

The Epidemiology of Infant Shaft Fractures of Femur or Humerus by Incidence, Birth, Accidents, and Other Causes

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Abstract

Background: Knowledge of femur and humerus shaft fractures during infancy is scarce. The purpose of this population-based registry study was to analyze both birth-related femur and humerus shaft fractures and those diagnosed later in infancy, as regards incidence, perinatal characteristics, other diagnoses, and reported accidents.

Methods: Children born in 1997–2014, diagnosed with a femur or humerus shaft fracture before age 1 year, were identified in the Swedish Health Registries. Rate of birth fractures were estimated by combining femur and humerus shaft fractures coded as birth-related with femur and humerus shaft fractures diagnosed during day 1-7 without registered trauma, abuse, or bone fragility disorders. Incidence was computed by comparing infants with femur or humerus shaft fractures to the total at-risk population.

Results: The incidence for birth-related femur shaft fractures was 0.024‰ (n=45) and that for birth-related humerus shaft fractures was 0.101‰ (n=188). The incidence was 0.154‰ for later femur shaft fractures (n=287) and 0.073‰ for later humerus shaft fractures (n=142). Birth-related femur shaft fracture was associated with preterm, multiple birth, breech, cesarean, and small-for-gestational age, while humerus shaft fracture was associated with preterm, multiple birth, birth weight > 4,000 g, and shoulder dystocia. A bone fragility diagnosis was recorded in 5% of those with birth-related or later femur shaft fractures. Among infants with birth-related humerus shaft fractures, 1% had a bone fragility diagnosis; the figure for later fractures was 6%. Maltreatment diagnosis was associated with both fracture types, especially among those aged < 6 months, where approximately 20% and 14% of cases, respectively, were associated with abuse. Fall accidents were reported in 73% and 56% among those with later femur and humerus shaft fractures, respectively.

Conclusion: This study provides data on epidemiology, birth, parental characteristics, and reported accidents in relation to femur and humerus shaft fractures during infancy. Few children had a bone fragility diagnosis. Fall accidents were the main contributor to femur or humerus shaft fracture during infancy; however, the proportion of fractures attributed to maltreatment was high in children under 6 months.

Background

Shaft fractures of femur and humerus during infancy can occur in connection with birth or later. Birth-related shaft fractures of femur and humerus are rare and reported as case reports or small case series. A few studies, which did not include fractures to other parts of the femur or humerus, have reported birth-related femur shaft fracture rates from 0.13‰ (number of patients = 7) [1] to 0.14‰ (n = 4) [2] and birth-related humerus shaft fracture rates from 0.04‰ (n = 4) [3] to 0.06‰ (n = 2) [2]. Femur shaft fracture among infants, aged under one year, had an annual incidence of 0.16‰ (n = 313) in Sweden in the years 1987 to 2005. However, birth-related fractures were excluded from that population-based registry study [4]. In a study analyzing treatment of femur shaft fractures in children aged 0 to 15 years of age (total study population n = 1,852), Talbot et al. reported an incidence of 0.06‰ in children aged under one year [5]. The incidence of humerus shaft fractures during infancy has, to our knowledge, not been addressed before.

Adverse birth and infant characteristics reported for birth-related femur and humerus shaft fractures include: multiple birth, low birth weight, breech extraction, cesarean section, vacuum extraction, and fetal osteoporosis [1, 2, 6–8]. To the best of our knowledge, the relationships between maternal obesity or preeclampsia and shaft fractures of femur and humerus during infancy have not been studied. Preterm birth (< 37 weeks) has been associated with a 2.1–2.3-fold increased risk of future hospitalization for femur fractures before the age of 6 months [9, 10].

Most femur and humerus shaft fractures diagnosed later during infancy are related to fall accidents [4, 5, 11] but both bone fragility [10] and non-accidental trauma [4, 5, 12] are correlated in this age group. A recent large multicenter hospital study reported that one third of humerus fracture (excluding supracondylar fractures) and one fifth of femur fractures (to any part of the femur), in children under the age of one year, were associated with probable abuse [12].

We have previously, in a study not specifically addressing shaft fractures, reported the incidence of birth-related fractures for infants born in Sweden in 1997 to 2014, that diagnosis can be delayed, and that other fractures diagnosed among neonates might also be associated with birth [6]. The objective of this population-based registry study was to study both birth-related and later shaft fractures of femur or humerus during infancy, in children under the age of one year. The aim was to analyze these fractures by sex, incidence, age at diagnosis, associations with prenatal exposure, birth complications, infants' characteristics, accidents, and other diagnoses during infancy.

Methods

Design

This was a nationwide population-based register study that comprised 1,855,267 infants born in Sweden between January 1997 and December 2014. We followed up the infants and their mothers until the children were one year of age using data from the National Patient Register (NPR), the Swedish Medical Birth Register (SMBR), and the Register of Children and Young Persons Subjected to Child Welfare Measures (RCWM) maintained by the Swedish National Board of Health and Welfare [13]. During the study period, the NPR used diagnosis codes from the Swedish version of International Classification of Diseases (ICD-10-SE).

Cases

Among all children born during the 18-year period, we selected cases diagnosed with Birth injury to femur (P13.2), Birth injury to other long bones (P13.3), Fracture of shaft of femur (S72.3), and/or Fracture of shaft of humerus (S42.3) (Table S1).

Femur shaft fractures

We found 39 infants with birth-related femur fractures (P13.2). Among them, 9 also had a femur shaft fracture diagnosis (S72.3), while 6 had S codes indicating fractures of other parts of the femur. We have assumed that the 24 birth-related femur fractures with no additional S code indicating which part of the femur was fractured were shaft fractures. One child had both a birth-related femur fracture and a femur shaft fracture on day 349 and was included in both of these categories. We found 299 infants with femur shaft fractures (S72.3) without a P code. Among them, 12 were diagnosed at the age of 1–7 days without a reported fall accident. We have assumed these fractures (none of them had a maltreatment diagnosis) were also birth-related. These 45 cases formed the study base for birth-related femur shaft fractures. We found 287 infants diagnosed with femur shaft fractures (S72.3), without a presumed birth-related femur shaft fracture, and these cases formed the study base for femur shaft fractures diagnosed later in infancy (Fig. 1).

Humerus shaft fractures

We found 144 infants with birth-related long bone fractures (P13.3). Among them, 54 also had a humerus shaft fracture diagnosis (S42.3), 84 had no S codes indicating other long bone fractures, 4 had both a humerus shaft fracture and a fracture of proximal or distal humerus, and 6 had S codes indicating fractures of other parts than the humerus shaft, but not humerus shaft fractures. We found 192 infants with humerus shaft fractures (S42.3) without a P code; among them, 50 were diagnosed at the age of 1–7 days without a reported fall accident. We have presumed these fractures were birth-related (none of them had a maltreatment diagnosis) and the estimated 188 cases formed the study base for birth-related humerus shaft fractures. We found 142 infants with humerus shaft fractures (S42.3), without a birth-related long bone fracture, and these cases formed the study base for humerus shaft fractures diagnosed later in infancy (Fig. 2).

Population

Population controls were all infants without shaft fractures of humerus or femur; for maternal and perinatal exposure (SBMR), the control population was 908,324, and for other diagnoses (NPR) during infancy, the control population was 1,855,267, as described in a previous publication [10].

Exposure

We categorized a number of exposures. The prenatal exposures included obesity (body mass index (BMI) at the start of pregnancy > 29.9 kg/m²) and preeclampsia. Delivery factors were dystocic labor and mode of delivery (spontaneous vaginal birth, vacuum-assisted, planned or emergency cesarean). The infant factors were sex, single or multiple birth, term or preterm birth (< 37 gestational weeks), birth weight ($> 4,000$ grams), small-for-gestational age (SGA) ($< 2.5^{\text{th}}$ or $< 10^{\text{th}}$ percentiles), and large-for-gestational age (LGA) ($> 2.5\%$). ICD codes for exposures, including accidental injury (± 7 days) and bone fragility, are defined in Table S1 [see Additional file 1]. Bone fragility disorders were defined as having a diagnosis of osteogenesis imperfecta, rickets, or vitamin D deficiency before the age of one year. Maltreatment was defined as having a maltreatment diagnosis (Additional file 1) or an entry in the RCWM during the first year of life.

Statistical analyses

Data are presented as descriptive statistics (mean and median). Time of birth-related fracture diagnosis during the first month was categorized into early or late neonatal period (1–7 days and 8–28 days). The age of each child with a later femur or humerus shaft fracture was categorized as < 6 months or 6–12 months. The incidence rate, the proportion of cases per 1,000 infants with 95% confidence intervals (CI), was computed by comparing the number of children with a fracture (ICD-10 code: S72.3, S42.3, P13.2, or P13.3) to the total number of children at risk. Fisher's exact test or Pearson's chi-squared test was applied to assess differences. Statistical significance was defined as $P < 0.05$. To assess sex differences in relation to the categories of femur and humerus shaft fractures, crude and adjusted odds ratios were analyzed. For the statistical analyses, we used the statistical software package SPSS, version 25.0 (IBM Corp, New York, USA).

Results

Incidence, sex, and time of diagnosis

The incidence rate per 1,000 children for the estimated 45 infants with birth-related femur shaft fractures (27 boys, 60%, p value .535) was 0.024 [95% confidence interval (CI), 0.017–0.031] (Table 1). Fractures were registered during both the early (n = 39) and late (n = 4) neonatal period; however, 2 were registered after the age of one month. The incidence rate per 1,000 children for the 287 infants with later femur shaft fractures (141 boys, 49.1%, p value .389) was 0.154 (95% CI, 0.138–0.170). Mean and median ages were 192 and 210 days. Over half (59%) were diagnosed during the sixth to twelfth month of life.

Table 1

Total number of femur and humerus shaft fractures, sex-specific, age-specific incidence rate per 1,000 children and 95% confidence intervals (CIs). Crude odds ratio (OR) and adjusted OR for association between sex (girl reference) and femur and humerus shaft fractures by age group.

| Age | Incidence per 1,000 children (CI) | | | Crude OR (CI) | Adjusted ^a OR (CI) |
|--|-----------------------------------|---------------------|---------------------|-------------------------|-------------------------------|
| | Girls | Boys | Total | | |
| <i>Femur shaft fracture</i> | | | | | |
| Birth-related | 0.010 (0.005–0.014) | 0.015 (0.009–0.020) | 0.024 (0.017–0.031) | 1.40 (0.77–2.55) | 1.33 (0.67–2.65) |
| < 6 months | 0.025 (0.018–0.032) | 0.030 (0.022–0.037) | 0.055 (0.045–0.065) | 1.09 (0.74–1.62) | 0.79 (0.59–1.07) |
| 6–12 months | 0.053 (0.043–0.064) | 0.046 (0.037–0.056) | 0.100 (0.086–0.113) | 0.81 (0.61–1.08) | 0.79 (0.60–1.07) |
| <i>Humerus shaft fracture</i> | | | | | |
| Birth-related | 0.035 (0.027–0.043) | 0.066 (0.055–0.078) | 0.101 (0.087–0.115) | 1.81 (1.34–2.45) | 1.65 (1.20–2.28) |
| < 6 months | 0.020 (0.014–0.027) | 0.031 (0.023–0.039) | 0.052 (0.042–0.062) | 1.43 (0.95–2.15) | 1.43 (0.94–2.19) |
| 6–12 months | 0.011 (0.006–0.016) | 0.014 (0.008–0.019) | 0.025 (0.018–0.032) | 1.11 (0.62–1.99) | 1.24 (0.67–2.31) |
| ^a Adjusted for large-for-gestational age (> 2.5th percentile), birth weight > 4,000 g, small-for-gestational age (< 2.5th percentile), preterm-born, shoulder dystocia. | | | | | |
| Significant odds ratios are written in bold. | | | | | |

The incidence rate per 1,000 children for the estimated 188 infants with birth-related humerus shaft fractures (123 boys, 66%, p value .000) was 0.101 (95% CI, 0.087–0.115); the adjusted odds ratio was 1.65 (95% CI, 1.20–2.28) (Table 1). Fractures were registered during both the early (n = 162) and late (n = 24) neonatal period; however, 2 were registered after the age of one month. The incidence rate per 1,000 children for the 142 infants with later humerus shaft fractures (83 boys, 59%, p value .106) was 0.073 (95% CI, 0.065–0.089). Mean and median ages were 116 and 98 days. Over half (72%) were diagnosed during the first to fifth month of life.

Maternal, birth and child characteristics

The infants with birth-related femur shaft fractures had several statistically significant differences in background characteristics compared with the general population, such as being born by cesarean (74%), born in breech (76%), preterm (25%), SGA < 10% (36%), SGA < 2.5% (12%), with shoulder dystocia (4%), or in a multiple birth (20%). However, no associations were noted for infant sex, dystocia, or vacuum-assisted delivery. Among the later femur shaft fractures at age < 6 months, there was a significantly higher proportion of children born preterm (18%), SGA < 10% (21%), SGA < 2.5% (7%), or in a multiple birth (11%) compared with the general population. Shaft fractures diagnosed at age 6 to 12 months had a higher proportion of SGA < 10% (16%) (Table 2).

Table 2

Femur shaft fractures diagnosed among infants born in Sweden 1997 to 2014, birth-related (n = 45) or later (n = 287), by maternal, birth, and child characteristics, accidents, and other diagnoses.

| | Population | Femur shaft fracture | | | | | |
|--|---------------------------|----------------------|----------|----------------------|----------|--------------------|----------|
| | | Birth-related | | | Later | | |
| | | | | Age (months) | | | |
| | n = 908,324 | (n = 45) | | < 6 (n = 102) | | 6 to 12 (n = 185) | |
| | n (%) | n (%) | p value* | n (%) | p value* | n (%) | p value* |
| Mother | | | | | | | |
| Obesity | 302,613 (37) | 16 ^a (46) | .299 | 38 (43) | .324 | 75 (45) | .021 |
| Preeclampsia | 28,706 (3.2) | 1(2) | 1.0 | 7 (7) | .043 | 10 (5) | .089 |
| Birth | | | | | | | |
| Dystocic labor | 107,225 (11.8) | 4 (9) | .816 | 10 (10) | .646 | 18 (10) | .423 |
| Shoulder dystocia | 2,005 (0.2) | 2 (4) | .005 | 0 | | 0 | |
| Cesarean section | 161,554 (18) | 34 (74) | .000 | 30 (29) | .003 | 32 (17) | .915 |
| Vacuum | 62,099 (6.8) | 4 (9) | .775 | 7 (7) | 1.0 | 18 (10) | .267 |
| Forceps | 2,473 (0.3) | 0 | | 1 (1) | .243 | 0 | |
| Child | | | | | | | |
| Boy | 469,344 (52) | 27 (60) | .535 | 55 (54) | .902 | 86 (47) | .369 |
| Multiple birth | 26,639 (2.9) | 9 (20) | .000 | 11 (11) | .000 | 10 (5) | .074 |
| Breech | 33,998 (3.3) | 31 (76) | .000 | 10 (10) | .005 | 5 (3) | .564 |
| Preterm | 56,521 (6.2) | 11 (25) | .008 | 18 (18) | .000 | 12 (7) | .878 |
| SGA ^b (< 10%) | 94,217 (10.4) | 15 (36) | .000 | 21 (21) | .003 | 29 (16) | .018 |
| SGA ^b (< 2.5%) | 20,561 (2.3) | 4 (12) | .041 | 6 (7) | .020 | 5 (3) | .610 |
| LGA ^c (> 2.5%) | 32,172 (3.7) | 1 (3) | 1.0 | 2 (2) | .776 | 6 (3) | 1.0 |
| Birth weight > 4,000 grams | 169,517 (19) | 5 (11) | .251 | 9 (9) | .013 | 33 (18) | .843 |
| Other diagnosis during infancy | | | | | | | |
| Osteogenesis imperfecta | 70 (0.004) ^d | 2 (4) | .000 | 5 (5) | .000 | 8 ^e (4) | .000 |
| Rickets or VDD ^f | 259 (0.01) ^d | 0 | | 2 (2) | .000 | 1 (0.5) | .000 |
| Maltreatment | 1,347 (0.07) ^d | 0 | | 21 ^g (20) | .000 | 6 (3) ^h | .000 |
| *Pearson's chi-squared or Fisher's test | | | | | | | |
| ^a Missing (n = 10), ^b Small-for-gestational age, ^c Large-for-gestational age, ^d Per 1,855,267, ^e Entry in the Register of Children and Young Persons Subjected to Child Welfare Measures (n = 1) in connection with the fracture and OI diagnosis 137 days after the fracture, ^f Vitamin D deficiency, ^g Maltreatment diagnosis (n = 10), entry only in the Register of Children and Young Persons Subjected to Child Welfare Measures (n = 11); other diagnoses: subdural hemorrhage (n = 2), retinal hemorrhage (n = 1), rib fractures (n = 4), skull fractures (n = 2), ^h Maltreatment diagnosis (n = 4), entry only in the Register of Children and Young Persons Subjected to Child Welfare Measures (n = 2). | | | | | | | |

Children with birth-related humerus shaft fractures had several statistically significant differences in background characteristics compared with the population. Half of the mothers had overweight/obesity. Adverse birth characteristics were shoulder dystocia (37%), vacuum-assisted delivery (25%), male sex (66%), birth weight of more than 4,000 g (44%), LGA (29%), breech (10%), preterm-born (13%), and multiple birth (7%). Among the children with later humerus shaft fractures at age < 6 months, there was a significantly higher proportion of

maternal obesity (54%), shoulder dystocia (17%), LGA (14%), and birth weight of more than 4,000 g (33%). Few of those diagnosed at age 6 to 12 months had adverse perinatal characteristics (Table 3).

Table 3
Humerus shaft fractures among infants born in Sweden 1997 to 2014, birth-related (n = 201) and later (n = 136) by maternal, birth, and child characteristics, other fractures, and diagnoses.

| | Population | Humerus shaft fracture | | | | | |
|--|---------------------------|------------------------|----------|----------------------|----------|--------------------|----------|
| | | Birth-related | | | Later | | |
| | | Age in months | | | | | |
| | (n = 908,324) | (n = 188) | | < 6 (n = 96) | | 6 to 12 (n = 46) | |
| | n (%) | n (%) | p value* | n (%) | p value* | n (%) | p value* |
| Mother | | | | | | | |
| Obesity | 302,623 (37) | 81 ^a (47) | .007 | 46 (54) | .001 | 21 (46) | .285 |
| Preeclampsia | 28,968 (3.2) | 9 (5) | .205 | 4 (4) | .550 | 3(7) | .177 |
| Birth | | | | | | | |
| Dystocic labor | 107,215 (11.8) | 43 (23) | .000 | 12 (13) | .753 | 8 (17) | .249 |
| Shoulder dystocia | 2,005 (0.2) | 69 (37) | .000 | 16 (17) | .000 | 0 | |
| Cesarean section | 161,554 (18) | 35 (19) | .782 | 25 (26) | .040 | 12 (26) | .174 |
| Vacuum | 69,101 (8) | 47 (25) | .000 | 10 (10) | .331 | 2 (4) | .581 |
| Forceps | 2,473 (0.3) | 1 (0.5) | .401 | 1 (1) | .230 | 0 | |
| Child | | | | | | | |
| Boy | 469,339 (52) | 123 (66) | .000 | 58 (60) | .230 | 25 (54) | .936 |
| Multiple birth | 26,645 (2.9) | 13 (7) | .004 | 4 (4) | .369 | 5 (11) | .011 |
| Breech | 34,007 (3.3) | 18 (10) | .000 | 5 (5) | .412 | 5 (11) | .028 |
| Preterm | 56,533 (6.2) | 24 (13) | .001 | 9 (9) | .201 | 2 (4) | 1.0 |
| SGA ^b (< 10%) | 94,244 (10.4) | 16 (9) | .541 | 14 (15) | .181 | 9 (20) | .052 |
| SGA ^b (< 2.5%) | 20,566 (2.3) | 3 (2) | .802 | 3 (3) | .478 | 4 (10) | .015 |
| LGA ^c (> 2.5%) | 32,141 (3.7) | 50 (29) | .000 | 13 (14) | .001 | 0 | |
| Birth weight > 4,000 grams | 169,479 (19) | 82 (44) | .000 | 32 (33) | .000 | 4 (9) | .089 |
| Other diagnosis during infancy | | | | | | | |
| Osteogenesis imperfecta | 76 (0.004) ^d | 1 (0.5) | .017 | 3 (3) | .000 | 4 ^e (9) | .000 |
| Rickets or VDD ^f | 261 (0.01) ^d | 1 (0.5) | .052 | 1 (1) | .000 | 0 | |
| Maltreatment | 1,347 (0.07) ^d | 0 | | 12 ^g (14) | .000 | 2 ^h (4) | .000 |
| *Pearson's chi-squared test, Fisher's test | | | | | | | |
| ^a 17 missing BMI, ^b Small-for-gestational age, ^c Large-for-gestational age, ^d Per 1,855,267, ^e Entry in the Register of Children and Young Persons Subjected to Child Welfare Measures (n = 1) in connection with the fracture and OI diagnosis 21 days after the fracture, ^f Vitamin D deficiency, ^g Maltreatment diagnosis (n = 9), entry in the Register of Children and Young Persons Subjected to Child Welfare Measures (n = 3); other diagnoses: subdural hemorrhage (n = 1), retinal hemorrhage (n = 1), rib fractures (n = 2), skull fractures (n = 1), ^h Maltreatment diagnosis (n = 2), entry only in the Register of Children and Young Persons Subjected to Child Welfare Measures (n = 0). | | | | | | | |

Osteogenesis imperfecta, rickets, vitamin D deficiency, abuse, and accidents

Two (4%) of the infants with a birth-related femur shaft fracture had osteogenesis imperfecta (OI). Neither had a later maltreatment diagnosis. Among later femur shaft fractures, 13 (5%) had OI; three of those cases were diagnosed 61 to 137 days after the fracture, of which one had a maltreatment diagnosis before the OI diagnosis. Also among later femur shaft fractures, 3 (1%) had rickets/vitamin D deficiency, one of which was diagnosed 97 days after the fracture; none of these children had a maltreatment diagnosis. Among the infants with later femur shaft fractures, 27 (9%) had a maltreatment diagnosis or entry in the RCWM. Among children < 6 months, 20% (n = 21) of cases were associated with abuse; other diagnoses among those were subdural hemorrhage (n = 2), retinal hemorrhage (n = 1), rib fractures (n = 4), and skull fractures (n = 2). A fall was registered as the cause of injury in 69% of the later femur shaft fractures; among specified fall accidents, a fall from being carried was the most common external cause (Table 4).

Table 4
Reported accidents causing later shaft fractures of femur or humerus.

| | No reported accident | Reported accident | | | | | | | | | |
|-------------------------------|----------------------|-------------------|------------|--------------------|-----------------|-----------------------------|----------|-------------|-------------|------------------------|-----------|
| | | Fall accident | | | | | | | | Other type of accident | |
| | | All | All | From being carried | From same level | From furniture or equipment | From bed | From stairs | Unspecified | Pinching | Transport |
| | | n (%) | n (%) | n (%) | n (%) | n (%) | n (%) | n (%) | n (%) | n (%) | n (%) |
| <i>Femur shaft fracture</i> | | | | | | | | | | | |
| < 6 months (n = 102) | 37 (36.3) | 65 (63.7) | 53 (51.9) | 16 (15.7) | 5 (4.9) | 4 (3.9) | 3 (2.9) | 0 | 25 (24.5) | 10 (9.8) | 2 (2.0) |
| 6 to 12 months (n = 185) | 30 (16.2) | 155 (83.8) | 145 (78.4) | 34 (18.4) | 24 (13.0) | 20 (10.8) | 13 (7.0) | 14 (7.6) | 40 (21.6) | 8 (4.3) | 2 (1.1) |
| All (n = 287) | 67 (23.3) | 220 (76.7) | 198 (69.0) | 50 (17.4) | 29 (10.1) | 24 (8.4) | 16 (5.6) | 14 (4.9) | 65 (22.6) | 18 (6.3) | 4 (1.4) |
| <i>Humerus shaft fracture</i> | | | | | | | | | | | |
| < 6 months (n = 96) | 48 (50.0) | 48 (49.9) | 44 (45.8) | 8 (8.3) | 1 (1.0) | 3 (3.1) | 2 (2.1) | 0 | 30 (31.3) | 3 (3.1) | 1 (1.0) |
| 6 to 12 months (n = 46) | 11 (23.9) | 35 (76.1) | 35 (76.1) | 3 (6.5) | 9 (19.6) | 7 (15.2) | 2 (4.4) | 0 | 14 (30.4) | 0 | 0 |
| All (n = 142) | 59 (41.6) | 83 (58.4) | 79 (55.5) | 11 (7.7) | 10 (7.0) | 10 (7.0) | 4 (2.8) | 0 | 44 (31.0) | 3 (2.1) | 1 (0.7) |

Out of the 188 infants with birth-related humerus shaft fractures, 1 had OI and 1 had rickets/vitamin D deficiency. None of these children had a later maltreatment diagnosis. Among children with later humerus shaft fractures, 7 (5%) had OI – one of whom had a maltreatment diagnosis before the OI diagnosis – and 1 (1%) had rickets/VDD. Out of the later humerus shaft fractures, 14 (10%) had a maltreatment diagnosis or entry in the RCWM; 12 (14%) were diagnosed before the age of 6 months and other diagnoses among those were subdural hemorrhage (n = 1), retinal hemorrhage (n = 1), rib fractures (n = 2), and skull fracture (n = 1). A fall was registered as the cause of injury in 56% of later humerus shaft fractures; a fall from same level or from furniture was the most common specified external cause (Table 4).

Discussion

To our knowledge, this is the first study addressing the epidemiology of shaft fractures of femur and humerus, studying both birth-related and later fractures during the first year of life, with account taken of association to perinatal characteristics. As far as we know, this is also

the largest population-based report on such fractures, and the first report on the incidence of humerus shaft fractures diagnosed during infancy but after birth.

The overall incidence of birth-related shaft fractures of femur and humerus in the present study was 0.024‰ and 0.101‰, respectively. These results differ substantially from previously reported hospital case studies with incidence rates of 0.13–0.14‰ [1, 2] and 0.04–0.06‰ [2, 3]. Shaw et al. [11] reported that 20% (n = 5) of humerus shaft fractures in children under the age of one year were birth-related, compared with 60% in our study. It is unlikely that previous studies have had hidden cases, while hidden cases may exist with our register design. However, both the small sample sizes of previous reports [1–3, 11] and the differing settings might have contributed to the discrepancies compared with our population-based findings. Another reason could be our definition of birth-related fractures, which also included fractures identified in the early neonatal period without fall accidents, but not coded as birth-related. The existence of such cases indicates a possible underestimation of shaft fractures that might be associated with birth.

One of the issues emerging from this study is the challenges related to epidemiologic surveillance of birth-related femur and humerus shaft fractures, especially birth-related long bone fractures. The current P codes of birth injury to the skeleton in ICD-10 encompass the categories femur fractures and other long bone fractures. The ICD-11 released in 2018 has only one category: Birth injury to long bones (KA45.6). Our findings support the use of double coding, i.e., adding S codes, for improved precision of shaft diagnoses. Diagnoses of birth-related fractures might be delayed for femur fractures, [1, 14] especially among the preterm-born, [15] and for humerus shaft fractures [3, 8, 11]. We have previously reported that some birth-related fractures of femur or other long bones are diagnosed after the first week of life and in some cases at even higher age [6]. Our findings show an overlap between birth-related femur fractures and other long bone fractures. Whether these double diagnoses are due to coding errors, the identification of a cause, or delayed diagnosis cannot be ascertained, due to the study design with no access to patient records. In a clinical perspective, assessment of whether a fracture is birth-related or not has less importance for treatment; however, if the diagnosis is delayed and abuse is a potential cause, this can be of greater importance [8, 16].

The incidence of femur shaft fracture not related to birth found in this study (0.154‰) was in line with previous findings in Sweden [4]. This is higher than what was reported from the UK by Talbot et al.; however, the authors stated that there might be an underestimation of the incidence due to missed cases in their dataset obtained from the Trauma and Audit Research Network and exclusion of children with open fractures and associated injuries [5]. In these previous studies, the risk of a femur shaft fracture was reported as higher among boys than among girls after the age of one year, but neither this study nor these past studies found any statistically significant sex differences during infancy [4, 5]. We cannot explain why boys in our cohort had a higher risk of birth-related diaphyseal humerus fractures but not of femur shaft fractures. Sex is not always reported but – interestingly – birth-related humerus shaft fractures previously reported by Basha et al. [2] and Sherr-Lurie et al. [3] were all in boys.

The current study found an incidence of humerus shaft fracture later during infancy of 0.073‰. A previous hospital study reported that among 101 children younger than 12 months treated at an emergency clinic, 2 patients were diagnosed with a humerus shaft fracture [17]. Another study reported an incidence of 0.17‰ (n = 3) in children aged < 24 months [18].

Our cases shared perinatal risk factors confirming the state of knowledge. For femur shaft fracture, these factors were breech presentation [1, 2, 6], multiple birth, and prematurity [1, 6], and for humerus shaft fractures, they were vacuum-assisted delivery, shoulder dystocia, and heavy birth weight, as reported for other birth-related long bone fractures [6]. A high proportion of cesarean delivery in connection with shaft fractures [1–3] was not evident from our findings. Among those with a femur shaft fracture during the first to fifth month, we found a preponderance of the perinatal risk factors breech, prematurity, and SGA, and among those with humerus shaft fracture, shoulder dystocia, and heavy birth weight. Few perinatal risk factors were seen among those with femur or humerus shaft fractures diagnosed from the sixth month of life. For the early femur shaft fractures, this might be interpreted as a sign of bone fragility; however, this cannot be ascertained, due to the study design. We have previously shown that long bone fractures, not specifically shaft fractures, among infants of less than 6 months of age, especially those without accidents reported, are associated with risk factors for a metabolic bone disease, such as being born preterm [10]. Case reports have described rickets as being associated with humerus shaft fracture [19, 20] and femur shaft fractures [20]. Case series have reported long bone shaft fractures associated with metabolic bone disease of infancy [21]. We found only few shaft fractures with a diagnosis of rickets or vitamin D deficiency, as previously reported for fractures of any type [22].

Femur and humerus shaft fractures are closely associated with abuse diagnosis among infants. Our proportion of femur shaft fractures attributed to abuse, 9%, is higher than in the Swedish nationwide study, which reported a figure of 4.2% [4] probably because we included those having an entry in the RCWM. These numbers can be compared with those in previous studies reporting that 21–31% of femur fractures (including fractures of the entire femur) in children under the age of 1 year were caused by child abuse [12, 23]. Shaft fractures of the humerus are also common in connection with abuse [24]. Our outcome of 10% of shaft humerus fractures having been assessed as

abuse is lower than in a U.S. hospital study and a UK multicenter hospital study, which both reported that one third of humerus shaft fracture in children under the age of one year were associated with probable or indeterminate abuse [11, 12]. Whether this discrepancy is due to different criteria for determination of abuse or a true difference cannot be ascertained.

In the absence of other abuse-suspected findings, e.g., bruises, the conclusion that a femoral fracture or humerus fracture in an infant has arisen as a result of abuse cannot be based solely on exclusion of accidental trauma. The importance of making a thorough risk factor analysis, including bone fragility disorders, is underlined by the fact that routinely used radiological methods have low sensitivity for detecting low bone mineralization [25]. Tests for metabolic bone disease such as measuring levels of calcium, phosphorus, alkaline phosphatase (ALP), parathyroid hormone (PTH), vitamin D, copper, and ceruloplasmin should be done routinely [26], and testing for genetic disease, e.g., osteogenesis imperfecta, should be considered.

Eight out of ten children with a femur shaft fracture occurring later in infancy had a reported accident; the vast majority had a reported fall accident as described earlier [4, 5], usually occurring in children aged 6–12 months. Our finding of 58% of children with a humerus shaft fracture having an accident reported is higher than the 29% reported by a hospital study (7/24) [11]. However, much like in the study on femur shaft fractures by Talbot et al. [5], 29% of the children in our study had no reported accident.

Strengths and limitations

A major strength of this study was the population design derived from diagnoses in a national patient registry considered to have high validity [27]. Although shaft fractures in infants have not been specifically validated in the patient registry, this fracture type can be considered to have high precision, although there is still the potential for misclassification bias or missing data. A strength was also the linkage to the RCWM to detect cases associated with abuse; we found cases that had no diagnosis of maltreatment, but had an entry in the out-of-home care register. A further strength was the linkage to a birth registry [28]. The rate of birth fractures was estimated by combining femur and humerus shaft fractures diagnosed in days 1–7 and fractures diagnosed as birth-related, in the absence of accidents in the first week of life. The major weakness of the study was that we did not have information from patient records regarding treatment and cause of injury. This also limited our analysis of the clinical circumstances of shaft fractures registered as both birth-related (*P coded*) and other during infancy (*S coded*), and of shaft fractures in the early neonatal period not diagnosed as birth-related. Furthermore, the extent of testing for biochemical markers or genetic disease could not be determined, meaning that there might have been cases with an abuse diagnosis disorder in which proper differential diagnostics had not been undertaken.

Conclusion

This study is the largest population-based report and provides data on epidemiology, birth, parental characteristics, and reported accidents in relation to femur and humerus shaft fractures during infancy and may serve as a basis for further studies. The present study identified reported accidents as the main contributor to femur and humerus shaft fractures during infancy, and that bone fragility factors and birth trauma might be of importance not only for fractures diagnosed as birth-related. The incidence of birth-related fractures differs substantially from previously reported hospital case studies. We recommend pediatricians and pediatric orthopedic surgeons to use double coding, i.e., adding S codes to P codes, for improved precision of the registration of birth-related fractures.

List Of Abbreviations

National Patient Register (NPR)

The Swedish Medical Birth Register (SMBR)

The Register of Children and Young Persons Subjected to Child Welfare Measures (RCWM) Swedish version of International Classification of Diseases (ICD-10-SE)

Small-for-gestational age (SGA)

Large-for-gestational age (LGA)

Body mass index (BMI)

Osteogenesis imperfecta (OI).

Alkaline phosphatase (ALP)

Parathyroid hormone (PTH)

Declarations

Ethics approval and consent to participate

The study was approved by the Regional Ethical Committee in Uppsala, Sweden (2014-11-19, No. 383).

Consent to publish

Not applicable

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors have no financial relationship relevant to this article to disclose. The authors have no conflicts of interest to disclose.

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Authors' contributions

JvH: Study design, interpretation of data, and manuscript preparation

IT: Study design, interpretation of data, and manuscript preparation

UH: Study design, data collection, statistical analysis, interpretation of data, and manuscript preparation

All authors approved the final version of the manuscript

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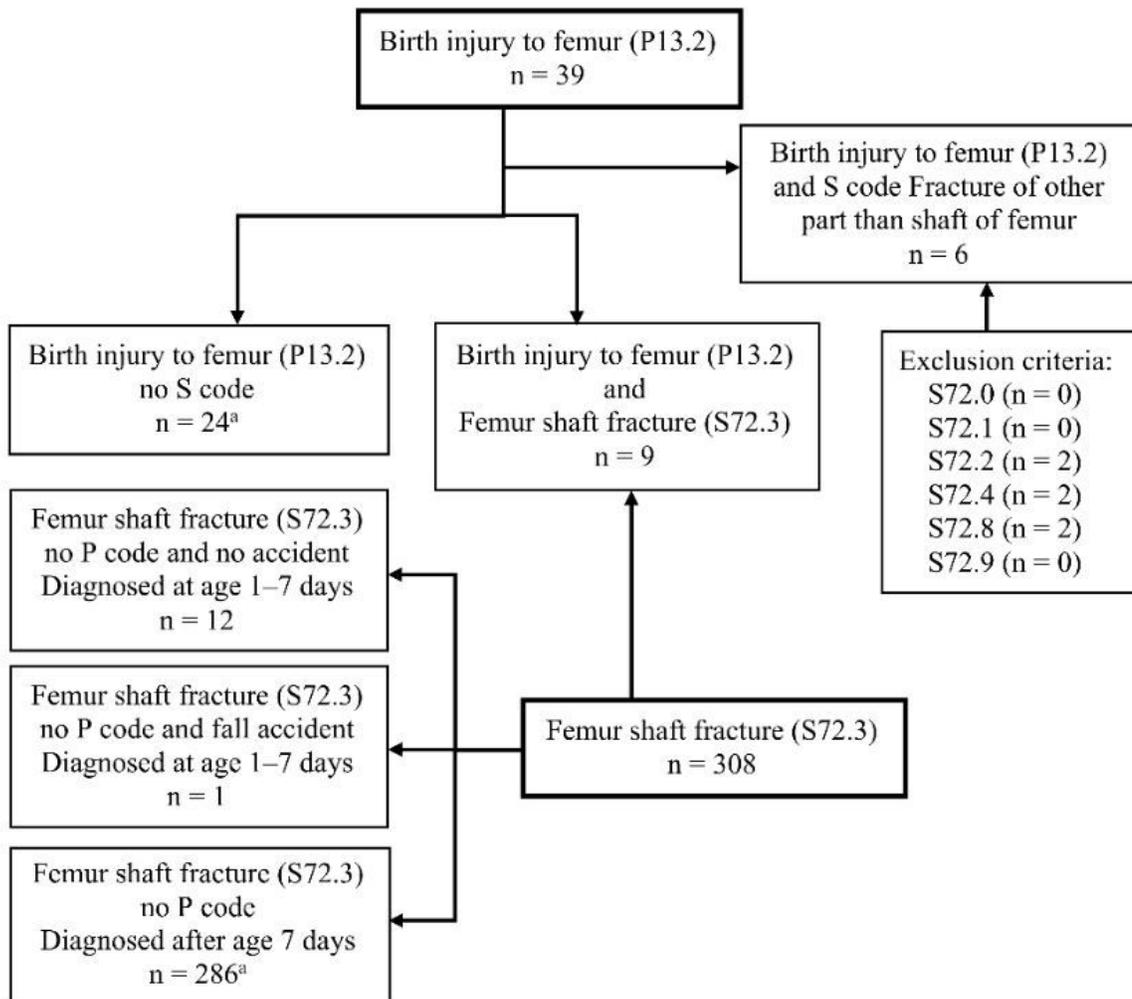
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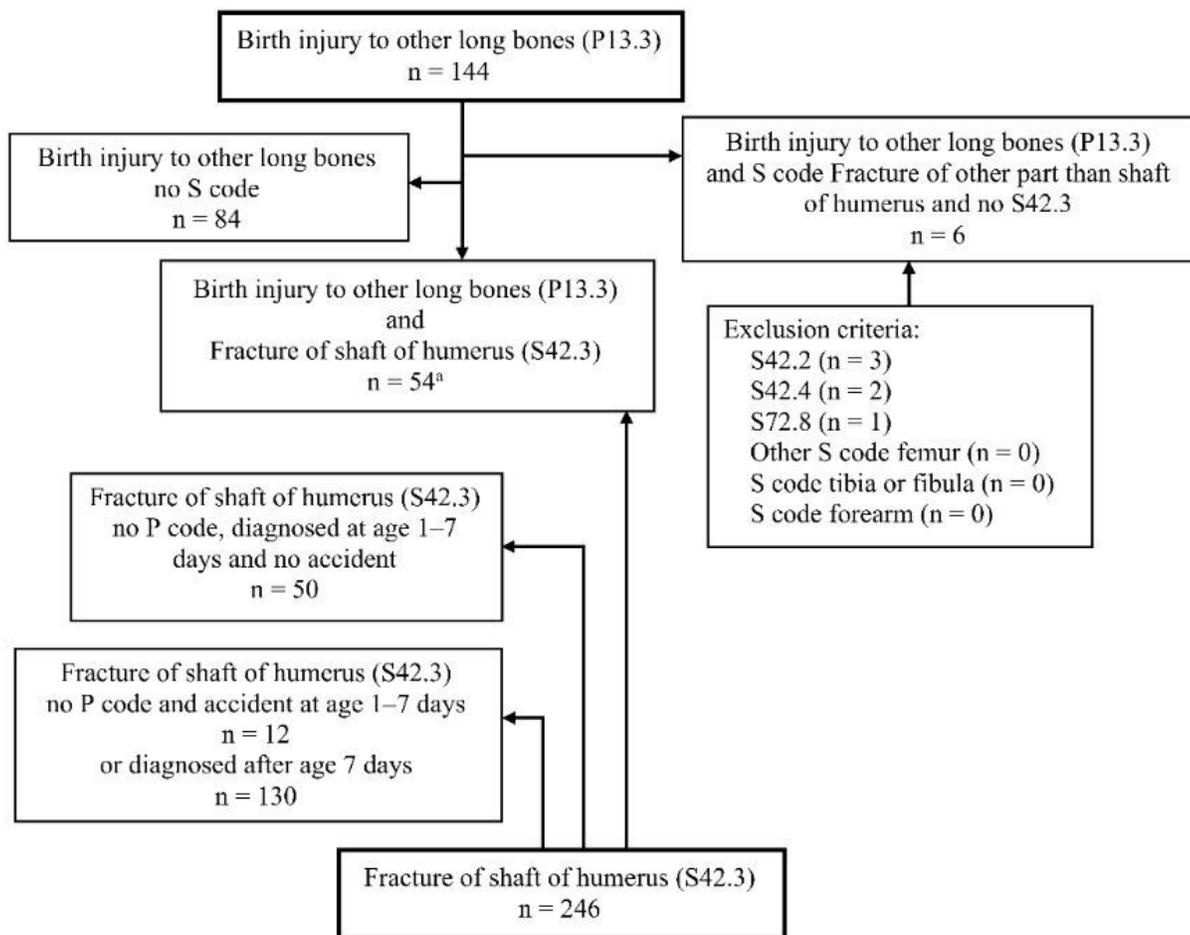
Figures



^aOne infant had a femur fracture on both day 1 and day 349.

Figure 1

Study eligibility flowchart of children under the age of 1 year with femur fractures, identified in the National Patient Register.



^aAlso included: humerus shaft fracture and fracture of humerus lower end (S42.2 +S42.3) (n = 3); humerus shaft fracture and fractures of upper and lower end of humerus (n = 1).

Figure 2

Study eligibility flowchart of children under the age of 1 year with humerus fractures, identified in the National Patient Register

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