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Long-term follow-up of Van Nes rotationplasty in patients with congenital proximal focal femoral deficiency

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Abstract

Van Nes rotationplasty may be used for patients with congenital proximal focal femoral deficiency (PFFD). The lower limb is rotated to use the ankle and foot as a functional knee joint within a prosthesis. A small series of cases was investigated to determine the long-term outcome. At a mean of 21.5 years (11 to 45) after their rotationplasty, a total of 12 prosthetic patients completed the Short-Form (SF)-36, Faces Pain Scale-Revised, Harris hip score, Oswestry back pain score and Prosthetic Evaluation Questionnaires, as did 12 age- and gender-matched normal control participants. A physical examination and gait analysis, computerised dynamic posturography (CDP), and timed 'Up & Go' testing was also completed. Wilcoxon Signed rank test was used to compare each PFFD patient with a matched control participant with false discovery rate of 5%.

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Supplementary material

A table detailing the results of the Prosthetic Evaluation Questionnaire for individuals who had a Van Nes rotationplasty is available with the electronic version of this article on our website www.bjj.boneandjoint.org.uk

There were no differences between the groups in overall health and well-being on the SF-36. Significant differences were seen in gait parameters in the PFFD group. Using CDP, the PFFD group had reduced symmetry in stance, and reduced end point and maximum excursions.

Patients who had undergone Van Nes rotationplasty had a high level of function and quality of life at long-term follow-up, but presented with significant differences in gait and posture compared with the control group.

Proximal focal femoral deficiency (PFFD) is a rare congenital deformity of unknown aetiology with hypoplasia or absence of the proximal femur, with an incidence of one in 50 000.¹ Patients with PFFD often have associated instability of the hip and rotational malalignment, poor hip musculature, contractures involving the hip and leg length discrepancy.^{2,3} There are many surgical and prosthetic options for patients with these complex problems. In 1950, Van Nes⁴ described a patient with congenital defects of the femur who had undergone a rotationplasty using the ankle and foot as a knee joint within a below-knee prosthesis. This procedure theoretically produces a functional joint at the level of the knee in order to allow a more normal gait pattern (Fig. 1). The procedure was later modified to include fusion of any residual knee joint,⁵ but it remains controversial because of the perceived poor cosmetic appearance with the foot facing backwards, associated difficulties in achieving a comfortable prosthesis and the possible need for subsequent rotational osteotomies as the limb might alter its rotatory alignment during growth.

There are few reports on the long-term outcome of patients who have undergone a Van Nes rotationplasty.^{2,6} These authors confined their observations to children and used gait analysis. In 1996, Fowler et al² compared the expenditure of energy during walking of nine patients who had undergone the operation with seven who had undergone a Syme amputation⁷ (both mean age of 12 years) at a minimum follow-up of two years, and found that those with a Syme amputation expended more energy. Fowler et al⁶ subsequently compared the gait mechanics at a minimum of two years of ten patients who had the Van Nes procedure (mean age 12.9 years) with nine who had undergone a Syme amputation (mean age of 15.1 years). Those with a Syme amputation were found to have more compensatory strategies on the non-prosthetic side.^{2,6} However, neither study reported activity levels or assessed quality of life (QoL).

Function, activity level and QoL have been assessed in patients who have undergone rotationplasty following the resection of a malignant tumour.⁵⁻¹⁵ In one study of gait in ten such children adverse kinetic parameters were reported when compared with controls, but the patients still functioned at a high level.¹² An adult study of similar size reported mild changes of gait and a reduced hip extension moment, but good functional results.¹⁰ It has also been reported that patients who have undergone rotationplasty after the resection of a tumour can continue to be involved in sports.¹⁶ Using measures of QoL including the Short Form (SF)-36,¹⁷ individuals who underwent rotationplasty, however, had lower physical function and body image scores.⁸ A further study reported that children who underwent resection of a tumour and rotationplasty perceived that they had no barriers to recreation, sporting or career goals and accepted their body image emotionally.¹¹

The purpose of the current study was to characterise the acceptance of rotationplasty by patients who were now adults, the presence of hip and/or back pain, range of movement (ROM) and strength of the prosthetic side, postural stability, abnormalities of gait, and overall health and well-being. We then compared those outcomes with those of age and gender matched controls. We hypothesised that the long-term QoL of individuals with PFFD who underwent a rotationplasty would be reduced compared with that of controls.

Patients and Methods

The study had ethical approval and the medical records of two community prosthetic companies were used to retrieve the contact information of 43 patients who underwent rotationplasty between 1940 and 1999 (Fig. 2). Attempts were made to contact all these patients but only 22 were found. Two were excluded as they had undergone disarticulation of the ankle as an adult. Exclusion criteria also included any further surgery of the lower limb or fracture within the past year; eight lived too far away and answered questionnaires by post. Thus, 12 patients with congenital PFFD who had undergone a Van Nes rotationplasty as a child and currently walked with a prosthesis participated fully in the study.

There were four women and eight men with a mean age at assessment of 31.6 years (16 to 57). They had undergone rotationplasty at a mean age of 6.5 years (2 to 12), and the investigation therefore represented a final follow-up of 25.1 years (11 to 45).

Radiographs (anteroposterior and lateral) of both lower limbs were taken. Based on the available radiographs taken at the time of surgery and last follow-up, the patients' hips were classified according to the Aitken classification of PFFD³: three had type A, none had type B, two had type C, four had type D and three had a congenitally short femur. A total of eight patients (67%) had a knee fusion.

A total of 12 age- and gender-matched adults (mean age 32.6 years (17 to 59)) without congenital musculoskeletal or neuromuscular deformities were enlisted as a control group.

Both groups underwent barefoot full body gait analysis using a 14 camera Vicon Motion Analysis System (Vicon, Oxford, United Kingdom). Two AMTI force plates (Advanced Mechanical Technology, Watertown, Massachusetts) were used to measure ground-reaction forces, from which joint moments and powers were calculated.

Computerised Dynamic Posturography (CDP), which isolates and assesses the sensory and motor components of standing postural control, was performed using the Neuro-Com SMART EquiTest (NeuroCom International Inc., Clackamas, Oregon). Force platforms are used to measure postural sway during the following protocols:

1. The Sensory Organisation Test (SOT), during which participants are exposed to six conditions, three trials of each that systematically target the contributions of visual, somatosensory and vestibular feedback on standing postural sway.
2. The Motor Control Test (MCT), during which participants are subjected to unexpected anterior-posterior force plate translations scaled to the participant's

height at small (2.8°/s), medium (6.0°/s), and large (8.0°/s) velocities. Using this protocol, a measure of weight-bearing symmetry was provided where a positive value indicates an increased weight-bearing compared with the non-prosthetic side. Response latency (ms) was reported as the time between the onset of force plate translation and the participant's response.

3. Limits of Stability (LOS), which assesses the participant's ability to shift their weight in all directions toward targets placed at a distance that takes into account the participant's height and the maximum sway that they can generate. The endpoint excursion score is the percentage of the participant's maximum distance travelled relative to their height. The composite score is a single numeric value that represents the average performance over all eight directions. The passive ROM in the lower limbs was recorded in both groups by a single clinician (AF) with a goniometer. The strength of the flexors, extensors, abductors, and adductors of the hip, and the dorsi- and plantar flexors of the ankle were assessed with a hand held dynamometer (HHD) (JTECH PowerTrack II Commander, Salt Lake City, Utah) using a standardised protocol.¹⁸ The score for each muscle was taken as the maximum value of three sets of measurements.

The SF-36 questionnaire¹⁷ and the Faces Pain Scale–Revised (FPS-R)^{19,20} were used to record general health, level of activity and pain. The SF-36 ranges from 0 to 100, with 100 being the best outcome and 0 the worst outcome. The FPS-R ranges from 0 to 10 with 0 being the best outcome and 10 the worst outcome. The revised Oswestry Back Pain Questionnaire²¹ was used to measure disability as a result of back pain, the Harris hip score²² to measure disability as a result of hip pain, the Prosthetic Evaluation Questionnaire (PEQ)²³⁻²⁶ to assess quality of life for prosthetic wearers and the Timed Up & Go (TUG) test^{23,27} to evaluate the risk of falling. The Oswestry Back Pain Questionnaire ranges from 0 to 100 with 0 as the best outcome and 100 as the worst outcome. The Harris hip score and PEQ both range from 0 to 100 with 100 as the best outcome and 0 as the worst outcome. The TUG is the number of seconds it takes to stand up from a standard chair, walk 3 m, turn, and return back to the chair and sit down. The shorter the time on the TUG, the better the balance ability.

Statistical analysis

A control participant was matched to each PFFD patient. The Wilcoxon Signed rank test was used to compare each PFFD patient with a matched control participant. A Benjamini-Hochberg 5% false discovery rate was applied to 57 comparisons, and results are considered statistically significant at $p < 0.0123$.²⁸ The analyses were performed using SAS statistical software (The SAS Institute, Cary, North Carolina).

Results

All patients in the PFFD group reported that they were satisfied with the outcome of surgery. Of the 12, five were currently students, five had office jobs, and two were manual workers. Of those who were no longer students, five were college graduates and two had attended some college in the past. A total of seven were single and five were married, two of

whom had children. All but one patient reported many close friendships and none reported that they avoided social gatherings, attending medical examinations or looking in mirrors. Avoidance of public changing rooms, gym and sports, swim suits, being physically close, photos or videos, change of posture, and hiding their leg differences with clothes was reported to occur at least occasionally for between one and three patients. Six undertook weight-lifting and nine exercised regularly to alter their shape; seven avoided some types of clothing such as tight jeans and shorts.

The Wilcoxon signed rank test demonstrated that the modified Oswestry disability index was similar in the PFFD and control groups (6.8% (0% to 28%) and 7.0% (0% to 38%), respectively; $p = 1.00$). On the day of testing, only one patient in the PFFD group reported mild low back pain on the FPS-R scale. The mean Harris hip score for the PFFD group was 92.7 (78 to 100) and two reported pain on their non-affected side with scores of 78 and 81, respectively (Table I).

The PFFD group scored a mean of 8.5 seconds (6.6 to 12.6) on the TUG, demonstrating a low risk of falling (high risk is defined as a TUG time > 14 seconds²⁹). The Wilcoxon signed rank test demonstrated that the control group had a statistically significant faster speed of 6.5 seconds (4.9 to 7.7) ($p = 0.001$). There were no further statistical differences between the groups in overall health and well-being (all $p > 0.057$; Table I).

The PFFD group had reduced hip flexion and ankle dorsiflexion ($p = 0.001$ and $p = 0.008$, respectively), and increased ankle plantar flexion on the prosthetic side compared with the control group ($p = 0.001$) (Table II).

The PFFD group had statistically weaker hip flexors ($p = 0.001$), hip abductors ($p = 0.008$) and ankle plantar flexors ($p = 0.009$) on the prosthetic side compared with the control group (Table II). The plantar flexors of the ankle in the PFFD group would be used to extend the knee on the prosthetic side. In comparison to the strength of the plantar flexors in the control group, the plantar flexor strength on the prosthetic side of the PFFD group was significantly less. Only the PFFD group completed the PEQ and they scored low in areas of satisfaction, appearance, and sounds of the prosthesis. However, they reported that others perceived them well and they did not see themselves as a social burden.

The PFFD group had a significantly reduced walking speed ($p = 0.0049$), cadence ($p = 0.0024$), stride time ($p = 0.0024$), foot off ($p = 0.0005$) and single support ($p = 0.0122$) compared with the control group (Table III). The Wilcoxon signed rank test demonstrated that anterior pelvic tilt throughout the gait cycle was greater in the PFFD group 21.2° (10° to 31.9°) vs 7.7° (4° to 12.3°) in the control group ($p < 0.001$). Peak hip extension was reduced in the PFFD group with the prosthetic side exhibiting the greatest reduction. At the knee, the PFFD group had peak knee flexion of 51.9° (10° to 31.9°) on the prosthetic side and 58.3° (19.5° to 73°) on the non-prosthetic side during swing phase. Both sides were only slightly reduced compared with the controls 62.2° (49.2° to 70.9°) ($p = 0.005$) (Fig. 3). In the PFFD group sagittal knee kinematics also revealed an absent loading response after initial contact and a mild decrease in extension of the during stance phase on the prosthetic side as well as delayed peak knee flexion during swing on the non-prosthetic side (Fig. 3). While walking

at a reduced speed the PFFD group non-prosthetic side produced on average 3.6 W/kg (1.6 to 6.0) during peak ankle power generation ("Push Off"), while the controls generated a mean of 4.0 W/kg (2.7 to 6.3) and the prosthetic side only a mean of 0.8 W/kg (0.1 to 3.2). Hip power generation ("Pull Off") was the same in the non-prosthetic side of the PFFD group and the controls had a mean of 1.4 W/kg (0.5 to 2.7) but reduced on the prosthetic side with a mean of 0.8 W/kg (0.5 to 2.4).

There were no differences in anterior-posterior standing sway under altered sensory conditions between the two groups in the SOT. In the MCT there were no differences in response latency following force plate translations between the two groups. The PFFD group had significantly increased weight-bearing on the non-prosthetic side (Fig. 4), and a significant reduction in the ability to shift their weight within the base of support compared with the control group in LOS. This was most pronounced when attempting to shift weight to their prosthetic side (Fig. 5).

Discussion

Despite the cosmetic defect of having a shortened limb facing backwards, all the patients in this study who had undergone Van Nes rotationplasty for congenital PFFD used their prosthesis all the time and maintained high levels of function, were well-adjusted emotionally and socially, and comfortable with their appearance and prosthetic design. Unexpectedly, there were no differences between the PFFD and control groups for any health related QoL subscales on the SF-36. These findings did not support the hypothesis that the PFFD group would have decreased QoL despite having notable deficits in ROM, strength, gait parameters and postural control.

Reduced ROM and strength of the hip on the prosthetic side may explain the decreased walking speed and postural and gait asymmetry. The relative weakness of the plantar flexors on the prosthetic side in our patients was comparable with that found in studies of those who had similar surgery after resection of a tumour.^{14,15}

The PFFD group used the non-prosthetic leg as the dominant side, spending more time during the gait cycle with the non-prosthetic foot on the ground; they also used this limb to generate more power at the ankle and hip. Our findings of reduced walking speed in these patients has been reported in the other papers reporting the outcome after excision of a tumour.^{9,12} We might speculate that if the PFFD group was encouraged to walk at the same speed as the control group they would have had nearly normal ankle push off power and possibly above-normal hip pull off power on the non-prosthetic side necessary to compensate for the lack of power on the prosthetic side.

The PFFD group's knee kinematics data support the proposition that rotationplasty provides a functional joint at the knee allowing single limb support and foot clearance during swing, even though an initial loading response was not always seen. The dramatic increase in anterior pelvic tilt in the PFFD group is interesting and may be a result of limited ROM and musculature of the hip. This compensatory strategy may assist in controlling the centre of mass, thus improving balance over the prosthetic limb.

CDP data demonstrated that although the PFFD group did not have postural control problems as a result of sensory deficit, they had asymmetrical weight-bearing and reduced ability to accept weight through the prosthetic side. The high degree of asymmetry in these patients is of concern, and demands further analysis of the association between postural instability and musculoskeletal impairment.

PFFD is a rare deformity. The sample size of our study was limited by the available population. Despite the small sample, however, we were able to assess a wide range of adults who had undergone rotationplasty, some 20 or more years previously, and who demonstrated good functional results. Further research on this population should include motion analysis with trunk markers to document lateral trunk lean and other trunk compensations, and electromyography to assess when and how effectively the rotated muscle groups are firing, as well as longer follow-up.

Regardless of deficits in gait and posture, the Van Nes rotationplasty should be considered as a viable option for selected children with unilateral PFFD. Adult participants in this study who underwent the surgery as a child were generally satisfied with the outcome and had a good quality of life.

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Fig. 1.
Photographs of a patient at 23 years after Van Nes rotationplasty for proximal focal femoral deficiency with and without prosthesis.

Fig. 2.
Flowchart of recruitment and participation.

Fig. 3. Sagittal knee angle plots for control group and proximal focal femoral deficiency (PFFD) group (prosthetic and non-prosthetic side) during one gait cycle. The prosthetic side in the PFFD group exhibited an absent loading response after initial contact, mild decrease in knee extension at stance, and mild decrease in knee flexion during swing.

Fig. 4.

Plot showing the mean symmetry of weightbearing of the proximal focal femoral deficiency (PFFD) and control groups. The PFFD group demonstrated significant increases in asymmetry, favouring the non-prosthetic side (* $p = 0.004$). Error bars denote the standard error.

Fig. 5.

Plots showing the mean maximum excursion (maximum distance travelled during conscious weight shifting) of the proximal focal femoral deficiency (PFFD) and control groups, for both composite data (left) and for prosthetic side only (right). The PFFD group demonstrated reduced percentages distance travelled in all directions (composite). Restrictions were most pronounced towards the prosthetic side ($p < 0.0123$).

Table I

Descriptive and comparative outcome measures for patients with a Van Nes rotationplasty (PFFD) and a control group

Mean outcome measure (range)	PFFD group	Control group	p-value
Faces pain scale (0 to 10)	0.2 (0 to 2)	0.8 (0 to 8)	0.750
Harris hip score (0 to 100)	92.7 (78 to 100)	100.0 (100)	0.063
Oswestry disability index (0 to 100)	6.8 (0 to 28)	7.0 (0 to 38)	1.000
Timed Up & Go (s)	8.5 (6.6 to 12.6)	6.5 (4.9 to 7.7)	0.001 *
Short-Form 36 subscore (0 to 100)			
Physical functioning	80.0 (25 to 100)	93.8 (60 to 100)	0.057
Role function	95.8 (50 to 100)	87.5 (0 to 100)	0.750
Bodily pain	89.2 (62 to 100)	84.0 (32 to 100)	0.574
General health	81.9 (62 to 100)	80.1 (22 to 100)	0.915
Vitality	71.7 (15 to 90)	73.3 (40 to 90)	0.601
Social functioning	90.6 (25 to 100)	95.8 (62.5 to 100)	1.750
Role emotional	86.1 (0 to 100)	100.0 (100)	0.500
Mental health	78.0 (24 to 100)	85.3 (68 to 100)	0.889

* statistically significant ($p < 0.0123$)

Table II

Description and comparison of lower extremity strength and range of movement for patients with a Van Nes rotationplasty (PFFD) and a control group

Mean (range)	PFFD group (prosthetic side)	Control group	p-value
Strength (N)			
Hip flexion	53.2 (22 to 147.4)	155.2 (96.8 to 389.4)	0.001 *
Hip abduction	86.2 (41.8 to 189.2)	125.5 (81.4 to 193.6)	0.008 *
Hip adduction	95.3 (41.8 to 354.2)	133.6 (66.0 to 264.0)	0.034
Hip extension	83.4 (46.2 to 259.6)	93.0 (48.4 to 242.0)	0.592
Ankle plantarflexion	92.4 (30.8 to 521.4)	149.9 (94.6 to 246.4)	0.009 *
Ankle dorsiflexion	78.8 (26.4 to 193.6)	78.1 (37.4 to 151.8)	0.690
Range of movement (°)			
Hip flexion	79.6 (50 to 115)	123.1 (115 to 140)	0.001 *
Hip abduction	34.2 (20 to 70)	44.2 (30 to 60)	0.547
Hip extension	-3.8 (0 to 50)	0.0 (0)	0.750
Ankle plantarflexion	62.1 (50 to 90)	41.9 (30 to 50)	0.001 *
Ankle dorsiflexion	-1.7 (-20 to 20)	10.6 (0 to 20)	0.008 *
Forefoot adduction	32.5 (0 to 70)	37.7 (20 to 60)	0.502
Forefoot abduction	23.8 (-10 to 60)	18.1 (0 to 50)	0.616

* statistically significant ($p < 0.0123$)

Table III

Description and comparison of temporal spatial gait parameters for patients with a Van Nes rotationplasty (PFFD) and a control group

Gait parameter (mean, range)	PFFD group (prosthetic side)	Control group	p-value
Cadence (steps/min)	101.3 (83.2 to 123.0)	116.0 (107.9 to 131.6)	0.0024 *
Stride time (s)	1.2 (1.0 to 1.4)	1.0 (0.9 to 1.1)	0.0024 *
Opposite foot off (% gait cycle (GC))	14.3 (11.1 to 21.3)	9.8 (6.3 to 12.3)	0.0005 *
Opposite foot contact (% GC)	49.8 (49.2 to 50.9)	50.1 (49.3 to 52.4)	0.470
Single support (% GC)	35.6 (23.1 to 45.9)	39.4 (34.9 to 53.7)	0.0122 *
Double support (% GC)	27.9 (34.9 to 53.7)	21.5 (16.1 to 28.2)	0.004 *
Foot off (% GC)	63.7 (59.8 to 70.5)	60.8 (59.0 to 62.8)	0.027
Stride length (m)	1.2 (1.0 to 1.5)	1.3 (1.2 to 1.4)	0.380
Step length (m)	0.6 (0.5 to 0.7)	0.7 (0.6 to 0.7)	0.380
Walking speed (m/s)	1.0 (0.7 to 1.3)	1.3 (1.1 to 1.5)	0.0049 *

* statistically significant ($p < 0.0123$)