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Original article

Pulmonary sequestration: an experience in a level III hospital

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Abstract

Background: Pulmonary Sequestration (PS) is a rare congenital malformation, with few cases reported. Thus, the prenatal and postnatal natural history of PS and the best management approach have not been fully characterized.

The aim of this study is to evaluate the experience with PS in our level III hospital, to improve our practice.

Methods: A retrospective review of all cases with PS admitted to our hospital between January 1996 and December 2016 was performed.

Results: Fifteen cases of PS were identified. Antenatal ultrasonography evaluation suggested the existence of 3 (20%) PS, 3 (20%) Congenital Pulmonary Airway Malformations (CPAM), 3 (20%) cases with no established diagnosis between CPAM and PS and 1 (6.7%) neuroblastoma. All newborns were admitted to Neonatal Intensive Care Unit and 7 (46.7%) became symptomatic. When postnatal X-ray and/or computer tomography scan were performed, the lesions corresponded to 12 (80%) PS, 1 (6.7%) CPAM, 1 (6.7%) suprarenal mass and 1 (6.7%) congenital diaphragmatic hernia. Eight (53.3%) cases had concomitant abnormalities. An invasive postnatal intervention was performed in 5 of 7 (71.4%) symptomatic patients and in 4 of 8 (50%) asymptomatic ones. Two patients underwent embolization. Surgery was performed in 8 (53.3%) cases (thoracotomy in 3 and thoracoscopy in 5). Respiratory morbidity was reported in 2 asymptomatic cases conservatively managed. Final diagnosis, based on histopathological examination or postnatal imaging when surgery wasn't applied, was: 9 (60%) extralobar sequestrations and 3 (20%) intralobar sequestrations; in 3 (20%) cases, it wasn't possible to confirm the type of lesion.

Conclusion: PS was associated to a high rate of congenital abnormalities. Concordance between prenatal and postnatal findings was poor. According to

our and other series, treatment of asymptomatic PS is controversial. Nevertheless an elective surgery should be preferred to prevent the risks of an urgent surgery. The outcome was generally good.

Keywords

Pulmonary sequestration, Neonatal Intensive Care Unit, pulmonary malformation, lung, prenatal diagnosis, newborn.

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Introduction

Pulmonary Sequestration (PS) is a rare pulmonary malformation characterized by a non-functioning and isolated portion of lung that receives its blood supply from one or more anomalous systemic arteries, rather than from the pulmonary circulation [1]. An aberrant arterial supply to the lung tissue was first described by Huber in 1777, and later, in 1946, Pryce introduced the term "sequestration", derived from the Latin term *sequestrare* (to separate) [2].

After Congenital Pulmonary Airway Malformation (CPAM), PS is the second most common congenital lung malformation, representing up to 6% of the reported congenital lung malformations, with no published population incidence [3]. The aetiology of this lesion is still not well understood, but the most commonly accepted embryological theory is the formation of an accessory lung bud in a distal position to the normal lung buds, occurring between the 4th and 8th gestational weeks [1].

PS is classified in two types based on their pleural investment: extralobar (ELS) and intralobar (ILS). ELS are masses of lung tissue that have their own pleural covering; so, they are anatomically separated from the surrounding normal lung. ILS share the pleural covering with the normal parenchyma; therefore, they are contiguous with the lung [3, 4]. A subset of PS share anatomical and histological features with CPAM and they have been described as hybrid lesions. This suggests that both CPAM and PS have similar embryological origin [5, 6].

With an increasing use and high quality prenatal imaging, PS is commonly diagnosed antenatally, usually at the 20th-22nd gestational week routine ultrasonography (US) [7-9]. The US imaging reveals a hyperechoic, solid, well-defined and homogeneous mass. The diagnosis can be verified by documenting an abnormal vascular supply, with Colour Doppler [10]. Additionally, a magnetic resonance imaging (MRI) could be suitable when vessels are not recognized in the US. In MRI, a well-defined mass with hypersignal at T2-weighted sequence is distinctive [11]. A postnatal imaging method should be performed to confirm the diagnosis, using an X-ray, followed by a contrast-enhanced computer tomography (CT) scan or a MRI [9, 12].

In newborns, there is a spectrum of different clinical presentations, from the asymptomatic to respiratory morbidity, which includes respiratory distress, recurrent infections or feeding difficulties. Therefore, the postnatal management of PS is of particular clinical importance [3]. When the newborns are symptomatic, the need for a surgical approach is consensual. However, the best approach in asymptomatic cases is controversial: while some clinicians defend a conservative approach through successive follow-up, others prefer an earlier surgical procedure with resection of the lesion [13-15].

The aim of this study is to evaluate the experience with PS in our level III hospital, in order to improve our practice in a prenatal diagnosis and clinical management of these patients.

Material and methods

We conducted a 20-year retrospective study. Cases of PS or hybrid lesions were selected from the newborn nursery and the Neonatal Intensive Care Unit (NICU) at "Centro Hospitalar São João", Porto, from January 1996 to December 2016, if they had suspicion of PS on prenatal US and/or a postnatal diagnosis of PS during the neonatal period. The description of the imaging examinations performed on the newborn or the histopathological examination in the cases submitted to surgery were analysed to establish the diagnosis. The follow-up was at least 12 months.

The charts of the neonates and their mothers were analysed and the following data were collected:

maternal age, gestational age at the time of diagnosis, prenatal US and MRI features, foetal interventions, gestational age at birth expressed in complete weeks, gender, birthweight, multiple pregnancy, pregnancy complications (pleural effusion, foetal hydrops or polyhydramnios), affected lung, type of lesion (intralobar or extralobar), need for NICU admission, final diagnosis, associated malformations, neonatal symptoms at delivery, including need for reanimation, ventilation or oxygen at birth, respiratory distress, respiratory infections, timing of surgery, type of surgery, histological diagnosis, morbidity and overall mortality.

This study was approved by the "Centro Hospitalar São João" Ethics Committee.

Data were collected by the same researcher and performed using SPSS Statistics® for Windows®, version 24.0. Due to the size of the sample, it was only possible to conduct a descriptive analysis.

Results

Fifteen neonates with PS were identified. Antenatal US evaluation suggested the existence of 3 (20%) cases of PS, 3 (20%) CPAM, 3 (20%) cases with no established diagnosis between CPAM and PS and 1 (6.7%) neuroblastoma. Five (33.3%) cases didn't have a prenatal diagnosis. **Fig. 1** presents a PS antenatally diagnosed with an US, corresponding to case no. 11. In 5 of these 10 cases, a prenatal MRI was performed in order to confirm the presence of the lesion. Two patients (cases no. 8 and 9) had further invasive prenatal testing, which showed a normal karyotype. The median gestational age at diagnosis of the foetal lesion was 22 weeks (range from 20 to 25 weeks). There were no instances of foetal hydrops, polyhydramnios or pleural effusion. Case no. 14 received prenatal corticosteroids therapy (preterm birth). Foetal intervention was not performed in any of the patients.

A summary of the data regarding the 15 newborns in the perinatal period is presented in **Tab. 1**. The median gestational age at delivery was 39 weeks (29-41 weeks), with a median birthweight of 2,970 grams (790-4,760 grams). There were no cases of foetal death or terminations of pregnancy. Eleven (73.3%) patients were male and 4 (26.7%) were

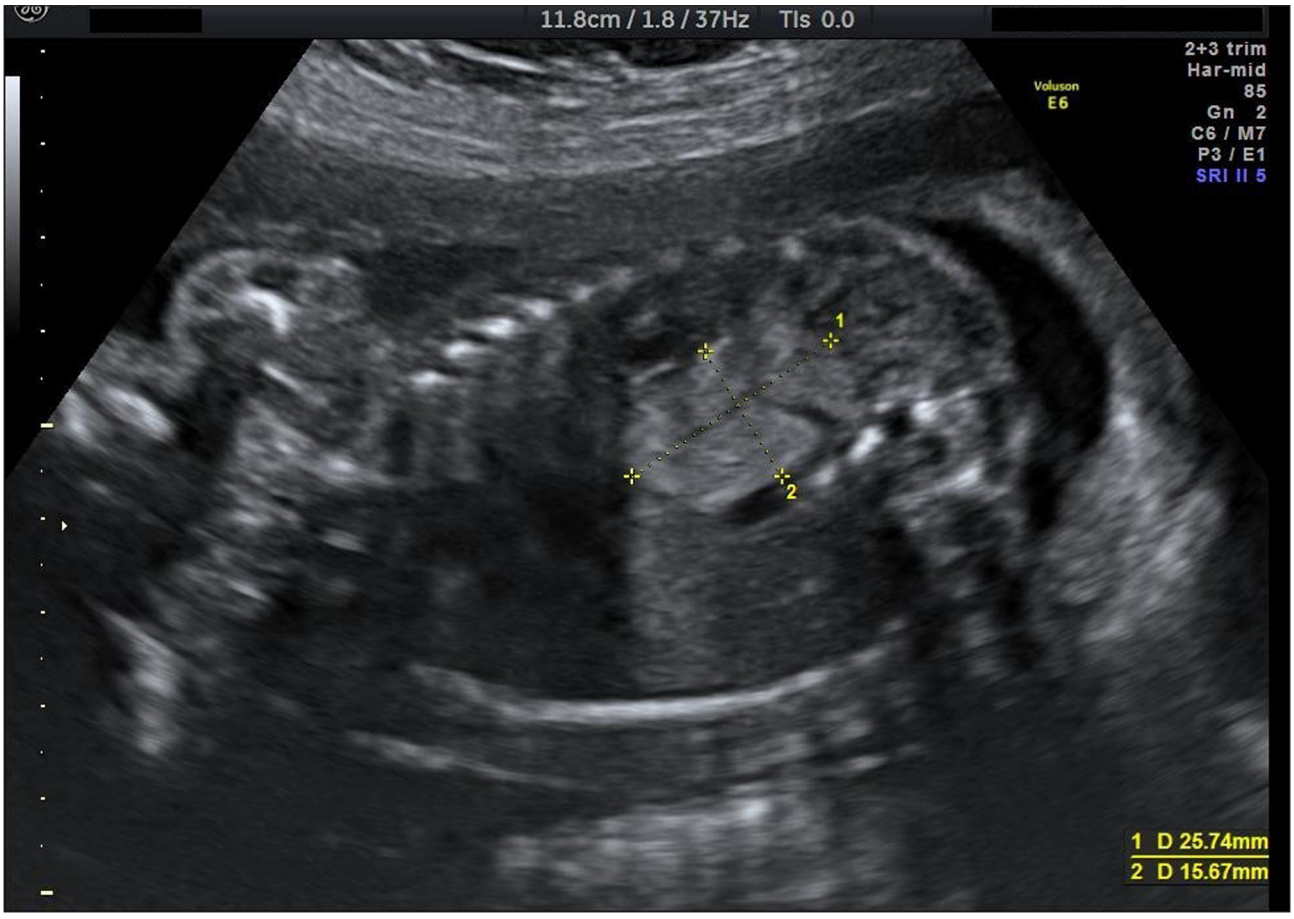

Figure 1. Extralobar Sequestration (ELS) – prenatal ultrasound of a foetus with 23 weeks of gestation showing an echogenic infradiaphagmatic mass on the left side (case no. 11).

female. Four (26.7%) cases were preterm birth, 3 of them corresponded to twin pregnancies. Case no. 14 (preterm of 29 weeks) needed reanimation, oxygen and ventilation in the delivery room.

Data on prenatal and postnatal diagnosis, NICU admission and associated abnormalities are reported in **Tab. 2**. All newborns were admitted to NICU. Clinically, 7 (46.7%) cases were symptomatic during the NICU stay: 2 (28.6%) presented with respiratory distress and 5 (71.4%) presented with both respiratory distress and feeding difficulties. Four patients needed oxygen and/or ventilation.

Table 1. Perinatal data.

Case no.	Year	Maternal age	Prenatal diagnosis	Gestational age at diagnosis (weeks)	Gender	Gestational age at birth (weeks)	Weight at birth (g)
1	2001	28	Neuroblastoma	24	M	40	3,460
2	2007	26	No	-	M	38	4,760
3	2009	20	No	-	F	39	2,838
4	2011	36	CPAM	22	M	36	2,550
5	2011	28	CPAM	21	M	41	3,620
6	2012	17	PS	22	M	39	2,970
7	2012	32	CPAM/PS	22	M	39	3,240
8	2013	35	CPAM/PS	21	F	41	3,630
9	2013	33	CPAM/PS	22	F	39	3,445
10	2013	35	CPAM	20	M	39	2,990
11	2015	38	PS	23	F	37	2,115
12	2016	49	No	-	M	36 (2nd twin)	1,865
13	2016	38	No	-	M	39	2,915
14	2016	36	No	-	M	29 (2nd twin)	790
15	2016	33	PS	25	M	36 (2nd twin)	1,970

CPAM: congenital pulmonary airway malformation; PS: pulmonary sequestration.

Table 2. Prenatal and postnatal diagnosis.

Case no.	Prenatal diagnosis	Admitted to NICU	Imaging postnatal diagnosis [a]	Affected lung	Associated congenital abnormalities
1	Neuroblastoma	Yes	Suprarenal mass	Left	No
2	No	Yes	PS	Right	CDH
3	No	Yes	CDH	Right	CDH
4	CPAM	Yes	ELS	Left	No
5	CPAM	Yes	Type II CPAM	Right	Cardiac abnormalities
6	PS	Yes	ELS	Right	No
7	CPAM/PS	Yes	ELS	Left	No
8	CPAM/PS	Yes	ILS	Right	No
9	CPAM/PS	Yes	ILS	Left	Cardiac abnormalities
10	CPAM	Yes	ILS	Right	Cardiac abnormalities
11	PS	Yes	ELS	Left	No
12	No	Yes	PS	Right	Cardiac abnormalities
13	No	Yes	ELS	Right	Polymarformative syndrome
14	No	Yes	ELS	Right	Cardiac abnormalities
15	PS	Yes	Extralobar hybrid lesion	Left	No

NICU: neonatal intensive care unit; PS: pulmonary sequestration; CDH: congenital diaphragmatic hernia; CPAM: congenital pulmonary airway malformation; ELS: extralobar sequestration; ILS: intralobar sequestration.
[a] Post-natal diagnosis based on post-natal X-ray and/or CT scan.

When postnatal X-ray and/or CT scan were performed, in 12 (80%) cases the lesions corresponded to PS, in 1 (6.7%) case to CPAM (type II, according to Stocker classification) and in another case (6.7%) to a suprarenal mass. Within the 12 cases of PS, 6 (50%) cases were classified as ELS, 3 (25%) were ILS, 1 (8.3%) was a hybrid lesion with features of both CPAM and ELS, and in 2 (16.7%), it was not possible to confirm the type of lesion. Case no. 3 had only been accidentally diagnosed with a PS during the surgery for a right sided congenital diaphragmatic hernia (CDH). Lesions were on the right side in 9 (60%) cases and on the left side in 6 (40%). **Fig. 2** and **Fig. 3** present, respectively, an MRI and a CT angiography performed in case no. 11 in order to confirm the diagnosis.

In 8 (53.3%) cases, concomitant abnormalities had been reported: 5 (62.5%) cases with cardiac malformations, 2 (25%) with a right sided CDH and 1 (12.5%) case with a polymalformative syndrome (ostium secundum-type interatrial communication, renal pelvis dilatation, horseshoe kidney and vertebral and costal defects). Within the cases with associated cardiac abnormalities, cases no. 5, 10 and 14 corresponded to patent foramen ovale, case no. 9 had heart failure with an interventricular communication and a patent foramen ovale and case no. 12 had an ostium secundum-type interatrial communication. Additionally, during this follow-up time, case no. 12 had recurrent respiratory infections and case no. 7 had two wheezing episodes.

Postnatal management of the patients is reported in **Tab. 3**. An invasive postnatal intervention was performed in 5 (71.4%) symptomatic patients (cases no. 12 and 14 are still under evaluation). In the asymptomatic ones, 4 (50%) were managed conservatively and the other 4 (50%) were managed invasively. Two (13.3%) patients underwent an embolization of the feeding vessel. Surgical resection was performed in 8 (53.3%) cases: 3 (37.5%) by thoracotomy and the other 5 (62.5%) by thoracoscopy. Three newborns underwent surgery in the first week of life and the remaining 5 patients between 3 months and 3 years of age. The

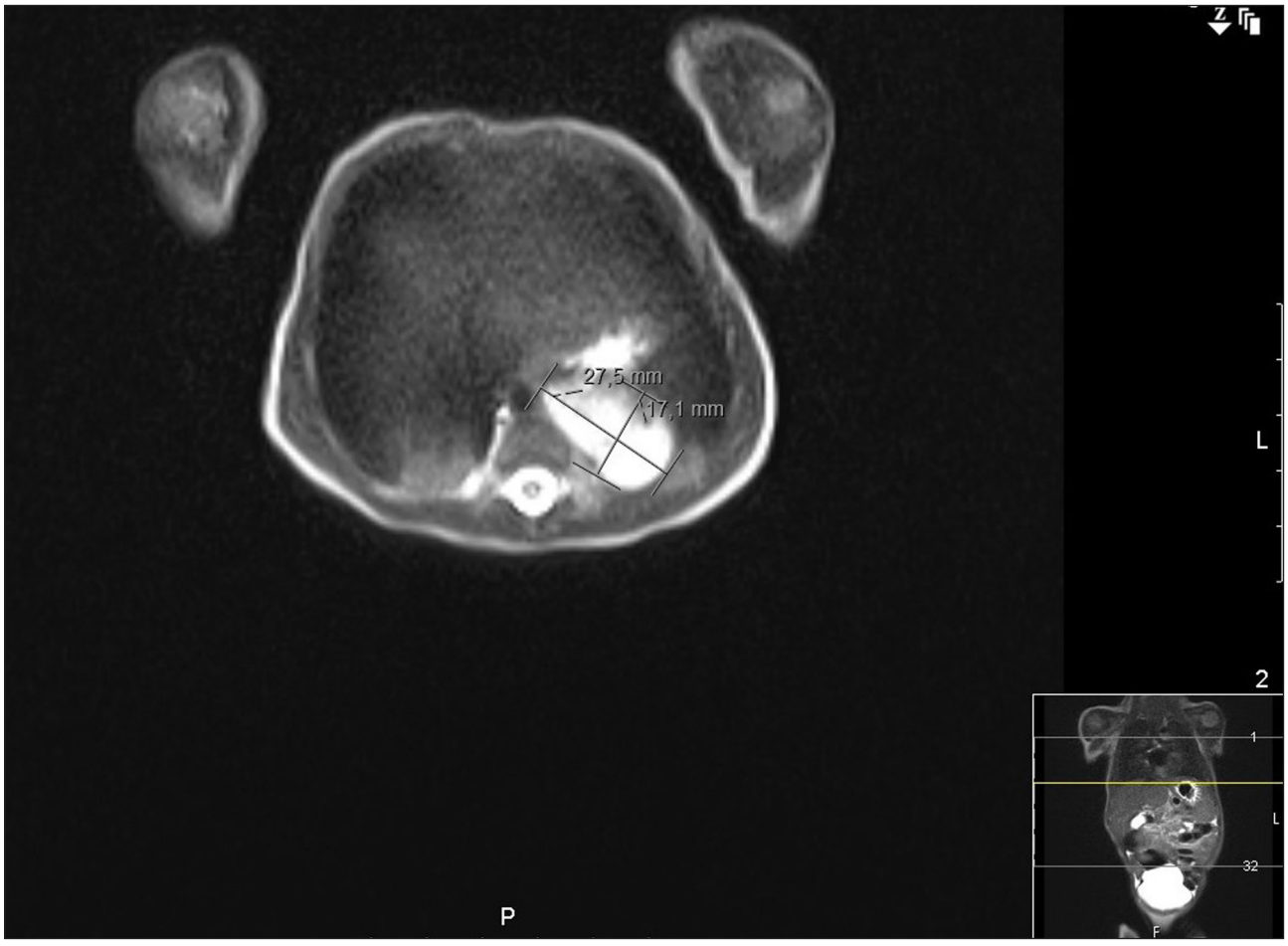

Figure 2. Extralobar Sequestration (ELS) – postnatal magnetic resonance imaging showing a left paravertebral homogeneous mass with hypersignal at T2-weighted sequence (case no. 11).

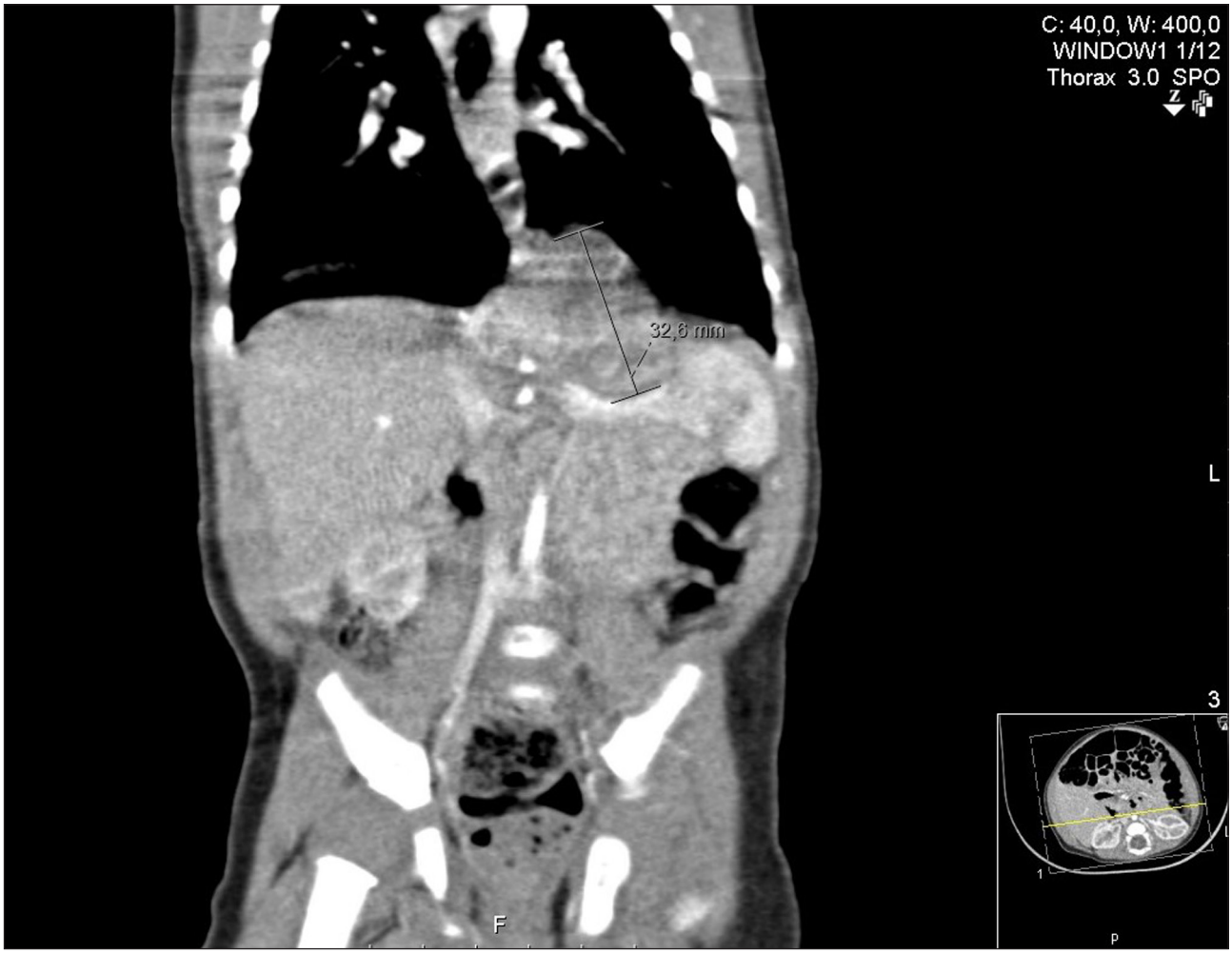

Figure 3. Extralobar Sequestration (ELS) – postnatal computed tomography angiography showing a solid mass on the left side (case no. 11).

median length of hospital stay was 5 days (range from 2 to 104 days) in the surgical managed group. After histopathological examination of the lesions, the lesions were classified as ELS (3 cases), intra-abdominal ELS (1 case) and ILS (2 cases), 1 of which with associated CPAM features. In 2 cases, it was not possible to establish a definitive diagnosis of the type of lesion. Post-operative follow-up was available for all patients ranging from 1 to 17 years of age. No post-operative morbidity was recorded.

Final diagnosis, based on histopathological diagnosis or imaging evaluation when a surgical procedure was not performed, is presented in **Tab. 3** and, in our series, there were 9 (60%) ELS, 3 (20%) ILS and in 3 (20%) cases, it was not possible to confirm the type of lesion.

The complications in this group of fifteen were two wheezing episodes in case no. 7 and respiratory infections in case no. 12, both managed conservatively, and a post-surgical pectus excavatum in case no. 2. In our series, no deaths were recorded.

Discussion

This study evaluated 15 cases of confirmed PSs observed in this NICU over a 20-year period. PS is a rare disease and this number is probably overestimated, since our hospital is a referral centre for management of congenital lung malformations. Therefore, some cases are referred to this hospital to ensure that the birth takes place in a tertiary neonatal care unit. According to a study performed in our hospital, respiratory abnormalities corresponded to 3.0% of all congenital abnormalities detected [16].

In our series, two thirds of the neonates had a prenatal US diagnosis of a thoracic mass. Prenatal detection is increasingly frequent due to routine foetal malformation scanning, in addiction to improvements in imaging resolution. On the other hand, a recent study has proved that the true incidence of congenital lung malformations has been increasing over the last ten years [17].

Prenatal detection is usually performed during the 20-22 week routine US [7-9] and our results

Table 3. Postnatal management of Pulmonary Sequestration (PS) cases.

Case no.	Symptoms at NICU	Postnatal intervention	Timing of surgery	Type of surgery	PS type [a]	Morbidity	Final diagnosis [c]
1	No	Surgery	6 days	Thoracotomy	intra-abdominal ELS	No	ELS
2	Yes	Surgery	6 days	Thoracotomy	ELS	Pectus excavatum	ELS + CHD
3	Yes	Surgery	6 days	Thoracoscopy	N/A	No	PS + CDH
4	No	Surgery	29 months	Thoracoscopy	ELS	No	ELS
5	Yes	Surgery	14 months	Thoracoscopy	ILS and CPAM	No	ILS + CPAM
6	No	No	-	-	-	Lost follow-up	ELS
7	No	No	-	-	-	Wheezing episodes	ELS
8	No	Embolization of the feeding vessel	-	-	-	No (under follow-up)[b]	ILS
9	Yes	Embolization of the feeding vessel and surgery	29 months	Thoracoscopy	ILS	No	ILS
10	No	Surgery	13 months	Thoracoscopy	ELS	No	ELS
11	No	No	-	-	-	No (under follow-up)[b]	ELS
12	Yes	No	-	-	-	Respiratory infections	PS
13	Yes	Surgery	3 months	Thoracotomy	N/A	No	PS
14	Yes	No	-	-	-	No (under follow-up)[b]	ELS
15	No	No	-	-	-	No (under follow-up)[b]	ELS + CPAM

NICU: neonatal intensive care unit; PS: pulmonary sequestration; ELS: extralobar sequestration; CDH: congenital diaphragmatic hernia; N/A: not available; ILS: intralobar sequestration; CPAM: congenital pulmonary airway malformation.
[a] Histological diagnosis; [b] cases no. 8, 11, 14 and 15 are still under evaluation in regular follow-up visits in our hospital, with no respiratory symptoms recorded up to the end of this study; [c] based on histopathological diagnosis or imaging evaluation (X-ray and/or CT) when a surgical procedure was not applied.

support this information, as all diagnosis occurred between 20 and 25 weeks of pregnancy.

According to our data, the diagnostic agreement among prenatal and postnatal imaging modalities and subsequent histopathological analysis is limited. As highlighted in **Tab. 1**, only 3 cases had a PS prenatal diagnosis. Actually, prenatal detection may be a challenge, particularly if the aberrant blood supply cannot be identified. The most common differential diagnosis for PS is CPAM and this series is consistent with this statement. Other common differential diagnosis are CDH, bronchogenic cyst or neuroblastoma [18]. In our study, in case no. 1, a mass was detected in the left adrenal gland during a 24 weeks prenatal US, which was maintained on subsequent evaluations, placing the hypothesis of a neuroblastoma; a postnatal CT was unable to distinguish the mass. Then, a thoracotomy was performed and histopathological examination showed an intra-abdominal ELS. Within PS, less than 10% are found under the diaphragm, mainly in the left upper abdomen [19]. Many authors describe the difficulty of diagnosing an adrenal mass and, when a homogenously echogenic solid mass is detected, blood supply of the lesion should be evaluated with colour Doppler [20].

Accurate prenatal diagnosis is essential both for guiding perinatal and postnatal management and for parent counselling, so, in recent years, MRI is increasingly being used to confirm the diagnosis suggested by US. However, in literature, there are divergences related to the utility of MRI. While some authors advocated the use of MRI as a helpful complementary diagnostic tool to differentiate between PS and CPAM [21], others defended that MRI isn't superior to US in the evaluation of congenital lung masses [22]. In our hospital, a prenatal MRI was performed in half of the cases, but, in each case, MRI did not provide any additional information.

According to literature, ELSs have a male predominance, in contrast with ILS that does not have a sexual predominance [23]. Our study shows that males are more affected by PS than females. The lesion type was not identified in all cases, so our data is unable to support this information.

In our series, all the neonates were admitted to NICU. This occurred most likely because they had

other neonatal pathologies associated or they were symptomatic. Some newborns were referred from another hospital to this tertiary centre for further evaluation.

As described before, there are two types of PS, being the ELS one most frequently found in the foetus [24]. Our data were in agreement with this, as the majority of PS diagnosed were ELS. Additionally, most PS cases were on the right side. Nevertheless, this finding is not supported by literature, because in previous studies lesions were found more commonly on the left side: around 90% of ELS and 60% of ILS affected the left side [4, 25].

PS is often associated with other congenital malformations, mainly ELS in which more than 60% have a co-existent anomaly, including CPAM, CDH, pulmonary hypoplasia, congenital lobar emphysema, cardiac abnormalities, vertebral anomalies and intestinal duplications [4, 26]. According to this, in our study, more than half of the patients had an associated developmental anomaly, mostly cardiac defects. Case no. 13 was diagnosed with a polymalformative syndrome, with both cardiac septal defects, renal pathology and vertebral and costal defects. There were also two cases with a CDH associated: in case no. 2, both PS and CDH were found postnatally during an X-ray and CT imaging, while, in case no. 3, PS was only found during a surgery performed for CDH. Actually, several cases of CDH associated with PS were previously reported, being PS found concomitantly in 15% to 40% of patients with CDH [27-29].

In our study, 5 of the 7 symptomatic patients had an invasive postnatal management; cases no. 12 and 14 didn't have. Concerning case no. 12, an embolization is scheduled. In case no. 14, an extremely low birth weight newborn (29 weeks of gestation), the evaluation was delayed until 12 months after birth and he is under follow-up without respiratory symptoms until the end of this study. In this case, the neonatal respiratory symptomatology was probably attributed to neonatal respiratory distress syndrome rather than to the lung mass and, therefore, the prognosis for PS was comparable to other cases. It is generally accepted that a surgical approach should be performed in symptomatic patients. However, there is no agreement amongst authors on how to manage asymptomatic lesions. The risk of morbidity in asymptomatic patients is quite small, but an elective surgery is associated with a better outcome and with fewer complications than an emergency surgery [14]. Peters et al. [30] conducted a survey to determine current practices of all members of the British Association of Paediatric Surgeons and they found that 20% of them never resected an asymptomatic lesion, while 24% always chose a surgical approach. In our experience, asymptomatic cases surgically managed had a good prognosis until the conclusion of this study, with minimal complications. In what concerns asymptomatic cases conservatively managed, respiratory morbidity was reported in one case.

Recently, minimally invasive thoracoscopy has replaced thoracotomy. A recent meta-analysis conducted by Adams et al. [31] showed a lower surgical complications rate after thoracoscopy, with a shorter length of stay, but associated with a longer operative time. In our hospital, a thoracoscopy approach has been chosen since 2009, except case no. 13 with a polymalformative syndrome in which an open resection was preferred by surgeons.

In the last years, new techniques have been developed. The target of embolization of the feeding vessel to sequestration is to decrease its size or its eventual regression. However, according to literature, only a few cases treated with trans-arterial embolization resulted in complete regression and most showed partial regression, with residual lesions [32]. In our data, this technique was applied in two children with ILS. In case no. 9, an embolization was first applied as the patient had multiple cardiac defects and, then, the surgery was performed 2 years later. Both cases had a good recovery.

With a suitable prenatal and postnatal management, the prognosis of PS is generally good [24, 33].

As PS is a disease with a small number of cases diagnosed each year, it is more difficult to study and have a good and effective knowledge of it. Consequently, it is important to conduct studies involving larger databases in order to achieve statistically significant conclusions about the best management and follow-up strategy.

This study has two major limitations: it is a retrospective study and it represents the results of a single level III centre, which explains the small size sample. Despite all these limitations, single-centre studies are extremely helpful to the physicians working in level III perinatal centres, allowing them to share experience and, consequently, improve the management of these infants.

Conclusions

In conclusion, PS, a rare pulmonary malformation, caused respiratory distress in newborns and was

associated to a high rate of congenital abnormalities. In our study, not all cases had a prenatal diagnosis, despite being essential, since it allows monitoring the prenatal and the postnatal period in specialised hospitals. A prenatal MRI didn't seem necessary, since it didn't provide any additional information. A postnatal imaging study should always be performed, since prenatal and postnatal diagnostic agreement was poor. According to our study and to other series in literature, treatment of an asymptomatic lesion is controversial. Nevertheless, to prevent postnatal complications, an elective surgical approach should be preferred in order to avoid the risks of an urgent surgery. We observed that patients with PS had a good outcome.

Declaration of interest

The Authors declare that there is no conflict of interest. There was no funding to perform this study.

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