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Rib fractures in infancy, case-series and register case-control study from Sweden

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Abstract

Objectives: To assess cases with rib fractures in infants under observation for or with an abuse diagnosis, and to compare risk factors with that in infants with fractures but no abuse diagnosis.

Methods: Design was case-series and register case-control of infants (aged <1 year) with rib fractures. Data was clinical records for the case-series (n = 26) and from national health registers for the case-controls (n = 28 and n = 31). Outcome measures were maternal and perinatal characteristics, reasons for appointment, examination, diagnoses, blood tests, and radiologic findings.

Results: The case-series had a median age of 70 days. A majority were detected through a skeletal survey for abuse investigation. Sixteen were boys, three were preterm and six were small-for-gestational age. Three carers had noticed popping sound from the chest; no infants showed signs of pain at physical examination. Mean number of fractures was 4.2, and 24 had callus. Bone mineralisation was scarcely reported. Metabolic panel was not uniformly analysed. The register case-control had a median age of 76 days, sharing risk factors such as maternal overweight/obesity, male sex, prematurity, and being small-for-gestational age. Cases more often had subdural haemorrhage, retinal haemorrhage, or long-bone fractures, controls more often had neonatal morbidity, respiratory infection, or a fall accident. Detection of fracture at time of a major surgery (n = 6) and rickets/vitamin D deficiency (n = 5) appeared in both groups, but was delayed among the cases.

Conclusions: Rib fractures in young infants, diagnosed as abuse, are usually asymptomatic and healing. A substantial

proportion had metabolic risk factors, suggesting false positive cases.

Keywords: abuse; infant; metabolic bone disease; radiology; rib fractures.

Learning points

- The case-series showed that a few infants presented with a popping sound, none had notes of admitted or witnessed shaking, none had pain, and physical examinations did not reveal any findings. Most had callus at time of diagnosis, no systematic radiology assessments of bone mineralisation were noted. Analysis of a metabolic panel was scarce.
- The register case-control study showed that rib fractures can be considered as incidental findings; for the cases, mainly from skeletal surveys to detect abuse, and for the controls through X-rays done for other reasons (neonatal morbidity, lower respiratory tract infections, major surgery, and accidents).
- Both the case-series and the register case-control study had risk factors for metabolic bone disease. The results indicated that there were false positive cases of maltreatment.

Introduction

A skeletal survey is recommended in the screening procedure for physical abuse. Symptomatic or incidental findings might be rib fractures, which are considered highly indicative of abusive head trauma (AHT) [1–7]. The possible non-accidental trauma mechanism causing rib fractures has not been fully established [8], but the most common theory is thoracic compression when the infant is violently shaken [2]. Rib fractures, including multiple fractures, are rarely reported in association with birth, only in one case-series [9] and one population register study [10]. In both studies, the infants had concomitant clavicle fractures. In the case-series, the clavicle fracture was ipsilateral to the rib fractures [9]. Incidental cases of occult rib fractures have been reported from neonatal units [11–13], and in infants in critical care for congenital

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heart disease [14], and in association to physiotherapy [15], whereas rib fractures caused by cardiopulmonary resuscitation (CPR) are rare [16]. Multiple rib fractures might occur in connection with severe thoracic trauma [17], and have been associated with metabolic bone diseases of infancy, such as nutritional rickets [18], vitamin D deficiency [19], Metabolic Bone Disease of Infancy (MBDI) [20], and osteopathy of prematurity [19, 21, 22]. Symptoms from rib fractures as part of severe thoracic trauma are dramatic (pneumothorax, haemothorax, pulmonary contusion, and lung lacerations or injury to mediastinum) [17]. However, specific symptoms of pain from rib fractures are rarely reported, whether in association with prematurity [22, 23] or during physiotherapy [15]. In one case-series, with rib fractures and clinical and medico-legal referrals, five out of 80 infants cried when they were picked up and, in three cases, parents had noted a popping sound [24].

We have previously shown that rib fractures in infants, before the age of six months, are associated with metabolic bone disease risk factors such as maternal obesity, male sex, being born preterm, multiple births, and diagnoses like rickets, vitamin D deficiency, and calcium metabolism disturbances [25]. However, that population register study did not address clinical history and findings among those with a maltreatment diagnosis, nor did it analyse rib fractures with or without a maltreatment diagnosis. To our knowledge, no previous nationwide study has addressed the care-seeking history, clinical presentation, or detection of rib fracture in infants. The objectives of this study were to describe cases of rib fractures in infants being observed for abuse or with a diagnosis of maltreatment by medical history, symptoms, clinical signs, and radiologic findings and to compare exposure of infants with rib fracture who have or do not have a maltreatment diagnosis.

Methods

Design

The design is a case-series and register case-control study.

Infants (1–365 days), born in the years 1997–2014 ($N = 1,855,267$), with a rib fracture diagnosis (Table 1) were identified in the Swedish National Patient Register (NPR) based on the 10th version of International Classification of Diseases (ICD-10-SE). Data were linked to the Swedish Medical Birth Registry (SMBR), Death Cause Register (DCR), and the Register of Children and Young Persons Subjected to Child Welfare Measures (RCWM), as previously reported [26, 27].

An eligibility flow chart of the case-series and register case-control study is presented in Figure 1.

Table 1: Definition of codes for diagnoses and accidents for the register case-control study, Swedish version of the International statistical Classification of Diseases (ICD10-SE).

Category	Diagnosis	Code (ICD10-SE)
Case definition	Rib fracture	S22.3
	Multiple rib fractures	S22.4
Infant abuse diagnoses	Observation for suspected abuse	Z03.8K
	Battered baby syndrome	Y07.9
	Maltreatment syndrome	T74.1
	Neglect and abandonment	Y06
	Other maltreatment	Y07
Other diagnoses	Retinal hemorrhage	H35.6
	Subdural hemorrhage	I62.9, S06.5
	Long bone fractures	S42.2, S42.3, S42.4, S42.7, S42.8, S52, S72, S82, T10, T12
	Clavicle fracture	S42.0
	Thoracic injury (pleura, lung, bronchus trachea)	S27
	Superficial injury of unspecified body region	T14.0
	Rickets	E55.0
	Vitamin D deficiency	E55.9
	Osteogenesis imperfecta	Q78.0
Accidents	Transport accidents	V01-99
	Fall accidents	W00-19

Case-series

Among 337 infants being observed for suspected abuse or with a maltreatment diagnosis (Table 1), for whom medical records were requested from the paediatric departments, 258 (77%) records were retrieved. Among the missing records, there were cases ($n = 12$) in the NPR having both diagnoses of maltreatment and rib fractures. Cases were those having rib fractures diagnosed by x-ray or chest CT ($n = 26$). Of those, 16 also had an ICD code for rib fracture in the NPR, while 10 did not. One case of rib fractures diagnosed solely by scintigram was excluded (Figure 1).

Medical records and written accounts of the radiologic examinations were reviewed for each patient. Digitalised radiology was not available. The following information was retrieved from the records: (1) reasons for seeking care, (2) symptoms, (3) local signs, (4) localisation, (5) number of fractures per child.

Specific information for rib fractures was: (a) one side or both sides, (b) localisation: posterior, posterolateral, antero-lateral, anterior/costochondral, (c) dating: 0–1 week (fresh), 2–4 weeks (“old”), more than 4 weeks (“older”).

Records were checked for any note on findings of abnormal bone mineralisation: (1) periosteal mineralisation abnormality (subperiosteal new bone formation [SPNBF]), (2) growth plate mineralisation abnormality, (3) ulnar cupping, (4) skull mineralisation

Figure 1: Study eligibility flow chart of children born in Sweden 1997 to 2014 ($N = 1,855,267$) with rib fractures before the age of 1 year identified in the National Patient Register (NPR), Swedish Medical Birth Register (SBMR), Register of Children and Young Persons Subjected to Child Welfare Measures (RCWM) and Death Cause Register (DCR). Case-series identified by record review ($n = 26$), register case-control identified by ICD10-SE codes ($n = 28$ and $n = 31$). ¹Among the missing records there were cases ($n = 12$) in the NPR having diagnoses of both maltreatment and rib fractures. ²Cases included in both the case series and the case-control study ($n = 16$). ³Cases with maltreatment diagnosis but not rib fracture diagnosis in the NPR ($n = 10$).

abnormality, (5) rib mineralisation abnormality, (6) vertebral mineralisation abnormality, (7) looser zones [20].

For the 16 cases also found in the NPR (Figure 1), eight had the ICD-10 code Rib fracture (Table 1), and eight had the ICD-10 code Multiple rib fractures (Table 1), with the number of fractures being 3.6 and 5.3, respectively (T-test 0.385) (not shown in the table).

Records were checked to determine if blood tests had been taken and a metabolic panel had been performed.

Register case-control

The register case-control sample had data only from the health registers. Cases were defined as those having ICD codes for rib fracture and maltreatment in the NPR (Table 1). Controls were defined by selection as those with the same fracture diagnoses, but without a diagnosis of maltreatment. Seven of the controls were excluded, six had an entry in the RCWM, one was a homicide from the DCR (Figure 1).

Co-variables

Co-variables for the register case-control sample were: age at diagnosis (days), maternal age and parity, maternal weight class by body mass index (BMI) (underweight $< 18.5 \text{ kg/m}^2$, normal $18.5\text{--}24.9 \text{ kg/m}^2$, overweight or obesity $\geq 25 \text{ kg/m}^2$), smoker in early pregnancy, pre-eclampsia, mode of delivery, infants' sex, gestational week, birth weight (g), and small-for-gestational age ($<10\%$). Co-variables at time of diagnosis of rib fracture were: any neonatal morbidity, subdural haemorrhage, retinal haemorrhage, long bone fracture, superficial injury of unspecified body region, fall or transport accidents, and any major surgery (head, chest, heart, gastro-intestines, kidney, or

ureters). Co-variables at time of or after diagnosis of rib fracture were: osteogenesis imperfecta, rickets, or vitamin D deficiency (Table 1).

Statistics

Descriptive statistics. Pearson's chi-squared test, Fishers' exact test, and T-test.

Ethics

The Regional Ethical Review Board in Uppsala approved the study (2014-11-19 No. 383). Register linkage was provided by the National Board of Health and Welfare. Approval for accessing medical records of those with a maltreatment diagnosis was obtained from the Regional Ethical Review Board in Uppsala (2015-11-18 No. 383/2).

Results

Case-series

We found 26 cases with rib fractures, based on the inclusion criteria (Figure 1). Median and mean age at diagnosis was 67 and 88.5 days, respectively (range 17–334). Five of the cases with rib fractures had subdural haemorrhage; one had retinal haemorrhage. The fractures were assessed as “old”, “older”, and “both callus and no callus”; one of these cases also had retinal haemorrhage.

Medical history

Out of all 26 cases, 16 (61%) were boys, three were born preterm, six were small-for-gestational age, and one was from a multiple birth. Four mothers had preeclampsia. Three had a vacuum extraction, and three had caesarean delivery. Maternal overweight/obesity was documented in nine out of 19. Two out of 20 were smokers (Table 2).

Reasons for seeking healthcare are shown in Table 3. One reason was carers' concern about possible injury from fall accidents ($n = 6$). The majority of the cases had other reasons for seeking health care, with fractures being incidental findings ($n = 20$): pain or tenderness in legs ($n = 4$), plain abdominal radiography because of vomiting ($n = 2$), and abusive investigation protocol ($n = 14$) because of convulsions ($n = 2$), vomiting ($n = 2$), bruises ($n = 3$), subdural haemorrhage ($n = 4$), and dead twin sister ($n = 1$). In none of those cases did the infant have local pain symptoms from the chest indicating rib fractures. In three cases,

Table 2: Maternal and birth characteristics of infants with rib fractures and maltreatment diagnosis ($n = 26$).

			n, %
Mother			
Age, years	< 20		4 (15)
	20–34		21 (81)
	35+		1 (4)
Parity	I		18 (69)
	II+		8 (31)
Weight class	Underweight		1 (6)
	Normal		9 (47)
	Overweight or obesity		9 (47)
	Missing		7
Smoker early pregnancy	No		18 (90)
	Yes		2 (10)
	Missing		6
Diabetes	No		24 (92)
	Yes		2 (8)
Preeclampsia	No		22 (85)
	Yes		4 (15)
Delivery	SVD ^a		20 (78)
	VE ^b		3 (11)
	CS ^c		3 (11)
Infant			
Sex	Boy		16 (62)
	Girl		10 (38)
Multiple birth			1 (4)
Gestational week	37+		23 (89)
	< 37 ^d		3 (11)
Birth weight	Normal		19 (73)
	SGA ^e < 10%		6 (23)

^aSVD: spontaneous vaginal delivery, ^bVE: Vacuum extraction, ^cCS: caesarean section, ^dGestational weeks: 25, 30, 32, ^eSGA: small-for-gestational age.

Table 3: Reasons for health care utilisation and doctor's physical findings in infants with rib fractures and maltreatment diagnosis ($N = 26$).

		n, %
Reasons for health care utilisation		
Local symptoms		
	Popping sound from the infant's chest	3 (12)
	Pain from the chest	0
Accidents		6 (23)
	Fall accidents	4 (15)
	Other accidents	2 (8)
Incidental		20 (77)
	Convulsions ^a	2 (8)
	Vomiting ^a	2 (8)
	Bruises ^a	3 (12)
	Subdural haemorrhage ^a	4 (15)
	Pain or tenderness in legs	4 (15)
	Swelling/lump over the clavicle ^a	2 (8)
	Dead twin sister ^a	1 (4)
	Plain abdominal radiography because of vomiting	2 (8)
Doctor's physical findings		
	Notes of popping sound/crepitation	1 (4)
	Notes of gripping marks, bruises, or swelling of the chest	0
	Notes of pain or tenderness at chest examination	0

^aFindings from routine protocol because of suspicion of abuse.

carers had noted a popping sound from their child's chest. There was no information on admitted or witnessed shaking of the infant in any case.

Table 3 expands on the doctors' findings. In 25 of the cases with rib fractures, there were no abnormal physical signs from the chest such as bruises, swelling or gripping marks noted by the doctor either before or after the X-ray. In one case, the doctor confirmed a popping sound with local crepitation. Results from analyses of blood tests in a metabolic panel are shown in Table 4. Results were available in less than half of the cases ($n = 12$), having not being performed in six cases, while no lab records were retrieved in eight cases. Tests of calcium and phosphate were all normal. Among 10 tests of alkaline phosphatase, eight showed elevated levels. Parathyroid hormone was elevated in one out of four samples. Vitamin D 25-hydroxy was low in one of three samples taken.

Radiology

Table 5 shows the radiologists' statements on the rib fractures. The 26 infants had a total of 104 rib fractures. However, in one case, only 'multiple fractures' was stated,

Table 4: Blood tests in a metabolic panel in case-series of infants with rib fractures (N = 26; any testing, n = 12, no testing done, n = 6, missing lab records, n = 8).

Blood test	Reference range	n	Normal	High	Low
Calcium	2.1–2.6 mmol/L	12	12	0	0
Calcium (ionised)	2.2–2.7 mmol/L	5	5	0	0
Phosphorus (phosphate)	1.3–2.5 mmol/L	10	10	0	0
ASAT (aspartate aminotransferase)	< 1.2 µkat/L	6	6	0	0
ALAT (alanine aminotransferase)	< 1.1 µkat/L	4	4	0	0
ALP (alkaline phosphatase)	1.2–4.8 µkat/L	10	2	8	0
PTH (parathyroid hormone)	10–65 ng/L, 1.6–9.6 pmol/L	4	3	1	0
Vitamin D 25-hydroxy	> 50 nmol/L	3	2	0	1
Copper	0.75–1.25 mmol/L	1	1	0	0
Magnesium	0.7–0.9 mmol/L	1	1	0	0

not the number. The mean number of rib fractures was 4.2; those having a reported fall accident had a mean of 3.3. Eight cases had four or more fractures, six cases had only one rib fracture. Eleven were left-sided, seven right-sided, and seven were two-sided. The majority had a dorsal/dorso-lateral localisation. In 24 cases, callus, or not fresh fracture, was noted. Out of all rib fractures, 85 had callus or were described as not fresh. Dating was explicitly stated in 13 cases: fresh (n = 2), 2–4 weeks (n = 7), and more than 4 weeks (n = 4). Dating with a reported fall accident was: fresh (n = 1, three fractures), 2–4 weeks (n = 1), and more than 4 weeks (n = 2).

Table 5: Radiographic findings in infants with rib fractures and maltreatment diagnosis in the case-series (n = 26^a).

Mean number of fractures	4.2
Localisation	
Posterior	5
Posterolateral	8
Antero-lateral	1
Not specified	9
Side	
Both sides	7
Left side	11
Right side	7
Callus/not fresh ^b	24
Not specified	1
Dating	
Fresh (0–1 week)	2
2–4 weeks	7
More than 4 weeks	4
Not specified	13
Pleural findings ^c	5

^aIn one case, we only had access to the paediatrician's record, not the radiologist's statement. ^bOne case had rib fractures both with callus and without callus. ^cPleural reaction (n = 3), pleural fluid (n = 1), pleural haematoma (n = 1); fresh (n = 2), one diagnosed on day 43 in connection with a fall accident, the other diagnosed on day 17, born at gestational week 25.

In four cases, either a pleural reaction, fluid, or haematoma was noted. Of those, two had only fresh fractures.

Five cases also had clavicle fractures; all were healing. In three cases, laterality was stated for both clavicle and rib fractures. Of those, two had bilateral rib fractures, and one had only ipsilateral.

In no case was there radiologic signs of pneumothorax or haemothorax. Seven of the infants had metaphyseal changes described as corner fractures, bucket handle fractures, or cupping interpreted in the records as classical metaphyseal lesions (CMLs). Other fractures were three humerus, two radius, and one skull.

Assessment of bone mineralisation was scarcely reported. In six cases, subperiosteal new bone formation (SPNBF) was noted; one case had cupping. Assessment of growth plate mineralisation, looser zones, or mineralisation of skull, ribs, or vertebrae were not mentioned in any of the radiologists' statements.

Out-of-home care

In all, 17 out of 26 were subjected to out-of-home care during a mean of 3.5 years with a range from 73 to 2,946 days. In four of the cases, the only finding was rib fractures. The other cases had fractures to the skull, radius, clavicle, or humerus. Four of the infants with rib fractures had a diagnosis of subdural haemorrhage; two of those cases had out-of-home care and two did not.

Register case-control

Table 6 shows data from the National Patient Registry. The sample had a mean age of 92 days at diagnosis and a median of 76 days. Cases and controls, in this sample, had a mean age of 82.6 days and 100.1 days, respectively, at diagnosis.

Table 6: Register case-control study of rib fracture diagnosis in infants. Cases are those having a maltreatment diagnosis, and controls those with no maltreatment or assault diagnosis, nor a decision on out of home care. Source: National Patient Register. Pearson chi-squared test, Fishers' exact test.

		All (n = 59)	Case (n = 28)	Control (n = 31)	
	Mean (days at diagnosis)	91.9	82.6	100.1	
	Median (days at diagnosis)	76.0	70.0	96.0	
		n (%)	n (%)	n (%)	p-Value
Mother					
Age, years	≤34	52 (88)	27 (96)	25 (81)	0.106
	35+	7 (12)	1 (4)	6 (9)	
Parity	I	32 (54)	16 (57)	16 (52)	0.795
	II+	27 (46)	12 (43)	15 (48)	
Weight class ^a	Normal	24 (50)	11 (52)	14 (50)	0.584
	Overweight or obesity	24 (50)	10 (48)	14 (50)	
Smoker early pregnancy	No	50 (93)	23 (89)	27 (96)	0.342
	Yes	4 (7)	3 (11)	1 (4)	
Diabetes	Yes	1 (2)	1 (4)	0	0.475
Preeclampsia	Yes	4 (7)	4 (14)	0	0.045
Birth					
Delivery	SVD ^b	38 (64)	20 (71)	18 (58)	0.011
	VE ^c	7 (12)	0	7 (23)	
	CS ^d	14 (24)	8 (29)	6 (19)	
Sex	Boy	46 (78)	20 (71)	26 (84)	0.348
	Girl	13 (22)	8 (29)	5 (16)	
Multiple birth		7 (12)	4 (14)	3 (10)	0.698
Gestational week	37+	46 (78)	23 (82)	23 (74)	0.677
	< 37	13 (22)	5 ^g (18)	8 ^h (26)	
Birth weight	Normal	50 (85)	21 (75)	29 (94)	0.075
	SGA ^e < 10%	9 (15)	7 (25)	2 (6)	
	Birth weight > 4,000 g	9 (15)	2 (7)	7 (23)	
Diagnoses at time of rib fracture					
	Neonatal morbidity day 1–7	6 (10)	0	6 ^f (19)	0.014
	Subdural haemorrhage	9 (15)	7 (25)	2 (7)	0.071
	Retinal haemorrhage	4 (7)	3 (11)	1 (3)	0.337
	Long bone fracture	17 (29)	15 (48)	2 (7)	0.000
	Clavicle fracture	7 (12)	5 (18)	2 (7)	0.235
	Any other thoracic injury	0	0	0	
	Superficial injury of unspecified body region	1 (2)	0	1 (3)	0.475
	Lower respiratory infection ^h	6 (10)	1 (4)	5 (16)	0.136
Major operation at time of rib fracture		6 (10)	4 (14)	2 (7)	0.051
Accidents at time of rib fracture					
	Fall ⁱ	12 (20)	2 ^j (7)	10 ^k (33)	0.012
	Transport	2 (3)	1 (4)	1 ^l (3)	0.673
Diagnoses during infancy					
	Osteogenesis imperfecta	2 (3)	1 ^m (4)	1 ⁿ (3)	1.0
	Rickets or VDD ^o	5 (9)	2 ^p (7)	3 ^q (10)	1.0

^aMissing (n = 11), ^bSVD: spontaneous vaginal birth, ^cVE: vacuum extraction, ^dCS: caesarean section, ^eSGA: small-for-gestational age, ^fNeonatal morbidity: extreme prematurity (n = 1), other birth-related fractures (n = 5), respiratory distress syndrome (n = 1), malformations of larynx or oesophagus (n = 3). ^g54 days after diagnosis of rib fracture, ^h1 case < 32 gestational weeks, ⁱcases < 32 gestational weeks, ^jPneumonia (n = 2), bronchitis (n = 3), ^kAccident on day of diagnosis of rib fracture or < 7 days before this day, ^lFall from being carried (n = 2), ^mfall from being carried (n = 3), fall from furniture (n = 3), unspecified (n = 3). Other injuries: concussion (n = 2), skull fracture (n = 4), subdural haemorrhage (n = 1), liver injury (n = 1), no other injuries (n = 4) ^lSkull fracture, subdural and subarachnoidal haemorrhage, spleen injury, fractures of pelvis and spinal column, ^mOI diagnosed 34 days after ⁿOI diagnosed 309 days after. ^oVDD: vitamin D deficiency ^p31 and 184 days after respectively, ^qSame day.

In all, including both cases and controls in this sample, 50% of the mothers had overweight or obesity, 78% of the children were boys, 22% were preterm born, 12% were multiple born, 15% had a birth weight of over 4,000 g, and 15% were small-for-gestational age. Four of the cases had mothers with preeclampsia. None were delivered by vacuum extraction and fewer cases than controls were born at less than 32 gestational weeks. At time of diagnosis, cases more often had subdural haemorrhage ($n = 7$), retinal haemorrhage ($n = 3$) and long bone fractures ($n = 15$), while controls at time of diagnosis more often had neonatal morbidity ($n = 6$), lower respiratory infection ($n = 5$) and considerably more fall accidents reported ($n = 10$). No other thoracic injury was diagnosed among the cases or the controls. There were both cases ($n = 5$) and controls ($n = 2$) with clavicle fractures. One case of superficial injury to an unspecified body region was noted among the controls, with none among the cases.

There were both cases ($n = 4$) and controls ($n = 2$) with rib fractures detected in connection with major surgery.

One case of osteogenesis imperfecta was diagnosed in each group, 34 and 309 days after diagnosis of rib fracture, respectively. Rickets or vitamin D deficiency were diagnosed for the controls at time of diagnosis of rib fracture ($n = 3$); for the cases this was 31 and 184 days after diagnosis of rib fracture and maltreatment, respectively ($n = 2$).

Discussion

We have shown that infants with rib fractures and an abuse diagnosis were very young, that fractures were often multiple, and that patients were usually diagnosed incidentally at a routine skeletal survey. Rib fractures were rarely associated with physical findings. Controls with rib fractures shared risk factors of metabolic bone disease with cases of abuse.

The present study confirms that rib fractures in connection with abuse diagnosis are found at young age [20], in parallel with AHT diagnosis [6]. We found a preponderance of males, young age, more multiple births, small-for-gestational age (<10%) and preterm born, both in the case-series and the register case-control study (both cases and controls), and bone fragility diagnoses in the register case-control study (both cases and controls), as previously reported [25], supporting an association with metabolic bone disease [13, 20, 25].

Rib fractures may occur during birth [7]. We have previously shown that rib fractures diagnosed among neonates have an association with difficult birth [10]; the

register case-control study supports this, as the controls were more often delivered by vacuum extraction and there was a higher proportion of big babies among controls. It is possible that there were birth-related rib fractures in the case-series, but this could not be ascertained. Out of the three cases with concomitant clavicle fracture, one case had only ipsilateral rib fractures [9].

The majority in the case-series, 20/26, were incidental findings from routine skeletal surveys, i.e., occult and without symptoms; in only one case were there clinical findings confirmed before the X-ray. Most of the detected rib fractures in the register case-control sample, among both cases and controls, can probably be described as incidental findings. For cases, findings were often a result of a screening protocol for abuse (concomitant findings of subdural haemorrhage, retinal haemorrhage, and CMLs), for controls, as a result of X-rays for other reasons (neonatal intensive care, respiratory infection and accidents), and for both cases and controls in connection with major surgery, something not previously reported, to our knowledge. No bruises, swelling, or gripping marks were noted as physical findings in the case-series, whereas in the register case-control, one control and none of the cases had bruises. This is consistent with the present knowledge that rib fractures seldom give symptoms and may not be clinically detectable [1, 19], thus challenging the idea of a traumatic origin for many of the rib fractures diagnosed as abuse.

Twenty in the case-series had two or more rib fractures, eight cases had four or more fractures. Neither the findings from the case-series nor those from the register case-control support a severe traumatic cause of the findings in our samples, in contrast to when rib fractures are found in connection with severe thoracic injury [17]. Multiple rib fractures are reported to be more prevalent in association with an abuse diagnosis [4], but are also reported in association with metabolic bone disease [20, 24]. Our case-series had mainly posterior or posterior-lateral fractures, which have previously been shown to be indicative of abuse [4, 7]. However, this not confirmed in a recent systematic literature review [5]. Posterior rib fractures are also associated with metabolic bone disease [22]. We found a preponderance of left-sided rib fractures in our case-series, which has been hypothesised to be associated with greater force of torso compression exerted by a right-handed perpetrator [28].

The current study found that 11 out of 13 had healing rib fractures, which is in accordance with proportions observed in the past, being from 60 to 89% [7, 29]. This finding may be interpreted as an anomaly in relation to the

proposed mechanism that violent shaking causes rib fractures, as cerebral symptoms of AHT are thought to occur soon after shaking [6].

Metabolic bone disease is in the differential diagnosis of rib fractures when there is no trauma reported. We have previously shown that metabolic bone disease risk factors are associated with rib fractures [25]. However, contrary to a case-series where periosteal mineralisation abnormality was found in 29 infants out of 55 having rib fractures [20], we found only six out of 26 had any note of abnormal mineralisation. One interpretation of this difference may be that in our study the radiologists did not include a systematic assessment of growth plate mineralisation, looser zones, and mineralisation in their reports [20]. However, in both the case-series and the register case-control study, risk factors of metabolic bone disease such as maternal overweight and prematurity were at hand, supporting the theory of an underlying bone fragility [25]. The link between maternal obesity and rib fractures could be a lower transfer of vitamin D to the foetus [30].

None in the case-series had any notes of admitted shaking or other abuse. This is in contrast to the suggested causal mechanism of rib fractures [2].

Considering the stated high predictivity of abuse for rib fracture, it might be unexpected to find an equal number of cases and controls in the Swedish setting. However, the majority of the rib fractures among the controls can be interpreted as incidental findings, i.e., occult fractures. Whether there were other circumstances supporting an abuse diagnosis for the cases, for example at time of diagnosis of a lower respiratory infection, major operations, or accident, cannot be ascertained with the register case-control design.

The majority in our case-series were subjected to out-of-home care, some with rib fractures as the only medical finding. The scientific foundation for a connection between rib fractures and abuse is considered to be solid [1, 2, 4, 5]; rib fractures have even been reported to have a positive predictive value of 100% for abuse [4]. However, methodological flaws, e.g., circular reasoning, might undermine the causality of violent torso compression and rib fractures [31]. A recent systematic review on rib fracture and abuse diagnosis with a critical approach to the choice of a reference test (reference group) concludes that there is insufficient scientific evidence to determine the diagnostic accuracy of rib fractures, as well as CMLs, being caused by abuse [32].

One fourth, both among those the case-series and among cases in the register case-control, had subdural haemorrhage and a few had retinal haemorrhage. Based on

the rationale that subdural haemorrhage and retinal haemorrhage are independent of fractures, while each of these findings is also considered as highly indicative of physical abuse, the co-occurrence of such findings most likely placed these infants in the maltreatment category. However, although intracranial findings and ocular findings are indisputably independent of fractures, certain kinds of non-traumatic subdural effusions, such as benign external hydrocephalus with or without acute haemorrhage, are associated with retinal haemorrhage [33, 34], and infantile rickets is associated with all types of hydrocephalus [35]. Thus, it is possible that concomitant intracranial pathology, ocular pathology, and certain kinds of fractures have a common non-traumatic underlying cause in infantile rickets. However, investigating this was outside the scope of the present study.

The diagnostic process when having a finding of a rib fracture, whether occult or symptomatic, warrants improvement. Our previous [25] and present findings suggest that metabolic bone disease needs to be assessed. Biochemical markers for metabolic bone disease should be measured as soon as possible. In the case-series, only two thirds had any taken blood tests for a metabolic panel, with no uniformity in the biochemical testing ordered, which might be explained by such testing not being recommended in the screening protocol for maltreatment during the study period [35, 36]. Nor could a consistent profile of metabolic bone disease from blood analytes be seen, as previously reported [20]. One reason for this might be that biochemical markers for metabolic bone disease are quickly normalised after birth through micronutrient supplements [37], and Vitamin D supplementation [38]. However, in the case-series, three cases had a delayed diagnosis of OI or rickets/VDD in another entry in the register than when a diagnosis of abuse or suspected abuse was made, indicating false positives for abuse. The assessment from skeletal X-ray can be standardised [20] or digital X-ray radiogrammetry can be applied [39]; alternatively, investigations through dual energy X-ray absorptiometry (DXA) or quantitative ultrasound (QU) might be of value.

Strength and limitations

The strength of the study is its national coverage and comprehensive case finding. We cannot exclude a detection bias in the case-series, as 23% of the records were not retrieved. The same may apply to the register case-control study, as there are some cases subjected to out-of-home

care that do not have a maltreatment diagnosis; however, they were excluded from the register case-control analysis. A strength of the study was the combination of the two study designs – case-series based on record review and population-based register case-control – as the findings supplement one another. The record review indicates that a considerable part of rib fractures is not reported to the NPR. However, we do not believe that this would have significantly changed our findings in either direction. Quality aspects of the ICD coding hampered analysis. The ICD codes Rib fracture and Multiple Rib fractures did not serve to discriminate between single rib fracture and multiple fractures, nor is laterality part of ICD. A limitation of the case-series was that we had no access to the radiographs. A weakness of the register case-control study was that we did not have access to the clinical records of the controls. Further research is thus warranted: a case-control study of rib fractures with or without a maltreatment diagnosis, with full access to medical records, metabolic panels, and radiographs would provide more in-depth knowledge.

Conclusion

Rib fractures are primarily detected by skeletal survey due to suspected abuse or from X-rays taken for other reasons in young infants without clinical symptoms from rib fractures, which were usually asymptomatic and healing, i.e., occult findings. A substantial proportion had risk factors for metabolic bone disease, but only a few had a later diagnosis of rickets or vitamin D deficiency. The lack of systematic radiologic assessments of bone mineralisation and metabolic panels would suggest that there were false positive cases of maltreatment. The current data highlight the importance of evidence-based assessment when determining the cause of unexplained rib fractures in infants.

What is already known on this topic

- Rib fractures are considered highly indicative of abusive head trauma and a skeletal survey of the chest is recommended when abuse is suspected.
- The possible non-accidental trauma mechanism has not been fully established.
- Incidental findings of rib fractures are reported from neonatal units. Rib fractures are reported in association with metabolic bone disease of infancy.

What this study adds

- The case-series showed that a few infants presented with a popping sound, none had notes of admitted or witnessed shaking, none had pain, and physical examinations did not reveal any findings. Most had callus at time of diagnosis, no systematic radiology assessments of bone mineralisation were noted. Analysis of a metabolic panel was scarce.
- The register case-control study showed that rib fractures can be considered as incidental findings; for the cases, mainly from skeletal surveys to detect abuse, and for the controls through X-rays done for other reasons (neonatal morbidity, lower respiratory tract infections, major surgery, and accidents).
- Both the case-series and the register case-control study had risk factors for metabolic bone disease. The results indicated that there were false positive cases of maltreatment.

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Informed consent: Informed consent was obtained from all individuals included in this study.

Ethical approval: Research involving human subjects complied with all relevant national regulations, institutional policies and is in accordance with the tenets of the Helsinki Declaration (as revised in 2013), and has been approved by the Regional Ethical Review Board in Uppsala approved the study (2014-11-19 No. 383). Approval for accessing medical records of those with a maltreatment diagnosis was obtained from the Regional Ethical Review Board in Uppsala (2015-11-18 No. 383/2).

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