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Contents

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ORIGINAL ARTICLE

Retrospective Study

784 Safety and outcomes of hip and knee replacement surgery in liver transplant recipients

Ahmed M, Abumoawad A, Jaber F, Elsafy H, Alsakarneh S, Al Momani L, Likhitsup A, Helzberg JH

791 Time of surgery and surgeon level in supracondylar humerus fractures in pediatric patients: A retrospective study

Albrahim IA, AlOmran AK, Bubshait DA, Tawfeeq Y, Alumran A, Alsayigh J, Abusultan A, Altalib A, Alzaid ZA, Alsubaie SS, Alzahrani MM

Basic Study

800 Automated decision support for Hallux Valgus treatment options using anteroposterior foot radiographs

Kwolek K, Gądek A, Kwolek K, Kolecki R, Liszka H

META-ANALYSIS

813 Long head of biceps tendon transposition for massive and irreparable rotator cuff tears: A systematic review and meta-analysis

Wan RW, Luo ZW, Yang YM, Zhang HL, Chen JN, Chen SY, Shang XL

CASE REPORT

827 Surgical treatment of atlantoaxial dysplasia and scoliosis in spondyloepiphyseal dysplasia congenita: A case report

Jiao Y, Zhao JD, Huang XA, Cai HY, Shen JX

836 Recurrent cyclops lesion after primary anterior cruciate ligament reconstruction using bone tendon bone allograft: A case report

Kelmer G, Johnson AH, Turcotte JJ, Redziniak DE

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Monthly Volume 14 Number 11 November 18, 2023

Contents

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ORIGINAL ARTICLE

Retrospective Study

Safety and outcomes of hip and knee replacement surgery in liver transplant recipients

Mohamed Ahmed, Abdelrhman Abumoawad, Fouad Jaber, Hebatullah Elsafy, Saqr Alsakarneh, Laith Al Momani, Alisa Likhitsup, John H Helzberg

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Abstract

BACKGROUND

Liver transplant (LT) is becoming increasingly common with improved life expectancy. Joint replacement is usually a safe procedure; however, its safety in LT recipients remains understudied.

AIM

To evaluate the mortality, outcome, and 90-d readmission rate in LT patients undergoing hip and knee replacement surgery.

METHODS

Patients with history of LT who underwent hip and knee replacement surgery between 2016 and 2019 were identified using the National Readmission Database.

RESULTS

A total of 5046119 hip and knee replacement surgeries were identified. 3219 patients had prior LT. Mean age of patients with no history of LT was 67.51 [95% confidence interval (CI): 67.44-67.58], while it was 64.05 (95%CI: 63.55-64.54) in patients with LT. Patients with history of LT were more likely to have prolonged

length of hospital stay (17.1% *vs* 8.4%, $P < 0.001$). The mortality rate for patients with no history of LT was 0.22%, while it was 0.24% for patients with LT ($P = 0.792$). Patients with history of LT were more likely to have re-admissions within 90 d of initial hospitalization: 11.4% as compared to 6.2% in patients without history of LT ($P < 0.001$). The mortality rate between both groups during readmission was not statistically different (1.9% *vs* 2%, $P = 0.871$) respectively.

CONCLUSION

Hip and knee replacements in patients with history of LT are not associated with increased mortality; increased re-admissions were more frequent in this cohort of patients. Chronic kidney disease and congestive heart failure appear to predict higher risk of readmission.

Key Words: Liver transplant; Hip replacement surgery; Knee replacement surgery

Core Tip: Patients receiving liver transplants (LTs) are having longer life expectancy. This resulted in an increasing number of patients with LTs requiring hip and knee surgery, with data about their outcomes being limited. The aim of this analysis is to evaluate the safety of these procedures in LT patients and provide more guidance on expected outcomes. This study concluded that LT patients are not at risk of higher mortality albeit increased morbidity.

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INTRODUCTION

Joint replacement surgery is one of the most performed elective surgical procedures. Both total hip arthroplasty (THA) and total knee arthroplasty (TKA) have proven to be highly beneficial orthopedic procedures[1]. Liver cirrhosis is an established risk factor for complications after THA and TKA. Patients with cirrhosis had longer lengths of stay, more frequent discharges to nursing facilities, increased 90-d readmission rate, and higher medical complications including urinary tract infections, acute kidney injury (AKI), need for blood transfusions, gastrointestinal hemorrhage, dislocations, infections, and revisions within 90 d[1]. One-year and longer-term mortality rates were higher in patients with cirrhosis. A model for end-stage liver disease score of 10 or higher predicted a three-fold increased likelihood of complications[1]. A recent meta-analysis examining 527 patients with history of liver pathology undergoing total joint arthroplasty (TJA) showed higher risk of infection and mortality[2].

Liver transplant (LT) is the only curative treatment for patients with liver cirrhosis and end stage liver disease[3]. The prevalence of liver transplantation, which ranks second only to kidney transplantation among solid organ transplantations in the United States, has increased[4]. The increasing 5-year graft survival rate of more than 70% has led to better outcomes with LT[3-5]. With increased life expectancy in the LT cohort, reduced bone mineral density, and higher risk of hip osteonecrosis from immunosuppressive drugs, this cohort becomes at higher risk of needed TKA[6-10].

Arthroplasty is generally safe in healthy, immunocompetent individuals, but its safety in LT patients remains controversial[11]. Previously, a meta-analysis examined the complication rates in 3024 LT patients undergoing THA or TKA [12]. This demonstrated that LT patients benefit functionally from both THA and TKA, but at the cost of increased infection-related complications, reoperation/revision, arthrotomy, and specific medical complications, including AKI and blood transfusion[12]. Other previous studies from small cohorts have addressed the issue of safety but had demonstrated conflicting results[13-17].

Current data describing the mortality rates and readmission rates of these patients after total joint replacement are insufficient to adequately assess complications due to the small cohort size. This makes it difficult to draw conclusions when considering the decision for joint replacement. As more LT patients seek evaluation for degenerative hip disease, a better understanding of the mortality rate and 90-d readmission rates in this high-risk cohort is needed. The aim of this study is to evaluate the mortality outcome and 90-d readmission rates in LT patients undergoing THA and TKA using the National Readmission Database (NRD). Compared to a control cohort, we hypothesize that LT patients undergoing THA or TKA will show a measurable increase in mortality and 90-d readmission rates.

MATERIALS AND METHODS

Patients with history of LT who underwent hip and knee replacement surgery between 2016 and 2019 were included in

the study. Patients were selected from the Healthcare Cost and Utilization Project databases (HCUP). The HCUP databases are sponsored by the Agency for Healthcare Research and Quality. The NRD database is the largest HCUP database and contains unweighted data from over 7 million hospital admissions each year. The data represent a 20% random sample of participating hospital discharges from 46 states. The NRD database is de-identified and available to the public. Thus, it is not considered human subject research and is exempted from review by the institutional review board. To assure a meaningful study cohort the investigators agreed upon a minimum study cohort of 250000 admissions. The International Classification of Diseases Code, 10th Revision Clinical Modification (ICD-10) was used to identify the patients. 90-d readmission rates and diagnoses were identified (Supplementary material). Multiple logistic regression model was used to identify the factors associated with readmission.

Statistical analysis

Continuous variables were described using mean ± SD, while categorical variables were described using proportion (percentage). Categorical variables were compared using the Rao-Scott χ^2 test, and continuous variables were compared using a survey-weighted Student's *t*-test. Multivariate analysis was performed using logistic regression models to decrease bias and adjust for possible confounding factors. Statistical analysis was performed by a biomedical statistician. All analyses were performed using STATA BE 17.

Inclusion criteria

The inclusion criteria including: (1) Patients > 18 years old; (2) Patient with history of LT; and (3) Patient needing hip and/or knee replacement surgery from 2016-2019.

Exclusion criteria

The exclusion criteria including: (1) Patient < 18 years old; (2) Patient needing any other kind of transplant; (3) Patients admitted for other type of orthopedic surgeries; and (4) Patients admitted after more than 90 d of initial admission.

RESULTS

General characteristics

A total of 5046119 hip and knee replacement surgeries were identified between the year 2016 and 2019. 3219 patients had prior LT. Mean age of patients with no history of LT was 67.51 [95% confidence interval (CI): 67.44-67.58], while it was 64.05 (95%CI: 63.55-64.54) in patients with LT. Patients without history of LT had mean length of hospital stay (LOS) 2.87 d (95%CI: 2.84-2.91) as compared to 3.86 d (95%CI: 3.62-4.11), $P < 0.001$. Medicare was the most common primary expected payer. Urban teaching hospitals were the most common (Table 1).

Outcomes

Patients with history of LT were more likely to have prolonged LOS (17.1% *vs* 8.4%, $P < 0.001$), develop AKI (11.1% *vs* 3.2%, $P < 0.001$), and have sepsis (1.2% *vs* 0.3%, $P < 0.001$). The mortality rate for patients with no history of LT was 0.22%, while it was 0.24% for patients with LT. No significant difference was found ($P = 0.792$). Patients with history of LT were more likely to have readmissions within 90 d of initial hospitalization: 11.4% compared to 6.2% in patients without history of LT ($P < 0.001$). The mortality rate between both groups during readmission was not statistically different (1.9% *vs* 2%, $P = 0.871$) respectively. Mortality was mainly caused by sepsis, pulmonary embolism and AKI. There was no significant statistical difference between the 2 groups in rate of prosthetic infections during readmission (10.1% *vs* 12.1%, $P = 0.34$) and gastrointestinal (GI) bleed (4.7% *vs* 5.9%, $P = 0.47$).

Multivariate analysis

Multiple logistic regression analysis showed that the odds ratio (OR) for 90-d readmission in LT patients was 1.54 (95%CI: 1.29-1.84). These readmissions were statistically associated with the presence of chronic kidney disease (CKD) (OR = 1.416, 95%CI: 1.39-1.44) and underlying congestive heart failure (OR = 1.72, 95%CI: 1.69-1.76) (Table 2).

DISCUSSION

Liver transplantation is a lifesaving treatment for patients with end-stage liver disease. With increasing advances in surgical technique, patient/graft selection, and immunosuppression, long-term survival after LT continues to improve. More LT patients are likely to become candidates for THA and TKA due to the increased risk of avascular necrosis and longer patient longevity. Postoperative complications and higher readmission rates are significant concerns in this cohort. While only a few small-cohort studies have reported postoperative complications in LT patients undergoing THA or TKA, no available data suggest readmission rates and predictors of readmissions. This study found 3219 LT patients identified among 5046119 patients who underwent hip and knee replacement surgeries between 2016 and 2019. The mean age of the LT cohort was 64.05 compared to 67.51 in the non-LT cohort. Overall, the LT cohort had a higher readmission rate at 90 d, longer hospital stays, and a higher risk of AKI and sepsis. CKD and congestive heart failure appear to predict a higher risk of readmission at 90 d. No significant difference in mortality rate, prosthesis infection, and gastrointestinal

Table 1 Compares demographics and main outcomes between the two groups during index admission

General characteristics	Patients with history of LT, *n* = 3218	Patients without LT, *n* = 5042901	*P* value
Mean age at admission	64.05 ± 0.49	67.51 ± 0.46	< 0.001
LOS	2.87 ± 0.03	3.86 ± 0.25	< 0.001
Mortality	8 (0.24%)	11154 (0.22%)	0.792
Primary expected payer			
1: Medicare	2104	3056650	< 0.001
2: Medicaid	189	212763	
3: Private	837	1592682	
4: Self-pay	13	26225	
5: No charge	4	3057	
6: Other	71	149442	
Location/teaching status of hospital			
1: Rural	191	498918	< 0.001
2: Urban nonteaching	509	1278335	
3: Urban teaching	2517	3268866	
CHF	263	260627	< 0.001
Renal failure	1461	434965	< 0.001
Coronary artery disease	454	661069	0.276

LT: Liver transplant; CHF: Chronic heart failure; LOS: Length of hospital stay.

Table 2 Predictors of readmissions at 90-d (multivariate analysis)

	Odds ratio, 95%CI	*P* value
Liver transplant	1.54 (1.29-1.83)	< 0.0001
Age	1.01 (1.009-1.01)	< 0.0001
Female	0.86 (0.85-0.87)	< 0.0001
DM	1.19 (1.16-1.22)	< 0.0001
CHF	1.72 (1.69-1.76)	< 0.0001
LOS	1.05 (1.04-1.06)	< 0.0001
Renal failure	1.42 (1.39-1.44)	< 0.0001
Rheumatoid arthritis	1.26 (1.23-1.29)	< 0.0001
Chronic pulmonary disease	1.34 (1.32-1.36)	< 0.0001
Obesity	0.98 (0.97-1.01)	0.78

Multivariate logistic regression model for predictors of readmissions at 90-d for patients with history of liver transplant. CI: Confidence interval; DM: Diabetes mellitus; CHF: Chronic heart failure; LOS: Length of hospital stay.

bleeding was observed.

Recent studies examining the outcomes of THA or TKA after LT are small and limited. There is little data on readmission rates and mortality in this population. It appears that LT patients were more likely to be readmitted within 90 d than control patients (11.4% *vs* 6.2%, $P < 0.001$). Multiple logistic regression analysis showed that the OR for 90-d readmission in LT patients was 1.54 (95%CI: 1.29-1.84). Predictors of increased 90-d readmission rates were also analyzed, including patient's age, LOS, chronic heart failure (CHF), CKD, lung disease, and diabetes mellitus with complications. Among these factors, CHF (OR = 1.72, 95%CI: 1.69-1.76, $P < 0.001$) and CKD (OR = 1.42, 95%CI: 1.39-1.44, $P < 0.001$) predicted a higher risk of readmission.

Mortality during this short follow-up period was also analyzed to better capture the mortality rate associated with TKA/THA compared to other causes. Few studies have reported short-term mortality risk. Inacio *et al*[18] reported a 1-year mortality rate of 0.5% in the 45-54 age group. Another meta-analysis reported that 1-year mortality year for the LT cohort undergoing THA or TKA was 4.35%. The observed one-year mortality rate in LT patients is more like cirrhotic patients undergoing THA and TKA, which was measured at 5% in 115 patients by Tiberi *et al*[1]. This study did not find an increased mortality rate among the LT group (0.24% in LT compared to 0.22 % in controls, $P = 0.792$). In addition, there was no significant difference in mortality rates between the two groups during readmission (1.9% *vs* 2%, $P = 0.871$).

Suppression of humoral immunity by lifelong immunosuppressants increases postoperative complications[17,19,20]. The incidence of systemic infections, particularly sepsis and AKI was significantly increased in the LT cohort after 90 d compared to the control group. The LT cohort in our study had a higher risk of AKI than controls (11.1% *vs* 3.2%, $P < 0.001$). A meta-analysis of 3024 LT patients reported a higher risk of complications in the LT group, with AKI being the highest OR among all complications[12]. The trend towards higher AKI rates has also been reported in patients with cirrhosis after arthroplasty. Tiberi *et al*[1] found a higher risk of AKI in patients with cirrhosis compared to controls after 90 d (10% *vs* 1%). In addition, significantly higher AKI rates were found in other types of transplantation. Choi *et al*[21] reported higher odds (OR = 22.25, $P < 0.001$) of developing AKI in the kidney and LT after THA compared to controls. Cavanaugh *et al*[22] reported a higher incidence in the kidney (OR = 3.48) and in cardiac, lung, and pancreas transplant patients (OR = 4.42) receiving TKA and THA, while Klement *et al*[19] reported significantly higher AKI rates in the kidney (OR = 6.03), lung (13.180), heart (19.660) and pancreas (7.780) transplanted patients who received TKA at 90 d. Post operative AKI can be linked to poor patient outcomes, including increased length of stay, cost and increased probability of discharging to extended-care facilities. Additionally, postoperative AKI is responsible for 25%-90% of in-hospital deaths[23].

Increased susceptibility to systemic and local infections is a major concern in LT patients undergoing THA or TKA. A meta-analysis by Han *et al*[12] reported higher rates of local and systemic infections both at 90 d and at all time points in the LT cohort compared to the control group. Additionally, this study found that LT patients undergoing TKA, compared to those undergoing THA, had a higher rate of joint infection at all time points. Onochie *et al*[24] had similar findings, a meta-analysis of chronic liver disease patients undergoing THA *vs* controls, demonstrated infection rates of 0.5% at a mean follow-up of 13.5 mo. The finding of higher rates of infection in patients with cirrhosis after arthroplasty is a general trend. While Jiang *et al*[25] found that a diagnosis of cirrhosis is the highest independent risk factor for periprosthetic infection, Deleuran *et al*[26] reported higher rates of infection in cirrhotic patients undergoing THA or TKA at one year with an incidence of 3.1% *vs* 1.4% in controls. Higher infection rates after arthroplasty are also observed in other types of organ transplant patients[19,22]. Our study found an increased risk of sepsis in the LT group (1.2% *vs* 0.3%, $P < 0.001$), but no significant difference between prosthetic infections was observed (10.1% *vs* 12.1%, $P = 0.34$). Delayed infection, however, remains a known risk.

The risk of bleeding tendency remains a paramount consideration with THA and TKA after LT. Hepatic reserve is a major concern, as a decrease in liver functions can lead to a decrease in platelets and clotting factors, and an increased risk of bleeding. Oya *et al*[16] study reported an increased incidence of intraoperative blood loss in a small cohort of 7 patients (303.6 mL *vs* 163.4 mL, $P < 0.01$). A few studies have also reported increased blood transfusion rates in the LT group[13,19,20,22]. This study reported no significant difference in GI bleeding between the two groups (4.7% *vs* 5.9%, $P = 0.47$). Nevertheless, careful assessment and evaluation of the bleeding profile should be performed in this population before considering THA or TKA.

In summary, LT patients appear to have longer hospital stays compared to nontransplant recipient patients (17.1% *vs* 8.4%, $P < 0.001$). This is consistent with the study of Aminata *et al*[13], which found a significant difference between the LOS in 33 LT who underwent THA compared to non-transplant patients (16 d *vs* 10 d).

Strengths of this study include an adequate sample size to report readmission rates, predictors of readmission, short-term mortality risk, and complication rates statistically and accurately in the LT cohort. To the authors' knowledge, this is the first study to examine the 90-d readmission rates in LT *vs* a control cohort for patients after THA or TKA. The coding and size of the database allowed comparison between the LT cohort and a large control cohort. This also enabled multiple logistic regression analysis to analyze predictors of readmissions.

This study is not free from limitations. Surgical variables unique to transplantation were not available, such as the regimen, dose and duration of immunosuppression, and the type of transplant (related donor). Also, the indication of joint replacement is not available. Another limitation is unavailability of the temporal relationship between the timing of LT and joint replacement in addition to degree of immunosuppression and performance status of patients and laboratory results.

Like any database query using ICD-10 codes, the quality of the data depends on the accurate coding at the time of the patient encounter. Although this data was collected by the United States NRD, this could encourage other countries with national registries to perform a similar analysis to see if these results can be generalized outside the United States. Finally, patient satisfaction, hip and knee outcome scores, and functional measurements were not included in this database.

CONCLUSION

In conclusion, THA or TKA after LT is not associated with measurably increased mortality but has an increased risk of 90-d readmission. The most prominent risk factors for readmissions are CHF and CKD. While the LT cohort showed an increased risk of AKI and sepsis, there was no increased risk of gastrointestinal bleeding and prosthesis infection. Careful patient selection and medical optimization can reduce readmission rates, risk of mortality, and postoperative complic-

ations in LT patients undergoing THA or TKA. Hopefully the results of this study will provide orthopedic surgeons with accurate readmission profiles to appropriately counsel their patients about the inherent readmission risks after THA and TKA, in LT patients.

ARTICLE HIGHLIGHTS

Research background

Solid organ transplants are rising with recipients having longer life span. This puts them at risk of needing joint replacement surgery during their life time. The outcomes of these surgeries are understudied which raises the need for studies to evaluate benefits and risks in this cohort.

Research motivation

The question is whether patients with liver transplant (LT) are at increased risk of developing complication or have a higher mortality when needing hip or knee replacement surgery.

Research objectives

The main objective is to prove the LT patients are not at increased risk of complications when needing hip or knee replacement surgery which will allow these patients to get this surgery when needed. Also, this study aims to identify factors associated with increased morbidity which can help modify these factors.

Research methods

Patients were selected from the Healthcare Cost and Utilization Project databases (HCUP). The HCUP databases are sponsored by the Agency for Healthcare Research and Quality. The International Classification of Diseases Code, 10th Revision Clinical Modification was used to identify the patients.

Research results

Patients with a history of LT undergoing knee or hip replacement have longer hospital stay, increase morbidity but no increase mortality as compared to patient with no history of LT.

Research conclusions

The results show that hip and knee replacement are safe procedures in patients with LT.

Research perspectives

More research is needed in identifying risk scores to stratify LT patients as either high or low risk for joint replacement surgery.

FOOTNOTES

Author contributions: Ahmed M contributed to the conception and literature review of the manuscript; Ahmed M, Abumoawad A, and Elsafy H designed this study; Ahmed M and Jaber F drafted the manuscript; Abumoawad A collected the data; Jaber F, Al Momani L, Likhitsup A, and Helzberg JH involved in the critical reviewing of the manuscript; Elsafy H contributed to the analytic plan; Helzberg JH supervised and edited the manuscript.

Institutional review board statement: This study is performed based on the Healthcare Cost and Utilization Project databases. The HIPAA Privacy Rule sets national standards for patient rights with respect to health information. This rule protects individually identifiable health information by establishing conditions for its use and disclosure by covered entities.

Informed consent statement: This study is performed based on the Healthcare Cost and Utilization Project databases. The HIPAA Privacy Rule sets national standards for patient rights with respect to health information. This rule protects individually identifiable health information by establishing conditions for its use and disclosure by covered entities.

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ORIGINAL ARTICLE

Retrospective Study

Time of surgery and surgeon level in supracondylar humerus fractures in pediatric patients: A retrospective study

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Abstract

BACKGROUND

Supracondylar humerus fractures account for more than 60% of all elbow fractures and about 1/5 of all pediatric fractures. Unfortunately, these fractures can be associated with risk of complications including neurovascular injuries, malunions and limb deformities. Controversy exists regarding the effect of time of surgical intervention and/or level of surgeon performing the surgery on outcome of these fractures.

AIM

To determine whether time of surgical intervention and/or surgeon level influence the outcomes of surgically managed pediatric supracondylar humerus fractures.

METHODS

We retrospectively studied 155 pediatric patients presenting with a supracondylar humerus fracture in a level 1 trauma center from January 2006 to December 2019. The data extracted included demographic data, fracture characteristics, surgical data, and follow-up outcomes. The collected data was analyzed and *P* values of < 0.05 were considered statistically significant.

RESULTS

Of the cohort, 11% of patients had documented post-operative complications, of which the majority occurred in surgeries performed after day time working hours and in fractures requiring open reduction. While the lowest complication rate was

found in surgeries performed by pediatric orthopaedic surgeons, this did not reach statistical significance.

CONCLUSION
In pediatric patients undergoing surgery for supracondylar fractures, we found a higher complication rate when surgeries were not performed during working hours. Surgeon level and training had no significant effect on the risk of post-operative complications.

Key Words: Supracondylar humerus; Fracture; Time of surgery; Level; Complications

Core Tip: In our pediatric cohort of surgically treated supracondylar humerus fractures we found a higher complication rate when surgeries were not performed during working hours. Surprisingly, surgeon level and training had no significant effect on the risk of post-operative complications.

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INTRODUCTION

Supracondylar humeral fractures (SCHF) account for 16% of all paediatric fractures and are commonly sustained by children younger than eight years of age[1-4]. These injuries commonly occur after a fall on an outstretched hand irrespective of forearm position[5-7]. The majority of these fractures require closed reduction and pinning, while open reduction is reserved for some complex patterns, delayed presentation or when closed reduction fails[8-10]. When compared to closed reduction and fixation, open reduction carries higher risk of complications, especially infection and loss of elbow range of motion[8,11-13].

Timing of surgical intervention for these injuries is an area of controversy, as proponents for emergent intervention prefer it before any surgical site swelling occurs to minimize complications including nerve injury and compartment syndrome. In addition, it is believed that early surgical management can improve the success of closed reduction in these patient[12]. Surgeons who prefer a more delayed but urgent approach believe that operating on these fractures with an experienced team and during the daytime hours will lead to a quicker and more efficient surgical experience and decrease the associated post-operative complications[14,15].

The purpose of our study was to explore outcomes of surgical management of SCHF in pediatric patients and to assess if timing of surgical intervention and/or surgeon level would influence the outcomes of these patients post-operatively.

MATERIALS AND METHODS

This is a retrospective cohort study conducted in a level 1 trauma center from January 2006 to December 2019. After ethical approval was obtained from the institutional review board in our institute (IRB-UGS-2019-01-333), all pediatric patients 1 to 14 years old who sustained a SCHF and were treated surgically with a follow up period of at least 24 mo were included. Exclusion criteria were patients who were lost to follow up, had incomplete data points in their charts or had inadequate radiographs. In addition, Gartland type I fractures were excluded as these all were managed non-surgically in our institution.

All surgeries were performed by pediatric orthopaedic trained consultants, board certified orthopaedic consultants, fellows, or residents (with consultant supervision). Cases were booked in the dedicated emergency room by the admitting consultant and surgery is performed according to urgency level decided by the admitting consultant. Cases admitted after day time working hours by non-pediatric orthopaedic consultants are repatriated to a pediatric orthopaedic consultant the next morning if not done. Operative fixation was performed under general anaesthesia, with the patients in supine position. The operative extremity was prepped and draped in standard fashion and pre-operative weight appropriate prophylactic antibiotics were administered. A trial of closed reduction was performed and if adequate percutaneous fixation with k-wires was performed (configuration and number were left to surgeon discretion). If closed reduction was unsuccessful, an open reduction was performed. After fixation, a sterile dressing was applied to the pin sites and an above elbow back-slap was applied in neutral position. Patients were followed up in clinic at 2 wk post-operatively, then at the 6-week post-operative mark where pins were removed, and patients were allowed to start range of motion of the elbow. Patients were then seen at 3-, 6- and 12-mo post-surgery.

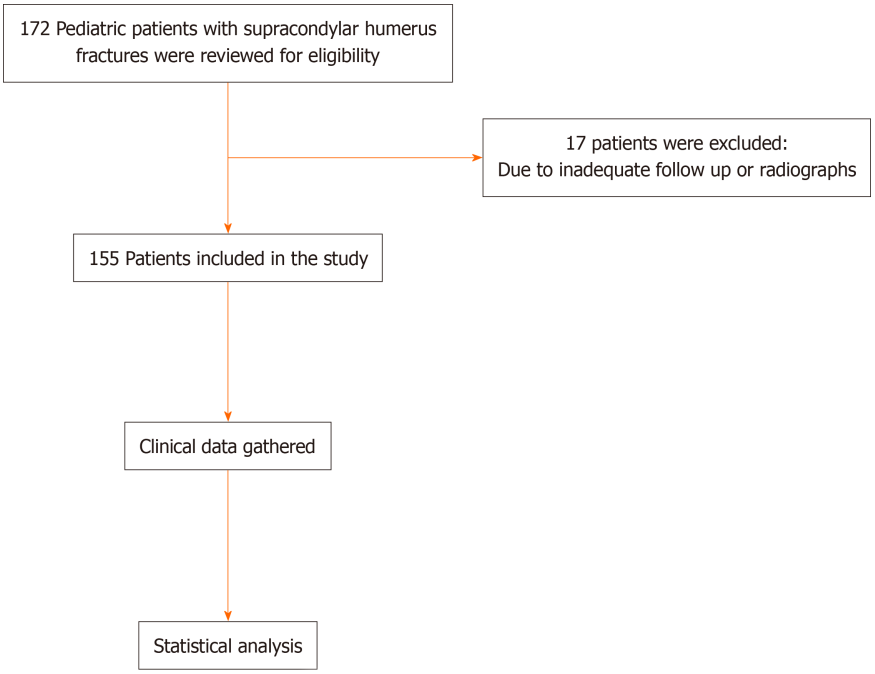

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Figure 1 Patient flowchart.

Data was extracted from the hospital electronic medical records, which included patient demographics data (age and gender), in addition to fracture characteristics and outcome variables. The collected variables included Gartland classification, time of surgery (during working hours "08:00-15:00" *vs* after day time working hours "15:00-8:00"), surgeon level (trainees "fellows or residents under supervision", non-pediatric orthopedic surgeons, and pediatric orthopedic surgeons), nerve or vessel injury (both pre- and post-operative), associated complications (including compartment syndrome and pin site infection) and fixation construct. Post-operative radiographic alignment was assessed on plain antero-posterior and lateral elbow radiographs. Inadequate reduction was defined according to Flynn's criteria (Baumann angle < 10°, displacement > 4 mm and anterior humeral line not bisecting the capitellum)[16].

A power analysis was performed with data from Saarinen *et al*[17], using a power of 80% and $\alpha = 0.05$, we estimated that we would require a sample size of 59 patients. Data was analyzed using Statistical Package for Social Sciences version 27 (IBM Corp, 2017). Categorical variables were described as frequencies and percentages, while continuous variables were described as mean and standard deviation. Bivariate analysis was tested using Chi-square test and ANOVA were applicable, with a *P* value of less than 0.05 considered as significant. Odds ratios with 95% confidence intervals were calculated to examine the association between the outcome variables and patient or fracture factors.

RESULTS

A total of 172 patients were reviewed and 155 were included in the study with a mean age of 5.36 years old (range 1-13 years old) (Figure 1). More than half of the patients were males 105 (67.7%). Of the patients included in the cohort, 2.6% had a documented nerve injury pre-operatively (half of which were managed during working hours), these injuries included two cases of median and two cases of radial nerve palsies. An absent radial pulse was found in 9% of patients (half of which were also managed during working hours), all patients regained pulses after manual reduction during the operative procedure (Table 1). More than half of patients (56.1%) were managed during working hours, while 43.9% were managed after day time working hours. The majority of fractures were managed by closed reduction and k-wire fixation (84.5%) and parallel K-wires were the most applied construct (48.4%) (Table 1).

Twenty-four cases (15.5%) required an open reduction, 3 of which had an absent pulse pre-operatively which was regained after fracture reduction. More than half of the open reduction cases were performed by trainees with consultant supervision (58.3%), while 20.8% were performed by pediatric orthopaedic surgeons and 20.8% performed by non-pediatric orthopaedic surgeons.

Of the cohort, 17 patients (11%) had documented post-operative complications. These complications included four cases of nerve palsy post-operatively, three of which were fixed with two lateral and one medial k-wires (two had a median nerve injury and one ulnar nerve injury) and one fixed with two lateral parallel k-wires (sustained a median nerve injury). The remaining complications were 12 cases of unacceptable angular deformity post-fixation and one case of bony spur requiring operative management. Patients who were managed after day time working hours were associated with more complications when compared with daytime surgeries (5.7% *vs* 17.6%, *P* value < 0.05). Patients who had their surgeries done after day time working hours were nearly at a 3 times higher risk of developing complications (OR = 2.8, *P*

Table 1 Demographic data of supracondylar fracture cohort

Variables, *n* (%)		
Time of surgery	During working hours	87 (56.1)
	After working hours	68 (43.9)
Surgeon level	Pediatric orthopedic consultant	44 (28.4)
	Non-pediatric orthopedic consultant	38 (24.5)
	Trainee	73 (47.1)
Gender	Male	105 (67.7)
	Female	50 (32.3)
Mechanism	Fall on outstretched hand	112 (72.3)
	Fall on flexed elbow	17 (11)
	Direct	15 (9.7)
	Motor vehicle accident	8 (5.2)
	Others	3 (1.9)
Pre-operative nerve injury	Yes	4 (2.6)
	No	151 (97.4)
Pulse	Intact	141 (91)
	Absent	14 (9)
Classification	Gartland type 2	32 (20.6)
	Gartland type 3	115 (74.2)
	Flexion type	8 (5.2)
Procedure	Open reduction	24 (15.5)
	Close reduction	131 (84.5)
K-wire arrangement	Parallel	75 (48.4)
	Medial & lateral	35 (22.6)
	Two lateral & one medial	34 (21.9)
	Others	11 (7.1)

value < 0.05) when compared to surgeries done during the daytime.

In addition, patient who required an open reduction had a higher rate of complications (33.3%) when compared to closed reduction (6.9%, *P* value < 0.001) (Table 2). Complications recorded with open reduction were 5 cases of angular deformity, one case of ulnar nerve palsy and one case of boney spur. The odds of a complication occurring was more than 6 times greater with an open reduction when compared to a closed reduction (OR = 6.8, *P* value < 0.005). We found no difference in rate of complication between Gartland type 2, type 3 and flexion type SCHFs.

Interestingly, although the lowest rate of complications was in surgeries performed by pediatric orthopaedic surgeons, this was not statistically significant (Table 2). There was no significant difference in Gartland classification, K-wire arrangement, pre-operative nerve injury and absent pulse between cases done by different levels of surgeons. Concerning timing of surgery and the surgeon performing the surgery, the majority (57.4%) of after day time working hours surgeries were performed by trainees, while only 25% were performed by non-pediatric orthopaedic surgeons and 17.6% by pediatric orthopaedic surgeons (*P* value < 0.05).

We also assessed the effect of surgeon level on complication rate in surgeries performed during working hours and surgeries performed after day time working hours, separately, and found no statistically significant difference.

DISCUSSION

The aim of this study was to investigate and analyze factors that could have an influence on outcomes of SCHF in pediatric patients, including time of surgery and surgeon level. In our cohort, we found that majority of complications occurred in cases performed after day time working hours, this may be attributed to that more severe injuries or cases with associated neurovascular compromise may be taken more urgently during the night. Aydoğmuş *et al*[18] reported

Table 2 Percentages of complications by variable and *P* value

Variables		Complication, *n* (%), *n* = 155		
		Yes, *n* = 16 (10.3)	No, *n* = 139 (89.7)	*P* value
Time of surgery	During working hours	5 (5.7)	82 (94.3)	**0.019**
	After working hours	12 (17.6)	56 (82.4)	
Surgeon level	Pediatric orthopaedic consultant	4 (9.1)	40 (90.9)	0.841
	Non-pediatric orthopaedic consultant	5 (13.2)	33 (86.8)	
	Trainee	8 (11)	65 (89)	
Gender	Male	10 (9.5)	95 (90.5)	0.404
	Female	7 (14)	43 (86)	
Classification	Gartland type 2	2 (6.3)	30 (93.7)	0.308
	Gartland type 3	13 (11.3)	102 (88.7)	
	Flexion type	2 (25)	6 (75)	
Procedure	Open reduction	8 (33.3)	16 (66.7)	**0.001**
	Close reduction	9 (6.9)	122 (93.1)	
K-wire arrangement	Parallel	6 (8)	69 (92)	0.154
	Medial & lateral	4 (11.4)	31 (88.6)	
	Two lateral & one medial	7 (20.6)	27 (79.4)	
	Others	0 (0)	11 (100)	

inferior fixation of 91 pediatric patients with operatively managed SCHF during after-hours when compared to cases performed during the daytime. In their cohort, they found no difference operative time, requirement of open reduction nor patient outcomes. When comparing early (< 12 h) and delayed (> 12 h) surgical fixation of pediatric SCHF, Suganuma *et al*[19] and Gupta *et al*[20] found no difference in rate of open reduction, post-operative complications nor surgical time between the two groups. In addition, Mehlman *et al*[15] found no difference in rate of open reduction, pin tract infection nor iatrogenic nerve injury between pediatric SCHF performed within 8 h of injury and more than 8 h of injury. Multiple other studies explored differences in outcomes and complication rates when surgery is done early or in a delayed fashion for SCHF, with the majority finding no statistically significant differences (Table 3).

Considering the higher complication rate in after day time working hours surgery and the lack of increased risk of adverse events in delayed fixation of SCHF, it seems clear that unless the extremity is pulseless with lack of perfusion, the fracture is open or there are signs of compartment syndrome, SCHF in pediatric patients should preferably be done during a reasonable time of day and not late after day time working hours.

While the majority of the complications in our cohort were in the surgeries that were conducted by trainees and non-pediatric orthopaedic surgeons, this was not statistically significant. When exploring the literature, we find that this is an area of controversy, as several studies have shown that the level and training of the surgeon has a significant effect on both outcome and complication rate of pediatric SCHF (Table 4). In addition, studies have also shown that when the procedure is supervised by a pediatric orthopedic surgeon, the outcomes are excellent with very low complication and revision rates irrespective of the level of the surgeon performing the surgery[21,22]. On the other hand, several studies have shown no difference in complication rate and risk of malreduction when the surgery is performed by a trained orthopaedic surgeon without pediatric fellowship training[17,23,24].

Limitations of the study include that it is a retrospective cohort, but we believe that we were successful in data collection from the patients' medical records with good accuracy. Another possible limitation is the small sample size, but we believe that our study is adequately powered based on the sample size analysis using previous published studies. In addition, the number of complications in our study was relatively low, but the proportion of complications was similar in previously published studies.

CONCLUSION

In pediatric patients undergoing surgery for SCHF, we found a higher complication rate when surgeries were not performed during working hours, or when an open reduction is required. Although pediatric orthopaedic surgeons had the lowest rate of complications, this difference did not reach statistical significance. We believe the data presented in this study can help in reaching a better-informed decision about the timing of surgery for pediatric patients with SCHF.

Table 3 Previous studies assessing effect of time of surgery on outcomes of pediatric supracondylar humeral fractures

Ref.	Sample size	Groups	Results
Suganuma *et al* [19], 2020	120 Gartland type II and III SCHF	Surgeries within 12 h of injury and > 12 h	No difference in: Operative time; Early complications; Post-operative radiographic parameters
Aydoğmuş *et al* [18], 2017	91 Gartland type III SCHF	Daytime *vs* after hours surgery	Poor fixation rate in after hour surgery; No difference in operative time or residual deformity
Schmid *et al*[25], 2015	343 Gartland type II and III SCHF	Surgeries within 6 h, 6-12 h, 12-24 h and > 24 h from injury	No difference in: Outcome; Complications; Requiring open reduction
Mayne *et al*[26], 2014	115 Gartland type II and III SCHF	Surgeries within 12 h of injury and > 12 h	No difference in: Infection rate; Iatrogenic nerve injury; Requiring open reduction
Larson *et al*[27], 2014	399 Gartland type II SCHF	Surgeries within 24 h of injury and > 24 h	No difference in complication rate
Yildirim *et al*[28], 2009	190 Gartland type III SCHF	Time from injury to surgery	4 fold increase in requiring open reduction for each 5 hour delay
Walmsley *et al*[29], 2006	171 Gartland type III SCHF	Surgeries within 8 h of injury and > 8 h	No difference in complication rate; Higher risk of requiring open reduction in > 8 h group
Sibinski *et al*[30], 2006	77 Gartland type III SCHF	Surgeries within 12 h of injury and > 12 h	No difference in: Operative time; Outcome; Risk of open reduction
Gupta *et al*[20], 2004	150 operatively treated SCHF	Surgeries within 12 h of injury and > 12 h	No difference in: Pin tract infection; Iatrogenic nerve injury
Mehlman *et al*[15], 2001	198 operatively treated SCHF	Surgeries within 8 h of injury and > 8 h	No difference in: Pin tract infection; Iatrogenic nerve injury
Iyengar *et al*[31], 1999	58 Gartland type III SCHF	Surgeries within 8 h of injury and > 8 h	No difference in: Open reduction rate; Clinical outcomes

SCHF: Supracondylar humeral fractures.

Table 4 Previous studies assessing effect of surgeon level on outcomes of pediatric supracondylar humeral fractures

Ref.	Sample size	Groups	Results
Fisher *et al*[23], 2021	231 patients who underwent CRPP for SCHF	Pediatric orthopaedic fellowship trained *vs* other orthopaedic surgeons	Shorter operative and fluoroscopy time; No difference in complications
Osateerakun *et al* [32], 2019	87 Gartland type II and III SCHF	Pediatric orthopaedic fellowship trained *vs* other orthopaedic surgeons	Higher risk of complications in Gartland type III when not performed by pediatric orthopaedic surgeon; Overall complication rate and acceptable alignment were similar
Saarinen *et al* [17], 2019	108 operatively treated SCHF	Residents, pediatric surgeons and orthopaedic surgeons	Orthopaedic surgeons had the least complications and inadequate reductions; Residents had less complications and inadequate reductions when compared to pediatric surgeons
Pesenti *et al*[33], 2018	236 Gartland type III SCHF	Surgeons with < 1 yr *vs* > 1 yr experience	Less experienced had longer operative time; No difference in complication and malalignment rate
Tuomilehto *et al* [34], 2018	210 operatively treated SCHF	Consultants *vs* registrars	Higher complications and poorer outcomes in surgeries done by registrars
Liu *et al*[22], 2011	654 operatively treated SCHF	Fellows progression through fellowship training	No difference in complications and malunions throughout the fellowship year; Spike of malreductions at case 7 which improves at case 15
Padman *et al*[21], 2010	71 Gartland type II and III SCHF	Consultants *vs* trainees	Poorer outcome and more complications in surgeries performed by trainees without consultant supervision
Farley *et al*[24], 2008	444 operatively treated SCHF	Pediatric orthopaedic surgeon *vs* non-pediatric orthopaedic surgeon	No difference in complication rate and outcomes

CRPP: Closed reduction percutaneous pinning; SCHF: Supracondylar humeral fractures.

ARTICLE HIGHLIGHTS

Research background

Pediatric supracondylar humerus fractures can be associated with risk of complications including neurovascular injuries, malunions and limb deformities. Surgical timing and level of surgeon performing the surgery may have an effect on outcome of these fractures.

Research motivation

Explore the effect of time of surgery and level of surgeon on pediatric supracondylar humerus fractures.

Research objectives

The objective of this study was to determine whether time of surgical intervention and/or surgeon level influence the outcomes of surgically managed pediatric supracondylar humerus fractures.

Research methods

We retrospectively studied 155 pediatric patients presenting with a supracondylar humerus fracture in a level 1 trauma center from January 2006 to December 2019. The data extracted included demographic data, fracture characteristics, surgical data, and follow-up outcomes. The collected data was analyzed and P values of < 0.05 were considered statistically significant.

Research results

Of the cohort, 11% patients had documented post-operative complications, of which the majority occurred in surgeries performed after day time working hours and in fractures requiring open reduction. While the lowest complication rate was found in surgeries performed by pediatric orthopaedic surgeons, this did not reach statistical significance.

Research conclusions

In pediatric patients undergoing surgery for supracondylar fractures, we found a higher complication rate when surgeries were not performed during working hours. Surgeon level and training had no significant effect on the risk of post-operative complications.

Research perspectives

We believe the data presented in this study can help in reaching a better-informed decision about the timing of surgery for pediatric patients with supracondylar humeral fractures.

FOOTNOTES

Author contributions: Albrahim IA, Alomran AK, and Bubshait DA contributed to concept; Albrahim I, Alomran AK, Bubshait DA, Tawfeeq Y, Alsayigh J, Abusultan A, Altalib A, Alzaid ZA, Alzahrani MM contributed to design; Albrahim I, Alomran AK, and Bubshait DA, Tawfeeq Y, Alumran A, Alsayigh J, Abusultan A, Altalib A, Alzaid ZA, and Alsubaie SS contributed to study execution; Albrahim I, Alomran AK, and Bubshait DA, Tawfeeq Y, Alumran A, Altalib A, Alzaid ZA, Alsubaie SS, Alzahrani MM contributed to manuscript writing and review; Alumran A contributed to statistical analysis; Alsubaie SS contributed to statistical analysis.

Institutional review board statement: Ethical approval was obtained from the Imam Abdulrahman Bin Faisal University review board (IRB-UGS-2019-01-333).

Informed consent statement: No informed consent was required for patients in this study as it was a retrospective chart review.

Conflict-of-interest statement: All the authors have no relevant conflict of interests.

Data sharing statement: Raw data and material are available as needed.

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ORIGINAL ARTICLE

Basic Study

Automated decision support for Hallux Valgus treatment options using anteroposterior foot radiographs

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Abstract

BACKGROUND

Assessment of the potential utility of deep learning with subsequent image analysis to automate the measurement of hallux valgus and intermetatarsal angles from radiographs to serve as a preoperative aid in establishing hallux valgus severity for clinical decision-making.

AIM

To investigate the accuracy of automated measurements of angles of hallux valgus from radiographs for further integration with the preoperative planning process.

METHODS

The data comprises 265 consecutive digital anteroposterior weightbearing foot radiographs. 181 radiographs were utilized for training (161) and validating (20) a U-Net neural network to achieve a mean Sørensen–Dice index > 97% on bone segmentation. 84 test radiographs were used for manual (computer assisted) and automated measurements of hallux valgus severity determined by hallux valgus (HVA) and intermetatarsal angles (IMA). The reliability of manual and computer-based measurements was calculated using the interclass correlation coefficient (ICC) and standard error of measurement (SEM). Inter- and intraobserver reliability coefficients were also compared. An operative treatment recommendation was then applied to compare results between automated and manual angle measurements.

RESULTS

Very high reliability was achieved for HVA and IMA between the manual measurements of three independent clinicians. For HVA, the ICC between manual measurements was 0.96-0.99. For IMA, ICC was 0.78-0.95. Comparing manual against automated computer measurement, the reliability was high as well. For HVA, absolute agreement ICC and consistency ICC were 0.97, and SEM was 0.32. For IMA, absolute agreement ICC was 0.75, consistency ICC was 0.89, and SEM was 0.21. Additionally, a strong correlation (0.80) was observed between our approach and traditional clinical adjudication for preoperative planning of hallux valgus, according to an operative treatment algorithm proposed by EFORT.

CONCLUSION

The proposed automated, artificial intelligence assisted determination of hallux valgus angles based on deep learning holds great potential as an accurate and efficient tool, with comparable accuracy to manual measurements by expert clinicians. Our approach can be effectively implemented in clinical practice to determine the angles of hallux valgus from radiographs, classify the deformity severity, streamline preoperative decision-making prior to corrective surgery.

Key Words: Computer-aided diagnosis; Artificial intelligence in orthopedics; Automated preoperative decision support; Deep learning; Medical imaging

Core Tip: This study presents an accurate method for automated assessment of angles of hallux valgus on high-resolution weight-bearing anteroposterior feet radiographs. Reference points are estimated according to the AOFAS standard on automatically segmented bones of the foot. The proposed method accurately calculates angles even in the case of significant toe deformity automating preoperative decision-making. Experimental results revealed high reliability of hallux valgus angle and intermetatarsal angle measurements between the proposed algorithm and medical doctors, achieving a correlation of almost 80%.

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INTRODUCTION

Hallux valgus (HV) is a foot deformity that affects a considerable percentage of the population[1,2]. It is a complex positional deformity of the first ray that leads to altered joint mechanics, dysfunction, and progressive pain. The technique of weightbearing dorsoplantar radiographs was standardized and determined in the AOFAS research committee report[3-6]. Orthopedic surgeons frequently use radiographic angles to make clinical decisions for patients with symptomatic HV[7-9]. Various radiographic measurements used in hallux valgus treatments were discussed[3,10]. The reliability of radiographic measurements in HV was also studied[11]. Through the use of WBCT scans, it has been demonstrated that up to 87% of hallux valgus cases exhibit metatarsal bone pronation, emphasizing the intricate multiplanar nature of this deformity. This metatarsal pronation explains the perceived metatarsal bone shape and the misalignment of the medial sesamoid bone in radiological studies, which has been recognized as a significant factor contributing to recurrence following treatment. As a result, distal metatarsal articular angle has proved unreliable, demonstrating a poor interobserver agreement[12,13]. Further research is needed to develop effective approaches for addressing the rotational deformity in individuals with HV[2,14,15].

Key angles utilized in clinical practice to establish the severity of HV are the hallux valgus angle (HVA) and the intermetatarsal angle (IMA)[8,12,13,16-18]. Intra- and inter-observer agreement for radiographic measurement of HVA/IMA is reportedly good using various digital techniques[11,17,19]. The HVA is between the longitudinal axes of the first metatarsal and the proximal phalanx (PP). The IMA is between the longitudinal axes of the first and second metatarsal bones (Figure 1). Many methods were used to facilitate and accelerate manual or computer-assisted determination of the longitudinal axes of the first, second metatarsal (1,2MT) and the hallucial PP bones, *e.g.*, establishing reference points, to make measurements more repeatable[7,20].

Traditionally, these angles were manually measured on hard-copy radiographs. Nowadays, computer-assisted measurement methods are being developed, that reduce the measurement error of HVA[21-23]. New possibilities for radiographic images analysis have emerged thanks to recent advances in clinical applications of deep learning[24-29]. This study is among the first forays into automated HVA/IMA measurements from radiographs.

Kwolek *et al*[30] introduced an algorithm for the automatic recognition of radiographs of the hallux valgus using U-Net neural network with promising outcomes. A study on hallux valgus measurement with a deep convolutional neural

network based on landmark detection has been discussed by Li *et al*[31]. In contrast to our approach based on the toe bones segmentation and reference points estimation, their method is based on a small number of landmark points. Moreover, their database contains mainly radiographs without hallux valgus (almost 50%) or with small deformation, *i.e.* only 5/340 (1.5%) radiographs have IMA > 16° (severe hallux valgus deformation).

In this study, we significantly expanded algorithms to automate HV assessment from foot radiographs[30]. The necessary bones (first, second metatarsal, and hallucial PP) were segmented and labelled by a U-Net to set reference points and calculate HVA/IMA automatically. Expert clinicians also determined these angles manually, with outcomes being compared later. Moreover, our algorithm was evaluated only on patients' radiographs who subsequently underwent hallux valgus surgery. Our dataset contains a considerable percentage of radiographs with severe forefoot deformations including toe overlap, severe pronation, and sesamoid dislocation. Our bone segmentation-based algorithm is sufficiently robust to handle even such challenging circumstances anatomy as toes overlapping.

Classification systems

Traditional classification methods rely upon weightbearing anteroposterior radiographs to determine the severity of HV based on the HVA, and IMA (Figure 1)[17]. More than 100 different operative techniques were described for the correction of HV[32-34]. The overall clinical picture together with the degree of deformity determine the surgical decisions made. A suitable intervention is selected by considering the overall clinical picture along with the degree of deformity, potential degenerative changes of the first metatarsophalangeal joint, size, and shape of the metatarsal, and joint congruency.

Our algorithm is based on operative treatment algorithm proposed by EFORT (Figure 2)[35]. A convolutional neural network was trained to segment bones, with subsequent image analysis to automatically estimate angles and recommend appropriate surgical decisions. Digital radiographs were managed using a picture archiving and communication system and the IMPAX software suite.

MATERIALS AND METHODS

Algorithm outline

The measurements of the HVA/IMA were performed automatically on bones segmented and labelled by the U-Net neural network (Figure 3)[36]. To achieve this, the U-Net was first trained using anteroposterior foot radiographs and corresponding images with manually segmented and labelled bones. By providing automatically segmented and labelled bones, the required reference points were likewise automatically measured and the HVA/IMAs ultimately calculated.

The U-Net was trained only on right feet radiographs to reduce the cost and time of model training. Radiographs with the left feet were mirrored and then incorporated into the database. At the angle measurement stage, the segmented images with the left feet were back-mirrored to perform the measurements on the feet in the original orientation.

Dataset

133 patients were randomly selected between 2014 and 2021. A total of 265 pre-operative (unilateral or bilateral) anteroposterior feet radiographs were sourced from the electronic database of the authors' institution (demographics in Table 1). Inclusion criteria were: available weight-bearing radiographs, sole indication: symptomatic hallux valgus. Exclusion criteria were: No available weight-bearing radiographs, prior osteotomies, radiographs with severe osteoarthritis and first metatarsophalangeal joint deformation, and/or severe, *e.g.*, rheumatoid forefoot deformations or Charcot diabetic foot, visible plates, and other artificial elements distorting the image of the bone. Radiographs were obtained using standard radiology equipment Eidos RF439 and Luminos DRF unit and digitally transmitted *via* a picture archiving and communication system.

The data was divided randomly into three subsets: training, validation, and testing (Figure 3B). Both the patient's right and left feet were included in these subsets. The training and validation subsets were used to train and validate the U-Net for bone segmentation, while the testing subset was used to evaluate the performance of the trained U-Net and automatically measure the HVA/IMA.

Training and validation subset

We initially applied a 71/29 percent random split between training and validating subsets to develop the U-Net. After achieving the Sørensen–Dice index (SDI) greater than the cutoff value, the final training set was established.

Testing subset

According to Zou *et al*[37], the minimum number of subjects (testing subset) to estimate the agreement of the measurements between the two methods is 80. Our testing subset consisted of 84 randomly selected anteroposterior foot radiographs. Apart from the input radiographs, the testing subset also contained manually segmented radiographs to evaluate the quality of bone segmentation by the U-Net network. We calculated the SDIs using radiographs with automatically and manually segmented bones. There were no duplicate patient radiographs between the training, validation, and testing subsets. The validation radiographs were used to select the best neural network model, and validate the performance of the selected network during its training. The HVA/IMA were estimated only on the testing radiographs.

Table 1 Demographics

	Age	Sex	Hallux valgus angle			Intermetatarsal angle		
			All feet	Right feet	Left feet	All feet	Right feet	Left feet
Number of subjects	133	133, 16M (12%)/117F (88%)	265	133	132	265	133	132
Number of measurements	-	-	243	120	123	212	105	107
Range	23-81 yr	-	4-68°	4-68°	10-61°	4-26°	4-26°	7-22°
Average	55.8 yr	-	31.2°	31.3°	31.1°	13.8°	14°	13.7°

M: Male; F: Female; -: Not applicable.

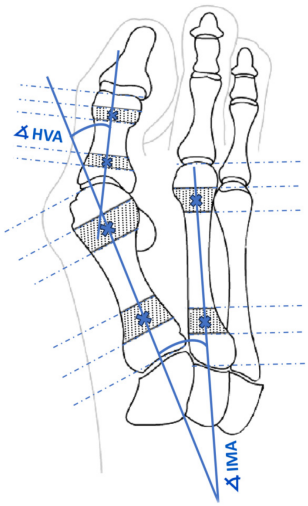

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Figure 1 Determination of angles of hallux valgus as described by AOFAS. HVA: Hallux valgus angle; IMA: Intermetatarsal angle.

Anonymization and manual labelling

The input radiographs were anonymized (Figure 3A) and stored in .png image format with lossless compression. Radiographs were digitally anonymized with unique IDs. To train a U-Net network that would achieve high bone segmentation accuracy, bones were manually annotated on original high-resolution radiographs. Initially, seventy radiographs were manually segmented and labelled by the first author in Adobe Photoshop. The radiographs with labelled bones were randomly split into a set of 50 training and 20 validation images. Manual segmentation of bones on radiographs is a very time-consuming task with considerable effort necessary to properly separate the border of bone from surrounding soft tissue. Considering the current understanding of pronation and variable shape of the first metatarsal head in hallux valgus deformation described by Wagner *et al*[14], the first metatarsal head and the sesamoid bones were delineated carefully and precisely by a foot surgeon to achieve precise measurements of the HVA/IMA[11]. The complex structure of bones in anteroposterior feet radiographs makes automated segmentation (delineation) particularly difficult[38]. Radiographs are contaminated by noise, artifacts, insufficient contrast, resolution, and/or intensity. These factors made preparing the dataset and developing the algorithm particularly challenging.

Various bone segmentation strategies were considered during algorithm development. We started with a binary segmentation of bones with bone extraction[30]. However, this approach produced clinically unreliable results in cases of cross-over toe with higher HVA. To overcome these difficulties, and simplify the algorithm to achieve robust automated separation of each required bone even in "difficult" radiographs, we established main regions on each foot radiograph *via* multi-class segmentation (Figure 3A). This approach allowed us to select and process just the three bones forming the HVA/IMA (1,2MT, and hallucial PP) and exclude all remaining structures and radiograph background. Considering that the region of interest on a given foot may have varying aspect ratios (height to width), some images were padded vertically with rows of black pixels to standardize the image size to 768 × 1024 pixels without changing the resolution (Figure 4).

U-Net training and validation

Radiographs pre-processing: The radiographs were prepared as described above to training a U-Net neural network[36]. We designed a U-Net neural network for bone segmentation that operates on grey images sized 768 × 1024 px (Figure 4). In contrast to the U-Net proposed by Ronneberger *et al*[36] our network is symmetric one, *i.e.* the input image size is equal to output map size, it performs multi-class segmentation, and relies on the Dice loss and score for training and evaluation,

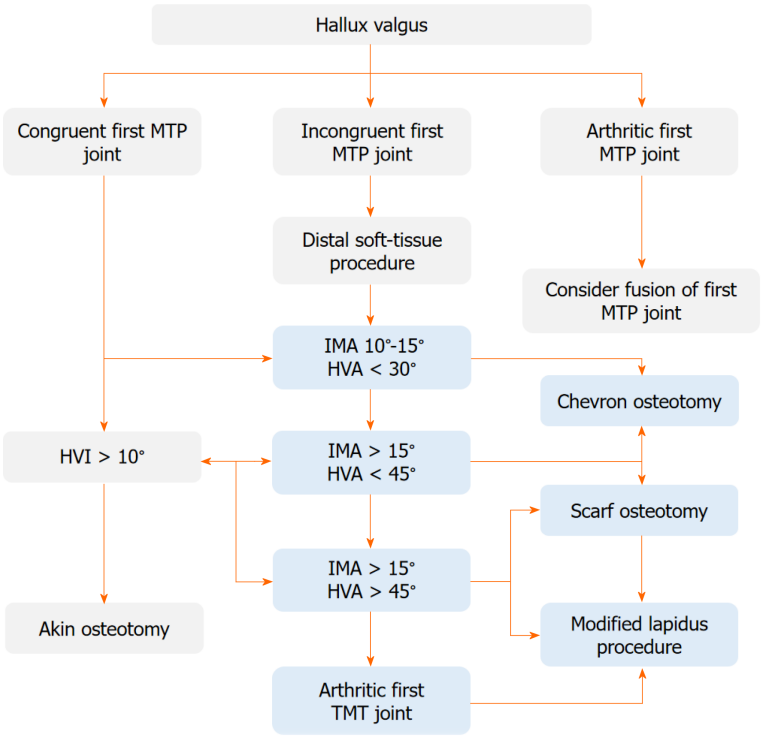

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Figure 2 Operative treatment algorithm of hallux valgus. D1 - decision for intermetatarsal angle (IMA) 10°-15° and hallux valgus angle (HVA) < 30°, D2 - decision for IMA > 15° and HVA < 45°, D3 - decision for IMA > 15° and HVA > 45°. HVA: Hallux valgus angle; HVI: Hallux valgus interphalangeus; IMA: Intermetatarsal angle; MTP: Metatarsophalangeal; TMT: Tarsometatarsal.

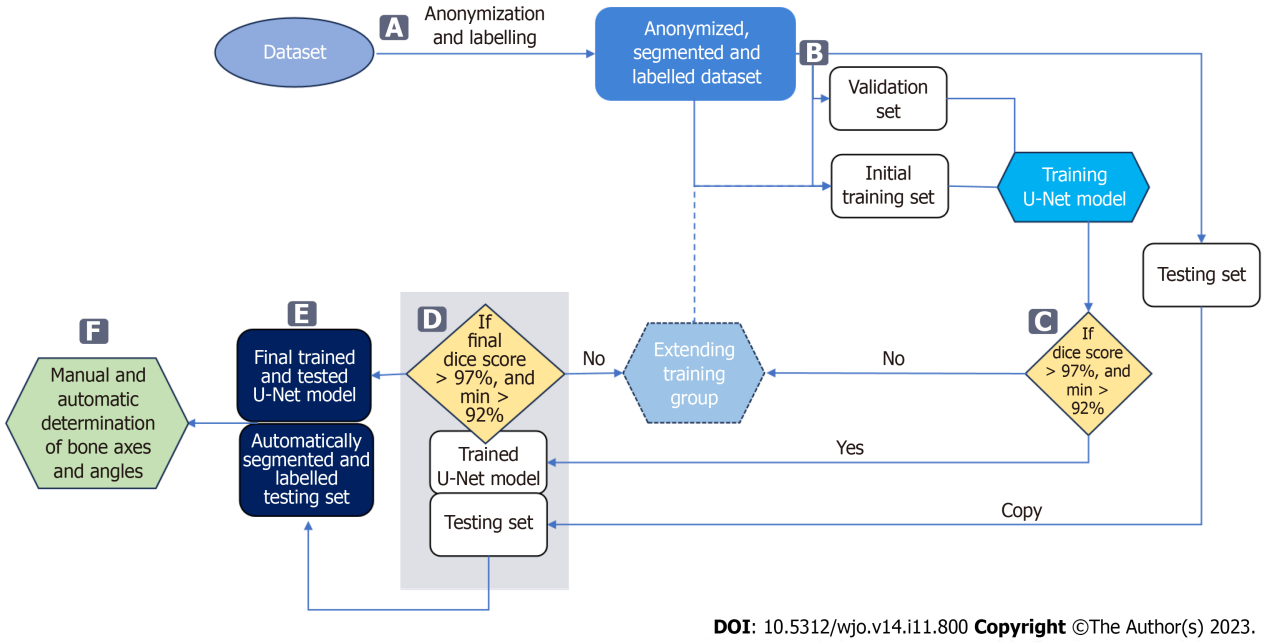

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Figure 3 Data flow in the proposed approach. A: Bones are manually segmented and labelled from anonymized input radiographs to perform multi-class segmentation using a U-Net neural network; B: Radiographs are then randomly assigned to three subsets: training, validation, and testing; C: The accuracy of bone segmentation in each training cycle of the U-Net is validated on a fixed validation subset consisting of 20 radiographs. The U-Net network is trained on a training subset initially consisting of 50 radiographs, which is increased by 10 each training cycle until achieving average Sørensen–Dice index (SDI) > 97% on the validation set; D: Once the network achieves an SDI > 0.97, calculated on the testing subset, the U-Net model completes. If SDI is not > 0.97, the training subset is extended and the U-Net is retrained; E: The final U-Net model is used to segment and label bones on all testing radiographs; F: These are used to automatically determine reference points and measure hallux valgus and intermetatarsal angles.

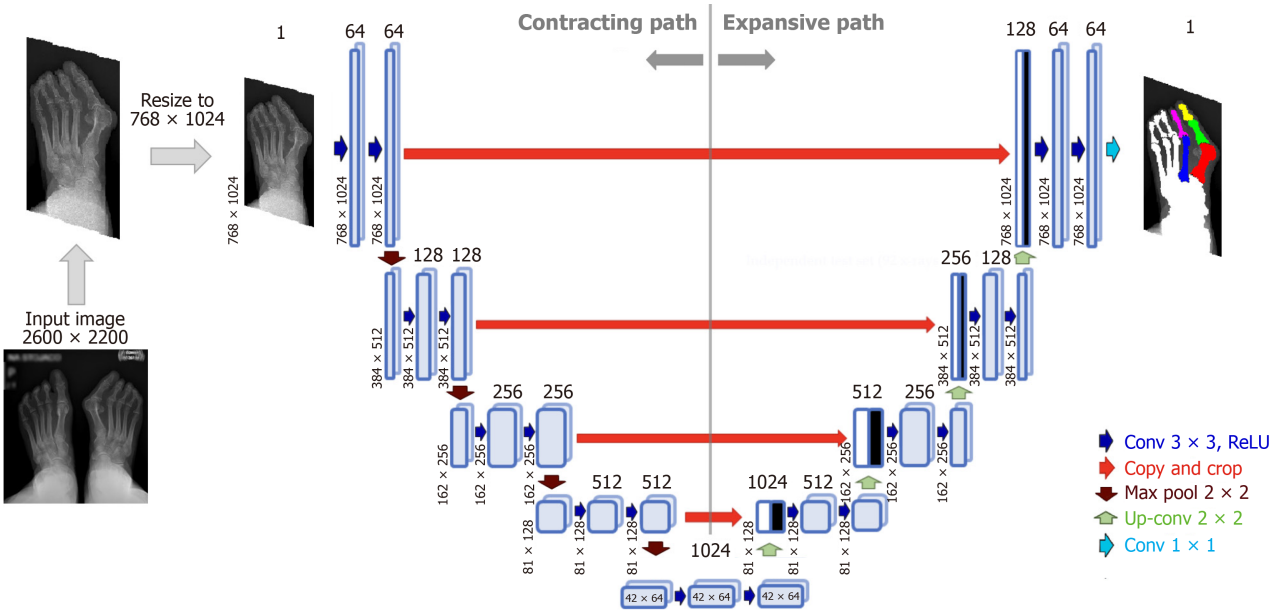

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Figure 4 Architecture of U-Net neural network for bone segmentation from radiographs. The network consists of an encoder (contracting path), which encodes an input image size of 768 × 1024 pixels to 42 × 64 × 1024 feature tensor at the bottleneck, and a decoder (expanding path) that decodes the feature tensor to the segmented image of the same size as the input image. The decoder follows the typical architecture of a convolutional network. Each block in the decoder consists of two 3 × 3 convolutions, each followed by a rectified linear unit (ReLU) and a 2 × 2 max pooling with stride 2 for down-sampling. Each down-sampling step of the encoder doubles the number of feature channels and decreases the image resolution by half. Every block in the decoder comprises upsampling the feature map followed by a 2 × 2 convolution that halves the number of feature channels - a concatenation with the correspondingly cropped feature map from the contracting path - and two 3 × 3 convolutions, each followed by a ReLU.

respectively. The accuracy of the bone segmentation was evaluated using SDI which is the most used metric in medical image segmentation[37,39]. We assumed that the threshold SDI should have a mean greater than 97%, with a minimum value greater than 92%. SDIs were determined only for the three bones required to estimate HVA/IMA. During U-Net training, the calculated SDI was used to check whether U-Net training should be stopped or continued on an extended training subset containing more images. After U-Net training was complete, the SDI was checked on the testing subset to verify whether the U-Net achieved the required generalizability. The initial training subset consisted of 50 anteroposterior foot radiographs with corresponding bone masks and labels. The initial training set was increased by 10 images after each training round until the threshold SDI was achieved on the validation subset (Figure 3C). The validation subset was fixed during training and used to compare the segmentation abilities of networks trained on incrementally larger training subsets. The threshold SDI was achieved on a training set of 150 radiographs with corresponding bone masks and labels. After adding an additional radiographs, the final training set consisting of 161 training images and 20 validation images (90% and 10%, respectively) was used to train the final U-Net. The dataset is available upon request.

Architecture and training U-Net: The neural network for bone segmentation follows the standard U-Net architecture established by Ronneberger *et al*[36]. Each U-Net encoder and decoder contains four layers (Figure 4). The validation SDI was calculated at the end of each epoch during the training of the U-Net, and the training was stopped when the SDI did not increase over 10 following epochs. This served as an early stop technique to avoid overfitting, where the value of early stop (patience) was set to 10. The U-Net was trained using Adam optimizer with Dice loss, learning rate (LR) set to 0.0001 (with reducing LR on plateau) and batch size equal to 8. The number of epochs was set to 80, and a callback was used to save the best U-Net model and its weights. The training data was augmented using mirroring, rotations, and contrast enhancement. Training of neural networks was performed on NVIDIA A100 GPU, whereas the testing was performed on the notebook's GPU (RTX2060).

Final validation of U-Net: Before measuring HVA/IMA, the trained U-Net was evaluated on the testing subset to assess its generalizability (Figure 3D). As the average SDI was larger than 97% on the testing subset with a minimal score larger than 92%, we used the trained U-Net to segment bones on all test radiographs (Figure 3E). In image post-processing, small holes in bones segmented by the U-Net were filled using morphological operations, and artifacts such as small blobs were deleted. Our algorithm first segmented and labeled bones that it then used to automate determining reference points and HVA/IMA measurements (Figure 3F, Figure 5). The programmer who trained the U-Net did not parti-cipate in manual measurements of HVA/IMA and did not see any results before statistical analysis.

Measurement of HVA and IMA

Automatic determination of reference points and angles: Using the anteroposterior feet radiographs, the U-Net segmented bones and labelled them with different colors (Figure 4). Using these labels our algorithm selected three bones

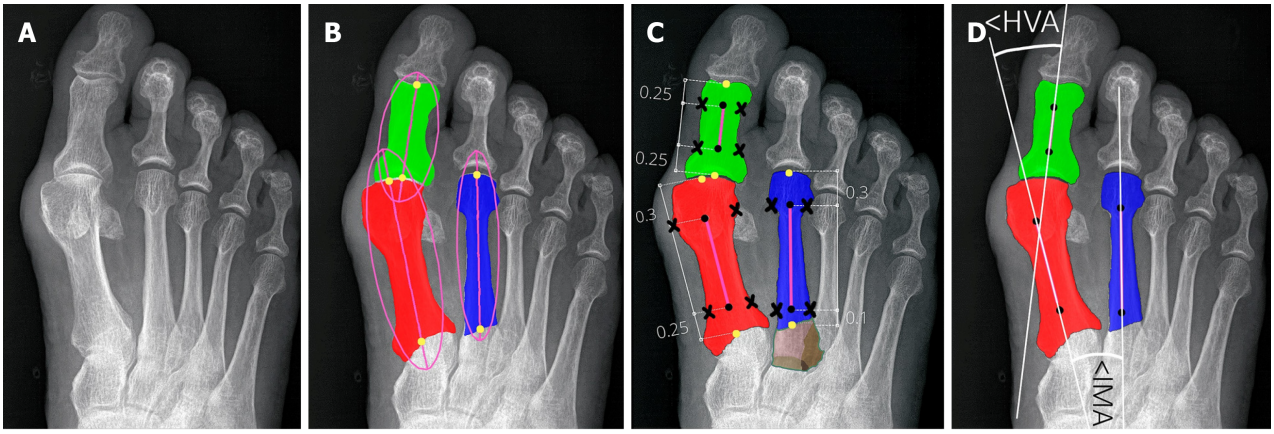

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Figure 5 Determining reference points of first and second metatarsals and hallucial proximal phalanx. A: Input radiograph; B: Three bones of interest: 1,2MT, and hallucial PP were approximated by ellipses to estimate bone axes, which were then used to determine bone endpoints; C: The elliptical axes were split into three parts with specific proportions to determine segment endpoints (marked by black dots) and reference points of the respective bone being established on a transverse line perpendicular to the longitudinal at a point equidistant from the outer border of the medial and lateral cortices (black crosses). Brown color show unsegmented (bone overlap) areas of the proximal epiphyseal of the second metatarsal bone; D: The central axes (white lines) were automatically determined through reference points (marked by black dots).

of interest: 1,2MT, and hallucial PP. HVA/IMA were automatically measured using reference points from these bones.

According to AOFAS (Figure 1) all reference points on the 1,2MT, and hallucial PP are the metaphyseal/diaphyseal points from which a guideline had to be determined to automate measurement of bone axes[18]. The final bone split ratios were selected following various combinations of bone split ratios to obtain the points closest to the diaphysis (Figure 5). For the 1MT, the reference points were located at 0.3 of the bone length proximal to the distal articular surface and at 0.25 of the bone length distal to the proximal articular surface. For the 2MT: 0.30 and 0.10, and for the hallucial proximal phalanx: 0.25 and 0.25, respectively.

Statistical analysis

Eighty-four radiographs of patients were used to measure HVA/IMA both manually by clinicians (reference method) and automatically by our algorithm. The reliability of the measurements between these two approaches was calculated using ICC and the standard error for a single measurement (SEM). Manual measurements (HVA/IMA) were performed by: an orthopedic surgeon (O_A) with 7 years' experience and repeated at 2 mo in blinded test (O_{A1} and O_{A2}), an orthopedic surgeon (O_B) with 15 years of experience; and by musculoskeletal radiologist (R) with 15 years of experience. Interobserver and intraobserver reliability coefficients (ICC) were calculated. The observers were not aware of any clinical results. Assessment of the HVA/IMA was performed according to the guidelines of the AOFAS *ad hoc* Committee on Angular Measurements (Figure 1) and digital technique using Radiant/Carestream[7,18,40]. Our algorithm then classified the appropriate severity (Figure 2) and operative decisions were compared against the orthopedic surgeon (O_{A2}). All statistical calculations were performed using MedCalc.

RESULTS

We proposed a novel automated HVA/IMA measurement method using deep learning algorithms. To measure these angles, the 1,2MT, and hallucial PP bones were automatically segmented, with reference points then automatically assigned. We obtained high interobserver and intraobserver correlations between manual measurements of HVA and IMA, and great agreement between artificial intelligence (AI) (our algorithm) and clinician angle measurements (Table 2). We analyzed HVA and IMA measurement errors for each patient's radiograph finding (Figures 6 and 7). Standard Error of the Mean for HVA was 0.26 and 0.16 for IMA. The accuracy of angles measured by the U-Net is similar to that of orthopedic surgeons.

A decision system was developed and tested according to the EFORT operative treatment algorithm (Figure 2)[35]. Operative decisions were taken (D1- chevron, D2- chevron or scarf, D3- scarf or Lapidus) based on calculated angles. The AI decisions were compared to O_{A2} decisions for concordance. The agreement of clinician decisions was also compared. The ratio of same pre-operative surgical decisions among AI and O_{A2} was almost 0.80 (67/84), which was higher than the ratio among clinicians (Table 2). A key achievement of our algorithm is that it saves radiologist and orthopedic surgeon's time while providing a clinically actionable HVA/IMA measurement that supports preoperative planning.

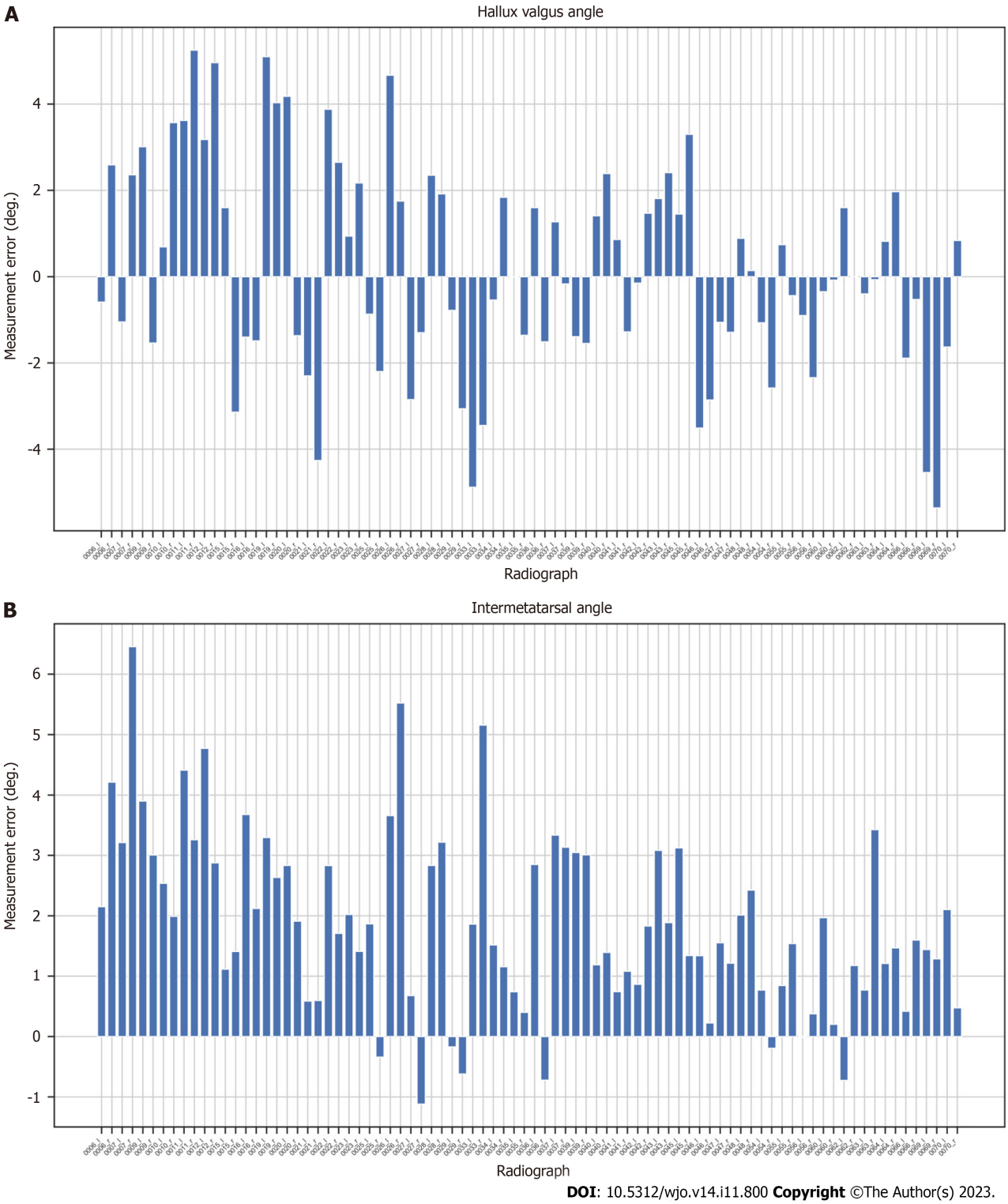

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Figure 6 Measurement errors (in degrees) for AI (our algorithm) compared to orthopedic surgeon (O_{A2}) for each radiograph. A: Hallux valgus angle; B: Intermetatarsal angle.

DISCUSSION

Some initial work on deep-learning radiographic and WBCT foot analysis was recently published[41-44]. While WBCT is arguably the future of hallux valgus preoperative qualifications, X-ray remains the standard as it is cheap, and widely available for symptomatic HV[38,45]. This work is in line with emerging research and substantially improves upon our previous algorithm. As demonstrated experimentally, the proposed approach can estimate HV angles on high-resolution radiographs and classify the severity of HV as a preoperative decision-making tool. Moreover, this work may expedite novel developments in forefoot surgery. This will provide a reliable opportunity to compare preoperative and posto-perative measurements and analyze the effects of surgical correction to produce better HV treatment standards.

Table 2 Correlation of hallux valgus angle, intermetatarsal angle, and pre-operative surgical decisions between clinicians, and against artificial intelligence

	Hallux valgus angle correlation (ICC)	Intermetatarsal angle correlation (ICC)	Pre-operative surgical decisions correlation
R-O_B	0.96	0.79	0.73 (61/84)
R-O_{A1}	0.96	0.81	0.62 (52/84)
R-O_{A2}	0.96	0.78	0.73 (61/84)
O_B-O_{A1}	0.96	0.91	0.75 (63/84)
O_B-O_{A2}	0.99	0.95	0.88 (74/84)
O_{A1}-O_{A2}	0.98	0.91	0.82 (69/84)
AI-O_{A2}	0.97 (AA-ICC)	0.89 (AA-ICC)	0.80 (67/84)
	0.97 (C-ICC)	0.75 (C-ICC)	

AI: Artificial intelligence; AA-ICC: Absolute agreement interclass correlation coefficient; C-ICC: Consistency interclass correlation coefficient; ICC: Interclass correlation coefficient; O_{A1}, O_{A2}, O_B: Orthopedic surgeons; R: Musculoskeletal radiologist.

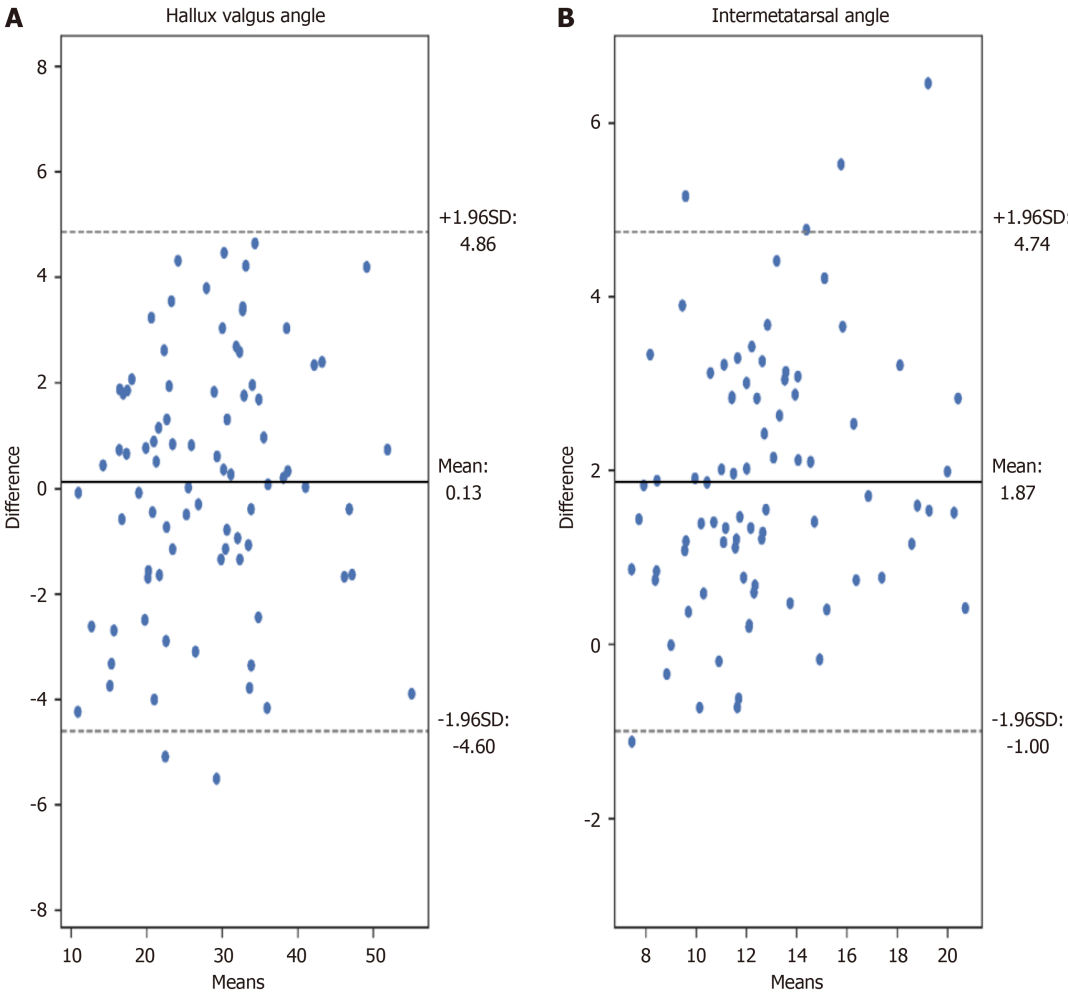

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Figure 7 Bland-Altman plots illustrating the differences between measurements achieved by our algorithm (AI) and orthopedic surgeon (O_{A2}). A: Hallux valgus angle measurements; B: Intermetatarsal angle measurements.

Coughlin *et al*[19] found that only 83.8% of IMA measurements made by physicians were within 3 degrees of concordance. AI overcomes the issue of clinician intra-observational and inter-observational reliability in terms of repeatable angular measurements of HV[46].

Considering that collecting a dataset of radiograms of patients with HV who were subsequently operated is not easy, we decided to rely on HV measurements on the segmented bones. Our initial research demonstrated that such an approach permits achieving better accuracy of HVA/IMA measurements on limited numbers of radiographs compared to key point-based ones. The difficulties associated with segmentation of proximal epiphyseal of 2MT bones due to anatomical overlap inclined us to apply a simplified segmentation with the exclusion of this bone area (Figure 5C). Consequently, the IMA measurements have an irrelevant bias (Figure 6B, Figure 7B).

Foot surgeons are aware that the decision to perform osteotomies or first tarsometatarsal joint (TMTJ) fusions (Lapidus procedure) depends on more than just HVA and IMA. Rather, it depends on the patients' clinical picture, concomitant deformities of the foot such as lesser toe deformities, pes planus, metatarsus adductus, first TMTJ instability, the width of the 1st MT shaft, pronation of the first ray, presence of first metatarsophalangeal joint osteoarthritis, and the surgeons' own skill level. The lateral view is also critical in evaluating the first TMTJ instability or presence of osteoarthritis which may necessitate a fusion rather than a 1st MT osteotomy. According to Lee *et al*[11] the HVA, IMA, interphalangeal angle, sesamoid rotation angle, and first metatarsal protrusion distance are worth measuring in HV considering three-dimensional role in this deformity. Presently the above-mentioned requirements may limit the applicability of our method in some cases. Nonetheless, our algorithm establishes itself as a fast and clinically effective tool in the assessment of many HV cases. In order to fully automate preoperative HV planning, further research and development remain necessary.

In future work, more radiographs will be labeled to train more advanced U-Net to distinguish bones under challenging areas better. A multi-center database of radiographs should be created. Due to recent developments and a deeper understanding of pronation, enhanced segmentation and further research on manual and automatic estimation of the distal metaphyseal/diaphyseal 1MT reference point is necessary. We plan to train and evaluate different networks on our dataset, which will be extended to new images from other hospitals.

CONCLUSION

The proposed automated, AI-assisted determination of angles of hallux valgus based on deep learning is an accurate tool that produces measurements comparable to manual measurements performed by experienced clinicians in significantly less time. Automation can be used in clinical practice to determine angles of hallux valgus on X-ray images, classify the degree of deformity, and streamline preoperative decisions-making prior to HV surgery.

ARTICLE HIGHLIGHTS

Research background

Recent advances in artificial intelligence and deep learning has spurred innovations in medical imaging modalities, resulting in enhanced visualisation possibilities. Additionally, there is a growing interest in the automation of regular diagnostic procedures alongside orthopedic measurements.

Research motivation

So far, no reliable and automated method has been developed for measuring angles of foot bones in significant deformities of the big toe from radiographs according to AOFAS. Likewise, there is no system for automated preoperative decision-making.

Research objectives

The aim of our research was to develop a robust automated method for measuring angles of hallux valgus on radiographs according to AOFAS guidelines, to determine the accuracy of this method, to compare it against expert clinician measurements, and to develop a preoperative decision-making systems.

Research methods

The bones which are necessary to determine the angles of hallux valgus, obtained on anteroposterior weight-bearing feet radiograms were segmented by a U-Net. The bone axes were determined, and then the reference points for determining the hallux valgus angles (HVA) and intermetatarsal angles (IMA) were found. The interclass correlation coefficient and standard error for single measurements were used to calculate the agreement between manual and automatic measurements. Finally, the correlation between the decisions of our algorithm and clinical adjudication for preoperative planning of hallux valgus was investigated.

Research results

The key foot bones were segmented from anteroposterior feet radiograms by the U-Net neural network with high accuracy (average Sørensen–Dice index larger than 97%). Such a precise segmentation enabled the accurate determination

of bone axes and the required reference points. Excellent agreement was achieved between manual and automated measurements of both angles. For HVA, absolute agreement interclass correlation coefficient (AA-ICC) and consistency ICC (C-ICC) were 0.97, and standard error of measurement (SEM) was 0.32. For IMA, AA-ICC was 0.75, C-ICC was 0.89, and SEM was 0.21. The proposed hallux valgus treatments based on HVA and IMA measured automatically correlated well with those proposed by orthopedic surgeons performing manual angle measurements.

Research conclusions

The proposed artificial intelligence powered automation for evaluating angles of hallux valgus through deep learning is a precise, yielding measurements akin to those conducted manually by experienced clinicians. This offers promising clinical applications such as facilitating the automated determination of angles of hallux valgus from X-ray images, categorizing the extent of deformity, and recommending a specific protocol for corrective surgery.

Research perspectives

Future research will focus on automating the measurements of remaining angles and parameters of forefoot deformation along its greater clinical implementation to further enhance diagnostic accuracy and improve patient outcomes.

FOOTNOTES

Author contributions: Kwolek Ko, Liszka H, Kwolek Ka designed research; Kwolek Ko, Kwolek Ka performed research; Kwolek Ko, Kwolek Ka elaborated analytic tools, Kwolek Ko, Liszka H, Gądek A, Kwolek Ka analyzed data; Kwolek Ko, Liszka H, Kolecki R, Kwolek Ka wrote the paper.

Institutional review board statement: This study protocol got an official statement that human and animal studies received waiver of the approval requirement from the ethics committee.

Informed consent statement: This study did not involve human experiments and does not require the signing of an informed consent form.

Conflict-of-interest statement: The authors have no conflict of interest concerning the materials or methods used in this study or the findings specified in this article.

Data sharing statement: No additional data are available.

ARRIVE guidelines statement: The authors have read the ARRIVE guidelines, and the manuscript was prepared and revised according to the ARRIVE guidelines.

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META-ANALYSIS

Long head of biceps tendon transposition for massive and irreparable rotator cuff tears: A systematic review and meta-analysis

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Abstract

BACKGROUND

Superior capsular reconstruction (SCR) with long head of biceps tendon (LHBT) transposition was developed to massive and irreparable rotator cuff tears (MIRCTs); however, the outcomes of this technique remain unclear.

AIM

To perform a systematic review of biomechanical outcomes and a meta-analysis of clinical outcomes after LHBT transposition for MIRCTs.

METHODS

We performed a systematic electronic database search on PubMed, EMBASE, and Cochrane Library. Studies of SCR with LHBT transposition were included according to the inclusion and exclusion criteria. Biomechanical studies were assessed for main results and conclusions. Included clinical studies were evaluated for quality of methodology. Data including study characteristics, cohort demographics, and outcomes were extracted. A meta-analysis was conducted of the clinical outcomes.

RESULTS

According to our inclusion and exclusion criteria, a total of six biomechanical studies were identified and reported an overall improvement in subacromial contact pressures and prevention of superior humeral migration without limiting range of motion (ROM) after LHBT transposition for MIRCTs. A total of five clinical studies were included in the meta-analysis of LHBT transposition outcomes, consisting of 253 patients. The results indicated that compared to other surgical methods for MIRCTs, LHBT transposition had advantages of more significant improvement in ROM (forward flexion mean difference [MD] = 6.54, 95% confidence interval [CI]: 3.07-10.01; external rotation [MD = 5.15, 95%CI: 1.59-8.17]; the acromiohumeral distance [AHD] [MD = 0.90, 95%CI: 0.21-1.59]) and reducing retear rate (odds ratio = 0.27, 95%CI: 0.15-0.48). No significant difference

in American Shoulder and Elbow Surgeons score, visual analogue scale score, and University of California at Los Angles score was demonstrated between these two groups for MIRCTs.

CONCLUSION
In general, SCR with LHBT transposition was a reliable and economical technique for treating MIRCTs, both in terms of biomechanical and clinical outcomes, with comparable clinical outcomes, improved ROM, AHD, and reduced the retear rates compared to conventional SCR and other established techniques. More high-quality randomized controlled studies on the long-term outcomes of SCR with LHBT transposition are required to further assess.

Key Words: Massive and irreparable rotator cuff tears; Long head of biceps tendon transposition; Rotator cuff repair; Superior capsular reconstruction

Core Tip: Superior capsular reconstruction (SCR) with long head of biceps tendon (LHBT) transposition was developed to massive and irreparable rotator cuff tears (MIRCTs). However, the outcomes of this technique remain unclear. SCR with LHBT transposition is a reliable and economical technique for treating MIRCTs, both in terms of biomechanical and clinical outcomes, with comparable clinical outcomes, improved range of motion, acromiohumeral distance, and reduced the retear rates compared to conventional SCR and other established techniques.

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INTRODUCTION

Rotator cuff tears are one of the most common musculoskeletal disorders, affecting between 15% and 50% of the population, and increasing in prevalence with age[1]. The prevalence of massive and irreparable rotator cuff tears (MIRCTs) is reportedly as high as 40% in rotator cuff tears[2,3]. The rotator cuff works as a dynamic stabilizer of the glenohumeral joint, and MIRCTs will cause the absence of this stabilizer, with the head migrating superiorly and anteriorly, leading to abnormal wear and tear of the head and glenohumeral joint[4,5]. If left untreated, the MIRCTs may lead to permanent pain and loss of function, eventually resulting in rotator cuff arthropathy.

The repair of MIRCTs still remains a surgical challenge due to muscle fat infiltration, tendon retraction, and tissue degeneration. Numerous surgical management options for MIRCTs are available, including debridement and sub-acromial decompression, partial rotator cuff repair, biceps tenodesis or tenotomy, an allograft or autograft (patch, fascia or dermis), tendon transfer (latissimus dorsi, pectoralis major, or pectoralis minor), balloon technique, and reverse total shoulder arthroplasty, have been reported[6-13]. If complete repair cannot be accomplished, partial repair may still improve pain and function of the shoulder. However, the risk of retear rate after partial repair is as high as 52%[14]. Compared to partial repair, arthroscopic patch grafting has better clinical efficacy, but patch grafting has no significant benefit for patients with high-grade fatty degeneration[15,16]. Tendon transfer is a good option for young, active patients with MIRCTs, minimal glenohumeral arthritis and severe functional limitations. However, the surgical trauma of tendon transfer is relatively large and the rehabilitation process is complicated. In patients with advanced cuff tear arthropathy and/or painful pseudoparalysis, a reverse total shoulder can provide predictable pain relief and function improvement but is associated with more complications and higher failure rate[17].

Superior capsular reconstruction (SCR) is considered a possible option for treating MIRCTs and restoring superior glenohumeral stability and shoulder function, which was first described by Mihata *et al*[18] in 2013. The graft used is a tensor fasciae lata (TFL) autograft that attaches medially to the superior glenoid and laterally to the greater tuberosity. Recently, the long head of biceps tendon (LHBT) has been proposed as an alternative to the standard SCR graft, which seems to overcome the problems of donor site morbidity, feasibility of grafting, and additional costs of allografting[19]. When performing the LHBT transposition technique, the native LHBT connection on the glenoid side was preserved. The proximal portion of the LHBT, was then transposed posteriorly and fixed on the supraspinatus tendon footprint as the SCR.

Currently, biomechanical studies have been carried out to observe the biomechanical effects of LHBT transposition for MIRCTs[20-22]. Meanwhile, few previous clinical studies have reported promising clinical outcomes after LHBT transposition for MIRCTs[23-25]; however, it remains unknown about the reliability of this technique. The purpose of this study was to perform a systematic review of biomechanical outcomes and a meta-analysis of clinical outcomes after LHBT transposition for MIRCTs. We hypothesized that LHBT transposition would effectively restore joint biomechanics compared with the unrepaired state and improve overall shoulder function, the acromiohumeral distance (AHD) and

decrease the retear rate of repaired rotator cuff.

MATERIALS AND METHODS

This meta-analysis and systematic review was written according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines[26].

Search strategy

Two independent authors (Wan RW and Luo ZW) performed an electronic search in three databases between their inception date and May 20, 2022: MEDLINE (PubMed), EMBASE (Ovid), and Cochrane Library. The Boolean search phrase was "(superior capsul* reconstruction OR superior capsul* repair OR superior labr* reconstruction OR superior labr* repair OR rotator cuff repair) AND (biceps)." The reference lists of correlational studies were also reviewed.

Eligibility criteria

Inclusion criteria consisting of biomechanical studies were as follows: (1) Cadaveric shoulders with massive or irreparable rotator cuff tears; (2) LHBT transposition was used in cadaveric shoulders; and (3) articles written in English or with an English translation.

Inclusion criteria consisting of clinical studies were as follows: (1) LHBT transposition was used to treat rotator cuff tears; (2) Rotator cuff tears were large to massive or irreparable; (3) Postoperative functional outcomes were reported; and (4) Articles were written in English or had an English translation.

The exclusion criteria were as follows: (1) Computational-based or animal studies; (2) editorial letters or letters to the editor, case reports, technical notes, expert consensuses, systematic and narrative reviews, pilot studies, unpublished manuscripts, book chapters, lectures, meeting abstracts, conference proceedings, or dissertations; (4) superior labrum anterior and posterior (SLAP) injury or LHBT injury; (5) SCR without using LHBT transposition; and (6) clinical studies not reporting preoperative and postoperative outcomes or without enough information for data analyses.

Study selection

After duplicates were removed, two independent authors (Wan RW and Luo ZW) evaluated all titles and abstracts for relative articles. If these data were inadequate, full texts were assessed to judge if studies met the inclusion criteria. If there was an objection concerning the inclusion of studies, studies were judged by the senior author (Shang XL) to make the final decision.

Data extraction

After assessing full-text articles for eligibility and applying the inclusion/exclusion criteria, the following information were extracted: study type, level of evidence, first author, publication year, country, number of patients, mean age, mean duration of follow-up, surgical technique, and postoperative clinical outcomes, and postoperative retear rates. The primary outcomes of interest were American Shoulder and Elbow Surgeons (ASES) score, visual analogue scale (VAS) score (0-10 [10 = severe pain]), University of California at Los Angles (UCLA) score, range of motion (ROM) including active external rotation at side (ER_0; degrees), active forward flexion (FF; degrees), AHD, and retear rates. For studies with insufficient information, the reviewers contacted the primary authors, when possible, to acquire and verify the data.

Quality assessment

Given the abundance of nonrandomized studies in the available literature, two independent reviewers (Wan RW and Luo ZW) critically appraised all eligible studies using Methodological Index for Nonrandomized Studies (MINORS) to evaluate their quality[27]. If a consensus was not achieved, a senior reviewer (Shang XL) made the final decision on the assessment. The MINORS instrument consists of 12 items: 4 for comparative studies only and 8 for noncomparative and comparative studies. A score of 0 (not reported), 1 (reported but inadequate), or 2 (reported and adequate) was suggested for each item, resulting in an ideal maximum score of 16 for noncomparative studies and 24 for comparative studies. For nonrandomized comparative studies, the methodologic quality was classified as follows: 0-12, low quality; 13-18, fair quality; and 19-24, good quality. The outcomes of the risk of bias and quality assessment offered context for the conclusions to be drawn from this review.

Statistical analyses

Statistical analyses were performed by using Review Manager, version 5.4 (Nordic Cochrane Centre, Cochrane Collaboration). The mean difference (MD) and odds ratio (OR) were used to compare continuous and dichotomous variables, respectively. All results were reported with 95% confidence intervals (CIs). $P < 0.05$ was considered statistically significant. I^2 test was performed to assess the impact of study heterogeneity on the results of the meta-analysis. According to the Cochrane review guidelines, if severe heterogeneity was present at $I^2 > 50\%$, the random effect models were chosen. If $I^2 \leq 50\%$, multiple similar studies were considered to be homogeneous, and the fixed effects model was used to combine the statistical values. The results are summarized in forest plots.

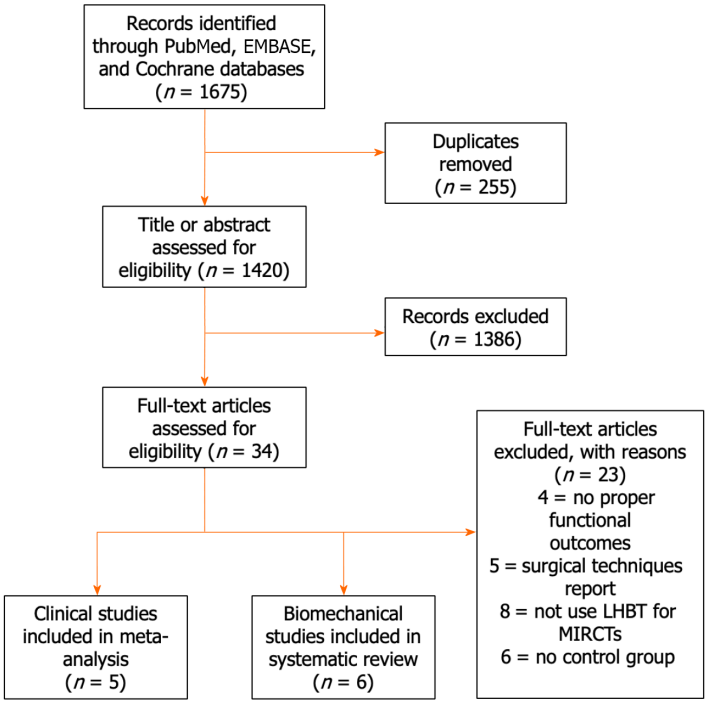

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Figure 1 Flowchart diagram of the electronic search. Based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. LHBT: Long head of biceps tendon; MIRCTs: Massive and irreparable rotator cuff tears.

RESULTS

Study selection

The initial literature search yielded 1675 articles (Figure 1). After removing duplicates, 1420 studies were screened for title and abstract, resulting in 34 full-text articles being assessed for eligibility. After the application of inclusion and exclusion criteria, six articles[21,28-32] reported biomechanical outcomes and five articles[33-37] reported clinical outcomes were identified for further analysis.

Risk of bias

The MINORS scores of two retrospective control clinical studies[33,34] were ≥ 19 points, indicating good quality of evidence. The MINORS scores of three other retrospective control clinical studies[35-37] were > 13 points, indicating fair quality of evidence (Table 1).

Systematic review of biomechanical studies

The characteristics and main results of included biomechanical studies are shown in Table 2. Because of the heterogeneity of testing conditions and outcome reporting, no meaningful statistical analyses could be performed. Overall, LHBT transposition for MIRCTs was reported to improve subacromial contact pressures and prevent superior humeral migration without limiting ROM.

Meta-analysis of clinical outcomes

A total of five studies meeting inclusion criteria were included in the meta-analysis of LHBT transposition outcomes, consisting of 253 patients. The study characteristics are presented in Table 3. We considered LHBT transposition for MIRCTS as the intervention group and other surgical methods for MIRCTs, *i.e.* the double-row repair, the transosseous-equivalent technique with absorbable patch reinforcement, the traditional SCR with a fascia lata autograft, arthroscopic rotator cuff repair (ARCR) alone, ARCR and tenotomy of LHBT, as the control group.

Patients were clinically assessed both preoperatively and postoperatively on a number of outcome-based scores that included ASES score, VAS score, UCLA score, ROM, AHD, and retear rates. ASES score was measured in five studies including a total of 253 patients: 127 in the LHBT transposition group and 126 in the control group. The results of the heterogeneity analysis indicated that these five studies had good homogeneity (P = 0.27, I^2 = 22%). A mean difference of 0.51, 95%CI: -1.91 to 2.93 was calculated, with a P value of 0.68. No significant difference was observed between the two groups regarding the ASES score (Figure 2A).

VAS score was measured in four studies including a total of 231 patients: 115 in the LHBT transposition group and 116 in the control group. The results of the heterogeneity analysis indicated that these four studies had good homogeneity (P = 0.33, I^2 = 13%). A mean difference of -0.13 (95%CI: -0.33 to 0.06) was calculated, with a P value of 0.18, indicating that no significant difference was found between the two cohorts (Figure 2B).

Table 1 Methodological Index for Nonrandomized Studies scores of the clinical studies

Ref.	Study type/LOE	A clearly stated aim	Inclusion of consecutive patients	Prospective collection of data	Endpoints appropriate to the aim of the study	Unbiased assessment of the study endpoint	Follow-up period appropriate to the aim of the study	Loss to follow up less than 5%	Prospective calculation of the study size	An adequate control group	Contemporary groups	Baseline equivalence of groups	Adequate statistical analyses	Score
Barth *et al* [37], 2020	III, retrospective control study	2	1	2	2	1	2	1	0	2	1	2	0	16
Kocaoglu *et al*[34], 2020	III, retrospective control study	2	2	2	2	2	2	2	0	2	2	2	0	20
Rhee *et al* [33], 2021	III, retrospective control study	2	1	1	2	2	2	1	1	2	2	2	1	19
Chiang *et al* [36], 2021	III, retrospective control study	2	1	2	2	1	2	1	0	1	1	2	0	15
Kawashima *et al*[35], 2022	III, retrospective control study	2	1	2	2	1	2	1	0	1	2	2	0	16

UCLA score was measured in three studies including a total of 163 patients: 89 in the LHBT transposition group and 74 in the control group. The results of the heterogeneity analysis indicated that these three studies had good homogeneity (P = 0.19, I^2 = 41%). A mean difference of 0.36, 95%CI: -0.67 to 1.39 was calculated, with a P value of 0.50. This suggested that no significant difference was found between the two cohorts (Figure 2C).

ROM was evaluated in five studies including a total of 253 patients: 127 in the LHBT transposition group and 126 in the control group. The results of the heterogeneity analysis in FF and ER0 indicated that these five studies had good homogeneity (P = 0.61, I^2 = 0%, P = 0.32, I^2 = 14%). In terms of FF, a mean difference of 6.54, 95%CI: 3.07-10.01 was calculated, with a P value of 0.0002, indicating that the FF was significantly better in the LHBT transposition group (Figure 2D). Regarding ER0, a mean difference of 5.15, 95%CI: 1.59-8.17 was calculated, with a P value of 0.005, indicating that the ER0 was significantly better in the LHBT transposition group (Figure 2E).

AHD was measured in four studies including 199 patients: 103 in the LHBT transposition group and 96 in the control group. The results of the heterogeneity analysis indicated that these four studies had good homogeneity (P = 0.13, I^2 = 48%). A mean difference of 0.90, 95%CI: 0.21-1.59 was calculated, with a P value of 0.01, indicating that LHBT transposition can significantly improve AHD (Figure 2F).

Retear rate was reported in five studies including a total of 253 patients: 127 in the LHBT transposition group and 126 in the control group. The results of the heterogeneity analysis indicated that these five studies had good homogeneity (P = 0.42, I^2 = 0%). An odds ratio of 0.27, 95%CI: 0.15-0.48 was measured (P < 0.0001), thereby indicating that the retear rate in LHBT transposition group was lower (Figure 3).

Table 2 Biomechanical outcomes after long head of biceps transposition

Ref.	Number of shoulders	Age, yr	Surgical technique	Testing conditions/groups	Testing method	Main results	Main conclusion
Park *et al*[22], 2018	9	58 (33-77)	ACR using autologous proximal biceps tendon for large to massive rotator cuff tears	(1) Intact; (2) Stage II tear (complete tear of the supraspinatus); (3) ACR for stage II tear; (4) Stage III tear (complete tear of the supraspinatus and anterior one-half of the infraspinatus); and (5) ACR for stage III tear	Range of motion, superior translation of the humeral head, and subacromial contact pressure were measured at 0°, 30°, 60°, and 90° of ER with 0°, 20°, and 40° of glenohumeral abduction	ACR for both stage II and stage III showed significantly higher total range of motion compared with intact at all angles. ACR significantly decreased superior translation for stage II tears at 0°, 30°, and 60° ER for both 0° and 20° abduction and for stage III tears at 0° and 30° ER for both 0° and 20° abduction. ACR for stage III tear significantly reduced peak subacromial contact pressure at 30° and 60° ER with 0° and 40° abduction and at 30° ER with 20° abduction	ACR using autologous biceps tendon biomechanically normalized superior migration and subacromial contact pressure, without limiting range of motion
El-shaar *et al*[21], 2018	10 (5 matched pairs)	63 (59-67)	SCR utilizing a LHB autograft or TFL autograft	(1) After a massive RC tear without SCR; and (2) After SCR with either a TFL autograft or an LHB autograft	Cadaveric demographics, mean force required to superiorly translate the humerus, and change in mean force when normalized to the torn condition were recorded	SCR with an LHB autograft required 393.2% ± 87.9% of the force needed for superior humeral migration in the massive RC tear condition, while SCR with a TFL autograft required 194.0% ± 21.8%. The LHB reconstruction group trended toward a stronger reconstruction when normalized to the torn condition	SCR with an LHB autograft is a feasible procedure that is shown to be biomechanically equivalent and potentially even stronger than SCR with a TFL autograft in the prevention of superior humeral migration
Han *et al*[30], 2019	7	50-65	SCR using the LHBT or using the LHBT with side-to-side repair	(1) Intact; (2) Simulated complete supraspinatus tendon tear; (3) Modified SCR using LHB; and (4) Modified SCR using LHB and side-to-side repair augmentation	Superior translation of the humerus, subacromial contact pressure and area, and glenohumeral range of motion were tested at 0°, 30°, and 60° of glenohumeral abduction	The complete cuff tear shifted the humeral head superiorly as compared to the intact shoulder. Subacromial peak contact pressure was also increased at 30° and 60° while contact area was increased at 0° and 30°. The modified SCR both with and without side-to-side repair shifted the humeral head inferiorly at 30° and 60°, with contact area further reduced at 60°. Both techniques had	The LHB with appropriate distal insertion on the greater tuberosity restores shoulder stability in irreparable rotator cuff tears by re-centering the humeral head on the glenoid

						comparable results for contact pressure and total rotational range of motion	
Han *et al*[29], 2020	8	65 (56-69)	PR, BR and BRSS	(1) Intact; (2) IRCT; (3) PR; (4) BR; and (5) BRSS	Total rotational range of motion was measured at 40°, then 20°, and finally 0° of glenohumeral abduction. Superior humeral translation and subacromial contact pressure were measured at 0°, 30°, 6 0°, and 90° of external rotation at each abduction angle	Superior humeral translation was significantly decreased in the BR and BRSS conditions compared with the IRCT and PR conditions at 0° and 20° of GH abduction ($P < 0.001$). BR and BRSS significantly reduced subacromial contact pressure compared with IRCT and PR at 0° of GH abduction ($P < 0.001$). There was no significant decrease in total rotational range of motion after BR at any abduction angle	BR biomechanically restored shoulder stability without over constraining range of motion in an IRCT model
Berthold *et al*[32], 2021	8	53.4 ± 14.2 (20-64)	SCR with V- shaped LHBT reconstruction, box-shaped LHBT reconstruction or single-stranded LHBT reconstruction	(1) Intact; (2) Irreparable psRCT; (3) V-shaped LHBT reconstruction; (4) Box-shaped LHBT reconstruction; and (5) Single-stranded LHBT reconstruction	ghST, MAA, maximum cDF, and sCP were accessed and recorded in each condition	Each of the 3 LHBT techniques for reconstruction of the superior capsule significantly increased MAA while significantly decreasing ghST and cDF compared with the psRCT. Additionally, the V-shaped and box-shaped techniques significantly decreased sCP compared with the psRCT. The V-shaped technique further showed a significantly increased MAA and decreased cDF when compared with the box-shaped and single-stranded techniques, as well as a significantly decreased ghST when compared with the box-shaped technique	Using the LHBT for reconstruction of the superior capsule improved shoulder function by preventing superior humeral migration, decreasing deltoid forces and sCP
Denard *et al*[31], 2021	8	62 (46-70)	SCR with box-shaped LHBT reconstruction or single-limb LHBT reconstruction	(1) Intact state; (2) A stage III MCT model (complete supraspinatus and anterior one-half of the infraspinatus); (3) Box Biceps SCR; and (4) Single-limb biceps	A custom testing system used to evaluate range of motion, superior translation, and subacromial contact pressure at 0°, 20°, and 40° of abduction	Range of motion was not impaired with either repair construct ($P > 0.05$). The box SCR decreased superior translation by approximately 2 mm compared with the MCT at 0°, but translation remained greater compared with the intact	A box-shaped SCR using the native biceps tendon partially restores increased superior translation and peak subacromial contact pressure due to MCT. The technique may have a role in augmentation of an IMCT

				state in nearly every testing position. The in situ tenodesis had no effect on superior translation. Peak subacromial contact pressure was increased in the MCT at 0° and 20°. Abduction compared with the native state but not different between the native and box SCR at the same positions

ACR: Anterior cable reconstruction; IRCT: Irreparable rotator cuff tear; PR: Partial repair; BR: Biceps rerouting; BRSS: Biceps rerouting with side-to-side repair; psRCT: Irreparable posterosuperior rotator cuff tear; ghST: Glenohumeral superior translation; MAA: Maximum abduction angle; cDF: Cumulative deltoid force; sCP: Subacromial peak contact pressure; SCR: Superior capsular reconstruction; LHBT: Long head of biceps tendon; TFL: Tensor fasciae lata; MCT: Massive rotator cuff tear; IMCT: Irreparable and massive rotator cuff tear; ER: External rotation.

DISCUSSION

The main finding of this investigation was that LHBT transposition for MIRCTs was generally reported to reduce subacromial contact pressures and prevent superior humeral migration without limiting ROM. On clinical outcomes, compared to other surgical methods for MIRCTs, LHBT transposition resulted in significant improvement in ROM, AHD, and reducing retear rate.

In the included biomechanical study, Park *et al*[22] reported that SCR with LHBT transposition significantly reduced the humeral head displacement caused by massive rotator cuff tears and decreased subacromial contact pressure. Meanwhile, El-shaar *et al*[21] found that SCR with LHBT transposition achieved equivalent and potentially even greater biomechanical stability than SCR using a TFL autograft in preventing humeral head migration. Han *et al*[30] found that SCR for LHBT transposition with or without side-to-side repair both shifted the humeral head downward and was a further reduction in the contact surface area of the acromion. Besides the results of both techniques were comparable in terms of contact pressure and total rotational ROM. A biomechanical study by Han *et al*[29] found that compared with partial repair after MIRCTs, LHBT transposition with or without side-to-side repair both significantly reduced the humeral head migration distance at 0° and 20° of glenohumeral abduction and effectively reduced the subacromial contact pressure at 0° of glenohumeral abduction. Three different SCR with LHBT transposition techniques were compared in the study of Berthold *et al*[32]: V-shaped, box-shaped, and single-stranded. Each of the techniques significantly increased maximum abduction angle while significantly decreasing glenohumeral superior translation and maximum cumulative deltoid force compared with the irreparable posterosuperior rotator cuff tear. The V-shaped technique further showed a significantly increased maximum abduction angle and decreased maximum cumulative deltoid force when compared with the box-shaped and single-stranded techniques, as well as a significantly decreased glenohumeral superior translation compared with the box-shaped technique. Meanwhile, Denard *et al*[31] found that ROM was not impaired with box-shaped and single-limb LHBT transposition and that there was no difference in subacromial contact pressure compared to an intact rotator cuff.

In the meta-analysis of the clinical study, compared to control group, LHBT transposition group improved FF, ER0, and AHD with a lower retear rate. No significant difference in ASES score, VAS score, and UCLA score was demonstrated between these two groups for MIRCTs. Two articles pioneered the treatment of MIRCTs with SCR using

Table 3 Overview of included clinical studies

Ref.	Country	Journal	Level of evidence, study type	Groups	No. of shoulder in group	Male:female sex	Age, yr	Follow-up, mo	Outcomes
Barth *et al*[37], 2020	France	*Am J Sports Med*	3, retrospective study	DR *vs* TOE with absorbable patch reinforcement *vs* SCR with LHBT autograft	28 *vs* 30 *vs* 24	15:13 *vs* 19:11 *vs* 16:8	63 ± 9 (48-83) *vs* 59 ± 7.6 (45-71) *vs* 60 ± 7 (47-81)	25 ± 2 (24-29) *vs* 27 ± 5 (24-36) *vs* 25 ± 2 (24-29)	ASES score, VAS score, constant score, range of motion, simple shoulder test, subjective shoulder value, muscle strength, retear rate
Kocaoglu *et al*[34], 2020	Turkey	*Orthop J Sports Med*	3, retrospective study	SCR with LHBT autograft *vs* SCR with a tensor fasciae lata autograft	14 *vs* 12	N/A	64.6 ± 8.4 *vs* 62.5 ± 6.5	28 *vs* 32	ASES score, VAS score, QuickDASH, range of motion, AHD, retear rate
Rhee *et al*[33], 2021	Korea	*Arthroscopy*	3, retrospective study	ARCR + BR *vs* ARCR	59 *vs* 52	32:27 *vs* 29:23	63.7 ± 6.5 *vs* 62.8 ± 6.9	15.1 ± 3.4 *vs* 25.1 ± 8.7	ASES score, VAS score, constant score, UCLA score, range of motion, muscle strength, AHD, retear rate
Chiang *et al*[36], 2021	China (Taiwan)	*Arthroscopy*	3, retrospective study	ARCR and SCR with LHBT autograft *vs* ARCR and tenotomy of LHBT performed at the insertion site	18 *vs* 22	7:11 *vs* 6:16	62.3 ± 7.5 *vs* 62.2 ± 6.1	26.6 ± 3.9 (24-38) *vs* 31.9 ± 6.4 (26-45)	ASES score, VAS score, UCLA score, rang of motion, AHD, retear rate
Kawashima *et al*[35], 2022	Japan	*Arthroscopy*	3, retrospective study	partial repair *vs* SCR with LHBT transposition	10 *vs* 12	6:4 *vs* 7:5	71.9 ± 7.5 *vs* 67.8 ± 2.0	37.2 (24-72) *vs* 24.8 (24-30)	ASES score, UCLA score, rang of motion, AHD, retear rate

AHD: Acromiohumeral distance; ARCR: Arthroscopic rotator cuff repair; ASES: American Shoulder and Elbow Surgeons; DR: Double-row technique; LHBT: Long head of biceps tendon; N/A: Not available; SCR: Superior capsular reconstruction; TOE: Transosseous-equivalent technique; UCLA: University of California at Los Angles; VAS: Visual analogue scale.

autologous broad fascia as the repair material, and the postoperative follow-up found that the patients' function was significantly improved with satisfactory clinical outcomes[18,38]. However, the autologous broad fascia retrieval requires additional incisions, which not only increases the trauma but also makes the operation complicated and technically demanding. Besides, more anchors are used to fix the broad fascia, which leads to increased costs. Some authors have suggested replacing fascial autografts with human acellular dermal patch allografts to avoid any additional skin incisions and any donor site morbidity[39,40]. A recent study reported by Shin *et al*[41] showed satisfactory outcomes with SCR using acellular dermal allograft. However, a systematic review showed high retear rate on SCR using acellular dermal allograft compared with fascia lata[42]. The mechanical strength of acellular dermal materials remains controversial and expensive.

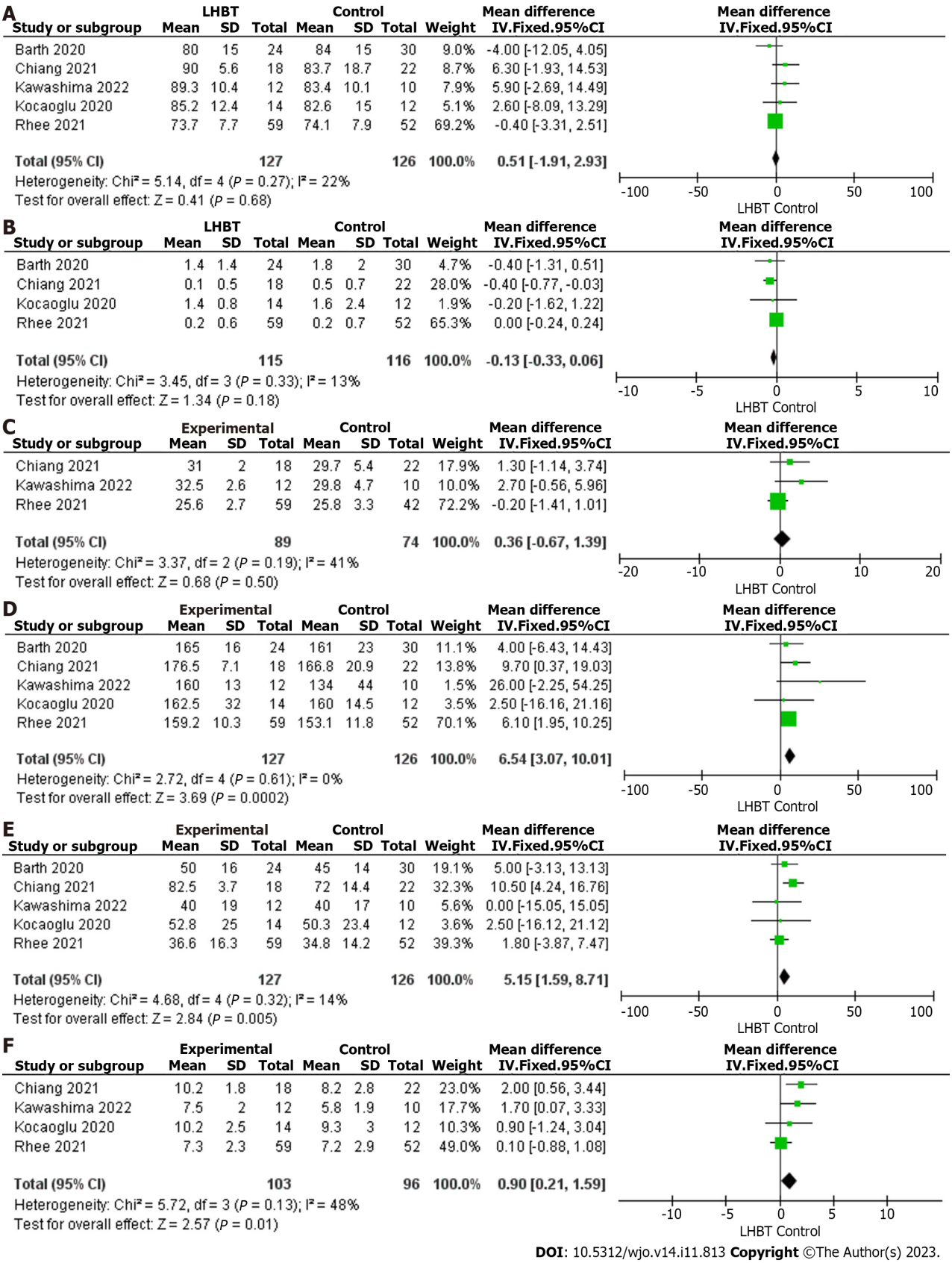

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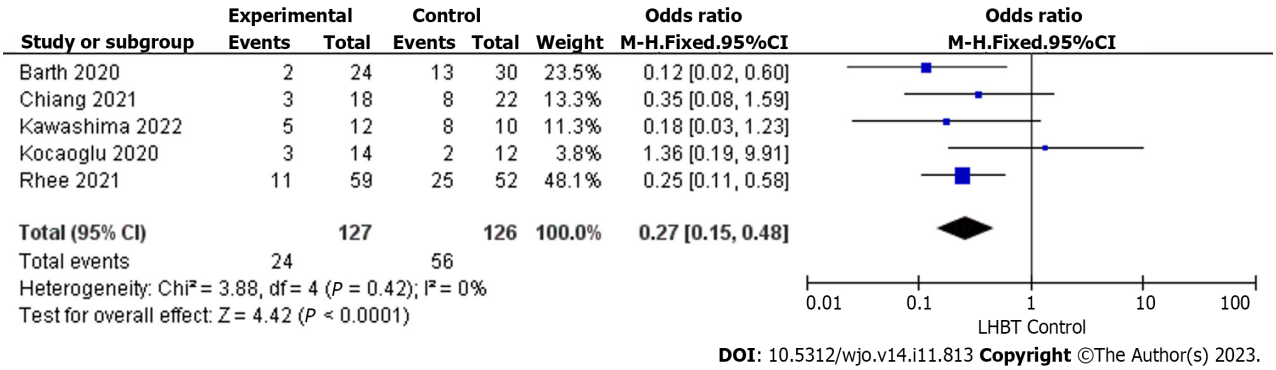
Figure 2 Standard differences in means. A: Standard differences in means for American Shoulder and Elbow Surgeons score between long head of biceps tendon (LHBT) transposition group and control group; B: Standard differences in means for visual analogue scale score between LHBT transposition group and control group; C: Standard differences in means for University of California at Los Angles score between LHBT transposition group and control group; D: Standard differences in means for forward flexion between LHBT transposition group and control group; E: Standard differences in means for external rotation between LHBT transposition group and control group; F: Standard differences in means for acromiohumeral distance between LHBT transposition group and control group. CI: Confidence interval; LHBT: Long head of biceps tendon.



DOI: 10.5312/wjo.v14.i11.813

Figure 3 Odds ratios for retear rate between long head of biceps tendon transposition group and control group. CI: Confidence interval; LHBT: Long head of biceps tendon.

SCR with LHBT transposition was proposed by Boutsiadis *et al*[19] first, and the LHBT without significant tears or severe degeneration was fully utilized in the operation, with the superior glenoid labral is preserved. LHBT was transferred laterally to the supraspinatus tendon footprint area for fixation. Finally, the distal LHBT is chosen to be cut or preserved according to its distal LHBT quality in order to simulate SCR, while the partially repaired rotator cuff can be bridged. The main advantage of this method is to borrow the LHBT to provide a tension-reducing scaffold to strengthen the anterior rotator cuff tissue mechanics and anterosuperior blocking effect to assist the massive rotator cuff repair and reduce the tension on the repaired rotator cuff tissue, thereby reducing the rate of postoperative rotator cuff retears and improving the AHD. This is consistent with the results of our meta-analysis.

Other advantages of SCR with LHBT transposition are that it not only avoids the trauma at the extraction site caused by taking the autologous broad fascia for SCR but also reduces the amount of anchor nails used, thus greatly reducing the cost and time of the procedure and decrease of infection. In addition, it is technically easier and more reproducible than SCR using fascial autografts or dermal allografts, which require a long learning curve. Finally, another possible advantage is the biological aspect of using a local autograft attached to the upper glenoid so that its vasculature may be preserved.

There are several potential limitations of this technique. The main condition is the availability of a relatively good quality LHBT, and SCR with other grafts should be considered when there is LHBT severe degeneration, LHBT rupture or partial tearing involving more than 50%, SLAP lesions > II, and some rare cases of anatomic variation or absence of the tendon[43]. However, in the case of chronic MIRCTs, they are often accompanied by LHBT damage. In addition, the LHBT is reportedly a pain generator in patients with rotator cuff tears, and the use of the LHBT as an autograft for SCR may, in theory, increase postoperative pain[37]. However, the results of clinical studies showed no difference in postoperative pain between the various compared techniques for either tenotomy or rerouting of the LHBT, suggesting that it can be safely used as an autograft[25,37].

This study had several limitations. First, the available studies or data about LHBT transposition used for MIRCTs were limited, only five studies with 127 patients in LHBT transposition group and 126 patients in control group. Second, there was insufficient high-quality comparative evidence, as the five included studies were all retrospective studies with a level of evidence 3, which may create recall or selection bias. Although the MINORS scores of these studies indicate good or fair quality evidence, they still fall short of rigorous randomized controlled trial studies. Third, the surgical approaches in the control group, although all of them are commonly used to treat MIRCTs, may have influenced the comparison of outcomes. Additional comparative trials, or even randomized controlled trials, are necessary in the future to determine which treatments are more advantageous in treating MIRCTs, and which modifications of the technique provide better outcomes. Moreover, prior investigations have revealed that distinct rehabilitation modalities and durations exhibit diverse prognostic implications for individuals undergoing arthroscopic repair of rotator cuff tears[44]. In this regard, forthcoming studies could potentially prioritize the evaluation of the influence of diverse rehabilitation approaches on the utilization of LHBT transposition as a therapeutic intervention for the management of MIRCTs. In addition, high-quality studies are necessary to evaluate the long-term outcomes of SCR with LHBT transposition, including postoperative pain, function and structural integrity. High-resolution ultrasound investigation may play an important role in this regard[45]. In a word, future high-quality research of SCR using LHBT transposition for MIRCTs is necessary.

CONCLUSION

This systematic review and meta-analysis demonstrated that SCR with LHBT transposition was a reliable and economical technique for treating MIRCTs, both in terms of biomechanical and clinical outcomes, with comparable clinical outcomes, improved ROM, AHD and reduced the retear rates compared to conventional SCR and other established techniques. To further evaluate the long-term effects of SCR with LHBT transposition, more high-quality randomized controlled studies are needed.

ARTICLE HIGHLIGHTS

Research background

Supracapsular reconstruction (SCR) combined with transposition of the biceps long head biceps tendon (LHBT) is an approach designed to meet the severe challenges posed by massive rotator cuff tears (MIRCT).

Research motivation

Although LHBT transposition has been adopted, its exact impact remains to be clearly elucidated.

Research objectives

There are gaps in our knowledge of the outcomes produced by this technique, and thus further research is needed to reveal its potential benefits and limitations.

Research methods

We conducted a methodical search of electronic databases to identify relevant literature based on inclusion and exclusion criteria. We first conducted a systematic review of the main findings and conclusions of the biomechanical studies. Subsequently, we conducted a comprehensive meta-analysis of the clinical outcomes of the included studies.

Research results

Biomechanical studies have reported that after performing LHBT transposition in MIRCT, there was a comprehensive improvement in subacromial contact pressure and a prevention of proximal humeral migration, without any resultant limitation in range of motion. The meta-analysis of LHBT transposition outcomes has encompassed five clinical studies demonstrated that, compared to other surgical methods for MIRCTs, LHBT transposition exhibited significant advantages in enhancing patients' ROM (forward flexion, mean difference [MD] = 6.54, 95% confidence interval [CI]: 3.00-8.08, external rotation [MD = 5.15, 95%CI: 1.59-8.17], acromiohumeral distance [AHD] [MD = 0.90, 95%CI: 0.21 to 1.59], and reducing the risk of retear [odds ratio = 0.27, 95%CI: 0.15-0.48]). There were no discernible differences between the two groups of patients in terms of American Shoulder and Elbow Surgeons scores, visual analogue scale scores, and University of California, Los Angeles scores.

Research conclusions

In summary, the utilization of LHBT transposition in SCR proved to be a dependable and cost-effective approach for addressing MIRCTs. This technique demonstrated favorable results not only in terms of biomechanical factors but also in clinical outcomes. It exhibited comparable efficacy to conventional SCR and other established techniques, while presenting notable improvements in ROM, AHD, and a reduced incidence of retear. Nevertheless, it is essential to emphasize the necessity for additional high-quality randomized controlled trials focusing on the long-term effects of SCR with LHBT transposition to further evaluate its efficacy.

Research perspectives

Future high-quality research of SCR using LHBT transposition for MIRCTs is necessary.

FOOTNOTES

Co-corresponding authors: Shi-Yi Chen and Xi-Liang Shang.

Author contributions: Wan RW and Luo ZW contributed equally to this study; Wan RW, Chen SY, and Shang XL participated in the study design, data analysis, and drafting of the critically revised the manuscript; Wan RW and Luo ZW were responsible for selecting articles for inclusion, and conducted the risk of bias assessment, and data extraction; Yang YM, Zhang HL, and Chen JN helped to revise the manuscript; Chen SY and Shang XL made invaluable contributions to conceptualizing the study and providing meticulous final review; All authors read and approved the manuscript.

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CASE REPORT

Surgical treatment of atlantoaxial dysplasia and scoliosis in spondyloepiphyseal dysplasia congenita: A case report

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Abstract

BACKGROUND

Spondyloepiphyseal dysplasia congenita (SEDC) is a rare autosomal dominant hereditary disease caused by COL2A1 mutations. SEDC primarily involves the skeletal system, with typical clinical manifestations, including short stature, hip dysplasia, and spinal deformity. Due to the low incidence of SEDC, there are only a few case reports regarding the surgical treatment of SEDC complicated with spinal deformities.

CASE SUMMARY

We report a case of a 16-year-old male patient with SEDC. He presented with typical short stature, atlantoaxial dysplasia, scoliosis, and hip dysplasia. Cervical magnetic resonance imaging showed spinal canal stenosis at the atlas level and cervical spinal cord compression with myelopathy. The scoliosis was a right thoracic curve with a Cobb angle of 65°. He underwent atlantoaxial reduction, decompression, and internal fixation from C1–C2 to relieve cervical myelopathy. Three months after cervical surgery, posterior correction surgery for scoliosis was performed from T3 to L4. Scoliosis was corrected from 66° to 8° and remained stable at 2-year follow-up.

CONCLUSION

This is the first case report of a patient with SEDC who successfully underwent surgery for atlantoaxial dysplasia and scoliosis. The study provides an important reference for the surgical treatment of SEDC complicated with spinal deformities.

Key Words: Spondyloepiphyseal dysplasia congenita; Surgical treatment; Atlantoaxial dysplasia; Scoliosis; Hip dysplasia; Case report

Core Tip: This study describes the case of a 16-year-old male patient diagnosed with spondyloepiphyseal dysplasia congenita (SEDC) and treated with surgeries for multiple spinal deformities. SEDC is a rare genetic disorder, which mainly affects skeletal development, with an incidence of approximately 3/1000000. Due to the low incidence, there are very few reports on surgical treatment of skeletal deformities in patients with SEDC. We believe that our study makes a significant contribution to the literature because this is the first case report of a patient with SEDC who successfully underwent surgeries for atlantoaxial dysplasia and scoliosis.

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INTRODUCTION

Spondyloepiphyseal dysplasia congenita (SEDC) is a rare autosomal dominant genetic disorder with an incidence of approximately 3/1000000[1]. SEDC affects bone development, particularly of the spine and the long bones of the limbs [2]. SEDC is caused by mutations in the COL2A1 gene, which regulates the synthesis of type II collagen, a major component of cartilage and other types of connective tissues[3]. Mutations in COL2A1 disrupt the normal production and assembly of type II collagen, which lead to abnormal bone growth and development[3,4]. The symptoms of SEDC vary widely, including short stature, atlantoaxial dysplasia, abnormal curvature of the spine (scoliosis or kyphosis), hip dysplasia, joint pain, early onset arthritis, and vision disorders[5]. Some individuals with SEDC may also have cleft palate or other craniofacial abnormalities[5]. There is currently no cure for SEDC, but surgical treatment for skeletal deformities can help manage symptoms and improve quality of life[6]. Although a few cases of atlantoaxial and hip dysplasia have been reported, surgical treatment of SEDC with scoliosis is very rare[7-10]. Herein, we report the case of a patient with SEDC who successfully underwent surgery for atlantoaxial dysplasia and scoliosis.

CASE PRESENTATION

Chief complaints

A 16-year-old male patient presented with a 5-year history of progressive scoliosis.

History of present illness

At the age of 11, the patient was diagnosed with scoliosis and was treated with a brace for more than 12 h daily and had regular check-ups. However, his scoliosis continued to deteriorate.

History of past illness

The patient experienced difficulty walking and had challenges with physical activities since the age of 1 year. His growth and development were delayed compared to those of his peers. Radiography of the hip joints revealed bilateral hip dysplasia.

Personal and family history

The patient denied any family history of similar diseases or malignant tumors.

Physical examination

Physical examination revealed that the patient was 128 cm in height, weighed 46 kg, and was unable to ambulate steadily. There was a right-sided thoracic spine curvature, resulting in a "razorback" deformity. The patient exhibited decreased proximal muscle strength in both lower limbs (grade IV), and bilateral Hoffman signs and Babinski signs were positive.

Laboratory examinations

Ventilation function, electrocardiogram, echocardiogram, and other preoperative examination results were normal. Blood calcium, phosphorus, vitamin D, and alkaline phosphatase levels were normal for bone metabolism.

Imaging examinations

During the initial outpatient examination, the spine X-ray indicated a leftward proximal thoracic curve of 32° and a rightward main thoracic curve of 65° (Figure 1A). Three months following atlantoaxial decompression and fixation surgery, and prior to scoliosis correction, the spine X-rays revealed the proximal thoracic curve was 36°, the main thoracic curve was 65°, the thoracic kyphosis was 7°, and the sagittal vertical axis (SVA) was 12.5 cm (Figure 1B and C). On the bending X-rays, the Cobb angle of the proximal thoracic curve was 33°and the main thoracic curve was 46° (Figure 1D

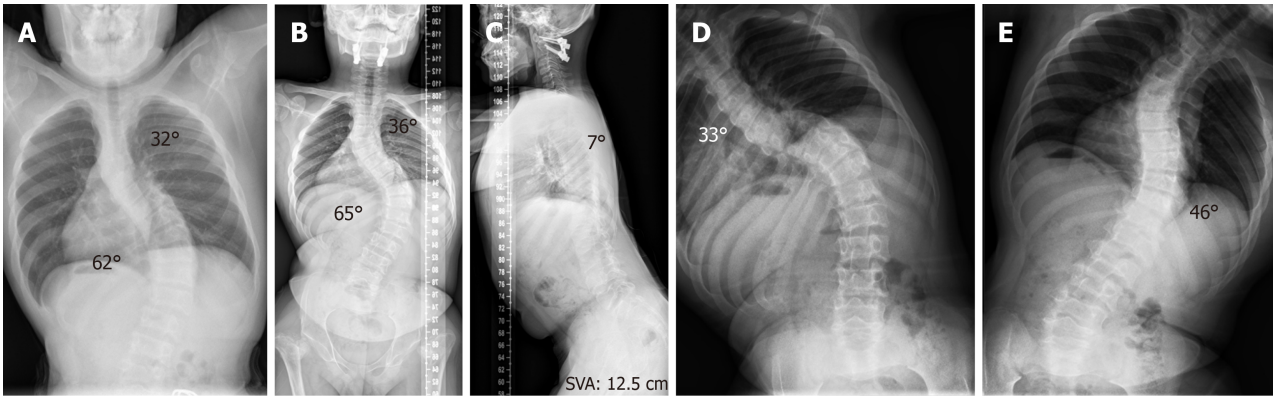

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Figure 1 Preoperative X-ray data of the spine. A: The spine X-ray taken during the first outpatient clinic indicates a leftward proximal thoracic curve (PTC) of 32° and a rightward main thoracic curve (MTC) of 65°; B: Before scoliosis correction, the standing spine X-ray reveals the PTC is 36° and the MTC is 65°; C: The thoracic kyphosis is 7° and the sagittal vertical axis is 12.5 cm; D and E: On the bending X-rays, the Cobb angle of the PTC is 33°and the MTC is 46°.

and E). Computed tomography (CT) three-dimensional (3D) reconstruction of the spine showed flattening of the vertebral body and widespread narrowing of the intervertebral space (Figure 2A-C). 3D CT of the cervical spine showed dysplasia of the odontoid process of the axis and the anterior and posterior arches of the atlas (Figure 3A and B). Spinal CT showed shorter pedicle screw lengths in the thoracic and lumbar vertebrae (2.94–5.17 cm), but the cancellous bone of the pedicles was well developed. A hip X-ray revealed dysplasia of bilateral hip joints and severe damage to the femoral heads. No obvious abnormality was found on the X-ray of bilateral knee joints and the full-length of both lower limbs (Figure 3C-E). Magnetic resonance imaging (MRI) of the spine indicated stenosis of the cervical spinal canal in the atlantoaxial region and myelopathy of the cervical spine (Figure 4C). The spinal MRI did not reveal other intraspinal abnormalities. The appearance of the patient before scoliosis surgery is shown in Figure 2D and E.

FINAL DIAGNOSIS

Combined with the patient's medical history and gene detection results (Figure 5), a diagnosis of SEDC was made.

TREATMENT

To avoid irreversible damage caused by cervical myelopathy, the patient was recommended to have atlantoaxial decompression and fixation surgery. Skull traction was implemented under anesthesia during the surgical procedure, and the atlantoaxial reduction was observed to be satisfactory. Consequently, one-stage posterior atlantoaxial reduction and fixation procedure was employed. During the procedure, lateral mass screws were placed on both sides of the atlas and pedicle screws were placed on both sides of the axis and fixed with connecting plates, and autogenous iliac cancellous bone were used for bone graft fusion. Three months after the operation, the patient underwent posterior spinal fusion, internal fixation, and bone graft fusion from T3 to L4 to treat his deteriorating scoliosis. During the surgery, Smith–Petersen osteotomies were performed within the fusion range to fully release the spine, and pedicle screws were inserted between T3 and L4. Autogenous combined with allogeneic bone particles were used for Moe bone grafting. Intraoperative blood loss was 450 mL, and the duration of surgery was 3.6 h. No abnormality was identified during intraoperative spinal cord monitoring.

OUTCOME AND FOLLOW-UP

The patient was successfully extubated and recovered smoothly after both surgeries. After the atlantoaxial decompression and fixation surgery, the compression of the cervical spinal cord was significantly relieved, and the instability of the atlantoaxial joint was improved (Figure 4). Before the corrective surgery of scoliosis, the patient's muscle strength of bilateral proximal lower limbs was significantly improved to grade V. The Hoffman signs and Babinski signs were negative. One week after corrective surgery for scoliosis, the spinal X-ray revealed significant correction of scoliosis with the main thoracic curve corrected from 65° to 10°, thoracic kyphosis corrected from 7° to 26°, SVA corrected from 12.5 cm to 3.2 cm, and considerable improvement in the back's unevenness (Figure 6A-D). At the postoperative 2-year follow-up, the patient reported no symptoms of discomfort, such as back pain and weakness of the lower extremities, the atlantoaxial joint reduction remained effective and spinal correction retained its stability (Figure 6E-H).

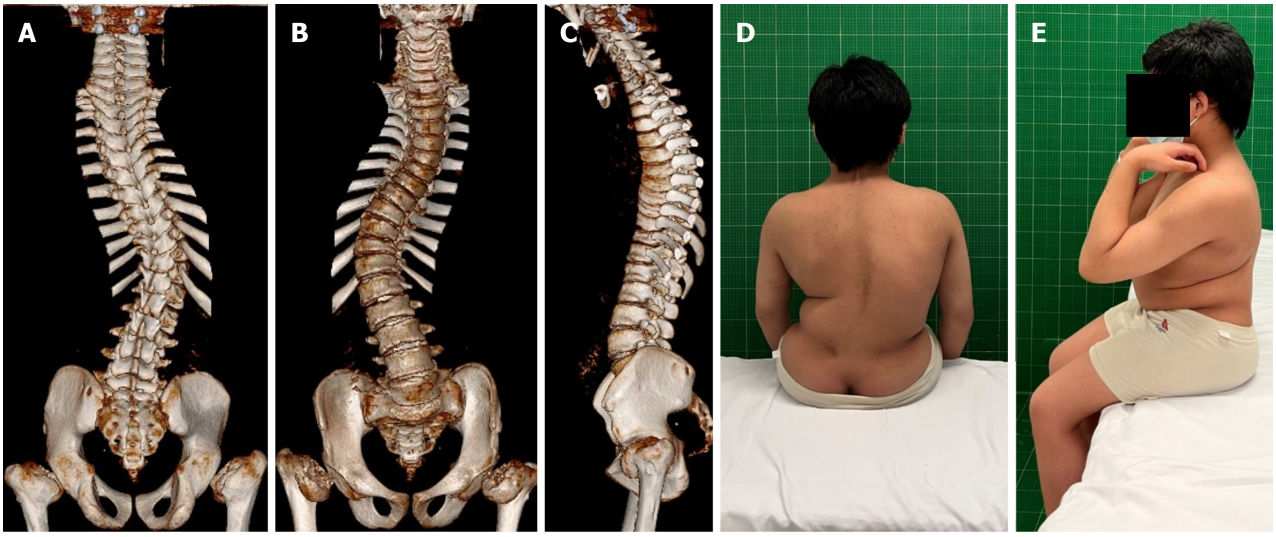

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Figure 2 Preoperative computed tomography three-dimensional reconstruction of the spine and preoperative appearance. A-C: Computed tomography three-dimensional of the spine shows postoperative changes of atlantoaxial vertebrae, scoliosis, flattening of the vertebral bodies, and dysplasia of the femoral head; D: Views from the back show unevenness of the back and right deviation of the trunk; E: Lateral views before the operation.

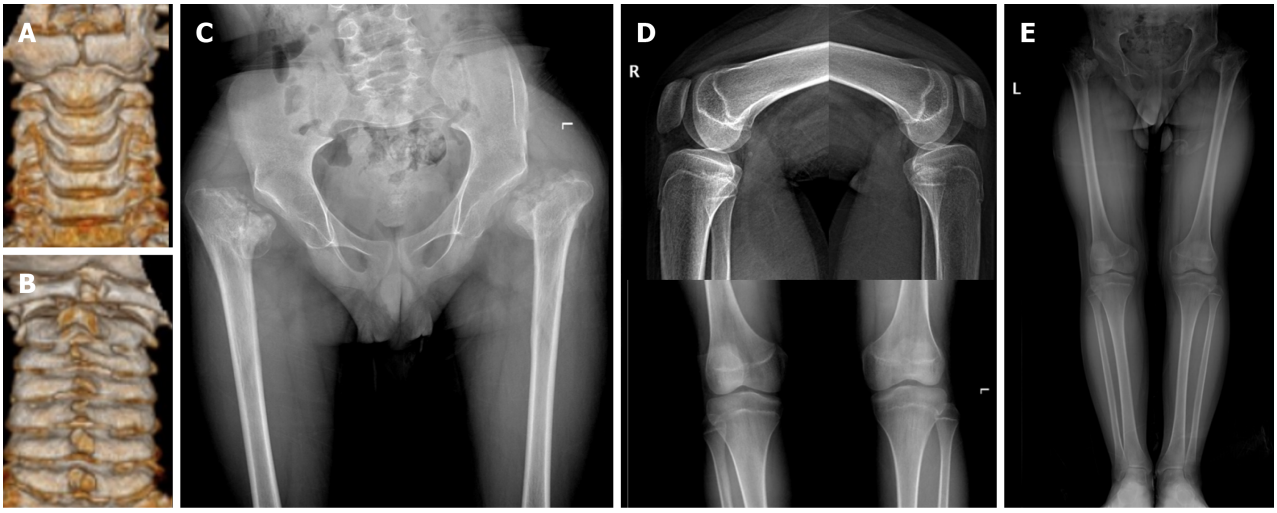

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Figure 3 Computed tomography three-dimensional reconstruction of the cervical spine and X-ray of the hip. A: The view from the front shows dysplasia of the anterior arch of the atlas and the odontoid process of axis; B: The view from the back shows dysplasia of the posterior arch of the atlas; C: A slight right inclination of the pelvis, dysplasia of bilateral hip joints, and severe damage to femoral heads are observed; D: No obvious abnormality is found in the X-ray of bilateral knee joints; E: X-ray of both lower limbs indicate that both lower limbs are equal in length.

DISCUSSION

SEDC is a rare genetic disorder that affects skeletal growth and development. Diagnosis of SEDC involves a combination of clinical examinations, radiographic imaging, and genetic testing[1,11]. The radiographic findings typically include flattened vertebrae, shortened long bones, and hip dysplasia. Due to the low incidence of SEDC and its main clinical feature of short stature, it can be easily confused with relatively common skeletal dysplasia that also cause short stature, such as osteogenesis imperfecta, multiple epiphyseal dysplasia, and achondroplasia[12-14]. In addition, mutations in COL2A1 can cause various skeletal dysplasia, such as Kniest and Stickler dysplasia, which can also be easily confused with SEDC[15]. Therefore, it is sometimes difficult to distinguish SEDC from these diseases based only on clinical examination and imaging findings. However, genetic testing can confirm the diagnosis and identify the specific genetic mutation responsible for the disorder. Consequently, genetic testing has become an important tool for the diagnosis of SEDC. Barat-Houari *et al*[15] analyzed the phenotypes and genetic diagnoses of over 700 patients with type II collagenopathy and identified 415 different COL2A1 mutations. Our patient presented with skeletal dysplasia in infancy; however, owing to the lack of genetic testing at that time, the diagnosis was not confirmed until the patient was 16 years of age. His genetic testing revealed a COL2A1 heterozygous mutation c.2965C>T(p.Arg989Cys), which is the most

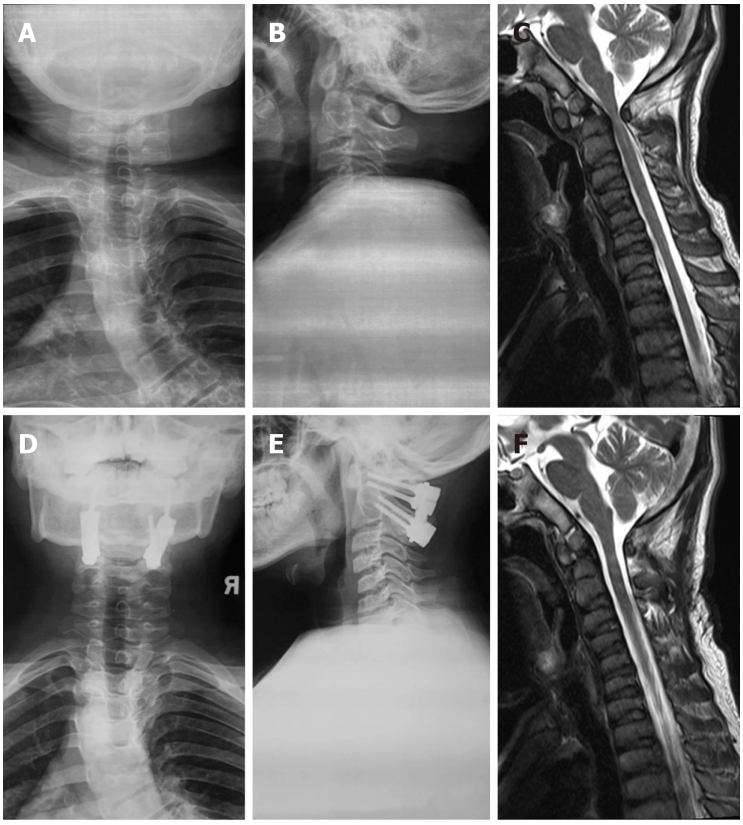

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Figure 4 Imaging data of atlantoaxial dysplasia before and after surgical treatment. A and B: Preoperative X-ray shows the widening of space between the anterior arch and odontoid process and dysplasia of the odontoid process; C: Preoperative MRI shows atlas-level spinal canal stenosis, spinal cord compression, and mild degeneration; D and E: Postoperative X-ray shows atlantoaxial fixation and fusion; F: The compression of the atlantoaxial spinal cord is relieved after the operation.

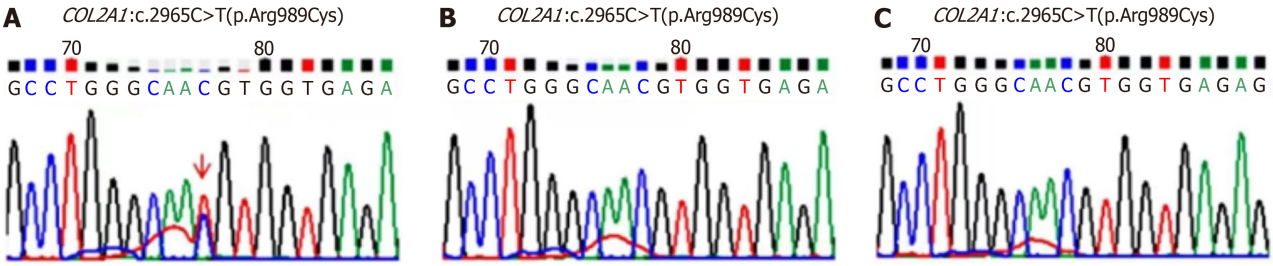

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Figure 5 Gene detection and Sanger sequencing in patients' pedigrees. A: The COL2A1 heterozygous mutation c.2965C>T(p.Arg989Cys) was found in the proband; B and C: The same pathogenic mutation was not found in the parents of the proband.

common mutation associated with SEDC[15]. The spectrum of gene mutations in SEDC will be further expanded and may guide the treatment of SEDC[16]. Genetic testing should be recommended for patients with clinical and radiological evidence suggestive of SEDC. Early diagnosis is crucial for providing appropriate follow-up treatment recommendations and genetic counselling for families affected by SEDC.

Currently, there are no specific therapeutic interventions for COL2A1 mutations, and treatments primarily focus on managing skeletal deformities. Surgical treatment for SEDC aims to correct deformities and prevent complications, such as joint pain, stiffness, and spinal cord compression. Treating hip dysplasia in SEDC can be challenging owing to the severe deformity and small volume of femoral heads, and often concomitant knee and spinal deformities. However, developmental dysplasia of the hip is an important factor that affects the walking ability of patients with SEDC. Shetty *et al*[17] performed bilateral valgus-extension osteotomy with hybrid external fixation in eight adolescent patients with SEDC who were followed up for 2 years and found significant improvements in the Harris hip score and postoperative mobility. Bayhan *et al*[10] reported that in children with SEDC, correcting the hip dysplasia by valgus hip osteotomy can maintain satisfactory walking function during 5 years of follow-up and improve hip mobility. Bisht *et al*[18] used CT to reconstruct the 3D structure of the hip joint and visualize the anatomical structure of the femoral head and neck, which was helpful for selecting implant sizes. Scheduled hip surgery can improve walking ability and delay the progression of

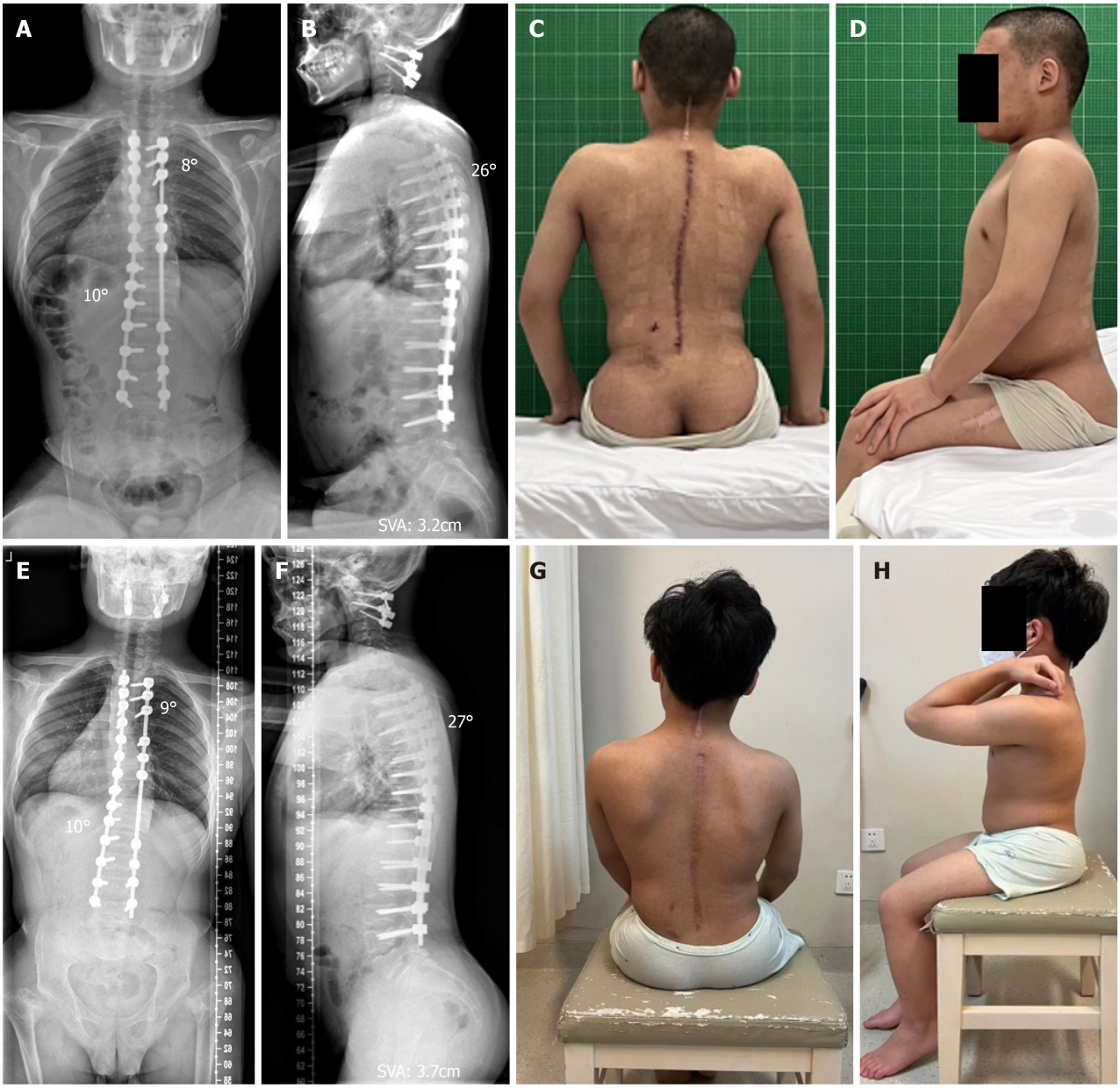

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Figure 6 Images and appearance at one week and two years after the operation. A: Spinal X-ray one week after operation showed that the Cobb angles of the proximal thoracic curve and main thoracic curve were 8° and 10°, respectively; B: The thoracic kyphosis was 26°, and the sagittal vertical axis (SVA) was 3.2 cm; C: Postoperative appearance from the back showed improvements in the unevenness of the back and balance of the trunk; D: Postoperative lateral appearance; E: Spinal X-ray two years after operation showed that the correction of scoliosis remained stable, with the Cobb angles of the proximal and main thoracic curves of 9° and 10°, respectively; F: The sagittal plane remained balanced with a SVA of 3.7 cm, and the thoracic kyphosis was 27°; G and H: The appearance of the back remained good.

hip joint osteoarthritis. However, our patient missed the opportunity for timely hip osteotomy surgery due to a lack of early diagnosis of SEDC. Even though the patient had severe bilateral hip joint damage, the presence of neurological damage due to atlantoaxial dysplasia makes it crucial to prioritize saving neurological function. Additionally, it was observed that the Cobb angle of scoliosis was still increasing three months after atlantoaxial decompression and fixation surgery. To avoid the potential need for more extensive surgery if scoliosis worsens, the decision was made to undergo scoliosis correction surgery before hip replacement surgery.

Among the various skeletal abnormalities associated with SEDC, atlantoaxial dysplasia is common and poses a serious threat to the patient's health. Liu *et al*[19] reported a case of SEDC complicated with atlantoaxial instability, spinal cord compression at the cranio-cervical junction, and quadriplegia. The patient underwent decompression of the cranio-cervical region and occipital-C4 fusion surgery, which improved neurological function after the operation. Serhan *et al*[20] performed upper cervical spine fusion with autogenous iliac crest bone grafting in 20 children clinically diagnosed with SEDC who were followed up for 8 years and reported that the instability of the upper cervical spine was significantly improved, and the bone graft fusion remained stable without any non-union. Miyoshi *et al*[9] analyzed seven patients with SEDC complicated with atlantoaxial subluxation who underwent reduction and fusion surgery and found that when

the diameter of the atlantoaxial spinal canal was less than 10 mm, atlantoaxial plate removal and occipital-cervical fusion were recommended. Al Kaissi *et al*[21] retrospectively analyzed 10 patients with SEDC and found that orthopedic treatment should begin with cervical spine evaluation to avoid severe neurological dysfunction or death. Multiple skeletal abnormalities were detected during the initial examination of our patient. However, atlantoaxial dysplasia exerted the greatest impact on the patient's quality of life. Therefore, atlantoaxial decompression and fixation surgery were recommended, resulting in improved muscle strength and the prevention of further spinal cord damage. From our perspective, surgery for atlantoaxial deformity should be performed promptly if atlantoaxial instability or cervical spinal cord compression is found.

Reports of successful scoliosis correction surgery in patients with SEDC are rare. Beighton *et al*[22] reported two cases of SEDC combined with scoliosis, both of which underwent posterior spinal fusion with Harrington instrumentation; however, the surgeries were unsuccessful. Winter *et al*[23] performed posterior spinal fusion with Harrington instrumentation in two patients with SEDC; during the follow-up period, one patient maintained good correction for 7 years, while the other experienced internal fixation failure due to pseudoarthrosis. Morita *et al*[7] used Luque rods to treat severe thoracolumbar kyphoscoliosis in a patient with SEDC. Although the correction was stable for 6 years postoperatively, scoliosis and kyphosis correction rates were only 10.5% and 25.9%, respectively. With the advancement of pedicle screws, posterior spinal fusion is believed to achieve a good corrective effect[24]. In this study, a high scoliosis correction rate of 84.6% was achieved through one-stage posterior spinal fusion, and there was no internal fixation failure or loss of correction during the 2-year follow-up. The high correction rate may be due to the use of a pedicle screw. In addition, the appropriate timing of treatment is also an important factor, as our patient's preoperative curve angle was 65°, which was smaller than in the five previously reported cases[7,22,23]. Furthermore, the preoperative spinal CT revealed that although the vertebral body in SEDC patients is small and flat, the cancellous bone of the pedicles developed well, suggesting that pedicle screw technology is suitable for scoliosis correction in SEDC. The selection of the upper and lower instrumented vertebrae (UIV and LIV) was also a crucial aspect of the pre-operative planning. Before surgery, the patient had balanced shoulders. The bending X-ray revealed that the proximal thoracic curve exceeded 25°, indicating the necessity for fusion[25]. Consequently, T3 was chosen as the UIV. As for the LIV, the center sacral vertical line (CSVL) passed between the pedicles of L4 in the bending X-ray. However, CSVL did not touch the pedicle of L3, so we opted to select L4 as the LIV. Our case suggests that a reasonable surgical plan, in combination with early treatment of scoliosis can achieve good results.

CONCLUSION

This is the first case report of a patient with SEDC who successfully underwent surgeries for atlantoaxial dysplasia and scoliosis. Early diagnosis of SEDC is important for providing appropriate treatment recommendations, and genetic testing is useful in confirming the diagnosis. Orthopedic evaluation of SEDC should begin with the cervical spine, as atlantoaxial dysplasia can be particularly detrimental. Early surgical intervention for hip dysplasia and scoliosis can result in favorable treatment outcomes. The use of pedicle screws may enhance the efficacy of scoliosis correction in patients with SEDC.

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FOOTNOTES

Author contributions: Cai HY collected data from medical records; Zhao JD reviewed the radiographs; Jiao Y wrote the manuscript; Huang XA provided intellectual support; Shen JX finalised the manuscript and was responsible for this; All authors reviewed the manuscript; Final approval of the manuscript has been obtained from all authors.

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CASE REPORT

Recurrent cyclops lesion after primary anterior cruciate ligament reconstruction using bone tendon bone allograft: A case report

Grayson Kelmer, Andrea H Johnson, Justin J Turcotte, Daniel E Redziniak

Specialty type: Orthopedics

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification
Grade A (Excellent): 0
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Abstract

BACKGROUND

Cyclops lesions are a known complication of anterior cruciate ligament (ACL) reconstruction, with symptomatic cyclops syndrome occurring in up to 11% of surgeries. Recurrent cyclops lesions have been rarely documented; this case study documents the successful treatment of a recurrent cyclops lesion.

CASE SUMMARY

A 28-year-old female presented following a non-contact injury to the right knee. Workup and clinical exam revealed an ACL tear, and arthroscopic reconstruction was performed. Two years later a cyclops lesion was discovered and removed *via* arthroscopic synovectomy. Seven months postoperatively, the patient presented with pain, stiffness, and difficulty achieving terminal extension. A smaller recurrent cyclops lesion was diagnosed, and a repeat synovectomy was performed. The patient recovered fully.

CONCLUSION

To the best of our knowledge, this is the first documented case of recurrent cyclops lesion after bone-patellar tendon-bone allograft ACL reconstruction presenting as cyclops syndrome.

Key Words: Anterior cruciate ligament; Cyclops lesion; Cyclops syndrome; Knee arthroscopy; Anterior cruciate ligament reconstruction; Case report

Core Tip: A 28-year-old female presented following a noncontact injury to the right knee. Workup and clinical exam revealed an anterior cruciate ligament tear, and arthroscopic reconstruction was performed. Two years later a cyclops lesion was discovered and removed *via* arthroscopic synovectomy. Seven months postoperatively, the patient presented with pain, stiffness, and difficulty achieving terminal extension. A smaller recurrent cyclops lesion was diagnosed, and a repeat synovectomy was performed. The patient recovered fully.

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INTRODUCTION

The cyclops lesion is a known complication following anterior cruciate ligament (ACL) reconstruction[1]. Initially described by Jackson and Schaefer[2] in 1990, a nodule of fibrous tissue with central granulation forms on the anterior aspect of the ACL graft and encroaches upon the intercondylar space. When this lesion is associated with a loss of terminal extension accompanied by symptoms such as pain, stiffness, or clunking with full extension, it is referred to as cyclops syndrome[2]. The incidence of cyclops lesions following ACL reconstruction is 25%-47%; however, many of these cases are asymptomatic, do not require surgical intervention, and are not associated with inferior clinical outcomes[3,4]. The rate of symptomatic cyclops lesions presenting as cyclops syndrome is much lower, at 0%-11%[5-8].

When a patient presents with an extension deficit following ACL reconstruction, magnetic resonance imaging (MRI) is the standard of diagnosis for cyclops lesions[3,5,8,9]. Physical therapy (PT) alone is not sufficient to restore full range of motion, arthroscopy with lesion excision is recommended[3,5,8]. Surgical management generally results in long-term resolution of the extension deficit[8-11]. In rare cases the cyclops lesion can recur. Only one such case was documented over two decades ago as part of a larger study on MRI of cyclops lesions[5]. In this report, we present the investigation of a recurrent cyclops lesion in a patient who sustained an ACL tear which was reconstructed using a bone-patellar tendon-bone allograft and ultimately underwent successful resection and rehabilitation.

CASE PRESENTATION

Chief complaints

A 28-year-old female patient presented to the clinic with a sports-related noncontact injury to her right knee.

History of present illness

The patient experienced a noncontact injury to her right knee while playing soccer nine days prior to presentation. She reports difficulty ambulating and instability in the knee.

History of past illness

No prior knee injuries.

Personal and family history

Personal and family history was non-contributory.

Physical examination

Examination revealed a painful right knee with effusion, tenderness along the lateral joint line, and positive Lachman test.

Imaging examinations

MRI demonstrated a complete tear of the ACL with associated bone contusions (Figure 1).

FINAL DIAGNOSIS

Recurrent cyclops lesion.

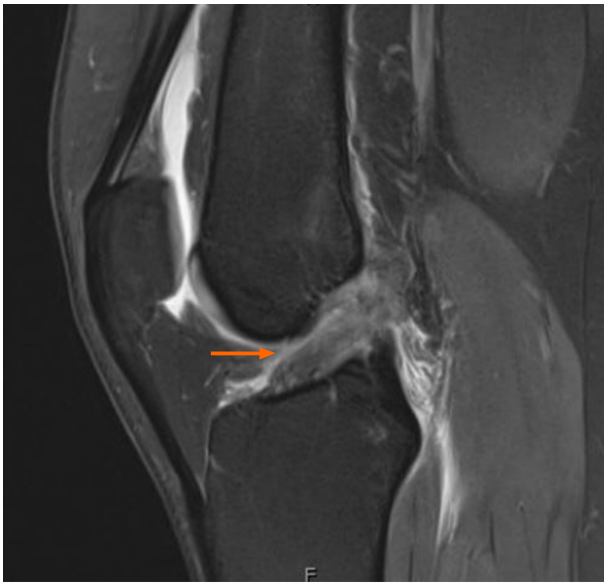

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Figure 1 Sagittal T2-weighted fast spin echo sequence magnetic resonance imaging (3 tesla magnet) revealed complete tear of the right anterior cruciate ligament (orange arrow).

TREATMENT

One week after initial presentation she was re-evaluated and arthroscopic ACL reconstruction was recommended, graft options were reviewed with the patient and a bone tendon bone allograft was selected as her graft of choice. She was provided a derotational brace, and referral to PT. Approximately 2.5 mo following the initial injury, reconstruction of the right ACL using bone-patellar tendon-bone allograft was performed. Body mass index (BMI) at the time of surgery was 29.9 kg/m^2. The femoral tunnel was prepared using an anteromedial portal technique utilizing a limited notchplasty. Bioabsorbable interference screws were used for both femoral and tibial fixation. The knee was placed in extension with a posterior drawer applied to the proximal tibia for final fixation. Figure 2 shows postoperative X-rays indicating appropriate tunnel placement. There were no complications and recovery and PT progressed as expected. She returned to all activities without restrictions.

Two years after the ACL repair, the patient sustained a repeat injury to her right knee, presenting about a month after this incident. The patient reported feeling a pop and medial-sided knee pain. Physical examination revealed swelling, medial joint line tenderness, and positive McMurray test. Radiographs were unremarkable. MRI demonstrated an intact ACL graft with a moderate-sized cyclops lesion estimated at a size of 16 mm × 17 mm × 11 mm (Figure 3). After 8 wk of PT, she still complained of stiffness and moderate pain with knee extension. Right knee arthroscopic synovectomy was recommended, and the patient proceeded with surgery approximately 1 mo later. BMI at the time of surgery was 26.7 kg/m^2.

Intraoperatively, there was an isolated cyclops lesion appreciated at the insertion of the ACL on the proximal tibia, which impinged in the intercondylar notch with terminal extension (Figure 4). A biter and mechanical shaver were used to remove the lesion. Both the ACL and posterior cruciate ligament (PCL) were noted to be intact. Femoral and tibial tunnels were found to be in appropriate position without evidence of notch impingement. Arthroscopic images were taken of the intact cyclops lesion, pre and post arthroscopic debridement. The patient was found to have a stable knee on examination under anesthesia. There were no surgical complications. Postoperatively PT was started immediately, emphasizing obtaining terminal extension with unrestricted passive and active motion. Knee Injury and Osteoarthritis Outcome Score (KOOS) was 76.33 6 wk following surgery.

Seven months postoperatively, the patient presented with complaints of tightness and stiffness of her right knee, difficulty achieving terminal extension, and pain in the anteromedial aspect of her right tibia. An MRI was obtained, demonstrating intact ACL reconstruction with a smaller recurrent mass estimated at a size of 5 mm × 9 mm × 10 mm along the anterior margin of the distal ACL consistent with a recurrent cyclops lesion (Figure 5). Recurrent arthroscopic debridement was recommended after 8 wk of PT failed to improve her symptoms. Biopsy at the time of surgical intervention was also recommended to evaluate for recurrent synovial lesions such as pigmented villonodular synovitis. Two months later, the patient proceeded with a right arthroscopy and debridement of the right knee. BMI at the time of surgery was 27.1 kg/m^2. KOOS one week prior to surgery was 44.91.

Repeat arthroscopy demonstrated a recurrent cyclops lesion adjacent to the anterior aspect of the ACL reconstruction (Figure 6). The ACL and PCL were intact. The medial and lateral compartments showed no meniscus tears or arthritis. A straight biter and grasper were used to obtain biopsies of the cyclops lesion. A mechanical shaver was then used to remove the cyclops lesion in its entirety from the intercondylar notch. Full range of motion with terminal flexion and extension was verified. Arthroscopic images were taken, and the recurrent cyclops lesion was sent for biopsy. Surgery was completed with no complications.

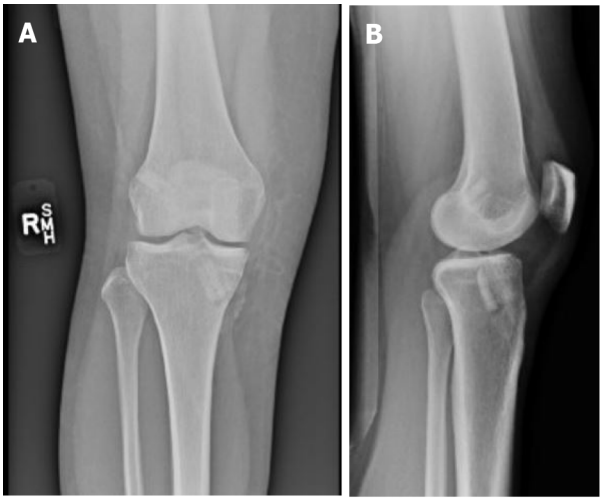

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Figure 2 Postoperative X-ray indicating the correct position of the femoral and tibial tunnels. A: Anteroposterior; B: Lateral.

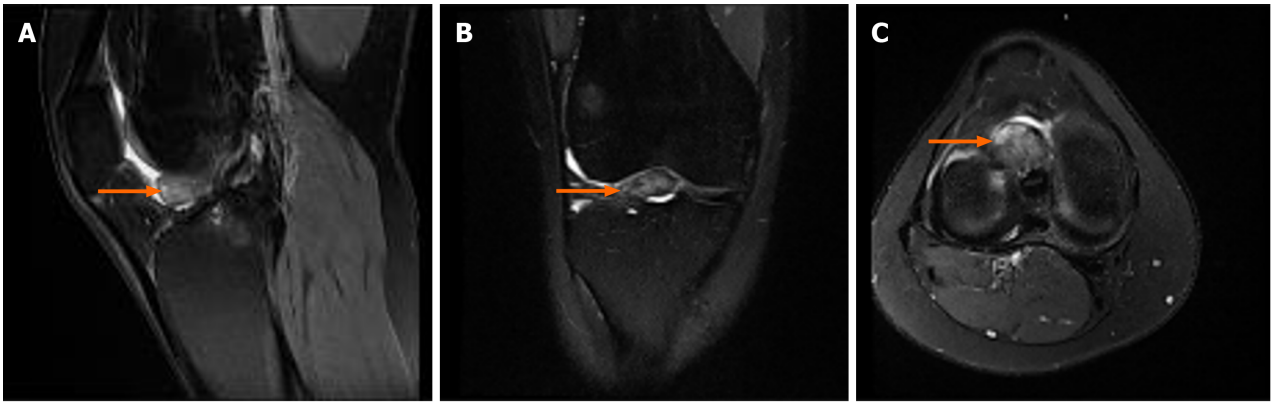

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Figure 3 Postoperative magnetic resonance imaging right knee. A: Magnetic resonance imaging films (3 tesla magnet) in sagittal proton-density fast spin echo with fat saturation (PD FSE FSAT) sequence (orange arrow); B: Coronal T2-weighted FSE FSAT sequence (orange arrow); C: Axial PD FSE FSAT sequence views taken after anterior cruciate ligament reconstruction with cyclops lesion measuring 16 mm × 17 mm × 11 mm (orange arrow).

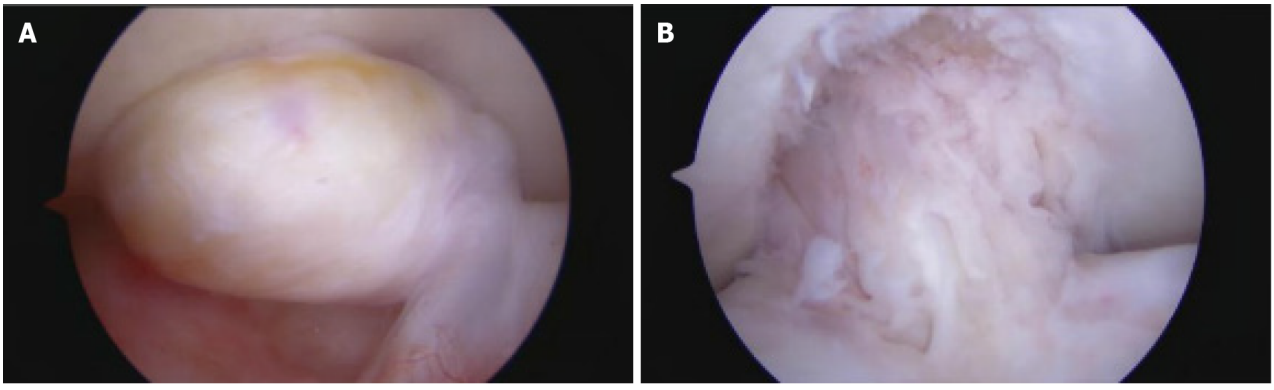

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Figure 4 Images taken during arthroscopic limited debridement of the right knee. A: Cyclops lesion at the proximal insertion of the anterior cruciate ligament; B: The intercondylar notch following removal of the lesion.

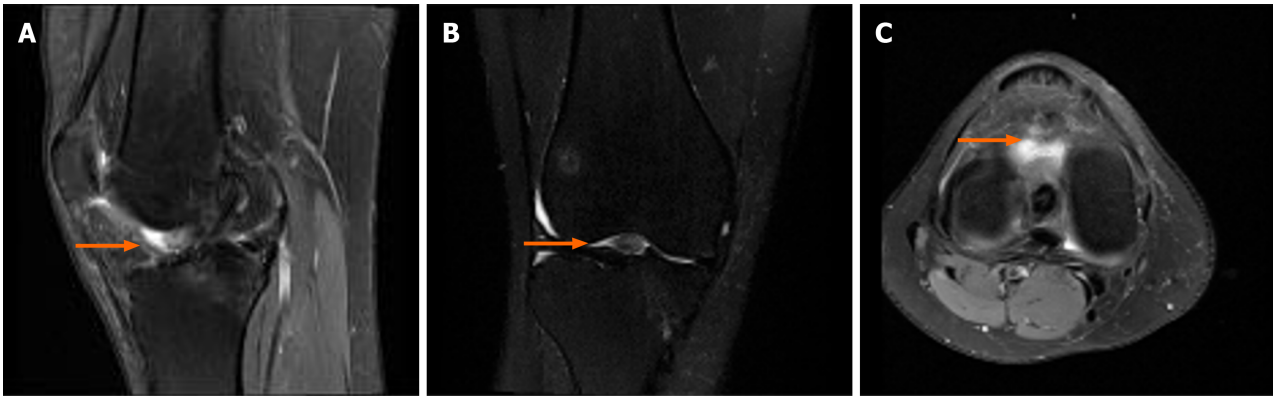

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Figure 5 Postoperative magnetic resonance imaging right knee. A: Magnetic resonance imaging films (3 tesla magnet) in sagittal proton-density fast spin echo with fat saturation (PD FSE FSAT) sequence (orange arrow); B: Coronal T2-weighted FSE FSAT sequence (orange arrow); C: Axial PD FSAT sequence views demonstrated a recurrent cyclops lesion of right anterior cruciate ligament graft with an estimated size of 5 mm × 9 mm × 10 mm (orange arrow).

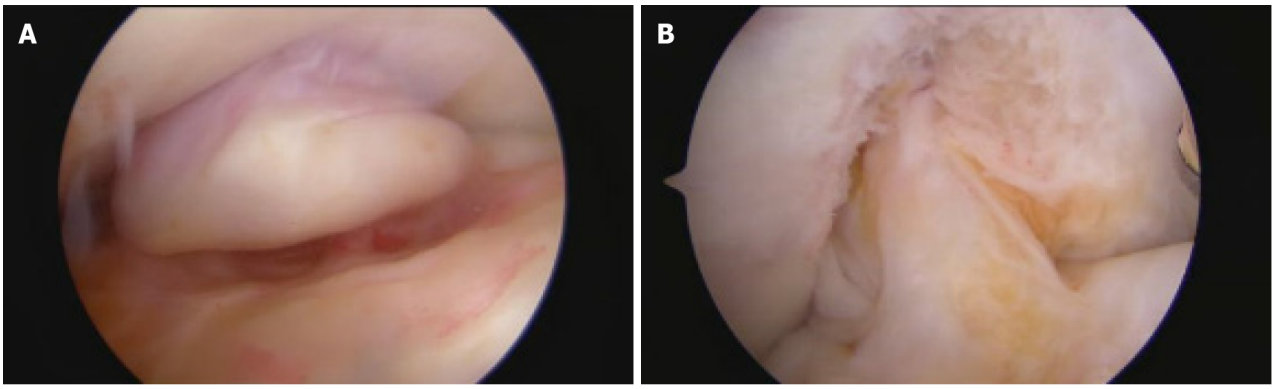

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Figure 6 Images captured during repeat arthroscopy of the right knee. A: Recurrent cyclops lesion; B: The intercondylar notch following debridement.

Pathologic analysis determined the excised mass was composed of synovial tissue with fibrosis and reactive fibrocartilage, consistent with a recurrent cyclops lesion. At 6 wk postoperatively, the patient was progressing through PT and was able to achieve terminal extension. The patient attended PT for 8 wk. By the end of PT, right knee active range of motion (ROM) reached 0-140 degrees, symmetric with the contralateral knee. Upon completion of PT she was able to resume all activities without restrictions.

OUTCOME AND FOLLOW-UP

Nineteen months after arthroscopy for recurrent cyclops lesion, the patient retained full function of the knee with no recurrent stiffness, full active and passive ROM, and able to perform all normal daily and athletic activities without pain; she denies any local recurrence to date. KOOS was 100 and Lysholm score was 100% at final follow up.

DISCUSSION

The mechanism of development and risk factors for cyclops lesions are still under investigation, and the incidence of recurrent cyclops lesions is sparsely documented. The commonly proposed origin of cyclops lesions is development as a result of an inflammatory reaction to remnants of the ruptured ACL[1,6]. Studies have found preservation of ACL remnants was not associated with cyclops lesions or cyclops syndrome, and minimal debridement during ACL repair did not increase incidence of cyclops lesions[4,6,8]. Tomihara *et al*[12] recently found that bone-patellar-tendon-bone autograft, female sex, and increased BMI were significant risk factors for cyclops lesions and syndrome. Conversely, Facchetti *et al*[3] reported no association of cyclops lesion incidence with factors such as graft type or sex but did find a positive correlation with lower patient BMI. Based on direct contradictions in the literature, it is difficult to theorize which demographic or surgical factors may have influenced the repeated development of cyclops lesions in our patient. Additional factors that have been associated with the development of cyclops lesions following ACL reconstruction

include increased graft volume, double-bundle grafts, smaller femoral and tibial tunnel diameter and excessively anterior tibial tunnels, although none of these appear to be contributory in our patient[7,13,14].

It is suggested that early loss of extension following ACL reconstruction allows space for the cyclops lesion to form[4,6,15]. Gohil *et al*[4] found that multiple patients who experienced loss of extension 2 mo postoperatively did not demonstrate cyclops lesion on MRI, but they subsequently developed cyclops lesions with smaller extension deficits. At 3 and 6 wk postoperatively, Pinto *et al*[15] reported an extension deficit was associated with a significant increase in the incidence of cyclops lesions. The major risk factor for cyclops syndrome discovered by Delaloye *et al*[6] was an extension deficit at 3 and 6 wk following ACL reconstruction. The patient presented in our case progressed well through PT following both her ACL reconstruction and initial cyclops lesion excision and at no point in time did she have an extension deficit.

The only other known case of a recurrent cyclops lesion was reported by Bradley *et al*[5] in their investigation of the utility of MRI for diagnosis of cyclops lesions; four months following excision of the initial lesion, one patient demonstrated a recurrent cyclops lesion. The major difference between this case and our case lies in the presentation; the Bradley *et al*[5] lesion was discovered incidentally on postoperative MRI, whereas our patient returned 7 mo postoperatively with cyclops syndrome. Previous literature reports cyclops lesions are predominantly asymptomatic, do not confer poorer clinical outcomes, and do not show an association between lesion size and pain or other symptoms[3]. It is unclear to what extent our patient's cyclops lesions contributed to her clinical presentation. A key aspect may have been the trauma of her repeated valgus injuries, the first causing her ACL tear and the second about two years after the repair. Both recurrent cyclops lesions were successfully managed with repeat arthroscopy and excision[5].

CONCLUSION

While cyclops lesions following ACL reconstruction are a common complication, recurrent cyclops lesions are minimally documented. Here, we describe what is, to the best of our knowledge, the first known case of a recurrent cyclops lesion presenting as cyclops syndrome. Future studies should be aimed at reconciling which demographic and surgical factors contribute to the development of cyclops lesions and syndrome.

FOOTNOTES

Author contributions: Kelmer G was responsible for investigation, data curation and writing-original draft; Johnson AH was responsible for conceptualization, investigation, data curation, writing-review and editing; Turcotte JJ was responsible for writing-review and editing and supervision; Redziniak DE was responsible for conceptualization, writing-review and editing and supervision.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

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