of odontogenic tumors and their relative frequency in the Sudan. The Sudan lies in the Central and Eastern African area with its peculiar diversities in the ethnic, geographical, and environmental characteristics. In this study the most frequent odontogenic tumor is ameloblastoma (97 cases, 64.7%) followed by odontogenic myxoma (12 cases, 8%), odontogenic fibroma (11 cases, 7.3%), ameloblastic fibroma (8 cases, 5.3%), and adenomatoid odontogenic tumor (7 cases, 4.7%).

Other less common lesions were shown in (Table 1). Similarly previous study of the epidemiology of oral neoplasm in the Sudan showed that ameloblastoma was the most frequent odontogenic tumor, constituted 88.3% of all odontogenic tumors collected in that study (31). This high rate of ameloblastoma in Idris’s series was for the reason that data was collected from Cancer Registry and RICK and ignored other odontogenic tumors (31). This result was in consistent with other African countries such as Ghana (91%), Nigeria (56-65%), Zimbabwe (79.1%), and Tanzania (73.7%) (15, 17, 18,19,22). Sawyer found an increased incidence
of ameloblastoma among African American over White American 

(16). This racial predilection was attributed to harvesting phenomenon (17), while Shear and Single (1978) suggested environmental factors such as carcinogens that might be present in diet or habits of African (52). Obviously, the Sudan has some characteristics in common with these countries such as racial, environmental, nutritional and geographical influences. Idris and his associates suggested the role of toombak in Sudanese patient as a risk factor for oral cancer, salivary gland neoplasm, and odontogenic tumors (31). Recent study regarded that HPV could be a possible etiologic factor of ameloblastoma (161). However, the role of other viruses and carcinogenic agents in diet were not well established up to now.

This result was also similar to data published from Japan (53%) and China (58.6%) (24, 43). In Turkey, however, the relative incidence of ameloblastoma drops to 36.5% (42). This study in contrast with the rates in series involving American and Canadian populations, in whom ameloblastoma was the second frequent odontogenic tumor accounting for 11% and 13.5%, respectively (28,29). Similarly, in the South American countries ameloblastoma was the second frequent odontogenic tumor after odontoma with a relative frequency varied of 34.6%, 30.7%, and 20.4% in Mexico, Brazil, and Chile, respectively (25-27). The variation in the incidence rates of ameloblastoma has been attributed to racial and
geographical factors (16). However, recently Fregnani suggested that data collected from Medical Hospitals have bias due to the referral pattern, as some odontogenic tumors such odontomas were underestimated while ameloblastomas were overestimated (59).

In this study ameloblastoma was more frequently located in the mandible 87.3% (55 cases) than maxilla 12.7% (8 cases). This result was consistent with Small’s report where in 81.3% of 1,000 cases, and Reichart’s report where in 83% of 3,677 cases the mandible was involved (35,39). Moreover, the site distribution of ameloblastoma in this study was comparable to corresponding data from Nigerian, Malaysian, Korean, American, and Brazilian patients where the mandible was involved in 91%, 93%, 78.3%, 88%, and 93.1%, respectively (17,40,41,44,48).

In this study ameloblastoma was more frequently located in the posterior part of the mandible and constituted 71% (45 cases) of cases, this result was in agreement with other reports from American, Nigerian, and Asian series (40,43,50). In contrast, Sawyer found no segmental predilection in White American (16). This may suggest another ethnic differences.

In this study ameloblastoma showed a male predilection 63% (58 cases), this finding is in conformity with African, American, and Asian series (28,42,48,50). In contrast, Martins in 29 cases of ameloblastoma found no gender predilection (41).
In this study the mean age was 35.62 years, this finding was comparable to other African, Afro-American, Malaysian, and Korean series \(^{(16,17,44,48,50)}\). In contrast, ameloblastoma affected older age group in Swedish and White American patients \(^{(36,40)}\). This also could be additional racial variation.

In the present study ameloblastoma showed peak incidence of 68.2% (54 cases) in the third, fourth, and fifth decades of life. This was similar to Nigerian and American series \(^{(15,40)}\). In contrast, the Malaysian series showed peak incidence (72%) in the second, third, and fourth decades of life \(^{(44)}\). In this study more than 30% of cases were presented after two years of the onset of the lesion. The delay of diagnosis of these lesions in the Sudanese patients could be attributed to the difficulty of transport and the lack of specialized oral surgeons in the distant States.

This study showed that the conventional ameloblastoma was the most common clinical type constituted 79.5% (35 cases) of cases, followed by the unicystic type 20.5% (9 cases), and the desmoplastic type 2.3% (one case). This result was in agreement with Reichart's series where in 3,677 cases of ameloblastoma 74.9% were conventional ameloblastoma, 15.7% were unicystic ameloblastoma, and 9.4% were peripheral ameloblastoma \(^{(39)}\). On the contrary, no peripheral type was found in this study, this probably because of the rarity of these lesions. The clinical resemblance to other gingival growths impart another
difficulty in identifying these lesions since majority of these lesions were excised without histological examination.

In this study the most common histological pattern was the follicular (20 cases) 58.9%, followed by plexiform pattern (7 cases) 20.6%, mixed pattern (5 cases) 14.7%, granular cell type (2 cases) 5.9%, and basal cell type (one case) 2.5%. Correspondingly, Reichart reviewing the world literature found that the follicular pattern was the most common pattern (33.9%) (39). The higher rates of the follicular pattern, was also noted in Nigerian patients (68.4% and 39.1%) in Mosadomi’s and Sawyer’s series, respectively (15,16). The authors also showed that the follicular pattern was by far the most common pattern in White American (50%) (16). In contrast, Asian and European series showed a remarkable increase in the incidence of plexiform pattern (34.2% and 45.16%, respectively) (36,44,48).

The mixed follicular and plexiform pattern was noted in 14.7% (5 cases) of cases, which was similar to that of Reichart's series (15.7%) (39). However, Sawyer and his coworkers noticed a slight increase in frequency of mixed patterns among Black American over White American (10%) and Black African (10.9%) (16). While, Asian series showed that the mixed pattern (17.7%) was more commonly noticed than the follicular pattern (16.5%) (44).

In this study the follicular pattern showed frequently cystic
degeneration and squamous metaplasia, however, no extensive keratinization was noted that warrant be designated as an acanthomatous type. Reichart reported that the acanthomatous type ranked the third in his series 11.3% (39). Anneroth and Hansen (1982) proposed a histological criterion for differentiation of acanthomatous pattern from other similar lesions (56).

In this study seven cases (20%) showed different types of hyaline deposits. In the follicular pattern two cases (5.7%) showed amorphous eosinophilic hyaline material in the area of degenerated stellate reticulum-like cells. While, two cases (5.7%) showed duct-like structures containing eosinophilic hyaline material. These duct like structures were noted in the area of stellate reticulum-like cells. Another type of hyaline deposit was also noted in the follicular pattern in the subepithelial area around the follicles. Similarly, Sapp and Jensvold (1983) showed that 32.2% of ameloblastoma cases studied were containing hyaline deposits. These deposits were seen in both the follicular cystic and the plexiform types. In the plexiform type they appeared as subepithelial deposits while in the cystic follicular type they appeared as thickening of basement membrane with calcification (57).

In this study a case of plexiform pattern showed hyaline deposits within the duct-like structures that were found in the stellate reticulum-like area. This pattern was similar to that described by Yamane (1984),
which was a rare variant known as pseudoglandular and accounted for 19% of the cases he studied. This variant formed of ductular and tubular epithelial proliferation\(^{(38)}\).

In this study three cases (8.6%) of ameloblastoma showed direct continuity with oral epithelium, either with gradual transition from oral epithelium to neoplastic epithelium or abrupt transition. In China, Wu (1985) showed that tumors with direct continuity with the oral epithelium displayed better prognosis\(^{(23)}\). However, this conclusion was unattainable in this report since it was a retrospective one.

This study showed a case that perforated the bone and invaded adjacent soft tissue in close proximity to salivary gland tissue, the lesion showed follicular pattern with cystic degeneration. In contrast, Larsson and Almeren noticed that the tumor grew in a trabecular and cellular manner with squamous metaplasia at the periphery rather than follicular or cystic pattern\(^{(36)}\).

In this study granular cell type constituted 5.9% (2 cases) of cases, however, Reichart showed that the granular cell type constituted 3.5% worldwide\(^{(39)}\). On the other hand Wladrone and El Mofty (1987) reported that the granular cell type was extremely a rare variant in USA (1%). While, Sawyer realized the absence of reported cases in both African and White American patients that were studied. The granular cell type, however, constituted about 10.9% in Nigerian patients\(^{(16)}\).
One case of basal cell type was reported (2.5%) in this study. Worldwide, the basal cell type was also extremely rare (1.4%) (39). In the USA, the basal cell type constituted 3% of reported cases (40), in Nigeria the basal cell type was also rare (6.5%), and no basal cell type was reported in African and White American in Sawyer’s series (16). Wu realized that the basal cell type showed more aggressive behavior (23).

In this study the average age for unicystic ameloblastoma was 25.7 years, similar to Robinson’s series, American, South African, and Chinese patients in which the average age was found to be 27.7 years, 22 years, 23.8 years, and 25.3 years, respectively (40, 65, 68, 71). This study showed slight female predilection with a male to female ratio of 1:1.25. Similarly, Philipsen and Reichart (1998) noticed female predilection in non-dentigerous variant (1: 1.8) and male predilection in the dentigerous variant (1.5:1) (70). In contrast, other world reports from American, South African, and Chinese patients showed male predilection (40, 68, 71). In this study all cases were located in the mandible, and 8 cases (88.9%) were located in the posterior region of it. Similarly, Waldron among the 12 cases he studied, all the cases were located in the mandible (40). However, in China and South Africa the mandibular lesion constituted 90.9% and 91.2%, respectively (68, 71).

In this study unicystic ameloblastoma was divided according to WHO classification into 3 groups (9). However, the third type was further
subdivided into early, intermediate and late variants for convenience. The histological classification of unicystic ameloblastoma was controversial. Gardner divided unicystic ameloblastoma into 4 types (66). However, Ackerman divided unicystic ameloblastoma into 3 groups (68). The clinical significance of this subdivision needs further studies.

In this study one lesion was diagnosed as desmoplastic ameloblastoma. This was in agreement with Waldron and Philipsen series (40,60). However, the mean age was lower than that in both series. The lesion was located in the posterior area of the mandible. In contrast, Waldron and Philipsen reported that the anterior maxillary area was the preferable site. However, a conclusion could not be drawn from this small sample.

Histologically, this case was consistent with previous reports (40, 60). However, it showed focal aggregation of multinucleated giant cells, cuffing of blood vessels, and chronic inflammatory cell infiltrate in certain areas. These features were not reported before, to the best of our knowledge, in desmoplastic ameloblastoma. The need for categorization of desmoplastic ameloblastoma as a separate variant of ameloblastoma was stressed by Waldron (40).

The only squamous odontogenic tumor in this study occupied the posterior maxilla and showed extensive invasion of the maxillary and palatal tissues and the deeper intracranial area. This feature was contrary
to the benign biological behavior of this tumor.

In contrast, both Goldblatt in his 16 cases and Baden in his 32 cases showed that the mandible was more involved especially the premolar-molar \((77,79)\). In this study the age was (22 years) consistent with Pullon’s result (mean age 25.8 years), slightly less than Goldblatt’s result (mean age 35.8 years), and markedly less than Baden’s result (mean age 40 years) \((75,77,79)\).

In this case no cystic degeneration was observed, although intraepithelial calcification was noted. Likewise, Pullon and McNeil noted intraepithelial calcification and cystic degeneration in the epithelial islands \((75,76)\). The tumor islands in this study were bounded by flattened to cuboidal cells. This was consistent with that reported by Goldblatt’s study \((77)\). In the present study prekeratin was noted within some tumor islands, and similarly Goldblatt showed crystalloid eosinophilic structures and suggested that these structures represent prekeratin or glycoprotein material \((77)\). In this study the connective tissue was highly collagenized with areas showed hyalinization. Similarly Baden noticed stromal hyalinization and hyaline collars around the epithelial islands \((79)\).

In the present study the relative frequency of the calcifying epithelial odontogenic tumor was 2%. However, the incidence is lower in China accounting for 0.9\% \((43)\). The site and age distribution in this study were comparable with Chinese patients \((43)\). The histopathology was
similar to other previous reports \(^{(9,82,85)}\).

In this study the mean age for odontoma, ameloblastic fibroma, and ameloblastic fibro-odontoma were 18.3, 20.3, and 27 years, respectively. Similarly, the average age of ameloblastic fibroma, ameloblastic fibro-odontoma, and odontoma were 15.5, 10.0, and 14.8 years, respectively \(^{(92,132)}\). This suggests that ameloblastic fibroma might not progress into odontoma but could progress into ameloblastic fibro-odontoma. Eversole, Slootweg, Gardner, Philipsen, and AL-Sebaei previously discussed this concept \(^{(91,93,95,99,103)}\). In contrast, recently Lopez considered ameloblastic fibro-odontoma as a variant of ameloblastic fibroma \(^{(101)}\). In this study both ameloblastic fibroma and ameloblastic fibro-odontoma showed male predilection, similarly Slootweg reported a male predilection \(^{(93)}\). In this study odontoma showed equal gender distribution. Similarly Toretti, Kaugars and Katz noticed equal sex distribution \(^{(133-135)}\).

In this study ameloblastic fibroma and ameloblastic fibro-odontoma showed a marked predilection for the posterior mandible. This was inconsistent with Trodahl and Slootweg reults \(^{(92, 93)}\). In contrast, Budnick and Toretti reported maxillary predilection for odontoma \(^{(132, 133)}\).

However, Kaugars described equal site distribution for both jaws \(^{(134)}\). The discrepancy in this study was due to the small size of the sample. The mean age of calcifying odontogenic cyst in this study was
26 years, which comparable to that found by Nagao and Buchner\(^{(123, 126)}\). In contrast the mean age was smaller than that found by Freedman, McGowan, and Johnson\(^{(120, 122, 129)}\).

This study showed a marked female predilection since all cases were seen in females. Freedman also reported a high female incidence (117), but McGowan, Johnson, and Nagao reported equal sex distribution (122, 123, 129).

In this study calcifying odontogenic cyst was frequently noted in the maxilla, similar to what was reported by Nagao and Lu\(^{(43, 123)}\). In contrast Freeman, McGowan, and Buchner reported no significant difference with respect to jaw distribution, while Johnson reported a mandibular predilection\(^{(120, 122, 126, 129)}\).

In this study all cases were the cystic form and only one case was solid. Furthermore, all showed the presence of ghost cells as described by other authors\(^{(9, 120, 122, 126, 129)}\).

In this study the mean age of adenomatoid odontogenic tumor was 30 years which was greater than that found by Courtney, Philipsen, and Awange who reported mean age of 16.5 years, 24 years and 16.2 years, respectively\(^{(108, 111, 112)}\). This higher mean age in this study was probably due to the small size of the sample and that most cases of adenomatoid odontogenic tumor were asymptomatic and accidentally discovered by routine X-ray. Furthermore, most cases have presented late to the clinics.
This study showed a female predilection (57.1%), similar to that reported by Giansanti, Courtney, Philipsen, and Awange \(^{107, 108, 111, 112}\). In contrast, Ajagbe and Arotiba reported male preponderance in Nigerian patients \(^{110, 114}\). This feature could be peculiar to Nigerian patients.

In this study the maxilla was the preferable site 87.5% \((7\text{ cases})\) and 75% \((6\text{ cases})\) were located in the canine area, while, 37.5% \((3\text{ cases})\) were associated with unerupted maxillary canine. This finding was consistent with that reported by Giansanti, Courtney, and Awange \(^{107, 108, 111}\). In contrast, Nigerian patients showed mandibular predilection \(^{110}\). This also could be a peculiar feature to Nigerian patients.

The histological feature of adenomatoid odontogenic tumor in this study was typical to that previously described, however, special stains were not used to detect the nature of eosinophilic hyaline materials. These materials were reported in this study in 3 different locations: within the duct-like structures, at the periphery of epithelial nodules, and at juxtaposition location of epithelial whorls. Although the nature of these eosinophilic materials was controversial, Courtney considered it as an excessive basement membrane formation, while Philipsen considered these materials as a form of enamel matrix \(^{117}\). Further investigation is needed to understand the nature of these materials.

In this study myxoma was the second commonest odontogenic tumor, which similar to that reported from African and Chinese studies
In contrast, this incidence was higher than that reported from North American countries \cite{28,29}.

The mean age in this study was 22.2 years, which was similar to that reported by African and Chinese studies \cite{14,43}. The peak incidence in this study was identical to that reported by Kaffe \cite{158}. This study showed marked male predilection (81.8%). In contrast, Gundlach, White and Kaffe found pronounced female predilection \cite{148,150,158}, while, Lu reported equal gender distribution \cite{43}. In this study the mandible was the preferable site (60%), similar to that reported by White, Gundlach, and Kaffe \cite{146,154,156}. However, Lu reported equal jaw distribution \cite{43}.

In the present study odontogenic myxoma showed spindle shaped cells lying in mucoid substances with abnormal calcification. This calcification was similar to that reported by Oygur \cite{158}. One case showed characteristics of malignant myxoma having large pleomorphic cells containing granular eosinophilic cytoplasm with mitotic figures, and large number of endothelium lined blood vessels. This case was identical to that reported by Lamberg \cite{152}.

In this study odontogenic fibroma was the third common odontogenic tumor accounting for 7.47% (11 cases) of the series. This incidence was higher than that reported from North American and Asian studies \cite{28,43}. In this study the mandible was the preferable site 66.7% (6 cases). This was in consistent with that reported by Kaffe \cite{158}. However,
no conclusion could be drawn from this in light of the paucity of cases.

This study showed gradual increase of odontogenic epithelial rests from small islands, thin strands, to thick strands and solid islands resembled squamous odontogenic tumor. However, variable amount of cellular fibrous tissue was seen. This wide spectrum of histological feature was found in central odontogenic fibroma WHO type. However, the clinical and biological behavior of these different presentations can not be judged in this study because of the small size of the sample and the difficulty of long-term follow-up.

In this study benign cementoblastoma was a rare tumor accounting for 0.7% of all cases. This was consistent with previous studies\(^{(163,164)}\). In contrast, the age of the patient was older than previous reports. This almost certainly because of the fact that majority of lesions were usually asymptomatic.

In this study only one case was reported as odontogenic carcinoma constituting 0.7% of all odontogenic tumors. This incidence was lower than that reported by Taylor who found that malignant odontogenic tumors accounting for 2.2% of all odontogenic tumors\(^{(171)}\). In contrast, Lu reported higher incidence of malignant odontogenic tumors among Chinese patients accounting for 6.1% of all odontogenic tumors. The histological features of this case was identical to that described by Slootweg, Kramer, and Dhir\(^{(9,166,172)}\).
4.2 Conclusion

This study documented the relative frequency and the clinico-pathological patterns of (n=151) of odontogenic tumors in Sudanese patients during the period 1994–2002 inclusive. The material was retrieved from the following settings Khartoum Teaching Dental Hospital, Omdurman Teaching Hospital, Medical Corp Hospital, and Police Central Hospital, the National Health Laboratory, and El Zahrawi Laboratory. The data was assessed and reclassified according to the Histological Typing of Odontogenic Tumors WHO (9).

Statistical analysis showed that ameloblastoma was by far the most frequent lesion followed by odontogenic myxoma, odontogenic fibroma, ameloblastic fibroma, and adenomatoid odontogenic tumor. The odontogenic myxoma and adenomatoid odontogenic tumor showed unique clinico-pathological patterns that probably peculiar to Sudanese patients. A further study in the tumorigenesis of these lesions is important.
4.3 Recommendations

The clinicians and pathologists should be aware of these lesions since they showed different histopathological and clinical characteristics. A careful histological examination of these lesions is important as well as deliberate clinical examination. We recommend further study at the molecular level to understand the tumorigenesis of these lesions.
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Fig. 1: Follicular ameloblastoma: Showing cystic degeneration and squamous metaplasia.

Fig. 2: Follicular ameloblastoma: Showing cystic degeneration and eosinophilic amorphous material in the area of stellate reticulum-like cells.

Fig. 3: Follicular ameloblastoma: Showing a large follicular containing duct-like structures filled with eosinophilic fibrillar material and granular cell changes.
Fig. 4: Follicular ameloblastoma with some follicles: Showing granular cell changes and sub-epithelial hyalinization around some follicles.

Fig. 5: Follicular ameloblastoma: Showing subepithelial hyalinization.

Fig. 6: Follicular ameloblastoma: Invading soft tissue in close proximity to salivary gland tissue.

Fig. 7: Plexiform ameloblastoma: Showing masses and strands of ameloblast-like cells, stromal degeneration was clearly noted.
Fig. 8: Plexiform ameloblastoma: Showing strands of ameloblast-like cells delineating stellate reticulum-like cells.

Fig. 9: Basaloid squamous cell carcinoma: Showing large masses of malignant squamous cells, bounded by basaloid cells. The lesion has direct communication with oral mucosa.

Fig. 10: Granular cell ameloblastoma: Showing granular cells occupying area of stellate reticulum-like cells, some granular cells were noted even at the periphery of follicles.
Fig. 11: Basal cell type of ameloblastoma: Islands are formed of basaloid cells which showed trabecular pattern of growth.

Fig. 12: Unicystic ameloblastoma: Showing a cystic lesion lined with stratified squamous epithelium with well polarized hyperchromatic columnar basal cells above which loose angular and stellate reticulum cells were noted.

Fig. 13: Unicystic ameloblastoma-plexiform variant: Showing a nodule formed of plexiform epithelial strands.

Fig. 14: Unicystic ameloblastoma: Showing a nodule of odontogenic epithelium projecting into the cystic cavity.
Fig. 16: **Unicystic ameloblastoma:** Showing invasion of connective tissue capsule by follicular odontogenic epithelium as well as small nodule projecting into the cystic cavity.

Fig. 17: **Desmoplastic ameloblastoma:** Showing dense collagenous connective tissue within which slender anastomosing strands and follicles were noted.

Fig. 18: **Desmoplastic ameloblastoma:** Showing a compressed follicle with cystic degeneration, the connective tissue showing dense infiltration of lymphocytes and few plasma cells.
Fig. 19: **Desmoplastic ameloblastoma**: Showing a follicle formed three duct-like structures containing eosinophilic fibrillar material lined by double rows of small basophilic epithelium. Also cuffing of blood vessels was noted.

Fig. 20: **Squamous odontogenic tumor**: Showing islands of odontogenic epithelium in highly collagenized fibrous connective tissue.

Fig. 21: **Squamous odontogenic tumor**: Showing islands of odontogenic epithelium lying in collagenized connective tissue showing hyalinization. Basophilic lamellated calcifications were noted within epithelium islands.
Fig. 22: **Ameloblastic fibroma**: Showing strands, finger-like projections of odontogenic epithelium embedded in primitive ectomesenchymal tissue. Some follicles showed cystic degeneration.

Fig. 23: **Ameloblastic fibroma**: Showing follicles and rosette of odontogenic epithelium lying in a delicate ectomesenchymal tissue with areas of hyalinization. The tumor was separated by fibrous tissue forming lobule, and surrounded by a fibrous connective tissue.

Fig. 24: **Odontogenic fibroma-WHO type**: Showing highly collagenized, highly cellular fibrous tissue with thick anastomosing strands and large solid islands of odontogenic epithelium.

Fig. 25: **Ameloblastic fibro-odontoma**: Showing islands of odontogenic epithelium lying in primitive ectomesenchymal tissue. Dysplastic dentine and cementum-like calcifications were noted.
Fig. 26: Calcifying odontogenic cyst: Showing calcification of ghost cells.

Fig. 27: Calcifying odontogenic cyst- a neoplastic variant: Showing follicles containing ghost cells.

Fig. 28: Calcifying odontogenic cyst lined by thin epithelium: Showed proliferation of thin anastomosing strands into adjacent connective tissue.
Fig. 29: Adenomatoid odontogenic tumor - a cystic variant: Showing intraluminal nodules and mural nodule within connective tissue.

Fig. 30: Adenomatoid odontogenic tumor: Showing dystrophic basophilic calcifications and small islands of odontogenic epithelium in the connective tissue of the tumor.

Fig. 31: Adenomatoid odontogenic tumor-solid variant: Showing nodules of odontogenic epithelium separated by eosinophilic hyaline material.

Fig. 32: Odontogenic fibroma-WHO type: Showing well collagenized moderately cellular tissue with scattered small islands of odontogenic epithelium and calcification.
Fig. 33: Odontogenic fibroma-WHO type: Showing very thin anastomosing strand of odontogenic epithelium.

Fig. 34: Odontogenic fibroma-WHO type: Showing very thin anastomosing strands and islands of odontogenic epithelium.

Fig. 35: Odontogenic fibroma-Simple type: Showing plump fibroblasts and storiform collagen fibers with scanty odontogenic epithelium.

Fig. 36: Ameloplastoma-granular cell type: showing predominant granular cell changes.
Fig. 37: Odontogenic myxoma: Showing stellar or spindle cells with thin delicate fibrillar processes lying in mucoid substance. Basophilic calcification and scattered odontogenic epithelium were noted.

Fig. 38: Odontogenic myxoma: Showing large pleomorphic cells with abundant granular eosinophilic cytoplasm. Large number of blood vessels were also noted.

Fig. 39: Odontogenic myxoma: Showing invasion of adjacent soft tissue.
Fig. 40: **Compound odontoma:** Showing small teeth-like structures and disorganized dental hard tissues.

Fig. 41: **Calcifying epithelial odontogenic tumor:** Showing an islands of large polyhedral cells with deeply staining nuclei-Amyloid-like material and small basophilic calcifications were noted.

Fig. 42: **Calcifying epithelial odontogenic tumor:** Showing solid sheets and islands of large polyhedral deeply staining cells. Amyloid-like material was noted.
Fig. 43: **Ameloblastic carcinoma:** Showing follicles of odontogenic epithelial cells embedded in fibrous connective tissue.

Fig. 44: **Ameloblastic carcinoma:** The follicle showing features of cytological atypia with areas of necrosis.