Transoral robotic surgery for chronic lymphocytic leukaemia in the lingual tonsils

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Transoral robotic surgery for chronic lymphocytic leukaemia in the lingual tonsils: A case report

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1. Introduction

Chronic lymphocytic leukaemia (CLL) is the most common leukaemia in the Western world, primarily affecting elderly men [1] and often presenting as asymptomatic lymphocytosis. Additionally, anaemia and/or thrombocytopenia may result from bone marrow invasion. CLL shares features with non-Hodgkin lymphoma, including lymphadenopathy, fatigue, fever, weight loss and night sweats, along with potential splenomegaly and hepatomegaly. Extranodal involvement, while less frequent, can occur in sites such as the skin and central nervous [2].

Limited research exists on extranodal sites in the head and neck region, with involvement of the lingual tonsils (LT) being very rare. In this case report, we describe the presentation, treatment, and clinical outcome of a patient who presented with extranodal CLL of the palatine and LT.

2. Case presentation

In January 2018, a 52-year-old male presented at our institution's internal medicine department with a neck mass and dysphagia lasting 6 weeks. He had no fever, weight loss or odynophagia, and had quit smoking 18 years ago and had no history of excessive alcohol intake. Medical history was unremarkable without medication use or surgical interventions. No family history of CLL was reported.

Clinical examination showed massive enlargement of the palatine tonsils, multiple enlarged cervical lymph nodes, one enlarged axillary lymph node and multiple slightly enlarged inguinal lymph nodes.

Laboratory tests showed lymphocytosis with a leukocytes count of 19.6×10^9/L, a lymphocytes count of 13.4×10^9/L with normal haemoglobin and thrombocytes count. Serology was negative for Epstein-Barr virus, human immunodeficiency virus, toxoplasmosis and showed a previous cytomegalovirus infection.

A CT-scan of the neck thorax and abdomen illustrated enlarged LT, multiple smaller cervical, axillar and para-iliac lymph nodes as well as limited splenomegaly Fig. 1. CLL was suspected and later confirmed by immunophenotypic analysis by flow cytometry. The patient was staged with RAI II, Binet B CLL, with no indication for systemic treatment. Upon referral to the otorhinolaryngology's, nearly kissing palatine tonsils and relatively large LT were observed. An extranodal localization of CLL in the palatine tonsils was suspected. Treatment consisted of two options, tonsillectomy or localized radiotherapy. Following comprehensive consultations with the patient, the haematologist, otorhinolaryngologist and radiotherapist, palatine tonsillectomy was chosen and confirmed colonization of CLL in the lymphoid tissue of the palatine tonsils. Follow up was uneventful, with CLL monitoring every 3 months.

Almost one year after tonsillectomy, the patient presented with dysphagia, globus, dyspnoea and lymphadenopathy. After initial antibiotics, symptoms improved but recurred weeks later.

Laboratory tests showed a leukocytes count of 26.6×10^9/L, a lymphocytes count of 19.7×10^9/L with normal haemoglobin and thrombocytes count. A CT-scan showed a progressive enlargement of the base of the tongue (BOT), while laryngoscopy demonstrated enlarged LT (Friedman grade 4). Biopsy confirmed a new localization of CLL. Treatment options included chemotherapy, radiotherapy or surgery.

Transoral robotic surgery (TORS) of the LT was performed by using the DaVinci® surgical system (Intuitive Surgical, Inc. – Sunnyvale, CA, USA), following the standard robotic protocol by O'Malley et al. [3]. A 5 mm monopolar cautery with spatula tip and a 5 mm Maryland forceps were used with a 30°upwards high magnification, three dimensional endoscope. The procedure proceeded without complications, resulting in the removal of 29 cc tonsillar tissue with histopathological proven CLL involvement.

2.1. Clinical outcome

Following resection of LT the patient experienced temporary throat pain, dysphagia, globus and hypogeusia which resolved within weeks to months post-surgery.

After 4 years of follow-up, there is no clinical sign of recurrence in the BOT area. Laryngoscopy showed no signs of residual LT tissue (Friedman grade 0). Blood counts remained stable without systemic therapy. Prior to surgery, the patient had an overall MD Anderson Dysphagia Inventory (MDADI) score of 64 (range 20–100) and an European Organization for Research and Treatment of Cancer Quality of Life Questionnaire (EORTC QLQ-C30) Global Health status of 58.3 (range 0–100). After TORS patient scored 93 and 83.3, respectively. To this date, no systemic or other localized treatment has been necessary.

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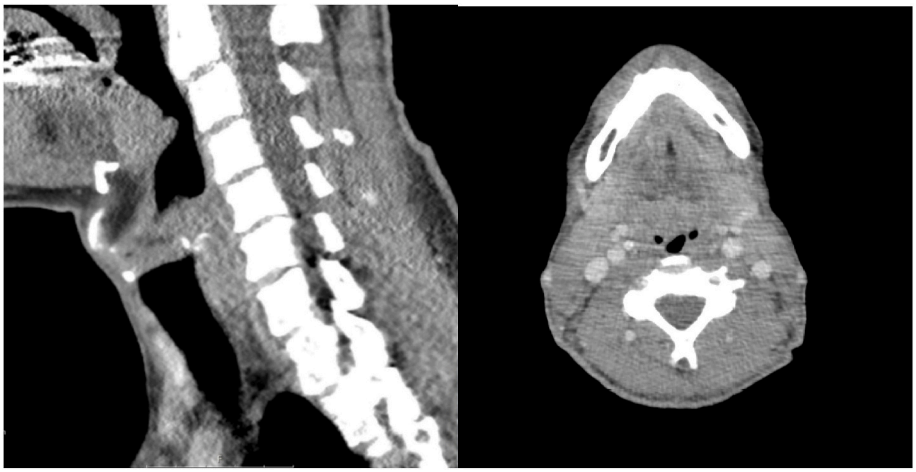

Fig. 1. CT-scan (left sagittal view, right axial view) of patient's neck with enlargement of the lingual tonsils.

3. Discussion

To our knowledge, only 2 reported cases of CLL in the LT exist, with patients undergoing treatment via chemotherapy or radiotherapy. One case described a 58-year-old female with CLL exhibiting an extranodal manifestation in the palatine tonsils [4]. Following tonsillectomy, she experienced recurrent enlargement of LT, necessitating chemotherapy. Despite initial treatment, the LT enlarged again, leading to subsequent radiotherapy, complicated by significant mucositis. Throughout the one-year follow-up, the patient received ongoing chemotherapy due to progressive lymphadenopathy, splenomegaly, and thrombocytopenia.

In another case, a 62-year-old woman with CLL presented with an extranodal manifestation in the BOT [5]. The patient received chemotherapy, resulting in complete response with no recurrence observed four years post-treatment.

Treatment of extranodal manifestations of lymphomas typically includes chemotherapy, radiotherapy, or concurrent chemoradiation. While aggressive lymphomas require systemic therapy combined with radiotherapy, indolent localized lymphomas may be treated with radiotherapy alone. However, lymphomas encompass a diverse range of neoplasms with varying morphology, clinical course and outcome. CLL, known for its indolent nature compared to other lymphomas, may necessitate unique therapeutic strategies in extranodal locations. Because of its rarity, there is a lack of research on optimal treatment for CLL extranodal manifestations in the head and neck.

In our case, the LT were surgically removed using a transoral robotic approach. Initially presented in early 2018, the only available surgical option was classical tonsillectomy. Following multidisciplinary consultation, the decision was made to perform tonsillectomy to mitigate CLL activity and postpone systemic therapy initiation. TORS has become an important treatment modality for malignant oropharynx tumours, benign tumours of the BOT and Obstructive Sleep Apnea (OSA). In our clinic TORS was introduced in the fall of 2018, allowing us to offer this approach to the patient with CLL affecting the LT.

In conclusion, extranodal CLL in the LT is very rare. While extranodal lymphomas are generally treated with chemoradiation and/or radiotherapy, our case demonstrates the successful use of TORS for CLL extranodal manifestations. This approach effectively postponed the indication of systemic therapy, highlighting the potential of TORS as a therapeutic modality for CLL patients in specific extranodal manifestations in the head and neck area.

Disclosure statement

No conflicts of interest.

Financial disclosure

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Ethical statement

The subject described in the manuscript "Transoral robotic surgery for chronic lymphocytic leukaemia in the lingual tonsils: a case report." gave their informed consent for use and publication of information and has signed a standard release form.

CRediT authorship contribution statement

Lisa W. Lekanne dit Deprez: Conceptualization, Formal analysis, Visualization, Writing – original draft. **Alexandra G.L. Toppenberg:** Conceptualization, Data curation, Supervision, Writing – review & editing. **Wouter L. Lodder:** Methodology, Project administration, Supervision, Validation. **Robert E. Plaat:** Investigation, Resources, Supervision. **Bas Franken:** Methodology, Project administration, Resources, Supervision. **Leonora Q. Schwandt:** Conceptualization, Data curation, Investigation, Supervision, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

[1] Sant M, Allemani C, Tereanu C, De Angelis R, Capocaccia R, Visser O, et al. Incidence of hematologic malignancies in Europe by morphologic subtype: results of the HAEMACARE project. Blood 2010;116(19):3724–34.
[2] Ratterman M, Kruczek K, Sulo S, Shanafelt TD, Kay NE, Nabhan C. Extramedullary chronic lymphocytic leukemia: systematic analysis of cases reported between 1975 and 2012. Leuk Res 2014;38(3):299–303.
[3] O'Malley Jr BW, Weinstein GS, Snyder W, Hockstein NG. Transoral robotic surgery (TORS) for base of tongue neoplasms. Laryngoscope 2006;116(8):1465–72.
[4] Dean M, Cunningham I, Dao L, Cole I, Mulliga S. Chronic lymphocytic leukaemia with upper airway obstruction. Leuk Lymphoma 1996;20(5–6):505–7.
[5] Bersabe AR, Romain JT, Ezzell EE, Renshaw JS. Chronic lymphocytic leukemia as an unusual cause of rapid airway compromise. Case reports in oncological medicine 2017;2017.

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