Multidisciplinary approach to improving concordance in diagnosis of odontogenic tumors

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Abstract

Background: The diagnosis of benign odontogenic tumors (BOTs) may occasionally be fraught with problems. Diagnosis of BOTs includes joint consideration of clinical features observed, appearance on radiographs and histopathologic slides. **Aim:** The aim of this study therefore is to ascertain the level of concordance between preoperative and postoperative histopathological diagnoses of surgically treated BOT and highlight modalities that improve it. **Materials and Methods:** A retrospective review of all cases with postoperative histopathological reports of BOT seen at the Dental and Maxillofacial Surgery Department of Aminu Kano Teaching Hospital, Kano, Nigeria, from January, 2012 to December, 2013 was done. Demographic information, clinical, radiological (plain radiographs), preoperative incisional biopsy and postoperative excisional biopsy results were collated. The preoperative incisional biopsy and postoperative excisional biopsy results were analyzed for concordance. **Results:** Thirty-three cases of BOT were reviewed (male:female = 1.4:1). Age ranged from 11 to 70 years (mean = 32 ± 18.1 years). An overall concordance of 78.8% was observed between preoperative and postoperative biopsy results. Twenty-eight histology request cards were reviewed and this showed that 14.3% of specimens were <1 cm while complete clinical information was provided in 50% of cases. **Conclusion:** The interaction between the pathologist and other relevant specialties that are required for effective management of BOT may be best nurtured when clinico-pathologic conferences are made a routine practice.

Key words: Benign odontogenic tumor, concordance, multidisciplinary

INTRODUCTION

The diagnosis of benign odontogenic tumors (BOTs) may occasionally be fraught with problems arising from similarities of these tumors when considering their clinical, radiologic or histologic appearance singly.[1] Oro-facial tissue proliferation is often by the multiplicity of structures from odontogenic apparatus, making lesions from derived structures bear close resemblance one to another not just clinically but also radiologically and histologically. The radiological appearance of BOTs with respect to bone density, expansion, borders, locularity, size of lesion and impact on adjacent teeth is seldom specific. Clinical behavior and histological appearance may sometimes be confusing because of a common primordial tissue source which is complex.[2] Proper diagnosis of BOTs includes a joint consideration of clinical features observed, appearance on radiographs and histopathologic slide. It is therefore relevant that a critically harmonized evaluation of all three diagnostic indices be done to arrive at correct diagnoses. An incisional biopsy is generally advocated for lesions larger than 5 cm or where there is a high index of suspicion for malignancy.[1] The rendered diagnosis thus helps in appropriate treatment planning, which is usually a therapeutic excisional surgery.

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Management of BOT with a wrong diagnosis may result in inappropriate treatment (which could be under-treatment or over-treatment) leading to possibility of recurrence, malpractice suits and unjustifiable morbidity, which may warrant reconstruction and rehabilitation. A preoperative biopsy is generally advocated to ensure surgeons have a right working diagnosis which is one of the critical steps

in their management, thus reducing the chances of wrong or inappropriate treatment. However, instances occur where there is discordance between the results of both the diagnostic preoperative and therapeutic postoperative biopsies. The aim of this study therefore is to ascertain the level of concordance between preoperative and postoperative histopathological diagnoses of surgically treated BOT and highlight modalities that improve it.

MATERIALS AND METHODS

A retrospective review of cases with postoperative histopathological reports of BOT at the Dental and Maxillofacial Surgery Department of the Aminu Kano Teaching Hospital, Kano, from January, 2012 to December, 2013 was done. Patients' case folders as well as incisional and final excisional biopsy reports were retrieved. Patients who did not have both biopsy records were excluded. The preoperative incisional biopsies were carried out by maxillofacial surgery residents while the therapeutic excisions were performed by consultant oral and maxillofacial surgeons.

Demographic information, clinical, radiological (plain radiographs), preoperative incisional biopsy and postoperative excisional biopsy results were collated. The preoperative incisional biopsy and postoperative excisional biopsy results were analyzed for concordance. Pathology records were also assessed to evaluate sizes of specimen submitted, clinical information provided on request cards and where indicated, review slides were made for re-assessment. Histopathology request forms were evaluated to determine whether adequate information was provided to assist with pathological diagnosis. To effect this, a point each was awarded for documentation of:

1. Presenting complaint;
2. Findings on clinical examination;
3. Radiographic features and
4. Provisional/tentative diagnosis on the histopathology request forms.

RESULTS

A total of 42 histological diagnoses of BOTs were made during the study period, 9 of which were excluded for not meeting the inclusion criteria. The remaining 33 cases comprised of 19 males and 14 females (ratio 1.4:1). The patients' ages ranged from 11 to 70 years while the average age was 32 ± 18.1 years.

Preoperative and postoperative histopathological diagnoses for the patients are presented in Table 1. The table showed an overall concordance of 78.8%. Twenty-eight histology request cards were available for review [Table 2] and this shows that 14.3% of specimens were <1 cm while the complete clinical information was provided in 50% of cases. Twenty-four (85.7%) of the 28 histopathology specimens were processed completely for histology.

Table 1: Post-operative histologic diagnosis, pre-operative incisional diagnosis and percentage concordance

Final histopathologic diagnosis	N	Number diagnosed at incisional biopsy	Concordance (%)	Previous diagnosis (N)
Ameloblastoma	23	19	82.6	Dentigerous cyst
Odontogenic myxoma	4	2	50	Odontogenic Fibroma
AOT	4	4	100	—
SOT	1	0	0	Acanthomatous Ameloblastoma
Ameloblastic Fibroma	1	1	100	—
Total	33	26	78.8	

N = Number, AOT = Adenomatoid odontogenic tumor, SOT = Squamous odontogenic tumor

Table 2: Characteristics of gross specimen submitted and details of information on accompanying request cards

Characteristics of specimen and request cards	Number	Percentage
Maximal diameter of submitted specimen <1 cm	4	14.3
Maximal diameter of submitted specimen >1 cm	24	85.7
Number with documented radiological features	14	50
Number with complete clinical history	14	50
Number with documented presenting complaint	27	96.4
Number with documented examination findings	24	85.7
Number with stated working diagnosis	26	92.9
Specimens completely processed	24	85.7

DISCUSSION

The overall discordance for preoperative and postoperative histological diagnosis of BOT in this study was 21.2%. This is lower than but comparable with the 30% reported by Guthrie *et al*.[1] who also compared concordance of pre- and post-operative histological diagnosis, as well as frozen section reports. It is however higher than the 9.8% to 16% discordance rates documented by others[2,3] working on similar theme as ours. Even though members of the International Head and Neck Scientific Group[4] have attributed these differences to sampling error in 70% of cases and interpretative errors in the remaining 30%, the import of this write-up however, is to explore specific factors underlying this discrepancy and how to aid their diagnosis.

Adenomatid Odontogenic Tumor and Ameloblastic Fibroma

In our study, a 100% concordance was found for adenomatoid odontogenic tumors (AOT) and ameloblastic fibroma (AF). Even though classical radiologic features of AOT were not documented in submitted request cards, confidence in the diagnosis of this entity on incisional biopsy material was because of adequacy of biopsy submitted as well as characteristic histology. Clinically it is described as the "two-third" tumor because 2/3 occurs in the maxilla with 2/3 being in young females. Two out of three times, it is in association with an unerupted tooth, 2/3 of which are canines. In the radiological diagnosis of this entity, Dare *et al*.[5] highlighted the limitations of panoramic radiographs and have rather recommended the intra-oral periapical view. This allows for easy perception of the radio-opacities characteristic of AOT. These opacities are seen as discrete foci having a flocculent pattern within areas of radiolucency. Histologically, a well-defined capsule encloses whorls and strands of epithelium among which are micro cysts. These may contain eosinophilic material and produce the adenomatoid appearance.

Ameloblastic fibroma is a rare odontogenic tumor seen mostly in the first and second decades of life. It is mostly located in the posterior mandible and has an association with unerupted tooth in 3/4 of cases. Radiologically, they are sclerotic and may be unilocular or multilocular. It also has a distinctive histological morphology composed of anastomosing cords of stellate reticulum cells with peripherally located palisaded columnar epithelium in a highly cellular stroma. AF may easily be mistaken for follicular ameloblastoma.[6] However, the stromal hyper-cellularity with spindle cells seen in AF is not seen in follicular ameloblastoma.[6]

Ameloblastoma versus Dentigerous Cyst

Ameloblastoma is the most common odontogenic tumor accounting for 10% to 30% of all odontogenic tumors. Typical clinical presentation includes solid multi-cystic, unilocular and extra osseous variants while the radiological presentation is usually as unilocular or multilocular radiolucencies. The histological presentation is more diverse with follicular, plexiform, basal, granular, acanthomatous and desmoplastic variants recognized.[7]

As evidenced by our concordance rate of 82.6%, the diagnosis of this lesion is usually straightforward and is based on the finding of stellate reticulum cells surrounded by palisaded ameloblast-like cells with reverse nuclear polarity. These cells either line cystic areas or are seen invading into accompanying stroma in cystic and solid variants in the different histologic patterns. This degree of concordance even though based on tiny incisional biopsies [Table 2] may reflect the generally adequate clinical and radiological information that accompanied submitted samples and possibly, epidemiology. Occasionally, as seen in this study, there is morphologic deviation from the classical pattern hitherto described.

These departures from the classic pattern on histology may underlie reasons for the misdiagnosis of the 4 of the 29 cases of ameloblastoma as dentigerous cysts. It may also be predicated on reasons of similarities in their clinical presentation [Table 3] and unavoidable sampling errors, particularly for unicystic ameloblastoma which, as shown in the study by Ackermann *et al*.,[8] were lined by variable often nondescript epithelium in 42% of their 57 cases.

Metaplastic changes, usually as nonkeratinizing squamous epithelium, is well documented for cystic ameloblastomas.[9,10] This phenomenon may result in misdiagnosis as a dentigerous cyst or other odontogenic cysts. The squamoid appearance may result from compression and attenuation of the epithelium by its fluid content as well as from chronic irritation. This should be borne in mind by the maxillofacial surgeon because of the tendency for late presentation by our patients, as well as iatrogenic insults by nonorthodox practitioners often consulted by these patients. Similarly, if a superficial biopsy is taken, the diagnosis may be missed as deeper and more representative components may not be included in the biopsy.[10]

In cases where the size of the sample is small, as was found for some of our cases, the pathologist has to resort to cutting deeper levels of processed tissue. However, this was not helpful in obviating misdiagnosis in our cases. Similarly, Dunsche *et al*.[11] concluded from their study of 101 specimens, that deeper step section of submitted tissue may be unhelpful in the evaluation for possible missed ameloblastoma. In the same vein, Oliver *et al*.[12] borne out of need to optimize histopathology service, concluded that the submission of a correctly handled representative biopsy *ab-initio* is the only panacea for correct diagnosis. In this vein, useful recommendations for specimen handling summarized in Table 4 and biopsy technique are well documented.[13] It is recommended that the length of the biopsy should be at least 3 times the width, thus ensuring deeper tissues are well represented.[14]

Table 3: Similarities in the presentation of Ameloblastoma and Dentigerous cyst

Features	Dentigerous Cyst	Ameloblastoma
Age	21-30 years	31.7 years ±15.6
Sex (M:F)	1.9:1	1.1:1
Relationship to impacted teeth	+++	++
Location	Mandibular posterior molar	Mandibular posterior molar
Radiology	Soap bubble pattern	Soap bubble pattern

Table 4: Recommendations for processing of tissue biopsies

Biopsy site selection
Remove sufficient tissue
Handle the specimen gently
Fix immediately
Clearly identify specimens and container
Submit a brief and thorough history and radiographs as appropriate
Submit legible and complete paperwork
Forward the specimen to the lab immediately
Always correlate the diagnosis with clinical impression

Odontogenic Myxoma versus Odontogenic Fibroma

Sampling bias may also explain the designation of one of the two odontogenic myxomas (OM) as an odontogenic fibroma (OF). The undifferentiated mesenchymal cells typical of OM have been described to possess the capability for fibroblastic differentiation.[15] Furthermore, clinically, where the radiological report is available; its presentation as a multilocular radiolucency may simulate ameloblastoma or keratocystic odontogenic tumor. Similar sampling bias, resulting in misdiagnosis of OM as OF, was documented by Sivakumar *et al*.[16] in a case report from India. Other features that have been described as characteristic of OM include hyalinization and calcification. Clinically most are young adults and most commonly present posterior mandibular lesions. To the radiologist, they present with multilocular, soap bubble radiolucency and to the pathologists as spindle to stellate cells in myxoid stroma with a variable fibrous tissue.

Squamous Odontogenic Tumor

The only case of squamous odontogenic tumor (SOT) in our series was misdiagnosed as an acanthomatous ameloblastoma. The likelihood of making this erroneous conclusion was also documented by Adebiyi *et al*.[17] in Lagos, Nigeria. In their study, the concordance of provisional diagnosis with final histology was also 0%. The rarity of this diagnostic entity, evidenced by only 5 cases in 20 years in the Lagos study, may underlie its misinterpretation for the more common acanthomatous ameloblastoma.

Its propensity for preferential localization in the posterior mandible is another feature it shares with ameloblastoma, more so that in the past it was considered a variant of ameloblastoma. The tumor is composed of variable sized nests and cores of benign appearing squamous epithelial cells with peripheral flattening or cuboidal cell arrangement in a background of fibrous tissue. Situations like these may, however, be more easily discernible by an oral pathologist who may be more experienced at handling such cases compared to a general pathologist, as was the case in this instance. This underlies the need for an increase in number of specialist oral pathologists in Nigeria.

From the foregoing it is obvious that discordance in clinical and pathological diagnosis may result from:

1. Inadequacy of sample sent for histology;
2. Adequacy of information given to other managing team members;
3. Awareness by the pathologist of not only the subtle morphologic differences in the various maxillofacial lesions but also their clinical and radiological features.

An interesting write-up by Powsner *et al*.[18] entitled "Clinicians are from mars and pathologists are from Venus" perhaps best illustrates the summary of issues raised by our study. The paper underscores the need for all members of the medical ecosystem to work together to facilitate optimized patient management. The clinician should be aware of limitations the pathologist faces when dealing with sub-optimally taken and handled biopsy specimens. He should also not be economical with relevant clinical information that will assist both the radiologist and the pathologist in serving him better. The pathologist should also not mind stepping out of his "niche" to interact with his clinical colleagues. This interaction is best nurtured when clinicopathologic conferences are made a routine practice.[4]

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