Original Article

Macroscopic Evaluation of Semi-Solid Cavity Contents in Ameloblastoma

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Abstract

Background: Intraosseous ameloblastoma is a benign odontogenic tumour with cystic cavitations which frequently contain tumour fluid and semi-solid materials. Clinical examination of ameloblastoma tumours by needle aspiration often turned out negative yields. There are claims that ameloblastoma could spill during surgery and contribute to tumour recurrence. **Aim:** The objective of the study is the macroscopic identification and prevalence of types of semi-solid cystic contents of ameloblastoma. **Patients, Materials and Methods:** This is a 10-year retrospective macroscopic evaluation of semi-solid contents from cavities of excised ameloblastoma. The subjects were consecutive patients treated at a tertiary hospital in Enugu. **Results:** The cystic cavities of 22 excision specimens were studied. Three types of semi-solid materials were observed alone or concurrently: clear or translucent gelatinous material alone in 50.0% (11), gray–white cheesy material alone in 31.8% (7), concurrent gelatinous material with cheesy material or blood clot in different compartments 13.6% (3), and blood clot alone in 4.6% (1). The hue (colour shade) distribution of the 14 gelatinous materials (alone and concurrent) was brownish 78.6% (11), greenish 14.3% (2), and pinkish 7.1% (1). There was no significant association between type of fluid aspirates at incisional biopsy and type of semi-solid cavity content, or type of semi-solid cavity content with age, recurrence, size, pain, or duration. Recurrent ameloblastoma was observed more with gelatinous materials though this was not statistically significant ($P = 0.68$). **Conclusion:** Clear: gelatinous, clots, and cheesy cystic contents could cause negative fluid aspirate yield and constitute the spillable tumour materials during surgery or cortical perforation of ameloblastoma.

Keywords: Ameloblastoma, blood clot, cystic cavity, gelatinous material, gray–white cheesy material

Introduction

Ameloblastoma of the jaws is the most common benign odontogenic tumour in Nigeria.[1] It could present clinically as solid, peripheral, unicystic, and metastatising.[2] The unicystic and solid types could appear unilocular or multilocular radiographically[3] and are associated with single or multiple cystic cavitations, respectively.[2,4] Cavities of ameloblastoma contain tumour fluids,[5] blood clots,[6] and other semi-solid materials (gray–white mass or gray–pink gelatinous mass).[3,6,7] The tumour fluids and the blood clots reported in ameloblastoma cavities could represent the progressive formation of interstitium with the expression of various vascular endothelial factors.[8] Negative fluid aspiration has often been observed during clinical examination of ameloblastoma.[5]

The tumour cystic cavities following jaw bone buccolingual expansion or cortical expansions appear as radiographic features of unilocular or multilocular radiolucencies.[9] Sonographic studies also showed the various cystic features with solid contents.[10] These features are observed macroscopically as multiple cavitations in surgical specimens of ameloblastoma.

The negative needle aspirate yields of 19.6% reported during clinical examination of ameloblastoma suggested solid or semi-solid cavity contents.[8] These solid or semi-solid cavity contents have rarely been studied or reported. The tumour fluid and semi-solid cavity contents are more likely to constitute tumour spill material due to their relative flowability and ease of detachment. Surgery-associated tumour spillage and tumour embolisation into lymphatic or blood vessels and

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aggressive tumour have been considered responsible for the high recurrence rate of ameloblastoma.[11]

The demonstration of semi-solid materials within the cystic cavities of ameloblastoma would substantiate the theory of spillage of potentially tumour-containing materials during surgery or cortical perforation[11,12] and also explain the negative aspirate yield during clinical examination of ameloblastoma cases.

Patients, Materials and Methods

This is a retrospective study of 22 excisional surgical specimens of jaws tumours with cystic cavities submitted to the Oral Pathology Unit and preserved in 10% formal saline. The histopathology study of the surgical specimen confirmed the diagnosis of ameloblastoma. The preserved excisional jaw segments were sectioned and macroscopically examined for semi-solid contents. The type of tumour fluid at biopsy was obtained from the laboratory forms of each specimen. The type of semi-solid cavity content, and the hue (colour shade) of the contents were documented. The clinico-pathologic information of each case such as gender, site of tumour, age at presentation, duration, complaint of pain, and histologic subtype of ameloblastoma was obtained from the biopsy forms, histopathology reports, and case files of the patient archived in the department.

A total of 206 cases of surgical specimens of ameloblastoma were identified in the archived specimen store and records over a 10-year period (June 2012–June 2022). There were 31 large excision specimens selected for the study out of which only 22 cases met the inclusion criteria of intraosseous ameloblastoma with semi-solid cavity content.

Inclusion and exclusion criteria

Included in the study were excisional specimens of intraosseous ameloblastoma that contained semi-solid materials in the cystic cavity or cavities. Excluded were excisional specimens of intraosseous ameloblastoma, including long standing cases, described as empty or contained only tumour fluids in the cystic cavities. Also excluded were large tumour specimens excised in fragments, excision specimens without cystic cavities, incisional specimens, and excisional specimens without adequate documentation or description of the cut surface.

The statistical software used for data analysis was the Statistical Product for Service and Solution IBM SPSS Statistics for Windows, version 24.0 (IBM Corp., Armonk, NY, USA). The analysis used descriptive statistics and results were presented as tables, means, frequencies, percentages, and standard deviation. The test for a statistical association between the variables at a 95% confidence interval was carried out using the Chi-square test, and $P < 0.05$ was considered as statistically significant. Approval was obtained from the Ethical Committee of the University of Nigeria College of Medicine Research Ethics Committee with Protocol No.: 0125/11/2021.

Results

A total of 206 ameloblastoma specimens were identified. There were 31 large specimens selected for the study out of which 22 surgical resection specimens met the inclusion criteria [Table 1]. The gender distribution was 40.9% (9) male and 59.1% (13) female. The multicystic cavities showed distinct bony or fibrous septa/walls separating cavities from each other [Figure 1]. Some of these cavities contained tumour fluids and/or semi-solid materials in the different compartments of the same tumour [Figure 1].

Semi-solid materials

Table 2 shows the three different types of semi-solid intracystic materials observed with their corresponding prevalence: (a) gelatinous material 50.0% (11) [Figure 1], (b) grayish–white friable cheesy material 31.8% (7) [Figure 2], and (c) blood clot 4.6% (1).

Three of the specimens contained gelatinous material with grayish–white friable cheesy materials or blood clot in different compartments of the same tumour, 13.6% (3) [Figure 3]. In concurrent cases, the gelatinous material was brown in two cases while being pink in the second case. The concurrent cheesy material occurred in two cases with gelatinous material, while the third specimen was blood clot with gelatinous material.

Table 3 shows the colour distribution of the gelatinous material including hues of brown, green, and pink. Brownish hue gelatinous colouration was most common, 78.6 (11). The cheesy material was coloured grayish white.

The corresponding tumour aspirates obtained from these ameloblastoma cases at incisional biopsy are shown in Table 4 with predominance of straw aspirates 45.0% (9), followed by the dark-brown aspirate 30.0% (6). Table 5 shows the semi-solid cavity material with the corresponding fluid aspirate at incisional biopsy. There was no statistically significant relationship, $P = 0.7$.

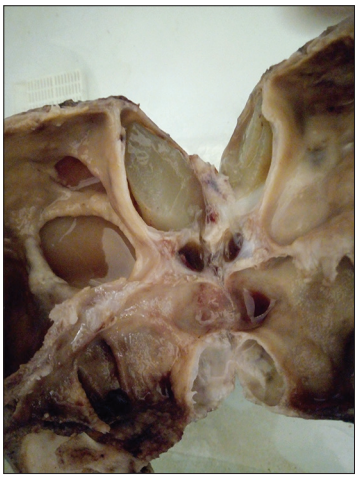

Figure 1: Subtotal mandibulectomy specimen of ameloblastoma. The cut surface is multicystic with gelatinous materials coloured gray-to-brownish hue within some cavities

Table 1: Clinical details of ameloblastoma cases with semi-solid cystic cavity contents

Gender/ age (years)	Duration (months)	Site	Widest tumour diameter (cm)	Histologic type	Recurrent	Semi-solid material	Colour shades/ hues of semi-solid materials	Fluid aspirate at biopsy
Female/25	72	Mandible posterior		Solid	No	Cheesy	Gray-white	Negative
Male/35	3	Mandible posterior	10	Solid	Yes	Cheesy	Gray-white	Dark brown
Male/20	12	Mandible anterior	12	Solid	No	Cheesy	Gray-white	Straw
Female/16	6	Mandible anterior	12	Solid	No	Cheesy	Gray-white	Straw
Male/13	36	Mandible posterior	12	Solid	No	Cheesy	Gray-white	Straw
Female/22	48	Mandible posterior	-	Solid	Yes	Gelatinous	Brownish-hue	Dark brown
Female/43	72	Mandible posttemporal bone	4	Solid	Yes	Cheesy	Gray-white	Negative
Female/39	168	Mandible posterior, condyle, maxilla	33	Solid	No	Gelatinous	Greenish-hue	Dark brown
Female/29	120	Mandible anterior	16	Solid	No	Gelatinous	Brownish-hue	Dark brown
Male/47	24	Mandible posterior	25	Solid	Yes	Gelatinous	Brownish-hue	Straw
Male/22	48	Mandible anterior	-	Solid	No	Gelatinous	Brownish-hue	Dark brown
Female/22	96	Mandible posterior	-	Solid	No	Mixed gelatinous and cheesy	Pinkish-hue	Dark brown
Female/17	36	Mandible posterior	-	Solid	No	Mixed gelatinous and cheesy	Brownish-hue	Negative
Male/16	60	Mandible bilateral	15.5	Solid	No	Gelatinous	Brownish-hue	Straw
Male/22	12	Mandible posterior	10	Solid	No	Gelatinous	Greenish-hue	Serosanguinous
Female/20	24	Mandible posterior	16	Solid	Yes	Gelatinous	Brownish-hue	Straw
Female/16	24	Mandible posterior	12	Solid	No	Gelatinous	Brownish-hue	Negative
Male/33	72	Mandible bilateral	16	Solid	Yes	Gelatinous	Brownish-hue	Straw
Female/15	48	Mandible bilateral	12.5	Unicystic (mural)	No	Gelatinous	Brownish-hue	Straw
Male/29	14	Mandible posterior	8	Solid	No	Blood clot	Dark	Straw
Female/19	15	Mandible posterior	6	Solid	No	Mixed gelatinous and blood clot	Brownish-hue and dark	Negative
Female/17	24	Mandible posterior	8	Solid	No	Cheesy	Gray-white	Straw

Table 2: Frequency of semi-solid cavity content at cut-up procedure

Cavity content	% (*n*)
Gelatinous material	50.0 (11)
Cheesy material	31.8 (7)
Mixed compartments (gelatinous, cheesy, blood clot)	13.6 (3)
Blood clot	4.6 (1)
Total	100.0 (22)

Age, duration, site, and clinical features

The overall mean age at clinic presentation was 25.1 ± 9.8 (range: 13–47) years. Estimations of age of tumour onset based on the duration showed that the mean age at tumour onset was 19.2 ± 7.3 (range: 10–35) years. The mean duration of lesion at clinic presentation was 49.8 ± 41.8 (range: 3–168) months. Majority of the ameloblastoma cases, 14 (63.6%), based on tumour duration had an early onset before 20 years of age, while at clinical presentation majority, 12 (54.5%) had moved into the 20–40-year age group. There was no association of the cavity semi-solid contents with age at onset, $P = 0.37$, or with age at presentation, $P = 0.95$, nor with tumour duration, $P = 0.20$.

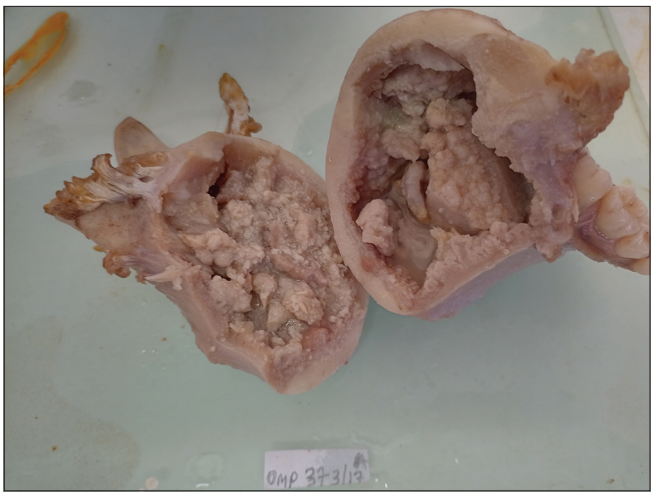

Figure 2: A hemimandibulectomy specimen with cut section showing a large cystic cavity filled with cheesy gray–white materials. The cortical bone is still intact

The frequency of the location of tumour with semi-solid contents exhibits predilection for the posterior mandible (60.0%) compared to other locations such as the anterior mandible 20.0% (4) and posterior maxilla/zygoma 20% (4). There was

no association of the semi-solid cavity contents with size of tumour (area), $P = 0.26$.

All the complaints of pain (7/7) were observed in patients with ameloblastoma containing gelatinous materials either alone or mixed with cheesy materials or blood clot (14/22). There was no association of the semi-solid cavity contents with pain, $P = 0.20$.

Only one case of ulceration was observed and although it occurred in the case with gelatinous material, it was not statistically significant, $P = 0.86$. There was no association of the semi-solid cavity contents with size of tumour (area), $P = 0.26$.

Histologic diagnosis

The histologic diagnoses showed that 95% (19) were solid ameloblastoma while only 5.0% (1) was mural unicystic ameloblastoma. Similarly, 30% (6) were recurrent tumours out of which 4/6 showed gelatinous contents, while 2/6 exhibited cheesy contents. The difference was not statistically significant, $P = 0.68$. There was no association of the semi-solid cavity contents with histologic diagnosis, $P = 0.83$.

Discussion

In this study, ameloblastoma with semi-solid cavity contents had predilection for the posterior mandible and a near equal gender distribution similar with reports on solid intraosseous ameloblastoma.[9] The observed mean ages at the onset of tumour and its clinic presentation of 19 and 25 years, respectively, suggest a youthful age of patients that develop semi-solid cavity contents. The duration of ameloblastoma in this study with a range of 3–168 months implies that semi-solid materials could develop early in tumour cystic cavities and may not be entirely dependent on long duration. Among excluded cases in the study were long standing tumours with empty cystic cavities or containing only tumour fluids. Tumour duration as a factor was not found to have any statistically significant association with the observation of semi-solid cystic contents.

The observation in this study that multicystic cavities were often distinctly separated by bony or fibrous septa made it possible to separately identify compartments occupied by either gelatinous, cheesy consolidated semi-solid materials, or both occurring simultaneously in the same tumour but in different compartments. The reason for the development of cystic compartments in ameloblastoma is unknown to the author.

This study observed three types of semi-solid contents: clear gelatinous material, gray–white cheesy material, and blood clot. While tumour fluids of ameloblastoma have been reported,[8,13] blood clots,[6] gelatinous material,[14] and gray–white cheesy material have only received scant mentions.[7] Previous clinicopathology studies of ameloblastoma did not discuss notable macroscopic features nor evaluate the cystic contents in depth[6,15-17] probably because the focus was on

Table 3: Frequency of colour shades of gelatinous material

Colour shades	% (*n*)
Brownish hue (one case from mixed compartment)	78.6 (11)
Greenish hue	14.3 (2)
Pinkish hue (from mixed compartment)	7.1 (1)
Total	100.0 (14)

Table 4: Frequency of aspirate type during incisional biopsy

Aspirate type at incisional biopsy	% (*n*)
Straw	45.5 (10)
Dark brown	27.3 (6)
Negative	22.7 (5)
Serosanguinous	4.5 (1)
Total	100.0 (22)

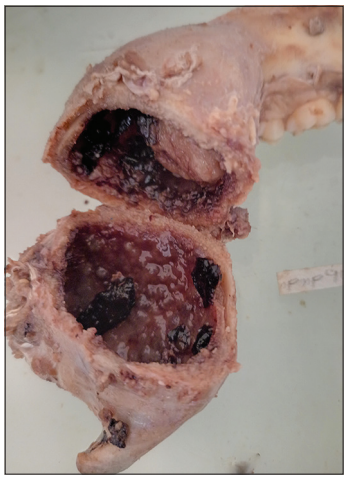

Figure 3: A hemimandibulectomy specimen of ameloblastoma with buccolingual expansion, with cut surface showing a cystic cavity containing gelatinous material and blood clots

Table 5: Type of semi-solid cavity material versus fluid aspirate at incisional biopsy

Cavity content	Straw	Serosanguinous	Dark brown	Negative	Total
Gelatinous material	5	1	4	1	11
Cheesy material	4	0	1	2	7
Mixed gelatinous and cheesy compartments	0	0	1	2	3
Blood clot	1	0	0	0	1
Total	10	1	6	5	22

P=0.7

the prevalence, diagnosis, and management.[15,18,19] Another reason is that ameloblastomas are sometimes detected early and treated before they form large cavities with semi-solid materials.

In this study, patients presented with tumour cavities containing semi-solid materials of which the process of formation is not presently known to the author. Nevertheless, the author suggests that the clear gelatinous materials could represent an immature enamel matrix product of a failed attempt at amelogenesis, while the gray–white cheesy materials could represent immature dentine matrix secretion in a failed attempt at odontogenesis accompanied with infiltration by inflammatory cells and tumour cells to give rise to the cheesy yellow/cream–white consolidated material. Therefore, the ameloblast-like peripheral tumour cells could be responsible for secreting the unmineralised gelatinous material. It is not clear if these semi-solid materials were more fluid-like at body temperature before excision, as opposed to the semi-solid form observed during laboratory cut-up.

The hues of colour exhibited by the gelatinous material were predominantly brownish, with a few appearing greenish and pinkish. The light green colouration of gelatinous material in ameloblastoma cavity was similarly observed by Kondamari *et al*.[14] in their report of ameloblastoma arising in the wall of dentigerous cyst. The cause of the colouration and the relevance are not obvious to the author. This study could not establish if the hues were acquired after the excision and contact with formal saline. The fixative was not expected to permeate gelatinous material and fix probable tumour cells embedded within and may therefore not have contributed to the different hues of colouration. If the fixation had any effect on the gelatinous colouration, the hues would then have been the same in all cases.

The relevance of the semi-solid materials is that they form the substrate for subsequent invasion by the inflammatory cells, vascular tissue, macrophages, and tumour cells. The perforation of the inter-cavity septa or the cortical bone plate of the jaws during tumour extension or surgery could lead to spill of the semi-solid cystic contents to the surrounding tissues. This has been a recognised path of recurrence in ameloblastoma.[11] The range of tumour duration in this study showed that the ability of ameloblastoma to produce spillable contents in primary and recurrent ameloblastoma takes a minimum of 3 months to acquire. Recurrent ameloblastoma was observed twice with gelatinous than cheesy materials though the difference was not statistically significant, but it supports the understanding that the more flowable gelatinous material has a higher potential for spillage and recurrence.

Despite multiple cavities in multicystic ameloblastoma with various contents, needle aspirations of tumour fluid during clinical examination tend to yield one specific tumour fluid type.[8] It has been reported that majority of tumours of ameloblastoma produce straw,[20] dark-brown,[21] and serosanguinous aspirates,[8] while negative yields could result from the needle insertion in cavities containing semi-solid material too thick for the needle gauge.[13] There was no significant association of the semi-solid content with the type of tumour fluid aspirated during clinical examination or incisional biopsy. This would suggest that the development of the semi-solid materials may be independent of the process producing the tumour fluids.

Conclusion

Gelatinous and cheesy cystic contents are possible factors in tumour recurrence since they could potentially spill during surgery or cortical perforation. Negative fluid aspirate of ameloblastoma during clinical examination or biopsy could be due to needle insertion into cavities with semi-solid contents. Further studies are needed to evaluate the potential for tumour invasion of the semi-solid cystic cavity materials in ameloblastoma.

This study is limited by the few number of cases and relied on visual identification of types, nature of semi-solid cystic cavity materials, and hues of the gelatinous materials.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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