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T-Cell Lymphoma after Silicon Injection in the Eyelids: Rare Case

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Abstract

Background: Lymphoma in the eye can affect the eyeball, orbit, tear glands, and eyelids. The involvement of lymphoma in the eyelids is about 5-20%, usually in the form of non-Hodgkin's lymphoma of 80% B lymphocytes, 14% T lymphocytes, and 6% NK cells. Generally, these lymphomas are unilateral and only 20% of cases are bilateral. In Indonesia, there are no reports of T-cell lymphoma in the eyelids. It is difficult to make an exact diagnosis of T cell lymphoma in the eyelids because it sometimes resembles an inflammation so it requires histopathological and immunohistochemical examination so that the therapy given will be more appropriate. **Case**: A 50-year-old woman with complaints of lumps on the left and right upper eyelids that have been getting bigger and bigger since 1 year ago. Initially, a lump on the left eyelid followed by a lump on the right eyelid. The patient with a history of using silicon several years ago was initially suspected of having silicone, then underwent surgery in 2018 with reactive hyperplasia results, then in 2019 the operation was carried out again with the results of low-grade non-Hodgkin malignant lymphoma, then continued with immunohistochemical examination with the results of T cell lymphoma eyelid region. **Conclusion:**Histopathological examination is sometimes difficult to distinguish between reactive hyperplasia and malignant lymphoma. This is because the picture is not typical so further examination with immunohistochemistry is needed for diagnosis using CD 20, CD 3 and ki67.

Keywords: Non Hodgkin Lymphoma, T cell lymphoma, Eyelid

Introduction

Lymphoma in the eye can affect three main parts of the eye, namely the globe, the orbit (the network that surrounds the eye), and the eye accessory (the tear glands and eyelids), known as the ocular adnexallymphoma(LAO). Most of the lesions in this area are Non-Hodgkin's Lymphoma (NHL), 80% are from B lymphocytes, 14% from T

lymphocytes, and only 6% are from NK-Cell.[1]The orbital lymphoma of the eye represents 6-8% of the entire orbit of the tumor and 10-15% of the magnetictumor. The involvement of the periocular location consists of the conjunctiva (20-33%), the orbit (46-74%), and the eyelids (5-20%).[1]The incidence of ocular adnexal lymphoma is highest among adults over the age of 50. There were no significant differences between men and women.[2] Incidences were more frequent in white than black races.[1]Most non-Hodgkin orbital and adnexal lymphomas showed low-degree types (84%) and only 16% histologically showed high degrees. Orbital lymphoma is generally unilateral and only 20% of cases are bilateral.Filler injections can also induce chronic inflammation that triggers the development of lymphoma. This case report aims to report one case of T cell Lymphoma a/r Ocular Sinistra in a 51-year-old woman with a left superior palpebra mass.

Case report

A female, fifty-one years old, Islamic religion, tribe of the Sundays, last education ended SLTP and worked as a householder, was taken with medication to the Oculoplasty and Oncology Reconstruction Police (ROO), Eye Hospital Cicendo Bandung, on April 17, 2019, with the primary complaint there were bumps on the upper left and right eyelids that increased day by day. From the history it is known that the patient came with complaints there were bumps of the top eyelid that were felt 1 year before entering the hospital, starting with a bumper of the left eyelid that was increasing, then followed by a bumper of the right eyelid since 5 months before entry into the hospital. The bump is loud and the eyes are watery. The patient had a history of silicone injections in the salon a few years ago. The previous patient had been hospitalized and underwent mass evacuation operations of silicone OS on July 13, 2018, at Cicendo Eye Hospital in Bandung. On April 18, 2019, the patient was hospitalized again and underwent surgery to debulk the mass of the OS lymphoma, silicone OS extraction, and OD xanthelasma extraction. The final results of the patient's biopsy were in the case we microscopically (Figure 1) showing fibrocolagen connective tissue stroma, glands that have undergone atrophy, among them hyperplastic lymphoid cells, diffused, small-sized, polymorphic core, hyperchromatic, and mitosis difficult to find, complicated reactive diagnosis of hyperplasia lymphoida/r ocular sinistra diagnosed comparing with low-grade non-Hodgkin malignant lymphoma.

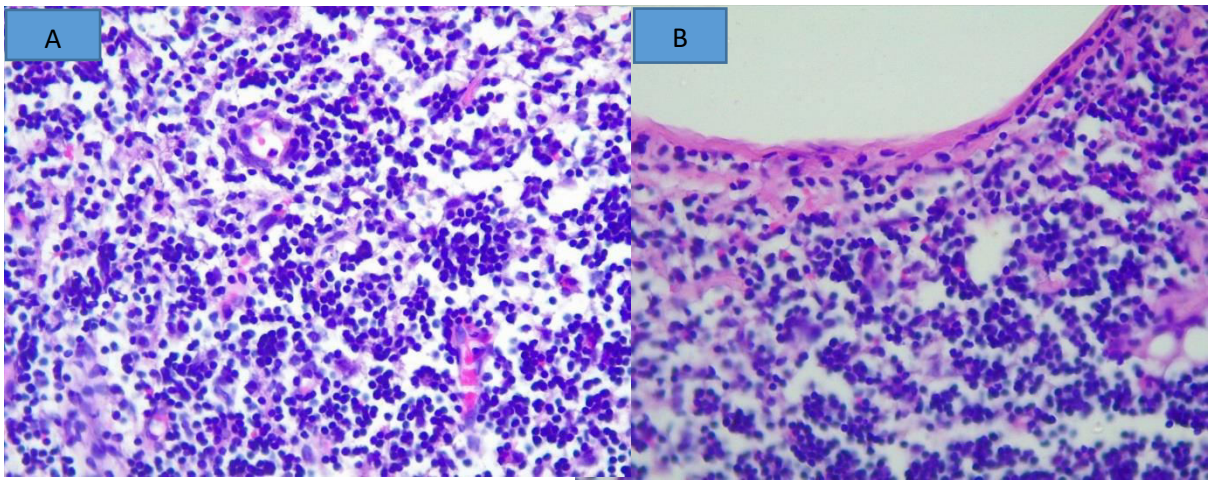
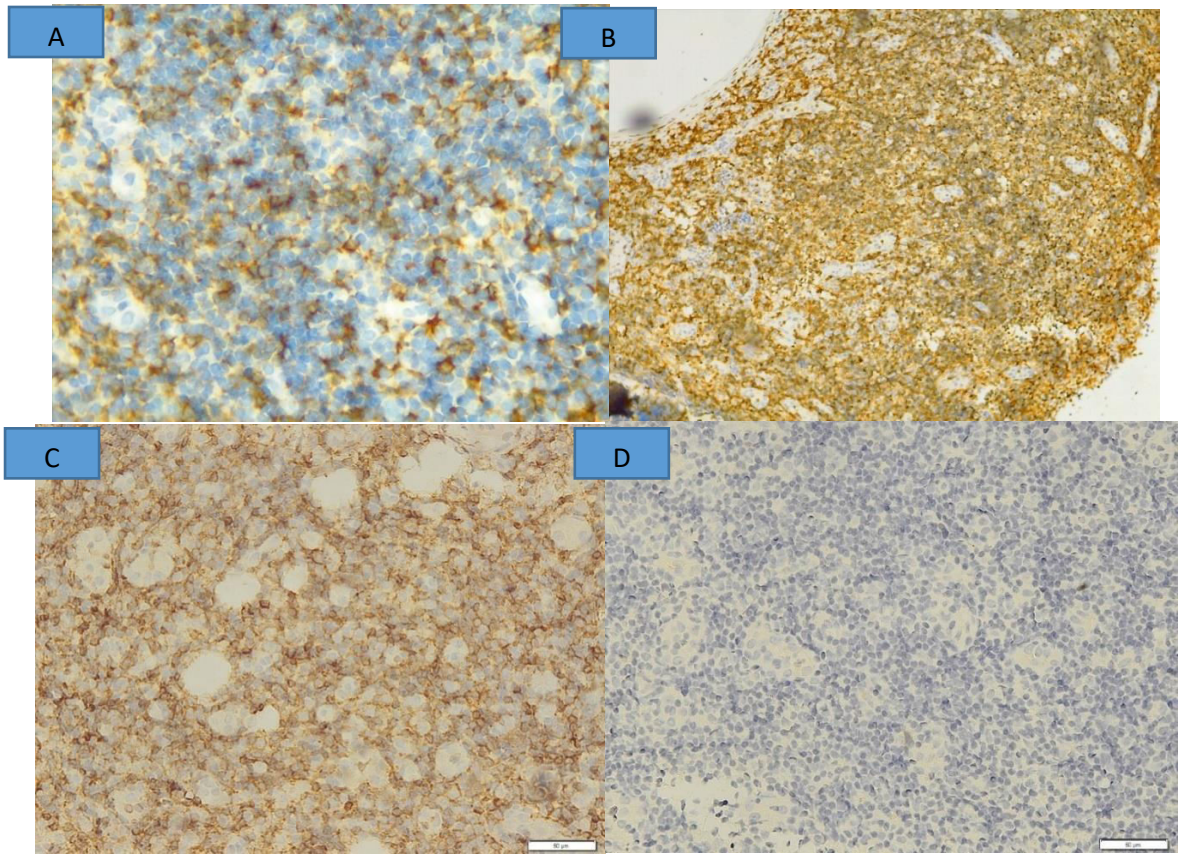


Figure 1. A and B (400x) malignant lymphatic cells spread diffused small size, surrounding blood vessels (blue arrows), and infiltrating into the epithelial layer (green arrow)

After the immunohistochemistry was performed, the results of the IHK examination (Figure 2) were obtained CD20 positive normal, CD3 positive, Ki67 positive high, CD5 positive and CD30 negative with the conclusion T cell lymphoma.



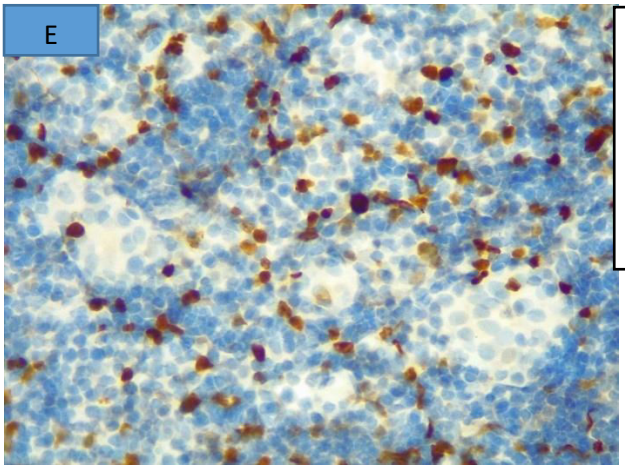


Figure 2.A(400x) CD 20 positive normal, B(100x) diffuse CD 3 positive, C (400x) diffused CD 5 positive, D (100x) CD 30 negative, E (400x) ki67 high proliferation

Discussion

Lymphoma in the eye is a lymphoma that can be located in orbit (40%), followed by the conjunctiva (35%-40%), the lacrimal gland (10-15%), and the palpebra (10%). The most common type of non-Hodgkin lymphoma may be a subtype of B-cell lymphoma or T-cell lymphoma.[4] The incidence of this lymphoma is highest in adults over 50 years of age, there is no significant difference between men and women, and it is more common in white people than in black people.[1,2] According to the National Cancer Institute Surveillance, Epidemiology and End Results, 2,390 cases were in the United States in 2008 (1.340 males and 1,050 females) and the incidence continues to rise every year.1 In Indonesia, the incidence of non-Hodgkin lymphoma is unknown. Basic Health Research data for 2013 a prevalence of lymphoma of 0.06%, or an estimated 14.905 people.[5] In the RSUD Dr.Soetomo recorded confirmed cases of adnexal ocular lymphoma of 78 cases during the period 2015-2019 with the greatest predilection regarding the orbital tissue (56%) and conjunctive (35%).[2] This corresponds to T cell lymphoma patients in this case who are over 50 years of age and occur in the upper left eyelid (unilateral).

The use of silicone oil as an injection filler has caused serious complications such as infection, granuloma[6,7] silicone migration[7] even the development of lymphoma.[8] Silicone migratory cases in the eyelids occur between 12 months to 19 years after injecting.[9] Injection of the filler can also induce chronic inflammation that triggers the development. Although the incidence of lymphoma after the injection of the filler is very rare.According to the patient in this case, the patient is an immunocompetent, but the patient has a history of using silicone injections several years ago then developed into T cell lymphoma on the eyelids.

This eye lymphoma has a slow onset and progressivity for years to show clinical symptoms. Symptoms vary depending on the location of the lesion. Symptoms include proctosis, mass in the ocular area, double vision (diplopia), impaired eye motility, and a sharp reduction in vision. The unusual picture included pain, inflammation and

rapidly growing mass with optic nerve disorders.[2] The lesions that occur can be unilateral or bilateral, involving the presepta of the eyelid. Lesions to the eyelids often result in swelling and prolapse of the eyepieces.[1]As in this case, the lymphoma appears as a slowly increasing mass, swelling in the eyeballs, and a sharp reduction in vision characterized by a decrease in visibility. Lymphoma of the eyelids typically concerns the upper eyelid, blurred as a soft mass and cranky with a clear rear border.2 In this case, macroscopically found a solid mass located on the superior palpebra ocular sinistra. With a tightly bounded white mass of 1.5cm x 1cm, so clinically suspected as a silicoma with a comparative diagnosis of lymphoma.

Microscopically, the T-cell lymphoma precursor shows immature cell immunophenotypes characterized by diffuse proliferation, nuclei of "Blast cells" with intermediate sizes of partially "convoluted" oval shapes, 0-1 nucleoli, smooth chromatin dispersed evenly, and found intranuclear terminal deoxynucleotidyltransferase (TdT) and variable T cell antigen expression. All T-cell precursors have an aggressive picture of the tumor.[10] In the case we microscopically (Figure 1) showed fibrocolagen connective tissue stroma, a gland that underwent atrophy, among them hyperplastic, diffuse, small-sized, polymorphic, hyperchromatic, and mitotic lymphoid cells.

Clinical examinations and X-rays alone cannot distinguish malignant lymphoma from lymphoid hyperplasia. Both reactive hyperplasias and malignancies are hypercellular proliferations with rare or nonexistentstroma components. Histologically, a light microscope can show a continuum from lymphatic hyperplasia, and low-grade lymphoma to high-grade severity.[11]Reactive lymphoidhyperplasias can resemble lymphomas, so it is difficult to mark the lesions just by histopathological examination alone. In such cases, immunopathological/immunophenotype and molecular studies are very helpful in further categorization.[12]

Unicentricpolyclonallymphoproliferation typically supports the reactive diagnosis of lymphoid hyperplasia compared to the multisentricity and monoclonality that are characteristic of the lymphoma.[13]In the case we based on biopsy showed a histopathological picture of reactive hyperplasia lymfoid in deferential diagnosis with low grade non hodgkin lymphoma malignant. To obtain a definitive diagnosis immunohistochemical examination to determine the subtype of lymphoma variants among others CD 20, CD 3, CD 30, CD 15, CD 5 and Ki67. The results of the IHK examination (Figure 2) obtained CD20 positive normal, CD3 positive, Ki67 positive high, CD5 positive and CD30 negative with the conclusion T cell lymfoma.

The pathological diagnosis of this lymphoma can be classified as Peripheral T-cell lymfoma, not otherwise specified. In accordance with the WHO classification, peripheral T cell lymphoma (PTCL), NOS, is a different category of nodal and extranodal mature T cell limphoma that does not match the classification of other

mature T-cell lymphocytes. PTCL, NOS, usually is characterized by the T cell phenotype (CD5 and CD7). The CD4+/CD8-phenotype dominates in nodal cases. CD4/CD8 double positive or double negative is sometimes found. The T cell receptor beta (beta F1) is normally expressed and is able to distinguish gamma delta T cell lymphoma from NK cell. CD15 may be positive, and can be expressed along with CD30 found in a small proportion of cases. Some cases may overlap with ALK-negative (ALK-) anaplastic large cell lymphoma (ALCL), but are still classified as PTCL, NOS, in current guidelines. CD15 expression is associated with poor prognosis.Proliferation is usually high, and the Ki-67 proliferation index >70% is linked to worse prognoses.The management of eye lymphoma therapy depends on the clinical picture, histological subtype, staging and systemic involvement. In general, treatment can be divided into systemic therapy or local therapy. In ocular adnexate lymphoma, systemic involvement requires management of systemic therapy with chemo, immunotherapy, radioimmune therapy, or a combination. In low grade lymphoma or lymfoma with low type without the presence of systemic involvement can be performed focal therapy i.e. cryotherapy, excision surgery and or therapy using radiation beam External beam radiation therapy can give a good therapeutic response because the lymphatic cells in the eye are radiosensitive.[2] Surgical therapy has been proven effective in the subtype of ocular lymphoma variants d'Emblee and lymfoma containing sarcoid-like granulomas.[14,15] In these patients, we chose for surgical treatment with the aim of reducing the volume of tumor mass and biopsy. The surgery was carried out due to the absence of systemic involvement in the patient. However, several publications have a total of 80 patients with surgical excisions without complementary therapies such as radiotherapy, suggesting that local recurrences occur more frequently.3 So that periodic and long-term follow-up as well as other modalities of therapy can be considered. Radiotherapy is an optional treatment for local tumors with additional chemotherapy for patients with general skin lesions or systemic spread.[16]If radiation therapy is used for eyelid lesions, then good care such as eye hygiene should beined. A common side effect of radiation of the upper eyelids is a damage to the lacrimal glands that leads to xerophthalmia. In addition, eyelid protectors should be used to help prevent radiation retinopathy.[17] Systemic chemotherapy may use high-dose methotrexate via intravenous continued orbital radiation. The chemotherapy response showed complete remission, although some had relapses. Complications of systemic methotrexate can be periorbitaledema, blepharitis, conjunctive hyperemia, lacrimation, and photophobia.[1] In this case, the patient did not receive radiation therapy or chemotherapy. Although radiation therapy is quite effective in controlling lymphoma, there is still a possibility of relapse and ocular toxicity or reduced visual acuity. Similarly with chemotherapy, although quite effective in the higher stages, aggressive administration of chemotherapies in elderly patients causes problems with the presence of comorbid disease. Kirsch a systemic T-cell lymphoma patient with

mass of eyelids treated with systemic chemotherapy and local radiation had some recurrences.[18]

The prognosis of patients with eye lymphoma is generally good (five-year survival rates between 50% and 94%). The location of the presentation of primary lymphoma of the eye is associated with the risk of systemic involvement.[3] Lymphomas of the eyelids are at the highest risk because of their more aggressive course. They are linked to an increase in the incidence of systematic lymphoma of approximately 67% to 100% compared to orbital and conjunctive lymphocytes.[2]In these cases, patients with T-cell lymphomaa/rpalpebra superior ocular sinistra require periodic and long-term follow-up. The patient is scheduled to return to control after one week of surgery.

Conclusion

Most of the lesions in this area are non-Hodgkin's Lymphoma (NHL), 80% are from B lymphocytes, 14% from T lymphocytes, and only 6% are from NK cells. Histologically, through a light microscope, it is tough to distinguish the reactive hyperplasia lymphoid from the malignant, because it has an unusual picture. In cases like this, immunohistochemical examinations to determine the subtypes of lymphoma variants, including CD 20, CD 3 and Ki67, are very helpful in further categorization.

Ethics: Informed Consent: Obtained.

Authorship Contributions: Concept, Design, Data Collection or Processing, Analysis or Interpretation, Literature Search, Writing: RJ, DI

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References:

1. Soebagdjo, H.D. Onkologi Mata. Surabaya: Airlangga University Press. 2019. 116-7.
2. Fatmariyanti S, Prastianti AJ. Limfomaadneksaokuli. Surabaya: KSM IlmuKesehatan Mata RSUD Dr. Soetomo. 2021:17-9.
3. Rahman A. Diagnosis danpenatalaksanaanlimfomaorbita. Padang: BagianllmuKesehatan Mata FakultasKedokteranUniversitasAndalas/RSUP Dr. M. Djamil. 2014;37(2):31-7.
4. D.J. Verret et al. T-cell lymphoma of the upper eyelid: A case report and review of the literature. Surgical Oncology (2006) 15, 135–139
5. Pusat Data danInformasiKementerianKesehatan RI. Data RisetKesehatanDasartahun 2013. BadanLitbangkes, Kemenkes RI dan Data PendudukSasaran. 2015; 3-5.

6. Altmeyer MD, Anderson LL, Wang AR. Silicone migration and granuloma formation. J CosmetDermatol. 2009;8:92-7.
7. Ellenbogen R, Rubin L. Injectable fluid silicone therapy. Human morbidity and mortality. JAMA. 1975;234:308-9.
8. Lee SH, Kim HC, and Kim YJ. B-Cell Lymphoma in a Patient With a History of ForeignBody Injection. J Craniofac Surg. 2017;28(2): 504–505.
9. Lee JH, Kim YD, Woo KI, Kong M. Subconjunctival and orbital silicone oil granuloma(siliconoma) complicating intravitreal silicone oil tamponade. Case Rep Ophthalmol Med. 2014;2014:686973.
10. Ioachim HarryL, Medeiros. Lymph Node Pathology, Fourth Edition, LippincotWiilliam& Wilkins ;2003: 294-303
11. Holds JB, Chang WJ, Dailey RA, et al. Basic and clinical science course, Section 7, Orbit, Eyelids, and lacrimal system. 2008–2009 ed. San Francisco, CA: American Academy of Ophthalmology, 2008:81–8.
12. Fukuhara J, Kase S, Ishijima K, et al. Conjunctivallymphoproliferative disorder. Ophthalmology 2011;118:423.e1–2.
13. Mannami T, Yoshino T, Oshima K, Takase S, Kondo E, Ohara N, et al. Clinical, histopathological, and immunogenetic analysis of ocular adnexal lymphoproliferative disorders: Characterization of malt lymphoma and reactive lymphoid hyperplasia. Mod Pathol 2001;14:641-9.
14. PileriS.A.Weisenburger D.D. Sng I. Muller-Hermelink HK. Chan WC. Nakamura S. Jaffe E.S. Peripheral T-cell lymphoma, NOS in WHO Classification of Tumors of Haematopoietic and Lymphoid Tissue. 2017: 403-7.
15. Guanche AD, Bohjanen K, Tope WD. Surgical treatment of obstructive palpebral tumors in d'emblee variant of cutaneous T-cell lymphoma. Dermatological Surgery 2004;30(12 Part 2):1568–71.
16. Behbehani RS, Bilyk JR, Haber MM, Savino PJ. Orbital lymphoma with concomitant sarcoid-like granulomas. Ophthalmological Plastic and Reconstructive Surgery 2005;21(6):458–61.
17. Onesti MG, Mazzocchi M, De Leo A, Scuderi N. T-cell lymphoma presenting as a rapidly enlarging tumor on the lower eyelid. Acta Chirurgia Plastica 2005;47(3):65–6.
18. Cook Jr. BE, Bartley GB, Pittelkow MR. Ophthalmic abnormalities in patients with cutaneous T-cell lymphoma. Ophthalmology 1999;106(7):1339–44.