Original Article

Increased Risk of Patellofemoral Instability Events and Surgical Management in Patients With Joint Hypermobility Syndromes: A Matched Cohort Analysis

Michael J. Kutschke, M.D., J. Alex Albright, B.S., Julia M. Winschel, B.A., M.P.H., Elaine W. He, B.S., Aristides I. Cruz Jr., M.D., M.B.A., Alan H. Daniels, M.D., and Brett D. Owens, M.D.

Purpose: To compare the odds of patellofemoral instability events requiring subsequent surgery and revision surgical intervention in patients with joint hypermobility syndromes (JHS) to that of a matched cohort. **Methods:** This is a retrospective cohort study using the PearlDiver Mariner Database. Records were queried between 2010 and 2021 with a diagnosis of JHS, including Ehlers-Danlos syndrome (EDS) and Marfan syndrome. Propensity matching was performed with a randomly generated control cohort without a diagnosis of JHS to account for age, sex, Charlson comorbidity index, diabetes, and obesity. Multivariable logistic regression was used to compare rates of patellar dislocation over a 1- and 2-year period between the 2 cohorts while controlling for previous knee injury or surgery. Patients who sustained a patellar dislocation over the 2-year period were followed to calculate rates of surgical intervention and subsequent revision. **Results:** In a population of 91,747, those with JHS experienced patellofemoral instability at a significantly increased rate at both a 1-year (adjusted odds ratio [aOR] 11.40; 95% confidence interval 9.23-14.25, $P < .001$) and 2-year (aOR 8.73; 7.36-10.44, $P < .001$) periods. The greatest risk was observed in patients with EDS at 1 year (aOR 16.32; 12.54-21.67, $P < .001$). Of those with an instability event, patients with JHS experienced a significantly increased rate of surgery at 1 year (aOR 3.20; 1.61-7.28, $P = .002$) and 2 years (aOR 3.18; 1.70-6.62, $P < .001$). Of those treated with surgery, there was no significant difference in the rates of revision surgical intervention between the JHS and control cohorts. **Conclusions:** Patients with JHS experienced significantly increased rates of patellofemoral instability and subsequent surgery. However, of those treated with surgery, there was no difference in rates of revision surgical intervention between those with or without joint hypermobility syndromes. **Level of Evidence:** Level III, retrospective cohort study.

The joint hypermobility syndromes (JHS) are a group of connective tissue disorders with joint laxity as a key area of phenotypic overlap. Two of the most common conditions are Ehlers-Danlos syndrome (EDS), a heterogeneous group of connective tissue diseases, and Marfan syndrome (MFS), an autosomal-dominant disorder of collagen production.[1] Generalized ligamentous laxity is known to contribute to joint instability, particularly of the patellofemoral joint.[2] Patellar dislocation is a relatively common orthopaedic condition, with the greatest incidence seen between the ages of 14 and 18 years, and is particularly common in pediatric patients in part because of the rapid skeletal growth that occurs during adolescence.[3-5] Patellofemoral instability is also common in patients with JHS. Studies have estimated that more than 80% of patients with EDS experience knee pain,[6] and more than one half of patients diagnosed with EDS experience patellar instability.[7] Among pediatric patients with EDS, the knee is the joint most often affected by pain or instability.[8]

Anatomic factors that influence stability of the patellofemoral joint have been widely investigated. Various studies also have focused on identifying risk factors for recurrent patellar instability, which include age at first dislocation, patella alta, elevated tibial tubercle to

From the Warren Alpert Medical School of Brown University, Providence, Rhode Island, U.S.A.

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Address correspondence to Michael J. Kutschke, M.D., 2 Dudley St., Suite 200, Providence, RI 02905, U.S.A. E-mail: m.j.kutschke@gmail.com

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trochlear groove distance, genu valgum, increased femoral anteversion, and trochlear dysplasia.[9] However, biologic considerations such as JHS remain less clearly understood.[10,11]

After nonsurgical management of patellar dislocation, many patients will develop recurrent patellofemoral instability.[12] A systematic review of overlapping meta-analyses by Erickson et al.[13] revealed an overall rate of recurrent patellar dislocation to be 34.7% after primary conservative management. Studies also have shown that this subsequent instability negatively affects patient-reported quality of life and physical activity level.[6] Further, recurrent instability of the patellofemoral joint increases the risk of early-onset osteoarthritis.[14] However, less is known about the risk of recurrent patellofemoral instability after surgical management of instability events, specifically in patients with JHS.

Surgical management of joint instability events in patients with JHS is complicated by muscle weakness and defective collagen function characteristic of such syndromes.[15] One study demonstrated good outcomes after medial patellofemoral ligament (MPFL) reconstruction for patellofemoral instability in pediatric patients with a previous diagnosis of JHS.[16] In contrast, other studies have found that patients with joint hypermobility experience significantly lower functional improvement after MPFL reconstruction compared with patients without joint hypermobility.[17] To help guide clinical decisions and achieve optimal patient outcomes, further research is needed to evaluate the efficacy of surgical intervention for patellofemoral instability in patients with JHS compared with those without connective tissue disorders.

The purpose of this study was to compare the odds of patellofemoral instability events requiring subsequent surgery and revision surgical intervention in patients with JHS to that of a matched cohort. We hypothesized that patients with a known diagnosis of JHS would have significantly greater rates of patellofemoral instability and subsequent surgery and revision.

Methods

Data Source

This is a retrospective cohort study. The PearlDiver Mariner Database (PearlDiver Technologies, Colorado Springs, CO) was used to conduct a retrospective analysis of deidentified data from January 2010 through December 2021 within the M157Ortho dataset. This is a multipayor administrative claims dataset generated and periodically updated using the insurance claims of more than 150 million patients. These data allow researchers to characterize and analyze short-, medium-, and long-term rates and trends of various conditions, postoperative complications, and sequelae using the *International Classifications of Diseases, Ninth Revision* (ICD-9) and *Tenth Revision* (ICD-10) and Current Procedural Terminology (CPT) codes. For the current study, these data were used to calculate and compare rates of index patellar dislocations, primary surgical stabilization (repair or reconstruction), and reoperation in those who underwent surgery with a diagnosis of JHS and a matched control population.

Creating the Cohorts

Three separate queries of the Mariner dataset were performed. One cohort was created from patients who had an insurance claim with an associated diagnosis code of only EDS (75683, Q79.6, Q79.60, Q79.61, Q79.62, Q79.63, Q79.69). A second cohort was created from patients with MFS (75982, Q87.40, Q87.410, Q87.418, Q87.42, Q87.430). A third cohort was created from the combination of all the aforementioned EDS and MFS diagnosis codes, referred to herein as the all JHS cohort. The cohorts were subsequently filtered to exclude patients who were diagnosed after 2019, those younger than 10 years old (given unique treatment considerations for young pediatric populations), and those not active within the dataset for at least 2 years after their diagnosis (i.e., loss of insurance, change of providers, mortality, etc.). These criteria allowed for a minimum of 2 years of follow-up for each patient included in the analysis. Patients diagnosed with a rheumatologic disease also were excluded. The 3 cohorts were subsequently matched using one-to-one nearest neighbor propensity score matching with a caliper of 0.2 to account for age at diagnosis, sex, Charlson Comorbidity Index (CCI),[18] diabetes, and obesity (body mass index >30 kg/m^2) to a randomly generated control cohort that met the inclusion criteria. This mode of matching allows for the creation of 2 groups that are statistically similar regarding the variables matched on while not losing patients in the experimental cohort to ensure maximum power. Of note, 3 separate cohorts and subsequent matched control cohorts were created (i.e., one for only EDS, one for only MFS, and one including all JHS). As such, the sum of the number of patients in the diagnosis-specific cohorts will not equal that of the all JHS cohort, as this cohort contains hypermobility diagnoses beyond only EDS and MFS.

Rates of Index Injury and Primary Surgery

The rates of index patellar dislocations were calculated over 1- and 2-year periods after the initial diagnosis of a JHS. These injuries were identified using ICD-9 and ICD-10 diagnostic codes (Appendix Table 1, available at www.arthroscopyjournal.org). The rates of primary surgical stabilization were identified using CPT codes (27420, 27422, 27427, 27418).

Rates of Revision Surgical Stabilization

A second query was performed to isolate all patients who underwent a primary surgical stabilization to

address patellar instability. In addition, only patients who underwent surgery with an associated ICD-10 code that specified laterality were included in the study in order to accurately track these patients to determine surgical failure and revision surgical stabilization. This cohort was also stratified by the presence or absence of a diagnosis of either EDS or MFS. A second matching process that followed the previously described protocol was performed with these cohorts. Rates of revision surgical stabilization within 1 and 2 years after the primary procedure were calculated and compared between the JHS cohort and the control.

Statistical Analyses

Multivariable logistic regression was used to account for residual differences in age, sex, CCI, diabetes, and obesity, while accounting for additional potential confounding variables, including tobacco use, previous ligamentous knee injury, previous cruciate ligament surgery, and previous meniscal surgery while comparing rates of patellofemoral dislocation, primary surgical stabilization, and revision surgery. Adjusted odds ratios (aORs) and 95% confidence intervals (CIs) were generated and reported for each comparison. The aOR was calculated using multivariable logistic regression controlling for age, sex, CCI, diabetes, obesity (body mass index >24.9), tobacco use, previous ligamentous knee injury, previous cruciate ligament surgery, and previous meniscal surgery. To protect patient identity, cohorts of fewer than 11 patients are reported as "−1" by PearlDiver and thus were reported as "<11" throughout this article. Statistical analyses were still able to be performed on these smaller cohorts, but the specific value is simply not reported. A P value of $< .05$ was determined to represent statistical significance a priori. All statistical analyses were performed using the R Statistical Package (version 4.2.1; R Core Team 2022; R Foundation for Statistical Computing, Vienna, Austria) embedded within PearlDiver.

Results

After we created and matched the 3 separate cohorts, the all-JHS cohort consisted of 91,747 patients, the EDS cohort had 65,175 patients, and the MFS cohort had 27,573 patients, with each of their respective control cohorts containing the same number of patients. Among each cohort, there were residual differences between the ages, sexes, CCIs, and rates of diabetes and obesity (Table 1).

After controlling for the residual differences in demographics and the noted potential confounding variables, patients in the EDS, MFS, and all JHS cohorts had significantly greater odds of being diagnosed with an index patellar dislocation at both the 1-year and 2- year marks after initial diagnosis of a JHS compared with control patients (Table 2). The greatest odds of being diagnosed with a patellar dislocation were seen in the EDS cohort at 1 year after initial EDS diagnosis (aOR 16.32, 95% CI 12.54-21.67, $P < .001$).

Table 1. Demographics and Comorbidities of the Experimental Cohorts and Control Groups With Comparisons.

Condition and Demographic	Experimental Cohort	Control Cohort	*P* Value
All JHS	n = 91,747	n = 91,747	
Age, yr, mean ± SD	32.6 ± 17.0	30.9 ± 18.7	**<.001**
CCI, mean ± SD	0.8 ± 1.3	0.5 ± 1.0	**<.001**
Sex (male), n (%)	27,774 (30.3)	25,799 (28.1)	**<.001**
Diabetes, n (%)	13,389 (14.6)	12,806 (14.0)	**<.001**
Obesity, n (%)	20,764 (22.6)	17,090 (18.6)	**<.001**
EDS	n = 65,175	n = 65,175	
Age, yr, mean ± SD	32.2 ± 16.3	31.2 ± 18.7	**<.001**
CCI, mean ± SD	0.8 ± 1.2	0.8 ± 1.2	**<.001**
Sex (male), n (%)	12,834 (19.7)	12,504 (19.2)	**.022**
Diabetes, n (%)	9,059 (13.9)	9,218 (14.1)	.208
Obesity, n (%)	16,767 (25.7)	14,056 (21.6)	**<.001**
MFS	n = 27,573	n = 27,573	
Age, yr, mean ± SD	33.3 ± 18.5	34.9 ± 19.2	**<.001**
CCI, mean ± SD	0.8 ± 1.4	0.8 ± 1.4	**.021**
Sex (male), n (%)	15,659 (56.8)	15,691 (56.9)	.790
Diabetes, n (%)	4,450 (16.1)	5,178 (18.8)	**<.001**
Obesity, n (%)	4,132 (15.0)	4,237 (15.4)	.217

NOTE. Bold values represent statistically significant results ($P < .05$).
CCI, Charlson Comorbidity Index; EDS, Ehlers-Danlos syndrome; JHS, joint hypermobility syndromes; MFS, Marfan syndrome.

Table 3 shows the rates of primary surgical stabilization after patellar dislocation for each cohort. Within the all JHS cohort, there were 1,189 patients who experienced a patellar dislocation. Of these patients, 177 (14.9%) underwent surgical stabilization to address their patellar dislocation within 1 year of their initial JHS diagnosis and 208 (17.5%) underwent surgery by 2 years post-JHS diagnosis. Within the EDS cohort, 1,027 patients experienced a patellar dislocation, with 162 (15.8%) of these patients undergoing surgery within 1 year of their initial EDS diagnosis and 185 patients (18.0%) underwent surgery within 2 years of EDS diagnosis. In the MFS cohort, 200 patients experienced a patellar dislocation. Twenty-two (11.0%) of these patients underwent surgery within 1 year of their initial MFS diagnosis and 30 (15.0%) patients underwent surgery within 2 years of MFS diagnosis. Of patients who experienced an instability event, the JHS cohort had significantly greater odds of undergoing surgery at 1 year (aOR 3.20, 1.61-7.28, $P = .002$) and 2 years (aOR 3.18, 1.70-6.62, $P < .001$). Although the EDS cohort had significantly greater odds of undergoing surgery at 1 year (aOR 3.52, 1.52-10.19, $P = .008$) and 2 years (aOR 2.99, 1.44-7.28, $P = .007$) compared with the control cohort, the MFS cohort did not at either 1 year (aOR 1.89; 0.49-12.43, $P = .418$) or 2 years (aOR 1.78; 0.56-7.92, $P = .378$).

Table 2. Rates of Being Diagnosed With a Patellar Dislocation Over a 1- And 2-Year Period in Patients With a JHS and a Comparison With a Control Cohort

Condition and Time Frame	JHS Cohort, n (%)	Control Cohort, n (%)	OR (95% CI)	*P* value	aOR* (95% CI)	*P* value
All JHS	n = 91,747	n = 91,747				
1 yr	979 (1.1)	94 (0.1)	10.51 (8.51-13.00)	**<.001**	11.40 (9.23-14.25)	**<.001**
2 yr	1,189 (1.3)	150 (0.2)	8.02 (6.76-9.50)	**<.001**	8.73 (7.36-10.44)	**<.001**
EDS	n = 65,175	n = 65,175				
1 yr	859 (1.3)	57 (0.1)	15.26 (11.67-19.95)	**<.001**	16.32 (12.54-21.67)	**<.001**
2 yr	1,027 (1.6)	99 (0.2)	10.52 (8.56-12.94)	**<.001**	11.31 (9.20-14.06)	**<.001**
MFS	n = 27,573	n = 27,573				
1 yr	152 (0.6)	21 (0.1)	7.27 (4.61-11.48)	**<.001**	6.28 (4.03-10.27)	**<.001**
2 yr	200 (0.7)	32 (0.1)	6.29 (4.33-9.14)	**<.001**	5.38 (3.72-8.02)	**<.001**

NOTE. Bold values represent statistically significant results ($P < .05$).

aOR, adjusted odds ratio; CI, confidence interval; EDS, Ehlers-Danlos syndrome; JHS, joint hypermobility syndromes; MFS, Marfan syndrome; OR, odds ratio.

Of the patients who underwent primary surgical stabilization, 52 patients required revision surgery to address their patellar instability (Table 4). There was no significant difference in the rates of revision surgical intervention between the JHS and control cohorts.

Discussion

The current study found that patients with JHS experienced significantly increased odds of patellofemoral instability and subsequent surgical stabilization compared with matched controls, which is congruent with our current understanding of hypermobility. Most notably, however, there was no significant difference among the 3 hypermobility groups and the matched controls with respect to rates of revision surgery in the management of recurrent patellofemoral instability after initial surgical intervention. In addition, the phenomenon of increased patellofemoral instability was most pronounced in the EDS cohort. Also, the MFS cohort uniquely only demonstrated increased odds of patellar instability events but no difference in odds of surgical intervention compared with those without joint hypermobility.

Patellofemoral instability is a common area of study, and the primary focus of many research efforts remains the anatomic and kinematic features that contribute to this condition.[19-22] Despite these advances in our understanding as well as available surgical techniques, the optimal algorithm for treating this problem still remains unclear. This lack of understanding in managing patellofemoral instability is further compounded when considering patients with abnormal connective tissues.

Our results supplement previous literature directed at understanding the management of patients at the confluence of syndromic joint hypermobility and patellofemoral instability. A review and meta-analysis by Pacey et al.[23] evaluated the risk of lower-extremity joint injury in the setting of adolescent and young adult athletes with generalized joint hypermobility. In the 18 included studies, generalized joint hypermobility was defined by 7 different clinical scales, the primary of which was the 9-point Beighton scale, although various cutoffs to define hypermobility were used throughout the studies. On the basis of the authors' standardized definition of hypermobility, participants with hypermobility engaging in contact sports demonstrated a

Table 3. Rates of Undergoing Surgery to Address a Patellar Dislocation Over a 1- and 2-Year Period in Patients With a JHS and a Comparison With a Control Cohort

Condition and Time Frame	JHS Cohort, n (%)	Control Cohort, n (%)	OR (95% CI)	*P* Value	aOR* (95% CI)	*P* Value
All JHS	n = 1,189	n = 150				
1 yr	177 (14.9)	<11 (N/A)	3.10 (1.50-6.44)	**.002**	3.20 (1.61-7.28)	**.002**
2 yr	208 (17.5)	<11 (N/A)	2.97 (1.54-5.74)	**.001**	3.18 (1.70-6.62)	**<.001**
EDS	n = 1,027	n = 99				
1 yr	162 (15.8)	<11 (N/A)	3.52 (1.42-11.26)	**.004**	3.52 (1.54-10.19)	**.008**
2 yr	185 (18.0)	<11 (N/A)	2.89 (1.32-7.50)	**.006**	2.99 (1.44-7.28)	**.007**
MFS	n = 200	n = 32				
1 yr	22 (11.0)	<11 (N/A)	1.85 (0.41-8.29)	.612	1.89 (0.49-12.43)	.418
2 yr	30 (15.0)	<11 (N/A)	1.71 (0.49-5.96)	.567	1.78 (0.56-7.92)	.378

NOTE. Bold values represent statistically significant results ($P < .05$).

aOR, adjusted odds ratio; CI, confidence interval; EDS, Ehlers-Danlos syndrome; JHS, joint hypermobility syndromes; MFS, Marfan syndrome; N/A, not available; OR, odds ratio.

Table 4. Rates of Requiring a Revision Surgery to Address Patellar Instability in Patients With a JHS Over a 1- and 2-Year Period in Patients With a JHS and a Comparison With a Control Cohort

Condition and Time Frame	JHS Cohort, n (%)	Control Cohort, n (%)	OR (95% CI)	*P* Value	aOR* (95% CI)	*P* Value
All JHS	n = 52	n = 52				
1 yr	<11 (N/A)	<11 (N/A)	1.23 (0.35-4.30)	1.000	1.04 (0.24-4.51)	.958
2 yr	<11 (N/A)	<11 (N/A)	1.57 (0.41-5.92)	.739	1.12 (0.31-4.01)	.861

aOR, adjusted odds ratio; CI, confidence interval; EDS, Ehlers-Danlos syndrome; JHS, joint hypermobility syndromes; MFS, Marfan syndrome; N/A, not available; OR, odds ratio.

combined odds ratio of 4.69 (95% CI, 1.33-16.52; $P = .02$) in experiencing a knee injury compared with their counterparts without hypermobility. This elevated odds ratio is similar, but to a lesser extent, to our current results, which revealed an aOR of 11.40 (9.23-14.25; $P < .001$) and 8.73 (7.36-10.44; $P < .001$) in patients with JHS at 1 and 2 years, respectively. This presents an interesting finding in that the patients in the meta-analysis by Pacey et al.[23] were contact athletes, which would intuitively produce a greater odds ratio compared with the general sample presented in the current study. In addition, the present study specifically only included patellofemoral instability, whereas the study by Pacey et al.[23] included all knee injuries; again, this would intuitively produce a perhaps greater odds ratio in a contact athlete population. This discrepancy suggests a potentially greater detrimental effect with respect to knee injuries in patients with specific JHS diagnoses compared with those with clinically hypermobile joints alone.

Much of the previous literature in this area has focused on smaller, specific study populations, whereas the current work represents a large-scale assessment of the diagnosis and treatment of patients with patellofemoral instability and JHS. Despite this inherent discrepancy in study population size, the results herein are relatable to smaller, clinical studies. Howells and Eldridge[17] performed a case-control study containing 25 adult patients with hypermobility (Beighton score $\geq$6) and a matched control group of 50 patients (Beighton score <4) all of whom were treated with a standard isolated arthroscopically aided MPFL reconstruction using semitendinosus autograft. Of note, 8 of the 25 patients in the hypermobility group had been diagnosed with a specific JHS. Although both groups improved significantly compared with their preoperative state, those in the hypermobility group had a significantly worse postoperative Kujala score ($P < .001$) and patient satisfaction ($P = .011$) compared with the control group. This result may be attributable to the possible disproportionate effects of approximately one third of their patients with hypermobility (8/25) reportedly known to have a specific JHS diagnosis causing a reduction in outcomes measures within the hypermobile arm of their study. Conversely, Hiemstra et al.[24,25] conducted a retrospective cohort analysis of 167 adult patients treated with isolated MPFL reconstruction with semitendinosus or gracilis autograft. Postoperative outcomes assessed by the physical domain of the Banff Patellofemoral Instability Instrument revealed no difference across all degrees of hypermobility (Beighton scores 0-9) without the mention of syndromic patients.[24] In conjunction with the results of the present study, these findings again offer additional support to the possibility of syndromic hypermobility potentially causing a greater impact on patellofemoral joint instability compared with patients who are classified as hypermobile on the basis of a joint mobility score alone. Also important to note is that both aforementioned studies used autograft to reconstruct the MPFL in patients with hypermobility, with good results. Although it seems counterintuitive to use diseased autograft to reconstruct an incompetent structure, this has not been clearly elucidated in the literature as there is support for both allograft and autograft reconstruction techniques.[14,26-29]

The present study included 3 experimental cohorts: patients with EDS, patients with MFS, and a combined cohort containing patients with EDS or MFS termed the all-JHS cohort. The results for the EDS cohort closely followed and were even more pronounced than those of the all-JHS cohort demonstrating increased odds of patellofemoral instability as well as subsequent surgery compared with matched controls. Interestingly, patients with MFS had greater odds of experiencing patellofemoral instability but were at no greater risk of proceeding on to surgical management compared with matched controls. One explanation is that this population of patients perhaps achieves greater results from nonoperative management strategies than their other JHS counterparts and do not require as frequent surgical management for this issue. As this result is not well explained by the current study, this discrepancy remains an avenue for further investigation.

The spectrum of JHS is wide; although there are many genetic causes of joint hypermobility, including MFS and EDS, joint hypermobility is also a characteristic of many other medical diagnoses.[30] It may be that one particular diagnosis constitutes the majority of these results, but the current study is not granular

enough to capture that detail. In addition, the possible anatomic factors contributing to patellofemoral instability included in the study were unable to be captured by our data source, nor could the rate and level of return to sport be ascertained. It is possible that the current study attributes the increased odds of patellofemoral instability and subsequent surgery in patients with JHS to their connective tissue diagnosis when it is truly multifactorial.

These findings have implications for clinical decision making in treating patients with JHS, as they may not be best managed by the same approach that is applied to patients without JHS. Because it can be challenging for patients to obtain a timely diagnosis of JHS,[31] clinicians should always be suspicious of JHS even if a patient lacks a formal diagnosis. Patients with JHS, particularly EDS, should be made aware of their increased risk of patellofemoral instability and the odds that surgical intervention will be subsequently pursued. It is paramount to set patient expectations accordingly on the basis of the presence of joint hypermobility. In addition, the results of the current study suggest that surgical intervention without significantly increased risk of revision surgery should be part of the initial treatment discussion in patients with JHS after an isolated patellofemoral instability injury.

Limitations

We acknowledge several limitations to this study. Although using PearlDiver allows for a large sample size, which increases the statistical power of our findings, the current study is limited in that patients included in analyses were not sampled randomly; possible sampling bias may limit the generalizability of our results. Further, because this study used ICD and CPT billing codes to identify patients with specific diagnoses, it is possible that some patients were miscoded, and that undiagnosed patients were not accounted for. Lastly, by nature of the database and intentionally broad nature of the query, granular details are lost with respect to patient anatomic factors as well as the specific surgical treatments provided for each patient.

Because of the study design, it is not possible to determine whether the first dislocation events observed during the study period represent an initial or recurrent patellar instability event, as instability events may have occurred prior to the study period. In addition, although this study represents a large cohort of patients with JHS, the *P* values produced by the statistical analysis can be disproportionately magnified by the very large study sample. Nonetheless, the use of the database allowed for construction of the cohorts to match in number of patients and possible confounding variables.

Another limitation pertains to the exclusion of young pediatric patients from this study. Patients younger than the age of 10 years are skeletally immature, thus typically requiring surgical techniques that spare the physis to reduce the risk of growth disturbance.[5] Because treatment considerations differ for young pediatric populations, they were not included in analyses which limits the application of our results to this age group.

Conclusions

Patients with JHS experienced significantly increased rates of patellofemoral instability and subsequent surgery. However, of those treated with surgery, there was no difference in rates of revision surgical intervention between those with or without joint hypermobility syndromes.

Disclosures

All authors (M.J.K., J.A.A., J.M.W., E.W.H., A.I.C., A.H.D., B.D.O.) declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

1. Malfait F, Hakim AJ, De Paepe A, Grahame R. The genetic basis of the joint hypermobility syndromes. *Rheumatol Oxf Engl* 2006;45:502-507.
2. Nomura E, Inoue M, Kobayashi S. Generalized joint laxity and contralateral patellar hypermobility in unilateral recurrent patellar dislocators. *Arthroscopy* 2006;22:861-865.
3. Fithian DC, Paxton EW, Stone ML, et al. Epidemiology and natural history of acute patellar dislocation. *Am J Sports Med* 2004;32:1114-1121.
4. Sanders TL, Pareek A, Hewett TE, Stuart MJ, Dahm DL, Krych AJ. Incidence of first-time lateral patellar dislocation: A 21-year population-based study. *Sports Health* 2018;10:146-151.
5. Popkin CA, Bayomy AF, Trupia EP, Chan CM, Redler LH. Patellar instability in the skeletally immature. *Curr Rev Musculoskelet Med* 2018;11:172-181.
6. Rombaut L, Malfait F, Cools A, De Paepe A, Calders P. Musculoskeletal complaints, physical activity and health-related quality of life among patients with the Ehlers-Danlos syndrome hypermobility type. *Disabil Rehabil* 2010;32:1339-1345.
7. Ainsworth SR, Aulicino PL. A survey of patients with Ehlers-Danlos syndrome. *Clin Orthop* 1993;286:250-256.
8. Stern CM, Pepin MJ, Stoler JM, Kramer DE, Spencer SA, Stein CJ. musculoskeletal conditions in a pediatric population with Ehlers-Danlos syndrome. *J Pediatr* 2017;181:261-266.
9. Pascual-Leone N, Ellis HB, Green DW. Patellar instability: Will my patella dislocate again? *Curr Opin Pediatr* 2022;34:76-81.
10. Redler LH, Dennis ER, Mayer GM, et al. Does ligamentous laxity protect against chondral and osteochondral injuries in patients with patellofemoral instability? *Orthop J Sports Med* 2022;10.

11. Thompson P, Metcalfe AJ. Current concepts in the surgical management of patellar instability. *Knee* 2019;26:1171-1181.
12. Surendran S. Patellar instability—Changing beliefs and current trends. *J Orthop* 2014;11:153-156.
13. Erickson BJ, Mascarenhas R, Sayegh ET, et al. Does operative treatment of first-time patellar dislocations lead to increased patellofemoral stability? A systematic review of overlapping meta-analyses. *Arthroscopy* 2015;31:207-1215.
14. Migliorini F, Trivellas A, Eschweiler J, Knobe M, Tingart M, Maffulli N. Comparable outcome for autografts and allografts in primary medial patellofemoral ligament reconstruction for patellofemoral instability: Systematic review and meta-analysis. *Knee Surg Sports Traumatol Arthrosc* 2022;30:1282-1291.
15. Rombaut L, Malfait F, De Wandele I, et al. Muscle mass, muscle strength, functional performance, and physical impairment in women with the hypermobility type of Ehlers-Danlos syndrome. *Arthritis Care Res* 2012;64:1584-1592.
16. Imerci A, McDonald TC, Rogers KJ, Thacker MM, Atanda A. Outcomes of medial patellofemoral ligament reconstruction and tibial tubercle osteotomy in syndromic adolescents with patellar dislocation. *J Clin Orthop Trauma* 2022;25:101770.
17. Howells NR, Eldridge JD. Medial patellofemoral ligament reconstruction for patellar instability in patients with hypermobility: A case control study. *J Bone Joint Surg Br* 2012;94-B:1655-1659.
18. Voskuijl T, Hageman M, Ring D. Higher Charlson Comorbidity Index scores are associated with readmission after orthopaedic surgery. *Clin Orthop* 2014;472:1638-1644.
19. Biedert RM. Patella alta: When to correct and impact on other anatomic risk factors for patellofemoral instability. *Clin Sports Med* 2022;41:65-76.
20. Imhoff FB, Cotic M, Dyrna FGE, et al. Dynamic Q-angle is increased in patients with chronic patellofemoral instability and correlates positively with femoral torsion. *Knee Surg Sports Traumatol Arthrosc* 2021;29:1224-1231.
21. Maas KJ, Warncke ML, Leiderer M, et al. Diagnostic imaging of patellofemoral instability. *ROFO Fortschr Geb Rontgenstr Nuklearmed* 2021;193:1019-1033.
22. Tan SHS, Chng KSJ, Lim BY, et al. The difference between cartilaginous and bony sulcus angles for patients with or without patellofemoral instability: A systematic review and meta-analysis. *J Knee Surg* 2020;33:235-241.
23. Pacey V, Nicholson LL, Adams RD, Munn J, Munns CF. Generalized joint hypermobility and risk of lower limb joint injury during sport: A systematic review with meta-analysis. *Am J Sports Med* 2010;38:1487-1497.
24. Hiemstra LA, Kerslake S, Kupfer N, Lafave MR. Generalized joint hypermobility does not influence clinical outcomes following isolated MPFL reconstruction for patellofemoral instability. *Knee Surg Sports Traumatol Arthrosc* 2019;27:3660-3667.
25. Hiemstra LA, Kerslake S, Loewen M, Lafave M. Effect of trochlear dysplasia on outcomes after isolated soft tissue stabilization for patellar instability. *Am J Sports Med* 2016;44:1515-1523.
26. Reddy G, Hayer PS, UlIslam S, et al. Outcomes of allograft medial patellofemoral ligament reconstruction in children and adolescents with hypermobility. *Int J Appl Basic Med Res* 2022;12:161-166.
27. Hohn E, Pandya NK. Does the utilization of allograft tissue in medial patellofemoral ligament reconstruction in pediatric and adolescent patients restore patellar stability? *Clin Orthop* 2017;475:1563-1569.
28. Flanigan DC, Shemory S, Lundy N, Stitgen M, Long JM, Magnussen RA. Medial patellofemoral ligament reconstruction with allograft versus autograft tissue results in similar recurrent dislocation risk and patient-reported outcomes. *Knee Surg Sports Traumatol Arthrosc* 2020;28:2099-2104.
29. Husen M, Milbrandt TA, Shah V, Krych AJ, Stuart MJ, Saris DBF. Medial patellofemoral ligament reconstruction using allografts in skeletally immature patients. *Am J Sports Med* 2023;51:1513-1524.
30. Tofts LJ, Elliott EJ, Munns C, Pacey V, Sillence DO. The differential diagnosis of children with joint hypermobility: A review of the literature. *Pediatr Rheumatol* 2009;7:1.
31. Terry RH, Palmer ST, Rimes KA, Clark CJ, Simmonds JV, Horwood JP. Living with joint hypermobility syndrome: Patient experiences of diagnosis, referral and self-care. *Fam Pract* 2015;32:354-358.

Appendix Table 1. List of Diagnosis and Procedure Codes for Patellar Instability and Stabilizing Surgical Procedures, Respectively

Diagnosis/Procedure	Code(s)
Patellar instability	Dislocation codes = {ICD-9-D-8363, ICD-10-D-S83014D, ICD-10-D-S83014S, ICD-10-D-S83004A, ICD-10-D-S83004D, ICD-10-D-S83004S, ICD-10-D-S83005A, ICD-10-D-S83005D, ICD-10-D-S83005S, ICD-10-D-S83006A, ICD-10-D-S83006D, ICD-10-D-S83006S, ICD-10-D-S83014A, ICD-10-D-S83015A, ICD-10-D-S83015D, ICD-10-D-S83015S, ICD-10-D-S83016A, ICD-10-D-S83016D, ICD-10-D-S83016S, ICD-10-D-S83094A, ICD-10-D-S83094D, ICD-10-D-S83094S, ICD-10-D-S83095A, ICD-10-D-S83095D, ICD-10-D-S83095S, ICD-10-D-S83096A, ICD-10-D-S83096D, ICD-10-D-S83096S}
	Subluxation codes = {ICD-10-D-S83001A, ICD-10-D-S83001D, ICD-10-D-S83001S, ICD-10-D-S83002A, ICD-10-D-S83002D, ICD-10-D-S83002S, ICD-10-D-S83003A, ICD-10-D-S83003D, ICD-10-D-S83003S, ICD-10-D-S83011A, ICD-10-D-S83011D, ICD-10-D-S83011S, ICD-10-D-S83012A, ICD-10-D-S83012D, ICD-10-D-S83012S, ICD-10-D-S83013A, ICD-10-D-S83013D, ICD-10-D-S83013S, ICD-10-D-S83091A, ICD-10-D-S83091D, ICD-10-D-S83091S, ICD-10-D-S83092A, ICD-10-D-S83092D, ICD-10-D-S83092S, ICD-10-D-S83093A, ICD-10-D-S83093D, ICD-10-D-S83093S}
Surgical procedure for patellar instability	CPT-27420 − Reconstruction of dislocating patella; (e.g., Hauser type procedure)
	CPT-27422 − Reconstruction of dislocating patella; with extensor realignment and/or muscle advancement or release (e.g., Campbell, Goldwaite type procedure)
	CPT-27427 − Ligamentous reconstruction (augmentation), knee; extra-articular
	CPT-27418 − Anterior tibial tubercleplasty (e.g., Maquet type procedure)

CPT, Current Procedural Terminology; ICD, *International Classification of Diseases*.