

## Lymphoma in the base of the tongue: a case report in Amazon

### Linfoma em base de língua: relato de caso na Amazônia

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#### ABSTRACT

Lymphoma is the third most incident malignant neoplasm in the world and accounts for 3 to 5% of malignant tumors. The lymph nodal location is very characteristic of HL, being rare outside this site. This study presents the case report of a patient attending a referral hospital in the Amazon region that was diagnosed with lymphoma on the basis of the tongue. The patient had undergone treatment with six cycles of chemotherapy followed by radiotherapy, so far with lesion regression and in outpatient follow-up.

**Keywords:** Cancer; Medicine; Hematology; Oncology.

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#### RESUMO

Linfoma é a terceira neoplasia maligna mais incidente no mundo e corresponde a 3 a 5% dos tumores malignos. A localização linfonodal é muito característica do LH, sendo rara fora deste local. Este estudo apresenta o relato de caso de um paciente atendido em um hospital de referência na região amazônica que foi diagnosticado com linfoma com base na língua. A paciente havia sido submetida a tratamento com seis ciclos de quimioterapia seguida de radioterapia, até o momento com regressão da lesão e em acompanhamento ambulatorial.

**Palavras-chave:** Câncer; Medicina; Hematologia; Oncologia.

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## INTRODUCTION

Lymphoma is the third most incident malignant neoplasm in the world and accounts for 3 to 5% of malignant tumors. Lymphomas are neoplasms of B or T lymphocytic cells that can be classified as Hodgkin's Lymphoma (HL) and non-Hodgkin's Lymphoma (NHL). In Brazil, an estimated 10,000 new cases of NHL and 2,500 of HL are present each year, with a higher prevalence in men for both types (BRASIL, 2018).

The lymph nodal location is very characteristic of HL, being rare outside this site. Meanwhile, extra nodal is common in NHL, occurring in up to 30% of cases (AMERICAN CANCER SOCIETY, 2019). The most common extra nodal sites are the gastrointestinal tract, the skin, the subcutaneous tissue, and the oral cavity. In the mouth, lymphomas can originate from the lymph node of the Waldeyer ring or may be of non-lymph node tissue, and tongue involvement is very rare, accounting for only 2% of lymphomas in the oral cavity (BECHIR et al., 2016).

In its histology, HL is characterized by the presence of Reed-Sternberg cells, which consist of multilobulated lymphoid cells with eosinophilic nucleus enveloped in a characteristic stromal and cellular context, whereas in NHL the monoclonal expansion of T or B lymphocytes is malignant.

NHL's clinic is diverse, presenting general symptoms, such as fatigue, weight loss, pruritus, night sweats, fever, in addition to bone pain and respiratory symptoms. At the physical examination, there is presence of non-painful adenomegaly in the neck, supraclavicular and axillary chains, and formation of nodal or extra nodal mass (COLLEONI et al., 2009).

Therefore, lymphomas are common malignancies that originate in cells of the immune system and can affect lymph node and extra nodal structures. However, its occurrence in the tongue (especially in its base) is an uncommon finding in oncological practice. Therefore, the present study aims to report a case of B-cell NHL on the base of the tongue that occurred at the oncology service of a tertiary hospital in the lower Amazon.

## METHODS

This is a quantitative, descriptive, and cross-sectional type study, carried out in the county of Santarém, located in the west of Pará State. Data were collected at the Hospital Regional do Baixo Amazonas - Dr. Waldemar Penna (HRBA). The study was performed over the medical records of a 30-year-old male patient who developed non-Hodgkin B lymphoma on the left base of the tongue, diagnosed and treated at the HRBA, and registered in April 2017 in the oncology department of the hospital. The research complied with the ethical precepts of Resolution 466/12 of the National Health Council. The project was approved by the Comitê de Ética em Pesquisa Envolvendo Seres Humanos (CEP UEPA/STM) under protocol number 3.128.669, in the date of 01 of February of 2019.

## CASE REPORT

C.S.C., male patient, 38 years old, brown, rural worker, native and derived from the Comunidade do Bananal, located in the county of Almeirim, state of Pará.

He opened the case with a complaint of “fangs” or “stitches” on the left wall of the pharynx in April 2017, five months before diagnosis, which the patient associated with the presence of a fishbone. As the symptom remained for about a month and evolved with the appearance of ulcerated lesion in the posterior region of the left base of the tongue, he sought prompt care in the city of Almeirim, where he was medicated with antibiotic therapy to treat possible pharyngotonsillitis. One week later, in a clinical reassessment, the physician requested the transfer to the Hospital Municipal de Santarém for otorhinolaryngological evaluation, due to the poor improvement of the condition and the non-characteristic presentation of the lesion. By this same period, pre-auricular adenomegaly is developed.

Two months after the onset of symptoms, the patient arrives at the Hospital Municipal de Santarém, where he was screened and hospitalized. On the same day, he has an appointment with the otolaryngologist, who requests an opinion from the Head and Neck Surgery service. In the latter, biopsied material was collected, which showed the following result: “invasive and ulcerated undifferentiated neoplasia”. For the histological definition of the lesion, an immunohistochemical examination was requested, which showed a positive result for high-grade B-cell lymphoma. Therefore, the patient

was diagnosed with non-Hodgkin B lymphoma on the left base of the tongue and referred to the hematology clinic of the Hospital Regional do Baixo Amazonas.

Regarding the physical examination at the admission to the hematology clinic, the patient was in good general condition, lucid, normocorated, hydrated, afebrile, atypical facies and with normal gait and posture. Oroscopy showed an impaired lesion in the lower left region of the tongue, in a regression of the ulcerative process reported by the patient. There was presence of single adenopathy in the left pre-auricular region. Examination of cardiac and respiratory systems was normal. Examination of the abdomen and limbs revealed no abnormalities.

A hemogram was performed with evidence of red cells count of  $5.34 \text{ mil/mm}^3$ ; hemoglobin of  $15 \text{ g/dl}$ ; hematocrit of  $45.6\%$ , normal hematimetric indexes and normal RDW,  $12,000/\text{mm}^3$  leukocytes,  $69\%$  ( $8,280$ ) of segmented,  $2\%$  ( $240$ ) of eosinophils,  $22\%$  ( $2,640$ ) of typical lymphocytes and  $7\%$  ( $840$ ) of monocytes; the platelet count was  $466,000/\text{mm}^3$ . Prothrombin time, activated partial thromboplastin time, and fasting glycemia were normal. Computed tomography of the face and neck showed an irregularly defined expansive process with a heterogeneous enhancement on the contrasting phase, located at the base of the tongue on the left, with no cleavage planes with the mucosal pharyngeal space and extending to the vallecula on the left and the presence of enlarged lymph node in the left jugulo-carotid chain, level IIA, in close contact, but maintaining a cleavage plane with the left parotid gland.

The patient was then approached by Head and Neck Surgery to perform a biopsy, whose material underwent pathological and immunohistochemical evaluation. The pathological evaluation showed evidence of Invasive and Ulcerative Neoplasia, and the immunohistochemical evaluation showed proliferation of large and atypical lymphoid cells with immunophenotype B (CD20 +) and high proliferation index ( $> 90\%$ ), which is consistent with High Grade B Cell Lymphoma CD20 +.

The patient was then treated with a 6-cycle chemotherapy regimen of Rituximab, Clotrimazole, Vincristine and Doxorubicin (R-CHOP) every 21 days. At the end of this therapeutic phase, radiotherapy was initiated with a total dose of  $40 \text{ GY}$  in 20 fractions.

When this stage was reached, a new clinical evaluation revealed regression of the lesions, maintaining only loss of taste. Radiologically, only an irregular lesion of probable

scarring character on the left side of the tongue base was noted. Therefore, the treatment was carried with outpatient follow-up for observation and control.

## DISCUSSION

The suspicion of lymphoma is not usually the first hypothesis suggested in lesions on the base of tongue due to the rarity of the cases. In addition to physical examination findings, CT and MRI can guide the reasoning for this clinical entity and for the extent of the affection when demonstrating an expansive process, usually of poorly defined contours. Therefore, histopathological and immunohistochemical studies should be carried out (MIREA et al., 2017).

Among extranodal non-Hodgkin's lymphomas, head and neck occupy second place in incidence, right behind the gastrointestinal tract ones. In the head and neck region, Waldeyer's Ring lymphomas are the most frequent, however, the primary site at the base of the tongue is significantly rare amongst them, with its incidence varying from 1% (TEH; JAYALAKSHMI; CHONG, 2014) to 9% (SAUL; KAPADIA, 1985) in the literature. Harabuchi et al. (1997) associated this topography with a worse prognosis in a study with 71 patients diagnosed with WR lymphoma.

Although the reported patient was diagnosed at age 38, tongue-base lymphoma predominantly affects older men. There are no specific risk factors consolidated in the literature for its development, but other authors discussed the involvement of *H. pylori* and cytomegalovirus (in HIV infected patients) in the pathogenesis of the disease, given the established correlation with lymphomas in other primary sites.

Among the types described, Diffuse B-cell Lymphoma is the most frequent (BECHIR et al., 2016), but marginal B-cell lymphomas (IFTIKHAR; SIDDIQUI; MINHAS, 2016), Burkitt's lymphoma, T cell lymphoma (CLARKE et al., 2019) and even Hodgkin's lymphoma (MAKA; SUBRAMANIAN; KILARA, 2014) have been described.

There are no clinical manifestations described in the literature that may be considered characteristics of the presence of lymphoma in the oral cavity; the symptoms most commonly present are local swelling, pain or discomfort and the presence of ulcerated lesion (SINGH et al., 2014). These were, in fact, the symptoms reported by our patient at the time of first medical visits. Many patients associate the symptoms with a sensation of foreign body presence in the oropharynx, which makes this description

frequent in the reports described in the literature (SIMSEK et al., 2016; IFTIKHAR; SIDDIQUI; MINHAS, 2016). The patient of this report initially believed the clinical features were associated with the presence of fishbone in the throat.

Another important point to consider is the presence of lymphadenopathy in clinical presentation, which is not regularly described in the literature. In the reported case, the patient presented with left preauricular lymphadenopathy, but in the reports described by Cerroni and Goteri (2003), and Singh et al. (2014), there was no presence of palpable lymph nodes, even in cases with over one year of symptoms. In the descriptions in which lymphadenopathy was present, there was a considerable variation in the affected chain, although all occurred in head and neck chains (LIM et al., 2012; PINNIX, 2016; SIMSEK et al., 2016).

The aspect of the lesion is also a point that draws attention. The reported patient presented with ulcerated lesion at the site of the neoplasia, and indeed this was the most frequent aspect in the descriptions on studies, exemplified by the reports of Pinnix (2016). The presence of only edema or nodulation / mass is also described in the literature (SIMSEK et al., 2016).

The small number of cases of this type of presentation of lymphoma makes it difficult to understand the pathophysiology and its response to treatment (LIM et al., 2012). Thus, there is still no consensus regarding its therapeutic approach, which may consist of chemotherapy, isolated radiotherapy, chemotherapy followed by radiotherapy and even isolated surgical excision, and these procedures are suggested according to the characteristics of the tumor and the patient (SONG et al., 2014).

In the reported patient, the regimen chosen was rituximab, cliclofosfamide, vincristine, doxorubicin and prednisone (R-CHOP) for 6 cycles. This chemotherapy regimen is the first indication of the American Cancer Society for Diffuse large B-cell lymphoma of stage III or IV (AMERICAN CANCER SOCIETY, 2019). After chemotherapy, radiotherapy was started, as indicated in several studies, which showed a better combination of R-CHOP plus radiotherapy for extranodal lymphomas, having a positive impact on remission and tumor recurrence and on patient survival (PINNIX, 2016).

In the last decade, slightly more than 10 cases of tongue-base lymphoma have been reported. These patients were treated with R-CHOP regimen followed by radiotherapy with complete remission, no complications, and no relapses during months

of outpatient follow-up (LIM et al., 2012; SIMSEK et al., 2016), a third case was treated with a chemotherapy regimen with epirubicine, also with a good therapeutic response (BECHIR et al., 2016).

Two other cases made use of the CHOP scheme followed by radiotherapy, but without rituximab, obtaining total remission of the lesion (LEE; LEE, 2014; HMIDI et al., 2012); however, there was recurrence in the orbit after 3 months from the end of the treatment, and the patient died 17 months later (LEE; LEE, 2014).

Isolated CHOP therapy plus intrathecal methotrexate was described as a choice in one article. With this approach, the patient also had complete remission of the lesion and without recurrence at around 17 months of follow-up (MANOLOPOULOS et al., 2003).

Isolated radiotherapy was used in two cases-reports, and both patients showed remission of the lesion without relapse. In one of them the patient developed xerostomia, which regressed within 2 months after treatment (SONG et al., 2014; JOVANOVIĆ, 2008).

Surgical resection was the chosen treatment in two other cases. In the first, the excision of the lesion was complete without loss of organ function and without recurrence during patient follow-up (SAKABE et al., 2003). In the second case, surgery was performed along with CHOP chemotherapy with total remission of neoplasia and no recurrence in 10 months (IFTIKHAR; SIDDIQUI; MINHAS, 2016). Three articles did not inform the methodology of treatment of the reported cases (WALID, 2009; CLARKE et al., 2019).

## CONCLUSION

It is concluded that there are still many difficulties in identifying and approaching not only tongue-lymphoma but also malignant and malignant lesions of the tongue in general. In the specific case of the lesion addressed by this study, it is observed that the lack of a database on the disease makes it difficult to establish a specific treatment line. Despite this, the treatment lines addressed in the reported cases, including this one, which include cycles of chemotherapy followed by radiotherapy, have proved to be efficient as initial therapy in patients with this diagnosis.

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