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ORIGINAL ARTICLE • HIP - PAEDIATRIC

A familial case series of valgus slipped capital femoral epiphysis

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Abstract
We present a familial case of valgus slipped capital femoral epiphysis (SCFE). Charts of members of the same family having the condition were retrieved. Clinical and radiological examinations were conducted after 10 years of the initial presentation and treatment. Two siblings, brother and sister, were initially diagnosed with valgus SCFE and treated surgically. Normal clinical examination was found for both siblings and both, treated and untreated hips. A radiological examination for the parents revealed signs of valgus SCFE in both hips of the mother. No association with metabolic, hormonal or neurological conditions was found. The review of the literature demonstrated that varus SCFE has a strong familial tendency. Our case series would suggest that, as in the classical SCFE, genetic inheritance could also be a contributing factor to valgus SCFE. Future radiological studies are needed to look for the true incidence of valgus SCFE in first- and second-degree relatives.

Keywords Slipped capital femoral epiphysis · Caput valga · Genetic inheritance

Introduction

First described by Muller et al., valgus slipped capital femoral epiphysis (SCFE) is considered a rare entity [1]. In 2006, a review performed by Loder et al. estimated the prevalence of epiphyseal coxa valga to be 1.9% among children presenting with idiopathic SCFE. Occurring mostly in girls (76% of cases) [2], the mean age at presentation was reported to be 2.3 years less than that of classical SCFE [3]. Patients with valgus SCFE were found to have shorter and less severe symptoms duration in comparison with varus SCFE [4]. A number of etiologies and risk factors have been postulated regarding the etiology of varus SCFE such as obesity, growth hormone deficiency, hypothyroidism and collagen disease within the physis, but none of such associations linked to valgus SCFE has been established yet [5, 6]. Valgus SCFE is defined as lateral displacement of the femoral head epiphysis in its hypertrophic zone [7], which is thought to be due to femoral neck retroversion [8]. Contrary to the classical SCFE, the Klein line in valgus SCFE usually intersects with the epiphyseal femoral line [9].

Demographics of some populations have been linked to varus SCFE; in black [10], white [11], Japanese [12] and Amish ethnicities [2]. Moreover, many familial reports, including first and second relatives, of classical varus SCFE have been published in the literature with a familial inheritance suggested to be following an autosomal dominant pattern with variable penetrance [13–21].

To our knowledge, no familial cases of valgus SCFE have been reported in the literature. In this study, we present the clinical and radiological findings of valgus SCFE encountered in members of the same family.

Patients and methods

We retrospectively reviewed the charts of three patients, members of the same family, all presenting with valgus SCFE. The clinical presentation, co-morbidities, radiological evaluation and treatment modalities were recorded. After approval by the institutional review board, written informed consents were obtained from both parents prior to the study.

Between 2007 and 2017, three members out a family of four were diagnosed with valgus SCFE by radiological examination; two siblings (1 boy and 1 girl) and their mother. The father did not present any clinical or radiological sign of

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this condition. Both children were symptomatic and had a surgical management, whereas the mother did not recall any past symptom related to the affected hip. The siblings were followed-up both clinically and radiographically for periods of 11 and 10 years, respectively, with X-rays at 1 month, 1 year, and at 10th/11th year. The mother was followed-up for 8 years with X-rays at the first consultation and at the 8th year. Only the last clinical and radiological evaluations were prospective.

Two independent orthopedic surgeons reviewed radiological studies performed at 1 month, 1 year and at final follow-up. The following radiological angles were calculated: the anteroposterior (AP) neck-shaft angle (NSA), the AP neck-physis angle (NPA), the AP physis-shaft angle (PSA), the AP physeal tilt (PT) and the posterior physeal tilt angle (PPT) on the frog lateral view. Radiological measurements were performed according to Shank et al.'s method [4].

Results

Clinical presentation

At the time of diagnosis, the boy was 16 years old complaining of right hip pain and locking that was increasing during 2 months prior to presentation. Pain was associated with limping and a sensation of giving way.

The girl was diagnosed 2 years after her brother; she was 15 years of age, did not complain of any locking or pain. The only complaint was a limp that was noticed by her parents for almost 2 weeks duration.

No leg length discrepancy was noted in both siblings.

The mother did not recall any symptoms throughout her life; no locking or limping was noticed. No co-morbidities were noted in any family member.

The demographic characteristics of the affected family members are summarized in Table 1. The mother or the father had no other children.

Radiological findings

Follow-up X-rays at 1 month, 1-year and 10-year postoperative intervals were retrospectively analyzed.

A summary of the radiographic measurements of the proximal femoral geometry is presented in Table 2. Figures 1, 2, 3 and 4 show the preoperative imaging of the girl and Fig. 5 shows the last postoperative X-rays of the girl. Figure 6 shows the postoperative X-rays of the boy. (The preoperative X-rays were lost.) The X-rays of the mother did not show any signs of osteoarthritis (Fig. 7).

Treatment modalities

Two symptomatic cases underwent surgical intervention. No prophylactic fixation of the contralateral side was performed in any of the siblings. The boy was treated with in situ percutaneous fixation using two cannulated screws.

Table 1 Demographic characteristics of the patients

Family members	Age (years)	Height (cm)	Weight (Kg)	BMI	Clinical symptoms	Side	Symptom duration	Fixation type	Follow-up period (years)
Mother	56	162	80	30.5	No symptoms	Left	Asymptomatic	None	–
Son	16	193	85	22.9	Pain, locking, limping,	Right	8 weeks	In situ fixation using 2x cannulated screws	12
Daughter	15	165	48	17.8	Limping	Right	2 weeks	In situ fixation using 1x cannulated screws	10

Table 2 Radiological signs of the patients at first presentation

Family member	Neck-shaft angle	Physis-shaft angle	Neck-physis angle	Lateral physeal tilt angle	Posterior physeal tilt angle
Mother	150°	90°	31°	12°	16°
Daughter	148°	104°	44°	20°	15°
Son	154°	90°	28°	15°	15°
Mean values	150.7 ± 2.5°	94.6 ± 6.6°	34.3 ± 6.9°	15.6 ± 3.3°	15.3 ± 0.47°

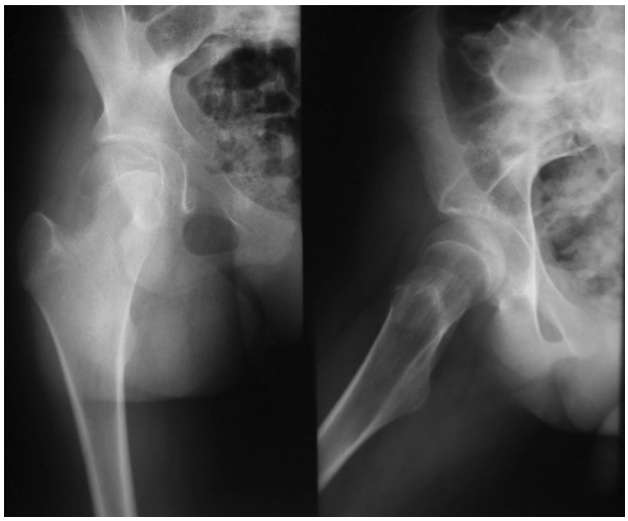

Fig. 1 Preoperative X-rays of the girl

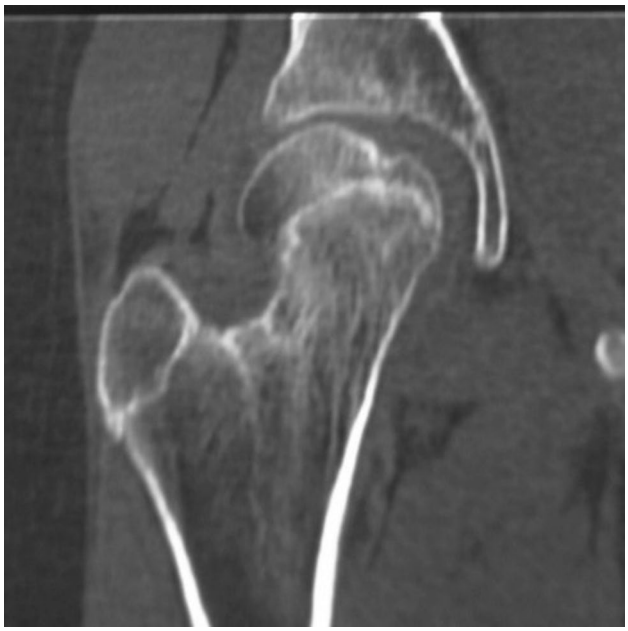

Fig. 2 CT scan of the girl

The daughter was treated with in situ percutaneous fixation using only 1 cannulated screw for fixation. No intervention was performed for the mother. No complications were noted both intra-operatively and postoperatively, and both patients were allowed full weight bearing immediately postoperatively.

Last clinical and radiological evaluation

Both siblings presented no symptoms at the last follow-up. The clinical examination of the affected hip demonstrated pain-free joints with a full range of motion, as was the case with the contralateral hip. Patients had no difficulties in daily and exercise activities. All measured angle values were similar at the last radiological assessment.

Discussion

The present study reports a familial case of valgus SCFE. Our literature search could not find a previously published paper on this topic. The condition is considered to be very rare. The results of Loder et al. [2] review concluded that valgus slip would represent 1–2% of all idiopathic SCFE. More recently, this prevalence value has been estimated at the prevalence of 4.7% in a series of 258 SCFE [4]. On the other hand, some authors expressed some skepticism over the existence of such entity; Griffith related the apparent lateral position of the epiphysis to a rotational malposition of the hip during an anteroposterior radiograph [22]. This argument has been overruled by three-dimensional computerized tomograms [8, 23].

While the exact cause of epiphyseal valga remains unknown, many risk factors are thought to be associated such as metabolic, hormonal and neurological disorders. However, this association has been found to be inconsistent. In the study of Shank et al., only four out of 12 patients were reported to have a hormonal disorder such as panhypopituitarism, hypothyroidism and growth hormone deficiency [4]. In our case series, none of the patients had a co-existing disease or condition. It is worthy to note that our boy had a height of 193 cm at presentation with a weight of 85 kg (at 16 years of age), but there were no clinical signs of Marfan or hypermobility syndromes. Both children were chronologically old for a slip; the age at the presentation was found to be higher than the reported mean age of 11 years [3]. Bone age determination to estimate skeletal maturity has not been performed. Interestingly, Chung et al. [24] reported in a very recent paper a high frequency of valgus SCFE (19 out of 22; 86.3%) in children with neoplasm having clinical and radiological signs of a slipped epiphysis.

Rennie has shown that varus SCFE has a strong familial tendency; whereas the individual incidence was only 0.05% in the general population, 7% of his patients had a close relative with an SCFE on radiographic evaluation [13]. Hägglund et al. [20] reported that almost one-third of 49 families with a history of one member having varus SCFE had the same condition in one or more first-degree relatives on radiographic examination. In addition, these authors found cases of varus SCFE in four families with second-degree relatives and that in ten families there was a second-degree relative with radiographically verified coxarthrosis. Those publications demonstrated that the familial accumulation was much higher than expected from incidence studies, indicating a

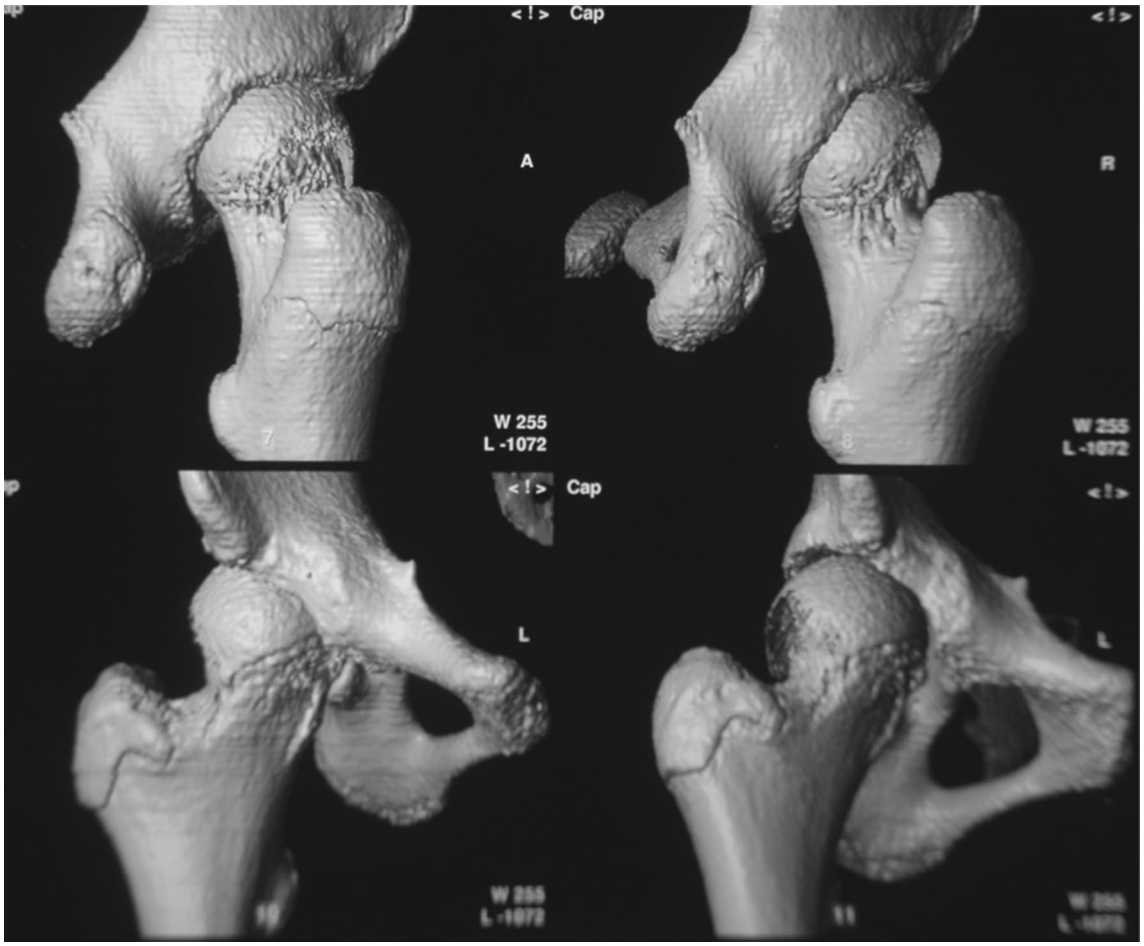

Fig. 3 3D scan of the girl

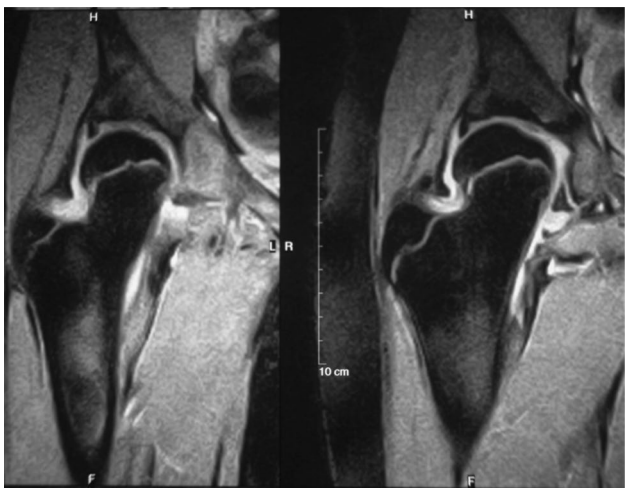

Fig. 4 MRI of the girl

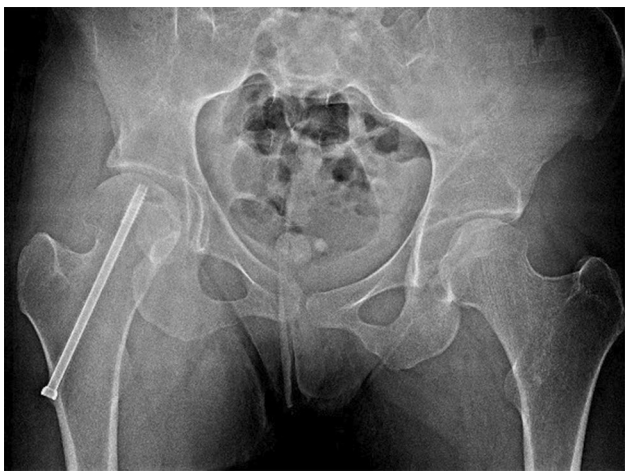

Fig. 5 Last postoperative X-rays of the girl

hereditary factor in the etiology. Our results suggest that the valgus type of SCFE could have an X-linked dominant inheritance since both children had the condition as their mother,

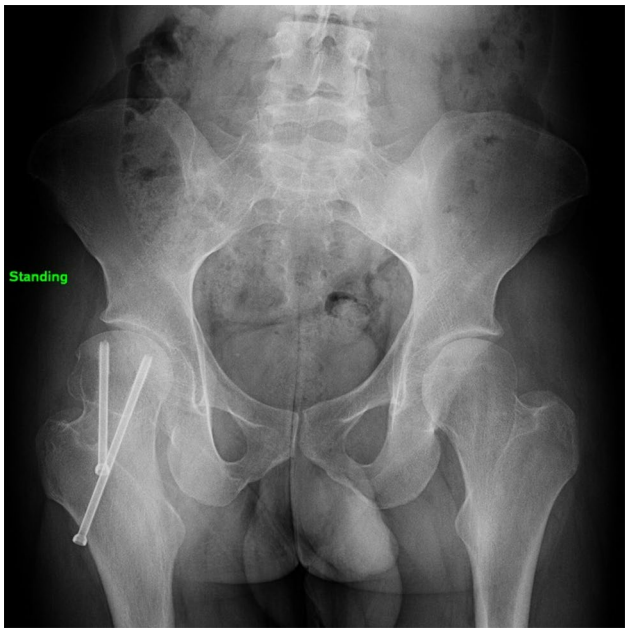

Fig. 6 Last postoperative X-rays of the boy

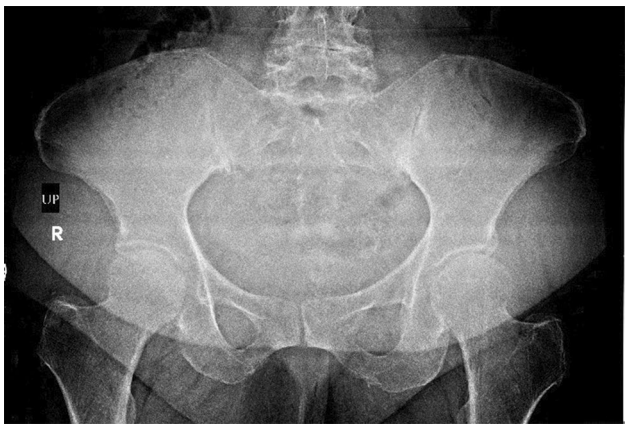

Fig. 7 X-rays of the mother

while the father did not. More studies including radiological assessment are needed in the future to look for the true incidence of valgus SCFE in first- and second-degree relatives and to comprehend its genetic inheritance.

When comparing our results with those reporting the normal values of the proximal femoral angular measurements, all our mean values were found to be superior to the mean values of normal hips as stated by Yngve et al. [25]. Only Shea et al. [26] reported a medial translation tilt of the femoral epiphysis with the similar value of normal hips. Additionally and besides the recent study of Kalhor et al. [27], many of our mean values were either close to the highest range or higher when compared to those reported in other studies investigating valgus SCFE. Most studies reported a lateral physeal tilt angle such as that observed in our study. However, our mean value of 15.6° was higher than that found with other authors who reported ranges between 6.7° [3] and 11° [26]. On the other hand, our mean posterior physeal tilt angle of 15.6° was found to be higher than the normal value of 12° [25] but lower than those reported in the literature, 22° [26] to 29.8° [3].

Despite the limitations of being a single case series with retrospective incomplete data collection, this paper could contribute to a better understanding of previously unknown risk factors such as the genetic factor in the development of valgus SCFE.

In sum, our study notes that genetic inheritance could be a contributing factor to valgus SCFE. The radiological findings could be of a major help in diagnosing this condition within families and relatives. Future clinical and radiological researches including first- and second-degree relatives are warranted.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval Ethical approval was obtained from the institutional board.

Informed consent Informed consent was obtained from all individual participants included in the study.

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