Midterm Survivorship and Complications of Total Knee Arthroplasty in Patients with Dwarfism

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Abstract:

Background:

Dwarfism is associated with skeletal dysplasias and joint deformities that frequently result in osteoarthritis requiring treatment with total knee arthroplasty (TKA). These surgeries can be challenging due to alignment deformities, poor bone stock, and smaller components. This study aims to compare TKA implant survivorship and complications between dwarf and non-dwarf patients.

Methods:

A retrospective case-control study was performed from 1997-2014 evaluating 115 TKAs in patients under the height threshold of 147.32cm. This cohort was compared to 164 patients of normal height, using propensity score weighting to balance gender, age, year of surgery, and comorbidities. Medical records were reviewed for demographics, surgical characteristics, and outcomes. Radiographic evaluation was performed to assess alignment, periprosthetic fractures, and loosening. All cases had 2-year minimum follow-up.

Results:

The revision rate was 8.7% in dwarfs compared with 3.7% in controls ($p=0.08$). The 2-, 5-, and 10-year implant survivorship in dwarfs was 96.4%, 92.5%, and 90.2%, respectively; and 96.6%, 95.6%, and 94.8% for controls, respectively ($p=0.24$). Dwarfs underwent significantly more manipulations for arthrofibrosis ($p=0.002$). There was greater femoral (17.4% vs. 2.1%, $p<0.01$) and tibial (6.5% vs 2.7%, $p<0.01$) component overhang in dwarfs compared to controls.

Conclusions:

Despite a two-fold increase in the revision rate of the dwarf cohort, the midterm survivorship is comparable between the dwarf and non-dwarf patients. However, dwarfs were more likely to become stiff and undergo manipulation; the increased propensity for stiffness may be associated with oversized components, as evidenced by greater component overhang, and an increased incidence of spinal pathology which has also been shown to lead to post-operative stiffness. Surgeons should be aware of this increased risk and may consider using smaller or customized implants to account for the morphological differences in this patient population.

Keywords: dwarf; total knee arthroplasty; outcomes; survivorship; complications

Level of Evidence: III

Introduction

Dwarfism can be a result of over 200 conditions, including endocrine disorders such as pituitary dwarfism and hypothyroidism, systemic disorders causing growth failure, genetic diseases, and skeletal dysplasias. Additionally, it is not uncommon for patients to be of idiopathic short stature, which is a height less than 147.32 cm according to the legal definition.[1] The most common form of genetic dwarfism, achondroplasia, accounts for 70% of all dwarfism and affects 1 in 15,000 to 1 in 40,000 people.[2] Among this population, skeletal dysplasias, such as achondroplasia, result in atypical load distribution in weight-bearing joints and can lead to orthopaedic complications such as joint deformity, particularly genu valgum; ligamentous laxity; and early degenerative joint disease.[3,4] As a result, these patients will often present as candidates for total knee arthroplasty (TKA).

Orthopaedic surgeons face several challenges when performing TKA in this population. Smaller components may be necessary, severe alignment deformities can be encountered, poor bone stock and soft tissue laxity or contractures may be present[5,6]. Despite these challenges, however, the body of literature regarding the clinical outcomes and complications of TKA in the dwarf population is not extensive. While many previous studies highlight a variety of potential intraoperative and postoperative complications, they are frequently of limited size due to the relative rarity of dwarfism.[5,7] Furthermore, a control group is often not present to serve as a comparative reference. Thus, the purpose of this study is to compare the revision rates and implant survivorship between dwarf and non-dwarf patients undergoing TKA.

Methods

A retrospective case-control study was performed between 1997 and 2014 on primary TKA patients under the height threshold of 147.32 cm (4’10”) using our institutional database. With these criteria, we identified 157 cases of primary TKA (156 females and 1 male). The average height was 146.43 cm and the mean age at the time of surgery was 70.7±10.7 years. We included all patients with a minimum 2-year follow-up (mean 6.2years, range 2.0-17.2), which left us with 115 TKAs in our final cohort. The primary etiology for TKA was osteoarthritis (112/115).

To obtain a balanced comparison with a control group of 164 patients with greater than 143.32 cm height, propensity score weighting was used to control for age, gender, Charlson comorbidity index[8], and year of surgery. The weights were generated using logistic regression to estimate the probability of being a dwarf based on the other variables, and then the weight was set to 1/prob[patient is dwarf] for patients who were dwarves, and 1/[1-prob[dwarf]] for non-dwarf patients. The weights were then normalized to a mean of 1.0. The weights ranged from 0.50 – 3.11; there were no extreme weights due to probabilities near 0 or near 1. Table 1 provides the demographics of the patient populations. All TKAs were done using posterior stabilized knees from three manufacturers (Zimmer [Warsaw, Indiana], Stryker [Mahwah, NJ], Depuy [Warsaw, Indiana]).

A manual review of the medical record was performed to identify patient demographics, surgical and hospital characteristics (operative time), and outcomes. The evaluated outcomes included any revision surgery and the reason for revision, subsequent procedures including manipulations under anesthesia, and intraoperative and postoperative complications, such as periprosthetic fracture, aseptic loosening, polyethylene wear/osteolysis, periprosthetic joint infection (PJI) defined by the International Consensus Meeting definition,[9] and dislocations.

Radiographic Analysis

Serial radiographic evaluation was performed of all anteroposterior and lateral radiographs by two independent orthopaedic surgeons on all preoperative, postoperative, and follow-up films. The inter-rater reliability (as measured by the concordance correlation coefficient) between the two orthopaedic surgeons was 0.94 (95% confidence interval [CI]: 0.91-0.96). Follow-up radiographs were also analyzed for radiolucent lines, periprosthetic fractures, and femoral and tibial component overhang. All measurements were obtained using digital imaging software, PACS (National Institutes of Health, Bethesda, MD) to obtain anatomic axis. Anatomic axis was measured using the angle formed by a line drawn from the center of the knee joint to the most proximal point of the mid-diaphyseal femur and a line drawn from the center of the knee joint to the most distal point of the mid-diaphyseal tibia. Normal femorotibial angles range from 174° to 178° depending on gender and race. While the tibial mechanical and anatomic axes are aligned, the femoral anatomical axis can be inclined 5-7° more than the mechanical axis. Further variation can result from tibial and femoral deformities and variation in hip angle. Cherian et al. discuss in greater detail the general principles behind radiographic axes and their application in TKA.[10] Radiographic loosening was defined by the presence or progression of component migration, change in position, subsidence, and complete radiolucent lines greater than 1mm[11]. Tibial component overhang was defined as any prosthetic material occurring outside the boundaries of a vertical line that extending from the cortex of the proximal part of the tibial plateau[12]. In contrast, femoral overhang was defined as component overhang >2mm in any of the 5 zones defined by the Knee Society[11,13].

Statistical Analysis

All statistical analyses were performed with R software 3.3.2 (R Foundation for Statistical Computing, Vienna, Austria) using an alpha level of 0.05 to determine significance. Kaplan-Meier survivorship curves were generated for 2-, 5-, and 10-year follow-up. Differences in survivorship were assessed using the log-rank test, while a Fisher's exact test was used to evaluate differences in revision rates. Student's t-tests were used to compare means between x-ray radiographic measurements. Our primary endpoint was the survivorship of the prosthesis or revision surgery for any reason. Secondary endpoints such as operative time, rate of manipulation procedures, and any significant radiographic differences between the groups were considered.

Results

Using propensity score weighting, the 5-, and 10-year survivorship was 92.5% (95% CI:87.8% -97.6%), and 90.2% (95% CI: 83.9% - 96.9%), respectively, for the dwarf cohort; and 95.6% (95% CI: 92.1% - 99.3%), and 94.8% (95% CI: 90.6% - 99.2%) for the non-dwarf cohort, respectively. The results were almost identical without the weighting. Overall, there was no difference in survivorship between the dwarf and non-dwarf cohorts (p=0.24, Figures 1 and 2). The revision surgery rate was 8.7% in the dwarf cohort compared with 3.7% in the control group. There was no statistically significant difference in the overall rate of revision (odds ratio [OR] 2.51, p=0.08), but the operative time was longer for dwarfs compared to controls (84.4 vs 74.6 min; p=0.01).

The reasons for revision in the dwarf group included aseptic loosening (n=3), PJI (n=3), patellofemoral arthritis (n=1), cement extrusion with pain (n=1), and periprosthetic fractures (n=2) (Table 2). Periprosthetic fractures were postoperative and included one tibial plateau fracture that had healed but required subsequent exchange of the tibial component, and one femur fracture that was treated with open reduction and internal fixation. However, the dwarf cohort underwent significantly more manipulations for arthrofibrosis (6.1% vs 0.0%, p=0.002). In the 7 patients that underwent manipulations under anesthesia, 29% (2/7) had femoral component overhang. In contrast, 18.5% (20/108) of TKAs that did not undergo manipulation had femoral component overhang.

In the control group, the pre- and post-operative anatomical axis values were 178.6º±6.5 and 175.9º±2.9, respectively. The pre- and post-operative anatomical axes in the dwarf group were 178.7º±8.7º and 176.3º±3.0º, respectively. There was no difference in pre-operative deformity between the dwarf and control cohorts (p=0.97), and there was no significant difference in postoperative alignment (p=0.62). However, there was greater femoral component overhang in the dwarf cohort (17.4%) compared to the control cohort (2.1%, OR 9.65, 95%CI: 5.40-17.27, p<0.01), and more tibial component overhang (6.5%) in dwarf patients compared to the control group (2.7%, OR 2.47, 95% CI: 1.36-4.49, p<0.01). For patients with tibial overhang, there was a trend towards a higher amount of tibial overhang in dwarfs (2.36 mm) compared to control patients (1.81 mm, p=0.09) (Table 3).

Discussion

While previous studies have shown that TKA is an effective treatment for degenerative disease in the joints of dwarfs, the literature regarding this unique and challenging population is limited. Orthopaedic surgeons must be cognizant that these patients may have poor bone stock, severe deformity necessitating soft tissue releases, and may require the use of smaller implants which can compromise surgery.

The results of the present study suggest that TKA in dwarfs demonstrate similar implant survivorship compared with a matched control cohort; however, surgeons should be aware that an increased rate of complications was found, although not statistically significant. TKA patients with dwarfism experienced greater post-operative stiffness resulting in a higher risk for manipulations, with approximately 1 in 20 dwarfs undergoing manipulation compared to none in the control cohort. This may be reflective of suboptimal component sizing, as component overhang was greater in the dwarf cohort. Also, since spine disease can lead to increased post-operative stiffness and manipulations, the increased incidence of spine pathology in dwarfs could also be a reason for the higher incidence of dwarf manipulations in this study. While dwarf patients were not predisposed to increased risk of malalignment compared with non-dwarfs, there was an association between dwarfism and longer operative times.

Questions have been raised about whether short stature can be indicative of poorer prosthesis survivorship and increased rates of complications.[5,6] Although prior studies have demonstrated that there are many benefits of the procedure, including functional outcome improvement,[7,14–19] they have not firmly established whether the results are comparable to those of normal stature. The use of a control group in our study allowed us to account for factors such as age, other relevant medical conditions (comorbidity index), and year of surgery for patients of normal stature. Although the older cases included may have used different surgical and anesthetic techniques with intrinsically higher risk for complication, it was our hope that controlling for year of surgery would help account for some of the temporal advancements in surgical technique and safety. In our study, TKAs performed in dwarfs demonstrated similar survivorship to that of the matched cohort. This is in contrast to Guenther et al., who found decreased 5 year survivorship in a case series of 138 TKAs in spite of overall improved functional outcome and International Knee Society (IKS) scores.[6] Although there were no statistically significant differences in revision rates in our study, our results indicate a significantly higher need for knee manipulations after TKA due to stiffness in dwarfs. While it has been argued that standard prostheses along with diligent preoperative planning can result in equally positive functional outcomes,[16] our results may demonstrate the contrary radiographically. Since many dwarf patients in our cohort had components that demonstrated overhang, the increased propensity for stiffness may be associated with oversized components; anterolateral overhang affects sagittal balance while mediolateral overhang can place excessive strain on the collaterals resulting in limited joint flexion motion[20,21]. Component overhang was greater for the femur, and there was a larger average distance of overhang of the tibial component in dwarf patients. Thus, ensuring proper component sizing for this population is critical and different implants may need to be used to accommodate this. Measures to prevent oversizing include using preoperative computed tomography, making femoral cuts by hand, or even utilizing custom-designed implants when appropriate to circumvent this complication.[5,7,19]

Our study had a number of limitations, and our findings should be interpreted in light of these issues. This study was retrospective, so we were limited to the data already available in the system, particularly lateral radiographs. In addition, long alignment x-rays were not available for most of these patients to measure anatomical and mechanical axes; thus, our measurements of anatomical axes may have been affected. In addition, a longer-term follow-up period on all the patients may have demonstrated some otherwise unseen results in the rates of revision, which has been demonstrated in the literature.[14,15] Furthermore, it was difficult to accurately determine the etiology of short stature in each individual, especially since it was poorly documented in the medical record or never diagnosed despite skeletal dysplasia. It is suspected that some of the patients naturally had short stature, as there was a significant presence of elderly females in our dwarf patient population. Due to low number of patients within the various etiologies for dwarfism, we could not analyze clinical outcomes stratified by diagnosis and thus could not differentiate how TKA might be affected in different subsets of skeletal dysplasia. However, this population still faces similar challenges to that of the dwarf population, as component sizing and poor bone quality must be taken into account. Lastly, we did not assess the functional outcome scores of our patients before and after the procedure. However, a recent study by Guenther et al. demonstrated that functional outcomes were significantly improved at 1 (67, $p<0.001$) and 5 year (65, $p<0.001$) postoperatively from admission (35). Although it has been established that postoperative knee function in dwarfs is significantly improved, it would have been interesting to see if the level of improvement is greater than or less than that of normal-height patients.

This study demonstrates that dwarfs undergoing TKA demonstrate no difference in midterm survivorship. However, patients with dwarfism were more likely to become stiff and may undergo manipulation. Surgeons should be aware of this increased risk and should ensure appropriate sizing with their surgical planning and technique. Further investigation should be performed to assess whether or not these general findings translate to specific conditions that can contribute to dwarfism and short stature.

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Table 1. Demographics of the patient population

	Sample size	**Age**	**Gender**	**BMI**	**Charlson score**	**Follow-up (years)**
TKA Dwarfs	115	70.2±10.8	115 females (100%)	31.2±7.3	4.0±1.4	6.2±3.6
TKA Controls	164	66.5±10.0	160 female (97.6%), 4 male (2.4%)	32.4±6.5	3.4±1.3	5.5±2.6

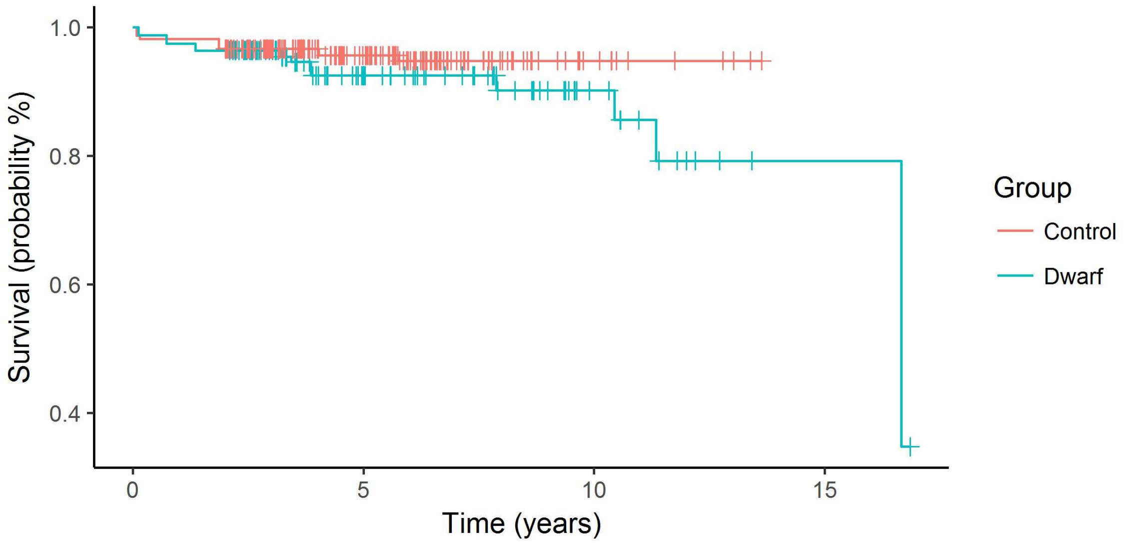
TKA=total knee arthroplasty; BMI=body mass index

Table 2. Total knee arthroplasty revisions in dwarfs and controls, with most common reasons for revision.

	Revision Rate (%)	**Periprosthetic joint infection (n)**	**Aseptic Loosening (n)**	**Periprosthetic Fracture (n)**	**Other (n)**
Dwarfs	8.7% (10/115)	3	3	2	2 (patellofemoral arthritis, cement extrusion with pain)
Controls	3.7% (6/164)	4	2	0	0
Odds Ratio	2.51	1.07	2.17	7.25	-
P-value	0.08	0.93	0.40	0.20	-

Table 3. Total knee arthroplasty x-ray measurements in dwarfs and controls.

	Dwarfs	**Controls**	**P-value**
Pre-operative Anatomic Axis (mean ± standard deviation)	178.7º±8.7º	178.6º±6.5º	0.97
Post-operative Anatomic Axis (mean ± standard deviation)	176.3º±3.0º	175.9º±2.9º	0.62
Tibial component overhang (mean ± standard deviation)	2.36mm±1.52	1.81mm±0.63	0.09
Tibial component overhang (%)	6.5%	2.7%	<0.01
Femoral component overhang (%)	17.4%	2.1%	<0.01


1.0
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Survival (probability %)
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Time (years)
Group
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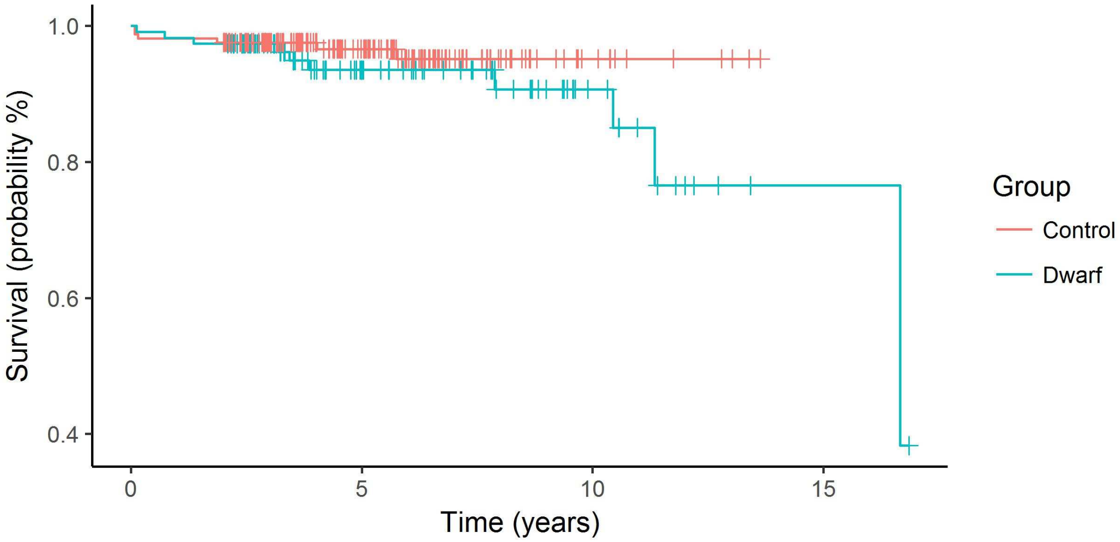
Dwarf


1.0
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Survival (probability %)
0
5
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15
Time (years)
Group
Control
Dwarf

Figure Legends:

Figure 1. Survivorship curve for patients undergoing primary total knee arthroplasty using propensity score weighting.

Figure 2. Survivorship curve for patients undergoing primary total knee arthroplasty without propensity score weighting.