INTRODUCTION

First reviewed by Irving and Hertig in 1937, the term placenta accreta was defined as “an abnormal adherence of all or part of the placenta to the underlying uterine wall”. Histopathologically it may be classified into three types according to the degree of invasion into the myometrium: placenta accreta vera, when the chorionic villi adhere to the myometrium; placenta increta, when the chorionic villi penetrate the myometrium; and placenta percreta, when these villi reach the uterine serosa. Heterogeneous definitions, with

Abbreviations:

FIGO - International Federation of Gynecology and Obstetrics
ICU - Intensive care unit
MRI - Magnetic resonance imaging
NICU - Neonatal intensive care unit
PAS - Placenta Accreta Spectrum disorders

Overview and aims: Placenta Accreta Spectrum disorders are the leading cause of emergency hysterectomy in developed countries and are associated with high maternal morbidity and mortality (60% and 7%, respectively). The objective of our study was to review cases of PAS disorders occurring in the last 8 years (2010-2017) in a tertiary care centre, evaluating the forms of treatment chosen and the maternal and fetal outcomes.

Methods: Retrospective cross-sectional study using hospital databases (Obscare®, SClinico®) to identify cases of placenta accreta during the study period. Diagnosis was made based on the histological study of placentas submitted for anatomopathological study.

Results: During the study period, 38 cases of a PAS disorder were diagnosed in 20,910 births (1.8 / 1000), a number that is possibly underestimated. There were 8 women with a previous caesarean section and 6 had placenta previa (2 of them with previous caesarean section). There was ultrasound suspicion of a PAS disorder in 3 cases; of the 4 most severe cases (3 incretas and 1 percreta), there was an ultrasound suspicion in 2. Regarding method of delivery, 21 births were by caesarean section and 16 by vaginal delivery. There was 1 case of spontaneous uterine rupture at 20 weeks’ gestation with fetal loss; gestational age of birth varied between 32 and 41 weeks; 5 peri-partum hysterectomies were performed; transfusions of blood derivatives were required in 8 women. As for the newborns, there were 8 cases of prematurity and 7 hospitalisations in the Neonatal Intensive Care Unit; fetal death was observed in 2 cases.

Conclusion: This case series demonstrate the high prevalence of PAS disorders and the great morbidity associated with it. Adequate antepartum care is essential in women with risk factors to make the diagnosis timely, thus enabling a multidisciplinary approach with better maternal and fetal outcomes.

Keywords: Placenta Accreta; Placenta Accreta Spectrum Disorders; Postpartum Haemorrhage; Postpartum Hysterectomy.
misleading terms and lack of adequate pathological correlation make adequate evaluation of its prevalence, accuracy of diagnosis and outcomes difficult. In an effort to standardise, in February 2018 the International Federation of Gynecology and Obstetrics (FIGO) published consensus guidelines, proposing the classification as Placenta Accreta Spectrum disorders (PAS), involving both the abnormally adherent placenta (placenta accreta) and the invasive placenta (increta and percreta). This classification has also recently been endorsed by the American College of Obstetricians and Gynecologists, the Society for Maternal-Fetal Medicine and the Royal College of Obstetricians and Gynaecologists.

The most cited article regarding PAS incidence, from 2005, describes it as 1 per 533 deliveries in the American population. Importantly, its incidence has been rising considerably in recent years, following the rise in caesarean section rates. In theory, the presence of a uterine scar, with disruption of the endomyometrial wall, whether caused by a caesarean section, probably the most important risk factor, or by a gynaecological procedure (such as curettage, myomectomy or hysteroscopy) is essential for the development of PAS. According to a decision-analytic model, if the caesarean rates in the United States of America continued to rise, by 2020 there would be an additional 4,504 cases of PAS disorders, and consequently 130 attributable maternal deaths annually. Another risk factor is the presence of a low-lying placenta. Placenta previa, without previous uterine surgery, is associated with a risk of 1%–5% for PAS. According to Silver, RM. et al., the risk of PAS ranges from 2% among women with a placenta previa, to 39%–67% when it is accompanied by two or more prior caesarean deliveries.

PAS is currently recognised as the leading cause of emergency hysterectomy in developed countries, and is associated with a maternal morbidity and mortality of 60% and 7%, respectively, mainly due to excessive blood loss. The average blood loss volume at delivery is 3,000–5,000 mL. Prenatal suspicion of PAS disorders enables planned surgery to be performed with the support of a multidisciplinary team to ensure lower maternal and fetal morbidity.

In Portugal, PAS disorders are generally under-reported and unrecognised prenatally. Between 2010 and 2016, caesarean section rates in public institutions, ranged from 32.9% to 36.3%, numbers that make our country highly susceptible to PAS disorders. We found no adequate report from any national institution regarding cases of PAS disorders, so we performed this study in order to estimate the prevalence of the PAS disorders in our institution, the adequacy of its diagnosis and maternal and fetal morbidity in the last 8 years in our centre.

METHODS

We conducted a retrospective medical record review of all PAS disorder cases diagnosed in our institution (a tertiary centre) from 2010 to 2017. Cases were identified by using hospital-based electronic databases (Obscare®, Sclinico®), with cross-referencing from different databases and multiple key terms searched (low-lying placenta; manual removal of the placenta; parturition hysterectomy; postpartum haemorrhage). All the cases included in our study had a pathological diagnosis of a PAS disorder, the majority of them in studies of the placenta and some in hysterectomy specimens. PAS disorders were diagnosed by the presence of myometrial tissue adjacent to the chorionic villi (with immunohistochemical confirmation) with focal interruption of the decidua basal and were classified as placenta accreta, increta or percreta as previously described. The placentas were sent for pathological study according to the attending medical team’s decision. As a consequence, the diagnosis of a PAS disorder was made by combining clinical characteristics (abnormal adherence of the placenta) with confirmation by pathologic examination of the placenta.

It is important to state that the majority of the women had pregnancy surveillance outside the hospital, with most of the ultrasounds (except the first- and second-trimester ultrasounds) performed in other centres. Only women with recognised risk factors (like placenta previa) were followed up in our centre.

Medical records were analysed and data collected included demographic data (age, body mass index, parity; mode of conception); risk factors for PAS disorders (placenta previa, prior caesarean section and number, history of other uterine surgery); timing of diagnosis, antepartum and intrapartum management; gestational age at delivery; birthweight, neonatal intensive care unit (NICU) admission, Apgar scores and umbilical cord blood pH (when applicable); metabolic acidosis defined according to FIGO consensus 2015 - arterial pH < 7,00 and base deficit > 12 mmol/L; intermediate maternal morbidity; recurrence of PAS disorders. Early and delayed re-operations were defined as...
urgent surgical procedures performed during the first 7 days after birth/spontaneous abortion or after 7 days, respectively. Early maternal morbidity was defined as the occurrence of one or more of the following: maternal admission to the intensive care unit (ICU), transfusion of packed red blood cells, coagulopathy, ureteral or bladder wall injury or early re-operation. Late maternal morbidity was defined as the occurrence of one or more of the following: hospital re-admission within 6 weeks or delayed re-operation. Hysterectomy was classified as scheduled if the surgery was performed by a multi-disciplinary team after appropriate planning in cases with antenatal suspicion and when deemed adequate by the obstetrical team; urgent, when there was high suspicion of a PAS disorder during caesarean section and there was no significant haemorrhage; emergent when the obstetrical team was faced with severe and refractory haemorrhage and hysterectomy was considered as a final solution. During the study period there were no cases treated by a conservative approach, where the placenta is left in situ or a segmental resection of the myometrium is performed.

As a consequence of the design of the study, no statistical analysis was performed.

RESULTS

Thirty-eight cases of PAS disorders were identified between January 2010 and December 2017 in a total of 20,910 births, with a prevalence of PAS of 1.8/1000 deliveries over the 8-year study period. There were thirty-four cases of placenta accreta, one of placenta increta and three of placenta percreta. Fourteen women were primiparous, the majority (10) of them without any identifiable risk factors. Eight women had previous caesarean sections (one woman with two previous caesarean sections, the remaining with one) and seven with a low-lying placenta (three also with previous caesarean section). There were four pregnancies after assisted reproduction techniques. Two women had been submitted to a hysteroscopic procedure – one myomectomy and one septotomy. Other risk factors as well as basic characteristics of the population are summarised in Table I. All women had at least one antenatal ultrasound performed. There was no antenatal suspicion of a PAS disorder in the majority of the cases - only three cases were suspected: one case of a placenta accrete (suspicion during the third trimester), one of an increta (also suspected during the third trimester) and one of a percreta, suspected already in the first trimester and confirmed on the second trimester ultrasound (Figure 1). However, in the most serious cases (three placenta percreta and one increta), ultrasound signs suggestive of a PAS disorder were present in two (half of the cases). Only two cases had magnetic resonance imaging (MRI) performed, also confirming the ultrasound suspicion of a PAS disorder. The gestational age of birth ranged from 32-41 weeks of gestation (median of 39 weeks). There was intrapartum suspicion of a PAS disorder in 25 patients;

<table>
<thead>
<tr>
<th>TABLE 1. PATIENT CHARACTERISTICS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Maternal age at delivery, years</td>
</tr>
<tr>
<td>Primiparous</td>
</tr>
<tr>
<td>Assisted reproductive techniques</td>
</tr>
<tr>
<td>Gestational age, weeks</td>
</tr>
<tr>
<td>Previous cesarean section</td>
</tr>
<tr>
<td>Zero</td>
</tr>
<tr>
<td>One</td>
</tr>
<tr>
<td>Two</td>
</tr>
<tr>
<td>Risk Factors</td>
</tr>
<tr>
<td>Previous PAS disorder</td>
</tr>
<tr>
<td>Previous cesarean sections</td>
</tr>
<tr>
<td>Placenta previa</td>
</tr>
<tr>
<td>Previous hysteroscopic procedure</td>
</tr>
<tr>
<td>Submucous myoma</td>
</tr>
<tr>
<td>Previous manual placental removal</td>
</tr>
<tr>
<td>Previous dilation and curettage</td>
</tr>
</tbody>
</table>

FIGURE 1. Placenta previa with lacunae and aberrant vascularisation of the cervix
other cases were diagnosed after pathologic examination of the placenta in cases of difficult manual removal of the placenta. The majority of births (21) were by caesarean section: twelve scheduled and nine urgent/emergent. There were 16 vaginal deliveries, all of them with manual placental removal. Table II reports peripartum characteristics of the study population. All the cases with antenatal suspicion of PAS disorders were delivered by scheduled caesarean section – attempts were made to deliver the placenta in two of them, one ending up in emergent hysterectomy. A peripartum hysterectomy after caesarean section with the placenta left in situ was performed in the other case. Five cases ended up with a hysterectomy (Figure 2).

Regarding the hysterectomies: a) in 2010 there was one case of a woman with a vaginal delivery who had only one identified risk factor (a previous dilation and curettage). After an attempt to manually remove the placenta, the patient started bleeding massively, refractory to medical attitudes and ended up with an emergent laparotomy performed with hysterectomy for bleeding control; pathology revealed a placenta percreta; b) there was one case, also in 2010, of a suspected PAS disorder during caesarean section due to placenta previa; no other risk factors were identified; an attempt to remove the placenta was made despite the intrapartum suspicion of an abnormally adherent placenta; hours after the caesarean the woman developed a coagulopathy and ended up with an emergent hysterectomy; pathology revealed a placenta percreta; c) during the year 2012, another woman had a scheduled caesarean section due to placenta previa with ultrasound suspicion of a PAS disorder; as risk factors, the woman had two hysteroscopic myomectomies and a pregnancy after assisted reproductive techniques; the placenta was forcibly removed and due to uncontrollable bleeding, and an emergent hysterectomy was performed; the patient had an iatrogenic ligation of the ureter and a laceration of the bladder, immediately corrected during surgery; pathology revealed a placenta increta; d) in 2013 there was a case of a woman with two previous caesarean sections and a placenta previa, with antenatal suspicion of a placenta percreta (by ultrasound and MRI); the patient had a scheduled caesarean hysterectomy, without any attempt of placental removal (fundal incision), with ureteral catheterisation and balloons in the common iliac arteries (inflated during surgery); surgery was performed by a multidisciplinary team – anesthesia, obstetrics, gynaecological oncology, urology and interventional radiology; deliberate cystotomy was required due to placental extension into the bladder wall; pathology revealed a placenta percreta; e) lastly, in 2015, another woman with a previous cesarean section and no other risk factors had a scheduled caesarean section; during surgery the placenta was found to be abnormally adherent and, as the patient desired definitive contraception, a hysterectomy without placental removal was decided; the surgery had no major morbidity; pathology revealed placenta accreta.

Table III summarises the maternal and neonatal morbidity in the entire cohort. There were 14 cases of postpartum haemorrhage; we found no maternal deaths due to a PAS disorder during the period of study. There was one re-admission, at the 10th day post-partum, requiring two suction curettages for complete removal of placental fragments. There were two unex-
Considered intrauterine fetal deaths that occurred before labour: one after a uterine rupture at 20 weeks of gestation, in a woman with a previous scheduled caesarean section and a hysteroscopic septotomy. This woman had a fundal placenta, probably inserted in the scar of the septotomy, and presented in the emergency department with haemoperitoneum, secondary to uterine rupture. The patient was submitted to emergent surgery, with delivery of a non-viable fetus and uterus reconstruction. The other fetal death was secondary to a rupture of a non-diagnosed vasa previa at 32 weeks of gestation.

There were two pregnancies after a diagnosis of a placenta accreta; in one of these cases there was recurrence, with another placenta accreta diagnosed.

**DISCUSSION**

During the 8-year study period, the overall prevalence of a PAS disorder in this tertiary centre was 1.8/1000 deliveries, in accordance with the published literature

...
removal of the placenta even though there was suspicion of a PAS disorder in 28 cases (25 intrapartum + 3 antepartum): in three cases it ended up in emergent hysterectomies with the associated morbidity. The ex-tirpative technique is largely contraindicated, especially when there is a high suspicion of a PAS disorder and the decision on caesarean hysterectomy had already been taken\textsuperscript{11,27,28}. Since 2014, with the creation of a specific guideline for this entity in our centre, when there is a suspicion of a PAS disorder, it is recommended no attempt be made to forcibly remove the placenta. The last hysterectomy performed during a caesarean section, in 2015, followed this recommendation and the patient ended up without significant bleeding and without the need for a blood transfusion. Some other options are reported in the literature, from the traditional approach of leaving the placenta \textit{in situ} to some conservative surgical procedures\textsuperscript{29-32}. In the period of study, there were no cases of conservative approaches where a segmental resection of the myometrium is performed or the placenta is left \textit{in situ}. Regarding conservative surgical procedures, it is still an option limited to some centres but not generalisable to other hospitals without such surgical experience. In the future, this technique can also be an option in selected cases, as the authors report significantly lower morbidity\textsuperscript{29,31,32}. Leaving the placenta \textit{in situ}, with a success rate ranging from 58-78% depending on the degree of invasion\textsuperscript{30,39}, is associated with important morbidity: primary postpartum haemorrhage, infection/sepsis and need for secondary surgical procedures including emergent hysterectomy. Complete placental resorption may take up to 60 weeks, making careful follow-up essential\textsuperscript{34}. There is also concern for the risk of recurrence of a PAS disorder in women who undergo conservative management. In our series of cases, two women became pregnant after a previous PAS disorder, with recurrence in one of the cases.

Our study has some weaknesses. First, our sample size and the design of our study make it impossible to perform any statistical analyses. It is a retrospective descriptive study with the inherent limitations. Second, a standardised protocol only became available in the last years of the study, with the first cases managed according to individual clinical criteria. Third, all our cases were managed in a tertiary care centre. Adequate blood banks, 24-hour anaesthesia coverage and specialists in maternal fetal medicine and gynaecological oncology and other specialities (Urology, Surgery, Interventional Radiology) were readily available. Thus, our data may not be generalisable to other centres with fewer resources, increasing the likelihood of even greater morbidity with the management of PAS disorders in those hospitals.

However, our study has some important strengths. Thorough research was done with multiple databases in order to try to gather all the possible cases during the period of study. Our centre has all the data in computer databases providing all the information necessary to complete the study. All the cases had pathological classification, making our estimate of the prevalence as close as possible to the real value. Although the definition of accreta remains controversial, we used the most commonly agreed definition: a placenta that is abnormally adherent to the uterus and with clinical features of a PAS disorder. It is somewhat different from the proposal of the FIGO consensus, which places less emphasis on the abnormally adherent placenta, but we believe this reflects the reality and the morbidity associated with this condition as it occurs in clinical practice more adequately\textsuperscript{3}. The morbidity from our cases strengthens the importance of any type of a PAS disorder.

Our series of cases report the great morbidity associated with any form of a PAS disorder. The prevalence in our centre is higher than previously assumed. It is important to be more attentive during the antenatal period in order to make the prenatal diagnosis, allowing planned multidisciplinary surgery. Our findings emphasise the importance of antenatal diagnosis for a PAS disorder and the need for increased attention to any possible risk factor in order to evaluate the placenta more adequately and optimise the care of these women. The best treatment mode will depend on the centre, the expertise of clinicians, the urgent/scheduled situation and the number of cases dealt with during a year. By changing our policy and focusing on antenatal diagnosis of a PAS disorder we believe our results will be better in the future.

REFERENCES

5. Jauniaux E, Alfirevic Z, Bhide AG, Belfort MA, Burton GJ,


Endercimento para correspondência
Pedro Viana Pinto
Centro Hospitalar São João
Portugal
E-mail: pedrovianapinto@gmail.com

Recebido em: 23/10/2018
Aceite para publicação: 18/12/2018