An autoimmune hemolytic anemia and an ovarian cyst:  
a case report

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Abstract: Autoimmune hemolytic anemia (AIHA) is a rare acquired disease caused by the antibody specifically against some antigens upon the surface of the own red blood cells (RBC). It may spontaneously occur or it can be rarely found as a paraneoplastic syndrome in the patients with various solid tumors. Herein we reported a case of AIHA diagnosed in a patient with an ovarian cyst. She was a 72-year-old Thai patient who had low-graded fever for a week. The physical examination revealed pallor and a large cystic mass occupying the entire left lower abdomen without tenderness. The blood tests were: Hb 8.4 g%, WBC 13,470/mm³, platelet 455,000/mm³, MCV 80.6 fl, reticulocyte 2.6 %, direct anti-globulin test 1+, indirect anti-globulin test 2+, ANA-negative, CA-125 50.4 U/ml, Hb analysis-normal, osmotic fragility-positive. The ultrasonography of the whole abdomen showed a 7.2x11.6x15.6 cm cyst at the left adnexa with internal fluid/fluid level and one septation. She was diagnosed as having AIHA and a large ovarian cyst. And she was treated with corticosteroid and responded well. Hb was 9.5 g% whereas the direct anti-globulin test became weakly positive within one month. AIHA in our patient could be much improved with just only the corticosteroid therapy although the resection of the cystic ovarian mass was not allowed. This outcome was different from nearly all other cases of AIHA associated with tumors. So, AIHA in our case looked likely to be the co-incidence more than the paraneoplastic syndrome of the ovarian tumor. Therefore in a case of AIHA happening in patients with various tumors and the operation could not be accessed due to any reason, the corticosteroid therapy should be considered one of the initial therapies of choice.  

Key Words: Autoimmune hemolytic anemia, Ovarian cyst
Introduction

Autoimmune hemolytic anemia (AIHA) is a rare acquired hemolytic disease caused by the antibody specifically against some antigens upon the surface of the own red blood cells (RBC) resulting in the destruction of the RBC and consequently hemolytic anemia\(^1\). The natural course may be gradual and asymptomatic or rapid and life threatening. To diagnose it depends on the documentation of the immunoglobulin upon the red blood cells by the direct anti-globulin test\(^2\). AIHA may spontaneously occur the so-called primary or idiopathic AIHA or it can be coincidentally found in many diseases/conditions or secondary AIHA such as the autoimmune diseases especially systemic lupus erythematosus (SLE), some drugs eg., alpha methyldopa, isoniazid, etc, the immunoproliferative disorders particularly chronic lymphocytic leukemia\(^3\) or lymphoma and various infections\(^4\).

For solid tumors, AIHA can be found as the rare paraneoplastic phenomenon in some cases with...
cancers of various origins. So far it has been recognized in cancers of the lung, kidney, colon-rectum, ovary, stomach and Kaposi’s sarcoma\(^{(5)}\). In contrast, among 101 Thai patients with primary and secondary AIHA, only one from five patients with some malignancies was found having an ovarian cancer\(^{(4)}\). And herein we reported a case of AIHA in an old Thai woman with a large ovarian cyst.

**Case Report**

A 72-year-old Thai woman was admitted at the medical ward because of the low-graded fever and dry cough for a week, no jaundice. The physical examination revealed the body temperature of 37.1 degree Celsius, pulse 102/min, pallor and an ill-defined mass occupying the entire left lower quadrant of abdomen, cystic consistency, no tenderness, but no peripheral lymphadenopathy.

The initial blood tests included: Hb 8.4 g\%, Hct 26.1 \%, WBC 13,470/mm\(^3\), N 78.5 \%, L 10.9 \%, M 10.1 \%, platelet 455,000/mm\(^3\), MCV 80.6 fl, MCH 25.9 pg, RDW 14.6 \%, normochromic normocytic RBC morphology in the blood smear, no nucleated RBC, no polychromasia, reticulocyte 2.6 \%, ferritin 527.3 ng/ml, serum iron 5.4 ug/dl, TIBC 134.0 ug/dl, Ca 8.6 mg\%, Mg 1.5 mg\%, P 2.8 mg\%, direct anti-globulin test 1+, indirect anti-globulin test 2+, Hb analysis: A\(_2\)A, Hb A\(_2\) 2.8 \%, Hb F 0.4 \%, Hb A 96.8 \%, single tube osmotic fragility test-positive.

FBS 114 mg\%, creatinine 0.58 mg\%, albumin 2.6 g\%, globulin 4.9 g\%, AST 26 U/L, ALT 13 U/L, alkaline phosphatase 61 U/L, direct bilirubin 0.1 mg\%, total bilirubin 0.4 mg\%, normal thyroid function test, morning cortisol 11.4 (normal 2.9-19.4 ug/dl), CPK 41 (normal <145 IU/L)

**Discussion**

Our case was diagnosed as having AIHA solely based on the positive direct anti-globulin test in the normocytic anemic patient although the signs of hemolysis such as the increased indirect bilirubin, the increased polychromasia could not be documented\(^{(6,7)}\). And she seemingly responded rather well to only corticosteroid therapy\(^{(8)}\) even though the concurrent ovarian tumor was not got rid because her Hb level not only did not further diminish but also it was raised from 8.4 to 9.5 g\% within one month of therapy.

AIHA could be occasionally found as a case report in patients with either benign ovarian cyst\(^{(9)}\),
ovarian teratoma\textsuperscript{(10,11)} or ovarian cancer\textsuperscript{(12)}. AIHA in all these cases seem poorly responsive to corticosteroid therapy and it will disappear only after the surgical removal of the ovarian lesions or chemotherapy for cancer successfully. In contrast, our case appeared more responsive to steroid although the ovarian cyst was left intact. With this outcome, AIHA and the ovarian cyst in our case looked like co-incidental more than having any association particularly the paraneoplastic syndrome.

When an ovarian cyst is unilocular with size $<13$ cm and serum CA-125 level $<35$ IU/ml in a postmenopausal woman, the possibility of a benign etiology is most likely. On the contrary, among 16 patients with CA-125 levels between 35 and 50 IU/ml, two cases with a unilocular cyst $>13$ cm and nine cases with multilocular cysts (3 cases $<13$ cm, 6 cases $>13$ cm) had borderline histopathology\textsuperscript{(13)}. Because many benign conditions that could result in higher level of CA 125 such as pregnancy, pelvic inflammatory disease ascites ovarian hyperstimulation syndrome\textsuperscript{(14)} were not found in our case, the combination of the size of the cyst $>15$ cm, multi-locular lesion, and the serum CA 125 $>50$ IU/ml, seemed to favor the malignant pathology in our case.

**Conclusion**

A 72-year-old Thai woman was diagnosed as having an autoimmune hemolytic anemia (AIHA) and a large ovarian cyst. AIHA well responded to cortico-steroid therapy although the ovarian was left intact, so it seemed that AIHA was likely to be the coincidence more than the paraneoplastic syndrome of the ovarian cyst.

**References**

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