Korean Journal of Oral and Maxillofacial Pathology 2018;42(4):95-98
ISSN:1225-1577(Print); 2384-0900(Online)
Available online at http://journal.kaomp.org
https://doi.org/10.17779/KAOMP.2018.42.4.002

조혈세포이식 후 재발한 백혈병 환자의 혀 종물

최성환[1], 박슬마로[1], 김현실[2,3], 조은애산드라[2,3]*

[1]연세대학교 치과대학 구강악안면외과학교실
[2]연세대학교 치과대학 구강병리학교실
[3]연세대학교 치과대학 구강종양연구소

〈Abstract〉

Tongue Mass Arising after Hematopoietic Cell Transplantation and Leukemia Relapse

*Sung-Hwan Choi[1], Slmaro Park[1], Hyun Sil Kim[2,3], Eunae Sandra Cho[2,3]**

[1]Department of Oral and Maxillofacial Surgery College of Dentistry, Yonsei University, Seoul, Korea
[2]Department of Oral Pathology College of Dentistry, Yonsei University, Seoul, Korea
[3]Oral Cancer Research Institute College of Dentistry, Yonsei University, Seoul, Korea

Oral examination in a patient with a history of acute lymphoblastic leukemia (ALL) and allogeneic hematopoietic cell transplantation (HCT) needs considerations of leukemia relapse and graft-versus-host disease (GVHD). Oral manifestations may contribute to early detection of relapse or systemic complications making accurate oral examination and diagnosis significant. We report a case of a large tumor like mass arising in a patient with a history of ALL and HCT. The patient had been diagnosed with ALL relapse and was being treated with chemotherapy, and furthermore was suspected of GVHD development.

Key words: Precursor cell lymphoblastic leukemia-lymphoma; HCT, GVHD, Mucocele

Ⅰ. INTRODUCTION

Acute lymphoblastic leukemia (ALL) is a hematopoietic malignancy of lymphoid progenitor cells[1]. Malignant lymphoid cells proliferation accumulated the bone marrow, peripheral blood and extramedullary regions with a rapid progression, including the oral cavity. ALL mainly arises in children and is the most threatful type of cancer under the age 20[1,2]. Advances in multiagent chemotherapy has increased the survival rate up to 90% in children, with less achievements of 30-40% in adult ALL[2]. Patients, especially those with disease relapse, are considered for allogeneic hematopoietic cell transplantation (HCT). Acute and chronic graft-versus-host disease (GVHD) are representative complications that may follow HCT[3]. Patients with a medical history of ALL are possible of experiencing various treatment levels of chemotherapy or HCT and are at risk for leukemia relapse which gives a wide range of considerations for dentists during oral examination. In this report, we discussed a case of a large pedunculated tongue mass in a patient with history of ALL and HCT.

* Correspondence: Eunae Sandra Cho, Dept. of Oral Pathology, Oral Cancer Research Institute, College of Dentistry, Yonsei University
Tel: +82-2-2228-3039, Fax : +82-2-392-2959
E-mail: sandra@yuhs.ac
ORCID: 0000-0002-0820-3019
Received: Jun. 23. 2018; Revised: Jul. 13. 2018; Accepted: Aug. 17. 2018

II. CASE REPORT

A 20-year-old male was referred from the department of Internal Medicine to the department of Oral and Maxillofacial Surgery (OMFS) for an exophytic soft tissue mass on the right lateral surface of tongue. The patient had been diagnosed with B-cell ALL and had gone through allogeneic HCT 17 months before initial visit at OMFS. Three months ago, relapse of ALL was discovered with suspicions of chronic GVHD in the skin and oral cavity, so chemotherapy was being treated. The patient had been suffering from dysphagia and nausea. There were neither any history of pain, spontaneous bleeding or trauma to the tongue including tongue biting. An exophytic soft tissue mass on the right lateral side of tongue was seen by oral examination. It was a pedunculated, multinodular red mass with an ulcerated surface. The size of the mass was 1.5×1.0×0.8cm (Fig. 1). The mass was excised for pathological diagnosis. Other than the tongue mass, several vesicles were observed on the soft palate and skin nodules were observed on the posterior neck and right subareolar region. Clinically, the vesicles of palate were thought to be superficial mucoceles while leukemic involvement was suspected at the skin and subareolar region.

On pathological examination of the excised specimen, a sparse central region with a fibrous region at the periphery was seen under low microscopic magnification (Fig 2A). Ulceration was surrounding the whole specimen. Higher magnifications of the sparse region revealed ingrowth of fibroblasts, blood vessels and normal inflammatory cells infiltrates in a mucinous background (Fig. 2B). The main composition of cells were foamy macrophages (Fig. 2C). The fibrous region was composed of granulation tissue and fibrosis. The diagnosis was organizing mucocele, extravasation type. There were no evidences of ALL relapse within the specimen. Recurrence of the mucocele at the tongue was not observed during a 1-year follow up.

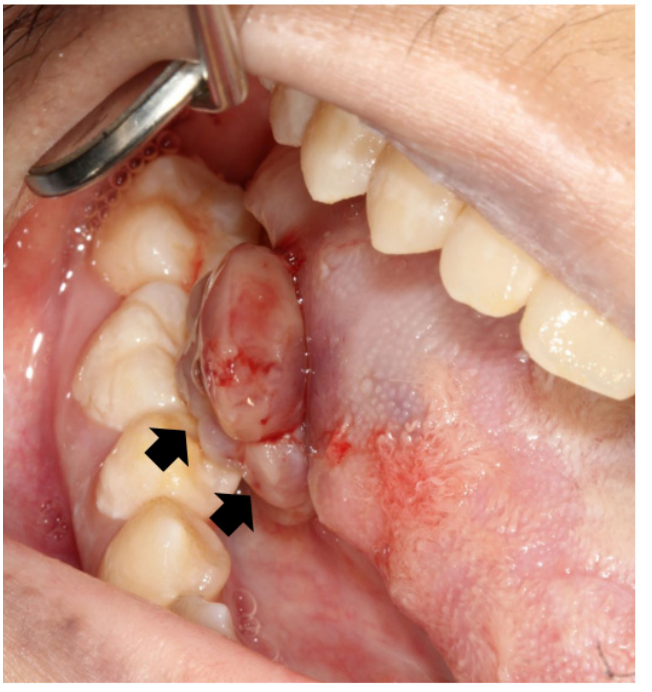

Fig. 1 A pedunculated, multinodular soft tissue mass on the right lateral tongue with ulceration (black arrows).

III. DISCUSSION

Abnormal oral symptoms, especially tumor like mass, may be worrisome in leukemia patients or those with a history of leukemia. If the patient had an additional history of chemotherapy or HCT, the differential diagnosis may be more complicated. ALL relapse, complications of chemotherapy and GVHD must be considered during clinical diagnosis.

Typical oral manifestations of leukemia and their treatments are mucositis, cervical lymphadenopathy, infections, periodontal diseases and gingival bleeding[4,5]. Extramedullary leukemic infiltration is mainly observed in acute myeloid leukemia (AML) rather than ALL. Myeloid sarcoma, the extramedullary tumor arising in AML patients, is composed of immature myeloblasts or granulocytic cell proliferation appearing as

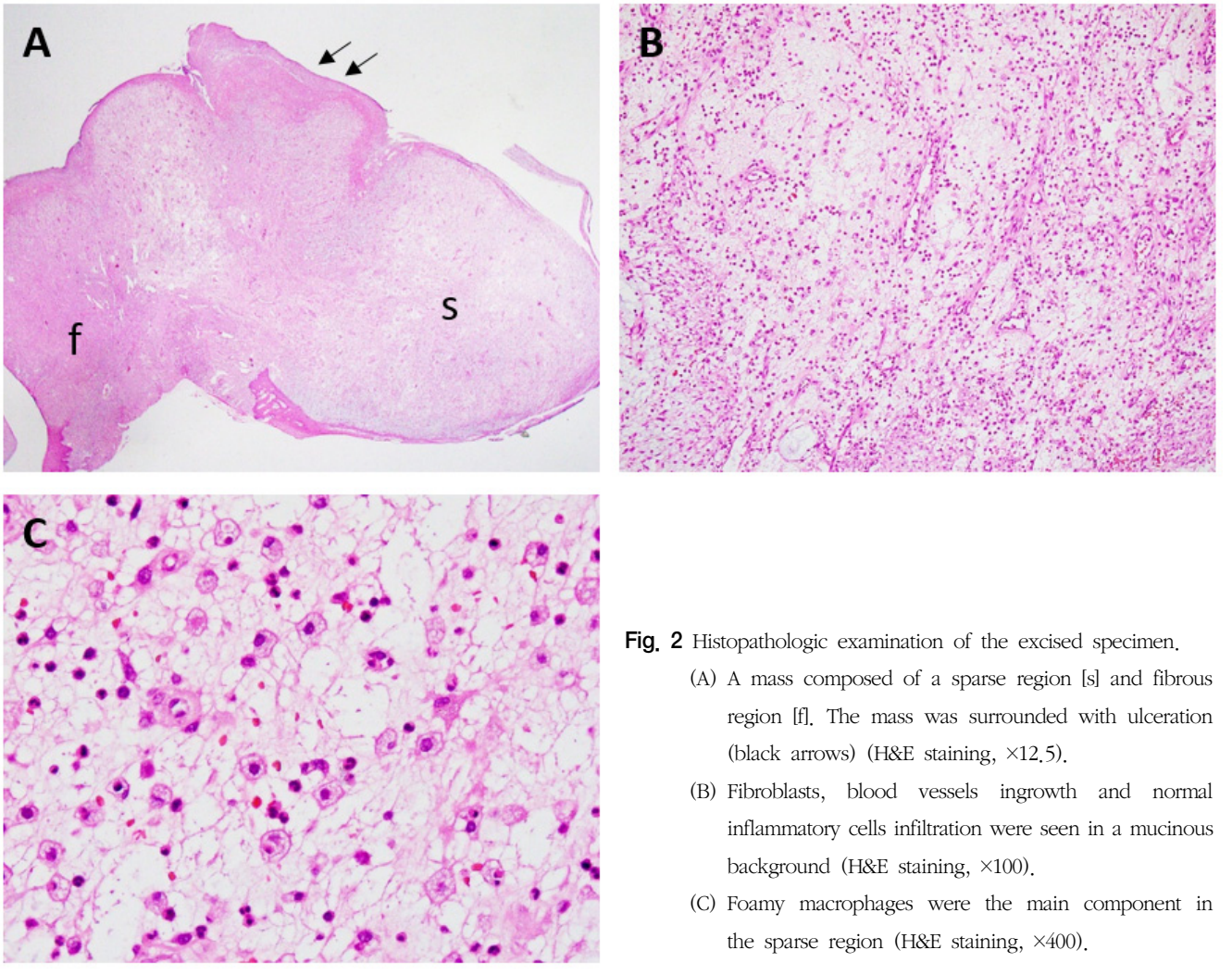

Fig. 2 Histopathologic examination of the excised specimen.
(A) A mass composed of a sparse region [s] and fibrous region [f]. The mass was surrounded with ulceration (black arrows) (H&E staining, ×12.5).
(B) Fibroblasts, blood vessels ingrowth and normal inflammatory cells infiltration were seen in a mucinous background (H&E staining, ×100).
(C) Foamy macrophages were the main component in the sparse region (H&E staining, ×400).

a painful ulcerated reddish-brown nodule[6]. Leukemic infiltration in ALL may exhibit leukemic infiltration in organs in features of hepatomegaly or splenomegaly, and rarely infiltrates in the major salivary glands[7,8]. Generalized gingival enlargement by leukemic infiltration is not as common as in AML. Unlike AML, a localized tumor mass in the extramedullary region is not typical in ALL. Therefore, a localized tumor like lesion is less likely to be a sign of ALL leukemic infiltration or relapse.

The soft tissue mass in our case turned out to be a large mucocele with untypical characteristics. Ulceration, pedunculated-multinodular morphology and reddish color were supposed to be due to the organizing state of the mucocele, clinically mimicking pyogenic granuloma or irritation fibroma. There are no specific issues of mucocele in known leukemia patients, but superficial mucoceles are well known to arise in post HCT patients with chronic GVHD. Unlike general mucoceles that typically occur in the lower lip by trauma, superficial mucoceles are thought to arise without trauma at the buccal mucosa or soft palate in mucosal lesions with chronic inflammation as in GVHD[9-11]. It is thought that persistent inflammation of the mucosa induces injury and leakage of mucin in the minor salivary glands with periodic recurrences. The presence of superficial mucoceles may be

an early sign of chronic GVHD in HCT patients and a full evaluation for GVHD must be considered. The tongue mucocele in our case was a single large mucocele with suspected traumatic features of ulceration despite that the patient denied any history of trauma at the tongue. The mucocele may have started as a superficial mucocele and increased in size due to organization and granulation tissue deposition by persistent inflammation and possible trauma during occlusion. The precise etiology of the tongue mucocele cannot be disclosed in this case, nevertheless our patient had additional superficial mucocele like vesicles on the soft palate supporting the possibility of GVHD. Post-HCT patients with mucocele(s) should be evaluated for GVHD, especially when a definite source of trauma was not identified.

ACKNOWLEDGEMENTS

This research was supported by Basic Science Research Program through the National Research Foundation of Korea (NRF) funded by the Ministry of Science, ICT & Future Planning (NRF-2017R1C1B1012464).

REFERENCES

1. Terwilliger T, Abdul-Hay M: Acute lymphoblastic leukemia: a comprehensive review and 2017 update. Blood Cancer Journal 2017;7:e577.
2. Hunger SP, Mullighan CG: Acute Lymphoblastic Leukemia in Children. N Engl J Med 2015;373:1541-1552.
3. Imanguli MM, Alevizos I, Brown R, Pavletic SZ, Atkinson JC: Oral graft-versus-host disease. Oral Dis 2008;14:396-412.
4. Valera MC, Noirrit-Esclassan E, Pasquet M, Vaysse F: Oral complications and dental care in children with acute lymphoblastic leukaemia. J Oral Pathol Med 2015;44:483- 489.
5. Hou GL, Huang JS, Tsai CC: Analysis of oral manifestations of leukemia: a retrospective study. Oral Dis 1997;3:31-38.
6. de Andrade BA, Farneze RB, Agostini M: Myeloid sarcoma of the oral cavity: A case report and review of 89 cases from the literature. J Clin Exp Dent 2017;9:e1167- e1171.
7. Saha A, Dandekar S, Milla S, Roman E, Bhatla T: Bilateral parotid gland enlargement and palpable nephromegaly in infant acute lymphoblastic leukemia: case report and review of the literature. J Pediatr Hematol Oncol 2014;36:246-248.
8. Al-Anazia, Al-Omar HM, Al-Shanqeeti AS, Al-Mubaraki A, AL-Sultan O, Mourad WA: Isolated extramedullary relapse of acute lymphoblastic leukemia in a submandibular salivary gland. 2006;2:109-113.
9. Balasubramaniam R, Alawi F, DeRossi S: Superficial mucoceles in chronic graft-versus-host disease: a case report and review of the literature. Gen Dent 2009;57:82-88.
10. Treister N, Duncan C, Cutler C, Lehmann L: How we treat oral chronic graft-versus-host disease. 2012;120:3407-3418.
11. Chi AC, Lambert PR, 3rd, Richardson MS, Neville BW: Oral mucoceles: a clinicopathologic review of 1,824 cases, including unusual variants. J Oral Maxillofac Surg 2011;69:1086-1093.