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Prenatal causes of an allantoic cyst with a patent urachus

Victor Cavillon[1-2], Anne Schneider[1], Poornima Donepudi[3], Raphael Moog[1], Isabelle Lacreuse[1], Anne Sophie Weingertner[2], Romain Favre[2], François Becmeur[1]

[1]Department of Pediatric Surgery, Hôpitaux Universitaires de Strasbourg, 67098 Strasbourg Cedex, France.
[2]Department of Obstetrics and Gynecology, Hôpitaux Universitaires de Strasbourg, 67098 Strasbourg Cedex, France.
[3]IRCAD, Hôpitaux Universitaires de Strasbourg, 67091 Strasbourg, France.

ABSTRACT

Prenatal diagnosis of an allantoic cyst with a patent urachus is rare. A urological etiology can sometimes be associated with it. We report 4 cases of allantoic cyst with a patent urachus and reviewed the literature to investigate for any etiology using the keywords “allantoic cyst” and “patent urachus”. Diagnosis of an allantoic cyst with a patent urachus was established during the first or second trimester of pregnancy. In two cases, rupture of prenatal cysts occurred and in one case, we described a prenatal vesicourethral abnormality. Surgery was performed with good results and no complications. A literature review collected 12 studies (15 fetuses) with a sex ratio of 3 boys/1 girl. The cysts ruptured in 2/3 of the cases, and in half of the cases, urinary abnormalities were found before birth (enlarged bladder, bladder prolapse). At birth, one boy had posterior urethral valve (PUV) and 4 boys had a bladder prolapse. The postoperative course was uneventful. We present our series with one case showing the etiology of a patent urachus during prenatal life. Cystoscopy seems to be valuable in the post-natal diagnosis. In the literature, we found one case of PUV. Surgery was performed with good results and no complications.

Key Words: Allantoic cyst; patent urachus; pseudocyst; urethral valves; pediatric urology.

Corresponding Author *Victor Cavillon*
Department of Obstetrics and Gynecology
Hôpitaux Universitaires de Strasbourg
1 Avenue Molière 67098 Strasbourg Cedex
France
E-mail: victor.cavillon@chru-strasbourg.fr

Anne Schneider
Department of Pediatric Surgery
Hôpitaux Universitaires de Strasbourg
1 Avenue Molière 67098 Strasbourg Cedex
France
E-mail: anne.schneider1@chru-strasbourg.fr
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Introduction

The urachus is a thin fibrous remnant of the allantois, a canal which runs between the top of the bladder and the umbilicus. It normally closes during the second part of pregnancy. Failure of closure of this communication results in various abnormalities; one of them are cysts, which can be detected on the umbilical cord using prenatal ultrasonography

[1]. However, an allantoic cyst with a patent urachus is extremely rare with an incidence of 3 per 1,000,000 live births in whom males are more often affected with a sex ratio of 3 boys for 1 girl [1].

We tried to identify a urological explanation to this pathology through 4 cases treated in our unit and a literature review in PubMed.

Material and Methods

A single centre review reported 4 cases of allantoic cyst with a patent urachus from 1999 to 2014. Retrospective data was collected and reviewed as follows: perinatal data, ultrasonography, clinical examination at birth, and urinary tract examination, type of surgical procedure, and outcome.

Descriptive data of the 4 cases are presented, and a review of the literature was performed using the following key words: "allantoic cyst" and "patent urachus". Only studies that talked about prenatal diagnosis were selected. We excluded those talking about urachus complications during childhood and after. We were looking for data about cyst size, rupture, congenital abnormalities, neonatal morbidity and surgery.

Case 1

A 34-year-old woman, gravida 3, para 2, was referred to our unit for the first trimester sonography. At 13 weeks of gestation (WG), the fetal bladder was enlarged and associated with a 16mm ventral cyst [**Fig. 1**]. During the 18-WG evaluation, the cyst size increased to 29mm. However, the size of the bladder was normal. The cyst's size was 30mm at 22 WG with an umbilical cord edema evidenced on Doppler. At the end of the second trimester, the allantoic cyst disappeared, the bladder was empty, and arterial flow on Doppler ultrasound was normal apart from an edema of the cord. She delivered a healthy 3,050g boy at term. There was a remnant bladder-umbilical communication through the cord.

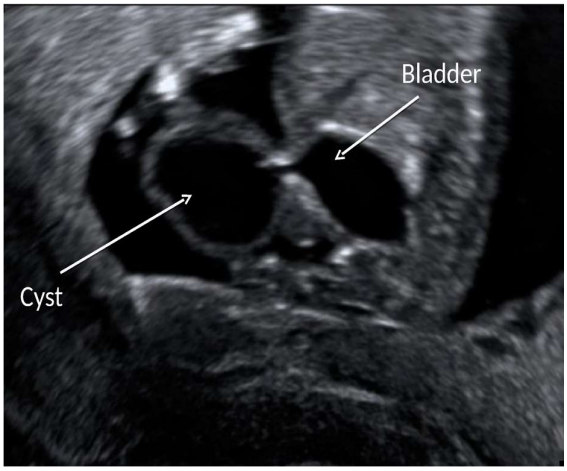

Fig. 1. Allantoic Cyst at 1st trimester.

Surgery was performed on the first day of life. Cystography and urethrocystoscopy revealed no bladder abnormality, no posterior urethral valve, and no vesicoureteral reflux (VUR). Nevertheless, cystoscopy revealed a marked bladder neck and a flattened veru montanum. The urachus was removed and umbilical vessels were ligated. Urethral catheter was left in place during 2 days and when removed, the boy had normal micturition. Post-natal ultrasound evaluation, 72 hours after surgery, confirmed normal kidneys, and normal bladder size one month later. One year later, micturition and urinary sonography were normal.

Case 2

A 23-year-old woman, gravida 2, para 1, had a first trimester ultrasound evaluation (16 WG). It revealed a 27mm ventral cyst and a normal bladder. It was not possible to distinguish between a vesico-allantoic cyst and bladder exstrophy. Sonography at 19, 23, and 31 WG confirmed the presence of an allantoic cyst,

which increased in size and measured 27mm, 28mm, and 30mm respectively [**Fig. 2**].

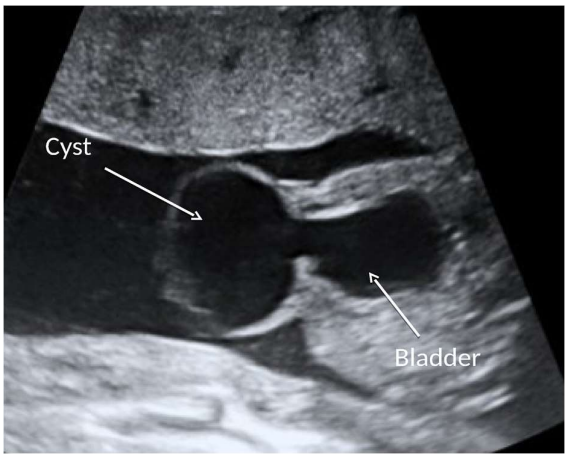

Fig. 2. Allantoic cyst at 2nd trimester.

Kidneys, bladder, and other organs were normal. The karyotype confirmed a 46 XY boy without chromosomal abnormality.

She delivered a baby boy weighing 3,470g at 39 WG. A bladder prolapse through the umbilicus was observed. Neonatal surgery using cystography and cystoscopy did not reveal any urinary explanation: normal bladder, and no posterior urethral valve (PUV). Excision-suture of the patent urachus was performed with success. We left urethral catheter during 4 days. Clinical and sonographic control one month and eleven months later showed normal micturition and sonography.

Case 3

A 34-year-old woman was seen for referral ultrasonography at 20 WG. A 30mm cyst could not be clearly distinguished between an allantoic cyst or an omphalocele.

She delivered a healthy boy with a 60mm umbilical cord cyst far from the abdominal wall. The umbilical cord was clamped without any further exploration. One month later, this boy was referred to pediatric surgery for a urinary leakage through the umbilicus. Surgical exploration revealed a communication with the bladder and no other abnormality. An excision of the urachus and suture of the top of the bladder were performed. We didn't leave urethral catheter. Histology confirmed the presence of an urachal cyst. The patient was discharged without any complication and a normal urinary ultrasonography. Clinical control 3 months later revealed no complication.

Case 4

A 27-year-old primipara was seen for referral ultrasound evaluation at 26 WG. A 20mm allantoic cyst was described.

A suspicious cord edema was noticed at birth. Ten days later, urinary leakage was suspected through the umbilicus. Pre-operative sonography revealed no kidney complication. The umbilicus could be catheterized. It revealed a communication with the bladder, which was excised and without any argument for an obstruction. We didn't leave any urethral catheter. Ultrasonography, one month later, revealed a small symmetric renal pelvic dilatation of 4mm with regression 6 months later. Three years after surgery, sonography revealed a small bilateral pyelectasis (4 mm); the micturition was normal.

Literature review

The literature review using PubMed with the words "allantoic cyst" and "patent urachus" gathered 12 corresponding studies between 1997 and 2013 gathering only 15 patients.

Among 15 cases, there were 11 male patients and 3 female patients. We did not find the information for one case. Mean maternal age was 31 years (range: 18 to 41). There were 14 singletons and one monochorionic biamniotic twin pregnancies. The diagnosis was made

between 12 and 27 WG. Cysts increased in size from the first to the second trimester. Prenatal cyst rupture occurred in 2/3 of cases. Seven prenatal abnormalities were described: 4 cases of enlarged bladder and 3 cases of bladder prolapse [Table 1].

STUDIES	MATERNAL AGE	PARITY	PREGNANCY	TERM AT DIAGNOSIS	CYST SIZE T1	CYST SIZE T2	CYST RUPTURE	SEX	PRENATAL URINARY ANOMALY
TOLAYMAT et al (1997)	34 yo	0	Singleton	19 WG	NA	50 mm	Yes	M	No
VAN DER BILT et al (2003)	38 yo	NA	Singleton	12 WG	15 mm	30 mm	Yes	F	Enlarged bladder
	31 yo	NA	Singleton	25 WG	Unseen	NA	Yes	M	No
SCHIESSER et al (2003)	32 yo	1	Singleton	14 WG	15 mm	58 mm	Yes	M	No
BUNCH et al (2006)	18 yo	0	Singleton	24 WG	Unseen	NA	No	M	No
MATSUI et al (2007)	34 yo	0	Singleton	14 WG	32 mm	NA	Yes	M	Bladder not seen / Exteriorized mass at umbilic
	36 yo	1	Singleton	19 WG	NA	40 mm	Yes	M	Bladder not seen / Exteriorized mass at umbilic
WEICHERT et al (2009)	21 yo	1	Singleton	14 WG	17mm	NA	Yes	M	Enlarged bladder at 20 WG
SEPULVEDA et al (2010)	31 yo	0	Singleton	12 WG	34 mm	80 mm	No	M	Enlarged bladder at 12 WG
	41 yo	2	Singleton	13 WG	45 mm	NA	NA	F	Enlarged bladder at 13 WG
BUREAU et al (2011)	29 yo	1	Twin MCBA	13 WG	30 mm	70 mm	Yes	M	No
GUPTA et al (2011)	19 yo	0	Singleton	27 WG	NA	37 mm	No	M	No
RASTEIRO et al (2012)	30 yo	0	Singleton	13 WG	33 mm	100 mm	No	NA	No
RAGA et al. (2012)	37 yo	0	Singleton	12 WG	18 mm	NA	Yes	M	Exteriorized mass at umbilic
MAZEAU et al (2013)	35 yo	1	Singleton	12 WG	26 mm	50 mm	Yes	F	No
CAVILLON et al (2016)									
Cas E.F.	34 yo	2	Singleton	13 WG	16 mm	30 mm	Yes	M	Enlarged bladder
Cas J.D.	23 yo	1	Singleton	16 WG	NA	28 mm	NA	M	No
Cas N.H.	34 yo	NA	Singleton	20 WG	NA	30 mm	No	M	No
Cas A.R.	27 yo	1	Singleton	26 WG	NA	20 mm	No	M	No

NA: Not Available

WG: Weeks of gestation

F: Female, M: Male

YO: Years old

MCBA: Monochorial biamniotic

At birth, one case of posterior urethral valve, 4 cases of bladder prolapse, 2 of vesicoureteral reflux, and one death (at 17 WG) with umbilical vessel thrombosis due to a cord edema were described [Table 2].

All newborns were operated on without complications [1-12].

STUDIES	POST NATAL MORPHOLOGIC ANOMALY	SUSPECTED DIAGNOSIS	NEONATAL COMPLICATIONS	EVOLUTION
TOLAYMAT et al (1997)	No	NA	No	Uneventful
VAN DER BILT et al (2003)	No	NA	No	Normal kidneys ultrasound
	No	NA	No	Normal kidneys ultrasound
SCHIESSER et al (2003)	No	NA	No	Uneventful
BUNCH et al (2006)	No	NA	No	Normal kidneys ultrasound
MATSUI et al (2007)	Bladder prolapse / bilateral VUR	NA	No	No VUR
	Bladder prolapse / bilatéral VUR	NA	No	Reduction of right VUR
WEICHERT et al (2009)	No	NA	No	Uneventful
SEPULVEDA et al (2010)	No	NA	No	Uneventful
	No	NA	Intra-uterine death at 14 WG	Umbilical vein thrombosis
BUREAU et al (2011)	PUV	PUV	No	Normal kidneys ultrasound
GUPTA et al (2011)	No	NA	No	Uneventful
RASTEIRO et al (2012)	NA	NA	NA	NA
RAGA et al. (2012)	Bladder prolapse	NA	No	Uneventful
MAZEAU et al (2013)	Bladder prolapse	NA	No	Abdominal muscle diastasis
CAVILLON et al (2016)				
Cas E.F.	Marked baldder neck and flattened veru montanum	Antenatal PUV	No	Uneventful
Cas J.D.	Bladder prolapse	NA	No	Uneventful
Cas N.H.	No	NA	No	Uneventful
Cas A.R.	No	NA	No	Uneventful

NA: Not Available

PUV: Postérior urethral valve

VUR: Vesicoureteral reflux

WG: Weeks of gestation

Discussion

In our series, prenatal diagnosis of allantoic cyst was established during the first and the beginning of the second trimester. All our fetuses were boys. Rupture of cysts occurred before birth in half of the cases (Table 1). In only one case, we could describe signs, which were in favor of a prenatal temporary obstruction of the distal urinary tract with a marked bladder neck and flattened veru montanum. The other cases did not bring any prenatal explanation. They did not have vesicoureteral reflux or other abnormality. We suspected, in the first case, one lower urinary tract obstruction that could have disappeared during pregnancy and that could explain this particular low urinary tract aspect and allantoic cyst, consequences of a high pressure in there. We founded no explanation for the 3 other patients.

Fetal MRI helps to differentiate between a real cyst, a pseudocyst, and an abdominal wall defect [1]. Pseudocysts are located in a paraxial situation. They are considered to be a result of Wharton's jelly degeneration. They are often associated with structural or chromosomal abnormalities such as trisomy 13 and 18 [13]. Additionally, "true" cysts preferentially lie in an axial situation between umbilical vessels and they are often related with normal fetuses. Bunch et al. propose the use of fetal MRI in case of uncertain diagnosis using sonography between "true" cyst and pseudocyst to gave parents the best information about fetal and neonatal risk. Systematic determination of prenatal karyotype is controversial. Indeed, Ross et al. propose that the search for chromosomal abnormalities should be offered in case of persistent cyst in the second and third trimester whereas Zangen et al. are more moderate in case of isolated anomaly [14]. Allantoic cysts with a patent urachus do not seem to be associated with a significant risk of chromosomal abnormality when isolated. Prenatal karyotype does not appear to be useful (including the complication of the sample), except in case of uncertain diagnosis.

In the reviewed studies, surgical management was the rule to prevent associated complications such as local infection, urinary infection, lithiasis or cancer. Teams often sutured umbilical vessels and excised the top of the bladder to prevent urachal cancer. Gleason et al. made a review of pediatric urachal anomaly and showed no severe risk of cancer with an extrapolated number required to treat more than 8,000 to prevent one hypothetic urachal adenocarcinoma [15]. On the other hand, Galati et al. observed a spontaneous resolution with non-operative management in patients younger than 6 months with small fistula [16]. Urethrocystogram and cystoscopy appeared to be essential in causal diagnosis as seen in our first case with suspected prenatal anomalies and to explore the possibility of posterior urethral valves.

It seems to be important to look for urinary tract evolution (sonography) and toilet training forfeiture. Indeed, Freedman and al. showed that some children with low urinary tract obstruction were not toilet-trained after 2 years old [17]. In our study, we had many patients lost to follow-up before toilet training, so we were not able to compare this parameter with literature. Sonography evolution was normal.

Conclusions

Allantoic cyst with a patent urachus is a rare pathology. Most of the time, no etiology is found but urethrocystogram and cystoscopy is often performed to look for posterior urethral valves or other urinary tract abnormality. Our

study has just suspected one case of prenatal lower urinary tract obstruction with a spontaneously correction. Surgery prevents complications such as infections or degeneration. There is no distinguished structural or chromosomal anomaly, which can substantiate systematic karyotype. Surgical management during the neonatal period can be discussed. However, medical supervision seems to be an acceptable alternative. In all cases, the neonatal evolution was normal with no surgical or kidney complications. Nonetheless, it is essential to follow these patients regularly until they are toilet-trained.

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References

[1]Bunch PT, Kline-Fath BM, Imhoff SC, Calvo-Garcia MA, Crombleholme TM, Donnelly LF. Allantoic cyst: a prenatal clue to patent urachus. Pediatr Radiol. 2006;36(10):1090–5.

[2]Van Der Bilt JDW, Van Zalen RM, Heij HA, Bilardo CM, Aronson DC. Prenatally diagnosed ruptured vesico-allantoic cyst presenting as patent urachus at birth. J Urol. 2003;169(4):1478–9.

[3]Bureau M, Bolduc S. Allantoic cysts and posterior urethral valves: a case report. Ultrasound Obstet Gynecol. 2011;38(1):116–8.

[4]Matsui F, Matsumoto F, Shimada K. Prenatally diagnosed patent urachus with bladder prolapse. J Pediatr Surg. 2007;42(12):e7–10.

[5]Rasteiro C, Ramalho C, Loureiro T, Pereira J, Matias A. Bladder emptying into an umbilical cord cyst: prenatal sonographic sign of allantoic cyst with patent urachus. Ultrasound Obstet Gynecol. 2013;42(2):239–40.

[6]Gupta N, Corbett H, Ismail R, Sathanantharajah R, Sivakumar S, McCarthy L. Allantoic cyst–an unusual umbilical cord swelling. J Surg Case Rep. 2011(4):5.

[7]Tolaymat LL, Maher JE, Kleinman GE, Stalnaker R, Kea K, Walker A. Persistent patent urachus with allantoic cyst: a case report. Ultrasound Obstet Gynecol. 1997;10(5):366–8.

[8]Weichert J, Chiriac A, Kaiser M, Thorns C, Axt-Fliedner R. Prenatal management of an allantoic cyst with patent urachus. Arch Gynecol Obstet. 2009;280(2):321–3.

[9]Sepulveda W, Rompel SM, Cafici D, Carstens E, Dezerega V. Megacystis associated with an umbilical cord cyst a sonographic feature of a patent urachus in the first trimester. J Ultrasound Med. 2010;29(2):295–300.

[10]Mazeau P, Curinier S, Kandem-Simo A, Delabaere A, Laurichesse H, Lemery D et al. Prenatal diagnosis and evolution of patent urachus. J Gynecol Obstet Biol Reprod. 2014;43(59:393–6.

[11]Schiesser M, Lapaire O, Holzgreve W, Tercanli S. Umbilical cord edema associated with patent urachus. Ultrasound Obstet Gynecol. 2003;22(6):646–7.

[12]Raga F, Bonilla-Musoles F, Castillo JC. SonoAVC: a new tool in early diagnosis of patent urachus with bladder prolapse. Ultrasound Obstet Gynecol. 2012;39(2):241–2.

[13]Ross JA, Jurkovic D, Zosmer N, Jauniaux E, Hacket E, Nicolaides KH. Umbilical cord cysts in early pregnancy. Obstet Gynecol. 1997;89(39:442–5.

[14]Zangen R, Boldes R, Yaffe H, Schwed P, Weiner Z. Umbilical cord cysts in the second and third trimesters: significance and prenatal approach. Ultrasound Obstet Gynecol. 2010;36(39:296–301.

[15]Gleason JM, Bowlin PR, Bagli DJ, Lorenzo AJ, Hassouna T, Koyle MA et al. A comprehensive review of pediatric urachal anomalies and predictive analysis for adult urachal adenocarcinoma. J Urol. 2015;193(2):632–6.

[16]Galati V, Donovan B, Ramji F, Campbell J, Kropp BP, Frimberger D. Management of urachal remnants in early childhood. J Urol. 2008;180 (4 Suppl):1824–7.

[17]Freedman AL, Johnson MP, Smith CA, Gonzalez R, Evans MI. Long-term outcome in children after antenatal i ntervention for obstructive uropathies. The Lancet. 1999; 31;354(9176):374-7.

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