



Contents lists available at ScienceDirect

## Taiwanese Journal of Obstetrics &amp; Gynecology

journal homepage: [www.tjog-online.com](http://www.tjog-online.com)

## Research Letter

## Postpartum flare up of systemic lupus erythematosus: Pulmonary diffused alveolar hemorrhage

Hsiao-Wen Lu<sup>a</sup>, Fu-Yun Wang<sup>a, b</sup>, Cheng-Kun Chang<sup>a, c</sup>, Sen-Wen Teng<sup>a, b, c, \*</sup>, Peng-Hui Wang<sup>d, e, f, \*\*</sup><sup>a</sup> Department of Obstetrics and Gynecology, Cardinal Tien Hospital-Hsintien, New Taipei City, Taiwan<sup>b</sup> Department of Midwifery and Women Health Care, National Taipei University of Nursing and Health Sciences, Taipei, Taiwan<sup>c</sup> School of Medicine, College of Medicine, Fu Jen Catholic University, New Taipei City, Taiwan<sup>d</sup> Department of Obstetrics and Gynecology, Taipei Veterans General Hospital, Taipei, Taiwan<sup>e</sup> Department of Obstetrics and Gynecology, National Yang-Ming University School of Medicine, Taipei, Taiwan<sup>f</sup> Department of Medical Research, China Medical University Hospital, Taichung, Taiwan

## ARTICLE INFO

## Article history:

Accepted 15 March 2018

## Dear editor:

Systemic lupus erythematosus (SLE) is a common autoimmune disorder that affects women during pregnancy [1]. The high level of prolactin and estradiol during pregnancy may contribute to lupus flare and associated complications. Diffuse alveolar hemorrhage (DAH) is a rare and life-threatening complication of SLE flare [2]. Here, we report a case of postpartum SLE flare that presents with DAH.

A 38-year-old primigravida woman with SLE had been treated regularly for eleven years. At the 32 + 3/7 gestation weeks, she was hospitalized for the management of severe pre-eclampsia. Emergent cesarean section was done soon because of continuous progression and deterioration of disease and at the same time, an attack of acute fetal distress. Postoperative condition was stable and uneventful. On day 3 postoperatively, she experienced sudden onset of dyspnea. The oxygen saturation dropped and metabolic acidosis developed. Chest X-ray (CXR) showed bilateral pleural effusion. High resolution computed tomography (HRCT) showed ground glass opacities with bilateral perihilar consolidation (Fig. 1).

Bronchoalveolar lavage revealed high red blood cell (RBC) count, which correlated with pulmonary alveolar hemorrhage. Postpartum SLE flare with DAH was impressed. This patient received an intensive care, including endotracheal tubing with mechanical ventilation, high dose of corticosteroid and empirical antibiotics, immediately. Acute renal failure occurred soon, and the patient was treated with plasmapheresis and emergent hemodialysis. After treatment, she recovered completely and discharged one month later.

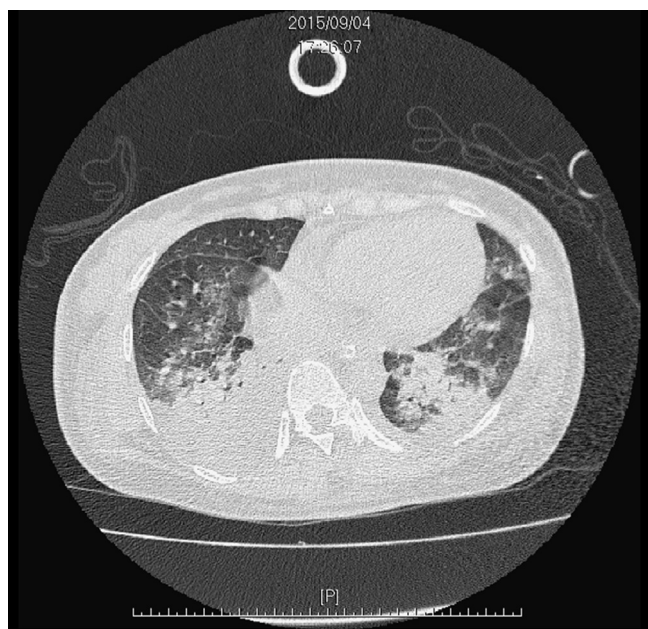
This case was interesting and worthy of further discussion. First, DAH is an extremely rare but severe complication of SLE, with a classic triad, including hemoptysis, rapid fall in hemoglobin over 24–28 h and alveolar or interstitial infiltrates [2], as demonstrated in the current case. Since DAH is often accompanied with acute respiratory failure, hypovolemic shock and sepsis, the mortality rate is very high (ranging from 23% to 92%) [2,3].

Second, it is important to emphasize the need of pre-pregnancy counselling for women with SLE. Pregnant women with SLE have an 18-fold higher risk for maternal mortality, 10-fold higher risk for major thrombosis, such as cerebrovascular accidents, pulmonary embolism or deep vein thrombosis, and 4-fold higher risk for infection [4]. The possible explanations include increased disease burden, the chronic use of immunosuppression rendering SLE women susceptible to infection and the increased pro-coagulant state [4–6]. In the current case, because of its referring characteristics, it is hard to evaluate her disease activity before and during pregnancy. However, severe form of pre-eclampsia accompanied with acute fetal distress of this pregnant woman suggested that SLE might be undertreated [7,8]. Some women with SLE have concerned very much for harmful effects of therapeutic drugs to the fetus [9]. A modest but significant effect of aspirin use to prevent preeclampsia in high-risk women has been suggested in the recent publication [10]. Treatment with low-dose aspirin in women at high risk for preterm preeclampsia can result in a lower incidence of this diagnosis than placebo [10]. To avoid flare up of SLE, we

\* Corresponding author. Department of Obstetrics and Gynecology, Taipei Veterans General Hospital, and National Yang-Ming University School of Medicine, 201, Section 2, Shin-Pai Road, Taipei, Taiwan.

\*\* Corresponding author. Department of Obstetrics and Gynecology, Cardinal Tien Hospital-Hsintien and College of Medicine, Fu Jen Catholic University, New Taipei City, Taiwan.

E-mail addresses: [senwen1955@yahoo.com.tw](mailto:senwen1955@yahoo.com.tw) (S.-W. Teng), [phwang@vghtpe.gov.tw](mailto:phwang@vghtpe.gov.tw) (P.-H. Wang).



**Fig. 1.** Bilateral perihilar consolidation with air bronchogram, along with some ground glass opacities at bilateral upper lobes of the lung.

recommended the consideration of aspirin use in routine for those pregnant women.

Third, the diagnosis and management of DAH is needed to be reviewed. To provide the current evidence in the DAH, we used the following strategy to target this topic and there are only a limited number of publication in the PubMed (<https://www.ncbi.nlm.nih.gov/pubmed/?term=Diffuse+alveolar+hemoorrhage%2C+pregnancy>) [11]. The most common extrapulmonary presentation was renal involvement (100%), which included clinical nephritis, nephritic syndrome or acute renal failure [11]. Characteristic pattern of CXR and HRCT and increased RBC count in bronchoalveolar lavage sample, as shown in our case, can confirm the diagnosis specifically [2,3]. Treatment with combined plasmapheresis and continuous venovenous hemofiltration, in addition to high dose corticosteroid can provide a better chance to save life [8]. Respiratory support and some other intensive therapies, such as the use of intravenous immunoglobulin and tacrolimus or rituximab, extracorporeal membrane oxygenation (ECMO), and human recombinant factor VIIa (rf VIIa) should be taken into consideration without hesitance [2].

We concluded that management of DAH in SLE women should be started early, including intensive immunosuppressive therapy and multiple modalities of extracorporeal organ support to provide a favorable outcome. In addition, pre-conception counseling for these high-risk women should always keep in mind [12].

#### Disclosure of potential conflict of interest

The authors declare that there are no conflicts of interest

#### Acknowledgements

This work was supported by grants from Taipei Veterans General Hospital (V107C-136).

#### References

- [1] Wong CH, Chen TL, Lee CS, Lin CJ, Chen CP. Outcome of pregnancy in patients with systemic lupus erythematosus. *Taiwan J Obstet Gynecol* 2006;45:120–3.
- [2] Kazzaz N, Coit P, Lewis E, McCune W, Sawalha A, Knight J. Systemic lupus erythematosus complicated by diffuse alveolar haemorrhage: risk factors, therapy and survival. *Lupus Sci Med* 2015;2, e000117.
- [3] Knight C, Nelson-Piercy C. Management of systemic lupus erythematosus during pregnancy: challenges and solutions. *Open Access Rheumatol Res Rev* 2017;9:37–53.
- [4] Teng YKO, Bredewold EOW, Rabelink TJ, Huizinga TWJ, Eikenboom HCJ, Limper M, et al. An evidence-based approach to pre-pregnancy counselling for patients with systemic lupus erythematosus. *Rheumatology (Oxford)* 2017 Nov 20. <https://doi.org/10.1093/rheumatology/kex374>.
- [5] Wu LS, Tang CH, Lin YS, Lin CP, Hung ST, Hwa HL, et al. Major adverse cardiovascular events and mortality in systemic lupus erythematosus patients after successful delivery: a population-based study. *Am J Med Sci* 2014;347: 42–9.
- [6] Wang PH, Teng SW, Lee FK. Disease activity of pregnant women with systemic lupus erythematosus. *J Chin Med Assoc* 2015;78:193–4.
- [7] Liu LC, Wang YC, Yu MH, Su HY. Major risk factors for stillbirth in different trimester of pregnancy—a systematic review. *Taiwan J Obstet Gynecol* 2014;53:141–5.
- [8] Lin LT, Wang PH, Tsui KH, Cheng JT, Cheng JS, Huang WC, et al. Increased risk of systemic lupus erythematosus in pregnancy-induced hypertension: a nationwide population-based retrospective cohort study. *Medicine (Baltimore)* 2016;95, e4407.
- [9] Chen YJ, Tseng JJ, Yang MJ, Tsao YP, Lin HY. Acute respiratory distress syndrome in a pregnant woman with systemic lupus erythematosus: a case report. *Lupus* 2014;23:1528–32.
- [10] Rolnik DL, Wright D, Poon LC, O’Gorman N, Syngelaki A, de Paco Matallana C, et al. Aspirin versus placebo in pregnancies at high risk for preterm preeclampsia. *N Engl J Med* 2017;377:613–22.
- [11] Chang MY, Fang JT, Chen YC, Huang CC. Diffuse alveolar hemorrhage in systemic lupus erythematosus: a single center retrospective study in Taiwan. *Ren Fail* 2002;24:791–802.
- [12] Lee YS, Peng MY, Ker CR, Chan TF. Management of pregnancy pancreas alone transplant recipient complicated with stage-4 chronic renal insufficiency and superimposed pre-eclampsia: case report and literature review. *Taiwan J Obstet Gynecol* 2017;56:700–2.