CASE REPORT

# Chloroma of the testis in a patient with a history of acute myeloid leukemia

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Chloroma, or granulocytic sarcoma, is a rare extramedullary solid hematologic cancer, found concomitant with acute myeloid leukemia. It is infrequently associated with other myeloproliferative disorders or chronic myelogenous leukemia. Chloroma of the testis after allogeneic bone marrow transplantation is particularly sparsely represented in the literature. It is suggested that an appropriate panel of marker studies be performed along with clinical correlation and circumspection to avoid misleading conclusions. We report an interesting case of a 32-year-old male with a clinical history of acute myelogenous leukemia, postallogeneic peripheral blood stem cell transplantation that was found to have chloroma of the right testis.

**Key words:** Acute myeloid leukemia, chloroma, testis

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## INTRODUCTION

Chloroma is a rare extramedullary neoplasm composed of immature myeloid cells (myeloid sarcoma)[1] presenting as single or multiple masses.[2] It would affect both sexes equally,[2] children more commonly than adults and 60% of the patients are younger than 15-year-old.[3] Myeloid sarcomas are associated with acute myeloid leukemia (AML) in 2%–8% of the cases, especially in situations of cytogenetic abnormalities such as t, inv, and 11q23.[4-6] The testicles are considered to be an uncommon site for myeloid sarcomas,[7,8] and there are a few cases with localization of chloroma in one of the testes.[9] Hereby, we would report an extremely rare case of chloroma of the testis in a patient with a history of AML.

## CASE REPORT

In the March of 2015, a 32-year-old man in Isfahan, Iran, had a history of AML sought care because of a painless right testicular mass. The AML, that was diagnosed 6 years earlier, was classified as M4 using the French–American–British system. The patient had received aggressive chemotherapy with multiple episodes of relapse and eventually underwent allogeneic peripheral blood stem cell transplantation 5 years ago. The testicular mass was detected 45 months after transplantation. The right testicular ultrasound showed two masses, 1.7 cm × 1.1 cm and 2.2 × 1.5, with increased blood flow. Then, the right orchiectomy was performed. No cytogenetic analysis was performed.

Macroscopic anatomopathological study of a sample from the right orchiectomy revealed two 1.7 and 2.2 cm masses with creamy color, relatively firm and solid sectional surface, placed at the outer half of the testis.

Histopathologic examination showed diffuse infiltration of immature monotonous large hyperchromatic neoplastic cells with scanty cytoplasm and round-to-oval nuclei [Figure 1].

Immunohistochemical studies consistently manifested the expression of leukocyte common antigen, BCL2, CD117, CD68 (focally), and Ki67 index about 60%, [Figures 2-6] but the expression of cytokeratin, placental alkaline phosphatase, CD10, CD3, CD20, CD3O, CD34,

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and ALK1 was negative and the diagnosis of granulocytic sarcoma was established. No cytogenetic analysis was performed. Afterward, patient's chemotherapy (cytarabine and hydroxyurea) was started, and now, he is well.

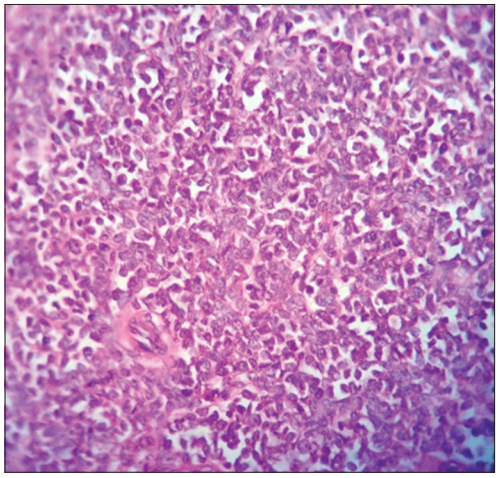

**Figure 1:** Histology shows diffuse infiltration of immature monotonous large hyperchromatic neoplastic cells with scanty cytoplasm (H and E, ×400)

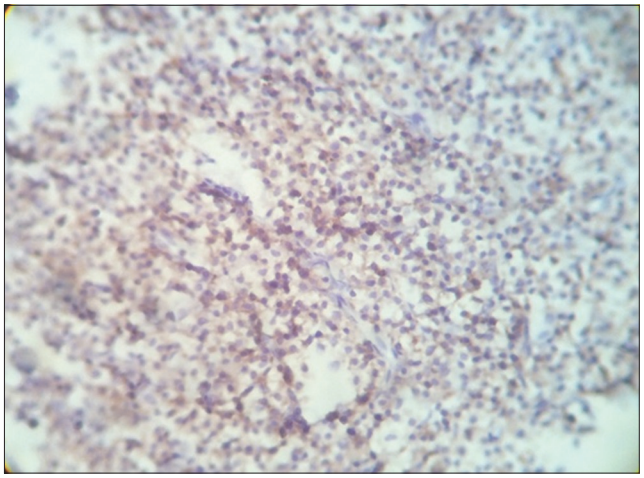

**Figure 3:** Immunohistochemistry shows positivity for CD117

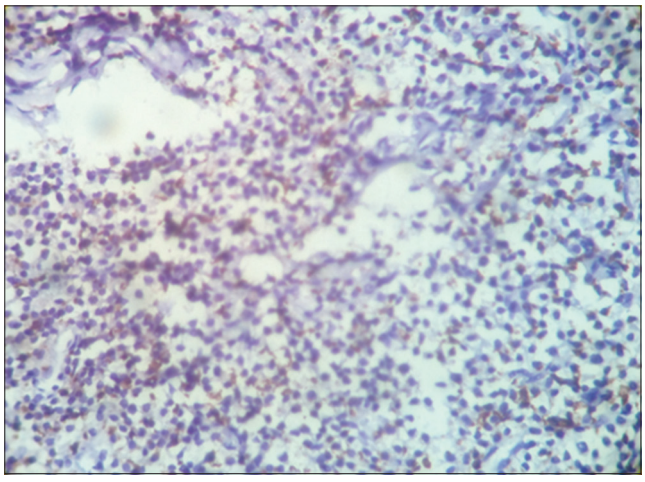

**Figure 5:** Ki67 index is about 60% of neoplastic cells (×400)

## DISCUSSION

Chloroma usually occurs as a secondary manifestation either before or simultaneously with AML.[2] Less frequently, it may appear after complete hematologic remission,[10] which

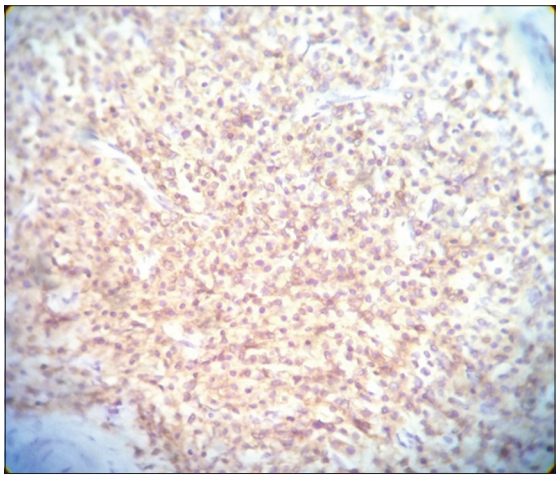

**Figure 2:** Immunohistochemistry shows positivity for leukocyte common antigen

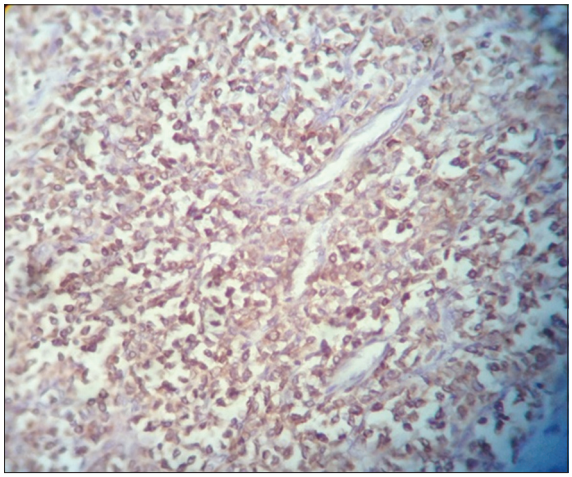

**Figure 4:** Immunohistochemistry shows positivity for BCL2

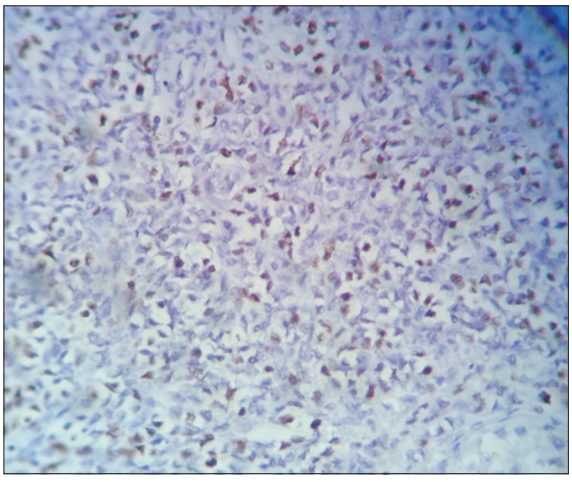

**Figure 6:** Ki67 index is about 60% of neoplastic cells (×400)

strongly indicates bone marrow or other extramedullary relapses.[10] It has also been observed as a primary chloroma which precedes AML by months or years[3,11,12] or it can be an independent entity, without progression to a hematologic disease.[7,13] Chloroma may also appear as a relapse in patients with a history of chronic myeloid leukemia (CML) after allogeneic peripheral blood stem cell transplantation and subsequent complete hematologic remission.[14] GS affects 2.5%–9.1% of the patients with AML[2] and 4.5% of those with CML.[15]

The most affected sites are bone structures (cranium, paranasal sinuses, sternum, ribs, vertebrae, and pelvis), central nervous system, soft tissues of the head and neck (especially the orbit), skin, lymph nodes, and breasts.[16,17] Initial presentation of AML with involvement of the testicles, as described in the present case, is considered to be uncommon, with a poor prognosis.[8,18] The correct histological diagnosis is based on identifying the granulocytic characteristics in the neoplastic cells. Because of high morphological variability and possible nonidentification of myeloid differentiation (variable), errors may occur especially when the neoplasia presents minimal myeloid differentiation such that it is composed of poorly differentiated cells distinct from the myeloblasts that are found in the bone marrow in cases of concomitant AML.[5,19]

Immunohistochemistry is of great value in identifying antigens associated with the myeloid lineage (CD13, CD33, CD43, CD117, lysozyme, and CD68). In the current case, the neoplasia showed positive reaction for CD68, which is the most commonly expressed markers of myeloid differentiation.[6,20]

## CONCLUSION

Despite the rarity of myeloid sarcoma, it should be taken into consideration in the differential diagnoses of undifferentiated neoplasia, with proper use of immunohistochemical techniques to make a rapid diagnosis and start treatment, regardless of the unsatisfactory response with frequent relapses, and evolution to acute leukemia.

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### Conflicts of interest
There are no conflicts of interest.

## REFERENCES

1. Vardiman JW, Thiele J, Arber DA, Brunning RD, Borowitz MJ, Porwit A, *et al.* The 2008 revision of the World Health Organization (WHO) classification of myeloid neoplasms and acute leukemia: Rationale and important changes. Blood 2009;114:937-51.
2. Pui MH, Fletcher BD, Langston JW. Granulocytic sarcoma in childhood leukemia: Imaging features. Radiology 1994;190:698-702.
3. Neiman RS, Barcos M, Berard C, Bonner H, Mann R, Rydell RE, *et al.* Granulocytic sarcoma: A clinicopathologic study of 61 biopsied cases. Cancer 1981;48:1426-37.
4. Khan MY, Hussein KK, Walter MG, Hasan MK, Kern W, Kharfan-Dabaja MA. Granulocytic sarcoma presenting with malignant anasarca in a patient with secondary acute myeloid leukemia. Int J Hematol 2004;79:250-2.
5. Bakst RL, Tallman MS, Douer D, Yahalom J. How I treat extramedullary acute myeloid leukemia. Blood 2011;118:3785-93.
6. Rocha Filho FD, Ferreira FV, Lima GG, Lima MG, Figueiredo AA. Apresentação incomum de sarcoma granulocítico mamário. Rev Bras Hematol Hemoter 2009;31:295-8.
7. Eggener SE, Abrahams A, Keeler TC. Granulocytic sarcoma of the testis. Urology 2004;63:584-5.
8. Valbuena JR, Admirand JH, Lin P, Medeiros LJ. Myeloid sarcoma involving the testis. Am J Clin Pathol 2005;124:445-52.
9. Pileri SA, Ascani S, Cox MC, Campidelli C, Bacci F, Piccioli M, *et al.* Myeloid sarcoma: Clinico-pathologic, phenotypic and cytogenetic analysis of 92 adult patients. Leukemia 2007;21:340-50.
10. Ghadiany M, Attarian H, Hajifathali A, Khosravi A, Molanaee S. Relapse of acute myeloid leukemia as isolated bilateral testicular granulocytic sarcoma in an adult. Urol J 2008;5:132-4.
11. Economopoulos T, Alexopoulos C, Anagnostou D, Stathakis N, Constantinidou M, Papageorgiou E. Primary granulocytic sarcoma of the testis. Leukemia 1994;8:199-200.
12. Kawashima H, Sakamoto W, Nishijima T, Hanada M, Mori K, Maekawa M. Granulocytic sarcoma of testis preceding acute myelocytic leukemia. Urol Int 1988;43:310-2.
13. Chiou SY, Chiou HJ, Chou YH, Tiu CM, Pan CC, Chang CY. Sonographic features of primary testicular granulocytic sarcoma. J Ultrasound Med 2003;22:1413-6.
14. Hu SW, Huang SP, Yang SF, Chai CY. Chloroma of the testis after allogeneic peripheral blood stem cell transplantation: A case report. Kaohsiung J Med Sci 2004;20:506-11.
15. Muss HB, Moloney WC. Chloroma and other myeloblastic tumors. Blood 1973;42:721-8.
16. Cheah KL, Lim LC, Teong HH, Chua SH. A case of generalised cutaneous granulocytic sarcoma in an elderly patient with myelodysplastic syndrome. Singapore Med J 2002;43:527-9.
17. Yamauchi K, Yasuda M. Comparison in treatments of nonleukemic granulocytic sarcoma: Report of two cases and a review of 72 cases in the literature. Cancer 2002;94:1739-46.
18. McIlwain L, Sokol L, Moscinski LC, Saba HI. Acute myeloid leukemia mimicking primary testicular neoplasm. Presentation of a case with review of literature. Eur J Haematol 2003;70:242-5.
19. Wong WS, Loong F, Ooi GC, Tse TC, Chim CS. Primary granulocytic sarcoma of the mediastinum. Leuk Lymphoma 2004;45:1931-3.
20. Campidelli C, Agostinelli C, Stitson R, Pileri SA. Myeloid sarcoma: Extramedullary manifestation of myeloid disorders. Am J Clin Pathol 2009;132:426-37.