Title page

Title: Patellofemoral joint alignment is a major risk factor for recurrent patellar dislocation in children and adolescents: a systematic review

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What is already known:

- Several structural and functional factors have been identified as possible risk factors for recurrent patellar dislocation in children and adolescents.

What are the new findings:

- There is a paucity of reported radiological parameters for healthy children and adolescents;
- Numerous studies report measurements of patellofemoral joint malalignment, making it a primary risk factor for recurrent patellar dislocation in this patient population;
- High TT-TG distance and high bony sulcus angle can confidently predict the risk of recurrent patellar dislocation in children and adolescents;

INTRODUCTION

Patellar dislocation is relatively common in the pediatric and adolescent populations, with an estimated annual incidence of 43 per 100,000 individuals in children[1] and a prevalence of six to 77 per 100,000[2-5] in adolescents. Furthermore, there is a significantly higher re-dislocation rate following an acute first-time dislocation in children and adolescents compared to adults[6, 7], and in patients aged less than 15 compared to patients aged 15-18[8]. Consequently, children and adolescents are likely to experience recurrent instability, at a rate ranging from 38.4% to 91%[9, 10].

Patellar dislocation is believed to be the result of an abnormal interplay between lower limb alignment, bony geometry of the trochlea and patella, passive restraints of ligaments and retinaculum and the action of the quadriceps. Medical imaging studies have identified anatomical parameters that may predispose adult individuals to patellofemoral joint (PFJ) instability[11, 12]. Other studies have attempted to describe the complex interaction between

bony and soft tissue (active and passive) restraints of the PFJ [4, 13-15] and their relative effect on PFJ stability throughout the tibiofemoral joint range of motion[16]. Nevertheless, the interplay of risk factors that predispose individuals to recurrent patellar dislocation remains poorly understood, because the parameters have been reported either in isolation or in sparse combination and in varying magnitudes.

Challenges associated with managing recurrent patella dislocations in the skeletally immature population include a paucity of normative and pathoanatomical data that contribute to PFJ instability. Surgical techniques that do not violate the physis have not been adequately assessed in terms of their specific indications, given the variability of factors that contribute to dislocation in this patient population.

The primary aim of this study was to summarize the current literature to characterize the lower limb alignment, patellofemoral morphology and soft tissue restraints of the PFJ through medical imaging measurements in children and adolescents with and without recurrent patellar dislocation. Our secondary aims were to synthesize the data to stratify the factors that influence PFJ stability and provide recommendations on the assessment and reporting of PFJ parameters in this patient population.

METHODS

Search strategy

To identify relevant papers on this topic, a systematic search was performed of the following computerized databases: CINAHL (2010-June 2017), the Cochrane Library (1979-June 2017), EMBASE (1955-June 2017), PubMed (1963-June 2017) and Web of Science (1955-June

2017). The adopted search strategy included MeSH terms for ‘knee joint’ AND ‘dislocation’ AND ‘(‘child’ OR ‘adolescent’)’. It was ensured that patellar dislocation was captured by use of the search terms ‘knee joint’ and ‘dislocation’. References from relevant papers were also screened to guarantee the inclusion of all key studies.

Inclusion and exclusion criteria

Studies which included typically developing children (1-12 years old) and adolescents (13-18 years old) who experienced recurrent patellar dislocation and also had direct measures of structural and functional factors were investigated (Population: children and adolescents; Intervention/Cause: structural and functional factors; Comparison: pre/post dislocation; Outcome: patellar dislocation). Studies which examined patients suffering from congenital or first-time acute traumatic patellar dislocation were excluded.

Study selection, methodological quality and measurement quality

After removing duplicates, the titles and abstracts of the papers obtained from the initial search were independently assessed by two authors (MB and SM). When the title and abstract were not sufficient the full text was screened. Any disagreement between the two reviewers was discussed until a consensus was reached. Full text papers were then retrieved and independently read again by the two authors (MB and SM). Conference abstracts were excluded. The methodological quality of the included studies was independently evaluated by two reviewers (MB and CC) based on a customized version of the Downs and Black checklist[17]. The 27-item checklist evaluates the quality of reporting, external validity, bias, confounding and power. Items 4, 8, 9, 13, 14, 19, 23, 24 and 26 were excluded due to inapplicability to the studies investigated. Each item is scored 0 or 1 (‘no’ or ‘yes’) except for item 5 which is scored from 0 to 2 (‘no’, ‘partially’ or ‘yes’). The scoring for item 27, dealing with statistical power,

was simplified from a 0 to 5 scale to a ‘yes’ or ‘no’ choice depending on whether the study had sufficient power to detect a clinically important effect[18, 19].

Data extraction

Data extracted included population demographics and measures of evaluated risk factors. Risk factors were grouped into four categories:

- *Lower limb alignment*, which included any mention of coronal or axial plane alignment;
- *PFJ alignment*, which included any measure of the relationship between the patella, femur or tibia;
- *Trochlea morphology*, which included any measure or angle contained within the trochlea itself;
- *Soft tissue restraints*, which included any mention of soft tissue influence or integrity.

The specific method of measurement for quantitative risk factors was documented, along with mean and standard deviation being derived, if not provided, as recommended by the Cochrane guidelines[20]. For measures that have been reported in more than one study, weighted averages and standard deviations were computed for patients and, when possible, for control participants. Meta-analysis was conducted in Review Manager (RevMan), version 5.2 for Windows, (The Nordic Cochrane Centre, The Cochrane Collaboration, Copenhagen, Denmark) on measures reported in two or more studies that included both a control and a patellar dislocator group. Weighted mean differences and 95% confidence intervals (CI) were calculated for each measure with a fixed effects model.

RESULTS

Search results

The initial search yielded 712 potentially relevant articles, with an additional six articles included that were references from key papers. After removing duplicates and screening of titles and abstract, 69 full-text articles were fully screened for eligibility. Of these, 49 were excluded predominantly due to the lack of direct measures and to the exclusion of recurrent patella dislocators from the patient population (Figure 1).

Figure 1. Systematic search strategy results.

Study participants

Of the 20 included studies, six included a control population, while three split the patients' group into subcategories based on undergoing treatment[9, 21, 22] (Table 1). Eight studies[21-28] only included patients suffering from recurrent patellar dislocation, while the other studies included a combination of single acute and recurrent dislocation[9, 29-31] or habitual and recurrent dislocation[32, 33]. Two[24, 28] of the 20 studies were case studies. Mean age of the participants ranged from 11.1 to 17.3 years.

Table 1. Population characteristics

First author (date)	Participants						
	Category	Number	Sex	Dislocation	Age		
		(n)	(m/f)	(A/H/R)	Mean	SD	Range
Aulisa (2012) ²³	P	14	4/10	0/0/14	11.1	-	9.2-13.1
Deie (2003) ³²	P	4	2/2	0/3/1	15.5	-	15-16
Dickens (2014) ³⁶	P	76	28/48	-	12.6	-	0-15
	C	495	220/275		12.6	-	0-15
Edmonds (2015) ²¹	P	20 (10 IR, 10 MPFLR)	-	0/0/20	15.6	2.2	-
	C	10	-		15.8	1.5	-
Horikawa (2011) ²⁴	P	1	0/1	0/0/1	15	-	-
Jaquith (2015) ²⁹	P	250	119/147 ^a	266 ^a /0/77 ^b	13.7	2.3	8-18
Ji (2012) ²⁵	P	17 ^c	-	0/0/17 ^d	15	-	12-18
Kan (2009) ²⁶	P	4	0/4	0/0/4	17.3	1.2	-
	C	4	0/4		14.5	1	-
Kumahashi (2012) ³³	P	5	2/3	0/1/4	13.6	-	11-15
Nelitz (2013) ²⁷	P	21	15/6	0/0/21	12.2	-	10.3-13.9
Nietosvaara (1997) ³⁴	P	33	11/22	-	15.6	-	12-17
	C	25	11/14		14.8	-	12-18
Palmu (2009) ⁹	P	62	16/46	64 ^a /0/44 ^b	13	2	-

			(27 CG, 35 SG)				
Pennock (2014) ³⁷	P	45	23/22	-	15.4	2	-
	C	180	104/76		16	2	-
Putney (2012) ³⁵	P	63	33/30	-	13.6	-	15-17
Regalado (2014) ³⁸	P	29	-	-	13	-	11-16
	C	10	-		13	-	11-16
Schoettle (2005) ²⁸	P	1	0/1	0/0/1	15	-	-
Seeley (2012) ³⁰	P	111	56/45	111/0/34 ^e	14.9	-	11-18
Vahasarja (1995) ³⁹	P	48	5/43	-	13.4	-	7.5-16
Yeoh (2016) ³¹	P	43	20/23	43/0/13 ^e	-	-	10-17
Zhao (2012) ²²	P	54	9/45	0/0/54	14.7	1.3	-
			(28 MRP, 26 VMP)		MRP	MRP	-
					15.2	1.6	
					VMP	VMP	

- = not reported; P = patients; C = controls; A = acute; H = habitual (i.e. involuntary patellar dislocation and relocation with every cycle of knee flexion and extension [71]); R = recurrent; IR = patients undergoing Insall realignment [72]; MPFLR = patients undergoing medio patellofemoral ligament reconstruction; ^a knees; ^b knees with evidence of acute dislocation which experienced also recurrent dislocation; ^c one patient lost to follow-up; ^d in the paper, the term “recurrent” have been interchanged with “habitual”, however, “recurrent” have been chosen according to Batra et al. [73]; ^e patients with acute dislocation who developed recurrent dislocation; CG = conservative group; SG = surgical group; MRP = patients undergoing medial retinaculum plasty; VMP = patients undergoing vastus medialis plasty.

Quality analysis

Methodologic quality had modified Downs and Black checklist scores ranging from 4 to 18, where the maximum score is 19 (Table 2). An overall score below 50% was assigned to five studies [24, 28, 32, 34, 35], two of which [24, 28] were case reports. Only 33% of the included studies [9, 23, 29, 30, 36, 37] scored above 70%. Generally, lower scores were in reference to distribution of principal confounders, sample representativeness, length of follow-up of patients, adjustment for confounding and power (20% of the included studies obtained the

127 highest score). On the other hand, the characteristics of the patients involved in the studies were
128 always clearly described and the main outcomes to be measured were accurately defined in
129 95% of the studies.

130

131 **Table 2.** Methodological quality assessment of included studies: Downs and Black checklist

Study	Downs and Black checklist [item/maximum score]																		
	1/1	2/1	3/1	5/2	6/1	7/1	10/1	11/1	12/1	15/1	16/1	17/1	18/1	20/1	21/1	22/1	25/1	27/1	Total/19
Aulisa et al. [23]	1	1	1	1	1	1	1	1	1	0	1	0	1	1	1	1	0.5	0	14.5
Deie et al. [32]	0	1	1	0	1	1	0	0	0	0	1	0	0	1	0	0	0	0	6
Dickens et al. [36]	1	1	1	0.5	1	1	1	1	0	1	0.5	0	1	1	1	1	0	1	15
Edmonds et al. [21]	1	1	1	1	1	0	0	1	0	1	1	0	1	0	1	1	0	0	11
Horikawa et al. [24]	1	0	1	0	0	0	0	0	0	0	1	0	0	0	0.5	0.5	0	0	4
Jaquith et al. [29]	1	1	1	1	1	0	1	1	0	1	1	0	1	1	1	1	0	1	14
Ji et al. [25]	1	1	1	1	1	0	0	1	0	1	1	0	1	1	1	1	0	0	12
Kan et al. [26]	1	1	1	1	1	1	1	0	0	0	1	0	1	1	0	0	0	0	10
Kumahashi et al. [33]	1	1	1	0.5	1	1	1	0	0	0	1	0.5	1	1	0.5	1	0	0	11.5
Nelitz et al. [27]	1	1	1	0	1	0	1	1	1	0	1	0	1	1	1	1	0	0	12
Nietosvaara et al. [34]	0.5	1	1	0	1	1	0.5	0	0	0	1	0	1	1	0	0	0	0	8
Palmu et al. [9]	1	1	1	2	1	1	1	1	0.5	0.5	1	1	1	1	1	1	1	1	18
Pennock et al. [37]	1	1	1	2	1	1	1	1	0.5	0	1	0	1	1	1	1	1	0	15.5
Putney et al. [35]	1	1	1	0	1	0	0	1	0	0	1	0	0	1	1	1	0	0	9
Regalado et al. [38]	1	1	1	0.5	1	1	1	0.5	0	0	1	1	1	1	1	0.5	0	0	12.5
Schoettle et al. [28]	1	1	1	0	1	0	0	0	0	0	1	0	0	0	0.5	0.5	0	0	6
Seeley et al. [30]	1	1	1	2	1	1	0	1	1	1	0	1	1	1	1	1	1	0	16
Vahasarja et al. [39]	1	1	1	0	1	1	1	1	0.5	0	1	0	1	1	1	1	0.5	0	13

Yeoh et al. [31]	1	1	1	0	1	1	1	1	0	0	1	1	1	1	1	1	0	0	13
Zhao et al. [22]	1	1	1	2	1	1	0.5	1	0.5	1	1	1	1	1	1	1	1	1	18

132 The customized Downs and Black checklist items refer to hypothesis/aim/objective (1), main outcomes (2), characteristics of the patients (3), principal
133 confounders (5), main findings (6), random variability in the data (7), probability values (10), source population (11), sample representativeness (12), blind
134 outcomes (15), data dredging (16), length of follow-up (17), statistical tests (18), accuracy of the main outcomes (20), case-control population source (21),
135 recruiting period (22), adjustment for confounding (25) and power (27).

Lower limb alignment

There were only two studies[27, 28] (one being a case report)[28] that had made an attempt to document overall alignment of the lower limb (Table 3). The other had specifically aimed to exclude patients with abnormal alignment parameters from their patient population. Schoettle et al.[28] reported measures of femoral anteversion and tibial torsion, measured on computed tomography (CT) scans, for a patellar dislocator diagnosed with lower limb rotational malalignment associated with trochlea dysplasia. However, neither of the two pathological conditions was addressed during surgery, and medial patellofemoral ligament (MPFL) reconstruction, together with lateral release, were chosen instead. Measurements of femoral anteversion, mechanical axis, and tibial torsion were reported only in one study, therefore it was not possible to derive an average value for any of these measurements.

152 **Table 3.** Measures of lower limb and PFJ alignment, trochlea morphology and soft tissue restraints.

Category	Measure (unit)	First author (date)	Patient group	Mean value [SD]	Mean value in controls [SD]	Medical imaging procedure	Knee flexion angle (°)
Lower limb alignment	Femoral anteversion (°)	Schoettle (2005)	AP	28.3 [-]	-	Computed tomography	-
	Mechanical axis (°)	Nelitz (2013)	AP	6 [1.5]	-	X-ray	-
	Tibial torsion (°)	Schoettle (2005)	AP	51.0 [-]	-	Computed tomography	-
PFJ alignment	Bisect offset ratio	Regalado (2014)	AP	0.84 [0.20]	0.54 [0.10]	Kinematic MRI -1.5T, 8mm slice thickness, sagittal/axial views, body coil	0
				0.77 [0.17]	0.53 [0.06]		10
				0.67 [0.14]	0.53 [0.07]		20
				0.64 [0.10]	0.53 [0.05]		30
	Congruence angle (°)	Kan (2007)	AP	20 [28]	-9 [12]	MRI- 3T, T1-weighted axial images, 5mm slice thickness, body coil	
		Kumahashi (2012)	RD	7.6 [10.2]	-		45
		Zhao (2012)	MRP	23.9 [17.4] ^a	-		0
	Lateral patellofemoral angle (°)	Vahasarja (1995)	AP	0.8 [6.1]	-	X-ray, axial view	20-30
	Lateral patellar deviation (mm)	Vahasarja (1995)	AP	4.6 [6.7]	-	X-ray, axial view	20-30
	Lateral stress ratio (%)	Kumahashi (2012)	RD	40.0 [20.5]	-	X-ray- 2kg stress skyline view	45
		Deie (2003)	RD	29.0 [1.4]	-	X-ray- 2kg stress skyline view	45

Medial stress ratio (%)	Kumahashi (2012)	RD	-6.4 [14.3]	-	X-ray- 2kg stress skyline view	45
	Deie (2003)	RD	-24.8 [0.4]	-	X-ray- 2kg stress skyline view	45
Patella alta						
<i>Insall-Salvati Index</i>	Deie (2003)	RD	1.65 [0.07]	-	X-ray, sagittal view	-
	Edmonds (2016)	IR	1.27 ^b [-]	1.2 ^b [-]	MRI, T1-weighted, sagittal view, knee coil	-
		MPFLR	1.35 ^b [-]			
	Kumahashi (2012)	RD	1.2 [0.2]	-	X-ray, sagittal view	-
	Nelitz (2013)	AP	1.20 [0.08]	-	X-ray, sagittal view	-
	Palmu (2008)	CG	1.3 [0.2]	-	X-ray, sagittal view	-
		SG	1.3 [0.2]			
	Regalado (2014)	AP	1.22 [0.23]	1.29 [0.16]	Kinematic MRI- 1.5T, sagittal and axial images,	0
			1.24 [0.21]	1.24 [0.17]	8mm slice thickness, body coil	10
			1.21 [0.21]	1.22 [0.13]		20
			1.20 [0.20]	1.21 [0.13]		30
	Vahasarja (1995)	AP	1.27 ^b [0.12]	-	X-ray, sagittal view	45
	<i>Caton-Deschamps Index</i>	Jaquith (2015)	AP	1.30 [-]	X-ray, sagittal view	-
Lateral patellar tilt (°)	Ji (2012)	AP	12.2 [2.8]	-	X-ray	-
	Kumahashi (2012)	RD	15.8 [4.8]	-	X-ray, Merchant view	-
	Regalado (2014)	AP	0 [12]	15 [5]	Kinematic MRI- 1.5T, sagittal and axial images,	0
			4 [9]	17 [4]	8mm slice thickness, body coil	10
			8 [7]	18 [4]		20
			9 [6]	18 [5]		30
	Vahasarja (1995)	AP	18.8 [6.7]	-	X-ray, axial view	45
	Zhao (2012)	MRP	25.3 [15.7]	-	Computed tomography, 2mm slice thickness	0

		VMP	26.5 [17.1]			
Patella-tibia/femur-tibia distance ratio	Zhao (2012)	MRP	1.4 [0.2]	-	Computed tomography, 2mm slice thickness	0
		VMP	1.2 [0.3]			
Q angle (°)	Aulisa (2012)	AP	19.4 [1.2]	-	-	-
	Horikawa (2011)	AP	20.0 [-]	-	X-ray	-
	Vahasarja (1995)	AP	9.0 [2.6]	-	-	-
TT-TG distance (mm)	Aulisa (2012)	AP	21.5 [1.75]	-	Computed tomography	-
	Dickens (2014)	AP	12.2 [2.4]	8.6 [1.7]	MRI, 1.5-3T, T2-weighted axial images	-
	Nelitz (2013)	AP	16.0 [2.8]	-	MRI, 1.5T, axial images	-
	Pennock (2014)	AP	16.3 [5.0]	11.7 [5.0]	MRI, 1.5T, axial images	-
	Schoettle (2005)	AP	24.0 [-]	-	MRI	-
	Yeoh (2016)	RD	17.4 [4.1]	-	MRI	-
	Zhao (2012)	MRP	16.8 [5.1]	-	Computed tomography, 2mm slice thickness	-
		VMP	15.9 [4.7]			
TT-TG index	Yeoh (2016)	RD	0.41 [0.08]	-	MRI	-
<hr/>						
Trochlea morphology	Dejour classification (%)	Jaquith (2015)	27% type A, 16% types BCD			
		Nelitz (2013)	47% type D			
		Schoettle (2005)	type C			
		Seeley (2012)	23% type A, 28% type B, 11% type C, 5% type D			

Lateral trochlea inclination (°)	Seeley (2012)	RD	12.7 [6.2]	-	MRI -1.5T, axial images, 3mm slice thickness, knee coil; articular joint surface	
Sulcus angle (°) <i>bony</i>	Aulisa (2012)	AP	162.1 [-]	-	Computed tomography	-
	Deie (2003)	RD	157 [1.4]	-	X-ray	45
	Horikawa (2011)	AP	170 [-]	-	Computed tomography	-
	Kan (2007)	AP	145 [10]	133 [10]	MRI- 3T, T1-weighted axial images, 5mm slice thickness, body coil	
	Kumahashi (2012)	RD	147.9 [15.1]	-	X-ray	45
	Nietosvaara (1997)	AP	157 [7]	145 [6]	Ultrasound	90
	Palmu (2008)	CG	153 [6]	-	X-ray, axial view	20
		SG	152 [8]			
	Regalado (2014)	AP	138 [11]	137 [7]	Kinematic MRI- 1.5T, 8mm slice thickness,	0
			137 [10]	139 [10]	sagittal and axial images, body coil	10
			138 [10]	136 [12]		20
			138 [10]	132 [23]		30
	Seeley (2012)	RD	144.7 [11.8]	-	MRI -1.5T, axial images, 3mm slice thickness, knee coil	-
	Vahasarja (1995)	AP	150.4 [5.9]	-	X-ray	
<i>cartilaginous</i>	Nietosvaara (1997)	AP	164 [9]	145 [4]	Ultrasound	90
	Seeley (2012)	RD	154.3 [9.4]	-	MRI -1.5T, axial images, 3mm slice thickness, knee coil	-

	Trochlear depth (mm)						-
	<i>bony</i>	Seeley (2012)	RD	2.6 [1.4]	-	MRI -1.5T, axial images, 3mm slice thickness,	
	<i>cartilaginous</i>			4.4 [2.1]	-	knee coil	
	Trochlear facet asymmetry (%)	Seeley (2012)	RD	52.1 [14.9]	-	MRI -1.5T, axial images, 3mm slice thickness, knee coil	-
Soft tissue restraints	Generalized ligamentous laxity (%)	Aulisa (2012)		62.5 %	-		
	MPFL injury (%)	Putney (2012)		53% tear, 47% stretching	-		
		Seeley (2012)		82.3 %	-		
		Zhao (2012)		92.6% sprain/tear	-		
	Adductor tubercle – VMO distance (mm)	Seeley (2012)	RD	16.44 [4.17]	-	MRI, 1.5T, T1 and T2-weighted sagittal images, knee coil	-
	Quadriceps force vector orientation (°)	Kan (2007)	AP			MRI, 3T, T1-weighted axial images, body coil, 5mm slice thickness	15
	<i>anterior/posterior ACSA</i>			5.6 [3.4]	2.0 [1.2]		
	<i>left/right ACSA</i>			5.2 [2.8]	2.0 [1.0]		
	<i>anterior/posterior PCSA</i>			5.8 [3.4]	2.1 [1.4]		
	<i>left/right PCSA</i>			5.3 [2.6]	2.0 [1.0]		
	PFJ reaction force/quadriceps force	Edmonds (2016)	IR MPFLR	0.94 [-] 0.92 [-]	0.76 [-]	X-ray, sagittal view; MRI- T1-weighted, sagittal view, knee coil	-

Patella	Edmonds (2016)	IR	1.21 [-]	1.18 [-]	X-ray, sagittal view; MRI- T1-weighted, sagittal	-
ligament/quadriceps		MPFLR	1.22 [-]		view, knee coil	
tendon force						
VLO pennation angle (°)	Kan (2007)	AP	14.5 [2.6]	18.7 [6.6]	MRI, 3T, T1-weighted axial images, body coil, 5mm slice thickness	15
VLO/VMO	Kan (2007)	AP			MRI- 3T, T1-weighted axial images, body coil, 5mm slice thickness	15
<i>ACSA</i>			1.9 [0.4]	1.4 [0.2]		
<i>PCSA</i>			1.6 [0.6]	2.1 [0.8]		
VLO/VMO volume	Kan (2007)	AP	1.7 [0.4]	1.6 [0.2]	MRI- 3T, T1-weighted axial images, body coil, 5mm slice thickness	15
VMO elevation	Seeley (2012)	RD	2.78 [3.35]	-	MRI- 1.5T, T1 and T2-weighted sagittal images, knee coil	-
VMO pennation angle (°)	Kan (2007)	AP	14.8 [1.6]	11.4 [2.0]	MRI- 1.5T, T1 and T2-weighted sagittal images, knee coil	15

153 - =not reported; AP = all patients; RD = recurrent dislocation group; MRP = patients undergoing medial retinaculum plasty; VMP = patients undergoing vastus medialis plasty; IR = patients
154 undergoing Insall realignment; MPFLR = patients undergoing medio patellofemoral ligament reconstruction; CG = conservative group; SG = surgical group; ^asign convention changed to make
155 results comparable; ^b the reciprocal of the value reported in the paper has been calculated to make results comparable.

PFJ alignment

Studies frequently reported abnormal measurements of PFJ alignment including congruence angle, patella alta, lateral patellar tilt, Q angle and lateral position of the tibial tubercle relative to the trochlear groove (TT-TG distance) (Table 3).

Measurements of congruence angle in recurrent dislocators were performed in three studies[22, 26, 33] by using three different imaging procedures. Kan et al.[26] found that control participants had slightly medially deviated congruence angles ($-9^{\circ}\pm 12$).

Eight studies[9, 21, 27, 29, 32, 33, 38, 39] quantified patella alta in their assessment of recurrent patellar dislocators, while Aulisa et al.[23] only commented that 43% of the patients were affected. All the articles used the Insall-Salvati Index[40] as their measurement tool, except for Jaquith et al.[29] who used the Caton-Deschamps Index[41]. Most of the measures were performed using lateral X-ray. The studies that compared the Insall-Salvati Index in patellar dislocators and healthy controls did not find significant difference between the two groups[21, 38].

Lateral patellar tilt angle was measured in five studies[22, 25, 33, 38, 39], mostly using X-ray. In contrast with the other studies, Regalado et al.[38] computed lateral patellar tilt as the angle between the lines joining the posterior femoral condyles and the maximum width of the patella, therefore obtaining lower values for patients with respect to controls.

Three studies[23, 24, 39] measured Q angle in their assessment of patients with recurrent patellar dislocation. However, there were no comparative measurements made in control subjects and the measurement method was not defined.

Seven studies[22, 23, 27, 28, 31, 36, 37] documented the TT-TG distance in recurrent patellar dislocators. The measurement was performed on MRI in five studies and on CT in two studies. Only Dickens et al.[36] tested the intra and inter observer reliability of the MRI measurement, finding excellent results. Significantly higher TT-TG distance was found in patients with

dislocation than controls[36, 37]. Contrary to Pennock et al.[37], Dickens et al.[36] showed that older children tended to have higher TT-TG distances and suggested an adjustment of the measure for age.

Patients with recurrent dislocation exhibited a laterally deviated congruence angle (Figure 2), with an average value of 23.6° (Table 4). Overall, alta was universally found in all dislocators, with an average Insall-Salvati Index of 1.28 ± 0.05 between studies. On the other hand, only 20 control participants were assessed, with an average Insall-Salvati Index of 1.20 ± 0.01 . The average lateral patellar tilt between the studies that performed the measurement in patellar dislocators using X-ray and CT scans was $20.8^{\circ} \pm 4.8$. A mean Q angle of $11.2^{\circ} \pm 4.2$ was calculated for the patellar dislocators across the studies (weighted mean patients' age: 12.9 ± 1 years, age range: 9.2-16 years). The mean TT-TG distance in patellar dislocators was 15.5 ± 2.5 mm (patients' age range: 0-17 years), while the average value for 675 controls was 9.4 ± 1.4 mm (weighted mean participants' age: 13.5 ± 1.5 years, age range: 0-15 years).

Figure 2. Description of most frequently reported measures of PFJ alignment and trochlea morphology. (a) Congruence angle: angle formed by bisecting the sulcus angle (α) and central patella ridge. (b) Lateral (left) and medial (right) stress ratios (%), defined as $BC/AB \times 100$. Diagrams of the 2kg stress skyline view at 45° of knee flexion. (c) Insall-Salvati Index, defined as the ratio between patellar length (PL) and patellar tendon length (PT). (d) Lateral patellar tilt: angle between the posterior femoral condyles line and the line of maximum width of the patella. (e) Q angle: angle between a line drawn from the anterior superior iliac spine to central patella (solid red line) and a line drawn from central patella to tibial tubercle (dashed blue line). The Q-angle can be measured in laying or standing. (f) TT-TG distance: distance between a line bisecting the tibial tuberosity (TT) and a line bisecting the trochlear groove sulcus (TG), both perpendicular to the posterior femoral condyles line. (g) Bony sulcus angle: angle between lines joining the highest points of the bony medial and lateral condyles and the lowest bony point of the intercondylar sulcus. (h) Cartilaginous sulcus angle: angle between lines joining the highest points of the cartilaginous medial and lateral condyles and the lowest cartilaginous point of the intercondylar sulcus.

Table 4. Weighted average of most frequently reported measures.

Category	Measure (unit)	Weighted average [SD]		Knees assessed	
		Patients	Controls	Patients	Controls
PFJ alignment					
	Congruence angle (°)	23.6 [4.4] ^{22,26,33}	-9.0 [12.0] ²⁶	62	4
	Lateral stress ratio (%)	36.3 [5.2] ^{32,33}	-	6	-
	Medial stress ratio (%)	-12.5 [8.7] ^{32,33}	-	6	-
	Patella alta				
	<i>Insall-Salvati Index</i>	1.28 [0.05] ^{9,21,27,32,33,38,39}	1.20 [0.01] ^{21,38}	197	20
	Lateral patellar tilt (°)	20.8 [4.8] ^{22,25,33,38,39}	-	132	-
	Q-angle (°)	11.2 [4.2] ^{23,24,39}	-	72	-
	TT-TG distance (mm)	15.5 [2.5] ^{22,23,27,28,31,36,37}	9.4 [1.4] ^{36,37}	254	675
Trochlea morphology					
	Dejour classification (%) ¹¹	50.2 [11.1] ²⁷⁻³⁰	-	133	-
	Sulcus angle (°)				
	<i>bony</i>	150.2 [6.4] ^{9,23,24,26,30,32-34,38,39}	140.4 [6.1] ^{26,34,38}	242	39
	<i>cartilaginous</i>	159.1 [4.9] ^{30,34}	145 [4] ³⁴	67	25
Soft tissue restraints					
	MPFL injury (%)	88.6 [0.2] ^{22,30,35}	-	88	-

Results from the fixed effects model (Table 5) showed that that the weighted mean for TT-TG distance was significantly different ($p < 0.01$) between recurrent patellar dislocators and controls.

Table 5. Weighted mean differences for measures reported in two or more studies that included a control cohort.

Category	Measure (unit)	Weighted Mean Difference [95% CI]	P
PFJ alignment			
	Patella alta		
	<i>Insall-Salvati Index</i> ^{21,38}	0.05 [-0.02-0.12]	0.14
	TT-TG distance (mm) ^{36,37}	3.71 [3.18-4.23]	<0.01
Trochlea morphology			
	Sulcus angle (°)		
	<i>bony</i> ^{26,34,38}	11.72 [8.54-14.9]	<0.01

Trochlea morphology

Characterization of trochlea morphology included predominantly measures of the sulcus angle and trochlea dysplasia, according to the Dejour's classification[11]. Ten studies[9, 23, 24, 26, 30, 32-34, 38] measured the sulcus angle in recurrent patellar dislocators. Four studies performed the measurement using X-ray, three using MRI, two using CT scans and one using ultrasound. The knee flexion angle at which the measurement was taken also varied across the studies, ranging from 15° to 90°.

Trochlea dysplasia in recurrent dislocators was considered in five studies[27-30, 35]. Putney et al.[35] reported that the incidence of trochlear dysplasia in their cohort was 19%, but they did not specify which criteria they used.

The weighted average across the studies for the bony sulcus angle was 150.2°±6.4 for dislocators and 140.4°±6.1 for controls. Nietosvaara et al.[34] and Seeley et al.[30] provided an additional measure of cartilaginous sulcus angle, which resulted being 159.1°±4.9 on average in patellar dislocators. Furthermore, it resulted that 50.2±11.1% of patients with recurrent dislocation had a form (types A, B, C, D) of dysplastic trochlea, according to Dejour's classification.

The weighted mean for bony sulcus angle was significantly different ($p < 0.01$) between recurrent patellar dislocators and controls (Table 5).

Soft tissue restraints

Few studies[21-23, 26, 30, 35] reported measurements of passive (ligaments) and active (muscle) soft tissue restraints of the PFJ, and these measurements were often performed only in one study. Indeed, only one study[23] reported measures of generalized ligamentous laxity found by the Carter-Wilkinson test[42]. Zhao et al.[22] also reported that most of recurrent dislocators had hyperlaxity, but they did not specify how this was determined. Three studies evaluated the integrity of the MPFL by visual inspection. Putney et al.[35] considered only patients with MPFL injury and he reported that, of those, 53% involved tears and 47% involved stretching. From two studies, it resulted that, on average, $88.6 \pm 0.2\%$ of recurrent dislocators exhibited either a MPFL strain or tear.

DISCUSSION

The paediatric cohort of PFJ dislocators reflects a highly variable group of patients who often have a constellation of predisposing biomechanical factors towards PFJ instability. The primary aim of this article was to quantify from the current literature which measurable radiological parameters were most likely to cause PFJ instability by comparing control and dislocator populations. The secondary aim was to stratify the individual parameters to determine which factor would be the most likely to contribute to instability in children.

Overall, we were able to find thirty-one reported parameters, however only ten of these measurements had been assessed in multiple articles. Only 4 of these parameters (congruence angle, patella alta, TT-TG distance, sulcus angle) had both dislocator and control population

results. Due to the paucity of data, it was difficult to draw conclusions based on this information, however it is possible to make recommendations that future studies in this population should quantify factors that cover all the main categories that can contribute to pathology, particularly the parameters that are considered relevant in the general literature.

In terms of extrinsic lower limb alignment, genu valgum is very commonly seen in paediatric patients as part of normal limb development. Cases of excessive valgus can be corrected by guided growth procedures with minimal morbidity[43] in older children with an intermalleolar distance of more than 8cm. In a recurrent patellar dislocator with excessive genu valgum, correction of valgus alignment contributes to improved patella tracking and provide an extrinsic method of improving stability by effectively decreasing the TT-TG distance.

Likewise, in children with persistent femoral anteversion or miserable malalignment contributing to patella instability, derotational osteotomy provides realignment of the extensor mechanism relative to the trochlea without violation of the knee itself[44]. This procedure, while more invasive, is also well tolerated in children compared to adults. There is no literature that supports how much anteversion is considered significantly abnormal, how much derotation is required or consequently at what level the procedures should be performed. Without consistent documentation of both coronal and axial plane alignment, it will be difficult to generate the body of information required to answer these clinical questions.

The most frequently reported radiological factors involving intrinsic PFJ alignment included congruence angle, patella alta, patella tilt and the TT-TG distance. It is well accepted that all of these factors play a role in the adult population however it was evident that patella alta was not as reliable a measure in the paediatric cohort of patients.

The average values of the Insall-Salvati Index were equal to or greater than 1.2 in both groups, suggesting that this factor was not able to discriminate between controls and dislocators in the

paediatric population. This might derive from the lack of ossification in the tibial plateau which generates a falsely high measurement in the control group of children. The poor reproducibility of the measure may also have been due to difficulty in determining the patellar tendon insertion point[45]. Conversely, the Caton-Deschamps Index has been shown to be a simple, accurate and reproducible measure to derive patellar height in children, but age-based values should be considered, given that the index increases as the age decreases[46]. An alternative measure of patellar height is the patellotrochlear index, which is believed to reflect the functional patellar height more accurately[47] and could be computed from MRI. These results might also be affected by the low number of control participants for which the measurement was computed.

The TT-TG distance appears to discriminate between patients and controls, with dislocators demonstrating higher TT-TG distances than controls. Previous studies[48, 49] used a 20mm threshold[11] as the rationale for medializing tibial tubercle osteotomy in skeletally mature patients. There have not been any studies to our knowledge that define the role of TT-TG distance prior to a Roux-Goldthwait style procedure for the skeletally immature patient[50]. The average TT-TG distance for patients reported in this review (15.55 ± 2.53 mm) is lower than expected, possibly because this parameter has been shown to increase logarithmically with age[36]. MRI values for the TT-TG distance have been shown to be smaller than those measured on CT. It is the authors feeling that CT in the paediatric population should be avoided and that MRI should be adopted as the standard tool for assessment, therefore values should be standardized for MRI. Differences in TT-TG distance measurements on MRI have been reported however, with significantly lower values when measured on higher resolution MRI[36, 51] due to flexion of the knee when positioned in a dedicated knee coil. The tibial tubercle is subsequently lateralized due to the screw-home mechanism. The TT-TG distance is also sensitive to small changes (5°) in knee adduction and abduction relative to neutral axis alignment, with an alteration of the measurement by as much as 40%[52]. Consequently, this

measure should be taken with the knee in full extension with images reconstructed in neutral axis alignment while using a body coil.

The results from this review suggest that a TT-TG distance threshold measured on MRI >15mm might be a good indication for medial tibial tubercle transfer in skeletally mature patients with recurrent patellar instability, as proposed by Schoettle et al [53]. Nonetheless, an adjustment for age is recommended.

It was surprising to find that Q angle was not well reported in the literature. There is considerable controversy about how to measure the Q angle as well as the implications of an elevated result[54]. Variability exists in the number of degrees of flexion the knee should be positioned in, as well as the state of contraction of the quadriceps muscle[55]. A significant negative relationship between Q angle and TT-TG distance was reported by Cooney et al.[56] in symptomatic patients with relaxed knees, implying that one cannot substitute the clinical for the radiological measure.

Morphologic features of the trochlea were primarily assessed by measuring the sulcus angle[57, 58]. With respect to patellar dislocators and controls, the dislocators exhibited shallower bony ($150.2^{\circ} \pm 6.4$) and cartilaginous ($159.1^{\circ} \pm 4.9$) sulcus angles. A shallow sulcus angle may be the consequence of abnormal biomechanics of the PFJ in the first instance, as cartilage shape occurs partly as a result of the applied forces. It has been shown[59] that bony and cartilaginous sulcus angles are age dependent, with general higher values for older participants with patellar instability. Therefore, this should be taken into account when establishing the predictive value of the measurement. Different imaging techniques and methods have been used to measure the sulcus angle. It has been shown that there is a reasonable level of inter-observer and intra-observer reliability and validity for this measurement when using CT and MRI[60, 61]. Van Huyssteen et al.[62] demonstrated that there is a highly significant difference between bony

and cartilaginous sulcus angle measured in patients with a dysplastic trochlea. Although the bony trochlea is dysplastic in these patients, the cartilage morphology can worsen this abnormal shape. MRI is therefore preferable to CT in facilitating surgical planning in a paediatric population.

Meta-analysis was only possible for three measures, the Insall-Salvati Index, TT-TG distance and bony sulcus angle, as these were the only parameters documented in comparisons of PFJ dislocators and controls in two or more studies. A statistically significant difference in weighted mean averages between controls and dislocators was found for the TT-TG distance and bony sulcus angle, indicating that, in paediatric patients, it is likely that these are the only two parameters we can confidently use to predict risk of recurrence.

The evidence base exhibited a number of substantial methodological limitations. Firstly, the majority of risk factors for patellar instability were measured from two-dimensional medical images collected in static postures. Construct validity is questionable, as patellar instability is a consequence of dynamic factors that change with many variables including joint kinematics and force vector contributions from the quadriceps. Secondly, normative values for potential predictors of patellar dislocation were difficult to derive, mainly because of the lack of matching controls in most studies. The heterogeneity of factors predisposing to patellar dislocation could also influence the categorization of values into normal and abnormal ranges. For example, increased severity of trochlea dysplasia has been shown to affect measurements of TT-TG distance, with an underestimation of the value up to 3mm[63] and a decreased inter and intra observer agreement[64]. Overall, none of the included studies conducted a comprehensive predictive analysis to estimate the relationship between lower limb and PFJ alignment, trochlea morphology and soft tissue restraints of the PFJ. Finally, the quality of the studies should be taken into account when evaluating the external validity and generalizability of the results. According to the quality assessment, only 35% of the included studies scored

above 70% on the Downs and Black checklist[17] and could therefore be deemed to be of sufficient quality to be considered for meta-analysis. However, there was not enough consistency between studies to attempt a meta-analysis.

Based on the current evidence, the authors believe that studies reporting on recurrent patellar dislocation in children and adolescents should include at the very minimum, assessment of (i) lower limb alignment in the coronal and axial planes; (ii) PFJ alignment to include TT-TG distance and congruence angle; (iii) trochlea morphology measurements to include sulcus angle and (iv) assessment of generalized ligamentous laxity. Laxity has been unanimously shown to predispose to recurrent patella dislocation[65, 66]. In this review it has only been reported in one study[23] and was found in 62.5% of patients. A standardized clinical Q angle protocol needs to be established and validated before including this measurement in the assessment of recurrent patellar dislocation. These recommendations could change in the future, when a more comprehensive dataset of factors predisposing to patellar dislocation in a paediatric cohort will be available.

The cause of recurrent patellar dislocation cannot currently be predicted from traditional statistical methods given the complex interplay between lower limb bone morphology, bony alignment and soft tissue restraints of the PFJ. Algorithms based on predictive analyses can only provide general information at a population level. Often, results are presented from mixed populations of children, adolescents and adults which may lead to inaccurate conclusions, since the lower extremity characteristics change according to age and skeletal maturity. Most of contemporary metrics are static measures of the PFJ alignment and are inappropriate to investigate the patellar dislocation mechanism during dynamic tasks. Clinical gait analysis[67] cannot provide patient-specific insights on PFJ (dys)function, because this joint is not included in currently employed models. Conversely, computational models of the musculoskeletal system[68, 69] can incorporate patient-specific lower limb bone models, and may allow us to

evaluate the PFJ dynamic function by combining gait analysis typical measurements with estimation of internal loads[70]. Such biomechanical methods may therefore provide insight into the complex etiology of patellar dislocation, help identify individualized risk factors for recurrent patellar dislocation, and quantitatively describe patellar stability throughout the knee range of motion in static and dynamic activities.

CONCLUSION

The findings of this review suggest that PFJ alignment is a major risk factor for recurrent patellar dislocation. Our meta-analysis showed that children and adolescents with recurrent patellar dislocation exhibited significantly higher TT-TG distance and bony sulcus angle with respect to age-matched control participants. These findings indicate that TT-TG distance and bony sulcus angle are the only two parameters we can confidently use to predict the risk of recurrence in paediatric patients. These results can streamline the patient evaluation and best inform clinical decision making.

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Competing interests

None declared.

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