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Sporadic Pediatric Burkitt's Lymphoma With Maxillofacial Expression: About Two Cases

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Abstract: *Burkitt's lymphoma (BL) is an aggressive type of mature non-Hodgkin's B-cell lymphoma. We report two rare cases of sporadic pediatric Burkitt's lymphoma (BL) with maxillofacial manifestation. Two boys, 9 and 3 years old, with nasopharyngeal BL initially manifesting as a cheek swelling. They received intensive chemotherapy treatment and obtained a complete response for one and death for the other as a result of his chemotherapy. These cases underscore the importance of maintaining high clinical suspicion when evaluating otolaryngological symptoms and the potential of Burkitt's lymphoma to lead to serious complications that may require emergency procedures. Also, the complications of the treatment that can lead in the worst cases to death.*

Keywords: Sporadic Burkitt's lymphoma, pediatric, maxillofacial

Introduction:

Described by Denis Burkitt in 1957 in Kampala (Uganda) Burkitt's Lymphoma (LB) is a malignant non-Hodgkin's lymphoma (LMNH) representing 35 to 50% of LMNH in children and 2% of LMNH in adults [1]. This is the first human malignancy in which the etiological role of a virus, the Epstein-Barr virus (EBV) has been demonstrated. Thus, opening the way to the search for other viruses implicated in the appearance of cancers such as: HTLV-1, HHV8, HCV and HIV [1, 2].

Case report:

First case

A 09-year-old male from a consanguineous marriage. On September 17, 2019, we were referred to the maxillofacial surgery emergency room for maxillo-mandibular swelling with a starting point under the right angle of the mandibule evolving for 2 months in a painful context and deterioration of general condition but without fever.

On examination, the latter showed more pronounced bilateral maxillo-mandibular swelling on the right associated with left exophthalmos, bilateral intraoral nasal obstruction, mandibular and palatal masses with tooth mobility.

Hospitalized in our department, a maxillofacial CT scan found 3 heterogeneous osteolytic processes, two mandibular centered on the horizontal branches and the largest measuring 48x19x37mm centered on the right maxilla and extending to the two nasal cavities, to the left orbit, upwards. the cerebral parenchyma via the base of the skull, behind the infra-temporal fossa and the nasopharynx with a tendency to obstruct the upper aerodigestive tract as well as numerous jugular carotid adenopathy.

A biopsy performed within 48 hours of admission completed with immunohistochemistry finding strongly positive anti-CD20, CD10 and Ki67 markers, a positive sign of Burkitt's lymphoma. Then the patient was referred to the pediatric oncology department on September 23, 2019 where the pre-therapeutic and extension assessment found no other tumor localization except the presence of anti-EBV antibodies, a sign of an old infection with the Epstein-Barr virus.

Given the rapid progression of the symptoms, the patient presented 3 days after his hospitalization in pediatric oncology with respiratory distress caused by the obstruction of the air ways by the extension of the tumor, which led to the performance of a tracheotomy.

He was then put on chemotherapy treatment according to the GFA LMB 05 protocol – VERSION 2009(3) with good clinical progress. Nevertheless, the patient will eventually die as a result of his chemotherapy 3 months after the start of treatment (fig1).

Second case

A 3-years-old, male, second in a family of 2 children from a non-consanguineous marriage. He had presented for 1 month a gingival swelling gradually increasing in volume in a context of deterioration of general condition with associated fever for which he was initially hospitalized in pediatrics as facial cellulitis. In view of the non-improvement, he was sent to us for maxillofacial surgery emergencies.

On admission to the emergency room, the patient presented with bilateral maxillomandibular swelling with left upper eyelid ptosis. Also, the patient had an oral breathing as a result of the nasal obstruction that he presented.

A CT scan founded, a homogeneous osteolytic process centered on the 2 bilateral nasal cavities measuring 65x45 mm extending laterally to the maxilla and to the infra-temporal fossae, behind the pharynx at the top at the base of the skull and to the orbits as well as lymphadenopathy bilateral jugular carotid.

A biopsy was performed, supplemented by immunohistochemistry, which found strongly positive anti-CD20, CD10 and Ki67 markers in favor of Burkitt's lymphoma, then referred to pediatric oncology for treatment.

An ultrasound performed as part of the extension assessment, found 3 renal nodules, two on the right and one on the left, the largest of which measured 1cm on the long axis.

He was then put under chemotherapy treatment according to the GFA LMB 05 – VERSION 2009 protocol [1]. with good clinical and radiological evolution and complete remission after the end of his cures (fig3).

Discussion:

Burkitt's lymphoma (BL) is an aggressive type of mature non-Hodgkin's B-cell lymphoma that can present in three major variants: endemic, sporadic and associated with immune deficiency. Endemic is associated with the Epstein Barr virus (EBV) and is most prevalent in central Africa, where it classically manifests as enlargement of the jaw or facial bones [1].

The sporadic form is found in non-endemic areas and usually manifests in the abdomen or bone marrow [4]. Nevertheless, this sporadic form involves the head and neck in less than a quarter of cases. Of these cases, the most common site is the cervical lymph nodes, although the mandible, maxilla, tonsils, nasopharynx, or nasal cavity may be involved [2, 3].

There is a third form related to immunosuppression in transplanted subjects or HIV positive [2, 3]. The disease preferentially affects male children [3, 5]. The diagnosis is based on histology and/or cytology and looks for specific markers related to the pathology [2, 3, 5].

The curative treatment is based on polychemotherapy with sometimes very significant side effects requiring care in an equipped environment not always available in low-income countries. [3, 5].

Conclusion:

As Burkitt's lymphoma is a rapidly evolving and potentially serious pathology in its maxillofacial location (invasion, compression of noble structures, etc.). Clinical or biological suspicion must always be appropriate and not ignored, even in non-endemic regions. Biopsies must be performed for lesions with rapid evolution to confirm the diagnosis as soon as possible in order to start treatment early.

List of figures:

Figure 1 : pictures showing the patient 3 days before starting chemotherapy and 1 month after treatment

Figure 2: Picture showing the patient before and after chemotherapy

Conflicts of interest

The authors declare no competing interest.

Authors' contributions

All the authors participated in the treatment of this patient and in the redaction of this article.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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