AN UNUSUAL COMPLICATION OF AN INVASIVE INTRAUTERINE THERAPY: 6 YEAR FOLLOW-UP

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Abstract

Shunt implantation in utero being a life-saving procedure, may be connected with a wide range of complications. One of them is catheter dislocation, a side effect which may impair the state of the fetus and child. We present a case of a boy with Congenital Adenomatoid Lung Malformation (CALM) type II. Invasive prenatal therapy, complicated by shunt migration did not influence our patient's health. Moreover, the abnormally located shunt stayed within the child's chest up to the age of 6 years. No symptoms linked to foreign body presence were observed. Although similar cases were published, we report a situation, in which the catheter was left in the patient's body for 6 years, for the first time..

Key words: CALM, Cystic Adenomatoid Lung Malformation, prenatal invasive therapy, shunt migration, foreign body

INTRODUCTION

Development of prenatal diagnostics and therapy brings success, failure or both. There is a shortage of information in the literature about the complications and failures. We describe an atypical complication after invasive therapy in utero with a 6-year follow up.

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CASE REPORT:

A 34-year-old gravida was referred to the hospital at 24 weeks of gestation, following the detection of lung anomalies. Ultrasonographic examination revealed cystic lesions up to 2 cm in diameter and hyperechogenic areas in the right enlarged lung. The final diagnosis of right unilateral CALM type II was established at the 25 week ultrasound. At 27 weeks of gestation, after thorough counseling of the patient, decompression of the fluid within the right lung was performed with simple needle aspiration (intervention: K.Sz.). At 28 weeks, a right pulmonary-amniotic double pig-tail catheter shunt was placed because of re-accumulation of fluid. After the intervention, ECHO examination showed that the fetus had stable cardiovascular function with mild right ventricular systolic and left ventricular diastolic dysfunction. USG + ECHO examination at 32 weeks' gestation confirmed normal biometric values, reduced cystic areas and improvement of haemodynamic state. At 39 weeks, a male infant weighing 3840g, with Apgar scores of 10, was delivered by elective cesarean section. The shunt was not located in either the neonate's chest wall nor within the placenta. After chest X-Ray examination, the shunt catheter was found in the right pleural cavity (Figure 1). In the neonatal, pediatric and preschool period no symptoms associated with the presence of a foreign body were noted. At the age of 6 years (before the school year) – (Figure 2), the decision for thoracoscopic shunt removal was made. The foreign body was removed under general anesthesia, and the clinically asymptomatic patient was discharged after 3 days of hospitalization.

COMMENT

Cystic Adenomatoid Lung Malformation (CALM) is a rare congenital malformation. The prevalence of this anomaly varies between 1:25 000 – 1:35 000 pregnancies and comprises more than 80% of fetal thoracic lesions[1,2]. Due to the variety of sonographic appearances of those lesions, a classification was developed[3] based on differences between the diameter of cysts and the place of origin. The prognosis is variable; lesions may regress or a cystic area may expand leading to pulmonary hypoplasia of the affected lung or non-immune fetal hydrops in approximately 40% of cases[2,4].

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Table 1. Statement of described complications after invasive therapy.

Year	Author	Diagnosis	Week of gestation**	Fetus follow-up	Delivery & Birth	Follow-up of the baby
2005	W.Sepulveda et al.	Left unilateral pleural effusion	22 26 34	Shunt migration into fetal chest, re-accumulation of fluid not observed	34 weeks Vaginal birth 2300g Apgar scores of 9 and 10 at 1 and 5 minutes	Confirmation of shunt dislocation, mild respiratory distress syndrome; age of 2 years asymptomatic; shunt still within the patient's chest
			27 30 39	Pleural effusion and mediastinal shift regression; shunt migration into fetal chest	39 weeks CS* 3400gApgar scores of 9 and 10 at 1 and 5 minutes	Confirmation of shunt dislocation; at age of 10 months asymptomatic; shunt still within the patient's chest
		Bilateral pleural effusion	24 30 (on the right) 33 (on the left) 35	Improvement in the right pleural cavity; in the left increasing effusion and subcutaneous edema; shunt dislocation into left pleural cavity	35 weeks CS*(Preterm rupture of membranes, preterm labor, preterm delivery) 3030g Apgar scores of 2 and 3 at 1 and 5 minute	Mechanical ventilation, left chest tube insertion and 30-day long hospitalization; at 2 years of age asymptomatic; shunt still within the patient's chest
1998	K.M.Lewis et al.	Fetal obstructive uropathy	19.6 30.6	Thrice implantation of a vesico-amniotic shunt with double shunt migration, evisceration, Preterm rapture of membranes, preterm labour and preterm delivery	30.3 Preterm rupture of membranes 30.6 vaginal birth 1700g Apgar scores of 2,5 and 6 at 1, 5 and 10 minutes	Evisceration, Massive bladder distention, bilateral intra-abdominal undescended Testes, prune-belly syndrome; Surgical intervention on the day of delivery: closure of abdominal wall defect with creation of a temporary vesicostomy; initial pulmonary and renal dysfunction with recovery; discharged on 61st day of postnatal life
2005	J.C.Shin et al.	Posterior urethral valves; Lower urinary tract obstruction	16 17 33	Shunt migration with increasing oligohydramnios	33 weeks CS* 3570g Apgar stores of 8 and 9 at 1 and 5 minutes	Newborn in good clinical condition, bilateral mild dilatation of renal pelvis on USG examination
2010	A.Springer et al.	Lower urinary tract obstruction (LUTO)	18 27 29	Shunt migration, moderate bilateral hydronephrosis and moderate urinary ascites	29 weeks CS* 1700g	Shunt migration led to development of fistula between fetal bladder and fetal peritoneal cavity; The newborn required mechanical ventilation; Continuity of fetal bladder wall was recreated; laboratory and clinical parameters remain normal

**CS = cesarean section*
*** Week of gestation, in which the diagnosis was made*
Week of gestation, in which the procedure of shunt implantation was made
Week of gestation, in which the infant was delivered

The case described demonstrated features of CALM type II with numerous cysts up to 2 cm in diameter. It is frequently associated with other developmental malformations such as pulmonary hypoplasia, heart failure, polyhydramnios and mediastineal shift[3, 5]. This malformation may be treated by puncture and decompression of the fluid but the outcome of this procedure may be transitory[6]. Such a situation was presented in our case, and re-accumulation of the fluid was the indication for shunt placement, allowing one of the cysts in the chest to drain into the amniotic cavity. Haemodynamic improvement was observed and the fetus remained stable. The ECHO examination showed only subtle cardiac dysfunction, insignificant from the clinical point of view and the newborn was born in good condition. The presence of the shunt within the infant's right pleural cavity was confirmed, a result of catheter migration.

Table 1. presents published cases pertaining to shunt migration after invasive fetal therapy. Similar to our case, were 3 cases described by Sepulveda, with good condition of the infants who were clinically asymptomatic up to the age of 2 years. Unfortunately further follow up is unknown for these infants. In contrast to the published cases, we present thoracoscopic removal of prenatal implanted shunt for the first time with a long term good outcome.

Malposition of the shunt may also be a complication following other fetal interventions, such as therapies for fetal uropathies. In these cases, the consequence of interruption of fetal organs or body continuity resulted in a fistula (Table1)[7,9].

In other cases, shunt dislocation and its migration into amniotic cavity may cause a situation, in which the shunt does not fulfill its task and is "delivered" with the baby[11].

In our case, despite the asymptomatic result of dislocation of the shunt after in utero intervention, the decision for minimally invasive thoracoscopic removal of the foreign body was made before the beginning of the school year and possible increase of physical activity, which is consistent and supported by the literature[12].

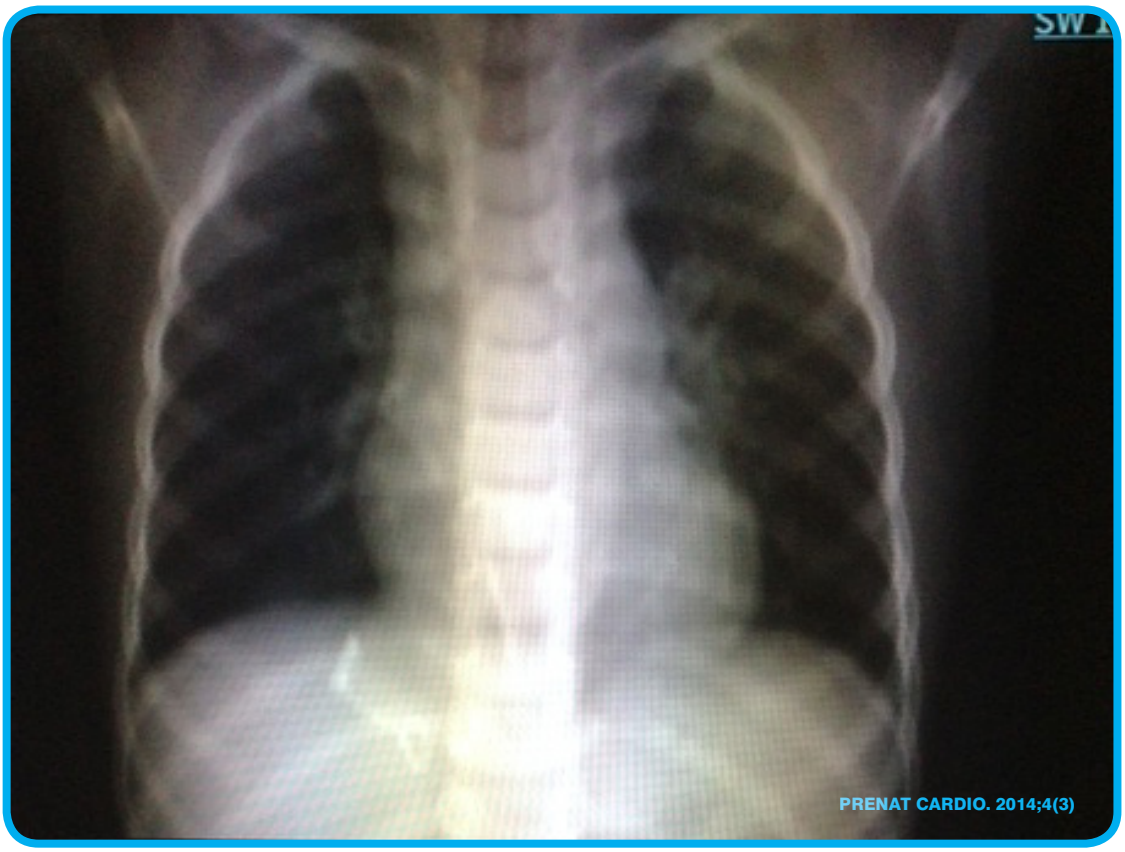

Figure 1. X-ray showing foreign body.

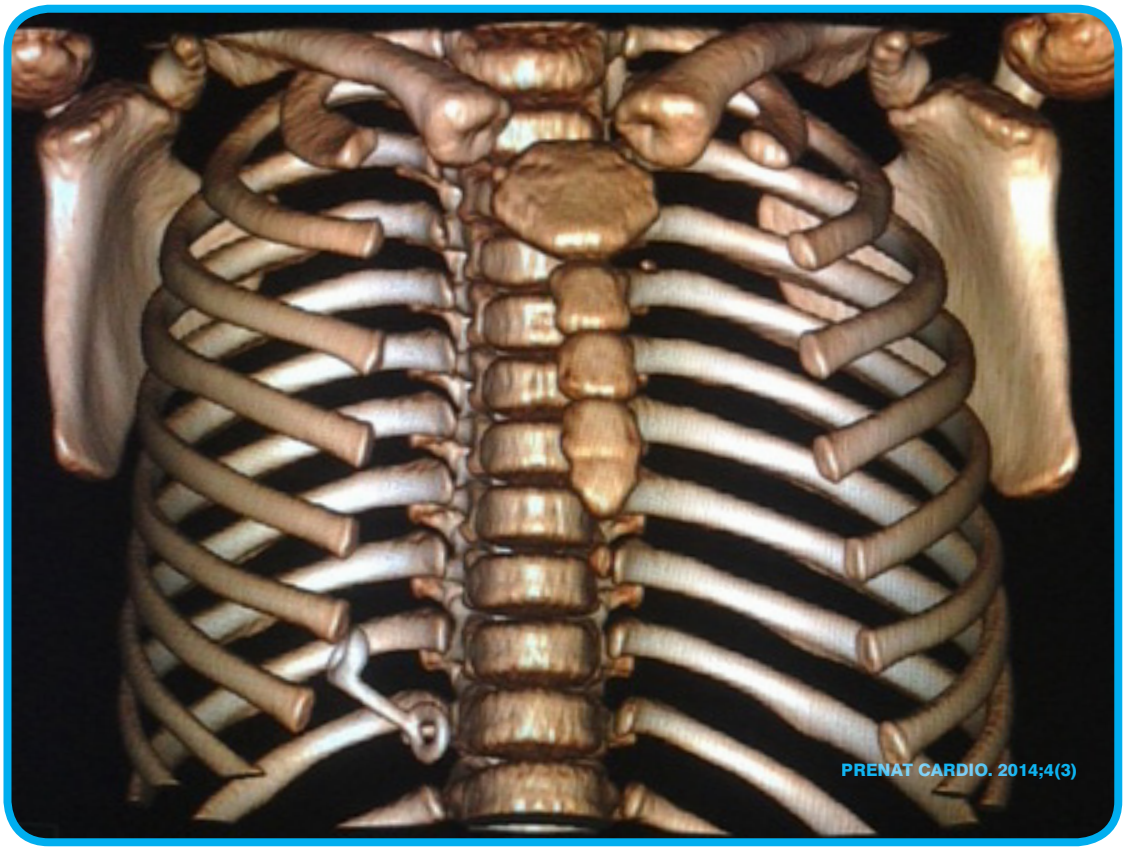

Figure 2. CT with foreign body.

CONCLUSION

One method for treating cystic adenomatoid lung malformation may be shunt implantation. This procedure may result in various complications including shunt migration, which may warrant later retrieval even if asymptomatic.

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Conflict of interest

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