

Traumatic diaphragmatic rupture in pediatric age: review of the literature

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Abstract

Purpose Traumatic diaphragm rupture (TDR) is a rare complication of trauma in pediatric age and may be easily missed by the severity of associated injuries so that delayed emergent presentation can occur with increased rate of morbidity and mortality. No review has been available to guide clinicians through the pitfalls and the initial diagnostic approach to pediatric TDR.

Methods A Medline thorough search on TDR was conducted using different queries. English language citations were identified during the period of January 2000 through December 2014 limiting the search to pediatric age (0–18 years). Abstracts were reviewed to determine eligibility and texts were obtained for further review. Differences were resolved by consensus and only reliable data were included.

Results Most frequently reported presenting symptoms of TDR are respiratory and abdominal. While respiratory symptoms are among the most frequently described at

the onset in pediatric and adult series, abdominal symptoms result to be more frequent in adult than pediatric patients. Chest X-ray (CXR) is the first-line imaging exam which is reported to show pathognomonic or suspect findings in 85 %. CT was the second main radiological technique used, in particular to confirm the suspicion of TDR.

Conclusions A high clinical index of suspicion is needed to diagnose and effectively manage diaphragmatic rupture. TDR should be kept in mind while dealing with patients assessed for abdominal or respiratory symptoms whenever there is history of trauma or blunt injury especially in children as the increasing of non-operative management of blunt abdominal trauma could result in missing important injuries as TDR.

Keywords Diaphragm rupture · Pediatrics · Pediatric trauma · Pediatric emergency surgery

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Background

Traumatic diaphragmatic rupture (TDR) is a rare complication of pediatric abdominal trauma.

The diagnosis of TDR is often elusive and difficult to confirm or rule out due to its frequently poor and non-specific clinical and radiological findings, to the severity of associated injuries and to the high index of suspicion required for its recognition. TDR may be easily missed; so, a delayed emergent presentation can occur with increased rate of morbidity and mortality [1].

Pediatric TDR has been described in the literature only by case reports and small series. The most numerous references available are the series by Ramos/Koplewitz and Okur [2–4].

According to the available data, TDR has a prevalence of 0.07 % of all pediatric trauma with a male predominance, that increases to 2.95 % when considering only blunt abdominal trauma (BAT) [2, 5, 6]. The mean incidence reported in the literature is of 0.9 new cases per year per center (range 0.54–1.8) [1, 2, 4, 5, 7–14]. TDR is mainly ascribed to blunt trauma, but also to penetrating trauma that represents 13.3–55 % of TDR etiology [2, 4]. Rare iatrogenic injuries related, for example, to the insertion of chest tube have been described [10].

The site and dimensions of TDR appear to be similar in both pediatric and adult patients with a more frequent occurrence on the left side of the diaphragm [15–29] and with bigger dimension of rupture in blunt trauma [17, 23, 26, 30, 31].

The highly reported presenting symptoms of TDR are respiratory and abdominal. Patients present often with dyspnea (86 %), abdominal pain (13 %) and with a reduction in breath sounds on the affected side (73 %) [10].

Diagnosis is often reached preoperatively and in less than 12 h [5, 10]. Imaging techniques mainly used are chest X-ray (CXR), and computerized tomography (CT). CT can enhance diagnostic sensitivity with imaging techniques as multiplanar reconstruction, three-dimensional reconstruction, and volume rendering. A positive CXR or with suspicious findings is reported to be effective for diagnosis in particular when CXR is associated to other imaging techniques [4, 9].

Laparotomy is the preferred surgical approach [1, 9, 10], sometimes associated with a thoracotomy [4, 5]. Complications are reported in 10–20 % of the cases that need surgery [5, 10]. Mortality ranges from 0 to 33 %, often due to associated injuries [2, 5, 9].

Pediatric literature about TDR consists mainly of case reports and small series with no clear indications about the management.

We present a review of the literature to elucidate the current data regarding etiologies, clinical presentation, diagnosis and treatment of blunt TDR in children and the principal differences with adult data. We will then highlight the relevant points for the pediatric patient presenting to the emergency department (ED) with a suspected abdominal trauma.

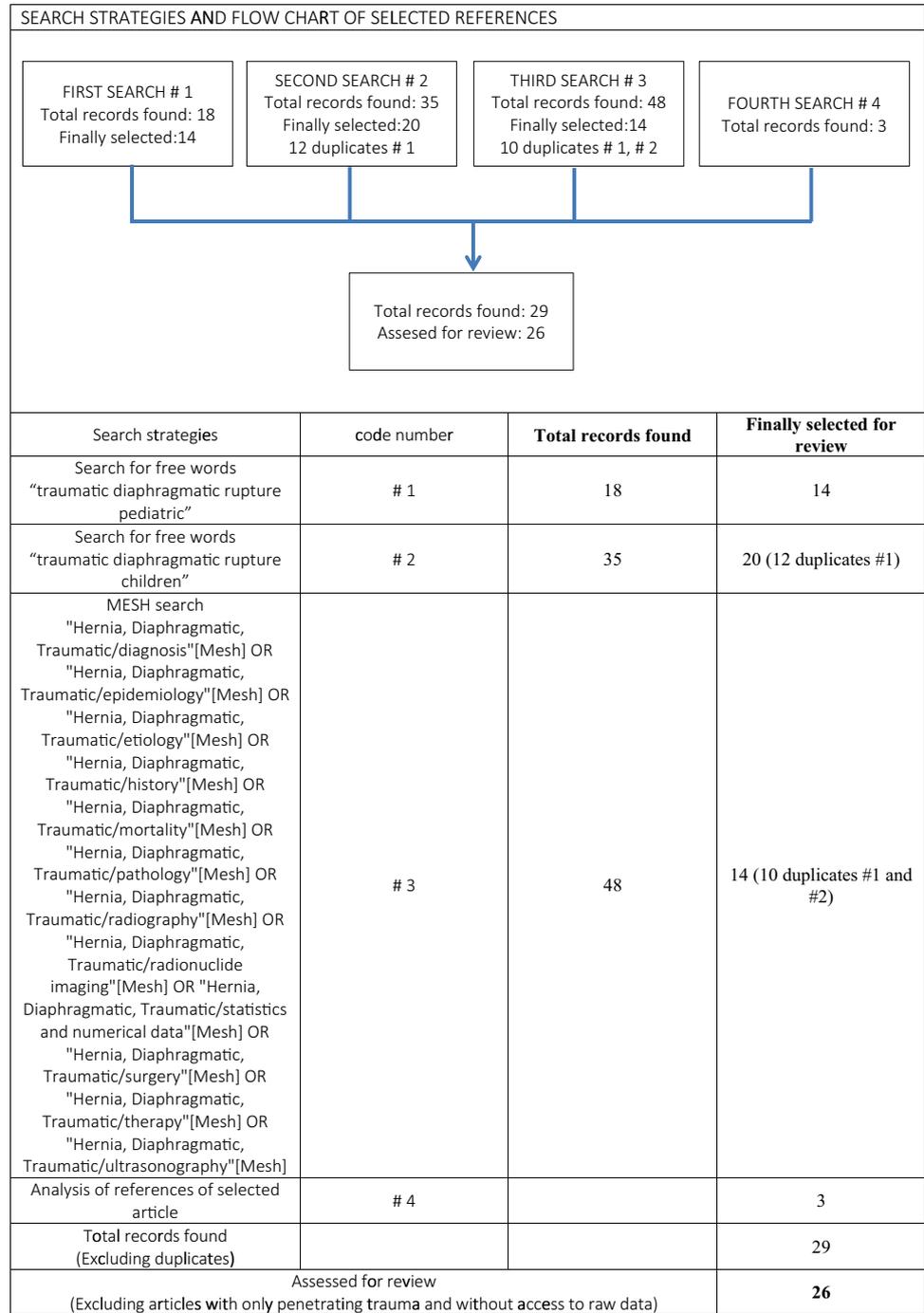
Methods

A Medline thorough search on TDR was conducted using different queries. Keywords were determined by running a broad search and noting vocabulary used to establish the concept of blunt diaphragmatic rupture. English language citations were identified during the period of January 2000 through December 2014 limiting the search to pediatric age (0–18 years). The search included even case reports and case series. We included adult series where data regarding pediatric patients could be extracted, while we excluded articles where the data of the TDR patients were not sufficient to allow an inclusion in the final pooled population. Abstracts were reviewed by two authors (NP, FM) to determine eligibility, and texts were obtained for further review if titles indicated any information regarding TDR. Differences were resolved by consensus and only reliable data were included. Our search identified a case report and a retrospective single-center study in which the characteristics of patients matched so that the patients described in the case report were apparently included in the retrospective study [9, 32]. In the second search, we found other two retrospective articles where the populations described were apparently the same [2, 3]. Koplewitz and Ramos [2, 3] described both 15 pediatric patients who sustained TDR, with same age, gender, mechanism of injury, associated injuries and management. We found disagreement in the side of injury in two patients and in the outcome in other two.

In both cases, we contacted the authors to ask for the raw data of the studies to exclude duplication of the patients described in our review, without obtaining answer. We decided to exclude from our review the case report by Rattan [32] and pooled the data from the two articles by Koplewitz and Ramos as one [2, 3].

Three articles selected for our review [4, 10, 12] reported mixed data of blunt and penetrating trauma. We contacted the authors to ask for the raw data and we obtained answer only from one of the three [10] who was included in our review, while the other two [4, 12] were excluded. We finally selected for this review 26 articles with a total of 95 patients included [2, 3, 5, 7–12, 14, 32–49] (Fig. 1).

Fig. 1 Search strategies and selected articles



Results

Characteristics of the studies included are summarized in Table 1.

The pooled pediatric population resulted from our review confirmed a higher prevalence of TDR in male (*n* 61) 64.2 %. The mean age was 7.4 years (range 3 weeks–18 years). The diaphragm was affected on the left side (*n* 55) in 57.9 % of the cases, followed by the right side

(*n* 32) 33.7 %. A small number of patients presented with bilateral (*n* 3) 3.15 % and central involvement (*n* 2) 2.1 %. In three cases, the affected side was not clearly reported (3.15 %). TDR due to BAT was determined, in order of descending frequency, to road accident (*n* 65) 68.4 %, fall (*n* 14) 14.8 % and crushing (*n* 6) 6.3 %. In a small sample of patients (*n* 10) 10.5 %, the mechanism of injury was not described. The mean size of the rupture had an overall mean of 8.8 cm. Lesions on the right side

Table 1 Article included in the review

Author	Number of patients	Mean age (year)
Kumar and kumar [33]	1	10
Al-Salem [11]	6	6.6
Kucera et al. [34]	1	11
Mirza et al. [35]	1*	8
Couselo et al. [36]	1	5
Picetti and Mergoni [37]	1	6
Rattan et al. [32]**	9	8.1
Rattan et al. [9]**	2	8.5
Jones et al. [38]	1	8
Oyinloye et al. [39]	1	4
Jain et al. [40]	1	3
Khan et al. [8]	8	6.9
Alper et al. [41]	1	5
Mulpuri et al. [42]	1*	10.7
Antao et al. [43]	1	5
Shehata and Shabaan [5]	10	8.5
Santschi et al. [44]	1*	12
Soundappan et al. [12]	8	7.6
Barsness et al. [7]	6	7
Friedlaender and Tsarouhas [45]	1	11
Machmouchi and Al Ibgahim [14]	4	6.25
Adler [46]	1	7
Sharma et al. [47]	2	8.5
Grover and Ratan [48]	1	8
Karnak et al. [10]	13	4.9
Cohen et al. [49]	1	15
Ramos et al. [2]**	13	8.4
Koplewitz et al. [3]**	13	8.4
TOT	95	7.4 (3w – 18y)

* Data extrapolated

** Different case series with apparently the same patients (see text)

were larger (mean 10.3 cm) than those on the left side (mean 7.3 cm).

The ISS was reported in 4 of the 26 studies with a total number of 21 patients, while the Pediatric Trauma Score (PTS) was indicated in one case report. The mean ISS was 27.5 (range 9–57), with great differences between patients with associated injuries (ISS 31.4) and patients with isolated injuries (ISS 15).

Clinical presentation

The clinical presentation was fully described in (*n* 54) 57.9 % of patients.

Respiratory signs and symptoms represented 60.5 % of the abnormalities described. The most frequent respiratory

findings reported were reduction of breath sounds (29 %), dyspnea (20.3 %) and respiratory distress (18.8 %) (Table 2).

In one case, bowel sounds were auscultated in the thorax, while in three patients, bilious or gastric fluid was drained from thorax (Table 2).

Associated injuries were described in (*n* 68) 71.6 % of the TDR patients. The liver (14.2 %) was the most frequent organ involved followed by the lung (12.3 %), the pelvis (10.35 %) and the kidney (7.7 %) (Table 3).

Fourteen patients sustained an isolated TDR while for 13 patients we did not find information about other concerning injuries except the TDR.

Imaging

Radiological investigations were reported in all except one child, who was dead on arrival to the ED. CXR resulted to be the principal imaging technique used in pediatric age. A total of 104 CXR were performed in the reviewed population. CXR resulted positive for TDR in (*n* 45) 43.3 % patients, negative for any finding related to TDR in (*n* 17) 16.3 % patients. CXR showed suspicious findings that led to further examination or surgical exploration in (*n* 42) 40.4 %.

In six cases, CXR was repeated more than once. Five of the 17 previously negative CXR showed TDR and one patient showed findings considered suspicious for TDR (e.g., elevation of diaphragm).

CT was the second main radiological technique used, in particular to confirm the suspicion of TDR or to study further the associated injuries. A total of 48 CTs were described in the pooled population. CT resulted negative in (*n* 12) 25 % of cases and positive in (*n* 33) 68.75 %, while in (*n* 3) 6.25 % showed uncertain findings for TDR, like elevation of hemi-diaphragm or irregularity of diaphragmatic contour. Other different imaging techniques (ultrasound, gastro-intestinal series, fluoroscopy, chest and abdominal XR with oral enema and thoracic drainage contrast) were used in 13 patients and resulted positive for TDR in (*n* 16) 88.9 % of the cases (Table 4).

Diagnosis

TDR was mostly (*n* 78, 82.1 %) diagnosed before the intervention. A fewer number of patients (*n* 14) 14.7 % had an intraoperative diagnosis. Three cases of autoptic diagnosis and one case of post-operative diagnosis during radiological follow-up were described. One patient with bilateral TDR rupture needed a second CXR to be fully diagnosed. One of 14 patients with intraoperative diagnosis required immediate intervention that did not allow imaging, while 13 patients performed imaging (CXR or CT) that resulted not informative or negative.

Table 2 Clinical findings in patients with TDR

Clinical findings													
Respiratory	% (n)	General examination	% (n)	Cardiac	% (n)	Abdominal	% (n)	Neurologic	% (n)	Drainage	% (n)	Negative examination	% (n)
Total	60.5 % (69)	Total	14 % (16)	Total	11.4 % (13)	Total	8.8 % (10)	Total	1.8 % (2)	Total	2.6 % (3)	Total	0.9 % (1)
Breath sound reduction	29 % (20)	Cyanosis/ Pallor	43.75 % (7)	Tachycardia	53.8 % (7)	Abdominal pain	50 % (5)	Loss of consciousness	100 % (2)	Bilious/ gastric fluid from thorax tube	100 % (3)		
Dyspnea	20.3 % (14)	Vomit	37.5 % (6)	Hypotension	30.8 % (4)	Abdominal tenderness	20 % (2)						
Respiratory distress	18.8 % (13)	Fever	12.5 % (2)	Hypertension	7.7 % (1)	Abdominal distension	20 % (2)						
Tachypnea	14.5 % (10)	Nausea	6.25 % (1)	Weak peripheral pulses	7.7 % (1)	Seat belt sign and bruising	10 % (1)						
Retraction	7.2 % (5)												
Respiratory failure	3 % (2)												
Thoracic pain	3 % (2)												
Bowel sounds	1.4 % (1)												
Tracheal deviation	1.4 % (1)												
Chest bruising	1.4 % (1)												

Table 3 Traumatic diaphragmatic rupture associated injuries and frequency

Associated injuries	% (n)
Liver	14.2 (22)
Lung	12.3 (19)
Pelvis	10.35 (16)
Kidney	7.7 (12)
Rib fracture	7.7 (12)
Long bone fracture	7.7 (12)
Spleen	5.2 (8)
Bowel	5.2 (8)
Emo/pneumothorax	4.5 (7)
Vertebra	3.9 (6)
Head	3.9 (6)
Pancreas	3.2 (5)
Major vessel	2.6 (4)
Stomach	1.9 (3)
Colon	1.9 (3)
Shoulder	1.9 (3)
Perineum	1.3 (2)
Ureter	1.3 (2)
Uterus, vagina, bladder, spinal cord, pericardium	0.65 (1) each

The range of time to diagnosis reported in the literature was extremely wide (Table 1). In our population, these data were available for 77 patients. Diagnosis of TDR was reached in the first 24 h in 45 children (58.4 %). The majority of these patients (97.8 %) were diagnosed in the first 12 h mainly due to the presence of a severe trauma that required an early radiological imaging or intervention (Table 1).

Surgical approach

Laparotomy was the preferred surgical approach in 77 patients (81.1 %). In the other cases, thoracotomy (*n* 7, 7.4 %) and laparo-thoracotomy (*n* 5, 5.3 %) were the surgical techniques that were described. Two patients had different surgical approach (thoracoscopy and thoracoscopy associated with laparotomy). In five patients, the medical or surgical

management was not reported. One patient with bilateral TDR had two different surgical approaches (Table 1).

Outcome

Overall mortality was (*n* 9) 9.5 %. The reasons of mortality were associated to hypovolemic shock, severe head trauma, multi-organ failure and cardiac arrest. A total of (*n* 83) 87.4 % patients survived and (*n* 60) 72.3 % of the survivors had no complications, while (*n* 23) 27.7 % had one or more complication. Pneumonia was reported as a frequent (*n* 6, 20.7 %) complication of TDR. The outcome of one patient was not documented, while two cases were described in more than one study considered for this review with different outcomes, as previously explained (Table 5).

We finally made an analysis to evaluate if the inclusion of diaphragmatic lesions caused by penetrating trauma may have changed or influenced the overall reported frequency of clinical signs at presentation, associated injuries, complications and outcome. In this population of children with blunt and penetrating diaphragmatic trauma, we did not find significant variation of the frequency described in the TDR population due to blunt trauma (Table 6).

Discussion

Diaphragmatic injury is a rare but serious condition in pediatric trauma patients. TDR occurs most often in patients who undergo multiple trauma with massive forces involved, even though ruptures have also been described as a complication of minor trauma [50].

A high clinical index of suspicion is needed to diagnose and effectively manage diaphragmatic rupture even in cases of a history of high-velocity motor-vehicular accident, road accident, apparently isolated lacerations of liver and spleen or pelvic fractures.

In pediatric age, we found a male–female ratio of 2.1:1, and also in adult patients there was a higher prevalence of TDR in male with a male:female (M:F) ratio ranging from 1.5 to 4.5:1 [4, 10, 15–18, 51].

Table 4 Imaging techniques

Imaging	Chest X-ray % (n)	Computerized tomography % (n)	Other techniques ^a % (n)
Total	<i>n</i> 104	<i>n</i> 48	<i>n</i> 18
Positive	43.3 % (45)	68.75 % (33)	88.9 % (16)
Suspicious	40.4 % (42)	6.25 % (3)	–
Negative	16.3 % (17)	25.0 % (12)	11.1 % (2)
Positive/suspicious only in late examination	7.7 % (8)	4.2 % (2)	5.5 % (1)

^a Ultrasound and GI series with or without contrast, barium enema, naso-gastric contrast, drainage contrast, fluoroscopy

Table 5 Outcomes and complications

Survivors	% (n)	Deceased	% (n)	Unknown/unclear	% (n)
Total	87.3 % (83)	Total	9.5 % (9)	Total	3.2 % (3)
With complications	% (n)	Presenting			
Total	27.7 % (23)	MOF	33.3 % (3)		
Complication	% (n)	Shock	22.2 % (2)		
Total	n 29	Head trauma	22.2 % (2)		
Pneumonia	20.7 % (6)	Cardiac arrest	11.1 % (1)		
Ileus	17.2 % (5)				
Intestinal occlusion	10.3 % (3)				
Pancreatitis	10.3 % (3)				
Wound infection	6.9 % (2)				
Intussusception	6.9 % (2)				
Paraplegia	6.9 % (2)				
Empyema	3.4 % (1)				
Renal failure	3.4 % (1)				
Cholestasis	3.4 % (1)				
Cardiac arrest	3.4 % (1)				
Kidney infarction	3.4 % (1)				
Peritonitis	3.4 % (1)				

Our review confirms that TDR localizes more frequently on the left (57.9 % in our review vs. 53.3–100 % reported in the literature) [9, 10].

While respiratory symptoms are the most frequently described at the onset in pediatric and adult series as TDR becomes clinically evident only after herniation of abdominal viscera into the thorax, abdominal symptoms result to be extremely more frequent in adult than pediatric patients (52 vs 8.8 %) [17].

Respiratory symptoms in pediatric patients are mainly nonspecific as they can be related to a variety of other lesions commonly associated with multiple trauma so that clear breath sounds and normal oxygen saturation do not rule out the possibility of a diaphragmatic hernia as herniation of abdominal viscera into the thorax can occur also months or years later.

The presence of bowel sound in the thorax, even if rare, could be considered pathognomonic of TDR.

In children, the diaphragmatic laceration is associated with other injuries in 71.5 %. This is in agreement with the available literature (50–100 %). A slight difference emerges looking at the most frequent injuries associated. Our review found that the lung appears to be the second organ involved as opposite to the literature where liver lacerations, pelvic fractures, major vessels rupture, stomach perforation, rib fractures, spleen lacerations, long bone fractures, kidney lacerations [2, 4–6] in descending order of frequency order, are the main associated injuries. The large number of associated injuries explains the Injury Severity Score (ISS) values reported in the literature. [2, 4, 5].

Imaging techniques mainly used are chest X-ray, computerized tomography and other peculiar CT imaging techniques (multiplanar reconstruction, three-dimensional reconstruction, volume rendering) associated to an increase of diagnostic sensitivity. A positive CXR or with suspicious findings is reported to be effective for diagnosis up to 54 % of cases when CXR is associated to other imaging techniques [4, 9].

In our review, chest X-ray resulted to be the first-line imaging exam. The pathognomonic sign of TDR is the visualization in the thorax of abdominal viscera. Because TDR can take place without herniation, CXR can often be negative or show nonspecific alterations. In our pediatric population, CXR is reported to show pathognomonic or suspect findings in 85 % as opposed to pediatric (64 %) and adult (40 %) [4, 18]. This results in the opportunity to use CXR as the first-line imaging technique and reduce the exposure of pediatric patients to radiation due to radiological exams [18].

Second-line imaging is represented by computerized tomography (CT) [17]. Any chest or abdominal CT ordered for a suspected post-traumatic injury should be extended to the diaphragm [29, 52].

Despite the poor specific clinical manifestation, we found that a preoperative diagnosis of TDR in children is reached in 82.1 % of cases. In the adult patient, TDR is more commonly diagnosed during the surgical approach (10–75 %) due to the associated injuries [15, 17–19]. In our review, we found a 14.7 % of intraoperative diagnosis of TDR.

Table 6 Comparison of frequencies between all cases of pediatric traumatic diaphragmatic rupture and selected cases of blunt traumatic diaphragmatic rupture

Sex	Side				Associated injuries				Imaging				Clinical findings				Diagnosis		Outcome			
	Male	Female	Left	Right	Liver	Lung	Spleen	Pelvis	CXR pos	CXR neg	CT pos	CT neg	Breath sound reduction	Dyspnea	Respiratory distress	Abdominal pain	Seat belt sign	Pre-operative diagnosis in 24h	Diagnosis in 24h	Survivors	Deaths	Complication
Blunt only (%)	64.2	34.7	57.9	33.7	14.2	12.3	5.2	10.35	43.3	40.4	68.75	25	29*	20.3*	18.8*	50*	10*	82.1	58.4	87.3	9.5	24.2
Blunt + penetrating (%)	67.7	31.6	63.9	28.6	13.7	10.2	8.3	8.3	43.9	15.1	72.7	21.2	28.3*	22.2*	15.2*	60*	16*	80.5	60.4	89.5	8.3	21.8

Comparison between blunt TDR (*n* 95 patients) and blunt + penetrating TDR (*n* 133 patients)

* See Table 2 for more details

In adult patients, TDR is mostly diagnosed in less than 12 h (89–92.5 %) probably because of the more frequent recourse to the surgical approach for other severe associated injuries. Pediatric patients diagnosed in less than 12 h in our review were 59.3 %.

Delayed diagnosis can be related to an occult TDR with no immediate herniation and can have a diagnostic delay also of several days, both in pediatric than in adult patients.

These differences regarding the diagnosis of TDR between children and adults can be attributed to the constant efforts made by pediatric specialists in being as less invasive as possible in the management of pediatric trauma.

Despite the poor specific clinical manifestation, a preoperative diagnosis of TDR in children is more common than adults who are diagnosed mostly during surgical procedures for the associated injuries. [15–17, 19, 24].

These differences can be attributed to the increasing non-operative management of blunt organ injuries in pediatric trauma patients combined with the wish to save children from radiation given by CTs and the related induced malignancy that could result in missing important injury as TDR.

In the available literature, laparotomy is the preferred surgical approach [1, 9, 10], sometimes associated with a thoracotomy [4, 5]. Thoracotomy access alone is described as the TDR repair technique in few cases [10].

Our review confirms that the surgical approach preferred for TDR correction was laparotomy followed by thoracotomy and this outcome fits what is reported in the adult literature, where laparotomy accounts for 35–96 % of surgical approaches, followed by thoracotomy [15, 17, 53–55]. We did not find any technical explanation, more than surgeon's preference, for the different surgical approach. This can logically be connected to the surgeon's experience and to the localization of associated injury for which the patient is brought to operating room. Complication and mortality rate in our pooled population appear to be the same as in adult series, where there is a wide range of both complications (11–62.9 %) [15, 56] and mortality (1–28.8 %) [18, 51], often due to the associated injuries or complications rather than TDR itself.

Traumatic diaphragmatic rupture is uncommon in trauma and often clinically occult. TDR can be masked by associated injuries in a polytrauma patient. Patients with TDR often have severe associated injuries that need to be diagnosed and managed promptly, as their severity and successful timely treatment can determine the patients' outcomes. Pediatric emergency physicians and emergency physicians should maintain a high index of suspicion of TDR while dealing with patients assessed for abdominal or respiratory symptoms and history of trauma occurred recently or in the past. In children, the diagnosis of TDR is more often delayed than adult. Early diagnosis and repair

help avoid the late complications due to herniation of intra-abdominal contents into the chest. Chest X-ray is the initial screening option followed by computed tomography to evaluate the diaphragm. Treatment of DI consists of early repair, mainly on laparotomy.

Death by diaphragmatic rupture alone is rare, and the main cases of death are caused by associated injuries to other organs. This review should be helpful in managing pediatric patients with traumatic rupture of the diaphragm.

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Compliance with ethical standards

Conflict of interest Authors declare that they have no conflict of interest.

Ethical approval The review presented did not involve human participants nor animals and did not require informed consent.

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