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Letter to the Editor / Editöre Mektup

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DETECTION OF HEMOGLOBIN D IN A PATIENT WITH HEMOLYSIS DUE TO DAPSONE TREATMENT

DAPSON TEDAVİSİNE BAĞLI HEMOLİZ GÖRÜLEN BİR HASTADA HEMOGLOBİN D SAPTANMASI

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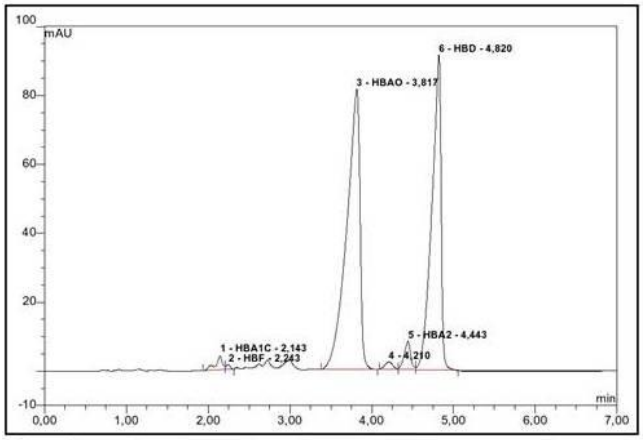
Letter to the Editor

Detection of Hemoglobin D in a Patient with Hemolysis due to Dapsone Treatment

Dear Editor,

Several mutations occur in the genes that encode globin chains which are a part of hemoglobin (Hb) structure. Such mutations give rise to formation of different hemoglobin variants.[1] One of these hemoglobin variants (Hb C, D, E, F and S) is Hb D [β 121 Glu→Gln (GAA→CAA)]. Hb D occurs in 0.2% in Turkey.[2] Molecular analysis has revealed many types of Hb D. Hb D-Los Angeles or Hb D-Punjab is the most common type.[3] Clinically, heterozygous types of Hb D do not produce any hematological manifestations. However, mild-to-moderate cases of hemolysis have been rarely reported in conjunction with its homozygous forms.[4] In this study, we present a case of hemolysis due to dapsone theraphy in coexistence with Hb D.

A 17-year-old female patient who had been on colchicin treatment for Behçet's disease for the last 3 years presented to the rheumatology department with persistent painful sores in her mouth. Following assessment of the patient, treatment with dapsone 50 mg tablets, three times daily was started by the rheumatology department. Two months after initiation of medical treatment, the patient presented to the hematology department with complaints of pallor, chills and fatigue. Her diagnostic work-up showed the following: Hb: 11.2 g/dL (11.9-14.6), MCV: 92.8 fL (81.8-95.5), PLT: 307 x 10^3/uL (173-390), reticulocyte count: 264.5 x 10^3/uL (17-63.8), reticulocyte%: 7.09%, LDH: 367 U/L (0-279), total bilirubin: 2.7 mg/dL (0.1-1.5), indirect bilirubin: 1.5 mg/dL (0.08-1.1) and haptoglobulin <30 mg/dL (30-200). Peripheral smear showed the presence of spherocytes and fragmented red blood cells. Direct and indirect coombs tests were negative. Papain antibody and glucose 6 phosphate dehydrogenase (G6PD) tests were negative. Her laboratory values before dapsone therapy were as follows: Hb: 14.2 g/dL, MCV: 85.2 fL, PLT: 309 x 10^3/uL. Initially, hemolytic anemia induced by dapsone was considered as possible diagnosis and dapsone treatment was discontinued. Laboratory work-up after three weeks showed a reticulocyte count of 125.6 x 10^3/uL, % reticulocyte of 3.49%. These results prompted a request for hemoglobin electrophoresis based on a preliminary diagnosis of thalassemia. Hb D was found as 41.65% (Figure 1). Moleculer genetic test (DNA analysis) was found to be Hb D-Los Angeles [c.3646>C(p.E122Q)(p.Glu.122Gln)(Heterozygous)].



No.	Retention Time min	Peak Name	Height mAU	Area mAU*min	% Relative Area
1	2,14	HBA1C	3,964	0,378	1,32
2	2,24	HBF	1,424	0,082	0,28
3	3,82	HBAO	81,389	15,319	53,31
4	4,21	n.a.	2,133	0,238	0,83
5	4,44	HBA2	8,183	0,747	2,6
6	4,82	HBD	91,39	11,969	41,65

Figure 1. Patient's hemoglobin electrophoresis

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Hemoglobin electrophoresis was performed from the blood sample of her father and revealed Hb D of 88.13% and HbA2 of 3.91%. Her father was diagnosed with beta-thalassemia minor and Hb D.

Drugs are a rare cause of hemolytic anemia. Dapsone is known to cause oxidative hemolysis. This situation has been reported to result from the direct toxic effect of a hydroxlamine derivative formed by N-hydroxylation to erythrocytes.[5] Hemoglobin electrophoresis was performed for the patient during follow-up due to persistent hemolysis and a rare Hb variant was detected. This was a surprising finding for us, the physicians who were following the patient because her previous hemogram and biochemical analyses were not suggestive of thalassemia. Thus, we were facing two rare conditions at the same time: drug-induced hemolysis and Hb D.

As a result; coexistence of hemolysis due to dapsone and Hb D is a rare condition. Since Hb D was found to be heterozygous in the patient, we think that this condition did not have clinical effect on hemolysis. To the best of our knowledge, this is the first case in which Hb D is detected in a patient with hemolysis due to dapsone.

Keywords: *Hemoglobin D, dapson, hemolysis*

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None declared

Conflict of Interest
None declared

Patient Consent
Informed consent (via Journal of Health Sciences of Kocaeli University informed consent form) was obtained from the patient.

Financial Disclosure
None declared

Author Contributions
MO: evaluated the case and wrote the paper, MHA: evaluated the case and made critical review, BB: evaluated the case and made critical review.

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