Endometriosis and spontaneous hemoperitoneum in pregnancy: ab uno disce omnes… Is it always true?

In the present issue, Benaglia et al. (1) report one case of spontaneous hemoperitoneum in pregnancy (SHiP) in a series of 348 women with endometriosis who were obtaining a pregnancy by in vitro fertilization (IVF) and conclude that this group of women had an extremely low risk of developing SHiP.

There is no doubt, and it has long been recognized, that SHiP is very uncommon. The first case was described by Doyle et al. (2) in 1957. Since then, several reviews (3, 4) highlighted that this dramatic complication, characterized by the breakage of a pelvic or abdominal vessel, causes massive internal hemorrhage and hypovolemic shock. In a series of 59 cases reported from 2008 to 2016, this disastrous complication was associated with perinatal and maternal mortality rates of 27% and 2%, respectively.

In their review, Brosens et al. (3) identified 45 articles encompassing 64 case reports and noted that 24 (38%) of the 64 selected cases occurred in pregnancies of women undergoing IVF, of whom 22 had endometriosis. They highlighted that a majority of patients experiencing SHiP in the IVF group had moderate or severe endometriosis. They concluded that severe endometriosis could be a risk factor for the development of SHiP during pregnancy after IVF, affecting the choice of treatment and suggesting preventive measures. The arguments for this possible link were the presence of multiple bleeding sites, the occurrence of decidualization, and sites of decidualization, which largely involved the parametrium and endometriomas. Pathologic evaluation of the bleeding sites frequently revealed decidualized stromal cells, and it was postulated that after controlled ovarian stimulation and embryo transfer, high, nonphysiologic progesterone levels in early pregnancy could exacerbate the decidualization process (3).

Porpora et al. (5) observed more obstetric complications during pregnancy in a series of 145 women with endometriosis than in a control group of 280 patients. They described only one case of SHiP, which was due to the probable rupture of an ovarian endometrioma in the third trimester of pregnancy, thus confirming that this complication is unusual.

The aim of the study by Benaglia et al. (1) was to provide more precise counseling to women with endometriosis requiring IVF. Their study observed a low incidence of SHiP but did not offer new insights into the pathophysiology of SHiP. The authors reported a single case of bleeding occurring soon after selective abortion of one fetus in a twin pregnancy, and a possible relationship with this procedure cannot be excluded. Of course, as mentioned by the authors, if the selective abortion had played a role, the frequency of SHiP would be overestimated in their study, and therefore the conclusion that this event is exceedingly rare would remain unchanged and a risk below 1.5% would seem too low to justify any prophylactic measures.

Nevertheless, if we consider that SHiP is extremely rare, a retrospective study evaluating a relatively small series (n = 348) may represent a source of bias. As suggested by Brosens et al. (3) and Lier et al. (4), the risk could be even higher in women with deep endometriotic lesions. Benaglia et al. (1) also noted that the risk was substantially higher in women presenting with deep lesions, but no precise estimate of the risk could be made due to the small number of patients in their series. Moreover, as stressed by the authors themselves, their study was monocentric, and inferences from findings obtained in a single center could be exposed to criticism. Another criticism of the Benaglia et al. study is the lack of histologic confirmation of endometriosis in one third of cases, even though the authors argue that the accuracy of sonographic diagnosis of endometriosis is well established. Another limitation is that risk factors for SHiP were not evaluated. The analysis of one case report obviously cannot offer further explanation.

Therefore, we recommend that these very experienced and competent healthcare providers initiate a national or, even better, a multinational study with a high number of patients with SHiP to identify factors associated with an elevated risk of SHiP. Furthermore, deep endometriosis is a major source of concern, and we agree with the authors that risk factors should be evaluated and prophylactic surgery should be discussed in this group of patients receiving counseling for infertility.

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