Case Report

Bimaxillary jaw involvement in pediatric Burkitt lymphoma: A rare case with radiologic–histopathologic–immunohistochemical correlation

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ABSTRACT

Burkitt lymphoma (BL) is an aggressive B-cell non-Hodgkin lymphoma characterized by rapid proliferation and high chemosensitivity. Bilateral jaw involvement in sporadic BL is rare and may pose a diagnostic challenge. Here, we report the case of a 12-year-old male who presented with rapidly progressive bilateral facial swelling following dental trauma and extraction, with onset noted within 2 days post-extraction. Clinical and radiographic findings revealed diffuse gingival enlargement, loss of lamina dura, and displacement of developing teeth. Histopathological examination demonstrated a characteristic "starry-sky" appearance, while immunohistochemistry confirmed CD20+, CD10+, and LCA+ B-cell lineage with a Ki-67 index approaching 100%. The patient was treated with multi-agent COPADM chemotherapy and showed marked regression within 1 month. Early recognition of atypical oral swellings, especially those with rapid onset following dental procedures, and prompt biopsy are critical for timely diagnosis and effective management of this aggressive yet highly curable malignancy.

Key words: Bilateral jaw swelling, Burkitt lymphoma, Immunohistochemistry, Pediatric lymphoma, Post-extraction swelling

Burkitt lymphoma (BL) is a highly aggressive B-cell non-Hodgkin lymphoma characterized by rapidly proliferating monomorphic lymphoid cells with a characteristic "starry-sky" appearance [1]. First described by Dennis Burkitt in 1958, it accounts for approximately 30–40% of childhood lymphomas [1]. BL is classified into three variants: endemic, sporadic, and immunodeficiency-associated [1,2]. While the endemic variant commonly involves the jaws, oral manifestations in the sporadic form are relatively uncommon, accounting for approximately 12–30% of cases [3]. When present, oral lesions may mimic odontogenic infections or other inflammatory conditions, often leading to diagnostic delays.

The present case is reported due to its unusual presentation of bilateral bimaxillary involvement in sporadic BL, which is rarely documented in the literature. Furthermore, the extremely rapid onset of swelling within 2 days following dental extraction adds to its clinical significance, as it closely resembles an aggressive odontogenic infection or hematological disorder, thereby posing a diagnostic challenge. This case highlights the importance of recognizing early oral manifestations and emphasizes the need for prompt biopsy and multidisciplinary evaluation for timely diagnosis and management.

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CASE REPORT

A 12-year-old male presented with bilateral facial swelling. He initially experienced pain in the lower right posterior tooth region but did not seek treatment. Fifteen days later, he sustained facial trauma while playing cricket. Ten days after the injury, mobility of the upper anterior teeth developed, leading to the extraction of tooth 11 at a private clinic. Within 2 days, progressive bilateral facial swelling developed (swelling has been present for 2 days). The patient was referred to the Department of Oral Medicine and Radiology for further evaluation.

On extraoral examination, facial asymmetry with diffuse bilateral swelling extending from the infraorbital rim to above the mandibular border was noted. The nasolabial folds were obliterated. The swellings were soft and tender, with palpable bilateral submandibular lymph nodes (Fig. 1a). Intraoral examination revealed diffuse gingival enlargement involving both jaws, with a lobulated swelling (~2 × 2 cm) on the maxillary gingiva. The left lateral incisor was displaced palatally, while the right canines were displaced buccally. The hard palate

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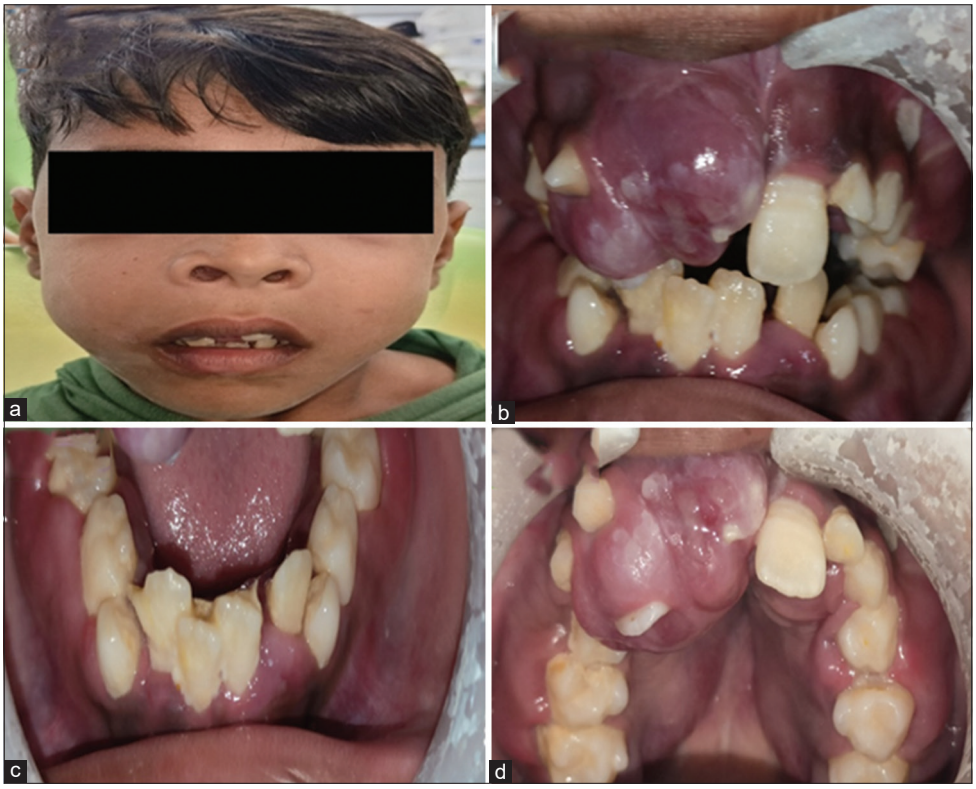

Figure 1: (a) Facial asymmetry with diffuse bilateral swelling. (b-d) Diffuse gingival enlargement involving both maxillary and mandibular arches

showed bilateral swelling near the midline. Multiple teeth exhibited segmental mobility (Fig. 1b-d).

Orthopantomograph revealed loss of cortication of the inferior alveolar canal, generalized bone loss, and loss of lamina dura. Developing mandibular molars showed occlusal displacement with cortical destruction (Fig. 2a). Paranasal sinus view showed haziness of the right maxillary sinus (Fig. 2b). Cone beam computed tomography revealed cortical plate destruction, complete loss of lamina dura, and involvement of developing tooth follicles (Fig. 2c and d), suggesting an aggressive malignancy. Vitality testing showed all teeth to be non-vital except 21. A provisional diagnosis of acute leukemia was considered, and the patient was referred to a cancer hospital.

Differential diagnosis was considered due to the rapid onset and aggressive nature of the bilateral jaw swelling. Langerhans cell histiocytosis (LCH) was considered due to its characteristic presentation with osteolytic lesions and "floating teeth"; however, LCH typically demonstrates well-defined radiolucencies and lacks the diffuse soft-tissue enlargement and rapid progression seen in the present case. Ewing sarcoma was included due to its occurrence in pediatric patients and aggressive bone destruction, but it often presents with periosteal reactions such as "onion-skin" appearance and is commonly associated with systemic symptoms like fever, which were absent here. Osteosarcoma was also considered, particularly due to cortical destruction; however, classical radiographic features such as "sunburst" appearance and mixed radiolucent-radiopaque lesions were not observed. A leukemia/lymphoma spectrum disorder was strongly suspected given the gingival enlargement, tooth mobility, and rapid progression; however, hematological parameters were within near-normal limits, making acute leukemia less likely, while histopathology and immunohistochemistry confirmed BL. Aggressive odontogenic infections (periodontitis, chronic periapical periodontitis, pericoronitis, and osteomyelitis) were also considered in view of the recent dental extraction and acute onset; however, the absence of suppuration, presence of diffuse non-localized swelling, displacement of teeth rather than localized bone destruction, and lack of response typical of infection favored a malignant etiology. Thus, the combination of clinical, radiographic, histopathological, and immunohistochemical findings led to the definitive diagnosis of BL.

An incisional biopsy revealed sheets of uniform lymphoid cells with a "starry-sky" appearance (Figs. 3a-d and 4a-e). Immunohistochemistry showed LCA, CD20, and CD10 positivity, confirming high-grade B-cell non-Hodgkin lymphoma. Cytokeratin and CD3 were negative, with a Ki-67 index near 100%, consistent with BL.

On follow-up after 1 month, hematological and biochemical parameters demonstrated overall stabilization following chemotherapy. Pre-treatment investigations revealed hemoglobin of 12.2 g/dL, total white blood cell (WBC) count of $8.18 \times 10^3/\mu L$, and platelet count of $250 \times 10^3/\mu L$, all within normal limits. Differential count showed neutrophils 51%, lymphocytes 39%, monocytes 8%, and eosinophils 2%. Biochemical parameters included serum creatinine 0.78 mg/dL, blood urea 39.7 mg/dL, total bilirubin 0.91 mg/dL, serum glutamic-oxaloacetic transaminase 38.3 IU/L, serum glutamic pyruvic transaminase 19.8 IU/L, and alkaline phosphatase 114.2 IU/L, which were within physiological range, with a slightly reduced globulin level (2.12 g/dL). Serological

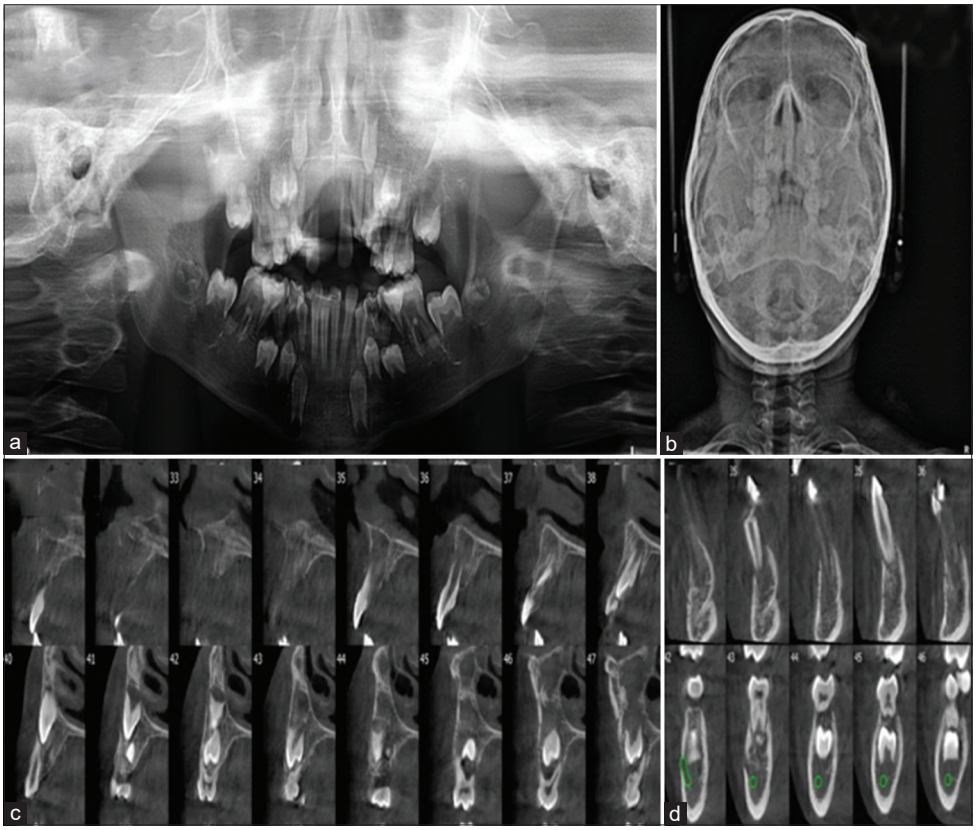

Figure 2: (a) Orthopantomogram showing loss of cortication of the inferior alveolar canal, generalized bone loss, and loss of lamina dura with displacement of developing molars. (b) Paranasal sinus view showing haziness of the right maxillary sinus. (c and d) cone beam computed tomography showing cortical plate destruction, loss of lamina dura, and involvement of developing tooth follicles

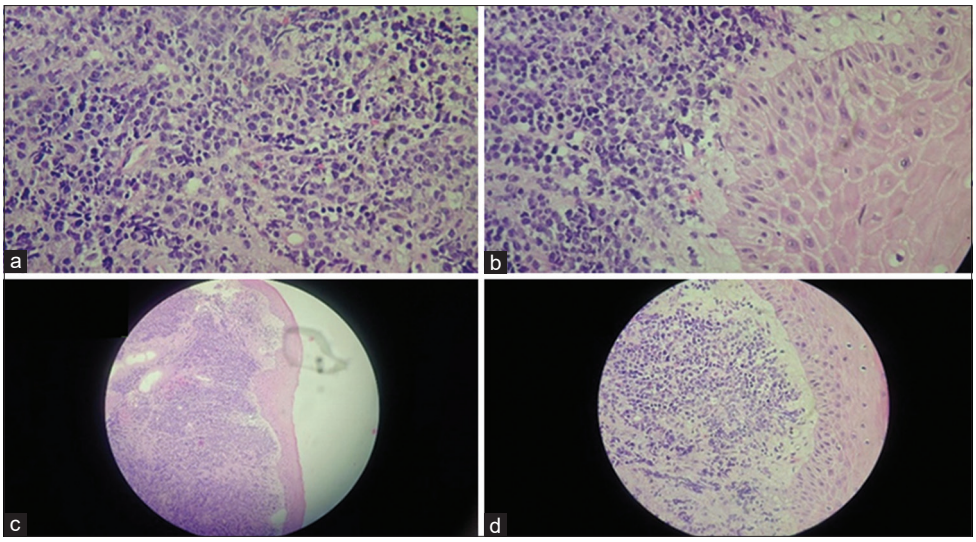

Figure 3: (a-d) Histopathology showing sheets of uniform lymphoid cells with "starry-sky" appearance

tests for human immunodeficiency virus, Hepatitis B surface antigen, and Hepatitis C virus were non-reactive.

Post-treatment investigations showed hemoglobin of 9.8 g/dL, red blood cell count 3.74 × 10^6/μL, and WBC count 6.24 × 10^3/μL, with platelet count maintained at 263 × 10^3/μL. Differential count revealed neutrophilia (70%) with lymphocytes 24%, monocytes 4%, and eosinophils 2%. Biochemical evaluation showed serum creatinine 0.41 mg/dL, sodium 136.2 mmol/L, potassium 4.18 mmol/L, and a markedly elevated LDH of 2329 U/L, consistent with high tumor turnover. Total protein (4.79 g/dL) and globulin (1.60 g/dL) were reduced, likely reflecting treatment-related changes.

The patient was referred for multi-agent COPADM chemotherapy. The patient completed three cycles with significant clinical improvement with regular follow-up for time period of 2 weeks and later for every 1 month and marked reduction in swelling extraoral and intraoral with reduction in pain (Fig. 5a-e).

DISCUSSION

BL is one of the most rapidly proliferating human malignancies, characterized by a doubling time of 24–48 h and a nearly 100% growth fraction [4]. Although it predominantly affects children, involvement

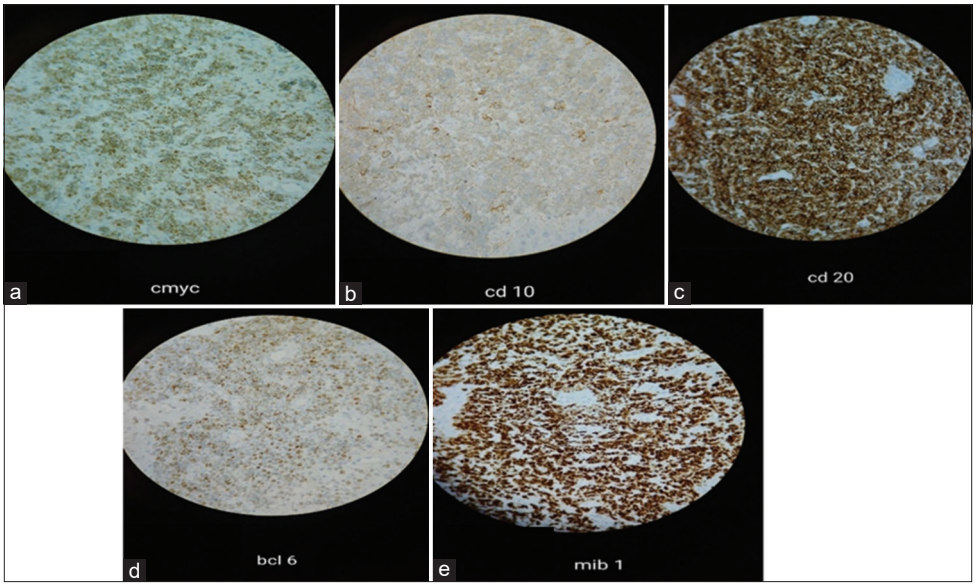

Figure 4: (a-e) Immunohistochemical staining positive for c-Myc, CD10, CD20, BCL6, and Ki-67

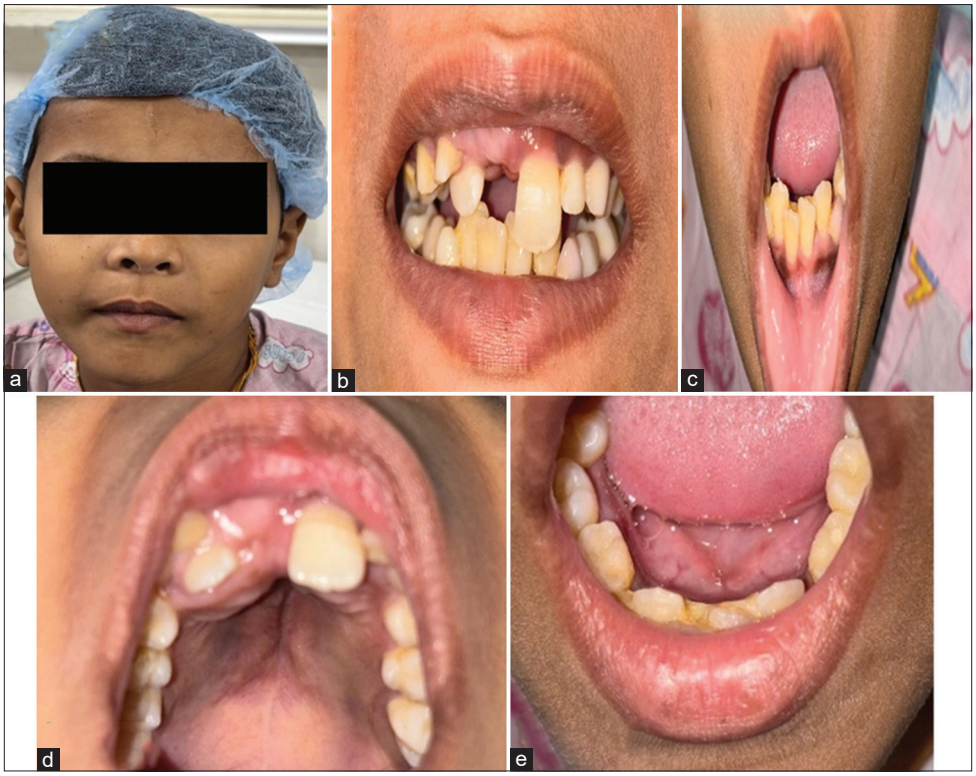

Figure 5: (a-e) Post-treatment images showing marked reduction in extraoral and intraoral swelling. (Follow-up photographs)

of the maxillofacial region in the sporadic variant is relatively uncommon and typically presents unilaterally or with abdominal disease [2,3]. Bilateral involvement of both the maxilla and mandible is exceptionally rare, with only a limited number of cases reported in the literature, including approximately four well-documented cases as highlighted by Alissa *et al*. [5] and the recent systematic review from Silva [2]. While isolated bilateral jaw involvement has been described, simultaneous bimaxillary distribution remains sparsely documented across case reports by Boraz [6], De Freitas Filho *et al*. [7], and broader reviews such as ECancer [8], Rodrigues-Fernandes *et al*. [9], and Ardekian *et al*. [10], thereby reinforcing the diagnostic and academic significance of the present case.

The present case underscores the diagnostic challenges associated with oral BL, which can clinically mimic benign or inflammatory conditions, leading to delays in diagnosis, as reported by Patankar *et al*. [11]. In this patient, the disease followed an unusual sequence: initial tooth pain, followed by trauma, extraction, and rapid post-extraction swelling. This pattern reflects the masked early phase of BL, which often resembles odontogenic infection or traumatic inflammation. Similar misinterpretations have been reported by Balasubramaniam *et al*. [12] and De Freitas Filho *et al*. [7], where early jaw lesions were treated as periapical abscesses or cellulitis prior to definitive diagnosis. Jan *et al*. [13] further emphasized that delayed biopsy following extraction is a major cause of

diagnostic failure in oral BL. Thus, the key clinical issue is not merely the aggressiveness of BL, but its ability to deceptively imitate routine dental conditions in its early stages.

Radiographically, the present case demonstrated loss of lamina dura, ill-defined osteolytic lesions, cortical destruction, sinus haziness, and displacement of developing teeth, consistent with the infiltrative nature of BL. Such findings should alert clinicians to the possibility of an underlying malignancy rather than routine odontogenic pathology. Comparable radiographic features, including loss of cortication and displacement of tooth buds, have been reported by El Gaouzi *et al*. [4] and De Coninck *et al*. [14], as well as in earlier observations by Freitas *et al*. [15]. Notably, mandibular involvement is more frequently described as osteolytic radiolucency, whereas maxillary lesions often extend into the sinus, producing diffuse radiographic changes, as discussed by Patankar *et al*. [11] and Silva *et al*. [2]. The bilateral distribution in the present case likely reflects extensive marrow infiltration, which has been associated with advanced or rapidly progressive disease, as noted by Alissa *et al*. [5] and ECancer [8].

Histopathologically, the characteristic "starry-sky" appearance observed in this case is consistent with established descriptions by El Gaouzi *et al*. [4], Patankar *et al*. [11], and De Freitas Filho *et al*. [7]. Immunohistochemistry confirmed a mature B-cell phenotype (CD20+, CD10+, LCA+, CD3−, cytokeratin−), with a Ki-67 index approaching 100%, reflecting the exceptionally high proliferative activity of BL. A near 100% Ki-67 index is considered a hallmark feature and aids in differentiating BL from other high-grade lymphomas, as emphasized by Balasubramaniam *et al*. [12] and Jan *et al*. [13]. Similar immunoprofiles have been consistently documented, including the diagnostic utility of advanced techniques such as flow cytometry highlighted by Gupta *et al*. [16], reinforcing the reliability of combined histopathological and immunohistochemical evaluation.

Epidemiologically, BL exhibits a male predilection and predominantly affects children between 5 and 15 years of age, as described by De Coninck *et al*. [14] and Freitas *et al*. [15]. The aggressive clinical course observed in this patient aligns with previous reports describing rapid progression within days to weeks, often extending into adjacent structures such as the paranasal sinuses and orbit, as reported by Alissa *et al*. [5]. Pediatric cases more commonly demonstrate jaw involvement and may present with bilateral or multifocal disease, as observed by De Freitas Filho *et al*. [7] and Arboleda *et al*. [17], whereas adult cases tend to be atypical and less frequently involve the jaws, as discussed by El Gaouzi *et al*. [4] and Jan *et al*. [13]. This distinction highlights the importance of maintaining diagnostic vigilance across all age groups.

Despite its aggressive behavior, BL is highly chemosensitive. The favorable response to multi-agent COPADM chemotherapy observed in this patient is consistent with previous studies demonstrating rapid tumor regression following early treatment initiation, as reported by El Gaouzi *et al*. [4], Patankar *et al*. [11], and Balasubramaniam *et al*. [12]. Early diagnosis, accurate staging, and prompt initiation of systemic chemotherapy remain critical determinants of prognosis.

This case further highlights the crucial role of dentists as first-line diagnosticians. Rapidly enlarging oral swellings, unexplained tooth mobility, non-healing extraction sites, and disproportionate radiographic bone loss should be considered key warning signs of malignancy rather than routine inflammatory disease, as emphasized by Patankar *et al*. [11], Balasubramaniam *et al*. [12], and Jan *et al*. [13]. Early radiographic evaluation followed by timely biopsy and immunohistochemical analysis can be life-saving, as delays may permit rapid tumor progression due to its high mitotic activity [4,7,14].

In synthesis, the novelty of the present case lies not only in the bilateral involvement but in the simultaneous bimaxillary distribution with aggressive clinical progression, a pattern rarely documented in existing literature. More importantly, it reinforces a critical clinical lesson: early recognition of atypical oral features, rather than reliance on disease rarity, is essential for timely diagnosis and improved outcomes in BL.

CONCLUSION

This case highlights a rare bimaxillary sporadic BL in a child, emphasizing early diagnosis, radiologic–histopathologic correlation for diagnosis; prompt multi-agent chemotherapy improves prognosis in this aggressive malignancy. Early recognition by dental professionals is crucial for timely intervention and improved prognosis in such aggressive, yet highly treatable, malignancies.

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