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A prospective case report of anterior cruciate ligament reconstructions in identical adolescent female twins with open physes using single versus double hamstring grafts

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ABSTRACT

Introduction: We present a unique scenario involving identical adolescent female twins with open physes who sustained right anterior cruciate ligament ruptures on consecutive days while playing touch football.

Case Report: Our study design was a prospective case report to assess and compare two-year post-operative outcomes of different, randomly assigned surgical reconstruction techniques performed by a single surgeon on the same day, two months post-injury. The twins subsequently underwent similar and concurrent rehabilitation with the same physiotherapist. Data was collected, including admission details, pre-, peri-, and post-operative self-reported function, quality of life, objective laxity, physical, functional, radiological, and psychological measurements.

Conclusion: Similar outcomes for quality of life and self-reported function. Similar outcomes for range of movement, dynamic balance, neuromuscular control, propulsion, two-dimensional motion analysis, and agility tests. Minor differences in post-operative analgesia requirements measured differences in the hamstring and anterior cruciate ligament cross-sectional area, and objective anterior-posterior tibiofemoral joint laxity between the twins.

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INTRODUCTION

Diagnosis and reconstruction of anterior cruciate ligament (ACL) injuries in the skeletally immature patient is rising [1]. Australian [2] and US [3] statistics report a rapid increase in the incidence of ACL injury between 12–14 years, with a peak between 15 and 18 years. Several factors are thought to play a role in this increased incidence including a rise in competitive sports participation, decreased incidental physical activity (e.g., climbing trees, running on uneven surfaces), increased clinical awareness of pediatric ACL tears, and more comprehensive diagnosis and evaluation with magnetic resonance imaging (MRI) [4–7].

Historically, conservative management of ACL tears was preferred in the skeletally immature patient [5, 8], however, current clinical wisdom promotes early reconstruction with minimal short-term complications [9–13]. Nonetheless, the ideal surgical technique and rehabilitation remains contentious with long-term

outcomes unclear. Indeed, adult patients can develop symptoms and/or signs of osteoarthritis within 10 years of ACL injury, and therefore a child injured at age 10 could potentially, and alarmingly, develop osteoarthritis in early adulthood [14, 15]. Mindful of these issues, the International Olympic Committee's recent consensus on prevention, diagnosis, and management of pediatric ACL injuries [14] recommended that future research focus on the efficacy of different surgical techniques in the skeletally immature patient.

In this case report we present identical adolescent twins with open physes who sustained ACL ruptures within a 30-hour period. The twins were randomly assigned one of two common surgical techniques used in the skeletally immature patient. Given the rarity of these combined presentations a search of the literature revealed only one previous study of identical adolescent twins who underwent ACL reconstruction. Astur et al. [16] presented a case series of twin brothers with open physes who underwent ACL reconstruction surgery on the same day; however, in this study, no functional outcome measures were performed pre- or post-operatively, and no comparative post-operative medical imaging was reported. In addition, the twins in Astur et al. [16] sustained their injuries 11 months apart, had non-homogenous injuries, and outcomes were not compared to age, sex, and activity level matched controls.

This investigation assessed two-year radiological and functional outcomes following ACL reconstruction in monozygotic twins treated with different surgical techniques; the primary aim being to identify any differences in surgical outcome, patient satisfaction, objective performance, or radiographic discrepancies. The secondary aim was to compare outcome measures of the monozygotic twins with their uninjured dizygotic female sibling.

CASE REPORT

The case report was comprised of a set of female triplets; two monozygotic twins and a dizygotic sibling. They were born at 31 weeks gestation, achieved typical development and had good health throughout childhood.

The monozygotic twins were labelled as "Twin 1" and "Twin 2" according to the order of surgical intervention. Their dizygotic sibling was recruited as the age and sex matched "control."

Twin 1 injured her right ACL on 10/05/2017 at 13:00 hours, playing touch football in school, in her first game of the week, at the start of the second half. At the time of the injury Twin 1 was 12 years and 2 months of age and was premenarchal. The mechanism described by her was non-contact (she was running in a defensive role, touched an opposition player, and fell to the ground). She described no previous knee or musculoskeletal injury history to note.

Twin 2 injured her right ACL on 11/05/2017 at 19:00 hours, playing touch football in a club match, in her second game of the week, at the end of the first half. At the time of the injury Twin 2 was 12 years and 2 months of age and was premenarchal. The mechanism described by Twin 2 was also non-contact (offensive position, carrying ball, side-stepped to the right to avoid touch and fell to the ground). Twin 2 also described no previous knee or musculoskeletal injury history to note. Both twins were experienced touch football players with several years of experience and competitive skill level.

The presentation of these identical twins posed an unprecedented opportunity to test different ACL reconstruction techniques on two essentially identical physiological specimens, with their non-identical uninjured triplet as a control. Medical imaging evidenced an equivalent preoperative condition of the knee joint. Both twins underwent surgery on 07/07/2017. Findings from pre-operative assessment, examination under anesthetic, and knee arthroscopy are shown in Table 1.

Table 1: Intra-operative findings for Twin 1 and 2

Parameter	Twin 1	Twin 2	Descriptor (qualitative)
Range of motion	5°–135°	5°–135°	–
Lachman (25°)	>10 mm	>10 mm	Gr III (severe)
Anterior endpoint	Soft	Soft	Abnormal
AP translation (@25°)	>10 mm	>10 mm	Gr III (severe)
AP translation (@70°)	>10 mm	>10 mm	Gr III (severe)
Posterior draw (@70°)	0–2 mm	0–2 mm	Normal
Medial joint opening	0–2 mm	0–2 mm	Normal
Lateral joint opening	3–5 mm	3–5 mm	Normal
Pivot shift test	++ clunk	++ clunk	Positive
Compartment findings	None	None	–
Meniscus tear	Yes	Yes	Lateral only
Height (cm)	158	157.5	–
Mass (kg)	38.7	38.5	–

AP: anterior-posterior

Surgery

Both twins underwent transphyseal ACL reconstruction techniques with debridement of a small lateral radial meniscus tear. The twins underwent the same intra-operative regime for antibiotics, use of tourniquet, antibiotic wrap for graft, use of intra-operative nerve block, as well as the same post-operative imaging (see Figure 1) analgesia prescription, mobilization, and physiotherapy intervention (see Table 2).

Figure 1: Day 1 post-operative X-rays for Twins 1 and 2.

Surgical techniques were allocated randomly with no deliberate consideration of which twin was receiving which treatment. Twin 1 was operated on first with a total operating time of 122 minutes and had a single hamstring semitendinosus graft, quintupled, FH Orthopedics tape locking screw (TLS augmented) [17]; graft width was 9 mm. Twin 2 was operated on second with a total operating time of 95 minutes and had a double hamstring semitendinosus/gracilis graft, quadrupled (Smith & Nephew endobutton/PEEK interference screw) [18, 19]; graft width was 7.5 mm.

Post-operative

The twins' peri-operative and acute post-operative comparisons are summarized in Table 2.

Twin 2 had a notably larger opioid analgesia requirement compared to Twin 1. Both patients received routine physiotherapy intervention day 1 post-operatively before being discharged home.

Table 2: Peri-operative details for Twin 1 and 2

	Twin 1	Twin 2
Operation time	• 122 minutes	• 95 minutes
IV antibiotics	• Vancomycin wrap for graft • 3× doses Cefazolin (intra + post-op)	• Vancomycin wrap for graft • 3× doses Cefazolin (intra + post-op)
Orthotic post-operatively	• Richards splint in extension	• Richards splint in slight flexion
Weight-bearing	• WBAT	• WBAT
Other	• PCA overnight • Dressing debulked 3/7 post-op • X-ray Day 1 post-op • Max pain score 3/10	• PCA overnight • Dressing debulked 3/7 post-op • X-ray Day 1 post-op • Max pain score 6/10
Physio day 1	• 1030hrs • Knee flexion 15° • Mobility NAD, independent crutches	• 1000hrs • Knee flexion 20° • Mobility NAD, independent crutches
In-patient medications	• Intra-op femoral nerve block • Nil anti-emetic • Ibuprofen 350 mg ×2 • Oxycodone 5 mg > r/o PCA • Paracetamol 500 mg ×4 • PCA 4.56 mg (bolus only)	• Intra-op femoral nerve block • Nil anti-emetic • Ibuprofen 350 mg ×2 • Oxycodone 5 mg > r/o PCA • Paracetamol 500 mg ×4 • PCA 22.8 mg (background + bolus)
Discharge analgesia	• Oxycodone • Paracetamol • Ibuprofen	• Oxycodone • Paracetamol • Ibuprofen

WBAT: weight bear as tolerated, PCA: patient controlled analgesia, NAD: no abnormality detected, r/o: removal of.

Rehabilitation

Twins 1 and 2 were followed up for two years and their progress was monitored by the same experienced physiotherapist. The twins attended appointments concurrently and performed similar rehabilitation programs. The total number of appointments attended over the two-year follow-up was 18 for Twin 1 and 19 for Twin 2.

Two-year post-operative clinical evaluation

At two years, all multi-disciplinary assessors were blinded to the twins' surgical technique and twin identity during both data collection and analysis. Institutional Human Research Ethics approval was granted, and participants provided informed consent.

Physical assessments were standardized for both twins and chosen according to our hospital's standard 12-month data collection procedure. Participants wore bike shorts, non-impeding shirts, and had similar footwear (Twin 1 and Twin 2 wore the same Nike shoes; the triplet wore similar shoes but different brand). Each participant performed an identical warm-up (jogging, hopping, and jumping on mini trampoline) and was provided scripted verbal instructions for each test with opportunity given for clarification. One practice test was allowed for each test and rest times between tests were standardized. The tests performed were the Y Balance Test (YBT), double jump for distance, vertical jump for height, triple/cross-over hop for distance (quantified with the Limb Symmetry Index—LSI), Forward Step-Down test (FSD), Landing Error Score System (LESS), modified pro-shuffle and the modified agility T-test. The same assessor (a physiotherapist) conducted the tests, and other individuals present only contributed to set-up, timing, distance measuring, and filming. In addition, the Pedi-IKDC (Pediatric International Knee Document Committee) subjective knee form, ACL-RSI (return to sport after injury), KOOS (knee injury and osteoarthritis outcome score), and HSS Pedi-FABS (hospital for special surgery pediatric functional activity brief scale) were used as subjective measures for patient satisfaction, progress, and outcome.

Radiologists reporting on imaging obtained were not informed of the two different reconstruction techniques and therefore reported the scans without bias. Two-year post-operative medical imaging for Twin 1 and Twin 2 was performed as follows:

- MRI (knee coil 3T with 2 mm slices) to assess chondral status, bone marrow (BM) signal, menisci status, ACL graft morphology, growth plate disturbance, position of tunnels and hardware,
- MRI with specific muscle sequencing from anterior superior iliac spine to proximal tibia to assess muscle morphology,
- Bilateral anterior-posterior (AP) lower limb weight-bearing X-rays to assess coronal plane alignment and leg lengths [mechanical axis deviation (MAD), lateral distal femoral angle (LDFA), medial proximal tibial angle (MPTA), leg length discrepancy (LLD)],
- Lateral knee X-rays (affected knee) to assess proximal posterior tibial angle (PPTA).

Anterior/posterior tibiofemoral joint laxity was quantified using the GNRB [20, 21], a device attached to the patient's leg offering precise assessment of ACL laxity, measuring tibiofemoral displacement by performing an automated Lachman test, and obtaining a force-displacement curve.

Two-year post-operative results

The relevant two-year post-operative measurements are summarized in Table 3. Radiographically, Twin 1 had an increased cross-sectional area (CSA) of her ACL graft when compared to both Twin 2 and their sibling control (17.68 mm^2 compared to 16.92 and 17.51 mm^2, respectively). This is not unexpected given that Twin 1 had a quintupled semitendinosus graft, measuring 9 mm in diameter intra-operatively. Magnetic resonance imaging also demonstrated that Twin 2 had a larger side-to-side difference in her medial hamstring bulk at 85.9% compared to 90% of Twin 1, which reflects the difference in harvesting between the two techniques (one hamstring tendon vs two). Long leg films demonstrated Twin 1's operative leg to have a MAD of 1.5 mm, a LDFA of 81.1, and MPTA of 89.8°, and a PPTA of 77° compared to a MAD of also 1.5 mm, LDFA of 83.9°, and a MPTA of 90.3° on the uninjured leg. Twin 2's operative leg had a MAD of 3.5 mm, a LDFA of 80.9°, and MPTA of 89.1°, and a PPTA of 76° compared to a MAD of 2.3 mm, LDFA of 83.1°, and a MPTA of 88.0 on the uninjured leg. We note that no pre- or post-operative lateral knee imaging was performed on the uninjured legs of the twins to obtain a PPTA for comparison, and no

Table 3: Two-year post-operative measurements for Twin 1, Twin 2 and triplet control

Parameter	Twin 1	Twin 2	Control
MRI knee	ACL graft intact PCL intact MCL intact LCL intact Medial mediscus normal Lateral meniscus minor fraying Cartilage normal BM normal	ACL graft intact PCL intact MCL intact LCL intact Medial menuscis normal Lateral meniscus minor fraying Cartilage normal BM normal	All normal
ACL CSA (mm^2)	17.68	16.92	17.51
Medial hamstring CSA (mm^2)	R = 981.31 L = 1091.96 Difference 90%	R = 1084.28 L = 1261.57 Difference 85.9%	-
Long leg films (mm)	R 809 L 810 LLD = 1 Neutral mechanical axis	R 802 L 807 LLD = 5 Neutral mechanical axis	-

ACL: anterior cruciate ligament, PCL: posterior cruciate ligament, MCL: medial collateral ligament, LCL: lateral collateral ligament, CSA: cross sectional area, LLD: leg length discrepancy.

radiology was performed for the control subject as it was not clinically indicated. Measurement of anterior-posterior tibiofemoral joint laxity demonstrated that Twin 1 had less anterior-posterior displacement in response to 150 N (4.9 mm) compared to Twin 2 (7.2 mm) but comparable to the control (5.5 mm average, see Figure 2). Furthermore, side-to-side differences were less when comparing the injured to uninjured knee in Twin 1 (1.4 mm) compared to Twin 2 (3.1 mm). The control demonstrated a very small difference between both of her knees (0.4 mm) showing that there are possibly some minor variations between each patient's native anatomy with regard to laxity about the knees, specifically the ACL, without any pathological cause.

Minor differences were observed between Twin 1 and Twin 2 on physical and functional assessment measures (see Table 4). Twin 1 scored higher on the double jump for distance, LSI (cross over hop), forward step down and agility tests, while Twin 2 scored higher on YBT, vertical jump for of height, and LSI (triple hop). Overall, there were no observed trends in the physical performance measures collected and any differences noted between the twins are unlikely to be clinically meaningful. This premise is supported by the twins' identical scores of 98.91% on the Pedi-IKDC.

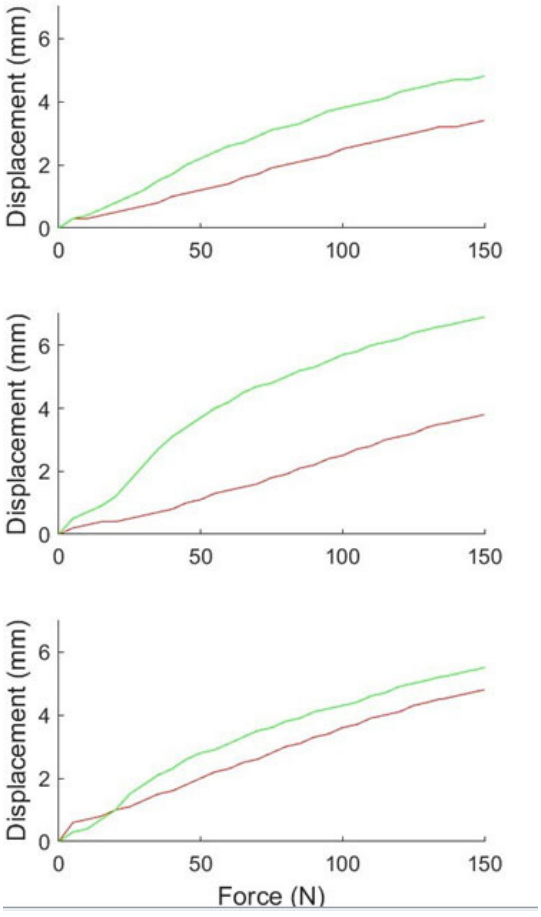

Figure 2: GNRB graft laxity data for Twin 1 (top), Twin 2 (middle), and control (bottom). Green line indicates the right side (i.e., the injury side for Twins 1 and 2) and red indicates the left side.

At the two-year post-operative mark, Twin 1 and Twin 2 had returned to sport, however both played less competitively and less frequently than pre-injury, perhaps a measured decision made by the siblings and their parents in order to maximize recovery and minimize risk of re-rupture.

Table 4: Two-year post-operative physical and functional testing for Twin 1, Twin 2 and triplet control

Parameter	Twin 1	Twin 2	Control
Height (cm)	164.5	162.6	159.8
Mass (kg)	48.7	48.0	50.9
BMI	17.8	18.3	19.8
Menarche	13y3m	13y7m	13y7m
Knee ROM	0–120°	0–120°	0–120°
Hip ROM			
[L/R] IR	75°/75°	80°/80°	60°/60°
[L/R] ER	75°/75°	80°/80°	75°/75°
Beighton score	3	2	1
R YBT	93.33	100.79	99.23
Double jump	3.03 m	2.78 m	3.58 m
Vertical jump	31 cm	35cm	30 cm
Power	589 watts	617 watts	605 watts
Limb symmerty index			
Triple hop	93.85%	96.61%	101.91%
Crossover hop	102.4%	95.28%	110.91%
R FSD	3	2	2
LESS	0	0	0
Agility tests			
R modified pro-shuffle	5.44	6.31	5.88
Modified agility T-test	8.94	9.47	9.59

BMI: body mass index, ROM: range of motion, IR: internal rotation, ER: external rotation, YBT: y balance test, LESS: landing error score system, FSD: forward step down test.

DISCUSSION

We report two-year surgical outcomes for a rare consecutive presentation of ACL-injured identical twins, who subsequently underwent surgery on the same day two months post-injury. Both surgical methods yielded good objective and subjective outcomes at two-year follow-up with no complications. Twin 1 had a longer surgical time (122 vs 95 minutes), while Twin 2 had more post-operative pain and analgesia use, as may be expected from the harvest of double hamstrings. Twin 1 had a thicker graft and hamstring bulk compared to the unaffected side. There was no evidence of chondral and meniscal pathology on MRI or coronal plane mal-alignment on long leg films

for both twins as demonstrated by negligible differences in MAD, LDFA, MPTA, and PPTA between operative and uninjured sides. Of note, Twin 2 had greater graft laxity evidenced from tibiofemoral excursion with a side-to-side difference of 2.2 mm, which may be used to inform further investigations. Quality of life and self-reported function were remarkably similar and analysis of multiple physical outcome measures like range of movement, dynamic balance, neuromuscular control, propulsion, two-dimensional motion analysis, and agility tests found no consistent differences or observable trends between the twins and were comparable to the triplet control [22–32]. Both Twin 1 and Twin 2 have returned to competitive touch football, albeit not at the same level or frequency as pre-operatively, which according to the literature may be advisable in this patient cohort [33–38].

We acknowledge some limitations in this case report. Firstly, although intra-operative findings suggest an equivalent clinical presentation at baseline, we did not collect a pre-operative objective measure of laxity and thus cannot compare our objective laxity results to baseline. Secondly, no pre-operative long leg alignment X-rays were taken, which are now standard protocol at our hospital to quantify the presence of growth disturbance relative to baseline. Thirdly, we note that no pre- or post-operative lateral knee imaging was performed on the uninjured legs of the twins, preventing us from monitoring/comparing PPTA to their normal uninjured leg over time. With regard to data robustness, we feel that our report would have benefited from physical and functional assessments at 6, 12, and 18 months, rather than just at 24 months. Finally, with regard to physical outcome measures; those used are not validated for children therefore normative values are unclear for this age group.

Our case report revealed the twins had similar outcomes following their ACL reconstruction surgeries evidenced by performance and subjective evaluations. There were some minor differences, of note, post-operative analgesia requirements differed for the twins. Also, we highlight objective differences in the twins' hamstring and ACL cross-sectional area as well as anterior-posterior tibiofemoral joint laxity. Historically, the primary outcome measurement used in prospective ACL reconstruction trials is side-to-side difference in passive anterior-posterior knee laxity, obtained via the Lachman and/or the anterior knee draw test. We recommend the use of an objective measurement tool such as the GNRB, as it allows an accurate and reproducible metric, which is likely to be more sensitive to between-group differences. Other measures that should be looked at in conjunction with laxity are growth disturbance rates, knee extension/flexion strength discrepancies, harvested hamstring musculotendon volume/cross-sectional area/length at donor muscle site, volume and cross-sectional area of graft on MRI, and patient-reported outcomes. Given that both ACL reconstruction techniques are hamstring grafts, the subtle difference in the harvesting technique could make enough of a difference post-operatively with regard to pain, rehabilitation, and function to justify one over the other.

CONCLUSION

This case report contributes to the current discussion in the orthopedic and physiotherapy communities regarding increasing rates of ACL rupture in the skeletally immature population, the historical ambiguity regarding management and rehabilitation, appropriate outcome measures to inform practice, and long-term knee health and quality of life outcomes. Careful thought with regard to graft harvesting, preparation technique, and reconstruction technique must be taken to ensure the best possible outcome for the patient while preserving as much of the patient's native anatomy as able and avoiding delays in return to rehabilitation or function post-operatively. Given this prospective case report and the paucity on literature on single versus double hamstring graft techniques in the pediatric population, it is clear that further information must be gathered. It would be of value to replicate the case report for identical male twins to see if outcomes, patient satisfaction, and laxity would be comparable; however, obtaining the same rare scenario of injury, timing, and treatment would be highly unlikely. Furthermore, although this case report examined operative treatment only, it would be of great value to investigate the long-term outcome measures for operative versus non-operative treatment in patients with open physes as there is no true consensus on this in the skeletally immature population.

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Author Contributions

Garrett Malayko – Conception of the work, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Kylie Bradford – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Guarantor of Submission

The corresponding author is the guarantor of submission.

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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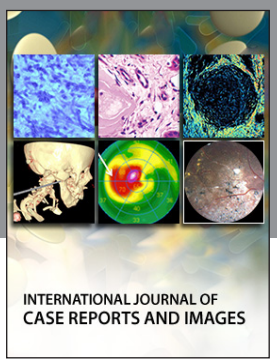

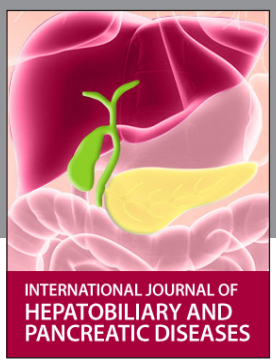

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