TWO CASES OF CHRONIC MYELOID LEUKEMIA IN PEDIATRIC AND YOUNG ADOLESCENT AGE GROUP

HOSSAIN MZ[1], HASAN P[2], AHMED B[3], MOZAZFIA KT[4], FERDUSH Z[5], TASNIM H[6], KABIR MA[7], KHAN MS[8]

Abstract

The incidence of chronic myeloid leukemia (CML) is 1-2 cases/100 000/year, without major geographical differences and the median age of presentation is 60–65 years1 or approximately 67 years. Commonly known as a disease of the elderly, CML rarely affects the pediatric group (0-14 years), or adolescent and young adults (15-29 years). Age specific rate of CML per 100,000 of population, have been reported in pediatric age group as 0.04 and in young adults and adolescent group as 0.22. There is also much scarcity of available literature on the characteristic, presentation, outcome and response to standard protocol treatment in this age group, although it has been noted recently by many researchers that, they differ significantly from the much commonly affected elderly population. We here report two cases of pediatric and young adolescent CML.

Keywords: *Pediatric, Young adolescents, CML, Chronic myeloid leukemia*

J Dhaka Med Coll. 2016; 25(1): 74-76

Introduction

The incidence of chronic myeloid leukemia (CML) is 1-2 cases/100 000/year, without major geographical differences and the median age of presentation is 60–65 years[1] or approximately 67 years.[2] Commonly known as a disease of the elderly, CML rarely affects the pediatric group (0-14 years),[3] or adolescent and young adults (15-29 years).[3–6] In India, Dikshit et.al. reported that during 2001-05, age specific rate of CML per 100,000 populations in pediatric age group was 0.04 and in young adults and adolescent groups was 0.22.[3] Such incidence rate or prevalence for Bangladeshi population is unknown, yet experience from clinical practice allow us to recognize the rarity of the condition. Incidence of CML in this age group merits much attention as outcome and response to standard treatment often differs significantly from the adult group.[7] We here report two cases of pediatric and young adolescent CML.

Case report

Case 1

A 22 years old Muslim Bangladeshi male presented with complaints of fever for 15 days and hemoptysis for same duration. Fever was high grade, continued, maximum temperature was 103º F, not associated with chills and rigors. He also complained of hemoptysis for same duration. On query, he also gave history of multiple purpuric spots for 10 days on both lower limbs. On examination, generalized lymphadenopathy and bony tenderness were readily found. Abdominal examination revealed there was hepatosplenomegaly. His family or drug history were non-significant. On investigation, his total count of WBC was 2,18,000/mm^3, Hemoglobin 5 g/dL, total platelet count was 10,000/mm^3. Peripheral blood film showed 20% myelocytes and 2% blasts, and along with other features was totally characteristic of CML. He was advised BCR-

1. Dr. Mohammad Zaid Hossain, Associate Professor, Department of Medicine, Dhaka Medical College, Dhaka.
2. Dr. Pratyay Hasan, Indoor Medical Officer, Department of Medicine, Dhaka Medical College Hospital, Dhaka.
3. Dr. Benozir Ahmed, Assiatant Registrar, Department of Medicine, Dhaka Medical College Hospital, Dhaka.
4. Dr. Kazi Tuba-E Mozazfia, Assistant Registrar, Dept. of Medicine, Shaheed Suhrawardy Medical College, Dhaka.
5. Dr. Zannatul Ferdush, Intern Doctor, Department of Medicine, Dhaka Medical College Hospital, Dhaka.
6. Dr. Homyra Tasnim, Intern Doctor, Department of Medicine, Dhaka Medical College Hospital, Dhaka.
7. Dr. Md. Sarwar Khan, Lieutenant Colonel & Medicine Specialist, BGB Hospital, Dhaka
8. Dr. Md. Alamgir Kair, OSD, DGHS, Mohakhali, Dhaka

Correspondence: Dr. Mohammad Zaid Hossain, Associate Professor, Department of Medicine, Dhaka Medical College, Dhaka. Email: zhvalentino@gmail.com

Received: *01 March 2016* ***Accepted:*** *20 March 2016*

ABL oncogene, and was found positive. It was noted that the patient had more or less normal weight, which persisted during the course of his illness between 50-60 kg.

Case 2

A 13 years old Muslim Bangladeshi male presented with fever for 7 days, bone pain for 7 days, 4 bouts of epistaxis. He had been previously diagnosed as a case of CML, by CBC and PBF, both of which showed findings characteristic of CML. He had been on imatinib mesylate for the last two years. He had had hepatosplenomegaly previously as evidenced by his medical records. Ha had none of them when he presented to us, but rather he presented to us with features of pancytopenia; total WBC 1,000/mm^3, total platelets 25,000/mm^3, Hb 6.5 g/dL. His bone marrow biopsy revealed bone marrow aplasia with lymphoblastic changes. This features suggested an exaggerated response to imatinib mesylate. It was noted that the boy was cachectic and had a weight of 35 kg.

Discussion

The two cases reported here presented with all the typical features of CML except for the age which belonged to much uncommon young adolescent group for the 1st case and pediatric age group for the 2nd case. There is also much scarcity of available literature on the characteristic, presentation, outcome in this age group, although it has been noted recently by many researchers that, they differ significantly from the much commonly affected elderly population.[7] Although in these two cases, reaching the diagnosis was easily possible, still recent increase in the incidence of CML is not in conformity with the existing experience of the clinicians in our country. This type of disease may pose as diagnostic dilemma for unsuspecting eyes.

It has been commented that generally, tyrosine kinase inhibitors, such as imatinib mesylate, is superior to allogeneic stem cell transplantation (SCT) as first-line therapy of CML, because of transplant-related mortality.[1] Raut et.al. suggested that imatinib mesylate is safe and effective in treatment,[8] in fact, Cortes et.al. suggested that younger patients tolerate tyrosine kinase inhibitors better than older patients.[9] However, Pemmaraju et.al. reported that response rate with imatinib mesylate for adolescent and young adult patients was lower than that seen in older patients. They also found that in this age group, second generation tyrosine kinase inhibitors conferred a better outcome compared to imatinib with a higher rate of complete cytogenic response (CCyR).[7] The reason behind this could be explained by many biological or psychological factors, which are yet not researched . For example, in 41 patients taking imatinib, it was found that body weight was the only variable that was significantly related to imatinib clearance and body weight can frequently be a rapidly changing parameter in many adolescents and young adult patients.[10] In our cases also, imatinib did not show expected outcome or was associated with adverse effects. In the second case, imatinib therapy resulted in undue decrease in total WBC count, and for which we had to switch to growth factor therapy. In this first case also, imatinib did not show expected outcome.

Conclusion

Further study should be undertaken to find out the true incidence of CML in pediatric and young adolescent age groups. As, the response to drugs used according to current protocols, may also differ, the clinicians should be cautious.

Conflict of interest: None

References

1. Baccarani M, Dreyling M. Chronic myeloid leukaemia: ESMO Clinical Practice Guidelines for diagnosis, treatment and follow-up. Ann Oncol. 2010 May 1;21(suppl_5):v165–7.
2. Jemal A, Siegel R, Xu J, Ward E. Cancer statistics, 2010. CA Cancer J Clin. 2010 Oct;60(5):277–300.
3. Dikshit RP, Nagrani R, Yeole B, Koyande S, Banawali S. Changing trends of chronic myeloid leukemia in greater Mumbai, India over a period of 30 years. Indian J Med Paediatr Oncol Off J Indian Soc Med Paediatr Oncol. 2011;32(2):96–100.
4. Thomas DM, Albritton KH, Ferrari A. Adolescent and young adult oncology: an emerging field. J Clin Oncol Off J Am Soc Clin Oncol. 2010 Nov 10;28(32):4781–2.

5. Morgan S, Davies S, Palmer S, Plaster M. Sex, drugs, and rock "n" roll: caring for adolescents and young adults with cancer. J Clin Oncol Off J Am Soc Clin Oncol. 2010 Nov 10;28(32):4825–30.

6. Wein S, Pery S, Zer A. Role of palliative care in adolescent and young adult oncology. J Clin Oncol Off J Am Soc Clin Oncol. 2010 Nov 10;28(32):4819–24.

7. Pemmaraju N, Kantarjian H, Shan J, Jabbour E, Quintas-Cardama A, Verstovsek S, et al. Analysis of outcomes in adolescents and young adults with chronic myelogenous leukemia treated with upfront tyrosine kinase inhibitor therapy. Haematologica. 2012 Jul;97(7):1029–35.

8. Raut L, Bohara VV, Ray SS, Chakrabarti P, Chaudhuri U. Chronic myeloid leukemia in children and adolescents: A single center experience from Eastern India. South Asian J Cancer. 2013;2(4):260–4.

9. Cortes J, De Souza C, Ayala-Sanchez M, Bendit I, Best-Aguilera C, Enrico A, et al. Current patient management of chronic myeloid leukemia in Latin America: a study by the Latin American Leukemia Net (LALNET). Cancer. 2010 Nov 1;116(21):4991–5000.

10. Peeters M, Koren G, Jakubovicz D, Zipursky A. Physician compliance and relapse rates of acute lymphoblastic leukemia in children. Clin Pharmacol Ther. 1988 Mar;43(3):228–32.