

Table S1. Data of fetuses with BPS managed expectantly.

| Author | n | GA at diagnosis | Hydrops | Fetal / Neonatal death | Survival | GA at birth | Surgery |
|-------------------------------------|-----|--------------------------------------|-------------------|------------------------|-----------------|-------------|---------------------|
| Langer <i>et al.</i> , 1995 [32] | 2 | 25 - 26 | - | - | 2 (100%) | 37 - 38 | 0/lost to follow-up |
| Abuhamad <i>et al.</i> , 1996 [33] | 2 | 18 | - | - | 2 (100%) | 39 – 40 | 0 |
| da Silva <i>et al.</i> , 1996 [34] | 3 | 25 - 34 | + (2) ascites (1) | - | 3 (100%) | 30 - 35 | 3 (100%) |
| Evans <i>et al.</i> , 1996 [35] | 3 | 25 - 34 | + (3) | - | 3 (100%) | 30 - 36 | 3 (100%) |
| Adzick <i>et al.</i> , 1998 [1] | 37 | 18 - 36 | + (1) ascites (1) | 1 (neonatal) | 36 | no data | 7 (19%) |
| Becmeur <i>et al.</i> , 1998 [10] | 8 | 20 - 33 | - | - | 8 (100%) | 37 - 40 | 8 (100%) |
| Bratu <i>et al.</i> , 2001 [36] | 13 | 24 | + (2) | 2 (neonatal) | 11 (84%) | no data | 11 (85%) |
| Chen <i>et al.</i> , 2003 [37-39] | 2 | 19 - 20 | - | - | 2 (100%) | 38 - 40 | 1 (50%) |
| Illanes <i>et al.</i> , 2005 [440] | 4 | 19 - 29 | - | - | 4 (100%) | no data | 2 (50%) |
| Ruano <i>et al.</i> , 2005 [41] | 3 | 21 - 33 | - | 1 (neonatal) | 2 (77%) | no data | 3 (100%) |
| Cavoretto <i>et al.</i> , 2008 [22] | 6 | 19 - 35 | - | - | 6 (100%) | 37 - 42 | 5 (83%) |
| Mallmann <i>et al.</i> , 2014 [17] | 29 | 20 - 27 | - | - | 29 (100%) | 34 - 39 | 16 (55%) |
| Stoiber <i>et al.</i> , 2015 [18] | 7 | 22 | - | | 7 (100%) | 39 | no data |
| Nunes <i>et al.</i> , 2015 [42] | 7 | [2 nd – 3 rd] | | | 7 (100%) | >35 | 4 (57%) |
| Pinto <i>et al.</i> , 2016 [43] | 2 | 20 - 22 | - | | 2 (100%) | 34 - 38 | 0 |
| Riley <i>et al.</i> , 2018 [19] | 100 | 19 - 37 | 5 | | 100 (100%) | 28 - 41 | 76 (76%) |
| Present series | 16 | 20 - 28 | - | | 16 (100%) | 37 – 40 | 4 (25%) |
| Total | 244 | | 15/244 [6.1%] | 4/244 [1.6%] | 240/244 [98.3%] | | 143/226 [63.3%] |

n=number of fetuses included in the study; GA = gestational age in weeks; Surgery = number of fetuses requiring postnatal surgery

Table S2a. Invasive management: data of fetuses with BPS treated with thoraco-amniotic shunt.

| Author | n | GA at interv. | Method of treatment | Presence of hydrothorax /hydrops | Effect | Fetal death | Neonatal death | Survival | GA at birth | Surgey |
|---|---|---------------|--|----------------------------------|----------------------|-------------|----------------|-----------|-------------|----------|
| Weiner <i>et al.</i> , 1986 [7] | 1 | 24 | Thoracocentesis + Thoraco-amniotic shunt | 1 (100%) | Reaccumulated (100%) | | 1 | 0 | 29 | 1 (100%) |
| Slotnick <i>et al.</i> , 1990 [8] | 1 | 32 | Thoracocentesis + Thoraco-amniotic shunt | 1 (100%) | Resolved (100%) | | | 1 (100%) | 34 | 1 (100%) |
| Hernanz-Schultzman <i>et al.</i> , 1991 [2] | 1 | 27 | Thoraco-amniotic shunt | 1 (100%) | Resolved (100%) | | | 1 (100%) | 36 | 1 (100%) |
| Favre <i>et al.</i> , 1994 [9] | 1 | 30 | Thoraco-amniotic shunt | 1 (100%) | Resolved (100%) | | | 1 (100%) | 38 | 1 (100%) |
| Adzick <i>et al.</i> , 1998 [1] | 2 | 29-30 | Thoraco-amniotic shunt | 2 (100%) | Resolved (100%) | | | 2 (100%) | 33 - 35 | 2 (100%) |
| Becmeur <i>et al.</i> , 1998 [10] | 1 | 30 | Thoraco-amniotic shunt | 1 (100%) | Resolved (100%) | | | 1 (100%) | 38 | 1 (100%) |
| Lopoo <i>et al.</i> , 1999 [11] | 2 | 23-30 | Thoraco-amniotic shunt | 2 (100%) | Resolved (100%) | | | 2 (100%) | 33 | No data |
| Salomon <i>et al.</i> , 2003 [12] | 1 | 34 | Thoraco-amniotic shunt | 1 (100%) | Resolved (100%) | | | 1 (100%) | 36 | 0 |
| Picone <i>et al.</i> , 2004 [13] | 1 | 19-36 | Thoraco-amniotic shunt | 1 (100%) | No data (100%) | | | 1 (100%) | 28-40 | No data |
| Hayashi <i>et al.</i> , 2006 [14] | 3 | 28-30 | Thoracocentesis + Thoraco-amniotic shunt | 3 (100%) | Resolved (100%) | | | 3 (100%) | 33-35 | 3 (100%) |
| Odaka <i>et al.</i> , 2006 [15] | 1 | 27 | Thoraco-amniotic shunt | 1 (100%) | Reaccumulated (100%) | | | 1 (100%) | 37 | 1 (100%) |
| Kitano <i>et al.</i> , 2006 [16] | 3 | 27-30 | Thoracocentesis + Thoraco-amniotic shunt | 3 (100%) | Resolved (100%) | | | 3 (100%) | 33-35 | No data |
| Mallmann <i>et al.</i> , 2014 [17] | 7 | 25 - 29 | Thoraco-amniotic shunt | 7 (100%) | - | 1 | | 6 (85.7%) | 30-37 | 5 (83%) |
| Stoiber <i>et al.</i> , 2015 [18] | 2 | 29-30 | Thoracocentesis + Thoraco-amniotic shunt | 2 (100%) | Resolved (100%) | | 1 | 1 (50%) | 33-36 | 0 |

| | | | | | | | | | | |
|-------------------------|----|-------|------------------------|--------------|-----------------|--|--|---------------|-------|-------------|
| Riley et al., 2018 [19] | 2 | 23-27 | Thoraco-amniotic shunt | 2 (100%) | Resolved (100%) | | | 2 (100%) | 33-35 | 1 (50%) |
| Present series | 5 | 20-25 | Thoraco-amniotic shunt | 5 (100%) | Resolved (100%) | | | 5 (100%) | 34-38 | 3 (60%) |
| | 34 | | | 34/34 [100%] | | | | 31/34 [91.2%] | | 21/26 [84%] |

n = number of fetuses included in the study; GA = gestational age in weeks; LB=life birth; IUD=intrauterine death; NND= neonatal death; Surgery = number of fetuses requiring postnatal surgery

Table S2b. Invasive management: data of fetuses with BPS treated with laser coagulation of the feeding vessel.

| Author | No of fetuses | GA at interv. | Method of treatment | Presence of hydrothorax/hydrops | Effect | Fetal death | Neonatal death | Survival | GA at birth | Surgery |
|---|---------------|---------------|---------------------|---------------------------------|---------------------------------------|-------------|----------------|-----------|-------------|----------|
| Oepkes et al., (2007) [20] | 1 | 23 | Laser coagulation | 1 (100%) | Resolved (100%) | | | 1 (100%) | 39 | 0 |
| Ruano et al., 2007 [21] | 1 | 29 | Laser coagulation | 1 (100%) | Resolved (100%) | | | 1 (100%) | 38 | 1 (100%) |
| Cavoretto et al., 2008 [22] | 8 | 23-32 | Laser coagulation | 8 (100%) | Resolved (100%) | | | 8 (100%) | 34-41 | 5 (63%) |
| Witlox et al., (2009) [23] | 1 | 23 | Laser coagulation | 1 (100%) | Resolved (100%) | | | 1 (100%) | 41 | 0 |
| Ruano et al., 2012 [24] | 3 | 24-29 | Laser coagulation | 3 (100%) | Resolved (66.7%) No change (33.3%) | 1 | | 2 (66.7%) | 30-39 | No data |
| Mallmann et al., 2014 [17] | 5 | 24 - 31 | Laser coagulation | 5 (100%) | Resolved (100%) | | | 5 (100%) | 38 - 40 | 1 (20%) |
| Cruz-Martinez et al., 2015, 2017 [25, 26] | 15 | 20-31 | Laser coagulation | 15 (100%) | Resolved (100%) | | | 15 (100%) | 35-40 | 0 |
| Kosinski et al., 2017 [27] | 2 | 22-27 | Laser coagulation | 2 (100%) | Resolved (100%) | | | 2 (100%) | 39-41 | 0 |
| Gottschalk et al., 2017 [28] | 11 | 25-34 | Laser coagulation | 11 (100%) | Resolved (100%) | | | 11 (100%) | 30-42 | 3 (27%) |

| | | | | | | | | | | |
|----------------|----|-------|-------------------|----------|-----------------|--|--|---------------|-------|---------------|
| Present series | 7 | 21-25 | Laser coagulation | 7 (100%) | Resolved (100%) | | | 7 (100%) | 39-41 | 2 (28.6%) |
| Total | 54 | | | | | | | 53/54 [98.1%] | | 12/51 [23.5%] |

N = number of fetuses included in the study; GA = gestational age in weeks; LB=life birth; IUD=intrauterine death; Surgery = number of fetuses requiring postnatal surgery

Table S2c. Invasive management: data of fetuses with BPS treated with intratumor injection of sclerosant or combined treatment.

| Author | No of fetuses | GA at interv. | Method of treatment | Presence of hydrothorax /hydrops | Effect | Fetal death | Neonatal death | Survival | GA at birth | Surgery |
|------------------------------|---------------|---------------|---|----------------------------------|-----------------|-------------|----------------|-------------|-------------|-------------|
| Nicolini et al. 2000 [29] | 1 | 27 | Alcohol injection + thoracoamniotic shunt | 1 (100%) | Resolved (100%) | | | 1 (100%) | 40 | 0 |
| Bermudez et al., 2007 [30] | 3 | 24-26 | Polidocanol injection | 2 (67%) | Resolved (100%) | | 1 | 2 (67%) | 38 | 2 (67%) |
| Ramos KS et al., 2010 [31] | 2 | 30-31 | Thoracoamniotic shunt + laser coagulation + thoracocentesis | 1 (100%) | Resolved (100%) | | | 2 (100%) | No data | 2 (100%) |
| Gottschalk et al., 2017 [28] | 1 | 25, 26 | Thoracoamniotic shunt + laser coagulation | 1 (100%) | Resolved (100%) | | | 1 (100%) | 40 | 0 |
| Present series | 1 | 25 | Thoracoamniotic shunt + laser coagulation | 1 (100%) | Resolved (100%) | | | 1 (100%) | 40 | 1 (100%) |
| Total | 8 | | | | | | | 7/8 [87.5%] | | 5/8 [62.5%] |

N = number of fetuses included in the study; GA = gestational age in weeks; LB=life birth; NND= neonatal death; Surgery = number of fetuses requiring postnatal surgery