Successful Treatment of a Patient with Refractory Anemia by Immunosuppressive Therapy: Another Case of 'Autoimmune Myelodysplasia'?

A.	Alessandro	Bucalossi
G.	Giuseppe	Marotta
P.	Piero	Galieni
E.	Egidio	Dispensa

Division of Hematology, 'A. Sclavo' Hospital, Siena, Italy

Dr. Alessandro Bucalossi, Divisione di Ematologia, 'A. Sclavo' Hospital, Siena, Via Tufi, 1, I-53100 Siena (Italy)

We read with interest the report by Davis and Farver [1] concerning the spontaneous recovery of anemia in a young patient with an atypical pure red cell aplasia. This is probably a further description of a peculiar disease already reported in two young patients by Muller et al. [2]. We observed a similar condition in a 19-year-old man referred to our hospital for a serious anemia. A complete blood count showed: Hb, 4 g/dl; MCV, 94 fl; reticulocyte count, 0.2%; WBC, $3.5 \times 10^9/l$; platelet count, $247 \times 10^9/l$. Bone marrow histologic and cytologic examination displayed a hypercellular picture with dysplastic erythro-poietic hyperplasia, characterized by maturation arrest at the level of proerythroblasts with rare basophilic erythroblasts, nuclear-cytoplasmic asynchrony, and megaloblastic features. Mild maturation defects were also present in both granulocytic and megakaryocytic lineages, with a blast count less than 5%. No chromosomal abnormality was detected. Moreover, there was no serological evidence for parvovirus B19 infection.

A diagnosis of refractory anemia was made after other causes of anemia had been excluded. Therapy with erythropoietin was started for 4 weeks (200 U/kg, three times weekly) without any effect on the transfusion frequency. According to previous reports that described response to glucocorticoid therapy in myelodysplastic syndromes (3, 4), we began treatment with high-dose methyl-prednisolone (2g/12h, 3 times). The same schedule was repeated after 1 week and followed by prednisone (1.5 mg/kg/day) gradually tapered off and withdrawn once the Hb level achieved a normal value. A bone marrow aspirate performed at this time showed an almost complete resolution of dyserythropoiesis with normal granulocytic and megakaryocytic maturation. In conclusion, the analysis of the cases reported in the literature [1, 2], along with our experience, provides further evidence about a rare subset of young patients with dyserythropoietic anemia characterized by a maturation block of erythro-poiesis, normal karyotype, good response to immunosuppressive treatment and perhaps liable to spontaneous remission. In this regard, it may be appropriate to distinguish this last subset of patients with dyserythropoiesis from clonal myelodysplastic syndromes and to consider it as one of the 'autoimmune myelodysplasias' according to Mieschner et al. [5].

References

Davis RB, Farver ML: Pure red cell anemia with erythropoietic maturation arrest and spontaneous recovery. *Acta Haematol* 1994; 92:29-32.

Muller EW, de Wolf JTM, Vellenga E: Successful immunosuppressive treatment after failure of erythropoietin therapy in two subjects with refractory anaemia. *Br J Haematol* 1993;83: 171-172.

Bagby GC, Gabourel JD, Linman JW: Glucocorticoid therapy in the preleukemic syndrome (hemopoietic dysplasia). *Ann Intern Med* 1980;92:55-58.

Motoji T, Teramura M, Takahashi M, Oshimi K, Okada M, Kusakabe K, Mizoguchi H: Successful treatment of refractory anemia with high-dose methylprednisolone. *Am J Hematol* 1990;33:8-12.

Mieschner PA, Favre H, Beris P: Autoimmune myelodysplasias. *Semin Hematol* 1991;28: 322-330.