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Abstract

Van Nes rotationplasty is a treatment option for individuals with congenital proximal focal femoral deficiency (PFFD), in which the extremity is surgically rotated to utilize the ankle and foot as a functional knee joint in a prosthesis. This case control study was done to determine the long-term function and quality of life (QOL) outcomes of these individuals. Twelve prosthetic participants (PFFD Group) and 12 control participants (Control Group) completed SF-36, Faces Pain Scale-Revised, Harris Hip Score, Oswestry back pain score and Prosthetic Evaluation Questionnaire© .Lower extremity physical exam, gait analysis, computerized dynamic posturography (CDP), and Timed 'Up & Go' testing was also completed. Wilcoxon Signed rank test used to compare each PFFD participant to the matched Control participant with false discovery rate of 5%. 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 showed decreased symmetry in stance, and reduced end point and maximum excursions. Individuals who underwent Van Nes rotationplasty showed a high level of function and QOL at long-term follow up, but presented with significant differences in gait and posturography parameters compared to the Control Group.

Introduction

Proximal Focal Femoral Deficiency (PFFD) is a rare congenital deformity of unknown etiology with hypoplasia or absence of the proximal femur. With a diagnosis of PFFD there are often associated

hip instability and rotational malalignments, insufficient hip musculature, hip joint contractures, and a significant leg length discrepancy^{1,2}. There are multiple surgical and prosthetic options for these complex patients. In 1950, Van Nes published a case describing the treatment option of rotationplasty surgery for congenital defects of the femur using the ankle of the shortened limb to control a prosthetic knee joint in a below-the-knee type prosthesis³. This procedure has the advantage of producing a functional joint at the level of the knee to allow a more normal gait pattern (Fig. 1). This procedure was later modified to include fusion of the knee⁴. Van Nes rotationplasty remains controversial though due to perceived poor cosmetic appearance with the foot facing backwards, issues with proper prosthetic fit and possible need for subsequent rotational osteotomies in the future due to derotation of the limb.

To our knowledge, there are limited published reports on the long-term outcome of individuals into adulthood with PFFD who underwent Van Nes rotationplasty. Most authors looked at children and utilized measures of function including formal gait analysis. In 1996, Fowler et al. looked at energy expenditure during walking for nine subjects who underwent Van Nes rotationplasty compared to seven subjects who had a Syme amputation (average age of 12 years)¹. They found that those children with a Syme amputation had increased energy expenditure during normal gait¹. Then in 1999, Fowler et al. compared gait mechanics of 10 subjects with PFFD who either underwent rotationplasty to nine subjects who had a Syme amputation (average age of 15 years)⁵. Individuals who underwent a Syme amputation demonstrated amplified compensatory strategies on their non-prosthetic side as compared to individuals who underwent a rotationplasty^{1,5}. However, within these reports there was no mention of activity level or quality of life (QOL).

Function, activity level and QOL has been assessed in individuals with malignant tumor resection who underwent rotationplasty⁶⁻¹³. In one study of gait in 10 children who had a tumor resection and a rotationplasty, differences were reported in kinetic parameters compared to controls, but the participants were still highly functional¹⁰. An adult study with a similar population revealed mild gait deviations and a reduced hip extensor mechanism, but good functional results as

well⁸. Another study reported that individuals who underwent rotationplasty for tumor resection were highly involved in sports¹⁴. Veenstra et al. used multiple measures of QOL including the SF-36. Individuals who underwent rotationplasty had lower scores in measures of physical function and body image⁶. An additional study reported that children who underwent rotationplasty for tumor resection perceived that they had no barriers to recreation, sporting or career goals. Also, they were emotionally accepting of their body image⁹.

There is a gap in the literature regarding the long-term function and QOL of adults with PFFD who underwent rotationplasty at a young age. Therefore, the purpose of the current study was to characterize the acceptance of the rotationplasty, presence of hip and/or back pain, range of motion (ROM) and strength of the prosthetic side, postural stability, gait deviations, as well as overall health and well-being. We then compared those outcomes with those of age and gender matched controls. We hypothesized that the long-term QOL of individuals with PFFD who underwent a rotationplasty would be decreased compared to that of the controls.

Materials and Methods

This case control study was approved by the institutional review board at RUSH University Medical Center. Medical record reviews and solicitation of two community prosthetic companies were used to retrieve patient contact information for 43 patients who underwent rotationplasty from 1940-1999 (Fig. 2). Attempts to contact all of these individuals by phone or letter were made, but only 22 were able to be located. Two of these individuals were excluded as they had undergone an ankle disarticulation as an adult. Eight of the 22 who consented lived too far away and were only able to answer a few questionnaires by mail. As a result, 12 individuals (mean age: 31.6 ± 13.5 years) with congenital PFFD who underwent a Van Nes rotationplasty as a child and currently walk with a prosthesis participated (PFFD Group) in person and the data of these 12 were analyzed. Rotationplasty was performed at an average age of 6.5 ± 3.9 years with follow up testing performed in this study 25.1 ± 11.2 years postoperatively. Exclusion criteria included lower extremity surgery or

fracture within the past year. Sample size was based on convenience as there was only a limited population to draw from. Twelve, age and gender matched adults without history of congenital musculoskeletal or neuromuscular deformities (mean age: 32.6 ± 14.1 years) were tested for comparison (Control Group).

Both groups of participants had 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.

Computerized Dynamic Posturography (CDP) was performed using the NeuroCom SMART EquiTest® (NeuroCom® International, Inc., Clackamas, OR, USA). CDP is a quantitative method used to isolate and assess the sensory and motor components of standing postural control. This is accomplished using force platforms to measure postural sway during the following testing protocols Sensory Organization Test (SOT), Motor Control Test (MCT), and Limits of Stability (LOS). During the SOT, participants were exposed to six conditions, three trials of each that systematically target the contributions of visual, somatosensory and vestibular feedback on standing postural sway. During the MCT, participants were exposed to unexpected anterior-posterior forceplate translations scaled to the participant's height at small (2.8 deg/sec), medium (6.0 deg/sec), and large (8.0 deg/sec) velocities. Using this protocol, a measure of weight bearing symmetry was provided where a positive value indicates an increased percentage of weight bearing to the non-prosthetic side. Response latency (ms) was reported as the amount time between the onset of forceplate translation and the participant's response. Finally, the LOS assessed the participant's ability to volitionally shift their weight in all directions toward targets placed at a distance that takes into account the individual's height and the maximum sway that they can generate. The endpoint excursion score is the percentage of the individual's maximum distance traveled relative to their height. The composite endpoint excursion is the average distance in all directions.

X-rays (A/P and lateral) of both lower extremities were taken for the PFFD Group. Based on available radiographs done at the time of the surgery and last follow up, participant's hips were classified per Aitken classification of PFFD². In this study, three participants had type A, no participants had type B, two participants had type C, four participants had type D PFFD, and three participants had congenital short femur. Of these participants, eight had a knee fusion and four did not. Lower extremity passive ROM measurements were taken of both groups by a single clinician (AF) with a goniometer using a standard protocol¹⁵. Lower extremity muscle strength of the hip flexors, extensors, abductors, and adductors, and ankle plantar flexors and dorsiflexors were assessed with a hand held dynamometer (HHD) (JTECH PowerTrack II Commander, Salt Lake City, UT) using a standardized protocol¹⁶. Strength scores for each individual muscle was the maximum value of three trials.

Additional activity and functional evaluation measures included Short Form-36 (SF-36) Medical Outcomes Study questionnaire¹⁷ to review general health, activity level and pain; Faces Pain Scale-Revised (FPS-R)^{18,19}; Revised Oswestry Back Pain Questionnaire²⁰ to measure disability due to back pain; Harris Hip Score²¹ to measure disability due to hip pain; Prosthetic Evaluation Questionnaire© (PEQ©)²²⁻²⁵ to assess quality of life for prosthetic wearers; and the Timed Up & Go (TUG)^{22,26} to evaluate fall risk.

Statistical analysis. A matched pairs study design, with one control participant matched to each prosthetic participant was used. The Wilcoxon Signed rank test was used to statistically compare each PFFD Group participant to the matched Control Group participant. A Benjamini-Hochberg 5% false discovery rate was applied to 57 comparisons, and results are considered statistically significant at $p < 0.0123$ ²⁷. The analyses were performed using SAS statistical software (The SAS Institute, Cary, NC).

Results

Demographics. The PFFD Group reported that they were satisfied with the outcome of the rotationplasty. Of the 12 participants: five were currently students, five worked at office jobs, and two worked in

manual labor. Of those who were no longer students, 5 were college graduates and 2 had attended some college in the past. Seven of the participants were still single and all but one had dated in the last year; and the five other participants were all married. Two of the married individuals had children of their own. All but one participant reported multiple close friendships.

Body Image. None of the 12 participants reported that they avoided social gatherings, medical exams or mirrors. Avoidance of changing rooms, gym and sports, swim suits, being physically close, photos or videos; and use of jewelry, change of posture, and hiding their leg difference with clothes was reported to occur at least occasionally for 8-25% of the participants. Half of the participants performed weight lifting activities and 67% exercised to alter their shape. Fifty-eight percent of participants stated that they avoided some types of clothing such as tight jeans and shorts.

Pain. The PFFD and Control Groups both reported similar low back pain with $6.8 \pm 9.7\%$ and $7.0 \pm 13.0\%$ disability respectively on the Oswestry back pain questionnaire. On the day of testing, only one participant in the PFFD group reported mild low back pain on the Revised-Faces Pain Scale. The average Harris Hip Score for the PFFD Group was 92.7 ± 9.2 out of 100, indicating excellent outcome. Two participants in the PFFD Group reported pain on their non-prosthetic hip (Table I).

TUG. The PFFD Group scored an average of 8.5 ± 1.6 seconds on the TUG, demonstrating a low fall risk. The Control Group scored statistically better with faster speed of 6.5 ± 1.0 seconds on average (Table I).

SF-36. There were no statistical differences between the Groups in overall health and well-being (Table I).

ROM. The PFFD Group presented with reduced hip flexion and ankle dorsiflexion, and increased ankle plantarflexion ROM on the prosthetic side compared to the Control Group (Table II). All other extremity

ranges on the non-prosthetic side for the PFFD Group were similar to the Control Group.

Strength. The PFFD Group demonstrated statistically weaker hip flexors, hip abductors and ankle plantarflexors on the prosthetic side compared to the Control Group (Table II). Ankle plantarflexors in the PFFD Group would be used to extend the knee on the prosthetic side. For the PFFD Group, strength on the non-prosthetic side was similar to the Control Group.

PEQ©. Only the PFFD Group completed the PEQ©. As a group, they scored low in areas of satisfaction, appearance, and sounds of the prosthesis. However, participants reported that others perceived them well and they did not see themselves as a social burden (Table III). A higher percentage score represents a positive response.

Gait Analysis. Temporal-spatial gait parameters for the PFFD Group demonstrated a significant decrease in walking speed, cadence, stride time, foot off, and single support compared to Control Group (Table IV). Kinematic data showed the PFFD Group had an anterior pelvic tilt throughout the gait cycle (average 21.2°) which was greater than Controls (average 9.3°) by 56%. Peak hip extension was reduced in the PFFD Group with the prosthetic side exhibiting the greatest reduction. At the knee, the PFFD Group displayed an average peak knee flexion of 55.3° on the prosthetic side and 60.5° on the nonprosthetic side during swing phase, both sides were only slightly reduced compared to the Controls (62.2°) (Fig. 3). In the PFFD Group, sagittal knee kinematics also revealed an absent loading response after initial contact and a mild decrease in knee extension during stance phase on the prosthetic side as well as delayed peak knee flexion during swing on the non-prosthetic side (Fig. 3). Kinetic analysis revealed that while walking at a reduced speed the PFFD Group non-prosthetic side produced on average 3.5 W/kg during peak ankle power generation ("Push Off"), while the Controls generated 4.1 W/kg and the prosthetic side only 0.9 W/kg. Hip power generation ("Pull Off") was equal between the PFFD Group non-prosthetic side and Controls (1.4 W/kg) but reduced on the prosthetic side (0.8 W/kg).

Computerized Dynamic Posturography. SOT: There were no differences in anterior-posterior standing sway under altered sensory conditions between the two groups. MCT: There were no differences in response latency following forceplate translations between the two groups. The PFFD group demonstrated a significant increase in weight bearing toward their non-prosthetic side (Fig. 4). LOS: The PFFD Group demonstrated a significant reduction in the ability to volitionally shift their weight within their base of support compared to the Control Group. This was most pronounced when attempting to shift their weight to their prosthetic side (Fig. 5).

Discussion

Despite the cosmetic defect of having a shortened limb facing backwards, individuals in this study who had rotationplasty for congenital PFFD were full time prosthetic users; and were overall functional, 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 compared to the Control Group, regardless of the PFFD Group's notable deficits in lower extremity ROM, strength, gait parameters and postural control.

Reduced hip range of motion and strength on the prosthetic side may have led to decreased walking speed as well as postural and gait asymmetry. The relative weakness of the prosthetic side plantarflexors in our participants was also comparable to that found in other studies of those with rotationplasty due to tumor resection^{12,13}, yet surprising given the fact that the plantarflexors on the prosthetic side now need to act as knee extensors in a prosthesis.

The PFFD Group used the non-prosthetic leg as the dominant side as evidenced by spending more time during the gait cycle with the non-prosthetic foot on the ground and also used this limb to generate more power at the ankle and hip. Our findings of reduced walking speed for the PFFD Group was also noted in other studies with individuals who underwent rotationplasty after bone tumor

resection^{7,10}. We estimate 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 nonprosthetic side necessary to compensate for the lack of power on the prosthetic side. The PFFD Group's knee kinematics support the notion that rotationplasty provides a functional joint at the level of 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 hip ROM and insufficient hip musculature. This compensatory strategy may assist in controlling the center of mass, thus improving balance over the prosthetic limb.

Computerized Dynamic Posturography data demonstrated that although the PFFD Group did not have postural control deficits related to sensory deficits, they did present with weight bearing asymmetry and decreased ability to accept weight through their prosthetic side. We did not find any studies that looked at stance symmetry for individuals with rotationplasty. The high degree of asymmetry noted in the current study was concerning and calls for a more in depth analysis of the association among postural instability and musculoskeletal impairments.

PFFD is a rare congenital deformity. The sample size of our study was limited by the available population. Despite the small sample size, we were able to assess a wide range of adults who had a rotationplasty a mean of 25 years later who demonstrated good functional results. Further research on this population should include motion analysis with trunk markers to document amount of trunk lateral lean and other trunk compensations, as well as electromyography to assess when and how effectively the rotated muscle groups are firing.

Research supports the use of rotationplasty for patients undergoing bone tumor resection but there is very limited information regarding long-term outcomes for individuals with PFFD. Our findings are important as they fill in a significant gap in the literature regarding QOL and long-term functional outcomes for individuals with congenital

PFFD and rotationplasty. Regardless of deficits in gait and posturography parameters, rotationplasty should be considered as a viable long-term option for selected cases of children with unilateral PFFD. Adult participants in this study who underwent rotationplasty as a child were overall satisfied with the outcome and demonstrated a good QOL as adults.

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Table I. Descriptive and comparative outcome measures for individuals with a Van Nes rotationplasty and a control group \pm SD.

Outcome Measure	PFFD Group	Control Group	p Value
Faces Pain Scale (0-10)	0.2 \pm 0.6	0.8 \pm 2.3	0.750
Harris Hip Score (0-100)	92.7 \pm 9.2	100.0 \pm 0.0	0.063
Oswestry Disability Index (0-100)	6.8 \pm 9.7	7.0 \pm 13.0	1.000
Timed Up & Go (seconds)	8.5 \pm 1.6	6.5 \pm 1.0	0.001*
SF-36: Physical Functioning (0-100)	80.0 \pm 20.9	93.8 \pm 11.5	0.057
SF-36: Role-Function (0-100)	95.8 \pm 14.4	87.5 \pm 31.1	0.750
SF-36: Bodily Pain (0-100)	89.2 \pm 13.0	84.0 \pm 21.8	0.574
SF-36: General Health (0-100)	81.9 \pm 13.6	80.1 \pm 21.8	0.915
SF-36: Vitality (0-100)	71.7 \pm 19.8	73.3 \pm 13.2	0.601
SF-36: Social Functioning (0-100)	90.6 \pm 21.4	95.8 \pm 11.1	1.750
SF-36: Role - Emotional (0-100)	86.1 \pm 33.2	100.0 \pm 0.0	0.500
SF-36: Mental Health (0-100)	78.0 \pm 23.0	85.3 \pm 9.9	0.889

*p< 0.0123

Table II. Description and comparison of lower extremity strength and range of motion for individuals with a Van Nes rotationplasty and for a control group \pm SD.

	PFFD Group (prosthetic side)	Control Group	p Value
Strength (Newtons)			
Hip Flexion	53.2 \pm 37.7	155.2 \pm 74.8	0.001*
Hip Abduction	86.2 \pm 39.9	125.5 \pm 29.0	0.008*
Hip Adduction	95.3 \pm 33.5	133.6 \pm 55.9	0.034
Hip Extension	83.4 \pm 28.6	93.0 \pm 42.6	0.592
Ankle Plantarflexion	92.4 \pm 43.8	149.9 \pm 46.2	0.009*
Ankle Dorsiflexion	78.8 \pm 43.3	78.1 \pm 34.0	0.690
Range of Motion (degrees)			
Hip Flexion	79.6 \pm 5.8	123.1 \pm 1.5	0.001*
Hip Abduction	34.2 \pm 4.2	44.2 \pm 1.6	0.547
Hip Extension	-3.8 \pm 4.5	0.00	0.750
Ankle Plantarflexion	62.1 \pm 3.7	41.9 \pm 1.4	0.001*
Ankle Dorsiflexion	-1.7 \pm 3.9	10.6 \pm 1.2	0.008*
Forefoot Adduction	32.5 \pm 5.6	37.7 \pm 2.1	0.502
Forefoot Abduction	23.8 \pm 7.4	18.1 \pm 2.2	0.616

*p<0.0123

Table III. Descriptive outcome of Prosthetic Evaluation Questionnaire© for individuals who had a rotationplasty.

Scale (max 100%)	Average % Scored ± SD
Satisfaction	69.5 ± 30.0
Ambulation	78.9 ± 17.5
Appearance	74.2 ± 11.7
Frustration	79.1 ± 27.9
Perceived Response of Others	90.2 ± 16.2
Residual Limb Health	67.5 ± 20.5
Social Burden	92.9 ± 10.5
Sounds	68.0 ± 24.5
Utility	76.9 ± 17.6
Well Being	86.1 ± 18.8

Table IV. Description and comparison of temporal spatial gait parameters for individuals with Van Nes rotationplasty and for a control group \pm SD.

Gait Parameter	PFFD Group (prosthetic side)	Control Group	p Value
Cadence (steps/min)	101.3 \pm 3.6	116.0 \pm 1.9	0.0024*
Stride Time (sec)	1.2 \pm 0.04	1.0 \pm 0.02	0.0024*
Opposite Foot Off (% GC)	14.3 \pm 0.9	9.8 \pm 0.5	0.0005*
Opposite Foot Contact (% GC)	49.8 \pm 0.2	50.1 \pm 0.2	0.470
Single Support (% GC)	29.8 \pm 4.1	38.9 \pm 0.8	0.0122*
Double Support (% GC)	23.3 \pm 3.5	21.0 \pm 0.7	0.204
Foot Off (% GC)	63.7 \pm 1.0	60.8 \pm 0.3	0.027
Stride Length (m)	1.2 \pm 0.05	1.3 \pm 0.03	0.380
Step Length (m)	0.6 \pm 0.03	0.7 \pm 0.01	0.380
Walking Speed (m/s)	1.0 \pm 0.05	1.3 \pm 0.03	0.0049*

*p<0.0123

GC = gait cycle

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Figure Legends

Figure 1. Subject with left PFFD who underwent Van Nes rotationplasty with and without prosthesis.

Figure 2. Flowchart of recruitment and participation.

Figure 3. Sagittal knee angle plots for Control Group and 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.

Figure 4. Weight bearing symmetry group averages and standard errors of the PFFD and Control Groups from the MCT are provided. The PFFD Group

demonstrated significant increases in asymmetry, favoring the non-prosthetic side. ** $p < 0.0123$.

Figure 5. Group averages and standard errors of the maximum distance traveled during volitional weight shifting of the PFFD and Control Groups from the LOS are provided. The PFFD Group demonstrated reduced percent distance traveled in all directions (composite). Limitation was most pronounced toward the prosthetic side. ** $p < 0.0123$

Each Author certifies that he or she has no commercial associations what might pose a conflict of interest in connection with the submitted article.

Ethical Review Board: This study has been approved by the RUSH Ethical Institutional Review Board

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