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Periradicular burkitt's lymphoma: a report of 2 cases from Nigeria and review of the literature

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Summary
This article reports two cases of periradicular Burkitt's lymphoma from Nigeria, to emphasize the difficulties in differentiating the early lesion from other periradicular lesions with similar clinical and radiological findings Case 1, is a 4-year-old boy who presented with a one-month history of a painless, hard, posterior mandibular swelling (right), which was causing loosening and displacement of deciduous teeth (84 and 85). Histopathological examination of periradicular tissues from extracted tooth (85), confirmed the diagnosis of early periradicular Burkitt's lymphoma. Case 2, is a 6-year-old boy who presented with one-week history of a loose, extruded right mandibular first molar tooth (46) and an exfoliated left mandibular first molar tooth (36). After two weeks of follow-up, the patient developed bilateral mandibular swelling at the molar region, as well as peri-orbital and bilateral pedal oedema. Incisional biopsy of the oral lesion at the region of exfoliated left first mandibular molar (36) was useful for histopathological diagnosis of early Burkitt's lymphoma of the jaw. In the face of limited diagnostic tools such as clinico-radiological assessment, cytology or incisional biopsy for incipient periradicular lesions, a high index of suspicion of Burkitt's lymphoma of the jaw may be helpful in early diagnosis and treatment of a lesion, presenting in a child as periradicular radiolucency or mixed radiolucency and radiopacity, with associated loosening and displacement of teeth.

Keywords: *Periradicular, Burkitt's lymphoma, Clinico-pathologic diagnosis.*

Résumé
Cet article rapporte deux cas de lymphome de Burkitt péri radiculaire au Nigeria pour exprimer les difficultés à différentier les lésions précoces d'autres lésions périradiculaire ayant des résultats cliniques et radiologique semblable. Le premier cas chez un garçon de 4 ans ayant eu un mois d'un enflement du mandibule droit postérieur solide et sans douleur qui causait une perte et un déplacement des premières dents (84 et 85). L'examination histopathologique des tissues périradiculaire extrait des dents (85) confirmait le diagnostic du lymphome de Burkitt périradiculaire. Le deuxième cas d'un garçon de 6 ans avec un semaine d histoire de perte du premier molaire du mandibule droit (46). Après deux semaines de suivi, le patient developait un enflement bilatéral mandibulaire de la région molaire periorbitale et une oedeme pédale bilatérale. La biopsie incisionale de la lésion orale au niveau du premier mandibulaire gauche de la molaire etait utile pour le diagnosie histopathologique précose du lymphome de Burkitt de la joue. Avec les problèmes d'outils de diagnostic limités tels que l'évaluation clinique et radiologique, cytologique ou la biopsie incisionale pour les lésions périradiculaire, un indexe élevée de suspicion du lymphome de Burkitt de la joue peut être utile dans le diagnostic précoce et les soins de la lésion, présente chez un enfant comme radiolucence périradiculaire ou radiolucence mixée et radiopacite avec une perte et déplacement des dents.

Introduction

Lymphomas have been reported as one of the malignant periapical lesions [1,2]. However, carcinomas are the commonest malignancies at the periapical region [3-9]. Whatever the type of periapical malignancy, there is a great cause for concern if there is misdiagnosis. Failure to make early diagnosis of a periradicular malignant lesion may reduce the chances of achieving a cure.

Occasionally, the early stages of malignant lesions like Burkitt's lymphoma can easily be misdiagnosed as a suppurative process or as benign lesion of the jaws [10]. Delayed diagnosis or initial misdiagnosis thus worsens the prognosis of the lesion. Therefore, routine histopathologic examination of periradicular tissues obtained from extracted teeth or during endodontic surgery, is necessary to confirm clinical diagnosis [11-17].

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Early diagnosis of periradicular Burkitt's lymphoma in our environment remains a great challenge, because of the difficulty in differentiating the early lesion from other periradicular lesions with similar clinical and radiological findings. However, the precise diagnostic tools for Burkitt's lymphoma includes: the determination of histological, immunophenotypic and genetic features, before administering appropriate therapy [18].

Most cases of Burkitt's lymphoma in Nigeria present as late stage lesions with extensive jaw swellings and severely displaced teeth [10,19,20]. This article reports 2 cases from Nigeria of early periradicular Burkitt's lymphoma, following routine biopsy of periradicular tissues from extracted teeth at the Dental Centre, University of Benin Teaching Hospital, Benin City, Nigeria. The findings were compared with previous reports.

Case report 1

A 4-year-old boy presented with a one-month history of a hard, painless, posterior right mandibular swelling, which measured 2cm x 3cm, causing loosening and displacement of teeth 84 and 85. Preoperative periapical radiograph showed a mixed radioapaque and radiolucent lesion around the roots of teeth 84 and 85. A clinical diagnosis of Burkitt's lymphoma was made (figure 1). Transalveolar extraction of affected teeth 84 and 85, under local anaesthesia and curettage of the socket was performed. Histopathologic examination of the recovered periradicular tissues confirmed the diagnosis of periradicular Burkitt's lymphoma (figure 2). The patient was referred to the Paediatric Oncology Unit of the hospital for expert management of the lesion. However, follow up of the case at the Unit showed

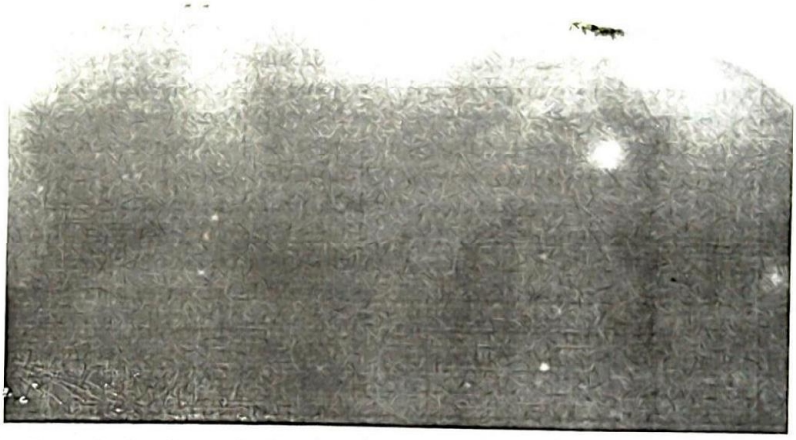

Fig. 1: Periapical radiograph showing periradicular radiolucency involving right deciduous first and second mandibular teeth.

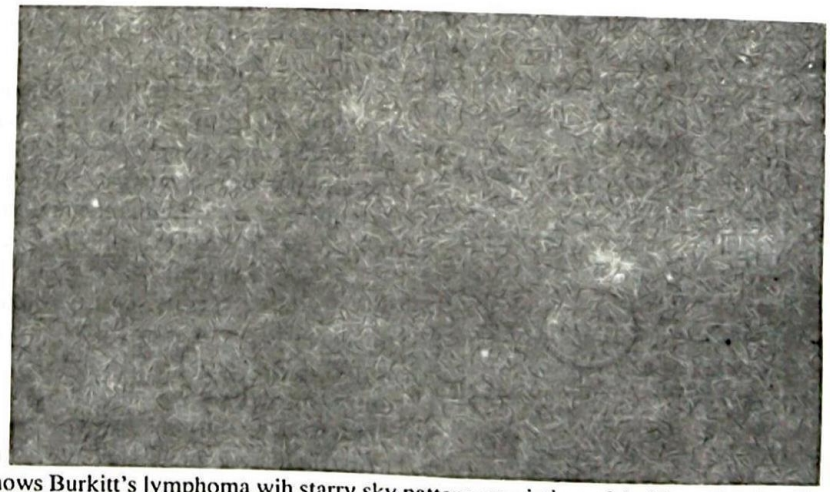

Fig. 2: Section shows Burkitt's lymphoma wih starry sky pattern consisting of darkly stained malignant lymphocytes and pale macrophages (x40, H & E)

that combination chemotherapy of methotrexate, oncovin, procarbazine and prednisolone (MOPP) was administered. The patient responded rapidly within a week of commencing chemotherapy.

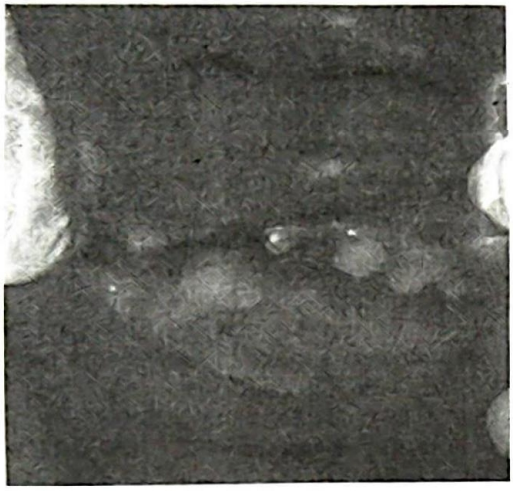

Fig. 3: Intra-oral photograph showing missing left mandibular deciduous second molar, lingually displaced left permanent fist molar.

Fig. 4: Panoramic radiograph showing early Burkitt's lymphoma with missing left mandibular deciduous second premolar, lingually displaced left permanent first molar and extruded right mandibular permanent first molar.

Case report 2

A 6-year old boy presented with one-week history of a loose, extruded right mandibular first molar tooth (46), exfoliation of left mandibular second premolar tooth (35) and lingually displaced first molar (36) [figure 3]. After two clinical review sessions over a 2-week period, to ascertain the nature of the bone resolving lesion using periapical radiographs, the patient developed bilateral mandibular swelling at the molar region, as well as peri-orbital and bilateral pedal oedema. Panoramic radiograph showed periradicular radiolucency with the resorption of the roots of the right mandibular first molar (46) and missing left mandibular second premolar (35) and displaced first molar (36) [figure 4]. Abdominal ultrasound showed liver, spleen and renal masses. An incisional biopsy of the oral lesion at the region of the exfoliated left mandibular second premolar tooth (35) was performed and a diagnosis of early Burkitt's lymphoma of the jaw was made (figure 5). The case was then referred to the Paediatric Oncology Unit of the hospital. A follow up of the case at the Unit showed that the patient died 4 days after commencing the first course of MOPP

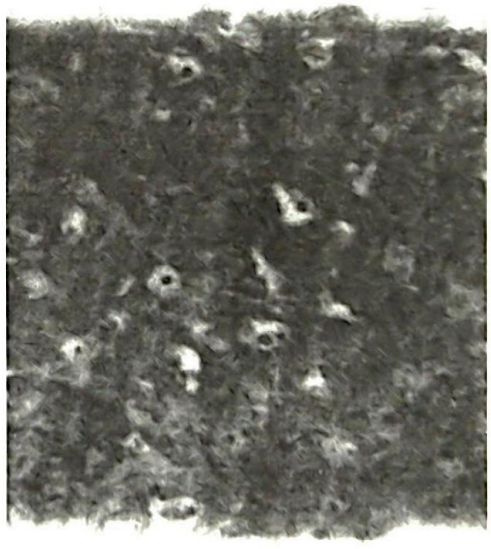

Fig.5: Section shows Burkitt's lymphoma with starry sky pattern consisting of darkly stained malignant lymphocytes and pale macrophages with areas showing engulfed lymphocytes (x40, H & E).

Discussion

There have been some controversies about the need for routine biopsy of periradicular lesions. Clinicians sometimes submit specimens of periradicular tissues for histopathologic examination, only when it is certain that the clinical diagnosis is doubtful, rather than as a routine biopsy to confirm clinical diagnosis. Whereas, some studies in recent times suggest that routine histopathologic evaluation of periradicular tissues

associated with extracted teeth or during endodontic surgery will ensure standard care and better treatment outcome [11-16,21-23]. These cases showed that histological evaluation remains the most reliable means of confirming clinical diagnosis and preventing misdiagnosis, particularly for periradicular malignant lesions. However, very few cases of periradicular lymphoma have been reported [1-2] amongst malignant periradicular lesion, which consist mainly of carcinomas [3-9].

There was difficulty in diagnosing both cases of periradicular Burkitt's lymphoma clinically and radiologically, because some periradicular suppurative or benign lesions have similar features [10,24]. The diagnosis of both patients was confirmed after histopathological evaluation. On the contrary, there may be need for careful histological evaluation, where there is radiological finding of enlarged dental crypts, a feature sometimes seen in early Burkitt's lymphoma [20]. This is to exclude the possibility of a dental cyst, particularly the early stage of dentigerous cyst being misinterpreted as an early feature of Burkitt's lymphoma [25,26].

Early histopathologic diagnosis of periradicular Burkitt's lymphoma was made in the first patient, who was immediately referred for follow up and further treatment. Similar to previous report [18], a remarkable response was observed after a week of administering chemotherapy, which indicates a better prognosis following early suspicion and diagnosis of periradicular Burkitt's lymphoma in this patient. Whereas, the second patient presented initially with a jaw lesion associated with tooth extrusion, displacement and exfoliation, as well as periradicular radiolucency and root resorption. These features are typical findings in early Burkitt's lymphoma [20], but the clinical and radiological features though suggestive of the lesion, were insufficient to make definitive diagnosis. Subsequently, there were signs of abdominal involvement, suggesting that patient's condition was rapidly deteriorating before a confirmatory histopathologic diagnosis was made. Unfortunately, the patient died a few days after commencing the first course of chemotherapy.

Although, there have been difficulties associated with previous attempts at correlation of clinico-radiological features with histopathologic diagnosis of early Burkitt's lymphoma [20], clinical, radiological, cytological and histopathological examinations are the only means of diagnosing Burkitt's lymphoma in our environment. Clinically, Burkitt's lymphoma occurs mostly as swelling(s) in children, with variable sex, age range and site [27-29] between African and non-African types, making it difficult to clinically differentiate the lesion from other periradicular lesions. Whereas, radiological features such as tooth root resorption, loss of lamina dura and enlarged dental crypts may be useful for early diagnosis of Burkitt's lymphoma [20,29].

In conclusion, even with limited diagnostic tools such as clinico-radiological assessment, cytology or incisional biopsy for incipient periradicular lesions, a high index of suspicion of Burkitt's lymphoma of the jaw may be helpful in early diagnosis and treatment of a lesion, presenting in a child as periradicular radiolucency or mixed radiolucency and radiopacity, with associated loosening and displacement of teeth.

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