

Analyzing risk factors for recurrence of developmental coxa vara after surgery

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Abstract

Purpose To evaluate the risk factors for developmental coxa vara (DCV) recurrence following valgus osteotomy of the proximal femur.

Methods We retrospectively reviewed records of 32 DCV patients (46 hips) treated surgically (2005 to 2012). Recurrence-related factors, including age at initial surgery, side, sex, fixation methods, diagnosis of coxa vara, premature capital femoral physeal closure and postoperative Hilgenreiner epiphyseal (HE) angle, head-shaft (HS) angle, medial femoral offset and posterior slope angle (PSA) were analyzed.

Results At 4.7-year mean follow-up, 12 hip deformities recurred (26%). Postoperative HE angle > 41° and negative offset were statistically significant univariate and multivariate risk factors for the deformity recurrence. Increased PSA was common preoperatively, which accounted for 59% of hips. Postoperative PSA > 20° was associated with a high recurrence rate in the univariate analysis. Age was another univariate risk factor for the recurrence. Recurrence rate was 52% in the < 6.5-year age group versus 4% in the > 6.5-year age group. Other factors were not statistically significantly related to recurrence.

Conclusion DCV is a 3D deformity. To prevent recurrence, HE angle should be restored to < 41° in the coronal plane. Sagittal malalignment (abnormal PSA) should be corrected concurrently, so that, the direction of surgical correction is along the true deformity plane. During valgus osteotomy, the distal fragment should be lateralized to maintain a normal mechanical axis.

Level of Evidence: IV

Cite this article: Bian Z, Xu YJ, Guo Y, Fu G, Lyu XM, Wang QQ. Analyzing risk factors for recurrence of developmental coxa vara after surgery. *J Child Orthop* 2019;13:361-370. DOI: 10.1302/1863-2548.13.180201

Keywords: coxa vara; valgus osteotomy; femoral offset; posterior slope angle; Heuter-Volkman law; recurrence

Introduction

Coxa vara is defined as a neck-shaft (NS) angle of < 110°.¹ Herring² had classified it into three groups: developmental coxa vara (DCV), coxa vara with congenital femoral deficiency and acquired coxa vara. DCV includes the isolated form and coxa vara associated with skeletal dysplasia. It is a relatively rare paediatric hip deformity. Patients often present with gait abnormalities and limb-length discrepancy (LLD). Surgical treatment with valgus osteotomy is indicated if the Hilgenreiner epiphyseal (HE) angle is > 60° or if the deformity progresses.¹

Proximal femoral valgus osteotomy has become an established procedure for the treatment of coxa vara. The aim of the surgical treatment is to restore the normal anatomy, thereby improving the mechanics and function of the hip joint. Generally, the HE angle should be corrected to < 35° to 40°,^{1,3,4} converting the poorly tolerated shear forces to more physiological and desirable compressive forces. Good results had been reported using Pauwels' Y-shaped osteotomy³ or Borden subtrochanteric end-to-side osteotomy⁵ or with modern paediatric hip plate (PHP) fixation.⁶ Nevertheless, these studies had either a small number of cases or short follow-up, and most studies included heterogeneous causes of coxa vara, including the acquired type. However, in some studies, the recurrent rate is still high, which is up to 50% to 75%.^{4,7} Moreover, most studies only identified that good correction of the HE angle is a single important factor related to recurrence.^{1,3,4,8,9}

As DCV is a 3D deformity, it occurs not only in the coronal plane but also in the sagittal and axial transverse planes.¹⁰ From the lateral view of the hip, the femoral epiphysis normally has mean 5° of retroversion, which is defined as the posterior slope angle (PSA).¹¹ With increase in PSA, there is an increase in the incidence of slipped epiphysis. Kim et al¹⁰ used 3D and 2D CT to demonstrate the presence of a marked femoral retroversion component, which often occurs in severe DCV.

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In the coronal plane, another radiological parameter that may be changed after the operation is the mechanical axis. Mechanical axis deviation (MAD) can affect the hip, knee and ankle joints.¹² For the knee joint, Shim et al¹³ noticed that genu valgum may subtly worsen over time in a growing child because of lateralization of the lower-extremity mechanical axis with respect to the knee joint, with the resulting abnormal Hueter-Volkman forces across the physis causing progressive genu valgum. For the hip joint, medialization of the femur may change the direction of the lower-extremity mechanical axis. It will change the direction and pressure of the proximal femoral physis, which may be related to the recurrence of the deformity.

The purpose of this retrospective study was to determine the risk factors of DCV recurrence by analysis of the clinical and radiological parameters. Our hypothesis was that deformity recurrence is related not only to HE angle but also to the PSA and medialization of the femur.

Patients and methods

We performed a retrospective review all of the coxa vara cases treated by valgus osteotomy at BeijingJiShuiTan hospital between 2005 and 2012. The inclusion criteria were: 1) patients with diagnosis of DCV – acquired coxa vara (developmental dysplasia of the hip, traumatic, pathology and slipped capital femoral epiphysis (SCFE)), which had diverse aetiologies, were excluded and coxa vara with congenital femoral deficiency was also excluded since the vara deformity was mainly at the neck shaft junction level; 2) patients who had at least two years of growth potential (male < 13 years; female < 11 years), so the proximal growth plate is still opening, which is the precondition for recurrence of the deformity.

A total of 32 patients were identified for inclusion (18 boys and 14 girls), 14 of whom had bilateral involvement. A total of 46 hips underwent valgus osteotomy. The mean age of patients was 7.3 years (3 to 13). In all, 26 patients (35 hips) had the infantile (isolated) type and six (11 hips) patients had coxa vara associate skeletal dysplasia (two multiple epiphyseal dysplasia, one cleidocranial dysplasia, two spondylosis epiphyseal dysplasia, one metaphyseal chondrodysplasia).

Valgus osteotomy was performed at the intertrochanteric or subtrochanteric level. Adductor tenotomy was routinely performed during the operation. Before 2010, several fixation methods were used in 25 hips, including single screw (two hips), dynamic compression plate (11 hips), T shape plate (eight hips), blade plate (three hips), and Kirschner-wire (one hip). After 2010, PHP was used as the standard fixation method (24 hips, including three revision cases). Femoral rotational component osteotomies were performed in two hips to correct the abnormal femoral anteversion angle.

Anteroposterior (AP) and frog-lateral radiographs of hips were used to evaluate the deformity preoperatively, postoperatively and during follow-up. Measurements (Fig. 1) included: 1) HE angle, defined as the angle between Hilgenreiner line and epiphyseal line in the AP view; 2) PSA, defined as the angle between the line parallel with the longitudinal neck-diaphyseal axis and the line perpendicular with the epiphysis in the lateral view or CT scan (normal < 10°); 3) head-shaft (HS) angle, defined as the angle between a line perpendicular to the proximal femoral epiphysis and a line through the middle of the femoral shaft (HS angle was used as a parameter instead of NS angle, because the NS angle may not adequately reflect the amount of deformity or prognosis for spontaneous progression¹); 4) femoral offset was used to evaluate the medialisation of the femur, which is defined as the perpendicular distance between the long axis of the femur and the centre of the femoral head in the AP view. It was regarded negative if the axis of the femoral shaft was medial to the centre of the femoral epiphysis. All radiographs were digitized and measurements were performed manually by two independent observers (Z.B. and X.M.L.) on Picture Archive and Communication System images. The results show good interobserver reliability using intraclass correlation (ICC) analysis (the ICC values were 0.98 (preoperatively)/0.97 (postoperatively) for HE angle, 0.98/0.97 for HS angle and 0.96/0.89 for PSA). The mean was taken as the final value.

An operative technique was recommended when using PHP (Fig. 2) as follow. 1) Choose an appropriate PHP – the 140° and 150° plate were most commonly used for valgus osteotomy – it will be helpful by mimicking the operation using a transparent tracing paper. 2) Calculate the positioning wire angle, which will determine the final degree of correction. First, the correction angle should be calculated with the aim of the HE angle to be less than 20° to 30°. Then, the angle between the positioning wire and the femoral shaft can be calculated by the screw angle minus the correction angle. 3) Where to put the positioning wire? In the AP view, the wire should be in the femoral neck as proximal as possible, so that osteotomy can be closed to the center of rotation of angulation (neck head junction) at the intertrochanteric level and distal medialization of the femur can be effectively avoided. In the lateral view, the wire should be in the middle of the neck and perpendicular to the physis. Then, the abnormal PSA can be corrected after operation with the extension or flexion component, so that the direction of correction is along the true deformity plane (maybe an oblique plane) other than the simple coronal plane.

Recurrence was defined as either worsening HE angle or progression to > 60°. If the proximal femur physeal was closed before age 12 years, it was regarded as premature

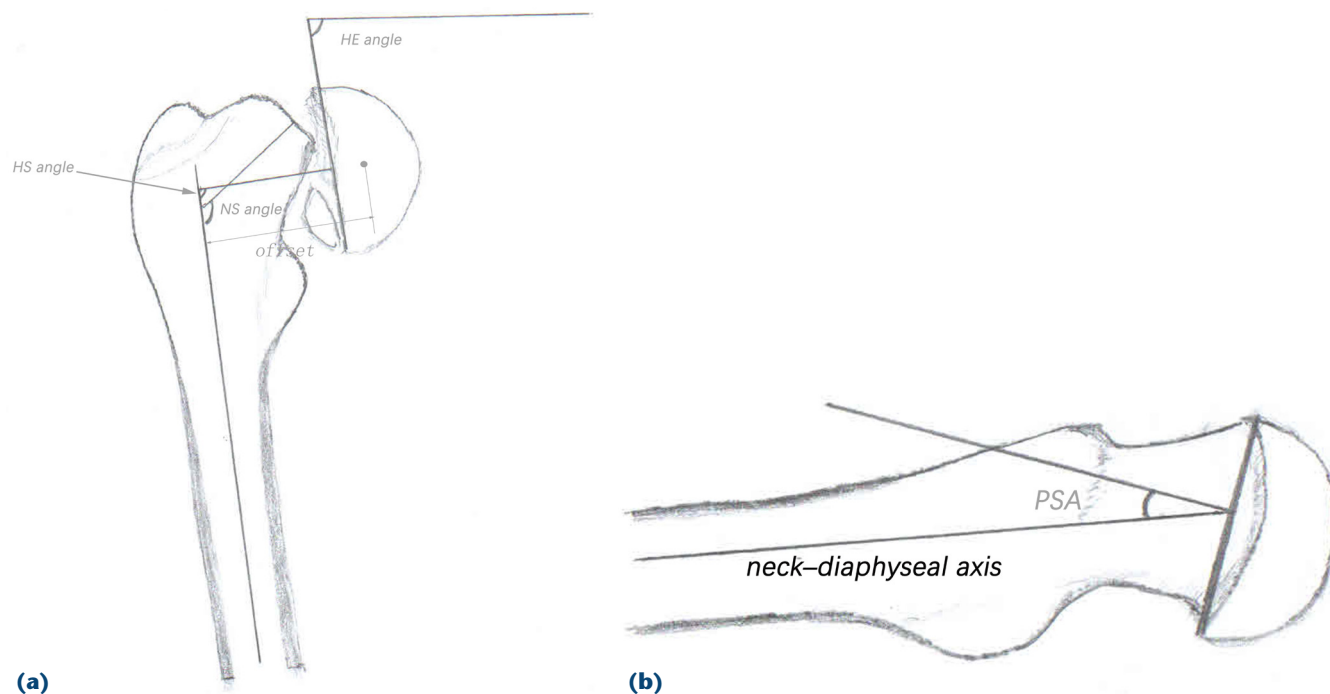


Fig. 1 Radiologic parameters: Hilgenreiner epiphysal (HE) angle, head-shaft (HS) angle, neck-shaft (NS) angle and femoral offset in anteroposterior view **(a)** and posterior slope angle (PSA) in frog lateral view **(b)**.

closure. Clinical assessment included evaluation of pain, range of movement and LLD.

Statistical analysis

Data were presented as means (sd) for continuous variables and as frequencies for categorical variables. Simple linear regression was used to evaluate the linear relationship between the HS angle and HE angle, and paired t-test or Wilcoxon signed-rank test was used for comparing the differences of angles pre- and postoperatively.

The receiver operating characteristic (ROC) curve was used to determine the optimal cutoff value corresponding to the maximum Youden index, including age, postoperative HA angle, HE angle and PSA, which were divided to two groups by the cutoff value. First, a univariate binary logistic regression analysis was performed between the non-recurrence and recurrence hips to determine the significance of the differences with regard to sex, affected side, diagnosis, age at surgery, fixation methods, premature closure, postoperative HS angle, HE angle and PSA and offset. Secondly, the radiological deformity parameters, which include coronal plane parameter, such as HE angle and offset, and the sagittal plane parameter, such as PSA, were analyzed by logistic multivariate regression analysis. All tests were two-sided, and a p-value < 0.05 was considered significant. SPSS version 21.0 (IBM, Armonk, New York) was used for all statistical analyses.

Results

The HE and HS angles were significantly improved after the operation (Table 1). The HE and HS angles had a linear relationship both preoperatively and postoperatively (Fig. 3). The mean follow-up time was 4.7 years (2 to 13). In all, 12 hip (26%) deformities recurred. All recurrences take place at around two to four years postoperatively, and most of them can demonstrate a change in the HE angle about one-year postoperatively. The mean follow-up time in the recurrence group was 6.1 years (2 years to 13 years) compared with 4.2 years (2 years to 12.7 years) in the non-recurrence group, and there was no statistical difference ($p = 0.076$). Revision surgeries were performed in nine hips.

Univariate analyses show that recurrence was not related to the diagnosis even if it was an infantile isolated type or coxa vara associated with skeletal dysplasia, not related to the fixation methods whether by PHP or other fixation methods and not related to the affected side and sex. On univariate analyses, postoperative HS angle, HE angle, PSA and offset were all significantly related to recurrence, and their cutoff values were 142°, 41° and 20°, respectively, which were calculated by the ROC curve corresponding to the maximum Youden index (Table 2).

Age was found to be related to recurrence in the univariate analysis ($p < 0.001$). The recurrence rate was 52% (11/21) in the < 6.5-year age group compared with the 4%

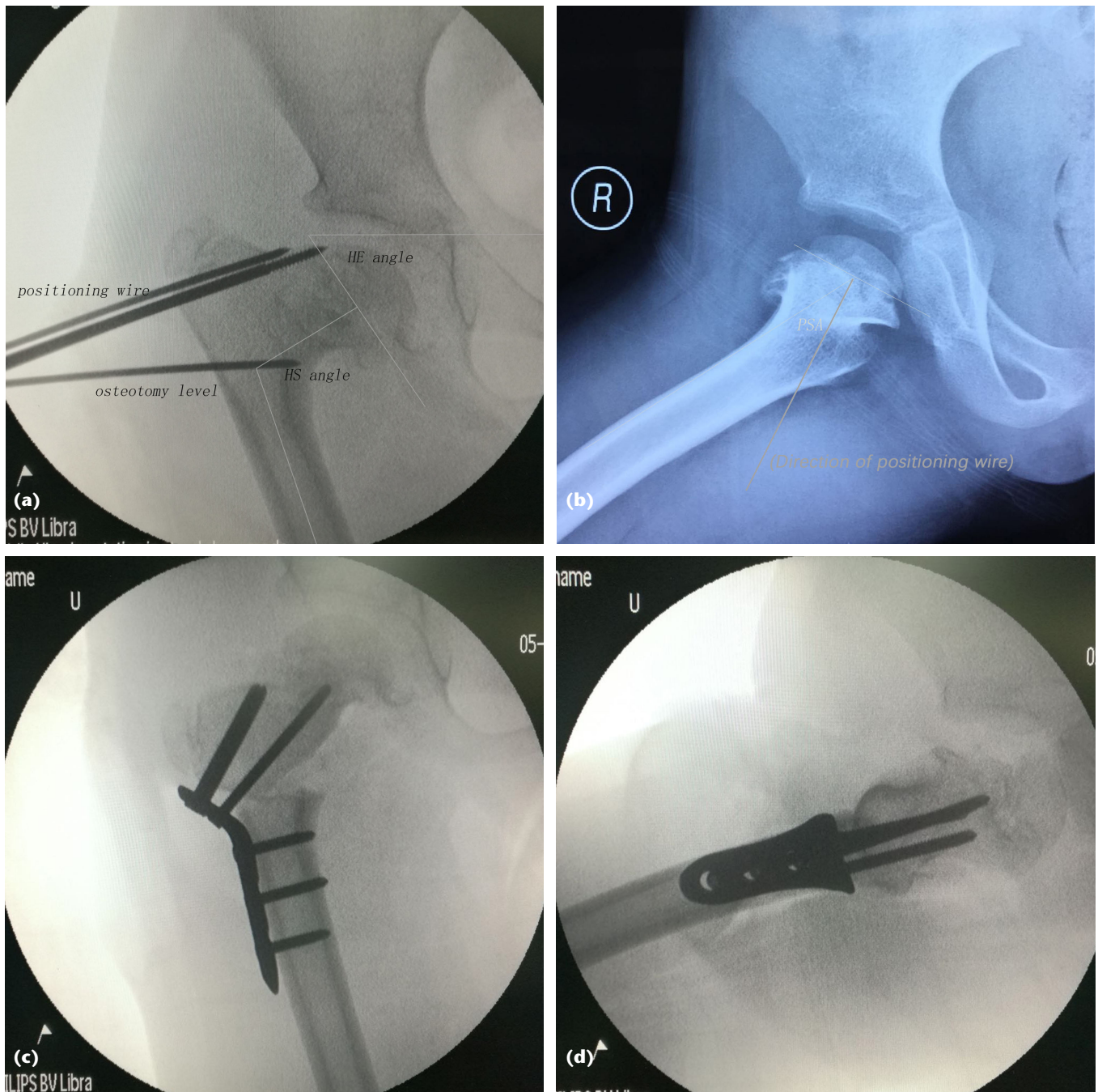


Fig. 2 Eight-year-old girl with developmental coxa vara treated with valgus osteotomy and paediatric hip plate fixation: **(a)** proximal positioning wire determines the degree of correction and position of plate, distal wire marks the intertrochanteric level of osteotomy, Hilgenreiner epiphyseal (HE) angle: 56° , head-shaft (HS) angle: 102° , 'metaphyseal blanch sign' and 'reduced height of the epiphysis' can be seen, which indicated abnormal posterior slope angle (PSA); **(b)** PSA: -31° preoperatively; **(c)** after valgus osteotomy, correct the HE angle to 24° , the height of epiphysis had been restored after corrected the PSA, and the femoral offset was maintained by lateralization of distal fragment; **(d)** restore the normal PSA.

(1/25) in the > 6.5 -year age group. The rate of premature capital femoral physal closure was high, which occurred in 23 hips (50%). The mean age at closure was 9.5 years (6.5 to 12). The premature physis may have prevented

the recurrence of the deformity. Age was also related to postoperative HE angle ($p = 0.03$), which means that undercorrection was more common in the younger age group.

Corona plane parameters

In the multivariate logistic regression analysis of three radiological deformity parameters, postoperative HE angle and femoral offset were found significant related to recurrence ($p = 0.003$ and $p = 0.007$). In the HE angle $> 41^\circ$ group, the recurrence rate was 48% compared with 15% in the $< 41^\circ$ group. Postoperatively, the femoral offset became negative in four hips, in which three hip deformities recurred compared with the 21% in the femoral offset positive group, showing significance (Fig. 4).

Sagittal and axial plane parameter

Preoperatively, PSA increased in more than half of the vara hips (59%) (12° to 56° , epiphysis retroversion), PSA decreased in two hips (epiphysis anteversion) and the other 17 hips had normal PSA value (0° to 10°). Postoperatively, in the univariate analysis, the PSA $> 20^\circ$ group was

associated with a high recurrence rate ($p = 0.021$) (Fig. 5), and it was also associated with the undercorrection of the HE angle ($p = 0.01$). Thus, in the multivariate analysis, it was not a significant risk factor ($p = 0.172$).

At the final follow-up, most patients had normal range of hip movement. Six patients (eight hips) had moderate restriction in the internal rotation of the hip joint. Six had obvious LLD, which was > 2 cm on clinical examination. An early sign of osteoarthritis was found in one patient.

Table 1 The change of Hilgenreiner epiphyseal (HE) angle and head-shaft (HS) angle pre- and postoperatively

	Preoperative	Postoperative	t	p-value*
HS angle	105.1 (SD 14.4)	147.6 (SD 13.2)	-16.42	< 0.001
Pre- to postoperative	-42.5 (SD 17.6)			
HE angle	68.8 (SD 14.8)	29.8 (SD 12.1)	16.235	< 0.001
Pre- to postoperative	39.0 (SD 16.3)			

*paired t-test

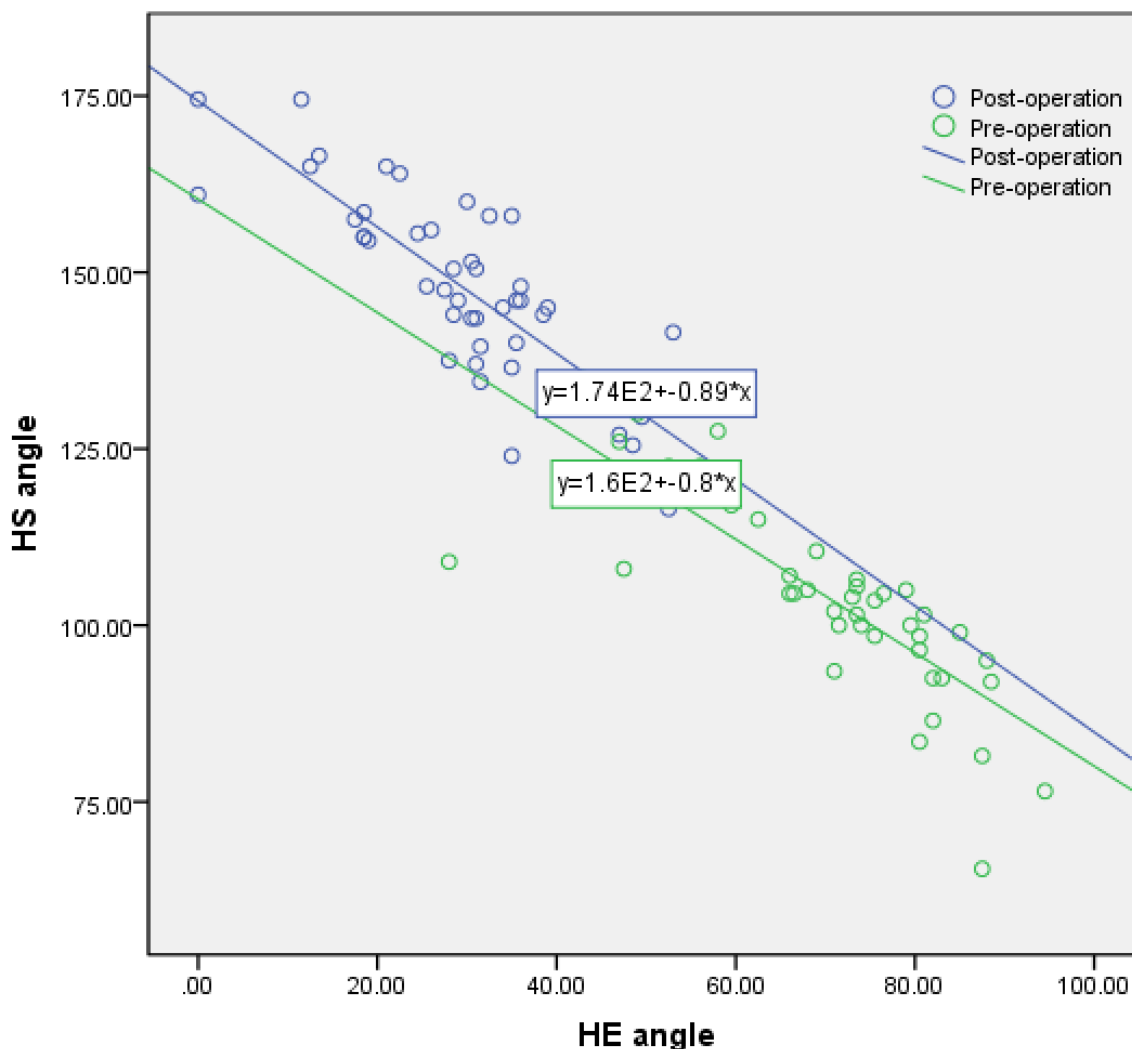


Fig. 3 Linear relationship of Hilgenreiner epiphyseal (HE) angle and head-shaft (HS) angle and changes postoperatively.

Table 2 Analyzing risk factors for recurrence of developmental coxa vara

	Recurrence number/total number		Univariate analysis p-value*	Multi variate analysis		
				β	OR (95% CI)	p-value**
Age (< 6.5 yrs/ \geq 6.5 yrs)	11/21	1/25	< 0.001	Not selected		
Sex (F/M)	6/23	6/23	1	Not selected		
Side (R/L)	5/21	7/25	0.747	Not selected		
Fixation (PHP/others)	5/21	7/25	0.747	Not selected		
Diagnosis (isolated/skeletal dysplasia)	10/35	2/11	0.494	Not selected		
Premature closure (yes/no)	4/23	8/23	0.186	Not selected		
HS angle (< 142°/ \geq 142°)	8/14	4/32	0.003	Not selected		
HE angle (< 41°/ \geq 41°)	6/39	6/7	0.003	3.790	44 (3.6 to 538)	0.003
PSA (< 20°/ \geq 20°)	5/32	7/14	0.021	1.414	4.1 (0.54 to 31)	0.172
Femoral offset (positive/negative)	9/42	3/4	0.048	3.695	10 (2.7 to 602)	0.007

OR, odds ratio; CI, confidence interval; PHP, paediatric hip plate; HS, head-shaft; HE, Hilgenreiner epiphyseal; PSA, posterior slope angle

*chi-square test

**Multivariate logistic regression

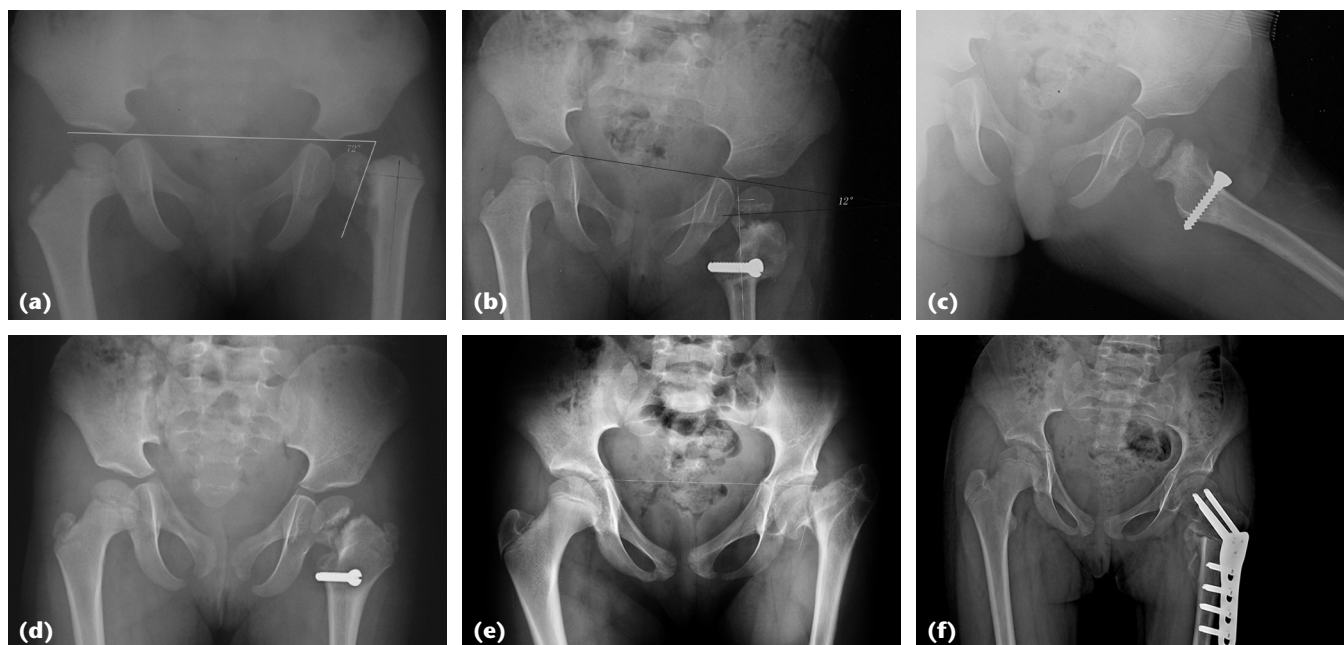


Fig. 4 Three-year-old girl, left developmental coxa vara: **(a)** Hilgenreiner epiphyseal (HE) angle: 72°; **(b)** valgus osteotomy was performed at intertrochanteric level: HE angle was fully corrected to 12°, however, the axis of femoral shaft was obvious medial to the centre of femoral epiphysis; **(c)** lateral view shows normal posterior slope angle; **(d)** five years, deformity progressively worsened; **(e)** 12 years, deformity recurrence, HE angle: 76°; **(f)** revision surgery was performed with paediatric hip plate fixation.

Discussion

Paediatric orthopaedics doctors may sometimes encounter recurrence when treating DCV. To our knowledge, this is the largest case series including 32 DCV patients (46 hips) treated with surgery and multiple factors were found related to recurrence. HE angle was the most important deformity parameter which should be corrected during surgery. However, even if the HE angle was fully corrected, a high number of cases still had a recurrence rate of 15% (six of 39 hips). Hence, other factors may have contributed to the recurrence in these cases. Coxa vara is a 3D deformity. The HE angle is a coronal

plane parameter. PSA, a sagittal and axial plane parameter, has been usually underestimated. The normal value of PSA was 5°. With increase in the posterior sloping angle, there is an increase in the incidence of slipped epiphysis. If PSA was > 12°, it had a high risk of slip, and prophylactic fixation was the recommended treatment of SCFE.¹¹ In our study, we find a high rate of increased PSA preoperatively (59%). Thus, the lateral view or CT scan is very important to find the axial plane deformity of coxa vara. In case of abnormal PSA, the radiographic characteristics of SCFE can be seen in the AP view for coxa vara, such as 'metaphyseal blanch sign' and 'reduced height of the epiphysis' (Fig. 2). Hence, in cases of increased PSA, valgus-flexion

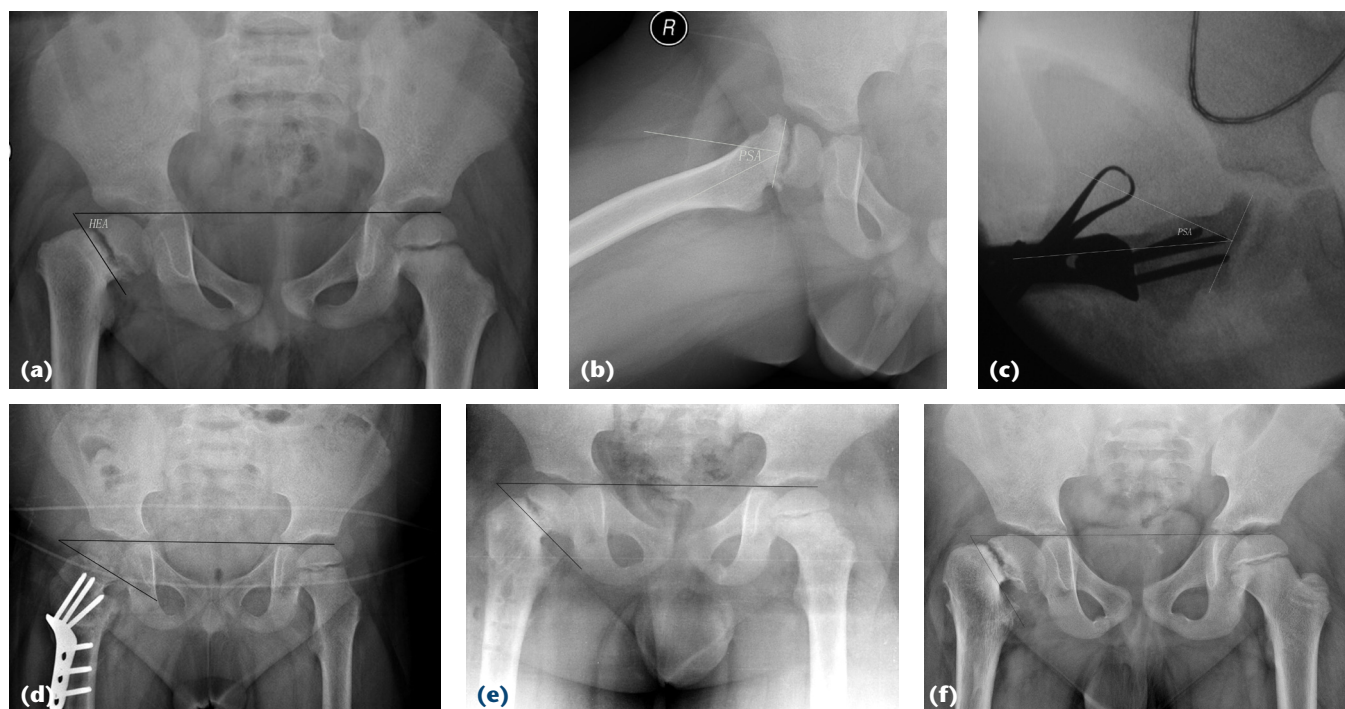


Fig. 5 Four-year-old boy, right developmental coxa vara: **(a)** Hilgenreiner epiphyseal (HE) angle: 61°; **(b)** posterior slope angle (PSA): 36°; **(c)** postoperatively, PSA was not corrected; **(d)** HE angle: 30° in anteroposterior view; **(e)** one-year postoperatively, deformity progressive worsens, HE angle: 44°; **(f)** four years postoperatively, deformity, recurrence HE angle: 60°.

type osteotomy is indicated to correct the PSA as well as the HE angle.

During valgus osteotomy, the HE angle was the most important parameter to evaluate the coronal plane deformity. Most studies had identified its important role with recurrence. Weinstein et al¹ first noted that the HE angle other than the NS angle could be more predictive of the natural history of coxa vara. The normal HE value was 16° (0° to 25°), and the goal of surgical treatment should be an HE angle of < 45°. Desai and Johnson⁸ reported that in their series of 12 patients (20 hips), a postoperative HE angle of ≤ 35° gave consistently satisfactory results without recurrence. Cordes et al³ reported 14 patients (18 hips) with ten years of follow-up. An excellent result (67%) could be expected in hips in which the HE angle was corrected to ≤ 40°, irrespective of the cause of coxa vara. Similarly, Carroll et al⁴ reported 26 patients (37 hips) with a recurrence rate of 50%, and Burns and Stevens⁹ reported nine (11 hips) patients with a recurrence rate of 16%. Both studies emphasized the importance of obtaining an intraoperative correction of HE angle to < 38°. Our study also supports this view. Both HE angle and HS angle can be used as postoperative evaluation parameters.

Another factor that correlated with recurrence was medialization of the femur. As a principle of angulation deformity correction, the osteotomy site is best at the

centre of the rotation angle. If the osteotomy is performed at a level proximal or distal to the apex and if not properly translated, it may lead to MAD. Subsequently, translation in addition to angulation is necessary to accurately correct the deformity¹² (Fig. 6). The mechanical axis of the lower limb should normally pass through the midpoint of the femoral head, knee, and ankle. However, after valgus osteotomy at the subtrochanteric level, lateral MAD with respect to the knee joint may occur if there was no lateral translation of the distal femoral shaft. Shim et al¹³ noted that excessive lateralization of the mechanical axis exacerbated occult genu valgum in three cases after valgus osteotomy of coxa vara.

For the hip joint, the medialization of the femur will change the direction of lower extremity mechanical axis (medial tilt). The new mechanical axis will initially pass the medial side of the capital physis, which increases the pressure of the medial physis. The Hueter-Volkman law¹⁴ stated that compressive forces inhibit growth and tension stimulates it. Thus, the growth of the medial physis may be inhibited. With the body weight gradually loading, the mechanical axis tends to move to the centre of the head in a more stable status, which will further cause the medial tilt of the capital physis (increase the HE angle). Both effects will increase the risk of deformity recurrence (Fig. 7). Our study showed that if there was a negative

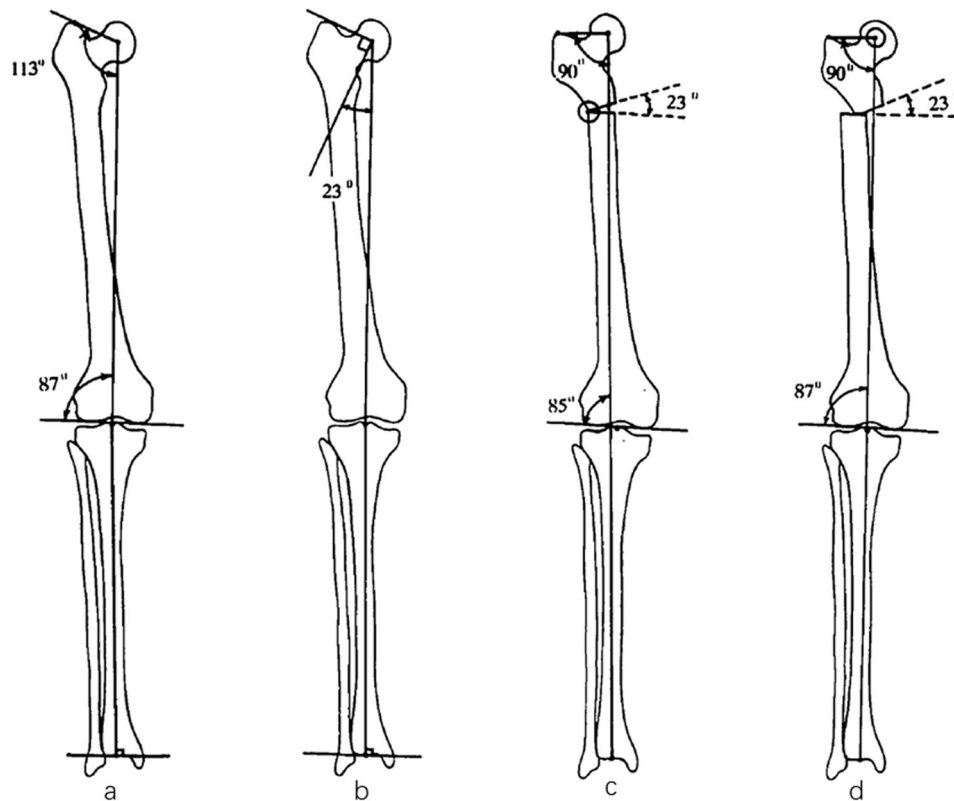


Fig. 6 Mechanical axis deviation during valgus osteotomy: **(a)** the mechanical axis is colinear between the hip, knee and ankle; **(b)** the joint orientation line at hip demonstrates a 23° varus deformity; **(c)** a simple opening or closing wedging correction at level of subtrochanteric produces a lateral translation deformity with valgus mechanical axis malalignment and malorientation of the hip and knee joint; **(d)** therefore, a valgus correction of the hip should be combined with lateral translation.

femoral offset, there would be a high recurrence rate at 75% (three of four hips).

Age at surgery has been hypothesized to influence surgical outcomes both negatively and positively. Carroll et al⁴ stated that age was not predictive of long-term correction but the best function outcomes were obtained in patients younger than age ten years. However, Weinstein et al¹ noted that 75% of the hips maintained over 80% of their surgical correction when surgery was performed after five years. Amstutz and Wilson¹⁵ suggested that the age at the time of index surgery affected the rate of recurrence, with younger children having an increased incidence of additional corrective surgery requirement. In our study, most cases with recurrence were in the younger age group (< 6.5 years). The reason may be multifactorial; except for the disease itself, under-correction was more common in younger age patients in our study. There is also a high rate of premature physeal closure, which may be another reason for the lower recurrence rate in the older age group. A similar finding was reported by Schmidt¹⁶ with 89% of hips having premature closure after valgus osteotomy.

Fixation method is another important issue during surgery. Recurrence was not related to the fixation method

if it is PHP fixation or other fixation. However, PHP was a convenient, rigid fixation method with locking system. In our practice, it is our standard fixation method. What should be noted during surgery is that all the deformity parameters should be appropriately addressed. Otherwise recurrence may occur regardless of the implant used.

The present study has some limitations. First, it is a retrospective, less than five-year mean follow-up study. Although two-year follow-up can pick up most of recurrence cases, some cases still need longer follow-up to observe if deformity will recur. Secondly, we did not record gait and detailed function outcome. Roberts et al¹⁷ found that a 72% rate of abnormal gait and 50% rate of fair-poor function if a longer follow-up was done. This may need to be added in a future study to provide more comprehensive evaluation of patients.

In conclusion, we have presented the largest case series of surgically treated DCV including 32 patients (46 hips). Under-correction (HE angle > 41°) and negative femoral offset was the risk factors of recurrence. Since DCV is a 3D deformity, to prevent recurrence, the HE angle should be restored in the coronal plane. Sagittal malalignment

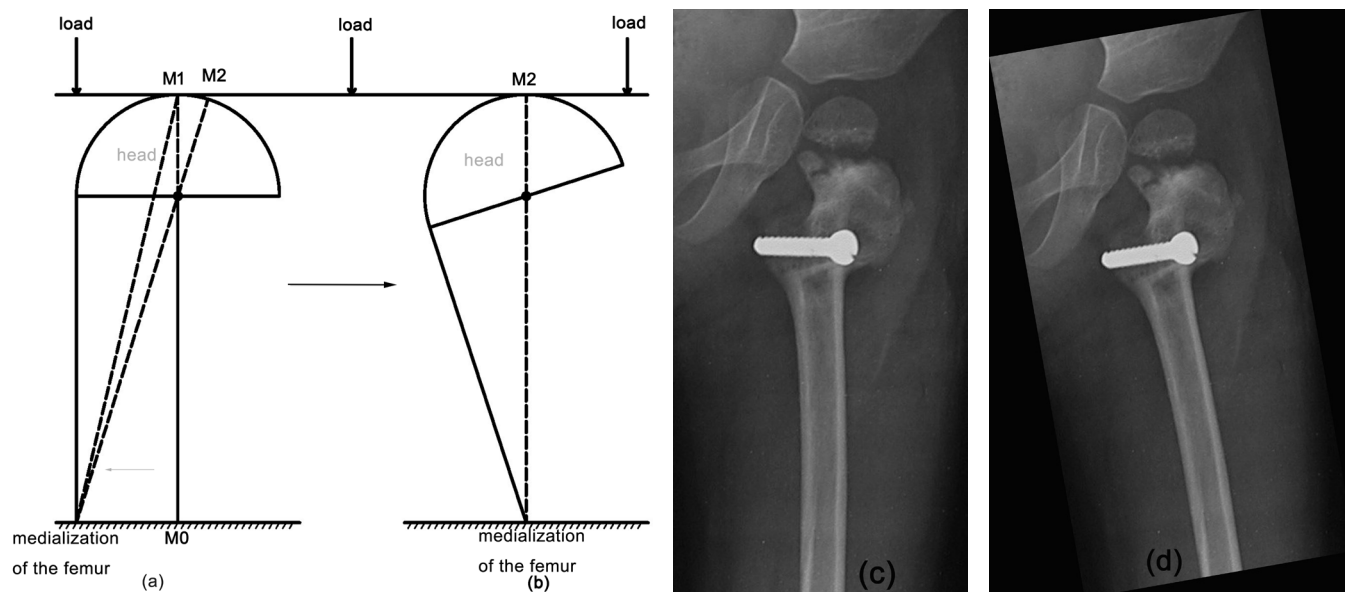


Fig. 7 Mechanical axis changes after medialization of the femur. **(a)** M0 is normal lower limb mechanical axis. The body weight passes the centre of head and perpendicular to the ground. M1 is a new mechanical axis when medialization of distal femur, which passes the medial part of physis (increase the medial pressure). **(b)** M1 tends to move to M2, which passes the centre of head in more stable status (increase the tilt of physis). **(c)** and **(d)** Demonstrated by real patient's X-ray.

(abnormal PSA) should be corrected concurrently, so that the direction of surgical correction is along the true deformity plane. During valgus osteotomy, the distal fragment should avoid medialization to maintain a normal mechanical axis. These techniques can be used as guidelines to improve surgical outcome and avoid recurrence.

Received 13 December 2018; accepted after revision 20 June 2019.

COMPLIANCE WITH ETHICAL STANDARDS

FUNDING STATEMENT

This study was funded by Beijing Hospitals Authority Youth Programme, code: QML20180401.

OA LICENCE TEXT

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ETHICAL STATEMENT

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Not required for this type of study.

ICMJE CONFLICT OF INTEREST STATEMENT

None declared.

AUTHOR CONTRIBUTIONS

ZB: Manuscript preparation, Performed measurements.

YJX: Study design.

YG: Cases collection, Study design.

GF: Study design, Manuscript preparation.

XML: Cases collection, Study design, Performed measurements.

QQW: Statistical analysis

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