# Le $oldsymbol{B}$ ulletin de la $oldsymbol{D}$ ialyse à $oldsymbol{D}$ omicile

### SIAMESE TWIN PREGNANCY IN PERITONEAL DIALYSIS: FIRST CASE IN THE WORLD

## GROSSESSE GEMELLAIRE SIAMOIS EN DIALYSE PERITONEALE : PREMIER CAS MONDIAL

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D.ElHamssili, T.Bouattar, S.Benbekha, R.Bayahia, L.Benamar

Service de Néphrologie – Dialyse – Transplantation Rénale. CHU Ibn Sina, Rabat. Faculté de Médecine et de Pharmacie de Rabat- Université Mohammed V, Rabat. Maroc

#### Résumé

Introduction: La probabilité de conception est faible en dialyse péritonéale (DP), et la probabilité de réussir la grossesse est encore plus faible. Près de 60 ans après le premier cas rapporté de grossesse réussie chez une patiente dialysée, de nombreuses questions concernant la grossesse en dialyse restent non résolues, à savoir la dose de dialyse nécessaire, le rythme de suivi, le choix de la technique de dialyse la plus appropriée.

Observation: Nous rapportons le premier cas mondial d'une grossesse gémellaire siamoise chez une patiente en dialyse péritonéale continue ambulatoire (DPCA). La grossesse a été diagnostiquée à 7 semaines d'aménorrhée (SA). La patiente était initialement sous 2 échanges par jour, par des poches de Dianeal de 2 litres de 1,36% de glucose. Dès la découverte de la grossesse, la prescription a été modifiée, mettant la patiente sous 3 échanges par jour par les mêmes concentrés. Le volume d'infusion a été maintenu à 2 litres jusqu'à 18 SA, puis il a été réduit à 1,5 litres du fait de la gêne ressentie par la patiente et de l'hyperpression abdominale. Les ultrafiltrations quotidiennes variaient entre 180 et 800 ml avec une diurèse à 1 litre. La pression artérielle était bien contrôlée, avec des chiffres en moyenne de 125/80 mmHg, sans

Mots clés : Dialyse péritonéale, Grossesse gémellaire siamoise, Âge maternel avancé

Abréviations:

PD: Peritoneal dialysis

CAPD : Continuous Ambulatory peritoneal dialysis APD : Automated peritoneal dialysis (on machine)

RRF : Residual renal function ESRD : end stage renal desease

IUFD: intrauterine foetal deathIUGR: Intauterine Growth

Retardation

WA: week anmenorrhea

recours aux traitements antihypertenseurs. Sur le plan biologique, le Kt / V était de 2,2, le nPCR était de 0,8, et la clairance hebdomadaire à 80 L/1.73 m<sup>2</sup>. L'hémoglobine moyenne était de 11.5 g/dl sous érythropoïétine 9000 unités par semaine. A 20 SA, l'échographie obstétricale a mis en évidence une grossesse gémellaire mono-choriale, mono-amniotique. Vu les risques maternel et fœtal élevés, un contrôle obstétrical plus rapproché a été indiqué. A 24 SA, l'échographie obstétricale morphologique couplée au doppler des artères utérine et ombilicale était en faveur d'une grossesse gémellaire monochoriale monoamniotique avec forte suspicion de siamois accolés par le pelvis, avec une malformation cérébrale chez un des deux fœtus. La patiente a présenté des contractions et douleurs pelviennes intenses menant à une césarienne en urgence avec issue de deux jumeaux siamois vivants, accolés par le pelvis, pesant 900g, décédés un jour après l'accouchement.

Conclusion: la grossesse en dialyse est à hauts risques maternel et fœtal. Il est nécessaire de préparer et de suivre cette grossesse pour en optimiser les chances de réussite. Ceci sous-entend une adaptation du traitement médical et de la prescription de dialyse, notamment le volume et la modalité des échanges. Il en va de même pour le suivi gynécologique et obstétrical qui doit être régulier et strict, afin de prévenir toute complication materno-fœtale.

#### Abstract

**Introduction:** The probability of conception is low in peritoneal dialysis (PD), and the probability of successful pregnancy is even lower. Nearly 60 years after the first reported case of successful pregnancy in a dialysis patient, many questions about pregnancy during dialysis remain unresolved, namely the required dialysis dose, the follow-up rhythm, the choice of the the most appropriate dialysis technique.

Case report: We report the first case worldwide of twin Siamese pregnancy in a patient on continuous ambulatory peritoneal dialysis (CAPD). The pregnancy was diagnosed at 7 weeks of amenorrhea (WA). The patient was initially under 2 exchanges per day, with 2-liter bags of Dianealof 1.36% glucose. As soon as the pregnancy was discovered, the prescription was modified, putting the patient under 3 exchanges a day with the same concentrates. The infusion volume was maintained at 2 liters up to 18 WA, and then reduced to 1.5 liters due to patient discomfort and abdominal hyperpressure. Daily ultrafiltration ranged from 180 to 800 ml with 1 liter diuresis. The blood pressure was well controlled, with an average of 125/80 mmHg, without need

Keywords: siamese, pregnancy, peritoneal dialysis

#### INTRODUCTION

Dialysis pregnancy is a rare event. It can be responsible for serious maternal-fetal complications. Nevertheless, in recent years, there has been an increase in the frequency of pregnancy and an improvement in its prognosis. The first case of dialysis completed pregnancy was reported in Italy by Confortini [1], and more and more cases of successful dialysis pregnancies are reported in the literature. The occurrence of a pregnancy in peritoneal dialysis (PD) is rarer and the probability that it is twin is exceptional. Since the launch of this technique, a few cases of peritoneal dialysis mono-fetal pregnancy have been reported. We report the experience of our center in the management of the first case worldwide of twin Siamese pregnancy in a patient in CAPD.

#### **OBSERVATION**

This patient is 41 years old, followed since 2017 for end-stage renal failure (ESRD) by accidental discovery, secondary to indeterminate nephropathy and treated by CAPD with two liters Dianeal PD4 1.36% glucose twice a day.

Clinically: Blood pressure was normal at 120/80 mmHg, with no edema of the lower limbs with an average weight gain of 800g per month. Diuresis was preserved at 1 liter / day with a residual renal function estimated at 3.8 ml / min.

of antihypertensive drugs. On a biological level, the Kt / V was 2.2, nPCR was 0.8, and the weekly clearance was  $80\,L/1.73\,m^2$ . The average hemoglobin was  $11.5\,g$  / dl under erythropoietin 9000 units per week. At 20 WA, obstetrical ultrasound revealed a mono-chorionic, mono-amniotic twin pregnancy. Due to high maternal and fetal risks, closer obstetrical control was indicated. At 24 WA, morphological obstetrical ultrasound coupled with Doppler examination of the uterine and umbilical arteries favored the diagnosis of mono-amniotic mono-chorionic twin pregnancy with strong suspicion of Siamese contiguous to the pelvis, with a cerebral malformation in one of the fetuses. The patient experienced intense contractions and pelvic pain leading to an emergency cesarean section with two surviving Siamese twins, contiguous to the pelvis, weighing 900g, but who died one day after delivery.

**Conclusion:** Pregnancy during dialysis is at high maternal and fetal risks. It is necessary to prepare and follow this pregnancy to optimize the chances of success. This implies an adaptation of the medical treatment and dialysis prescription, including the volume and modality of exchanges. The same holds true for gynecological and obstetrical follow-up, which must be regular and strict, in order to prevent any maternal-fetal complications.

Dialysis: Daily ultrafiltration ranged from 0 to 200 ml / day, KT / V was 1.5, weekly creatinine clearance was 70 L/1.73 m<sup>2</sup>, and nPCR was 0.5.

On the obstetrical side, the patient had three gestures and two parities. The first two pregnancies proceeded without complications and the renal function was normal.

The third pregnancy occurred after 7 months of oral contraceptive with micro progestin while being on peritoneal dialysis. She was diagnosed at 7 weeks of amenorrhea, suspected due to late menstruation. A B-HCG assay was positive, but it was not reliable; the pregnancy was confirmed by an endovaginal ultrasound showing the existence of a gestational sac with cardiac activity.

Our attitude was to follow the patient in consultation each week, to monitor clinical parameters, and perform a blood test, urine and dialysate tests every two weeks. We increased the number of Dianeal 1.36% glucose bags to 3 exchanges per day with an infusion volume maintained at 2 liters. At 16 weeks of amenorrhea, due to the progressive increase in intra-abdominal pressure, the infused volume was reduced to 1.5 liters and a transition to automated peritoneal dialysis was planned at 24 weeks of amenorrhea.

Medically, the patient received 6g of calcium carbonate per day, 1g of furosemide, 20 mg of omeprazole and 9000 IU erythropoietin per week. Platelet antiaggregant

was prescribed at a dose of 75 mg per day as soon as the diagnosis of pregnancy was made, to avoid the risk of pre-eclampsia.

On a biological level, the blood urea level was on average 0.8 g / l with a KT / V at 2.2, a weekly clearance of creatinine at 80 L / 1.73 m<sup>2</sup> and an nPCR at 0.79. The average hemoglobin level was 11.5g / dl. On the obstetrical side, the patient had a consultation every two weeks with ultrasound control. At 20 weeks of amenorrhea, obstetrical ultrasound showed a mono-chorale, mono-amniotic pregnancy. Due to high maternal and fetal risks, a closer obstetrical control was indicated. At 24 weeks of amenorrhea, a Doppler-associated morphological obstetric ultrasound of the uterine and umbilical arteries showed monoamniotic monochorinal twin pregnancy, with strong suspicion of Siamese contiguous to the pelvis, with a cerebral malformation in one of the two fetuses. The patient experienced intense contractions and pelvic pain the same day, requiring hospitalization in the Gynecology Department and emergency cesarean section, giving birth to live Siamese twins, contiguous to the pelvis, weighing 900 g, who died one day after delivery (Figure 1).



Fig. 1: Siamese twins alive, contiguous to the pelvis, weighing 900 g, died one day after childbirth.

Temporarily, the patient was placed on intermittent hemodialysis for one month until wound healing of caesarean section, then she was resumed peritoneal dialysis with the same prescription: two exchanges of 2 liters bags of Dianeal PD4 at 1.36% glucose with still 1.5 liter urine volume and residual renal function at 4 ml/min.

#### DISCUSSION

The first successful pregnancy in a dialysis patient was reported 47 years ago by Confortini [1]. However, the first sustained pregnancy in peritoneal dialysis was only reported 12 years later in a woman who conceived after 30 months of CAPD. Pregnancy was maintained until 30 weeks of amenorrhea, then the patient delivered a stillborn child [2].

Over time, the rate of pregnancy success increased from 23% in 1980 - reported by the European Dialysis and Transplantation [3] association to more than 70% in 2008 reported by some case series [4, 5]. Pregnancy in women with ESRD is relatively difficult because fertility is significantly reduced due to anovulation and amenorrhea, associated with decreased libido. It is therefore not surprising that the conception rate is low at around 0.3- 4.1% [6, 7].

ESRD patients have higher rates of morbidity and mortality during pregnancy, either in the mother or the fetus compared to women with normal renal function [8]. PD and hemodialysis are the two methods of replacing kidney function in pregnant women with ESRD. Although hemodialysis allows for more precise control of water-overload, it can lead to marked hemodynamic instability and arterial blood pressure fluctuations that can affect placental blood flow. PD, on the other hand, provides a less aggressive, continuous dialysis mode with fewer variations in maternal blood volume.

It is not easy to complete a hemodialysis pregnancy, because patients must accept to to be dialysed at night for up to 8 hours per night or have daily sessions of 4 hours. This intensive hemodialysis schedule is one of the main reasons for improving the results seen over the last decade. We report the experience of our service regarding pregnancy in hemodialysis for 10 years. The mean age at conception was 34 years and the average duration of dialysis before conception was 76 months. The diagnosis of pregnancy was made on average at 10.4 weeks of amenorrhea. Among 23 patients of childbearing age, we recorded 11 pregnancies in 8 patients. The evolution was marked by the fetal in-utero death in one case, the therapeutic interruption of pregnancy in a second case and four spontaneous abortions. No maternal complications were found. In the long term, fetal psychomotor development was normal [9]. According to Okundaye et al, the occurrence of pregnancy is more common in women on hemodialysis than in those on PD (2.4% vs. 1.1%). However, the chances of a successful pregnancy remain better in PD than in HD [10]. This is due to several factors, namely: the persistence of a higher residual renal function, a more stable metabolic environment and the absence of intradialytic hypotension that could potentially cause IUGR and IUFD. Because PD depends on the peritoneal dialysis catheter, increasing the size of the uterus during pregnancy can alter the position of the catheter and reduce the effectiveness of the prescribed therapy [11].

There is also uncertainty regarding the recommended target for pregnant patients on peritoneal dialysis. It has been suggested that Kt / V for peritoneal dialysis patients should be 2.2 to 2.4 for better pregnancy outcomes, but this remains to be proven. To reach this Kt / V target, a treatment volume of up to 20 L per day has been recommended [12]. This approach was not practiced in our patient because of the high cost of therapy and the unavailability of an APD machine. Most nephrologists rely on clinical and laboratory parameters to monitor and adjust the PD prescription as needed. In addition, a closely coordinated follow-up between the treating nephrologist and the obstetrician specializing in high-risk pregnancies remains crucial [13].

#### **CONCLUSION**

Pregnancy on dialysis is at high risk of maternalfetal complications, but the prognosis has changed considerably in recent years. This is due to better management and monitoring of this population. It is no longer lawful to formally contraindicate pregnancy in PD. However, it is necessary to prepare the pregnancy to optimize the chances of success by adapting the drug treatment, the volume of exchanges and their modality.

#### **CONFLICTS OF INTEREST**

the authors declare that they have no conflict of interest in this article.

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