Management of Spontaneous Pneumothorax in Children: A Systematic Review and Meta-Analysis

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Abstract

Introduction Management of primary spontaneous pneumothorax (PSP) is mainly based on adults. Data are controversial with regards to its management in children. We aimed to assess: (1) the length of hospital stay (LOS) between conservative management (i.e., observation with O_2 administration), aspiration/chest drain, and surgical management; (2) the risk of recurrence after nonsurgical treatment versus surgery; (3) the risk of recurrence in the presence of bullae.

Materials and Methods Using a defined search strategy, three independent investigators identified all the studies on the management of PSP in children. Case reports, opinion articles, and gray literature publications were excluded. The study was conducted according to the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. A meta-analysis was performed using RevMan 5.3. Data are expressed as mean ± SD.

Results Of 3,089 abstracts screened, 95 full-text were analyzed, 23 were included in the quantitative analysis, and 16 were included in the meta-analysis (1,633 patients). LOS was similar between conservative and surgical management (6.2 ± 0.8 days vs. 5.9 ± 1.4 days; p = ns). Recurrence of PSP was significantly higher among children with a nonsurgical management (32%) versus those surgically treated (18%; $p = 0.002$). The incidence of recurrence was slightly higher in patients managed by aspiration/chest drain (34%) compared with those with a conservative management (27%; $p = 0.05$). Risk of recurrence in patients with or without documented bullae was not significantly different (26 vs. 38%, respectively; p = ns).

Conclusion Given the lack of a standardized management of pediatric PSP, the present study seems to demonstrate a better outcome in children treated with surgery as first-line of management.

Level of Evidence This is a Level III study.

Keywords

- spontaneous pneumothorax
- children
- systematic review
- meta-analysis

Introduction

Primary spontaneous pneumothorax (PSP) is defined as a pneumothorax occurring neither with an underlying lung disease nor a trauma.[1–5]

The incidence of PSP ranges between 4.7 and 28/100,000/year in men and 1.2 and 6/100,000/year in women, with a higher prevalence in young, tall, and thin males.[1,2,4–9]

The incidence of PSP in the pediatric population is low and it has been reported to be around 3.4/100,000 children, with a male to female ratio of 4:1 and a peak of incidence during the adolescence.[4,10–13] However, it has been reported a higher recurrence rate in the pediatric population compared with adults (50–60% vs. 30–50%).[5,7,10,12,14,15]

Air containing lesions (blebs or bullae) are thought to play an important role in the etiopathogenesis of PSP.[1,7–9,15–17]

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Guidelines exist from the British Thoracic Society and American College of Chest Physicians regarding the management of PSP in adults. However, there is not a consensus regarding the strategy of treatment of PSP in children up to now.[1,5,9,15,18]

Moreover, there are three reported methods for calculating the size of pneumothorax on chest X-ray in adult population: the Light, Rhea and Collins algorithms.[8] The British Thoracic Society guidelines suggest defining a pneumothorax as large if there is a $\geq$2 cm gap between the lateral lung edge and the chest wall at the level of the hilum.[9] The American College of Chest Physicians guidelines on the other side consider a large pneumothorax when there is an apical distance $\geq$3 cm between the thoracic wall and the lung.[18]

Up till now, the management of pediatric PSP is mainly based on the clinical conditions of the patients, with some authors suggesting an early surgical procedure due to the high risk of recurrence.[7,11,14] Conversely, others prefer an initial nonoperative treatment (with O_2 administration, needle aspiration and/or chest drain), reserving surgery in case of recurrence or persistent air leak.[3,5,12,17,19]

Moreover, controversies exist on the role of the blebs/bullae as predicting factors for recurrence.[7,12,15]

With the present study we aimed to assess: (1) the length of hospital stay (LOS) between nonsurgical treatment (defined as observation alone with O_2 administration, needle aspiration and/or chest drain) and surgical management; (2) the risk of recurrence after nonsurgical treatment versus surgery; (3) the risk of recurrence in the presence of air-containing lesions (blebs/bullae).

Materials and Methods

Data Sources and Study Selection

This study was registered on PROSPERO—international prospective register of systematic reviews (registration number: CRD42019131588).[20] The systematic review was drafted according to the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) statement.[21]

A systematic review of the literature was performed using a defined search strategy (►Table 1). Three investigators (M.E.M., G.La, and A.R.) independently screened scientific databases (PubMed, Medline, Cochrane Collaboration, Scopus, and Ovid) looking for studies reporting on spontaneous pneumothorax in children. Medical Subject Headings (MESH) and terms used are "spontaneous pneumothorax," "spontaneous pneumothorax AND management," and "spontaneous pneumothorax AND children" (►Supplementary Table S1, available in the online version). Reference lists were examined to identify relevant cross-references. Case reports, opinion articles, experimental studies, and case series with less than 10 patients were excluded. All gray literature publications (i.e., reports, theses, conference proceedings, bibliographies, commercial documentations, and official documents not published commercially) were excluded. Full text articles of potentially eligible studies were retrieved and independently assessed for suitability by three investigators (M.E.M., G.La, and A.R.). We included all studies (trials, cohort, and case-control) that reported at least one outcome of interest. Furthermore, we included in the meta-analysis only those studies comparing different managements of PSP in children. If two or more studies reported overlapping patient cohorts, for each outcome measure we included only the article with the largest number of patients. Any disagreement over the eligibility of a specific study was resolved through the discussion with the fourth author (G.Li). Moreover, the size of PSP of all included studies has been reported, when mentioned.

Table 1 Inclusion criteria of systematic review

Publication	
Language	Any
Date	After 1950
Subject	Human studies
Study type	Retrospective
	Prospective
	Case control
	Cohort
Excluded	Case reports
	Case series
	Letters
	Editorials
	Gray literature
Keywords	Primary spontaneous pneumothorax
	Spontaneous pneumothorax
	Children
	Nonsurgical management
	Surgical management

The outcome measures for the present study were:

1. The LOS between conservative management (i.e., observation with O_2 administration), aspiration and/or chest drain, and surgical management;
2. The risk of recurrence after conservative treatment versus aspiration and/or chest drain versus surgery;
3. The risk of recurrence in the presence of air containing lesions (blebs/bullae) at computed tomography (CT) scan.

Statistical Analysis

Categorical variable frequencies were compared using Pearson's chi-square test or the two-tailed Fisher exact probability test, as appropriate. When median and range were reported, mean $\pm$ SDs were estimated, as previously reported.[22] Meta-analysis of comparative studies was conducted using RevMan 5.3.[23] Data are presented as risk ratio (RR) for categorical variables, and mean differences (MD) for continuous variables, along with 95% confidence intervals (CI) using the random-effects model, with p-values shown for Z-test for overall significance and I^2 statistic for heterogeneity. A p-value <0.05 was considered statistically significant. Data are expressed as mean $\pm$ SD.

Quality Assessment

Risk of bias for individual studies was assessed in duplicate (M.E.M. and A.R.) using the methodological index for nonrandomized studies (MINORS).[24] Differences between the two reviewers (M.E.M. and A.R.) were resolved through consensus and discussion with the third author (G.La). The total score for this 12-item instrument ranges 0 to 24 points with a validated "gold standard" cut-off of 19.8. We assessed the methodological quality for each outcome by grading the quality of evidence using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) methodology.[25] Quality of evidence was rated as high, moderate, low, and very low for each outcome. Observational studies start with a low quality of evidence. The quality of evidence was rated down in the presence of risk of bias, inconsistency, indirectness, imprecision, and publication bias. For assessment of risk of bias in observational studies, we used the MINORS instrument. Inconsistency was determined according to heterogeneity. We produced I^2 values to assess heterogeneity. I^2 value of 0 to 40, 30 to 60, 50 to 90, and 75 to 100% were considered as low, moderate, substantial, and of considerable heterogeneity, respectively. Imprecision was assessed using optimal information size, which was based on 25% relative risk reduction, 0.05 of α error, and 0.20 of β error.[26]

Results

Study Selection and Characteristics

Of 3,049 abstracts screened, 95 full-text were analyzed, 23 were included in the quantitative analysis,[2–7,10–17,19,27–34] and 16 were included in the meta-analysis (1,633 patients, ►Fig. 1, ►Table 2).[2,3,6,7,11–15,17,19,27,28,31,33,34]

The LOS was similar between nonsurgical and surgical management of PSP (5.9 ± 1.4 vs. 6.2 ± 0.8 days, respectively; p = ns, MD 0.27 [95% CI −0.11, 0.64], I^2 = 0%; ►Fig. 2A). Similarly, the LOS was similar between conservative management of PSP compared with aspiration and/or chest drain (4.7 ± 1.2 vs. 7.3 ± 1.5 days, respectively; p = ns, MD 1.41 [95% CI −0.78, 3.60], I^2 = 97%; ►Fig. 2B).

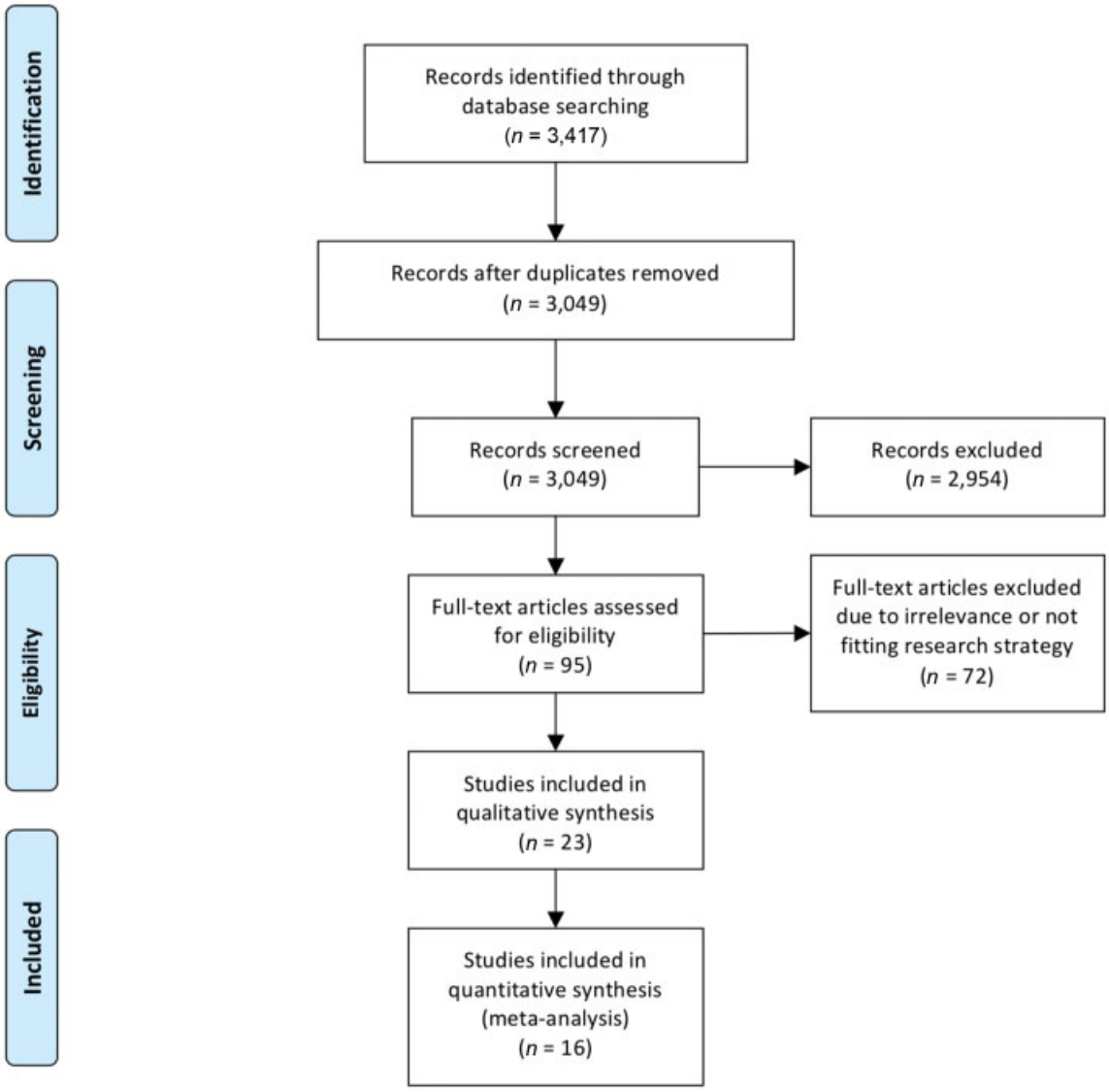

Fig. 1 Diagram of workflow in the systematic review and meta-analysis.

Table 2 Studies included in the meta-analysis

Author	Year	Type of study	LOS (days)			Surgery during 1st admission		Recurrence			Recurrence (bullae)	
			O_2	A/CD	Surg.	O_2 (%)	A/CD (%)	O_2 (%)	A/CD (%)	Surg. (%)	Bullae (%)	No bullae (%)
Yu et al[34]	1975	R	n.r.	n.r.	n.r.	n.r.	n.r.	0/8(0)	0/2(0)	n.r.	n.r.	n.r.
Davis et al[33]	1993	R	n.r.	n.r.	n.r.	n.r.	n.r.	0/1(0)	5/5(100)	1/6(17)	n.r.	n.r.
Poenaru et al[14]	1994	R	2.9	5.5	n.r.	n.r.	n.r.	3/23(13)	7/35(20)	n.r.	n.r.	n.r.
Wilcox et al[17]	1995	R	n.r.	n.r.	n.r.	0/3(0)	4/14(29)	0/3(0)	3/14(21)	n.r.	n.r.	n.r.
Qureshi et al[19]	2005	R	n.r.	n.r.	n.r.	n.r.	n.r.	20/37(54)		4/14(29)	n.r.	n.r.
Chung et al[13]	2009	R	n.r.	n.r.	n.r.	n.r.	9/15(60)	n.r.	0/15(0)	n.r.	0/7(0)	0/8(0)
Nathan et al[6]	2010	R	n.r.	n.r.	n.r.	n.r.	n.r.	6/25(24)			3/14(21)	3/11(27)
Lee et al[3]	2010	R	n.r.	n.r.	n.r.	n.r.	n.r.	4/12(33)	30/59(51)	0/6(0)	1/26(4)	3/8(37)
Zganjer et al[2]	2010	R	n.r.	n.r.	n.r.	n.r.	n.r.	n.r.	0/2(0)	1/14(7)	1/12(8)	0/4(0)
Seguier-Lipszyc et al[7]	2011	R	n.r.	n.r.	n.r.	n.r.	n.r.	10/18(55)	8/18(44)	0/10(0)	5/13(38)	15/33(45)
Smith et al[31]	2011	R	4.9(3–25)[a]	11.7(5–32)[a]	n.r.	n.r.	n.r.	n.r.	n.r.	n.r.	n.r.	n.r.
Lopez et al[12]	2014	R	n.r.	n.r.	n.r.	8/25(32)	28/73(38)	10/25(40)	25/73(34)	13/89(15)	n.r.	n.r.
Young Choi et al[15]	2014	R	4.5 ± 1	3.6 ± 1	n.r.	n.r.	n.r.	18/42(43)	36/72(50)	n.r.	12/14(80)	42/100(42)
Soler et al[11]	2018	R	n.r.	n.r.	n.r.	n.r.	n.r.	11/33(33)	19/33(58)	2/14(14)	n.r.	n.r.
Williams et al[28]	2018	R	4.1 ± 4	7.2 ± 7	6.2 ± 4	53/336(16)	157/497 (32)	140/497 (28)	82/366 (22)	42/207(20)	n.r.	n.r.
Williams et al[27]	2018	R	5.5(3–7)[b]	6 (3–7.5)[b]	5.5(5–7)[b]	5/8(62)	14/28(50)	3/8(37)	10/28(34)	4/10(40)	n.r.	n.r.

Abbreviations: a, median (range); b, median (IQR); A/CD, aspiration and/or chest drain; n.r., not reported; O_2, conservative management with oxygen administration; P, prospective cohort study; PD, prospective database; R, retrospective; Surg., surgery.
Note: Data are expressed as mean $\pm$ SD.

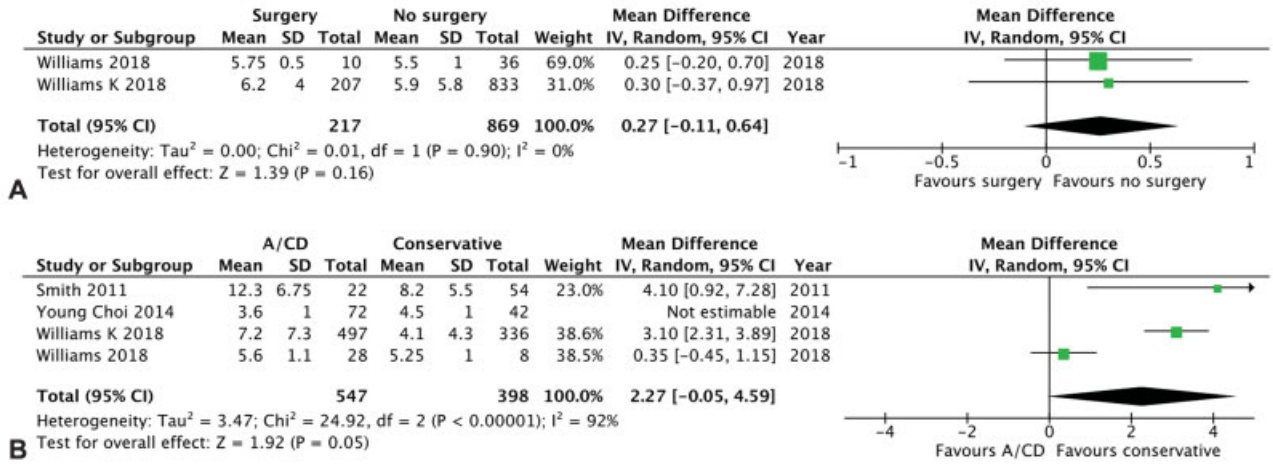

Fig. 2 (**A**) Forest plot comparison of the length of hospital stay (LOS) between nonsurgical and surgical management of primary spontaneous pneumothorax (PSP). (**B**) Forest plot comparison of the LOS between conservative management and aspiration and/or chest drain (A/CD) of PSP.

Recurrence of PSP was significantly more frequent among children with a nonsurgical management (379/1,185 patients, 31.9%) versus those surgically treated (67/370 cases, 18.1%; $p = 0.002$, RR 0.52 [95% CI 0.35, 0.79], $I^2 = 37\%$; ►Fig. 3A). Moreover, the incidence of recurrence was higher albeit not significant in those managed by aspiration and/or chest drain (283/836 patients, 33.8%) compared with those with a conservative management (139/509 cases, 27.3%; $p = 0.05$, RR 1.18 [95% CI 1.00, 1.40], $I^2 = 0\%$; ►Fig. 3B). However, the risk of a surgical procedure during the first hospital admission was similar in those managed by aspiration and/or chest drain (203/612 patients, 33.2%) compared with those with a conservative management (66/372 cases, 17.7%; $p =$ ns, RR 1.36 [95% CI 0.79, 2.33], $I^2 = 62\%$; ►Fig. 4).

The risk of recurrence in children with documented bullae at CT scan (22/86 patients, 26%) was not significantly different compared with those with no bullae detected (63/164 cases, 38%; $p =$ ns, RR 0.84 [95% CI 0.30, 2.36], $I^2 = 78\%$; ►Fig. 5).

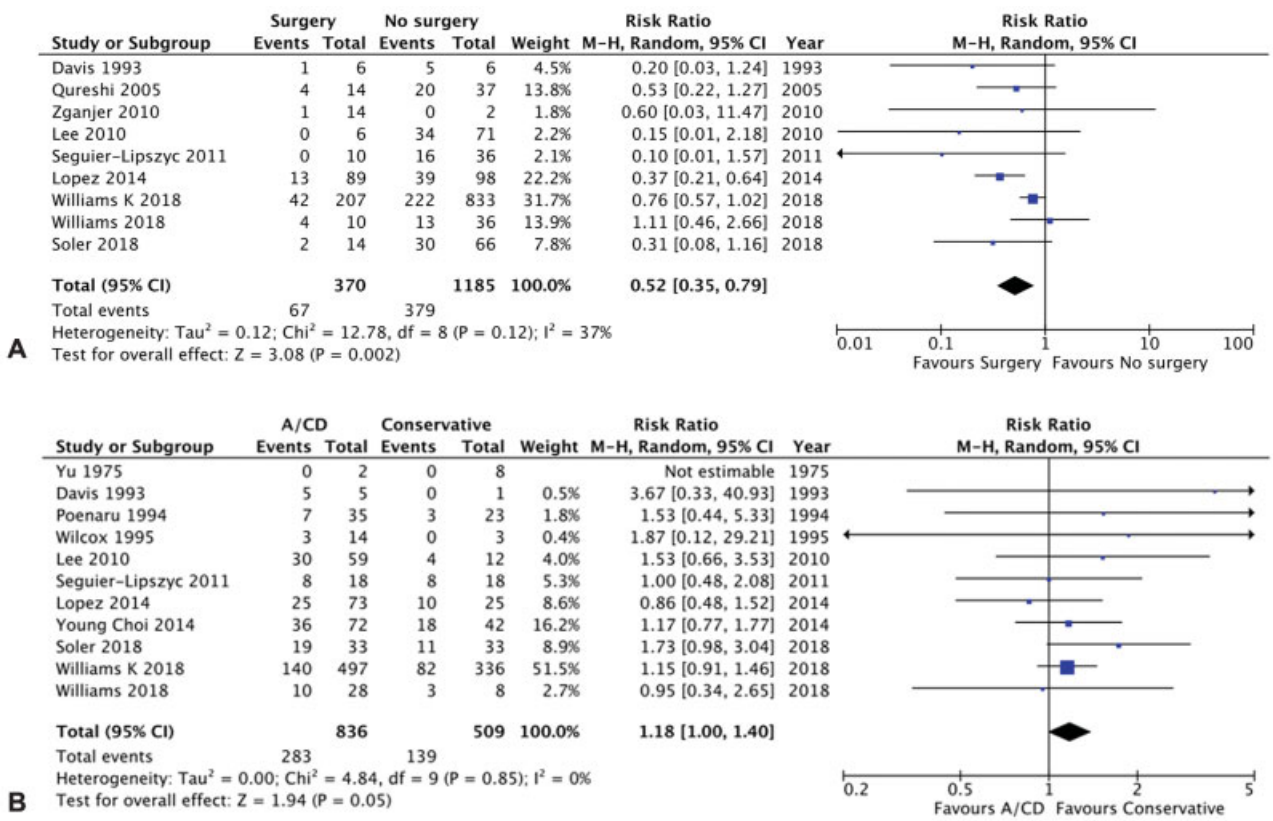

Fig. 3 (**A**) Forest plot comparison of recurrence of primary spontaneous pneumothorax (PSP) among children with a nonsurgical management versus those surgically treated. (**B**) Forest plot comparison of recurrence of PSP among children managed by aspiration and/or chest drain (A/CD) compared with those with a conservative management.

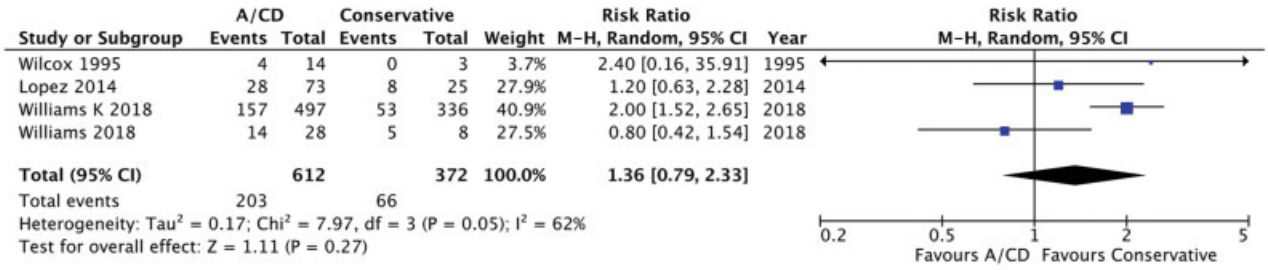

Fig. 4 Forest plot comparison of the risk of surgical procedure during the first hospital admission between children managed by aspiration and/or chest drain (A/CD) compared with those with a conservative management.

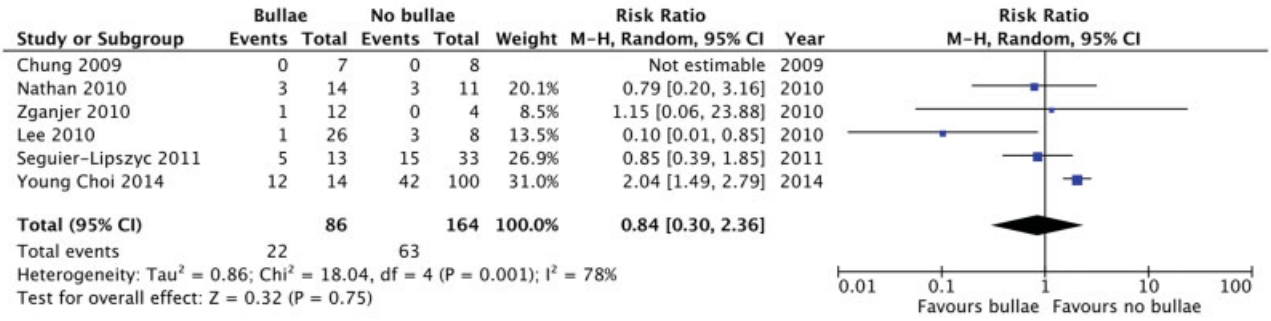

Fig. 5 Forest plot comparison of the risk of recurrence in children with documented bullae at computed tomography scan compared with those with no bullae detected.

Size of Pneumothorax

Only 5 out of 16 studies included in the meta-analysis analyzed the size of the PSP (►Table 3). Moreover, different methods have been used to determine this measure.

Discussion

The correct management of pediatric PSP has always been object of controversies.

Several studies have been published on this topic up to now; however, they are mainly retrospective with a limited cohort of patients or case series.[2–8,10,12–17,19,27–34] Moreover, data from children are often included in larger adult series, thus resulting difficult to be deduced.

Therefore, pediatric guidelines are still lacking and children with PSP are commonly managed according to the experience and preference of different clinicians.[5,8]

Possible managements consist of observation with O_2 administration, needle aspiration, chest drain insertion, or thoracotomic/thoracoscopic surgery (blebectomy/apicectomy with pleurodesis).[9]

The incidence of pediatric PSP is lower than the one reported in adults and it is estimated around 3.4/100,000 children. Boys are more commonly affected than girls, with a male:female ratio of 4:1.[4,10–13] However, Poenaru et al in 1994 showed that boys and girls under 9 years of age are equally affected.[14]

The incidence of PSP among the pediatric population has a bi-phasic distribution: there is a first peak of incidence in the neonatal period and a second one throughout adolescence.[13]

The higher incidence of PSP among neonates (mainly those preterms) may be explained by the high transpulmonary pressure generated at the first breath,[31] while the second peak of incidence during adolescence has been connected to the rupture of apical bullae/blebs.[1,7–9,15–17]

Moreover, even if it has been reported a higher incidence of PSP in adults, this has not been confirmed with regards to the recurrence rate. As a matter of fact, the incidence of recurrence has been reported to be around 30% in adults and up to 50 to 60% in children.[5,12,15,35]

On this basis, questions have raised whether children should undergo a more aggressive initial treatment or not.[10,19]

Historically, the first-line of management of PSP in children has been conservative, since traditional thoracotomy was more invasive and painful. Moreover, thoracotomy usually required a lengthened hospital stay.[12,17,33] However, the advent of the video-assisted thoracoscopic surgery (VATS), guaranteed a less invasive surgical approach with a shortened hospital stay. These improvements produced a shift toward an earlier surgical management to prevent recurrences.[12]

Controversies exist also in terms of quantification of pneumothorax. As mentioned, few methods for calculating the size of PSP have been reported.[8,9,18] However, all of them are complexes and not very accurate.[8] Based on the different guidelines used, the management of the pneumothorax can differ. Moreover, both the British Thoracic Society and the American College of Chest Physicians guidelines enlighten the importance of the clinical features of the patients more than the size of the pneumothorax in the management of the PSP. In fact, a clinically stable large pneumothorax can be

Table 3 Methods of quantification of pneumothorax

Author	Year	Pneumothorax quantification
Yu et al[34]	1975	N/A
Davis et al[33]	1993	N/A
Poenaru et al[4]	1994	N/A
Wilcox et al[17]	1995	N/A
Qureshi et al[19]	2005	N/A
Chung et al[13]	2009	N/A
Nathan et al[6]	2010	Degree 1: partial slide of pneumothorax Degree 2: complete pneumothorax Degree 3: complete pneumothorax with contralateral shift of the mediastinum
Lee et al[3]	2010	Light index; Collins formula
Zganjer et al[2]	2010	Small: rim of air <2 cm Moderate: lung collapsed halfway to the heart border Large: lung completely collapsed
Seguier-Lipszyc et al[7]	2011	N/A
Smith et al[31]	2011	N/A
Lopez et al[12]	2014	N/A
Young Choi et al[15]	2014	Distance from the chest wall to the pleural line: Minimal: <1 cm Small: between 1 and 2 cm Moderate: distance between small and large Large: distance > width of the remaining lung Complete: when the lung mass was restricted to hilum
Soler et al[11]	2018	Small: <2 ribs visible on chest-X ray or <15% Moderate: 2–4 ribs or 15–30% Large: >4 ribs or >30%
Williams et al[27]	2018	N/A
Williams et al[28]	2018	N/A

managed conservatively while a patient with dyspnea and chest pain, even if with a small pneumothorax should undergo aspiration and/or chest drain.[9,18] Furthermore, the management of the PSP mainly depended on the presence of symptoms more than on the size of the pneumothorax: asymptomatic patients were always treated with a nonoperative management (i.e., observation alone and/or O_2 administration).

Based on the current adults guidelines, clinically stable patients with PSP should be treated conservatively, with observation alone or O_2 administration, thus reserving active intervention for breathless patients. Even if British Thoracic Society guidelines proposed needle aspiration as first-line management for symptomatic PSP, American College of Chest Physicians suggested the use of chest drain because of the high risk of additional procedures and persistent air leaking following the needle aspiration.[9,18]

Commonly, when symptoms such as respiratory distress occurred, needle aspiration or chest tube insertion was performed.[2,6,11] In case of a persistent air leaking, children were candidate for surgery.[2,3,6,11,15]

Nonetheless, Young Choi et al suggested that asymptomatic patients with small pneumothorax should be managed with O_2 administration, while small, symptomatic pneumothorax or pneumothorax of any other size should be managed with chest tube insertion.[15] Also, Lee et al inserted a chest tube unless the pneumothorax was "just a small rim of air around the lung."[3]

Furthermore, a retrospective multicenter study by Soccorso et al in 2015 based on 50 pediatric cases showed that 53% of children treated by needle aspiration recurred and ultimately required a chest drain. Thus, the authors suggested to directly insert a chest tube in children to reduce the risk of repeat procedures and anesthesia.[29]An early VATS has also been proposed to avoid the risk of recurrence and thus to reduce the total hospital stay in children who should undergo a chest tube insertion for the first episode of PSP.[7,27,28]

Williams et al reported that the length of stay was similar between patients treated by surgery or aspiration and/or chest drain at their first episode.[27] The results of our meta-analysis seem to corroborate this outcome.

Moreover, it has reported a recurrence rate after the primary surgery between 4 and 40%.[7,12,19,27,28] These data, even if low, seem not justify an initial surgical approach.

In our study we found a significantly lower risk of recurrence between patients treated by primary surgery versus those nonsurgically managed (18 vs. 32%, respectively; $p = 0.002$) and among the latter, those managed by observation alone had a slightly reduced risk of recurrence than those

Table 4 Risk of bias assessment for individual studies using methodological index for nonrandomized studies (MINORS)[24]

Item	Yu et al[34]	Davis et al[33]	Poenaru et al[14]	Wilcox et al[17]	Qureshi et al[19]	Chung et al[13]	Nathan et al[6]	Lee et al[3]	Zganjer et al[2]	Seguier-Lipszyc et al[7]	Smith et al[31]	Lopez et al[12]	Young Choi et al[15]	Soler et al[11]	Williams et al[28]	Williams et al[27]
1. A clearly stated aim	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
2. Inclusion of consecutive patients	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
3. Prospective collection of data	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
4. End points appropriate to the aim of the study	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
5. Unbiased assessment of the study end point	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
6. Follow-up period appropriate to the aim of the study	1	2	0	1	0	0	2	2	2	2	0	0	1	0	0	0
7. Loss to follow-up less than 5%	0	1	0	0	0	0	1	2	0	0	0	0	1	0	0	0
8. Prospective calculation of the study size	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
9. An adequate control group	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
10. Contemporary groups	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
11. Baseline equivalence of groups	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
12. Adequate statistical analyses	0	0	0	0	2	0	2	2	2	2	2	2	2	2	2	2
Total score	13	15	12	15	14	12	17	18	16	16	14	14	16	14	14	14

0 = not reported.
1 = reported but inadequate.
2 = reported and adequate.

Table 5 GRADE evidence profile[25] for the management of PSP in children

Quality assessment							No. of patients		Effect	Quality	
No. of studies	Study design	Risk of bias	Inconsistency	Indirectness	Imprecision	Other considerations	Cases	Controls	Relative (95% CI)	Absolute (95% CI)	
LOS in surgery vs. no surgery							*Surgery*	*No surgery*			
2	OS	Moderate[a]	Substantial	Not serious	Serious[b]	None	217	869	–	MD 0.27 higher (0.12 lower to 0.71 higher)	⊗OOO VERY LOW
LOS in A/CD vs. conservative							*A/CD*	*Conservative*			
4	OS	Moderate[a]	Considerable	Not serious	Serious[b]	None	547	398	–	MD 1.41 higher (1.44 lower to 6.64 higher)	⊗⊗OO LOW
Recurrence in surgery vs. no surgery							*Surgery*	*No surgery*			
9	OS	Moderate[a]	Low	Not serious	Serious[b]	None	67/370 (18.1%)	379/1,185 (31.9%)	RR 0.52 (0.35, 0.79)	138 fewer per 1,000 (from 93 fewer to 210 fewer)	⊗⊗OO LOW
Recurrence in A/CD vs. conservative							*A/CD*	*Conservative*			
11	OS	Moderate[a]	Low	Not serious	Serious[b]	None	283/836 (33.8%)	139/509 (27.3%)	RR 1.18 (1.00, 1.40)	65 more per 1,000 (from 55 more to 77 more)	⊗⊗OO LOW
Surgery at first admission in A/CD vs. conservative							*A/CD*	*Conservative*			
4	OS	Moderate[a]	Considerable	Not serious	Serious[b]	None	203/612 (33.2%)	66/372 (17.7%)	RR 1.36 (0.79, 2.33)		⊗⊗OO LOW
Recurrence in bullae vs. no bullae							*Bullae*	*No bullae*			
6	OS	Moderate[a]	Considerable	Not serious	Serious[b]	None	22/86 (25.6%)	63/164 (38.4%)	RR 0.84 (0.30, 2.36)	128 fewer per 1000 (from 46 fewer to 360 more)	⊗⊗OO LOW

Abbreviations: A/CD, aspiration and/or chest drain; CI, confidence interval; LOS, length of hospital stay; MD, mean deviation; OS, observational study; PSP, primary spontaneous pneumothorax; RR, risk relative.
[a]Bias due to possible confounding.
[b]OIS, optimal information size not met
Note: GRADE working group grades of evidence.
High quality: Further research is very unlikely to change our confidence in the estimate of effect.
Moderate quality: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.
Low quality: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.
Very low quality: We are very uncertain about the estimate.

who underwent aspiration and/or chest drain (27 vs. 34%, respectively; $p = 0.05$).

The reduced number of procedures can explain the shorter hospital stay in those primarily treated by surgery, thus eliminating the time-lapse between the first nonsurgical management and the surgical one.

Moreover, the risk of a surgical procedure did not differ between those children initially managed either by conservative or operative treatment. This might support the choice of a primary conservative management followed by an early surgery if there is a persistent air leak (>3–5 days) or a recurrence, in clinically stable patients.[8,35]

The role of air-containing lesions (blebs/bullae) in the etiopathogenesis and in the risk of recurrence of PSP has also been debated, along with the need of performing a high-resolution CT in children with a first episode of PSP. It has been reported an incidence of air-containing lesions at high-resolution CT ranging from 31 to 100% in children with PSP. Moreover, the recurrence rate of pneumothorax in these children has been reported to range between 50 and 100%, thus suggesting an important role of the blebs in the risk of recurrence.[7,15]

A retrospective single center study on 114 children from Young Choi et al showed a 55% incidence of bullae on high-resolution CT in patients with a first episode of PSP with a risk of recurrence in this cohort of 60%.[15] Soccorso et al, in 2015, reported a 100% incidence of bullae in the surgical specimens of children who underwent wedge resection after a first episode of PSP.[29] On the other side, some authors do not recommend the use of high-resolution CT for all the children with PSP due to its low sensitivity as screening test and its high radiation dose.[8,27]

In our study we reported that the presence of bullae did not increase the risk of recurrence of PSP, thus supporting the hypothesis that a high-resolution CT seems not to be mandatory after the first episode of PSP.

Limitations

We are aware of the limitations of our study, which rely on the quality of the studies and data available in the literature, as any other meta-analysis. Moreover, we highlighted the lack of consensus in the definition of the size of pneumothorax. Therefore, a limitation of our study is the inability to measure the effect of the size of the PSP. Moreover, all the studies included in the meta-analysis were retrospective and only few of them have reported a follow-up period appropriate to the aim of the study. None of the papers provided sample size calculations, and most of the studies lack in few of the outcomes considered (►Table 2). As a consequence, none of the studies included reached the gold standard cut-off on MINORS of 19.8 out of 24 (►Table 4). According to the GRADE methodology, the quality of evidence was low for the reported outcomes (►Table 5). However, when independently assessed by two authors (G.Li and P.L.C.) using AMSTAR (A Measurement Toll to Assess Systematic Reviews),[36] the present systematic review and meta-analysis received a relevant score (►Supplementary Table S2, available in the online version). The PRISMA checklist was then completed (►Supplementary Table S3, available in the online version).

Conclusion

This is the first systematic review and meta-analysis comparing the outcomes between conservative management, aspiration and/or chest drain, and surgical procedure of PSP in children. Given the lack of high-quality studies and the paucity of information related to the size of the pneumothorax, primary surgery seems to reduce the risk of recurrence. However, a surgical procedure could be safely performed as early as possible either after a primary conservative management or aspiration and/or chest drain, without an increased LOS.

The presence of bullae seems not to predict a higher risk of recurrence, thus not supporting the need of a high-resolution CT to all pediatric patients after the first episode of PSP.

However, further data given by high-quality studies would be needed to corroborate our preliminary results.

Conflict of Interest
None declared.

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