East African Medical Journal Vol. 79 No. 1 January 2002
ODONTOGENIC TUMOURS AND TUMOUR-LIKE LESIONS IN TANZANIA
E.N.M. Simon, DDS, Lecturer, Department of Oral Surgery and Oral Pathology, Muhimbili University College of Health Sciences, P.J.W. Stoelinga. DDS, MD, PhD, Professor and Chairman, Department of Oral and Maxillofacial Surgery, University of Nijmegen, The Netherlands, E. Vuhahula, DDS, PhD, Senior Lecturer, Department of Pathology and Morbid Anatomy, Muhimbili University College of Health Sciences and D. Ngassapa, DDS, MSc, DSc, Professor and Head, Department of Anatomy and Histology, Muhimbili University College of Health Sciences, Dar es Salaam, Tanzania.

Request for reprints to: Dr. E.N. M. Simon, Faculty of Dentistry, Muhimbili University College of Health Sciences, P. O. Box 65014, Dar-es-Salaam, Tanzania.

ODONTOGENIC TUMOURS AND TUMOUR-LIKE LESIONS IN TANZANIA

E.N.M. SIMON, P.J.W. STOELINGA, E. VUHAHULA and D. NGASSAPA

ABSTRACT

Objectives: **To retrospectively document the pattern of occurrence of odontogenic tumours in Tanzania over fifteen years.**

Design: **The histologic types, site, age and sex distribution of odontogenic tumours in Tanzania from 1982 to 1997 were reviewed. Records of patients who presented to the four referral centres in Tanzania and who had histologically proven oral tumours and tumour-like conditions were examined.**

Results: **Odontogenic tumours comprised about 12.2% of all oral tumours and tumour-like conditions. The majority of odontogenic tumours (55.3%) were seen in patients below 30 years of age and they more commonly affected the mandible than maxilla. Ameloblastoma was the most commonly seen odontogenic tumour (73.7%), followed by odontogenic myxoma (10.3%). The site, sex, and histologic distribution of ameloblastoma did not differ from other African studies. Over 50% of patients with ameloblastoma presented to hospital late (after three or more years).**

Conclusion: **In order to improve on the treatment outcome, the need for early detection and referral of patients by medical personnel and dentists is stressed.**

INTRODUCTION

Odontogenic tumours form a group of lesions essentially occurring in the mandible and maxilla, but on very rare occasions may be found in other sites(1-6). They are classified according to the World Health Organisation's Histological Typing of Odontogenic Tumours(7). Their natural history varies widely(1) and generally there are no known aetiological factors. Odontogenic tumours constitute about 0.002% of all neoplasms(8); between 1.11 and 1.3% of all oral biopsy specimens(1,9) and between 2.5 and 15% of all oral neoplasms(10,11). There are only a few reports available on the relative frequencies of odontogenic tumours, the incidence of the various histologic types and the geographic and racial distribution(6,9,10,12-17). The majority of them are true benign odontogenic tumours, however, occasionally malignant variants are encountered.

Ameloblastomas, although benign in nature tend to invade tissues locally and this makes them prone to recurrence. A neoplasm in which both the patterns of an ameloblastoma and cytological features of malignancy are shown by the primary growth in the jaws and/or by any metastatic growth is called a malignant ameloblastoma(18). While the ameloblastoma has been found to be the most common odontogenic tumour in Africa and Asia, this is different in Germany, The United States, Canada and Mexico where the most common odontogenic tumour was odontoma(1,6, 9,11,13,14,17). Two studies suggest a possible higher incidence rate of ameloblastomas in black Africans(19,20). In Tanzania Slavin and Cameron(15), found the relative frequency of ameloblastoma to be 0.7%. This figure may, however, be misleading because in their study they calculated this on the basis of the frequency of malignant tumours.

Shear and Singh(20), based on an age-standardised study among South African Blacks and Whites concluded that the incidence rate of ameloblastomas was much higher among Blacks than Whites, that is 1.96, 1.20, 0.18 and 0.44 for Black males and females; and White males and females respectively. Larsson and Almeren(21) in Sweden, estimated the incidence rate of ameloblastoma to be 0.6 per million and Muller(22) in The Netherlands reported a similar figure.

Only a few East African studies on the relative frequency and incidence rate are available(15,16,23). Unfortunately, the epidemiological terms used were often confusing making it very difficult to extract reliable figures for comparison. In order to be able to plan on the number of specialist centres that are able to treat these rather devastating tumours, it seemed prudent to make an inventory in Tanzania about the potential needs. It is, therefore, the aim of the present study to determine the relative frequency and incidence rates of different odontogenic tumours in the Tanzanian population over a fifteen-year period and to compare this data with those from studies in other parts of the world.

MATERIALS AND METHODS

The medical records of patients with oral tumours attending the four referral centres in Tanzania, Bugando Medical Centre (BMC), Kilimanjaro Christian Medical Centre (KCMC), the Muhimbili Medical Centre (MMC) and Mbeya Referral Hospital (MRH) over a fifteen-year period (1982 to 1997) were reviewed. Most patients with oro-facial tumours were usually referred to these centres where biopsies were taken and delivered to MMC or KCMC histopathology laboratories for histological examination. The records from these centres, therefore, are closely representative of the whole of the Tanzanian population. The reports of the histologic examinations of biopsies taken from the patients and the files of the patients formed the only source of information.

RESULTS

Twenty eight patients' files were not available for re-examination which implies that the location and size of the tumour was not known but information on sex, age and histology were known through the histology reports. In 37 of the cases the specific age was not known because it was only indicated as either adult or child.

The different tumours and tumour-like lesions encountered in this study are shown in Table 1. Except for the odontogenic keratocyst, all the tumours and tumour-like lesions of which the total number in the whole study was less than ten (for example, malignant melanoma, verucous carcinoma, chodromyxoid fibroma and haemangio- endothelioma) were grouped together as "others". Those tumours for which a definite histologic diagnosis could not be made were categorised as non-specified.

Table 1

Distribution of different oro-facial tumours or tumour -like lesions seen in Tanzania from 1982 – 1997

Tumour	No.
Ameloblastoma	157
Squamous cell carcinoma	152
Kaposi's sarcoma	148
Burkitt's lymphoma	137
Epulis	56
Pleomorphic adenoma	56
Fibrous dysplasia	46
Haemangiona	43
Malignant lymphoma	30
Fibroma	29
Adenocystic carcinoma	29
Ossifying fibroma	28
Radicular cyst	22
Odontogenic myxoma	22
Papiloma	21
Other odontogenic cysts	20
Other cysts	16
Mucocoele	16
Dentigerous cyst	14
Dermoid cysts	11
Odontogenic keratocyst	7
Others	421
Non specified	271
Total	1751

Table 2

Distribution of odontogenic tumours by sex

Tumour	Male	Female	Total	%
Ameloblastoma	76	81	157	73.7
Odontogenic myxoma	9	13	22	10.3
Cementoma	8	3	11	5.2
Odontoma:				
Compound	2	4	6	2.8
Complex	3	2	5	2.3
Calcifying epithelial Odontogenic tumour	1	2	3	1.4
Squamous odontogenic tumour	2	0	2	0.9
Adenomatoid odontogenic tumour	0	2	2	0.9
Ameloblastic fibroma	2	0	2	0.9
Cementifying fibroma	1	1	2	0.9
Calcifying odontogenic cyst	0	1	1	0.5
Total	104	109	213	100

Odontogenic tumours comprised only 12.1% of the total number of oro-facial tumours. Ameloblastoma appeared to have been the most common odontogenic tumour followed by odontogenic myxoma and odontomas. Table 2 shows the distribution of the different types of odontogenic tumours by sex. Males and females were almost equally affected.

Table 3 shows the distribution of the odontogenic tumours by site. Most tumours occurred in the mandible (90.2%) as compared to the maxilla (9.7%). The location of ameloblastomas in the mandible was most often in the posterior region, that is, premolar molar-ramus region (62.4%).

Table 3

Distribution of odontogenic tumours by site

Tumours	Maxilla		Mandible		
	Anterior	Posterior	Anterior	Posterior	Both anterior /posterior
Ameloblastoma	3	9	24	63	14
Calcifying epithelial of odontogenic tumour	–	–	–	3	–
Squamous of odontogenic tumour	–	–	–	2	–
Odontoma					
Compound	–	1	–	5	–
Complex	–	1	1	3	–
Adenomatoid odontogenic tumour	–	–	1	1	–
Calcifying odontogenic cyst	–	1	–	–	0
Ameloblastic fibroma	–	–	1	1	–
Cementifying fibroma	–	–	2	–	–
Cementoma	–	–	1	10	–
Odontogenic myxoma	3	4	3	9	2
Total	6	16	33	97	16

NB: 1. In three cases of ameloblastoma it was not stated which jaw was affected.
2. In 3 8 cases of ameloblastoma of the mandible and three cases of the maxilla the exact location in the jaws was not stated.
3. The location of one odontogenic myxoma was not stated.

Table 4

Distribution of odontogenic tumours by age

Tumour	Age in years					
	0-9	10-19	20-29	30-39	40-49	>50
Ameloblastoma	3	16	42	22	20	17
Calcifying epithelial odontogenic tumour	0	0	0	2	1	0
Squamous odontogenic tumour	0	0	1	1	0	0
Odontoma						
Compound	1	4	1	0	0	0
Complex	0	2	2	1	0	0
Adenomatoid odontogenic tumour	0	1	1	0	0	0
Calcifying odontogenic cyst	0	0	1	0	0	0
Ameloblastic fibroma	0	2	0	0	0	0
Cementifying fibroma	0	1	1	0	0	0
Cementoma	0	1	1	0	0	0
Odontogenic myxoma	0	5	10	6	1	0
Total	4	31	63	35	26	17

Table 5

Time elapsed between the appearance of first signs and symptoms in patients with ameloblastoma and the time of presenting to hospital

Years	No.	%
0 - 1	23	14.6
2 - 3	54	21.6
4 - 5	27	14
6 - 7	10	6.3
8 - 9	5	3.2
10 - 11	3	1.9
12 - 13	2	1.2
14 - 15	3	1.9
>15	2	1.2
Unspecified	28	17.8
Total	157	100

Some of these tumours, however, extended to the anterior area or even occupied the whole horizontal part of the mandible (13.8%) and the remaining tumours were located solely in the anterior part from canine to canine (23.8%). Table 4 shows the distribution of the odontogenic tumours by age. The age range was from seven to 71 years. More than half (55.3%) of the patients who presented with odontogenic tumours were below 30 years of age. Ameloblastomas most often affected patients in the ages between 20 and 40 years.

Histomorphologically, the majority of the ameloblastoma diagnosed were of follicular pattern (38.2%) followed by plexiform (27.4%) and unicystic type (5.1%). The remaining 29.3% were diagnosed as ameloblastoma without further characterisation. Most patients with ameloblastoma had clinical signs and symptoms for more than a year before reporting to hospital. About one third of the patients presented within the first three years after the onset of signs and symptoms, but a considerable number of patients presented much later (Table 5). Based on the population of Tanzania (31 million), an incidence rate of odontogenic tumours of 0.4 per million and ameloblastoma of approximately 0.3 per million can be calculated.

DISCUSSION

This study focussed on the relative frequencies of the different odontogenic tumours and the incidence rates of ameloblastoma as seen in Tanzania over a fifteen-year period. The fairly large group of non-specified tumours forms a serious flaw in this retrospective study. One reason may be that some of the specimens may not have been representative of the lesions removed. It cannot be ruled out, however, that a number of these tumours in fact were odontogenic tumours, which may have lowered the total number of odontogenic tumours found.

Odontogenic tumours formed a relatively small proportion of orofacial tumours seen during this period comprising only 12.2%. This is in keeping with available African reports, which have shown relative frequencies of odontogenic tumours to vary between 2.5 and 19.1%(5,11,24). Among odontogenic tumours, ameloblastoma was most commonly seen, with a relative frequency of 73.7%. This is also in keeping with most African studies. Chidzonga *et al*(25), in Zimbabwe reported a relative frequency of 78%, while Odukoya *et al*(5) and Mosadomi(11) reported relative frequencies of 58.5% and 65.5%, respectively in Nigeria. Asian studies have presented similar figures. In China, Lu *et al*(6) and Wu and Chan(17) found relative frequencies of ameloblastoma of 58.6% and 45%, respectively. In Turkey, although the frequency of ameloblastoma was slightly lower than in the African and Asian studies it also ranked highest among odontogenic tumours with a relative frequency of 36.5%(12). These figures differ from European and American studies, which reported relative frequencies between 10% and 23.7%, second to odontoma, which was the most common odontogenic tumour(1,9,13,14).

It is apparent from these figures that the relative frequencies of ameloblastoma as found in Africa and Asia are the highest. Racial predilection, as proposed by Sawyer *et al*(19), has been disputed by others who have attributed this higher relative frequency in Africans to a "harvesting effect"(11). Tumours other than ameloblastoma and myxofibroma may also reach considerable size but will still present little discomfort to the patient which precludes him/her to seek medical treatment. Ameloblastomas and myxofibromas, however, usually present with more disfigurement causing problems, which actually forces the patient to present to hospital. It is also likely that the readily available dental services in American and Western European countries, where oral radiographic examinations are done on a routine basis, accounts for early detection of jaw tumours such as odontomas that to a large extent are symptomless. This is supported by the results of this study.

From this study it is clear that many patients with ameloblastoma in Tanzania presented themselves to the

hospital after considerable delay. The time that elapsed between the appearance of clinical signs and symptoms and reporting to hospital ranged from one year to more than fifteen years in some cases. In one case a woman reported to have had the tumour for 22 years. In addition, due to pressing socio-economic priorities in many instances patients tend to go to hospital only when there is unbearable pain, discomfort or gross interference with function. In the Tanzanian society oral health is also given a low priority compared to more life threatening medical conditions such as AIDS, malaria and malnutrition. Other factors that might have contributed to the delays are the distances one had to travel to the appropriate centre and the financial implications, the introduction of cost sharing (user fees) in the health sector in recent years, ignorance of patients and failure of health workers at primary health facilities to diagnose and refer patients(26).

Histologically, like in other studies(1,5,11,17), the follicular type of ameloblastoma dominated followed by the plexiform type. None of the other types were reported. During most of the period covered in this study there was no oral pathologist in Tanzania, therefore, the biopsy specimens were examined by general pathologists. This may be the reason why more rarely encountered histologic types of ameloblastoma such as the desmoplastic, malignant, basal cell or granular cell types were not reported. There is also a possibility that some unicystic ameloblastomas were diagnosed and treated as odontogenic cysts on the basis of clinical and radiological findings.

Only 9.7% of the ameloblastomas were located in the maxilla, 90.3% were in the mandible and in three patients it was not stated whether they were maxillary or mandibular. This is in keeping with other studies done elsewhere(5,6,12,17,25). The equal distribution of ameloblastomas between males and females is also in agreement with available data from other studies(6,12,16,17). The clinical presentation of ameloblastoma by site does not differ from other studies either. Similar findings have been reported(5,6,14, 17,25,27). However, it differs from other studies(24,28,29) where ameloblastoma was found to be located in the anterior part more often than in other parts of the mandible.

The calculated incidence rate of ameloblastomas in Tanzania seems somewhat low in the light of the suggested higher rates among African blacks. Some of the cystic ameloblastomas, however, have probably been removed without histological examination, which would affect the reported numbers in an unfavourable way. It is also likely that some patients may have been operated in some regional hospitals without input from the oral-maxillofacial surgeons, while some patients may have refused treatment in the referral centres.

Odontogenic myxoma was the second most commonly seen odontogenic tumour. Odukoya(5) in Nigeria, and Lu *et al*(6) in China also found myxoma to be the second most commonly seen odontogenic tumour after ameloblastoma with frequencies of 11.8% and 8.4%, respectively. In Turkey, although it came third after ameloblastomas and odontomas, odontogenic myxomas have a higher relative frequency (13.8%). Of the 22 myxomas seen, the majority were located in the mandible, nine of them posteriorly. This is in keeping with the present knowledge that odontogenic myxoma has a predilection for the mandible(30). The majority of odontogenic myxomas were found within the age of 20 to 40 years and none was found in children below the age of ten years. This compares with available literature(27).

The relative frequency of odontomas, both compound and complex variants was 5.2% and the majority were located in the posterior mandibular region. This low frequency contrasts with studies from Europe and America(9,13,14,19) but is in keeping with other African and Asian studies (5,6,25). There was an almost equal sex distribution and they were seen mostly between the ages of 10 to 30 years. Cementoblastoma was seen with the same relative frequency as that of odontomas also with a predilection for the mandible.

Ameloblastic fibroma, cementifying fibroma, squamous odontogenic tumour and adenomatoid odontogenic tumour (AOT) were all seen with a relative frequency of about 0.9%. Of these the AOT was seen in two females in their early twenties. This particular tumour had been seen with higher frequencies in Nigeria, 6.2%, Mexico 7.1%, China 8.3%, Germany 4%, Canada, 3.6% and United States 3.4%(1,5,6,9,13,14). The low relative frequency of most of these tumours, as seen in this study, is probably attributable to the fact that they are largely symptomless unless they reach considerable size and, therefore, patients did not present themselves. It is also possible that the specimens were not always sent for histologic examination, because of the obvious benign nature. Since expert histopathologic service was only available at the MMC during most of the period covered in this study, clinicians were not encouraged to submit specimens to avoid the cumbersome procedures involved in obtaining results.

The extremely low relative frequency of odontogenic keratocysts can probably be explained by a "reversed harvesting effect". It is known that the majority of keratocysts give rise to very few signs and symptoms such as swelling and pain. These cysts tend to hollow out the mandible instead of giving rise to expansion(31). They also tend not to be easily infected. In the Tanzanian circumstances this means patients do not see the need for medical treatment unless acute inflammatory reaction or pathologic fractures force them to report for treatment.

Ameloblastomas formed the majority of odontogenic tumours in this study. The peak age of ameloblastoma was between 20 and 29 years. Many of the patients reported to hospital late with large tumours that had already caused gross bone destruction. In order to get better treatment results, there is need to educate the population on the nature of the lesions and advantages of early treatment. The dentists and medical personnel can facilitate this through early detection and referral of the patients. A prospective study with the intention to categorise the various tumours more precisely and to calculate the incidence rate more accurately is recommended.

ACKNOWLEDGEMENTS

We would like to express our appreciation and gratitude to the administrations of the Muhimbili Medical Centre, Kilimanjaro Christian Medical Centre, Bugando Medical Centre and the Mbeya Referral Hospital for permitting us to use these data. We are also grateful to the members of staff of the Pathology and Dental departments of these Centres for their help during data collection.

REFERENCES

1. Regezi, A.J., Kerr D.A. and Coutney R.M. Odontogenic tumours: an analysis of 706 cases. *J. Oral. Surg.* 1978; **36**:771-778.
2. Adler C.P. Tumour-like lesions in the femur with cementum-like material. *Skeletal Radiol.* 1985; **14**:26-37.
3. Cohn H.C., McPherson T.A., Barnes L. and Kennedell J.S. Cemento-ossifying fibroma of the ethmoid sinus manifesting as proptosis. *Ann. Ophth.* 1985; **14**:173-175.
4. Jelic J.S., Loftus M.J., Miller A.S. and Cleveland D.B. Benign ameloblastoma: Report of an unusual case and analysis of 14 additional cases. *J. Oral Maxillofac. Surg.* 1993; **51**:1033-1037.
5. Odukoya O. Odontogenic tumours: an analysis of 289 cases. *J. Oral Path. Med.* 1995; **24**:454-457.
6. Lu Y., Xuan M., Takata T., *et al.* Odontogenic tumours. A demographic study of 759 cases in a Chinese population. *Oral Surg.* 1998; **86**:707-714.
7. Kramer I.R.H., Pindborg J.J. and Shear M. WHO Histologic Typing of Odontogenic Tumours. Berlin:Springer-Verlag 1992: 7-9.
8. Beken E., Odontogenic tumours. *Path. Ann.* 1971; **6**:475-568.
9. Daley T.D., Wysocki G.P. and Pringle G.A. Relative incidence of odontogenic tumours and oral and jaw cysts in a Canadian population. *Oral Surg.* 1994; **77**:276-280.
10. Kovi J. and Laing W.N. Tumours of the mandible and the maxilla in Accra Ghana. *Cancer* 1966; **19**:1301-7.
11. Mosadomi A. Odontogenic tumours in an African population. *Oral Surg.* 1975; **40**:502-521.
12. Gunham O., Erseven G. and Ruacan S., et al. Odontogenic tumours; A series of 409 cases. *Austr. Dent. J.* 1990; **35**:518-522.
13. Mothes P., Kreusch Th. and Harms D., et al. Die Haufigkeit odontogener Tumoren im Wachstumsalter. *Dtsch. Zahnartztl Z.* 1991;**46**:18-19.
14. Mosqueda - Taylor A., Ledesma - Montes C. and Caballero - Sandoral S. *et al.* Odontogenic tumours in Mexico: a collaborative retrospective study of 349 cases. *Oral Surg.* 1997; **84**:672-675.
15. Slavin G. and Cameron H. M. Ameloblastomas in Africans from Tanzania and Uganda. A report of 56 cases. *Brit. J. Cancer* 1969; **23**:31-38.
16. Onyango J.F., Awange D.O. and Wakiaga J.M. Oral tumours and tumour-like conditions II. Age, sex and site distribution. *East Afr. Med. J.* 1995; **72**:568-576.
17. Wu P.C. and Chan K.W. A survey of tumours of the jaws in Hong Kong Chinese. *Brit J. Oral Maxillorac. Surg.* 1985; **23**:92-1023.
18. Slootweg P.J. and Muller H. Malignant ameloblastoma or ameloblastic carcinoma? *Oral Surg.* 1984; **57**:168-176.
19. Sawyer D.N., Mosadomi A., Page D.G., *et al.* Racial predilection of ameloblastoma? A probable answer from Lagos, (Nigeria) and Richmond Virginia (USA) *J. Oral. Med.* 1985; **40**:27-31.
20. Shear M. and Singh S. Age-standardized incidence rates of ameloblastoma and dentigerous cyst on the Witwatersrand, South Africa. *Comm. Dent. Oral Epidem.* 1978; **6**:195-199.
21. Larsson A. and Almeren H. Ameloblastoma of the jaws, an analysis of consecutive series of all cases reported to the Swedish cancer registry during 1958-1971 *Acta Pathol. Microbiol. Scand.* 1978; **86**:337.
22. Muller H. Het ameloblastoom, een klinisch en histopathologisch onderzoek. Thesis, University of Utrecht, The Netherlands; 1983:p 16.
23. Awange D.O. Ameloblastoma of the jaws in East Africa. *Odontostomatol. Trop.* 1994; **17**:15-21.
24. Akinosi J.O and Williams A.O. Adamantinoma in Ibadan, Nigeria. *West Afr. Med. J.* 1968; **17**:45.
25. Chidzonga M.M., Perez V.M.L. and Alvarez A.L.P. Ameloblastoma: The Zimbabwean experience over 10 years. *Oral Surg.* 1996; **82**:38-41.
26. Asamoa E.A., Anyanlere A.O. and Olaitan A.A., Paediatric tumours of the jaws in Northern Nigeria, Clinical presentation and treatment. *J. Cranio-Maxillofac. Surg.* 1990; **18**:130-135.
27. Reichart P.A., Philipsen H.P. and Sonner S. Ameloblastoma: Biological profile of 3677 cases. *Eur J. Cancer* 1995; **31**:86-99.
28. Arotiba G. A study of Orofacial tumours in Nigerian Children. *J. Oral Maxillofac. Surg.* 1996; **54**:34-38.
29. Olaitan A.A., Adeola D.S. and Adekeye E.O. Ameloblastoma: Clinical features and management of 315 cases from Kaduna, Nigeria. *J. Cranio-Maxillofac. Surg.* 1993; **21**:351-355.
30. Van der Waal I. Diseases of the jaws: Diagnosis and treatment. Chapt 10 p 204 Munksgaard, Copenhagen, 1991.
31. Stoelinga P.J.W. Long-term follow-up on keratocysts treated according to a defined protocol. *Int. J. Oral Maxillofac. Surg.* 2001; **30**:14-25.