OBSTETRICS

Pseudoamniotic band syndrome: a rare complication of monochorionic twins with fetofetal transfusion syndrome treated by laser coagulation

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OBJECTIVE: The purpose of this study was to assess the incidence and risk factors of limb constriction defects that are related to pseudoamniotic band syndrome (PABS) after selective fetoscopic laser surgery (FLS) in fetofetal transfusion syndrome (FFTS).

STUDY DESIGN: All consecutive cases of FFTS that were treated by selective FLS between 1999 and 2006 were examined prospectively for PABS at the time of delivery. Incidence and characteristics of PABS were reported. Univariate analysis was conducted to look for potential risk factors of developing PABS.

RESULTS: The 438 consecutive FFTS cases were treated at 15-26 weeks of gestation; PABS developed in 8 cases (1.8 %). The affected twin was always the former recipient. The diagnosis was made prenatally in 2 of 8 cases (25%). All cases survived the perinatal period. PABS affected fetal leg, arm, and foot in 3, 4, and 1 cases, respectively. In 5 (62.5%) and 7 (87.5%) cases, PABS occurred after premature rupture of membranes and intrauterine death of the donor, respectively. In 4 cases (50%), there was both premature rupture of membranes (PROM) and intrauterine fetal death; in 3 cases (37.5%), there was intrauterine fetal death alone, and in 1 case (12.5%), there was PROM alone. In the remaining 430 cases, PROM occurred in 62 cases (14.4%) and 66 cases (15.3%) within and after 3 weeks after surgery, respectively. PROM was significantly more frequent within the group that was complicated with PABS than within the rest of the cohort ($P = .05$). No maternal, fetal, or perioperative risk factor could be identified.

CONCLUSION: Awareness and targeted serial ultrasound evaluation in this high-risk group may improve prenatal diagnosis, counseling, and management of PABS after FLS.

Key words: fetofetal transfusion syndrome, fetoscopic laser surgery, limb constriction, pseudoamniotic band syndrome

Cite this article as: Winer N, Salomon LJ, Essaoui M, Nasr B, Bernard JP, Vile Y. Pseudoamniotic band syndrome: a rare complication of monochorionic twins with fetofetal transfusion syndrome treated by laser coagulation. Am J Obstet Gynecol 2008;198:393.e1-393.e5.

Amniotic band syndrome is the sporadic association of severe congenital malformations that are related to entanglement or amputation of fetal parts by multiple strands of a ruptured amniotic sac.[1] Its incidence is 1 of 1200 to 1 of 15,000 live births.[2,3] The most common anomalies include multiple constrictions or amputations of the limbs and craniofacial and body wall defects.[1,4] Risk factors are ill-defined but include bleeding in early pregnancy.[5] Pseudoamniotic band syndrome (PABS) is a iatrogenic complication that causes entanglement of fetal parts in a constrictive sheet of detached or ruptured amniotic membrane after an invasive procedure that includes amniocentesis,[6-8] amnioreduction,[9] or septostomy in twins.[10] Selective fetoscopic laser surgery (FLS) of intertwin anastomoses on the chorionic plate has proved to improve survival in severe fetofetal transfusion syndrome (FFTS) presenting before 26 weeks of gestation.[11] FLS is now offered widely as the first-line treatment in these complicated monochorionic pregnancies.

To our knowledge, the incidence of and risk factors for PABS after fetoscopy-guided laser have not been documented. These issues were addressed in a large cohort of consecutive cases of FFTS that underwent laser surgery.

Materials and Methods

We reviewed all consecutive cases of FFTS that were referred to our institution for FLS over a 7-year period (1999-2006). Selective laser coagulation of placental anastomoses on the chorionic plate was performed as previously described[11] and has become standard of care in our institution in women with FFTS between 15 and 26 weeks of gestation. In all cases, initial evaluation included a detailed ultrasound examination of both twins, with biometric, morphologic, and Doppler examination. Inclusion criteria included polyuric polyhydramnios in the recipient twin, with the deepest vertical pool of at least 8.0 cm ≤20

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Received April 23, 2007; accepted Sept. 17, 2007.

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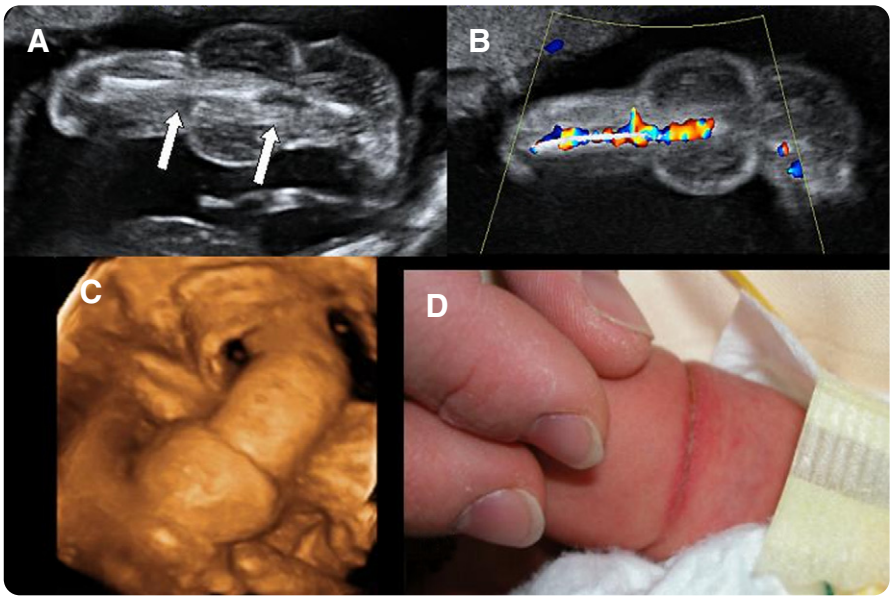
0002-9378/$34.00

doi: 10.1016/j.ajog.2007.09.030

FIGURE
Right fetal forearm with 2 strictions



Right fetal forearm with 2 strictions (*arrows*) that caused edema as shown by **A,** 2-dimensional and **C,** 3-dimensional ultrasound scanning shows adequate vascularization on **B,** color Doppler imaging; **D,** postnatal appearance at birth.

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weeks of gestation and 10.0 cm from 20 weeks onwards with a distended bladder, together with oliguric oligohydramnios in the donor twin, with the deepest vertical pool measuring at most 2.0 cm. Exclusion criteria from treatment were fetal death, major fetal anomaly, ruptured membranes, maternal condition mandating delivery, and any previous invasive procedure. All procedures were performed percutaneously, under local anesthesia.[11] The site of entry on the maternal abdomen was chosen to allow access to the insertion of the intertwin membranes on the placental surface through the recipient's sac with a 3.3-mm cannula that was loaded with a trocar under continuous ultrasound guidance. A 2-mm fetoscope (50,000 pixels, model 11630; Karl Storz GmbH & Co KG, Tuttlingen, Germany) and a diode laser with a 400- to 600-μm fiber were used. Intertwin anastomoses were identified and coagulated with a nontouch technique with an output of 30-60 W. Amniotic fluid was drained subsequently through the canula until the deepest pool reached a maximum of 5-6 cm on ultrasound examination.

Cases in which PABS was identified in utero and/or at birth were recorded. Information regarding pregnancy course and outcome were collected in all cases and included maternal age, history of bleeding in early pregnancy, gestational age at laser surgery, gestational age at delivery, the type of multiple pregnancy, postoperative preterm premature rupture of membranes, intrauterine death of 1 twin, and the occurrence of PABS. The location of the placenta (anterior or posterior), the need for a transplacental approach, the occurrence of intraamniotic bleeding, and technical difficulties were also noted. The procedure was called difficult when good visualization was impaired because of cloudy amniotic fluid or perioperative intraamniotic bleeding or when there was an incomplete visualization of the vascular equator caused by the fixed position of the donor twin or that of the intertwin membranes.

Statistical analysis

The distribution of cases between donors and recipients was compared on the basis of a binomial distribution. Potential predictors for PABS were investigated with univariate analysis of maternal, fetal, and perioperative characteristics that were associated with the occurrence of PABS. Risk factors that were identified on univariate analysis were intended to be included in a multivariate logistic regression model. Qualitative data were compared by means of chi-square test or Fisher's exact test, as appropriate. The Student *t* test was used for parametric variables. Continuous variables were reported as mean ± SD. The Welch correction was applied in cases of unequal SDs. Statistical analyses were performed with Stata software (version 9.2 for Windows; StataCorp LP, College Station, TX), Statistica software (version 6.0; StatSoft, Inc, Tulsa, OK), and Excel 2000 (Microsoft Corporation, Redmond, WA). All tests were 2-tailed, and a probability value of $<.05$ was considered statistically significant.

RESULTS

Four hundred thirty-eight FFTS cases were treated during the study period and formed our study group. There were 420 (95.7%) and 18 (4.3%) monochorionic twin and dichorionic triplet pregnancies, respectively. Mean (± SD) maternal age was 30.3 ± 5.4 years. Laser surgery was performed at a mean gestational age of 21 ± 2.9 weeks. There were 188 cases (42.8%) with an anterior placenta.

Eight cases were complicated by PABS, and only 2 of these had been diagnosed prenatally. Bleeding in early pregnancy was not reported in any of the 8 cases. The affected twin was the recipient in all cases ($P = .004$), and the donor had died in utero after the procedure in all cases but 1. PABS affected leg, arm (Figure), and foot in 3, 4, and 1 cases, respectively. PABS occurred after premature rupture of membranes at 25-33 weeks of gestation in 5 cases (62.5%). In the remaining 430 cases, premature rupture of membranes occurred in 62 (14.4%) and 66 cases (15.3%) within and after 3 weeks after surgery, respectively. Premature rupture of membranes was significantly more frequent within the group that was complicated with PABS than within the rest of the cohort ($P = .05$). Mean gesta-

TABLE 1
Characteristics of the 8 cases with PABS

	Case							
Characteristic	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**
Maternal								
Type of multiple pregnancy	[a]	[a]	[a]	[a]	[b]	[a]	[a]	[a]
Maternal age at surgery (y)	31	24	32	26	26	38	21	34
Gestational age at surgery (wk)	21	23	16	21	19	19	20	16
Gestational age at birth (wk)	26	34	33.5	30	25.5	33	30	31
Bleeding in early pregnancy	No	No	No	No	No	No	No	No
Fetal								
Position of the placenta	Posterior	Anterior	Anterior	Anterior	Anterior	Posterior	Posterior	Posterior
Affected fetus	Recipient	Recipient	Recipient	Recipient	Recipient 1	Recipient	Recipient	Recipient
Intrauterine death after LFS	Donor	Donor	Donor[c]	Donor	Donor	Donor	2 Alive	Donor
Quintero staging	1	3	3	2	3	1	1	3
Premature rupture of membranes	No	No	Yes	Yes	Yes	No	Yes	Yes
Affected limbs	2 Legs	Left arm	Right leg	Right leg	Right foot	Left arm	Left arm	Right hand

LFS, laser fetoscopic surgery.

[a] Monochorionic biamniotic twin pregnancy.

[b] Monochorionic triamniotic triplet pregnancy with 2 recipients and 1 donor.

[c] Fetal death by cord coagulation 10 days after laser therapy.

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tional age at delivery in cases that were complicated with PABS was 30.4 ± 3.2 weeks. Characteristics of the cases are shown in Table 1.

Univariate analysis of potential risk factors for PABS was performed (Table 2). We could not identify any factor that could help to predict the occurrence of PABS; placental position, duration of surgery, gestational age at laser surgery, and technical difficulties showed similar distributions in cases of FFTS with or without PABS. Therefore, a multivariate regression model was not built.

Comment

Laser coagulation of the intertwin anastomoses on the chorionic plate has proved to be the best first-line treatment in FFTS when the syndrome develops before 26 weeks of gestation.[11] Complications at least partly attributable to FLS in FFTS include fetal death and preterm premature rupture of membranes.[12] Our study demonstrates that PABS can also occur after FLS in approximately 2% of cases.

PABS have also been reported after other invasive prenatal procedures (eg, amniocentesis[6,8] and septostomy[10]) in twins. Fetoscopy and laser coagulation were performed in the recipient sac and were followed by amnioreduction in all cases. This possibly could lead to rupture of the amnion or of the intertwin membrane unrecognized in the perioperative period by creating holes in the intertwin membrane or free-floating sheets that could trap parts of the most mobile twin after fetoscopic surgery. This could explain the reason that the recipient was always the affected twin in our series ($P = .004$). The high frequency of clinically diagnosed preterm premature rupture of membranes in our series (5 of 8 cases; 62.5%) is also consistent with a particular role of a defect in the amnion in this condition.

Sporadic cases of PABS in monochorionic twins with and without FFTS have been reported, but the diagnosis was made before any invasive procedure was performed.[13-16] PABS has also been reported in an acardiac twin and in the pump twin; both fetuses showed multiple severe malformations, which suggests an early endogenous origin of amniotic band syndrome.[17] Cord entanglement within an amniotic band in monochorionic diamniotic twins has also been reported after spontaneous antepartum septostomy.[10,18] Serial amnioreduction and septostomy have been associated with severe complications that were related to intrauterine amniotic rupture, including fetal and neonatal death that resulted from cord entanglement, preterm premature rupture of the membranes, preterm labor and delivery, and amniotic band syndrome.[19-21] Our report suggests that PABS may occur in up to 2% of the cases after FLS. This low incidence may explain the reason that the examination of a large cohort was necessary to identify

TABLE 2
Characteristics of the overall FFTS population

Characteristic	FFTS		
	With laser surgery (n = 430)	Complicated by amniotic bands (n = 8)	*P* value[a]
Age (y)[b]	30.3 ± 5.4	29 ± 5.7	.50
Gestational age at laser (wk)[b]	21 ± 2.9	19.3 ± 2.3	.10
Placental location (n)			
Anterior	184	4	.73
Posterior	241	4	
Not identified	5		
Gestational age at delivery (wk)[b]	29.4 ± 5.6	30.4 ± 3.2	.61
Duration of laser surgery (min)[b]	32.3 ± 12.5	36.6 ± 20.8	.32
Site of entry (n)			
Transplacental	92	1	.69
Extraplacental	285	7	
Undetermined	53		
Anastomoses detected and coagulated endoscopically (n)[b]	8.7 ± 4.6	7.2 ± 4.2	.33

[a] $P < .05$ was considered significant.

[b] Data presented as mean ± SD.

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this rare but potentially severe complication.

We reviewed PABS cases retrospectively and therefore may have missed some additional cases, particularly in cases with double fetal death.[22] In cases with single fetal death, delivery and pathologic examination occurred after a long time interval, and bands may have been missed in macerated fetuses, even after careful examination. Our large cohort did not identify any predictor of the occurrence of PABS, although there was a nonsignificant trend for earlier gestation at surgery in the affected group ($P = .1$). Many factors have been related to the occurrence of PABS.[23] However, we were not able to evaluate all of them because of the retrospective design of our study and the low prevalence of PABS. Factors that were associated with difficult FLS (such as anterior placenta and transplacental entry) were not associated with the development of amniotic bands. This is in agreement with a previous report that could not identify any risk factor for premature rupture of the membranes or miscarriage in FFTS that was treated by FLS.[12] Finally, our study highlighted the poor performance of prenatal diagnosis of PABS because only 2 of 8 cases were identified prenatally. Pregnancies that are treated by FLS for FFTS, especially with intrauterine fetal death of the donor, should raise awareness of PABS and may justify targeted ultrasound follow-up examination that would include Doppler studies and 3-dimensional sonography,[24,25] because this may contribute to better diagnosis, counseling, and treatment of cases that are complicated with PABS. ■

ACKNOWLEDGMENT

We thank S. Boulet for the postnatal picture.

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