

# Sirenomelia in twin pregnancy

## A case report and literature review

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### Abstract

**Rationale:** Sirenomelia is a very rare congenital malformation and characterized by fused lower extremities, oligohydramnios, renal agenesis, absent urinary tract and external genitalia, single umbilical artery, and imperforate anus. Ultrasonography is an optimal method for prenatal screening and diagnosis of sirenomelia. The incidence of sirenomelia in the twin pregnancy is extremely low.

**Patient concerns:** We reported a case of 1 twin with sirenomelia in dichorionic-diamniotic twin pregnancy after in vitro fertilization and embryo transfer.

**Diagnoses:** The sirenomelia twin was diagnosed at the 2nd trimester by ultrasonic examination and complicated with oligohydramnios and a single umbilical artery, another twin was normal.

**Interventions:** A regular and careful antenatal care was conducted. The parents refused to examine the chromosome of sirenomelia twin, and the chromosomal microarray analysis of the amniotic fluid sample was only achieved in the normal anatomy twin after extensively counseled by the multi-disciplinary team.

**Outcomes:** At 34+2 gestational weeks, the demise of the malformed twin occurred, while fetal heart rate monitoring of the normal twin was abnormal, and an emergency cesarean section was performed. A healthy male baby was delivered with Apgar scores of 10 and 10 at 1 and 5 minutes, respectively. The mother and the baby were followed up and are in good health until now.

**Conclusion:** Sirenomelia is a lethal condition in the perinatal period. Early antenatal diagnosis is very important. Voluntary selective termination of sirenomelia 1 in twin pregnancy may be advised. Expecting parents should be counseled by the multidisciplinary team about the management and prognosis of the sirenomelia.

**Abbreviations:** IVF-ET = in vitro fertilization and embryo transfer, HIV = human immunodeficiency virus, VDRL = venereal disease research laboratory, HBsAg = hepatitis B surface antigen, NICU = neonatal intensive care unit, Bmp-7 = bone morphogenetic protein-7, IUGR = intrauterine growth retardation, MRI = magnetic resonance imaging.

**Keywords:** literature review, sirenomelia, twin

### 1. Introduction

Sirenomelia, also called sirenomelia sequences, mermaid syndrome, or mermaid malformation, is a very rare congenital malformation. It is characterized by complete or incomplete fused lower extremities, renal agenesis, oligohydramnios, absent

urinary tract and external genitalia, single umbilical arteries, imperforate anus, etc.<sup>[1]</sup> It is a lethal condition in the perinatal period. The survival and quality of life in sirenomelia live birth is extremely poor due to multiple congenital anomalies. The reported percentage of elective termination of pregnancy for the fetal anomaly is about 49.5%.

The proportion of born alive, premature, and weight <2500 g in sirenomelia was 47%, 71.2%, and 88.2%, respectively.<sup>[1]</sup> More than half of sirenomelia was stillbirth and the born alive sirenomelia usually die within 1 or 2 birthdays for abnormal kidney and bladder complications.<sup>[2]</sup> Only 1% of cases can survive the 1st week after birth.<sup>[1]</sup>

The incidence of sirenomelia in the twin pregnancy is extremely low. However, compared with singleton pregnancy and dizygotic twins, the incidence of sirenomelia in the monozygotic twin is more than 100-fold.<sup>[3,4]</sup> Therefore, the diagnosis and management of sirenomelia in the twin pregnancy is a worthy matter to be discussed.

We reported a case of twin sirenomelia in a patient after in vitro fertilization and embryo transfer (IVF-ET). Written informed consent was obtained from the couple before the procedure and manuscript publication. The treatment procedure followed ethical principles, all data were collected from chart reviews, and approval was obtained from the Institutional Review Board. In addition, we used a list of keywords including “Ectromelia,” “Sirenomelia,” and “Mermaid Malformation” to perform an extensive search and make a literature review. Twenty-three

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**Figure 1.** Prenatal ultrasonography image of the sirenomelic twin.

similar cases were selected in our study after we reviewed full text, and these are summarized herein.

**2. Case report**

A 33-year-old woman, gravida 1, para 0, underwent IVF-ET and 2 embryos were transferred to the uterus. After ET, ultrasonography revealed a dichorionic diamniotic twin pregnancy. The mother had progesterone treatment for 3 months after IVF-ET. The patient’s serology was negative for human immunodeficiency virus (HIV), venereal disease research laboratory (VDRL), and hepatitis B surface antigen (HBsAg) and she had no diabetes mellitus. The couple had no reported history of medication, substance abuse, and family history of congenital anomaly. Prenatal ultrasonography identified twin pregnancy with 1 fetus displaying fused lower limb at 24+3 weeks of gestation. Given this condition, the patient was transferred to our department.

The ultrasonography in our hospital revealed 1 twin normal anatomy with normal amniotic fluid volume and the S/D value of umbilical artery blood flow was 2.6; another twin had abnormal spinal sacral and tail physiological curvature, fused lower extremities with only 4.4cm echo of 1 lower extremity (2 long bones), absent fibula and tibia, oligohydramnios, single umbilical artery, duodenal atresia or stenosis (“double bubble sign” in abdominal cavity), and absent kidneys, ureters, and bladder (Fig. 1). A diagnosis of twin pregnancy with 1 sirenomelia twin was then made.

The couple was extensively counseled by the multi-disciplinary team regarding the diagnosis, treatment, and prognosis of the sirenomelia twin, and sirenomelia fetus would most likely to die due to multiple congenital anomalies in the uterus or after birth. We also recommend the couple of selective termination of the sirenomelia fetus. The parents preferred expectant management. Due to the couple’s refusal to exam the chromosome of sirenomelia twin, the chromosomal microarray analysis of the amniotic fluid sample was only achieved in the normal anatomy twin and the result was normal, karyotype:46, XY.

At 28 weeks’ gestation and 3 days, ultrasonography revealed sirenomelia twin complicated with persistent absent end-diastolic flow of umbilical artery, which persistent presented during the ongoing pregnancy. Close monitoring was performed. At 34 weeks’ gestation and 2 days, the demise of the malformed twin occurred, while fetal heart rate monitoring of the normal twin was abnormal, and an emergency cesarean section was performed. A healthy male baby weighing 2890g was delivered with Apgar scores of 10 and 10 at 1 and 5 minutes, respectively. The dead co-twin was 652g, fused lower limb with no feet, single umbilical artery, and absent external genitalia and anus (Fig. 2). Parents refused to do any further postmortem investigation (autopsy and radiological examinations). The normal twin stayed in the neonatal intensive care unit (NICU) for 5 days and discharged. The mother and the baby were followed up and are in good health until now.



**Figure 2.** Image of the sirenomelic twin. (A) Anterior views of the sirenomelic twin. (B) Posterior views of the sirenomelic twin.

**Table 1**  
**Clinical characteristics of selected trials.**

Reference	Age	Mode of conception	Twin type	Diagnosis GA	delivery GA	Mode of delivery	Maternal diabetes
Roberge, 1963 <sup>[16]</sup>	20	NS	MCDA	32	32	VD	NO
Wright and Christopher, 1982 <sup>[17]</sup>	20	NS	MCDA	NS	36	VD	NO
Biswas et al, 1985 <sup>[18]</sup>	25	NS	MCDA	NS	28	VD	NO
Oguz et al, 1986 <sup>[19]</sup>	19	NS	MCDA	NS	38	VD	NS
Schwaibold et al, 1986 <sup>[20]</sup>	25	NS	MCDA	NS	38	VD	NS
Di Lorenzo et al, 1991 <sup>[21]</sup>	NS	NS	MCDA	18	38	NS	NO
Kapur et al, 1991 <sup>[22]</sup>	34	NS	DCDA	17.5	Termination of pregnancy		NS
Rodriguez et al, 1991 <sup>[23]</sup>	40	NS	MCDA	25	32	VD	NO
Liu et al, 1998 <sup>[24]</sup>	27	NS	NS	35+1	35+1	CS	NS
Akbiyik et al, 2000 <sup>[25]</sup>	23	NS	NS	NS	38	CS	NO
Managoli et al, 2002 <sup>[26]</sup>	30	Spontaneous	DCDA	NS	38	VD	NS
Shonubi et al, 2002 <sup>[27]</sup>	22	NS	MCDA	NS	35	VD	NO
Assimakopoulos et al, 2004 <sup>[28]</sup>	37	IVF	DCDA	19	32	CS	YES
Horikoshi et al, 2005 <sup>[29]</sup>	33	IVF	DCDA	24	36	CS	NS
Akl and Saleh, 2006 <sup>[30]</sup>	21	NS	MCDA	18	34+4	CS	NO
Nisenblat et al, 2007 <sup>[31]</sup>	26	NS	DCDA	17	38	CS	NO
Guen et al, 2008 <sup>[32]</sup>	30	NS	DCDA	16	35	VD	NO
Chen et al, 2011 <sup>[33]</sup>	32	IVF	DCDA	31	35	CS	NO
Chowdhury et al, 2015 <sup>[34]</sup>	19	Spontaneous	MCMA	32	32	NS	NO
Nokeaingtong et al, 2015 <sup>[35]</sup>	20	IVF	DCDA	NS	27	CS	NO
Rodriguez et al, 2017 <sup>[36]</sup>	31	NS	MCDA	16	Termination of pregnancy		NO
Turgut et al, 2017 <sup>[37]</sup>	25	NS	DCDA	2nd trimester	32	CS	NO
Yaqoubi et al, 2018 <sup>[38]</sup>	32	Spontaneous	MCDA	25	36	VD	NO
Present case	33	IVF	DCDA	24+3	34+2	CS	NO

CS = cesarean section, DCDA = dichorionic diamniotic, GA = gestational age (wk), IVF-ET = in-vitro fertilization and embryo transfer, MCDA = monochorionic diamniotic, MCMA = monochorionic monoamniotic, NS, not specified, VD = vaginal delivery.

### 3. Discussion

Sirenomelia is a very rare and lethal congenital malformation. A retrospective cohort study (investigated 19 programs from North and South America, Europe, China, and Australia) showed that the global incidence of sirenomelia is 0.98/100,000.<sup>[11]</sup> The exact etiology of sirenomelia is unclear. Risk factors associated with sirenomelia were as follows: maternal age <20 years or >40 years,<sup>[11]</sup> maternal diabetes, prenatal exposure to retinoic acid, cadmium, cyclophosphamide,<sup>[5]</sup> cocaine, landfills water, or lamotrigine.<sup>[6]</sup>

The vascular steal theory and defective blastogenesis are 2 main and accepted hypotheses. Vascular steal theory<sup>[7]</sup>: aberrant abdominal artery arises from the high level of the aorta with no branches of the renal artery or iliac artery and carries much blood to the placenta. Poor perfusion of caudal will lead to the abnormal development of the lower part body. Defective blastogenesis<sup>[8]</sup>: defective blastogenesis of caudal mesoderm between 13th days and 22nd days may lead to the lower extremity merging, malrotation, and/or dysgenesis.

Only 2 published cases of sirenomelia fetus were with the abnormal chromosomal result (a de novo balanced translocation<sup>[9]</sup> and a triploidy mosaic,<sup>[5]</sup> respectively), others were with the normal chromosome. Genetic basis of sirenomelia mice demonstrated that Cyp26a1, a key enzyme in the degradation of retinoic acid, may reduce bone morphogenetic protein-7 (Bmp-7) signaling in the caudal embryonic region.<sup>[10,11]</sup>

A regular antenatal ultrasound examination is recommended to scan and diagnose this congenital malformation in time. The transvaginal scan is a modality for better visualization of detailed anatomy.<sup>[8]</sup> Although ultrasonography is an optimal method for diagnosis of sirenomelia<sup>[12]</sup> at 1st-trimester or early 2nd-trimester,<sup>[8,13,14]</sup> visualization of sirenomelia by ultrasound may

be impaired by the coexisting oligohydramnios and/or intrauterine growth retardation (IUGR). Three-dimensional sonography and magnetic resonance imaging (MRI) also play key roles in sirenomelia diagnosis. Postnatal X-ray and autopsy are also recommended to confirm the diagnosis.<sup>[8]</sup> In the present case, sirenomelia was diagnosed by ultrasonography in the late 2nd trimester.

A more adequate classification of sirenomelia is reported by Stocker and Heifetz<sup>[15]</sup>: I, all thigh and leg bones are present; II, single fibula; III, absent fibulae; IV, partially fused femurs, fused fibulae; V, partially fused femurs, absent fibulae; VI, single femur, single tibia; VII, single femur, absent tibiae. In this case, sirenomelia twin was classified as type III according to ultrasonography and clinical examination, fused lower extremities, absent fibula, and tibia.

This case was dichorionic diamniotic twin pregnancy with sirenomelia of which limited studies have been reported. We conducted a search and reviewed the full literature pertaining to cases of sirenomelia in twin pregnancy after a thorough search of concerning literature in several databases. There were 23 published articles<sup>[16-38]</sup> reported cases of sirenomelia in twin pregnancy and 1 article<sup>[2]</sup> of sirenomelia in triplet pregnancy in English and Chinese. In our study, we only selected literature with sirenomelia in twin pregnancy. So, combing our case together, the number of sirenomelia in monochorionic diamniotic twin, dichorionic diamniotic twin, and the monochorionic monoamniotic twin was respectively 11, 10, and 1 due to 2 articles did not mention.<sup>[24,25]</sup> The maternal age range from 19 to 40 years and most cases conceived naturally. Diagnosis sirenomelia was between 16 and 32 weeks' gestation and delivery was from 27 to 38 gestational weeks except 2 cases terminated pregnancy at 2nd trimester. No abnormal gene or chromosome was found in the

**Table 2**

**Perinatal fetus outcomes of twin pregnancy in selected trials.**

Study ID	First twin			Second twin			Sirenomelia karyotype
	Sex	Birth weight, g	Newborn outcome	Sex	Birth weight, g	Newborn outcome	
Roberge, 1963 <sup>[16]</sup>	Female	1100	Sirenomelia, died 1 h and 34 min after delivery	Female	1022	Sirenomelia, died 1 h and 39 min after delivery	NS
Wright and Christopher, 1982 <sup>[17]</sup>	Female	2165	Normal anatomy and did well following delivery	Female	930	Sirenomelia, died 2 h after delivery due to respiratory arrest	46,XX
Biswas et al, 1985 <sup>[18]</sup>	Male	1300	Deficient cephalic and parietal bone formation, died after few hours of birth	NS	1500	Sirenomelia, grossly deformed without the signs of life at birth	NS
Oguz et al, 1986 <sup>[19]</sup>	Male	NS	Normal, alive	Male	1500	Sirenomelia, died of renal insufficiency during the 1st day of life	46, XY
Schwaibold et al, 1986 <sup>[20]</sup>	female	2800	Alive	Unknown sex	980	Sirenomelia, stillborn	NS
Di Lorenzo et al, 1991 <sup>[21]</sup>	Male	NS	Normal anatomy, alive	Male	2725	Died	NS
Kapur et al, 1991 <sup>[22]</sup>	—	—	Gestational sac disappeared at 8.5 gestational wks	—	—	Sirenomelia, termination of pregnancy at 18 wks	46, XY
Rodriguez et al, 1991 <sup>[23]</sup>	Female	1540	Abdominal distension, died 2 d after surgery due to severe disseminated intravascular coagulation	Unknown sex	2200	Sirenomelia and anencephaly, hydroptic stillborn	NS
Liu et al, 1998 <sup>[24]</sup>	Male	NS	Alive, normal anatomy	Male	2150	Sirenomelia, died within 5 min after birth	46, XY
Akbiyik et al, 2000 <sup>[25]</sup>	Male	NS	Healthy after underwent operation for imperforate anus	Male	NS	Sirenomelia, died on the 17th day	46, XY
Ugwu et al, 2011 <sup>[2]</sup>	Male	1000	Alive and well	Male	1000	Died soon after birth	NS
Managoli et al, 2002 <sup>[26]</sup>	Male	2050	Normal anatomy, alive	Unknown sex	1400	Sirenomelia, died after 5 h after birth	NS
Shonubi et al, 2002 <sup>[27]</sup>	Male	2450	Normal anatomy, alive	Male	1600	Sirenomelia, died after 20 h of life	NS
Assimakopoulos et al, 2004 <sup>[28]</sup>	Male	2010	Normal anatomy and fetal biometry	Male	1390	Sirenomelia, died 3 h after delivery	NS
Horikoshi et al, 2005 <sup>[29]</sup>	Female	2806	Normal anatomy	Male	1600	Sirenomelia, died 5 h after birth	46, XY
Akl and Saleh, 2006 <sup>[30]</sup>	Male	2100	Normal anatomy, alive	Male	1835	Sirenomelia, died 5 d after birth	NS
Nisenblat et al, 2007 <sup>[31]</sup>	Female	3240	Normal anatomy, alive	Male	1470	Sirenomelia, died within minutes after birth	46, XY
Guyen et al, 2008 <sup>[32]</sup>			Normal, alive	female	1800	Sirenomelia, died after 24 h	NS
Chen et al, 2011 <sup>[33]</sup>	Female	2128	normal anatomy, alive	Female	1576	Sirenomelia, died soon after birth	46, XX
Chowdhury et al, 2015 <sup>[34]</sup>	NS	1200	Sirenomelia, died within hour after birth	NS	1000	Sirenomelia with ancephaloc, died within hour after birth	NS
Nokeaingtong et al, 2015 <sup>[35]</sup>	NS	NS	Alive, normal anatomy	Male	716.5	Sirenomelia, died 30 min after birth due to inability to ventilate	NS
Rodríguez et al, 2017 <sup>[36]</sup>	—	—	—	—	—	Sirenomelia, termination of pregnancy	46, XY
Turgut et al, 2017 <sup>[37]</sup>	NS	NS	Alive, normal anatomy	Male	1040	Sirenomelia, died 4 h after birth	46, XY
Yaqoubi et al, 2018 <sup>[38]</sup>	Female	2500	Alive, normal anatomy	Female	1300	Sirenomelia, died 4 h after birth	46, XX
Present case	Male	2890	Alive	Unknown sex	652	Sirenomelia, died before caesarean section	NS

NS = not specified.

included literature. The survival rate of sirenomelia was very few and most was died from multiple congenital anomalies, while normal anatomy fetus had a high alive rate in previously reported cases and our case. Detailed information of sirenomelia in twin pregnancy is summarized in Tables 1 and 2.

Källén et al<sup>[39]</sup> conducted an epidemiologic study of rare malformations (nearly 10.1 million births) and found 97 cases with sirenomelia; however, among these 35 was stillbirths and 62 live births dead shortly after birth.

Jaiyesimi et al<sup>[40]</sup> found that the frequency of main defects in sirenomelia as follows: vertebrae/sacrum/pelvis/lower limb (100%), anorectal (97%), renal (93%), genital (85%), lower urinary tract (57%), single umbilical artery (79%), cardiac (26%), radial limb (21%), esophageal atresia or/and tracheo-oesophageal fistula (5%), respiratory tract (24%), and central nervous system (8%). In our present case, it was dichorionic-diamniotic twin pregnancy, and the sirenomelia twin was

complicated with oligohydramnios, single umbilical artery, fused lower extremities with the only echo of 1 lower extremity (2 long bones), absent fibula and tibia, duodenal atresia or stenosis, and absent kidneys, ureters, and bladder.

The cost of treatment in live-born sirenomelia is huge due to renal transplantation, life support and other repeated surgeries. We advised expecting parents should be counseled by the multidisciplinary team about the management and prognosis of the sirenomelia. Voluntary selective termination of sirenomelia twin can be advised to the parents based on the local law, religious, and ethical principles to reducing the rate of maternal complications in twin pregnancy such as preterm labor, PPRM, or mental trauma<sup>[8]</sup> if sirenomelia could be diagnosed early.

In summary, the survival and quality of life in live-born sirenomelia is extremely low due to multiple congenital anomalies. It is a lethal condition in the perinatal period. Early antenatal diagnosis is important. Expecting parents should be

counseled by the multidisciplinary team about the management and prognosis of the sirenomelia. Based on the local law and ethical principles, voluntary selective termination of sirenomelia 1 in twin pregnancy may be advised to the parents.

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## Author contributions

Tingting Xu carried out the retrospective review of the case, participated in the design, writing, and organization of the manuscript. Haiyan Yu conceived of the whole study, overall supervised the work, and carried out the study design and correction of the manuscript. Xiaodong Wang and Hong Luo participated in the design of the study. All authors read and approved the final manuscript.

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