

ORIGINAL ARTICLE

ODONTOGENIC TUMOURS IN BENIN CITY, NIGERIA: analysis of 220 cases and review of literature

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ABSTRACT

OBJECTIVE: Recent reports suggest a high incidence of odontogenic tumours in Nigerians, particularly ameloblastoma. This study further evaluates the age, gender, duration, site of occurrence and various histopathologic types of odontogenic tumours in the South-South Nigerian population.

METHOD: The patients' case notes, biopsy reports and histopathology slides of 1298 diagnosed cases of orofacial lesions between January 1991 and April 2014 were retrospectively reviewed. Patients with odontogenic tumours were selected and the age, gender, duration, site and histopathologic types of the lesion were analyzed.

RESULTS: Two Hundred and twenty (17.0%) of the patients had odontogenic tumours among the diagnosed orofacial lesions within the study period. The patients' ages ranged from 7 to 73 years, with a mean age of 32 ± 1.4 years and the peak age group was the 3rd decade of life (n=63, 28.6%). There were 114 (51.8%) males and 106 (48.2%) females, giving a ratio of 1.1: 1. The mean duration of the lesions on presentation was 43 ± 5.6 months and posterior mandible was the commonest site (n=139, 63.2%) of the lesions. Ameloblastoma (n=167, 75.9%) was the most common histopathologic type of the odontogenic tumours, which was significantly associated with the posterior mandible (n=107, 48.6%) [p=0.000]. This was distantly followed by odontogenic myxoma (n=11, 5.0%).

CONCLUSION: This study shows a high incidence of ameloblastoma and a lower overall incidence of odontogenic tumours in younger age groups with variable gender distribution and fairly similar site predilection of the various histopathologic types of odontogenic tumours compared to previous reports.

Keywords: Odontogenic tumours; incidence; histopathologic types

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INTRODUCTION

There is still controversy surrounding the incidence of odontogenic tumours, particularly ameloblastoma in black Africans compared to the Caucasian race.¹⁻⁴ Earlier reports⁵⁻⁷ indicated a low incidence of jaw tumours in Africans and attributed the apparently high incidence to “harvesting phenomenon.” This theory relates the late presentation of patients for treatment to the slow growing, painless and non-life threatening (except the malignant tumours) nature of the tumours. Consequently, a large number of Africans with jaw tumours including odontogenic tumours were seen at newly established hospitals and treatment centres over a relatively short period of time.

There are a few Nigerian studies so far on this subject, with earlier reports indicating a high incidence of ameloblastoma and a relatively low incidence of other odontogenic tumours in Nigerians.^{5,6,8,9} Arotiba et al⁴ reported a relatively higher incidence of odontogenic tumours, consisting mainly of ameloblastoma together with adenomatoid odontogenic tumour and fibromyxoma, when compared to previous Nigerian reports. Whereas, recent studies in another black African population^{10,11} confirmed a high incidence of ameloblastoma and low incidence of odontogenic tumours similar to what obtains in several European and North American studies. It is therefore obvious that the lack of consensus surrounding the true incidence of odontogenic tumours in Africans may warrant many more studies in the future.

The purpose of this study is to further evaluate the age, sex, duration, and site of occurrence of the various histopathologic types of odontogenic tumours, seen over a 23-year period in the South-South region of Nigeria and the findings of this study would be compared with those of previous Nigerian, other African and Caucasian reports, to ascertain the true incidence of these lesions.

PATIENTS AND METHODS

Approval was obtained from the Hospital Ethical Committee to retrospectively review of the clinical case notes, biopsy reports and histopathology slides of patients with diagnosed orofacial lesions over a 23-year period (January 1991 to April 2014) in the Department of Oral Pathology and Medicine, University of Benin Teaching Hospital, Benin City, Nigeria. Based on the World Health Organization (WHO) 2005 criteria for histological classification of odontogenic tumours;¹² the histopathologic diagnosis of the odontogenic tumours was reviewed.

The patients with confirmed diagnosis of odontogenic tumours were further analyzed for age, gender, duration and site of occurrence of the various histopathologic types of the lesions. The data collected were analyzed using SPSS version 16 and statistical correlation of the variables was performed using Pearson’s Chi-square, with a confidence level of 95% and probability values (P-value) of $P < 0.05$ regarded as significant.

RESULTS

A total of 220 (17.0%) patients had odontogenic tumours among the 1298 diagnosed orofacial lesions within the study period. The patients’ ages ranged from 7 to 73 years, with a mean age of 32 ± 1.4 years and the peak age group was the 3rd decade of life ($n=63$, 28.6%). There were 114 (51.8%) males and 106 (48.2%) females, giving a ratio of 1.1: 1 (Table 1). The mean duration of the lesions on presentation was 43 ± 5.6 months and posterior mandible was the commonest site ($n=139$, 63.2%) of the lesions. Ameloblastoma was the most common histopathologic type ($n=167$, 75.9%), followed by odontogenic myxoma

Table 1: Age and gender distribution of the odontogenic tumours

Tumour	Age and Sex																		Total	Total	%
	0 – 9		10 - 19		20 – 29		30 – 39		40 – 49		50 – 59		60 – 69		=/ <u>≥</u> 70		Total				
	F	M	F	M	F	M	F	M	F	M	F	M	F	M	F	M	F	M			
Ameloblastoma	1	1	16	22	27	20	16	24	13	5	3	13	2	3	-	1	78	89	167	75.9	
Fibromyxoma	-	-	-	3	1	-	3	1	2	1	-	-	-	-	-	-	6	5	11	5.0	
AOT	-	-	3	1	-	1	-	-	-	-	-	-	-	-	-	-	3	2	5	2.3	
CEOT	-	-	-	-	2	1	-	1	-	-	-	-	-	-	-	-	2	2	4	1.8	
COC	-	-	-	-	-	-	1	1	-	-	-	-	-	-	-	-	1	1	2	0.9	
GCOC	-	-	-	-	-	1	1	-	-	1	-	-	-	-	-	-	1	2	3	1.4	
Ameloblastic Fibroma	-	-	2	1	1	-	-	-	-	-	-	-	-	-	-	-	3	1	4	1.8	
Ameloblastic Carcinoma	-	-	-	-	-	1	1	-	1	2	-	1	1	1	-	-	3	5	8	3.6	
Odontogenic Fibroma	-	-	-	1	1	1	-	-	-	-	1	-	-	-	-	-	2	2	4	1.8	
Compound Odontome	-	1	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-	1	1	0.5	
Keratocystic Odontogenic tumour	-	-	1	-	3	2	2	-	-	1	-	-	-	1	-	-	6	4	10	4.5	
Odontogenic sarc.	-	-	-	-	1	-	-	-	-	-	-	-	-	-	-	-	1	-	1	0.5	
Total	3		50		63		51		26		18		8		1		106	114	220	100	

Note: CEOT = Calcifying epithelial odontogenic tumour; COC = Calcifying odontogenic cyst; GCOC = Ghost cell odontogenic carcinoma; Odontogenic sarc.= odontogenic sarcoma

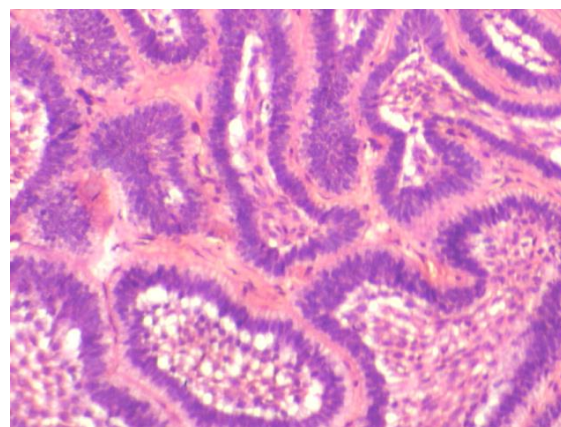
(n=11, 5.0%), keratocystic odontogenic tumour (n=10, 4.5%) and ameloblastic carcinoma (n=8, 3.6%). The uncommon odontogenic tumours were calcifying epithelial odontogenic tumour, ghost cell odontogenic carcinoma, calcifying odontogenic cyst, adenomatoid odontogenic tumour, ameloblastic fibroma, odontogenic sarcoma and compound-composite odontome [Table 2].

Ameloblastoma

There were 167 (75.9%) ameloblastoma, with male (n=89) to female (n=78) ratio of 1.1:1. The ages ranged from 7 to 73 years (mean=30 years) with a peak incidence in the third decade of life (n=47, 21.4%), [Table 1]. There were 141 (64.1%) mandibular lesions, 20 (9.1%) maxillary lesions and 6 (2.7%)

gingival lesions. The most common site was posterior (body/angle/ramus) mandibular region (n=107, 48.6%) [Table 2]. The duration on presentation was 2 months to 10 years, with mean duration of 24 months (Figure 1).

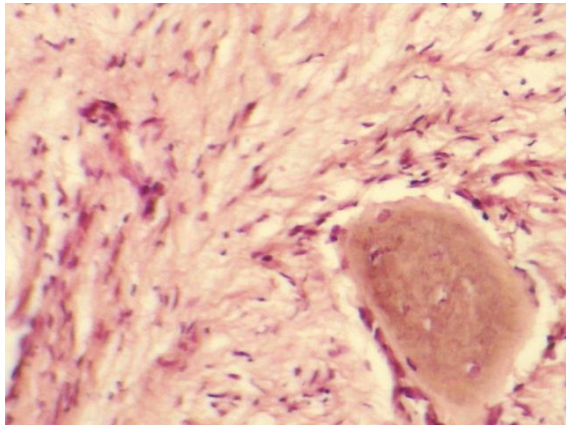
Figure 1: Follicular ameloblastoma



Odontogenic myxoma

There were 11 (5.0%) cases of odontogenic myxoma, with female (n=6) to male (n=5) ratio of 1.2:1. The age range was between 14 to 41 years (mean=29), with peak incidence in the fourth decade of life (n=4, 1.1%), [Table 1]. The lesion was slightly commoner in the mandible (n=6, 2.7%) than the maxilla (n=5, 2.3%), [Table 2]. The duration on presentation was 1 months to 3 years, with mean duration of 15 months (Figure 2).

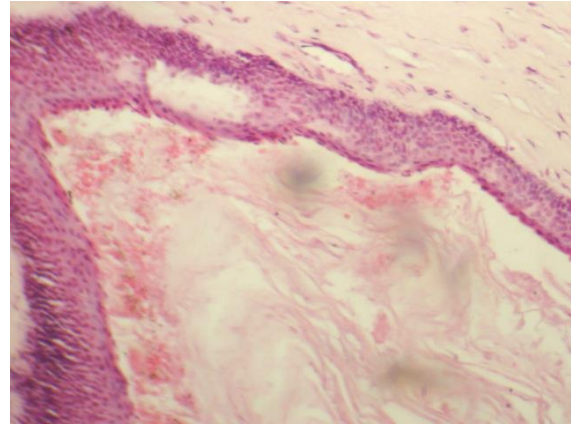
Figure 2: Odontogenic myxoma showing myxoid stroma, with odontogenic epithelial rest and residual bone



Keratocystic odontogenic tumour

There were 10 (4.4%) cases of keratocystic odontogenic tumour, with female (n=6) to male (n=4) ratio of 1.5:1. The age range was between 18 to 61 years (mean=24 years), with peak incidence in the third decade of life (n=5, 2.3%) [Table 1]. The lesion was found in the posterior mandible (n=8, 3.6%). The duration on presentation was 3 months to 5 years and the mean duration was 30 months (Figure 3).

Figure 3: Keratocystic odontogenic tumour lined by thin stratified squamous epithelium and thick fibrous capsule, with keratin squames the cystic lumen



Ameloblastic carcinoma

There were 8 (3.6%) cases of ameloblastic carcinoma, with male (n=5) to female (n=3) ratio of 1.7:1. The age range was between 27 to 66 years (mean=43 years), with peak incidence in the fifth decade of life (n=3, 1.4%) [Table 1]. The lesion was found in the posterior mandible (n=5, 2.3%). The duration range of symptom was 3 to 15 months and the mean duration was 8 months (Figure 4).

Figure 4a: Ameloblastic carcinoma with focal squamous metaplasia and keratinization

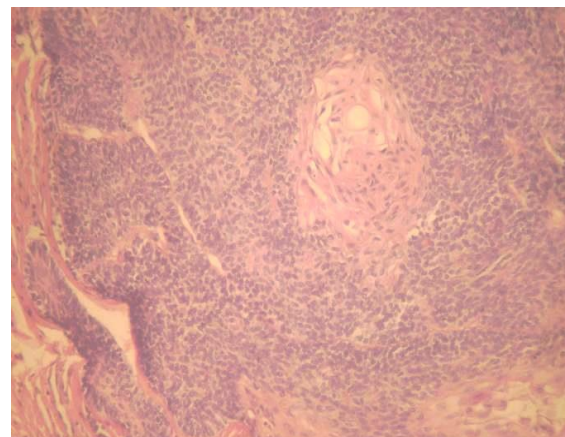
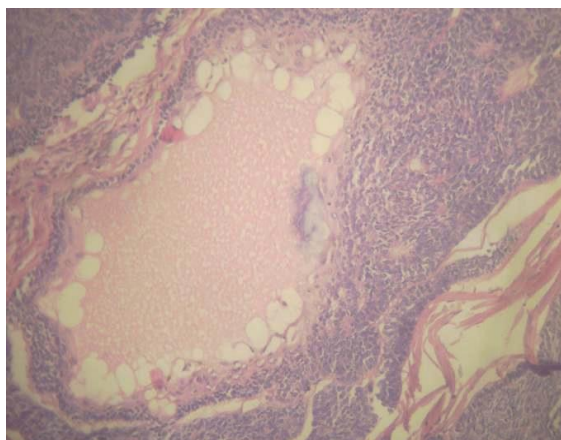


Figure 4b: Ameloblastic carcinoma with focal necrosis



Other odontogenic tumours

The other odontogenic tumours found in this study were adenomatoid odontogenic tumour (n=5, 2.3%), calcifying epithelial odontogenic tumour (n=4, 1.8%), ghost cell odontogenic carcinoma (n=3, 1.4%), calcifying odontogenic cyst (n=2, 0.9%), ameloblastic fibroma (n=4, 1.8%), odontogenic fibroma (n=4, 1.8%) and compound composite odontome (n=1, 0.5%)[Tables 1, 2 and 3].

DISCUSSION

Odontogenic tumours constitute a small group of all tumour and tumour-like lesions of the jaws and oral mucosa in adults.^{6,9} As low as 12.9% was reported in one study,⁶ although a higher incidence has been reported in children.¹⁴⁻¹⁶ So far, there has been disagreement over the actual incidence of these tumours, especially ameloblastoma in black Africans. 'Epidemiological obstacles' are still hampering the possibility of determining the accurate incidence of the jaw tumours in many African countries.^{5,11} Although attempts are now being made to determine the true incidence of these tumours, it is still difficult to draw conclusion from the few studies from Africa in the literature because of the use of different sampling methods, classification and the wide variation in the findings of the studies,

^{7,10,11} especially in the Nigerian population.^{4-6,9}

This study is useful because histopathologic diagnoses of odontogenic tumours at the centre were based on current WHO criteria.¹² Consequently, it provided a more reliable data of the various histopathologic types of these tumours seen in this centre. In this study, odontogenic tumours constituted 17.0% of all histopathologically diagnosed orofacial lesions. Compared to previous reports, there appears to be a fairly consistent low incidence of odontogenic tumours amongst orofacial tumour and tumour-like lesions in Nigerians,^{5,6,9} except in the series reported by Arotiba et al.⁴ (30%, from Ibadan) and Adebayo et al.¹⁷ (32%, from Kaduna) with a relatively higher incidence. In most African series a high incidence of odontogenic tumours has been consistently reported, when compared to those from Caucasians studies.^{1-6,9-11,17}

In some Caucasian studies,^{3,13} female predilection for odontogenic tumours has been reported, although there was almost equal gender distribution of the tumours in this study. This finding is in keeping with those of previous Nigerian studies on these tumours^{4,5}. Recent reports indicate a male preponderance of patients with ameloblastoma in Nigeria,^{4,9,17} which was ascribed as possible reason for overall equal gender incidence of odontogenic tumours in the study by Arotiba et al.⁴ Similarly, this study shows slight male predilection for ameloblastoma, ameloblastic carcinoma and ghost cell odontogenic carcinoma; while slight female predilection was observed for fibromyxoma, keratocystic odontogenic tumour, adenomatoid odontogenic tumour and ameloblastic fibroma. The variation in the gender distribution of the various histopathologic types of odontogenic tumours, suggests that there may be no strong gender predilection for them.

Most odontogenic tumours in this study presented in younger ages, with peak incidence in the third decade of life. This agrees with reports from previous African studies,^{4,5,16,17} except in one series reported by Simon et al,¹¹ which indicated a higher average age similar to those reported from European studies.¹⁸⁻²⁰ This apparent older age was alleged to be due to late presentation for treatment.⁵⁻⁷ This study also showed an overall mandibular predilection, and the jaw distribution of odontogenic tumours is fairly similar to those previously reported from both Nigerian and other African studies.^{4,5,9-11}

Ameloblastoma had the highest incidence amongst the various histopathologic types of odontogenic tumour in this series. This agrees with previous reports from Nigeria,^{4-6,9} Tanzania^{10,11} and Hong-Kong.¹³ The apparent higher incidence rate of ameloblastoma in black Africans may be to some yet to be discovered genetic and environmental factors. Studies on oncogenes, growth factors and molecules involved in abnormal cell proliferation will assist in predicting why ameloblastoma is commoner in blacks than Asians and Caucasians. However, some studies have attributed the apparent higher incidence rate of ameloblastoma in black Africans to the wide symptom duration before presentation for treatment.^{5,11}

Perhaps, a lower incidence of ameloblastoma may be found in black Africans, when the incidence rate is calculated per million of the population. Simon et al¹⁰ observed a lower incidence rate (0.3 %) of ameloblastoma, when the initial 73% incidence rate of the lesion in an earlier Tanzanian study was calculated per million of the total population of Tanzania. Whereas in a more recent Tanzanian study by Simon et al,¹¹ they found

a similar incidence rate of ameloblastoma comparable to those reported in several European studies.^{1, 3,18,20,21}

However, when we compared the incidence rate (75.9%) from our study per million of the Nigerian population (recent population figure of 140 million), we found a rate of 0.5% which is slightly higher than Simon et al's finding. This observation suggests a relatively higher incidence of ameloblastoma in Nigeria compared to Tanzania.

This study also shows an almost equal gender incidence, with slight male predilection for ameloblastoma, similar to recent Nigerian studies which reported a male preponderance for ameloblastoma.^{4,9,17,23-25} However, slight female predilection was previously reported from Nigeria,⁵ Tanzania¹¹ and most Caucasian series.^{1,3,21,22} Although, some studies from Nigerian and other African countries have reported that ameloblastoma more often affects the symphyseal region in black Africans,^{11,16,22,24,26} the posterior mandibular region was the commonest site of the lesion in this study. This agrees with previous findings in most Nigerian and African studies.^{4,5,9,11,17,25,26} However, the reason for this is still uncertain, although Akinosi et al²⁷ suggested that calculus deposits and sepsis might be implicated, following the difficulty in ensuring good oral hygiene in the posterior region.

Odontogenic myxoma was the second most common odontogenic tumour, with a low incidence and a younger mean age compared to previous reports.^{4, 10,11,28} The lesion was slightly commoner in female and located in the posterior mandibular region. These findings compared with earlier reports by Minderjahn²⁹ and Adekeye et al,³⁰ and recent Nigerian reports;^{4,9,17} suggests variable gender distribution and slight mandibular predilection of the lesion in Nigerians.

Table 2: Sites of the odontogenic tumours

Mandible	Maxilla
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Tumour type	No. of Patients			Gingival		
		Anterior	Posterior		Anterior	Posterior
Ameloblastoma	167	34	107	6	5	15
Fibromyxoma	11	-	6	-	2	3
CEO	4	1	2	-	-	1
COC	2	1	1	-	-	-
GCOC	3	-	2	-	-	1
AOT	5	-	-	-	4	1
Odontogenic Fibroma	4	-	2	1	-	1
Ameloblastic Fibroma	4	-	4	-	-	-
Ameloblastic Carcinoma	8	-	5	-	-	3
Compound Odontome	1	-	1	-	-	-
Keratocystic Odontogenic tumour	10	-	8	-	-	2
Odontogenic sarcoma	1	1	-	-	-	-
Total	220	36	139	7	11	27

Note: AOT = Adenomatoid odontogenic tumour; CEO = Calcifying epithelial odontogenic tumour; COC = Calcifying odontogenic cyst; GCOC = Ghost cell odontogenic carcinoma

The third most common odontogenic tumour in this study was keratocystic odontogenic tumour. The lesion was found mostly in young adults with slight predilection for females and a predominance of the lesion in the posterior mandibular site. Although, there is agreement in the age and site of the lesion, there is variation in the gender predilection of the lesion in this study compared with previous report.³¹

Ameloblastic carcinoma was the fourth most common odontogenic tumour in this study, with predilection for elderly male patients and in the posterior mandibular region. These findings are similar to previous reports.³¹ This lesion had the shortest mean duration on presentation compared to the other odontogenic tumours in this study. The patients may have sort treatment earlier because of the usual rapid growth and sometimes the trismus and dysphonia associated with this malignant lesion.^{31, 32}

Among the uncommon odontogenic tumours in this study, adenomatoid odontogenic tumour and ameloblastic fibroma were found

mostly in females; odontogenic fibroma and calcifying epithelial odontogenic tumour occurred equally in both gender, while ghost cell odontogenic carcinoma had a slight male predilection. These lesions occurred mostly in the young adults and posterior mandibular region, except adenomatoid odontogenic tumour with predilection for the anterior maxilla. The low incidence of these lesions is consistent with those of previously reported African series.^{4,9,11,17, 33} There is disagreement in the literature on the age and gender incidence of these lesions.^{17, 34}

In conclusion, this study shows a high incidence of ameloblastoma and a lower overall incidence of odontogenic tumours amongst Nigerians; with a younger mean age, variable gender incidence and fairly similar site predilection of the various histopathologic types of the lesions compared to previous reports. Apart from ameloblastoma in the blacks and odontomes in Caucasians,^{1,3,35} there appears to be no consistent pattern of presentation of the various histopathologic types of odontogenic

tumours in both African and Caucasian series.

Conflict of Interest: None declared

Table 3: Comparison of percentage incidence of odontogenic tumours

Tumour	Present study (Benin City) %	Arotiba et al ⁴ (Ibadan) %	Mosadomi ⁵ (Lagos) %	Odukoya ⁹ (Lagos) %	Adebayo et al ¹⁷ (Kaduna) %	Simon et al ¹⁰ (Tanzanian) %	Regezi et al ³ (Michigan) %	Wu and Chan ¹³ (Hong-Kong) %	Gunham al ¹ (Turkey) %
Ameloblastoma	76	59	66	59	73	80	11	62	37
Adenomatoid Odontogenic tumour	2	13	7	6	3	1	3	4	3
Odontogenic myxoma	5	16	0	12	12	7	3	1	13
Ameloblastic - fibroma	2	3	7	5	3	2	2	0	5
- fibrosarcoma	0	1	0	<1	0	0	0	0	0
- carcinoma	4	0	0	0	0	0	0	0	0
Calcifying epithelial cyst	1	2	3	2	3	2	2	2	1
Calcifying epithelial Odontogenic tumour	2	2	0	<1	1	2	<1	0	2
Ghost cell odontogenic Carcinoma	1	0	0	0	0	0	0	0	0
Central odontogenic fibroma	2	2	0	5	1	2	0	4	5
Peripheral odontogenic fibroma	<1	0	0	0	0	0	0	4	0
Keratocystic odontogenic tumour	5	0	0	0	0	0	0	0	0
Ameloblastic fibro-odontoma	0	1	3	0	<1	0	2	1	1
Odontome- compound	<1	0	10	4	2	3	37	6	9
-complex	0	0	0	0	0	0	30	0	9
Cementoblastoma (Cementoma)	0	0	7	1	0	2	0	17	0
Clear cell odontogenic carcinoma	0	0	0	5	1	1	0	0	0
Intraosseous carcinoma	0	2	0	0	0	0	0	0	0
Squamous odontogenic tumour	0	0	1	<1	0	0	0	0	0
Odontogenic sarcoma	<1	0	0	<1	<1	0	0	0	0
Total number of cases	220	128	29	289	318	116	706	82	409

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