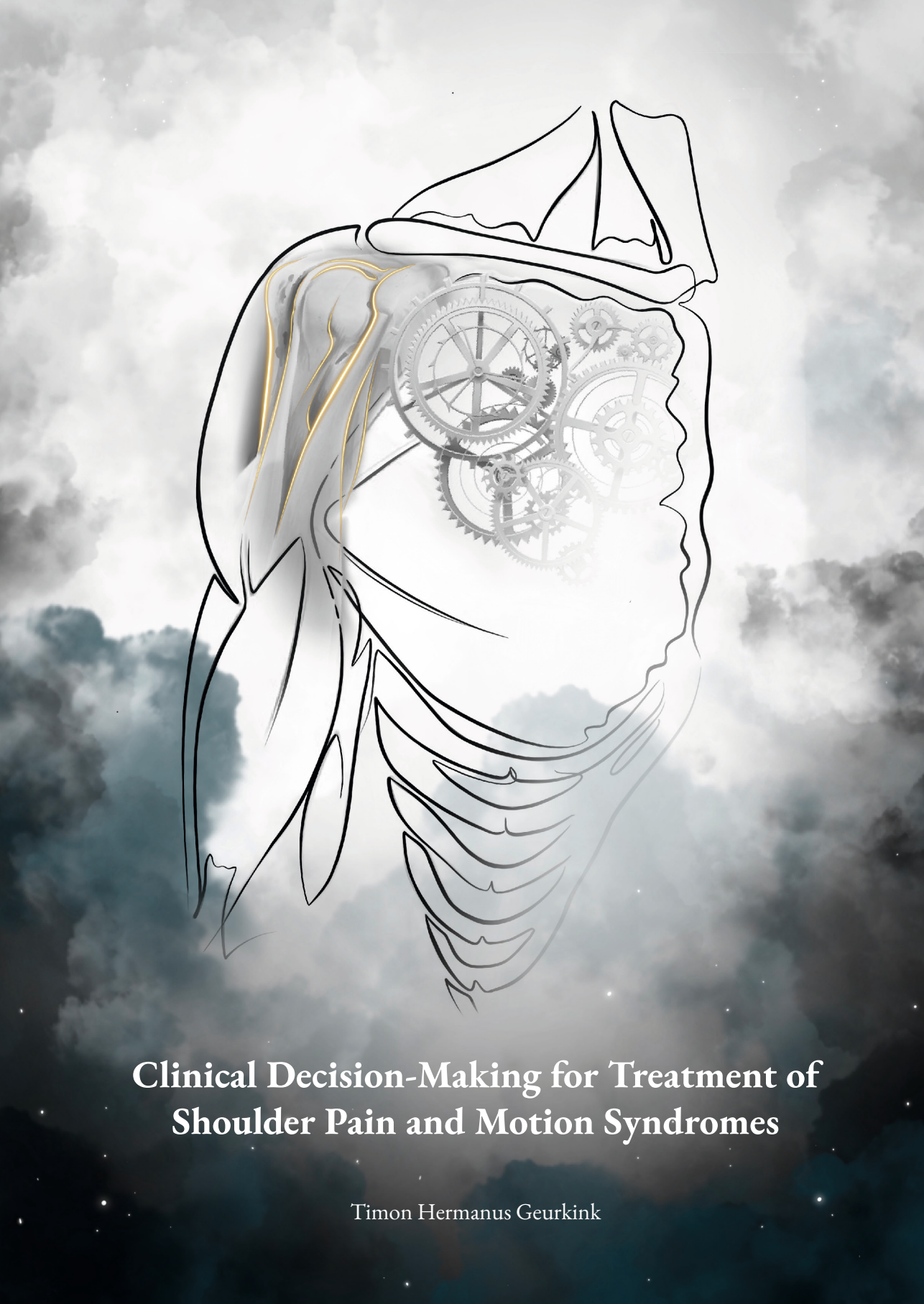

Clinical Decision-Making for Treatment of Shoulder Pain and Motion Syndromes

Timon Hermanus Geurkink

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Promotores:

Prof. Dr. R.G.H.H. Nelissen

Dr. P.J. Marang-van de Mheen

Copromotor:

Dr. Ir. J.H. de Groot

Leden promotiecommissie:

Prof. Dr. I.B. Schipper

Prof. Dr. S. Repping, Amsterdam UMC

Prof. Dr. M.P.J. van den Bekerom, OLVG

Prof. Dr. H.M.M. Vos

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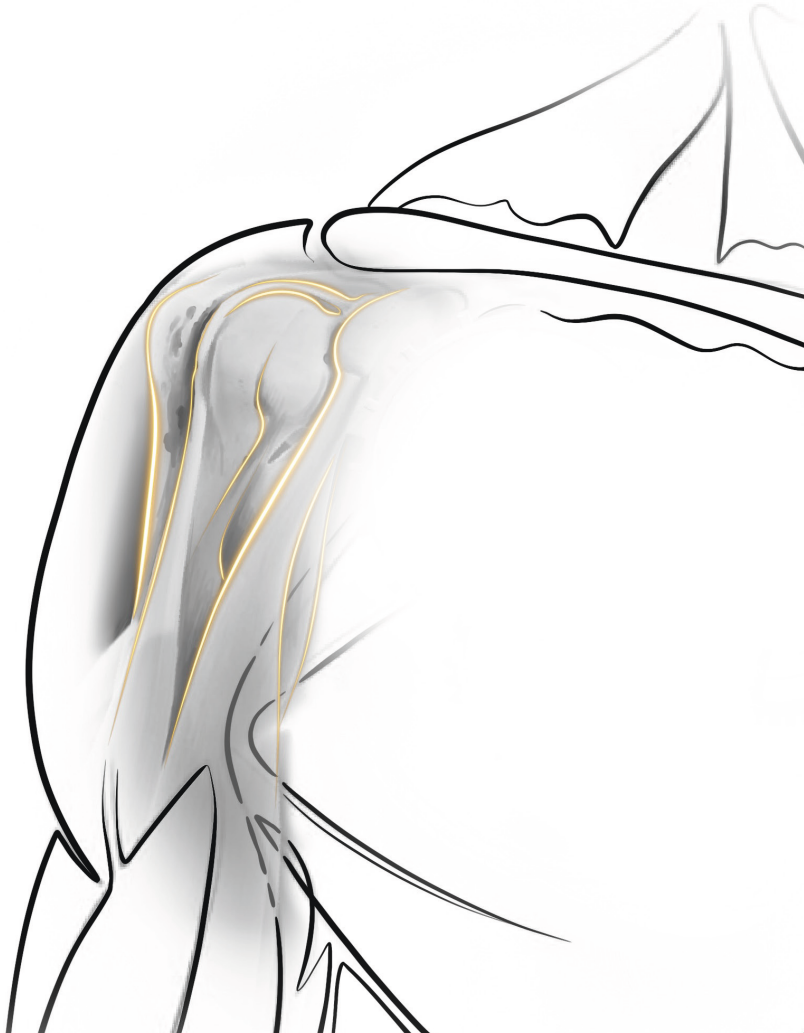

Chapter 1

General introduction and thesis outline

Background

The shoulder joint stands out as the most mobile joint within the human body. This incredible mobility allows an athlete to perform complex athletic manoeuvers but is also needed to perform routine tasks in daily life. Shoulder movements result from a complex, synergistic interplay between the thorax, clavicle, scapula and humerus, collectively forming the bony framework of the shoulder. Key joints include the sterno-clavicular, acromio-clavicular, gleno-humeral and scapulo-thoracic articulations. Of these, the glenohumeral joint, a "simple" ball-and-socket joint, primarily contributes to the enormous mobility of the shoulder joint, although the substantial role of scapulothoracic gliding plane should not be overlooked[1]. Muscles acting on these joints, such as those that form the rotator cuff (i.e. subscapularis, supraspinatus, infraspinatus and teres minor muscle), the deltoid, biceps brachii, coracobrachialis, triceps brachii, pectoralis major, serratus anterior, rhomboid and trapezius muscle facilitate shoulder movement. However, the extensive range of motion comes at a price: more susceptibility to injury.

The true importance of the remarkable freedom of shoulder movement in daily tasks often becomes evident only when individuals experience shoulder pain, shoulder instability, or encounter limitations in mobility. In the Netherlands, the shoulder joint ranks as the second most frequently reported anatomic site of musculoskeletal pain, following the lower back, with a prevalence of 17%[2, 3]. Globally, incidence rates of shoulder pain range from 8 to 62 per 1000 persons per year, and approximately 2% of the adult population seeks medical consultation with new shoulder pain annually[4]. Notably, a significant proportion of patients with shoulder pain, around 50%, continue to report symptoms even six months after onset[5]. Both pain and functional disabilities interfere with work, hobbies and social activities thereby significantly affecting the quality-of-life[6]. In the Netherlands, musculoskeletal disorders constitute the second most expensive disease group for healthcare costs, representing 6% of the overall healthcare expenditure[7]. Consequently, shoulder disorders substantially contribute to the economic burden, while the large proportion of patients with continued pain highlights the challenges clinicians face in diagnosis and treatment decisions[8, 9].

Challenges in the diagnosis and treatment of shoulder complaints

While the clinical presentation of pain and limited motion is relatively easily defined, diagnosing and treating shoulder complaints is challenging. Pinpointing the exact cause of shoulder pain and dysfunction can be difficult not only due to the complex shoulder anatomy through which the shoulder enables mobility, encompassing a diverse range of potential anatomical sources, but also since pain may be referred from different anatomical locations and patient may have e.g. pain sensitisation[10]. Focussing on anatomy, only few shoulder disorders can be attributed to a straightforward anatomical origin, such as neurogenic scapular winging resulting from a loss of motor function of either the serratus anterior or trapezius muscle due to pathology of the long thoracic or spinal accessory nerve

respectively[11, 12]. Such a clear-cut culprit of shoulder pathology provides the clinician with specific treatment targets. However, these shoulder disorders are rare and the low number of cases often translates into a scarcity of high-quality evidence regarding optimal treatment strategies and their (long-term) outcomes. Clinical decision-making in these cases therefore often relies heavily on expert opinion and anecdotal evidence, and this limited evidence-based guidance may lead to uncertainty and variability in clinical decision-making.

In most cases of shoulder complaints, however, such a clear-cut cause of the shoulder complaints is lacking. Clinicians therefore struggle to accurately identify the underlying cause due to the subjective nature of symptoms, the complex shoulder anatomy, the overlap in clinical features between different shoulder disorders, and the lack of reliable diagnostic tests for shoulder pathology[13, 14]. Relying solely on imaging for the diagnosis of shoulder complaints also poses a risk, as it may reveal findings unrelated to the patients' complaints since asymptomatic degenerative changes increase during life; for instance, acromioclavicular osteoarthritis is seen by MRI in about 30-40% of asymptomatic patients[15]. Similarly, rotator cuff tears are observed in 50% of asymptomatic individuals by the age of 70[16]. The latter emphasizes our incomplete understanding of the natural, physiological, processes in the shoulder, making it even more complex for clinicians to differentiate between normal physiolocial (i.e. "healthy" ageing) and pathological conditions due to the lack of a clear reference "of what can be considered normal". A decline in proprioception, for example, is associated with various shoulder disorders, such as rotator cuff disease, frozen shoulder and subacromial pain syndrome, but the association between ageing and proprioception in the asymptomatic shoulder is still unclear[17-19]. Alongside these diagnostic hurdles, the optimal treatment for the individual patient with shoulder problems -particularly when there is no clear culprit- remains uncertain due to the inherent lack of a specific treatment target. Adding to the complexity, each patient necessitates a personalized approach that considers a broader spectrum of factors such as age, activity level, comorbidities, treatment preferences as well as participation role in society. Lastly, the ever-evolving landscape of evidence-based interventions and surgical techniques aimed at improving outcomes, does also add another layer of complexity to the clinical decision-making process in these often not well defined shoulder pathologies.

Emerging new evidence and its impact on clinical decision-making

In the majority of shoulder complaints, the precise causes of these complaints remains unknown, which hinders decision-making on what is considered the optimal treatment. Subacromial pain, for example, accounts for the majority (up to 70%) of shoulder pain[20]. Historically, it was thought to be the result of impingement of rotator cuff tendons between the humeral head and the acromion, i.e. subacromial impingement syndrome. This turned out to be a too simplistic thinking as the pathogenesis of subacromial pain results from a variety of factors and not just mechanical impingement[21, 22 14, 23]. Due to the limited understanding of its origin, clinicians concluded in 2014 that it was more fitting to label it as a pain syndrome,

i.e. subacromial pain syndrome (SAPS)[14]. Nonetheless, treatment strategies for SAPS still lean heavily on the historical perspectives of the impingement theory.

Subacromial decompression (SAD) surgery, for example, aims to relieve impingement of the rotator cuff by resecting the anteroinferior part of the acromion. Various high-quality studies, however, found that SAD surgery provides no significant improvement in pain nor functionality in SAPS patients when compared with placebo surgery or non-surgical management, consistent with the current understanding that impingement is not the main cause of (subacromial) shoulder pain[24, 25]. Consequently, SAD surgery is now deemed "low-value care", a term reflecting procedures with little or no benefit, and even potential harm to patients[26]. The extent to which publication of such high-quality evidence affects clinical decision-making in daily clinical practice is still unclear. Timely implementation of evidence is, however, crucial for both clinicians and their patients since performing low-value procedures not only results in suboptimal treatment with potential side effects to patients, but also results in increasing healthcare costs[27].

Despite evidence indicating no benefit, SAD surgery is still frequently performed as shown by increasing trends observed in the United Kingdom, the United States, and Australia[28-30]. In the Netherlands, approximately 10.000 SAPS patients underwent SAD surgery in 2016[31]. There is a gap in knowledge on what factors drive clinicians towards the continued use of low-value care procedures. Reasons for such decision-making may include specific patient characteristics, surgeon characteristics, surgeons' perception of benefit or harm of surgical intervention, their knowledge and interpretation of guidelines, or a lack of alternative treatment options[32-34]. Several initiatives have been launched in the Netherlands that aim to influence clinical decision-making towards reducing the use of SAD surgery, such as clinical guideline changes and active disinvestment strategies (withdrawal of reimbursement through a policy change)[31, 35]. Such initiatives have, however, shown to be a complex undertaking and the mechanism through which they may exercise their effect on clinical decision-making remains unclear[36, 37].

Aim and outline of this thesis.

This thesis aims to contribute knowledge into the factors that are needed for and drive clinical decision-making on treatment of shoulder pain and motion syndromes with and without well-defined pathological entities. **Chapter 2** systematically reviews the current available evidence on both surgical and non-surgical management of neurogenic scapular winging, where individual studies may be hampered by low numbers of patients, to guide clinicians on optimal treatment for this rare motion syndrome. **Chapter 3** further builds upon this by investigating and presenting data on the long-term outcomes of patients who underwent pectoralis major transfer for neurologic scapular winging due to long thoracic nerve palsy, of which there is little knowledge. In **Chapter 4**, we explored the role of proprioception in shoulder pathology by assessing the association between ageing and proprioception in

an asymptomatic population, thereby providing clinicians with a reference of what can be considered normal kinematics of the shoulder in an ageing population. Furthermore, we explored how we could best implement new knowledge once it becomes available, particularly for complex disorders such as SAPS. We examined the impact of publishing high-quality evidence on clinical decision-making in **Chapter 5**, by assessing the effect of two randomized controlled trials showing no benefit of SAD surgery on volumes of SAD surgery in six hospitals across five countries. In **Chapter 6,** we aimed to understand the variability in decision-making and the factors driving clinicians to the continued use of SAD surgery using a case-vignette study with identical scenarios, thereby ruling out differences in patients explaining difference in treatment decisions. Lastly, we explored through what mechanisms a specific intervention (i.e. active disinvestment strategy by healthcare insurers) aiming to reduce SAD surgery for SAPS was affecting the clinical decision-making process in daily clinical practice (**Chapter 7)**. A summary of the main result of the studies described in this thesis and a general discussion including future perspectives is provided in **Chapter 8.**

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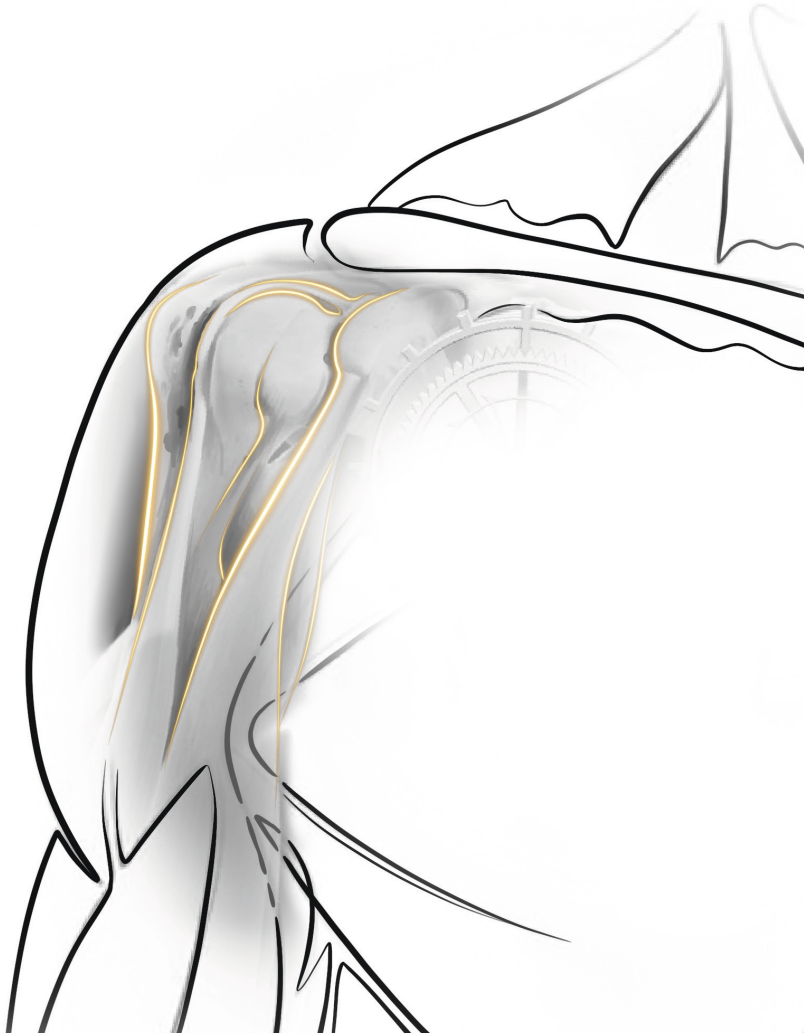

Chapter 2

Treatment of neurogenic scapular winging: a systematic review on outcomes after nonsurgical management and tendon transfer surgery.

Timon H. Geurkink MD[1,5], Hamez Gacaferi MD[2], Perla J. Marang-van de Mheen PhD[3], Jan. W. Schoones[4], Jurriaan .H. de Groot PhD[5], Jochem Nagels MD[1], Prof Rob G.H.H. Nelissen MD PhD[1]

1. *Department of Orthopaedics, Leiden University Medical Center, Postbus 9600, 2300 RC, Leiden, the Netherlands.*
2. *Botnar Research Centre, Nuffield Department of Orthopaedics, Rheumatology and Musculoskeletal Sciences (NDORMS), University of Oxford, Windmill Road, Headington, OX3 7LD, Oxford, United Kingdom.*
3. *Department of Biomedical Data Sciences, Medical Decision Making, Leiden University Medical Centre, Leiden, the Netherlands.*
4. *Directorate of Research Policy (formerly: Walaeus Library), Leiden University Medical Center, the Netherlands.*
5. *Laboratory for Kinematics and Neuromechanics, Department of Orthopaedics and Rehabilitation, Leiden, University Medical Center. The Netherlands.*

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ABSTRACT

Background:
Scapular winging is a rare condition of the shoulder girdle that presents challenging treatment decisions for clinicians. To inform clinical practice, clinicians need guidance on what the best treatment decision is for their patients and such recommendations should be based on the total evidence available. Therefore, the purpose of this review was to systematically review the evidence regarding nonsurgical management and tendon transfer surgery of patients with neurologic scapular winging due to serratus anterior (SA) or trapezius (TP) palsy.

Methods:
PubMed, Embase, Web of Science, Cochrane Library, Emcare and Academic Search Premier were searched up to April 5, 2022, for studies reporting on clinical outcomes after nonsurgical management and tendon transfer surgery of scapular winging due to weakness of the SA or TP muscle**.** The ICROMS-tool was used to classify the quality of the studies. Primary outcomes were: the fraction of patients with spontaneous recovery after nonsurgical management and improvement in shoulder function, pain- and shoulder scores after tendon transfer surgery. Data were pooled if data on the same outcome were available for at least three studies, using random effects meta-analysis.

Results:
Twenty-three (ten moderate-quality (MQ) and thirteen low-quality) studies were included. Six studies (three MQ; 234 shoulders) reported on outcomes after nonsurgical management of SA palsy, whereas twelve (six MQ; 221 shoulders) and six studies (one MQ; 80 shoulders) evaluated the outcome of tendon transfer for SA- or TP palsy(one study addressed both). Spontaneous recovery of scapular winging with nonsurgical management varied between 21 and 78% across studies after a median follow-up of 72 months. For surgical management of SA palsy, pooling data in a meta-analysis showed that patients on average improved by 47 degrees (95% CI: 34-61, P <0.001) in active forward flexion, had lower VAS-scores for pain(mean difference (MD): -3.0, 95% CI: -4.9- -1.0, P = .003) and had substantial improvements in American Shoulder and Elbow Surgeons (MD: 24, 95% CI: 9-39, P = 0.002) and Constant scores (MD: 45, 95% CI: 39-51), P <0.001). Patients with TP palsy on average improved by 36 degrees (95% CI: 21-51, P <0.001) in active forward flexion after tendon transfer. Statistical pooling was not possible for other outcome measures as insufficient data were available.

Conclusion:
A substantial part of nonsurgically managed patients with scapular winging seem to have persistent complaints, which should be part of the information provided to patients. Data pooling demonstrated significant improvements in shoulder function, pain- and shoulder scores after tendon transfer surgery, but higher quality evidence is needed to allow for more robust recommendations and guide clinical decision-making on when to perform such functional surgery.

INTRODUCTION

Scapular winging, or scapula alata, is a rare scapulothoracic disorder with altered motion and positioning of the scapula (i.e. scapular dyskinesis), characterized by medial border prominence of the scapula with respect to the thorax either at rest or during motion[1, 2]. This abnormal scapular motion originates from the inability of the scapulothoracic muscles to stabilize the scapula against the thorax and can be caused by various different causes, including neurologic injury, soft-tissue and bone abnormalities or it may be secondary to other disorders of the shoulder joint[3, 4]. Frequently, it has a neurologic origin that results in a loss of motor function of either the serratus anterior (SA) or trapezius (TP) muscle, because of pathology of the long thoracic or spinal accessory nerve respectively[5, 6]. Scapular winging has been associated with a great variety of underlying etiologies (e.g. trauma, inflammation, iatrogenic injury, myopathy) [7-9] and therefore it is often misdiagnosed in clinical practice[10-12].

In clinical practice, scapular winging is often associated with pain, weakness and decreased active range of motion of the shoulder[5, 9]. Most patients with a functional deficit due to scapular winging are thought to recover spontaneously within 24 months, but this is based on only a few studies[6, 13]. Nonsurgical management (e.g. prevention of overuse, physical therapy) can be given to relieve symptoms and maintain shoulder function[3]. Tendon transfer surgery can be considered for patients without functional recovery after two years of nonsurgical management[6, 14]. These surgical techniques aim to restore scapulothoracic motion by transferring the pectoralis major (PM) or the rhomboids and levator scapulae muscles (Eden-Lange procedure) to the scapula as a substitute for the loss in SA or TP function, thus improving shoulder functionality[3, 5, 6].

Several studies have examined the outcomes after nonsurgical management and tendon transfer surgery for scapular winging due to SA or TP palsy but these often included small numbers of patients. Therefore the results of individual studies may not be generalizable to the general population and provide limited information for clinician decision-making. To inform clinical practice, clinicians need guidance on what the best treatment decision is for their patients and such recommendations should be based on the total evidence available. Therefore, the purpose of this study was to systematically review the evidence regarding the effect of nonsurgical management and tendon transfer surgery in adult patients with scapular winging due to SA or TP palsy and to increase the statistical power by pooling data. Specifically, we wanted to assess (1) which fraction of patients recover with nonsurgical management and over what period of time, and (2) to evaluate to which extent patients improve after tendon transfer in pain, function and shoulder scores as well as the amount of complications.

METHODOLOGY

Protocol and registration

This review has been conducted following the published guidelines by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[15]. The review protocol was registered at PROSPERO - international prospective register of systematic reviews (registration number: CRD42020203579) before conducting the search[16].

Search strategy and selection

In cooperation with a trained information specialist (JS), a search strategy was composed. The following databases were searched up to April 5, 2022: PubMed, Embase (OVID version), Web of Science, Cochrane Library, Emcare (OVID version), and Academic Search Premier (EBSCOhost). The query consisted of the phrasing of various variants of scapular winging (see Appendix S1). In addition, we checked the reference lists from all included studies for any potentially related articles not identified by the initial search.

After the primary search, the list of references was imported to EndNote (Version X9, Clarivate Analytics, Philadelphia, USA) to remove duplicates and subsequently exported to Rayyan for study selection[17]. Two researchers (TG, HG) independently screened all titles and abstracts identified by the search strategy. All articles reporting on clinical outcomes after nonsurgical management or tendon transfer surgery for scapular winging in adult patients due to weakness of the SA or TP were assessed for eligibility. The exclusion criteria were: cadaveric or animal studies, adolescent population (under 18 years of age; single cases in a larger group were no reason for exclusion), scapular winging caused by myopathic disorders (e.g. facioscapulohumeral muscular dystrophy; single cases in a larger group were no reason for exclusion), other surgical interventions than tendon transfers (e.g. nerve surgery or scapulothoracic fusion) and insufficient clinical outcome data reported (i.e. nonsurgical management studies had to report on the fraction of patients who recover with nonsurgical management, while at least one of the following outcomes had to be reported for surgical studies: pain scores, range of motion or shoulder scores). Meta-analysis and systematic reviews were not included, but were checked for individual studies that could be included. Letters to the editor, meeting abstracts, and case-reports were also excluded as these do not contain empirical data, have insufficiently detailed information or precede a fully published article. In addition, publications in other languages than Dutch or English and articles in non-peer-reviewed journals were also excluded. All eligible studies were assessed for study population overlap. In case of overlapping populations in different publications, the author was contacted to verify this and the study with most complete data was included. Reasons for exclusion at each stage were recorded and are shown in a PRISMA flow diagram (Figure 1). In case of uncertainty regarding the eligibility of an article, disagreements were solved by means of discussion with a third independent reviewer (JN).

Assessment of methodological quality

Quality assessment was performed independently by two researchers (TG, HG) using the Integrated quality Criteria for Review Of Multiple Study designs (ICROMS) tool[18]. This grading system allows for the assessment of a large range of study designs. The ICROMS tool scores seven domains on an ordinal scale, for which the specific criteria considered in every domain are described in Appendix S2. Each specific criterion was assessed as being met (2 points), unclear (1 point), or not met(0 points). Studies must meet mandatory criteria (indicated in bold in Appendix S2) and a minimum score to be included for evidence synthesis, depending on the study design. For non-controlled before-after studies, which is the design of all studies included in the present systematic review, the minimum score is 22. However, we opted to include all studies as valuable information can be lost because of the exclusion of studies, but used the risk of bias score to interpret the quality of individual studies. Studies scoring at least 22 points and fulfilling the mandatory criteria were therefore classified as high quality (HQ) studies. Studies scoring at least 22 points, but failing to fulfil the mandatory criteria were classified as moderate quality studies (MQ). Studies scoring less than 22 points were classified as low quality (LQ) studies. Disagreements were solved via discussion with a third reviewer (JN) and reaching consensus.

Data extraction and synthesis of results

Two researchers (TG, HG) independently extracted the data from the included articles using a standardized data-extraction sheet. The following data were extracted from all articles: author, title, year of publication, study design, diagnostic criteria for scapular winging, intervention, number of patients/shoulders, patient characteristics (e.g. age, sex, causative factors of winging, symptom duration and the extent of scapular winging), duration of follow-up and clinical outcomes, including fraction of patients with spontaneous recovery (i.e. resolved scapular winging as defined in each study), time to recovery, shoulder function, pain scores, shoulder scores, residual winging as defined in each study (See Appendix S3) and complications. Outcomes of studies were collected in the original units including range, confidence intervals (CIs) or standard deviations (SD).

Data were pooled in a meta-analysis if a specific outcome measure was reported in at least three articles studying the same intervention, using a random-effects model, in RevMan v5.4[19]. The differences in outcomes before and after the intervention were calculated and analyzed using weighted mean differences along with the 95% confidence intervals (95% CIs). If the 95% CIs were lacking, they were calculated from reported P values[20]. Forest plots were used to present the results from individual studies and the pooled effect size. Heterogeneity in the pooled effect was evaluated using the I^2 index. We considered I^2>50% to indicate substantial heterogeneity[21]. A P value of < .05 was considered significant. The synthesis without meta-analysis (SWiM) was used for the narrative description of data that did not allow pooling[22].

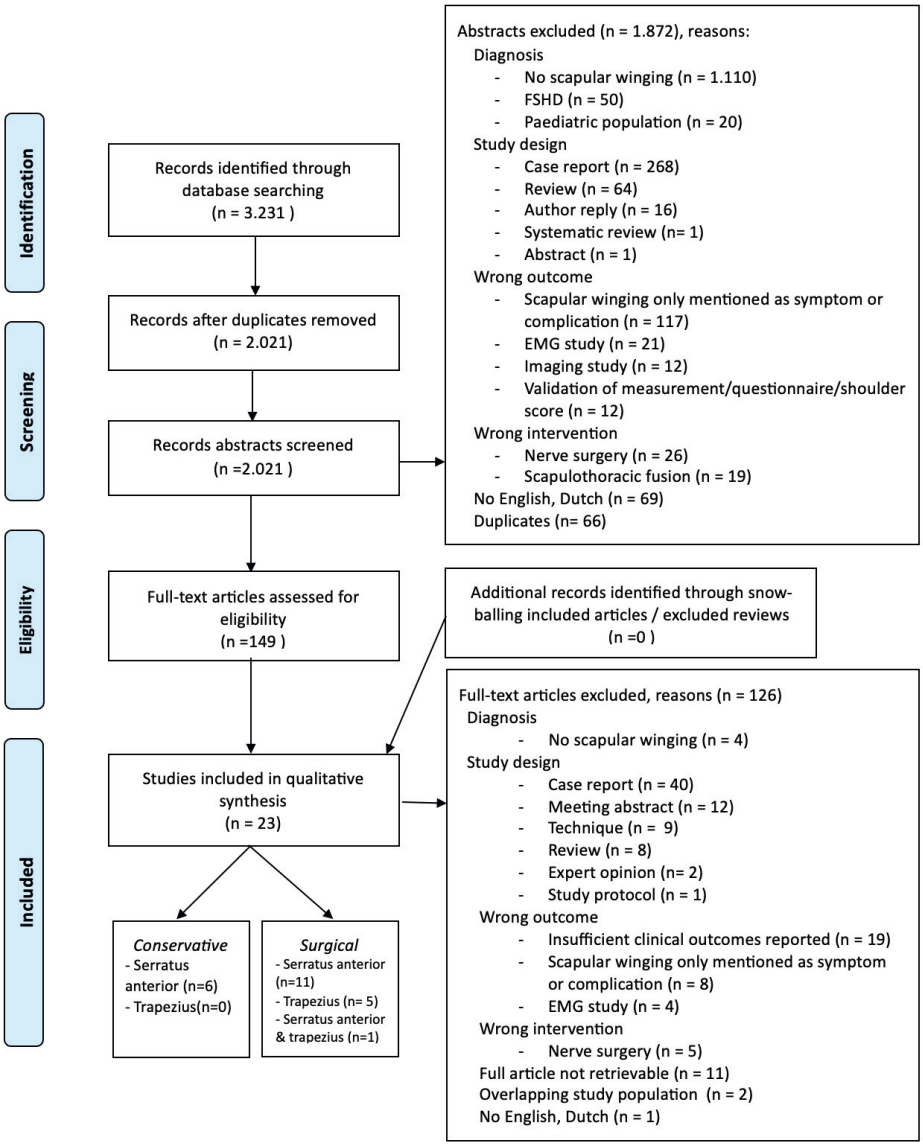

Figure 1. Flow diagram

RESULTS

Study selection

The search yielded 3.231 records of which 2.021 unique records. After screening abstracts for eligibility, 1.872 records were excluded, leaving 149 full-text papers to be screened on eligibility. A total of 11 papers (mostly old, range: 1945-1998), could not be retrieved and were excluded from analysis. Two studies were excluded after consultation of the authors because

they described overlapping populations[23, 24] with more complete papers with regard to the research questions[13, 25]. Finally, 23 studies were included for evidence synthesis (Figure 1). No additional studies were found by checking references of included studies. Six studies evaluated nonsurgical management[8, 13, 25-28] and seventeen studies evaluated surgical treatments[5, 29-44]. No studies compared nonsurgical management with surgical treatment.

Quality assessment
All 23 studies were non-controlled before-after studies. Only ten studies (43%) achieved the minimum ICROMS score (22 points), but all failed to meet all specific mandatory criteria and were therefore labeled as MQ studies (Table 1). ICROMS scores of the other studies ranged from 4 to 20 points, and were labeled as LQ studies. Six of the ten MQ studies were published in 2015 or more recently, compared with only one of the thirteen LQ studies.

Study characteristics
Characteristics of all included studies are shown in Table 2. In all studies, diagnosis was based on clinical assessment of patients, but diagnostic criteria were poorly described (see Appendix S3) and differed significantly between studies (e.g. the number of patients with diagnosis confirmed by electromyography). Across all studies and treatments, 535 shoulders with scapular winging were included. In total, 455 patients (85%) had scapular winging due to SA palsy, mostly caused by trauma (48%). These patients had a mean age of 34 years (SD 3.3), of whom 46% were female. Of these 455 patients, 234 (51%) received nonsurgical management, whereas 221 patients (49%) underwent PM transfer. In the remaining 80 patients (15%) scapular winging was caused by TP palsy, with the majority the result of iatrogenic injury (68%). These patients had a mean age of 35 years (SD 6.8), of whom 55% were female. All 80 patients underwent the (modified) Eden-Lange procedure.

Nonsurgical management
Six studies reported clinical outcomes after nonsurgical management for SA paralysis, three MQ and three LQ studies including a total of 234 shoulders[8, 13, 25-28]. Two MQ and three LQ studies described the natural course of SA palsy without any particular treatment with physical therapy as cointervention in part of these patients, but did not report on the same clinical outcomes. One MQ study evaluated the effect of bracing therapy (Table 1).

All studies reported on the number of patients with recovery of their winging. Completely resolved scapular winging after nonsurgical management varied between studies from 21-78% after a median follow-up of 72 months. Three of these studies (two MQ and one LQ) described the average time to recovery, which varied from 13 to 25 months respectively[8, 13, 27]. Active forward flexion was reported in two MQ studies and improved from 144 and 137 to 161 and 156 degrees respectively after nonsurgical management[13, 25]. Three studies (two MQ and one LQ) described shoulder function in terms of persistent functional limitations after nonsurgical management, with the number of patients with persistent functional limitations varying between 18-42%[13, 25, 26]. Three studies (two MQ and one LQ) reported

on pain: all studies showed that only few patients were completely pain-free with nonsurgical management (12%, 18%, and 30% of patients)[13, 25, 28]. No studies reported on clinical outcomes after nonsurgical management for TP palsy.

Surgical management of SA palsy
Twelve studies (221 shoulders; six MQ and six LQ studies), reported on clinical outcomes after a tendon transfer for SA paralysis and a median follow-up of 47 months(Table 2)[5, 29-37, 39, 40]. Overall, 98 shoulders had a direct PM transfer, whereas 123 shoulders had a PM transfer with augmentation of an allograft or autograft tendon(e.g. tibialis anterior, fascia lata, semitendinosus, gracilis).

Preoperative and postoperative active forward flexion was reported in twelve studies, of which seven studies provided sufficient information for their data to be pooled (Figure 2a)[5, 31, 35-37, 39, 40]. On average, forward flexion improved significantly after PM transfer with a mean improvement of 47 degrees (95% CI: 34-61, P=<0.001), but substantial heterogeneity was present between studies (I^2 = 66%). Studies not providing sufficient information, mostly because the SD was not reported and could not be calculated, showed comparable improvements in means as included studies and were mostly older studies. Six studies (four MQ and two LQ) presented pre- and postoperative VAS scores for pain, of which four MQ studies contributed data to the overall effect in a forest plot(Figure 2b)[35-37, 40]. Overall, VAS scores for pain were significantly lower postoperatively (mean difference: -3.0, 95% CI: -4.9--1,0, P = 0.003, I^2 = 88%). Pooled estimates also showed significant improvements in both American Shoulder and Elbow Surgeons (four studies, mean difference: 24, 95% CI: 9-39, P = 0.002, I^2 = 90%) (Figure 2c)[5, 36, 37, 40] and Constant scores (three studies, mean difference: 45, 95% CI: 39-51), P <0.001, I^2 = 0%) (Figure 2d)[31, 35, 39]. Across all studies, residual winging was observed in 16% of patients who underwent PM transfer. The overall complication rate was 20%, which included infection (3%), failure of the transferred tendon (3%), neurological complaints (4%) and postoperative frozen shoulders (5%) as the most frequently reported. All reported study outcomes can be found in Appendix S3.

Surgical management of TP palsy
There were six studies (80 shoulders), one MQ and five LQ studies, that investigated the clinical outcomes after a tendon transfer for TP paralysis with a median follow-up of 41 months (Table 1)[5, 38, 41-44].

Preoperative and postoperative active forward flexion were reported in one MQ and two LQ studies[5, 38, 44]. Forward flexion improved significantly after Eden-Lange procedure with a mean improvement of 36 degrees (95% CI: 21-51, P <0.001), but substantial heterogeneity was present(I^2 = 73%) (Figure 2e). Statistical pooling was not possible for other outcome measures (pain and shoulder scores) as insufficient data were available. Two LQ studies showed improvements in VAS scores for pain from 7.8 and 7.0 to 1.6 and 2.3 respectively[5, 44]. In addition, substantial improvements in both American Shoulder and Elbow Surgeons

(two LQ studies) and Constant scores (one MQ study) were described after surgery[5, 38, 44]. Across all studies, residual winging was seen in 15% of patients who underwent the Eden-Lange procedure, while 4 complications (8%) were reported. All reported study outcomes can be found in Appendix S3.

TABLE 1. ICROMS Risk of Bias (RoB) Assessment.

Author	1A*	1B*	1C	2C*	3E	3F	4C	5A	5D*	6C	7A	7B	7C	7D	7E	ICROMS score
Li (2017)[45]	2	2	2	2	2	2	1	2	0	2	2	2	2	0	2	25
Elhassan (2015)[39]	2	2	2	2	0	2	2	2	0	2	2	2	2	0	2	24
Vastamaki (2015)[25]	2	2	2	2	0	2	2	2	0	2	2	2	2	0	2	24
Elhassan (2015)[38]	2	2	2	2	0	2	1	2	0	2	2	2	2	0	2	23
Noerdlinger (2002)[32]	2	0	2	2	2	2	2	0	0	2	2	2	2	0	2	22
Tauber (2008)[35]	2	2	2	2	0	2	2	2	0	2	2	2	2	0	0	22
Pikkarainen (2012)[13]	2	2	0	2	0	2	2	2	0	2	2	2	2	0	2	22
Streit (2012)[36]	2	2	0	2	0	2	2	2	0	2	2	2	2	0	2	22
Chalmers (2015)[37]	2	2	2	2	0	2	0	2	0	2	2	2	2	0	2	22
Ng (2021)[8]	2	2	0	2	0	2	2	2	0	2	2	2	2	0	2	22
Galano (2008)[5]	2	2	0	2	0	2	2	2	0	0	2	2	2	0	2	20
Steinmann (2003)[33]	2	0	0	2	2	2	2	0	0	0	2	0	2	0	2	16
Amroodi (2018)[44]	2	2	0	2	0	2	2	2	0	0	2	0	2	0	0	16
Perlmutter (1999)[31]	2	2	0	2	0	2	2	1	0	0	2	0	2	0	0	15
Teboul (2004)[43]	1	0	0	2	0	2	2	2	0	2	1	0	2	0	0	14
Connor (1997)[29]	2	0	0	2	0	2	1	1	0	0	2	0	2	0	0	12
Romero (2003)[42]	2	0	0	2	0	2	2	0	0	2	0	0	2	0	0	12
Warner (1998)[30]	2	0	0	2	0	2	0	2	0	0	0	0	2	0	0	10

TABLE 1. ICROMS Risk of Bias (RoB) Assessment. (continued)

Author	1A*	1B*	1C	2C*	3E	3F	4C	5A	5D*	6C	7A	7B	7C	7D	7E	ICROMS score
Bigliani (1996)[41]	2	0	0	2	0	0	2	1	0	0	2	0	2	0	0	**9**
Kaupilla (1996)[28]	2	1	0	2	0	0	2	0	0	0	0	0	2	0	0	**9**
Foo (1983)[27]	1	0	0	1	0	0	2	1	0	0	0	0	2	0	0	**7**
Goodman (1975)[26]	1	0	0	2	0	0	2	0	0	0	0	0	1	0	0	**6**
Stein (2006)[34]	1	0	0	2	0	0	1	0	0	0	0	0	0	0	0	**4**

Points should be given to every criterion. *Indicates the mandatory criteria and these criteria are darker colored.
0=Did not fulfil the criteria; 1=Unclear if criteria is fulfilled; 2=Did fulfil the criteria.
ICROMS = Integrated Quality Criteria for Review of Multiple Study Designs.
The green highlighted study has an ICROMS score ≥22, and fulfils the mandatory criteria, and was therefore classified as low RoB/high quality. The grey highlighted studies have an ICROMS ≥22 points, but do not fulfil the mandatory criteria, and were therefore classified as moderate RoB/moderate quality. The red highlighted studies have an ICROMS <22 points, and do not fulfil the mandatory criteria, and were therefore classified as high RoB/low quality.

Table 2. Study characteristics and outcomes.

Conservative (Serratus anterior palsy)

Study (year)	Shoulders (n)	Mean age in years (range)	%Female	Mean follow-up in months (range)	Etiology	% EMG-proven	% Previous surgery
Goodman (1975)	12	NR (5-55)*	58	54 (12-84)	Tra: 3 Iat: 2 Idi: 3 Oth: 4	67	NR
Foo (1983)	20	38 (18-70)	60	62 (6-144)	Tra: 3 Oth: 17	NR	NR
Kaupilla (1996)	26	37 (16-71)*	NR	72 (24-132)	Iat: 26	100	NR
Pikkarainen (2012)	37	34 (12-54)*	43	204 (24-360)	Tra: 12 Oth:25	100	NR
Vastamaki (2015)	55	30 (15-52)*	24	264 (120-336)	Tra: 46 Idi:8 Oth:1	100	NR
Ng (2021)	84	38 (15-77)*	35**	NR	Tra: 33 Iat: 2 Oth: 2 NAM: 47	100	NR

Surgical (Serratus anterior palsy)

Study (year)	Shoulders (n)	Mean age in years (range)	% Female	Mean follow-up in months (range)	Etiology	% EMG-proven	% Previous surgery
Connor (1997)	11	34 (20-52)	64	27 (12-60)	Tra: 9 Iat: 1 Idi: 1	91	64
Warner (1998)	8	32 (24-43)	50	40 (12-86)	Tra: 8	63	63
Perlmutter (1999)	16	33 (20-55)	56	40 (16-132)	Tra: 7 Iat: 7 Idi: 2	100	56
Noerdlinger (2002)	15	33 (17-44)*	40	NR	Tra: 12 Iat: 1 Idi: 2	73	53
Steinmann (2003)	9	34 (21-47)	44	34 (24-60)	Tra: 7 Iat: 2	100	22

Intervention	Co-intervention (%)	Mean time to recovery in months (range)	Reported outcome measures	%Persistent winging	Complications
No specific intervention	Physical therapy (83)	NR	-	75	NA
No specific intervention	Physical therapy (25)	13 (6-24)	-	25	NA
No specific intervention	Physical therapy (50) Bracing (65)	NR	-	73	NA
No specific intervention	Physical therapy (46)	16 (2-30)	RoM	22	NA
Bracing	Physical therapy (44)	NR	RoM	31	NA
No specific intervention	NR	Tra cohort (n=33): 25 (6-48) NAM cohort (n=47): 16 (3-36)	-	Tra cohort: 79 NAM cohort: 53	NA

Surgical technique PM transfer (n)	Graft (type)	Mean time to surgery in months	Reported outcome measures	%Persistent winging	Complications (n)
Indirect	Fascia lata (autograft)	27	RoM VAS ASES	36	-
Indirect	Semitendinosus and gracilis (autograft)	32	RoM	0	Infection requiring debridement and removal of graft (1)
Indirect	Fascia lata (autograft)	40	RoM CS	25	Transfer rupture (2)
Indirect	Fascia lata (autograft)	NR	RoM ASSES	60	Frozen shoulder (2) Muscle bulging (1)
Indirect	Fascia lata (autograft)	70	RoM ASES	33	Frozen shoulder (2) Seroma (1)

Table 2. Study characteristics and outcomes. (continued)

Surgical (Serratus anterior palsy)

Study (year)	Shoulders (n)	Mean age in years (range)	% Female	Mean follow-up in months (range)	Etiology	% EMG-proven	% Previous surgery
Stein (2006)	10	NR	NR	NR	NR	100	NR
Galano (2008)	11	34 (18-48)	82	72 (24-240)	Tra: 6 Iat: 2 Idi: 2 Oth: 2	91	27
Tauber (2008)	12	42 (27-75)	42	24 (18-56)	Tra: 5 Iat: 6 Idi: 1	100	NR
Streit (2012)	26	33 (15-53)*	57	58 (12-120)	Tra: 6 Idi: 20	100	38
Chalmers (2015)	24	30 (NR)	63	29 (NR)	Tra: 18 Iat: 2 Idi: 4	71	46
Elhassan (2015)	51	31 (14-65)*	45	NR	NR	58	NR
Li (2017)	28	38 (22-56)	46	21 (14-30)	Tra: 15 Iat: 5 Idi: 8	100	NR

Surgical (Trapezius palsy)

Study (year)	Shoulders (n)	Mean age in years (range)	% Female	Mean follow-up in months (range)	Etiology	% EMG-proven	% Previous surgery
Bigliani (1996)	22	32 (8-74)*	73	90 (24-168)	Tra: 7 Iat: 15	Unclear	32
Romero (2003)	12	25 (11-43)*	75	408 (348-456)	Tra: 1 Iat: 14 Oth: 1	33	67
Teboul (2004)	7	39 (25-65)	NR	29 (14-54)	Iat: 5 Idi: 2	NR	NR

Surgical technique PM transfer (n)	Graft (type)	Mean time to surgery in months	Reported outcome measures	%Persistent winging	Complications (n)
Direct	NA	NR	RoM	10	Transfer rupture (1)
Direct	NA	72	RoM VAS ASES	0	Infection (1) Transfer rupture (1)
Direct	NA	93	RoM VAS CS	8	Neurologic (1) Transfer rupture (1)
Direct (n=4) Indirect (n=22)	Semitendinosus (autograft)	22	RoM VAS ASES	19	Hematoma (1) Neurologic (4)
Direct (n=10) Indirect (n=14)	Direct: Achilles (allograft) Indirect: Tibialis anterior tendon (allograft)	29	RoM VAS ASES SST	8	Infection (2) Persistent pain (1) Frozen shoulder (1)
Direct	NA	NR	RoM CS SSV DASH	12	Infection (2) Hematoma (5) Persistent pain (3) Frozen shoulder (3) Neurologic (3) Transfer rupture (1)
Indirect	Semitendinosus (autograft)	21	RoM VAS ASES	0	Frozen shoulder (4) Seroma (1)

Intervention	Version	Mean time to surgery in months	Reported outcome measures	%Persistent winging	Complications (n)
Eden-Lange	Modified	34	ASES (pain)	27	Neurologic (1)
Eden-Lange	-	NR	CS	NR	None
Eden-Lange	-	28	RoM	NR	NR

Table 2. Study characteristics and outcomes. (continued)

Surgical (Trapezius palsy)							
Study (year)	**Shoulders (n)**	**Mean age in years (range)**	**% Female**	**Mean follow-up in months (range)**	**Etiology**	**% EMG-proven**	**% Previous surgery**
Galano (2008)	6	40 (18-54)	50	47 (11-89)	Tra: 3 Iat: 3	100	17
Elhassan (2015)	22	NR	41	35 (13-26)	Tra: 8 Iat: 11 Oth: 3	NR	NR
Amroodi (2018)	11	41 (25-59)	27	34 (24-48)	Tra: 2 Iat: 9	100	NR

*Some cases were under 18 years of age and therefore did not meet the inclusion criteria. **Average mean from larger cohort including patients with scapular winging due to myopathy. Follow-up of conservative treatment: starting from first presentation in hospital; Follow-up of surgical treatment: starting from day of surgery. Abbreviations: EMG-proven; diagnosis confirmed with electromyography; NR = not reported; NA = not applicable; Tra= traumatic; Iat= Iatrogenic; Idi = Idiopathic; Oth = other, NAM = neuralgic amyotrophy, RoM = Range of motion; WORC= Western Ontario Rotator Cuff Index; VAS = Visual analogue scale for pain; ASES = American shoulder and elbow surgeons score; SSV = Subjective shoulder value; CS = Constant score; DASH = Disability of the arm, shoulder and hand questionnaire.

	Intervention	Version	Mean time to surgery in months	Reported outcome measures	%Persistent winging	Complications (n)
	Eden-Lange	Modified	54	RoM VAS ASES	0	Infection (2)
	Eden-Lange	Modified	48	RoM CS SSV DASH	5	Transfer rupture (2)
	Eden-Lange	-	10	RoM VAS ASES	NR	NR

Figure 2 | Forest plots.

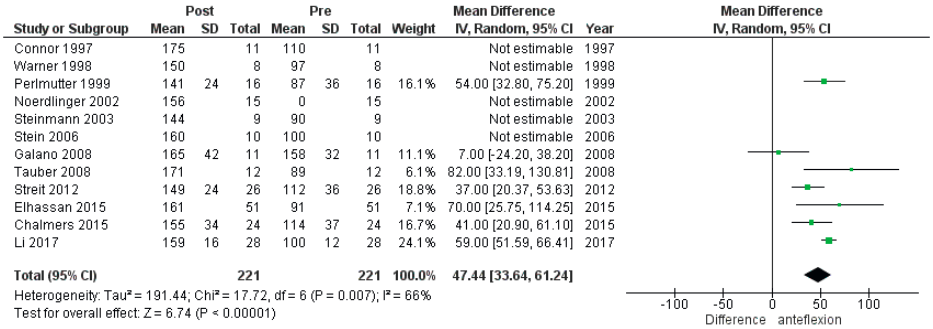

Figure 2a. Improvement in forward flexion after pectoralis major transfer for serratus anterior palsy.

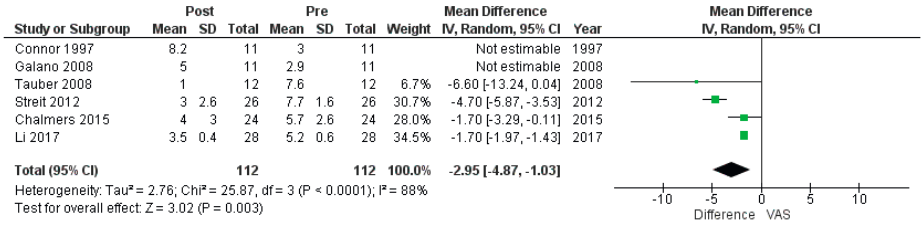

Figure 2b. Improvement in VAS score for pain after pectoralis major transfer for serratus anterior palsy.

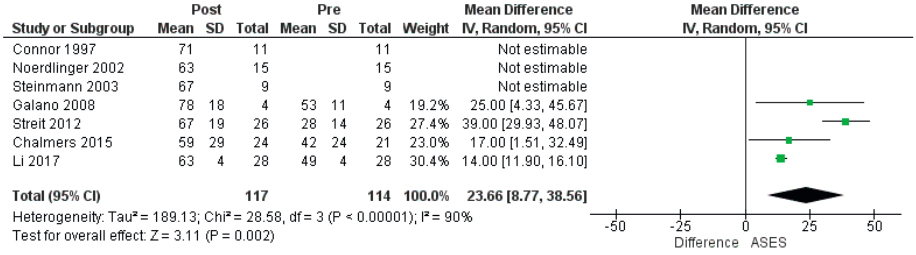

Figure 2c. Improvement in ASES score after pectoralis major transfer for serratus anterior palsy.

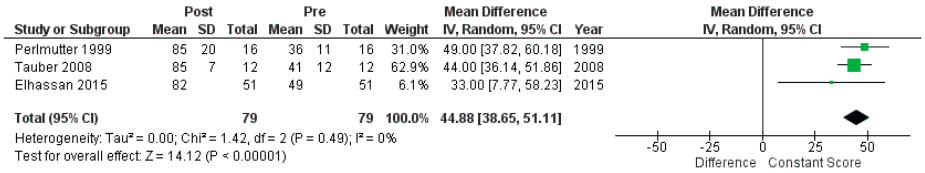

Figure 2d. Improvement in Constant score after pectoralis major transfer for serratus anterior palsy.

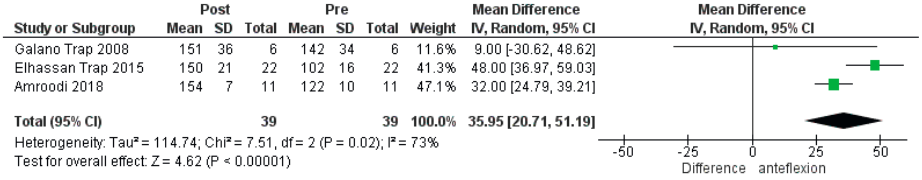

Figure 2e. Improvement in forward flexion after the Eden-lange procedure for trapezius palsy.

DISCUSSION

The present review found that only few studies reported on nonsurgical management for scapular winging due to SA palsy. In most of these nonsurgical management studies, there was no specific intervention, but only observation of the natural course. Scapular winging, functional limitations and pain persisted in a substantial percentage of these patients, which is indicative that recovery of scapular winging due to SA palsy after nonsurgical management is often only partial. The latter may also reflect that patients included in these studies may have had a more-severe clinical presentation, but data on background characteristics were often not presented within studies thus this remains unclear. No study reported on nonsurgical management for scapular winging due to TP palsy. For tendon transfer surgery, significant improvements in function, pain- and shoulder scores were shown for both SA- and TP palsy suggesting that a tendon transfer is a viable option for patients not recovering after initial nonsurgical management. However, substantial heterogeneity in reported outcomes was found which can likely (at least in part) be explained by the variety in diagnostic criteria of scapular winging and difference in quality between studies. Therefore, the overall effect estimates should be interpreted with caution, when discussing this with patients in clinical practice. Nevertheless, this review compiles the best available data on this low prevalent entity of scapular winging due to SA- or TP palsy.

This study had some limitations. First, as with any systematic review, the inherent weaknesses of individual studies translate into limitations of this review. In this systematic review, only noncontrolled case-series were included. Noncontrolled studies are prone to potential bias, although they can still offer useful information about the effectiveness of an intervention

aiming to improve patient safety if the risk of bias is low. None of the studies met the mandatory criteria of the ICROMS tool for noncontrolled studies, whereas only ten studies (43%) met the minimum score of 22 points and mostly concerned the more recent studies. All included studies were therefore labeled as MQ at best. Secondly, a clear description of the diagnostic criteria for scapular wining due to SA- or TP palsy was often lacking within studies and not all studies confirmed the diagnosis by electromyography and not in all of the patients. Electromyography, however, is considered of crucial importance to confirm the diagnosis of scapular winging originating from neurologic abnormalities[34]. Therefore, the diagnostic accuracy of scapular winging due to SA- or TP palsy may be questioned in several studies and it is possible that some of the included patients might have been misdiagnosed as scapular winging may have been secondary to other causes than SA- or TP palsy[10]. Furthermore, scapular winging is only a symptom and does not specify the nature of the disorder that has a large variety in etiology (e.g. traumatic, inflammatory, iatrogenic, myopathic). Both the potentially misdiagnosed patients and the variety in etiologic factors may be (partially) responsible for the heterogeneity in the overall effect estimates. Third, it is possible that publication bias contributed to the relative paucity of studies investigating the outcomes of nonsurgical management in comparison with studies evaluating the outcomes of surgical treatment. Also, we did not evaluate the effect of other surgical procedures, such as nerve surgery and scapulothoracic fusion as this was beyond the scope of this review. Lastly, although we used a systematic methodology, it is possible that different search terms would have resulted in additional studies meeting our inclusion criteria. On the other hand, no additional records were added by checking references of included studies, suggesting the search strategy has comprehensively captured all studies.

To our knowledge this is the first systematic review that summarized the existing evidence on outcomes after both nonsurgical and surgical treatment for scapular winging caused by SA- or TP palsy. Only Elsawi et al. performed a systematic review on the clinical outcomes after surgical treatment (i.e. nerve surgery or Eden-Lange procedure) of TP palsy[46]. However, three studies reporting on the outcomes of the Eden-Lange procedure for TP palsy were added by the present study, thereby giving a more complete overview [38, 41, 42]. Chalmers et al. performed a systematic review on the clinical outcomes of PM transfer for SA palsy, but only compared direct with indirect PM transfer and did not assess the risk of bias of the included studies[37]. The present systematic review therefore adds to the literature by providing a more complete overview on the overall evidence available of both nonsurgical management and tendon transfer surgery for scapular winging, while taking the quality of studies into account, which can be used to inform patients during a shared decision making process with their physician in clinical practice.

In literature, there is general consensus that scapular winging should be treated nonsurgically for at least 24 months, consistent with the average time to recover reported by three studies in the present review[8, 13, 27]. A variety of nonsurgical intervention options (e.g. physical therapy, bracing) have been suggested to relieve symptoms and maintain shoulder function,

but no therapy is universally accepted as being effective[6, 14]. Only one study investigated the efficacy of a specific physical therapy program and reported a significant improvement in the Western Ontario Rotator Cuff score after treatment, but did not report on other clinical outcome scores (e.g. persistent winging, function or pain) and did not have a control group[47]. Other studies investigating bracing therapy have reported conflicting results as well as low patient compliance[25, 27, 30, 48]. Some authors advice to start corticosteroid treatment to alleviate symptoms and stimulate recovery in case of scapular winging secondary to neuralgic amyotrophy[49]. Despite nonsurgical management being the first treatment option, surprisingly few studies were identified reporting on its outcomes. Only two of these studies reported on objective clinical outcome measures (i.e. range of motion), but not on pain- and/or shoulder scores. Consequently, the optimal nonsurgical management strategy of scapular winging remains unclear and many interventions are applied without clear evidence.

Although it must be noted that most included nonsurgical management studies only described the natural cause of scapular winging without a specific intervention, the present study showed that a substantial part of nonsurgically managed patients with scapular winging keep experiencing winging, functional limitations and/or pain in the long run. Therefore, it is possible that some patients with scapular winging would benefit from surgical treatment at an earlier stage. Unfortunately, this population cannot yet be identified as little is known about prognostic factors or the role of etiology that may predict sufficient spontaneous recovery of scapular winging[24]. Initial electromyographic examination does not seem to predict clinical outcomes[23, 50]. Only few studies investigated the influence of SA palsy etiology on the outcomes of spontaneous recovery and suggested that a traumatic or iatrogenic etiology carry a poorer prognosis in treatment outcomes, whereas palsies caused by infection recovered better[24, 26, 50]. Future studies are warranted to identify prognostic variables that may help predict (non) recovery after nonsurgical management and thus identify those patients who might benefit from surgical treatment at an earlier stage.

Treatment of scapular winging should be individualized and based on severity and chronicity of the patients' symptoms. As a variety of disorders can cause scapular winging, it is important that the correct diagnosis be established as the basis for any type of intervention[4, 51]. Nonsurgical management is always warranted in the beginning of the pathology as many patients will experience resolution without the need of surgery, as shown by this systematic review. However, because a substantial part of patients keep experiencing residual complaints, this should also be part of the information provided to patients. The same is true for tendon transfer surgery, for which the present review showed substantial heterogeneity in outcomes that can be used to inform patients so that they have reasonable expectations on surgical treatment. The provided estimated treatment effects of the best available evidence may thus guide clinicians and patients in treatment decisions, but given the overall low to moderate quality of evidence the results should be interpreted with caution.

Higher quality evidence is needed to supplement this evidence base and enable stronger recommendations. This review showed that diagnostic criteria for scapular winging varied widely between studies. In addition, large variability in reported outcome measures was present between studies that were often subjective (e.g. degree of winging) and therefore made it difficult to evaluate and compare treatment effects. Until a more precise an widespread method of classification and categorization of scapular winging is established, little meaningful information can be accumulated to help guide clinical decision-making. Future studies should establish clear criteria for the diagnosis of scapular winging and report on standardized (objective) outcomes so that data can be pooled across studies. In particular, this review shows a research gap regarding outcomes of nonsurgical management of scapular winging. Furthermore, more knowledge of prognostic factors that predict poor outcome after nonsurgical management may help to identify patients who would benefit from surgical treatment at an earlier stage. As scapular winging is a rare entity, this will likely not be feasible for a single institution or country, so that large (inter)national collaboratives are needed to further substantiate these findings and guide clinical decision-making.

CONCLUSION

This systematic review compiles the best available evidence on nonsurgical management and tendon transfer surgery for scapular winging caused by SA- and TP palsy. This review showed that scapular winging, functional limitations and pain persisted in a substantial percentage of nonsurgically managed patients, indicating that spontaneous recovery of scapular winging is often only partial. For tendon transfer surgery, significant improvements in function, pain- and shoulder scores were shown for both SA- and TP palsy suggesting that a tendon transfer is a viable option for patients not recovering after initial nonsurgical management. Higher quality evidence is needed to further substantiate these findings and further guide clinical treatment.

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Appendix S1. Search strategy

Database	Search Strategy	Number of references	Number of unique references
PubMed	(”Scapula Alata”[tw] OR ”Scapular winging”[tw] OR ”Scapula winging”[tw] OR ”Scapular wing*”[tw] OR ”Scapula wing*”[tw] OR ”winged scapula”[tw] OR ”winged scapula*”[tw] OR ”winging scapula*”[tw] OR ”winging of scapula”[tw] OR (scapula*[tw] AND (wingi*[tw] OR winge*[tw])) OR ”long thoracic nerve block”[tw] OR ”long thoracic nerve entrapment”[tw] OR ”long thoracic nerve injury”[tw] OR ”long thoracic nerve lesion”[tw] OR ”long thoracic nerve neuropathy”[tw] OR ”long thoracic nerve palsies”[tw] OR ”long thoracic nerve palsy”[tw] OR ”long thoracic neuropathy”[tw] OR ((”long thoracic nerve*”[tw]) AND (”pals*”[tw] OR ”lesion*”[tw])) OR ”serratus anterior muscle palsy”[tw] OR ”serratus anterior muscle paralysis”[tw] OR ”serratus anterior palsy”[tw] OR ”serratus anterior paralysis”[tw] OR (”serratus anterior”[tw] AND (”Paralysis”[mesh] OR ”palsy”[tw] OR ”paralysis”[tw] OR ”palsies”[tw] OR ”paraly*”[tw])) OR ”trapezius muscle palsy”[tw] OR ”trapezius muscle paralysis”[tw] OR ”trapezius palsy”[tw] OR ”trapezius paralysis”[tw] OR ”trapezial palsy”[tw] OR ”trapezial paralysis”[tw] OR ((”trapezius”[tw] OR ”trapezial”[tw] OR ”Accessory Nerve”[Mesh] OR ”accessorius”[tw] OR ”accessory nerve”[tw] OR ”accessory nerves”[tw] OR ”Eleventh Cranial Nerve”[tw] OR ”Eleventh Cranial Nerves”[tw] OR ”Nerve XI”[tw]) AND (”Paralysis”[mesh] OR ”palsy”[tw] OR ”paralysis”[tw] OR ”palsies”[tw] OR ”paraly*”[tw]))) NOT ((“Case Reports”[ptyp] OR “case report”[ti]) NOT (“Review”[ptyp] OR “review”[ti] OR “Clinical Study”[ptyp] OR “trial”[ti] OR “RCT”[ti]))	884	884
Embase (OVID version)	(”Scapula Alata”.mp OR ”Scapular winging”.mp OR ”Scapula winging”.mp OR ”Scapular wing*”.mp OR ”Scapula wing*”.mp OR ”winged scapula”.mp OR ”winged scapula*”.mp OR ”winging scapula*”.mp OR ”winging of scapula”.mp OR (scapula*.mp ADJ3 (wingi*.mp OR winge*.mp)) OR ”long thoracic nerve block”.mp OR ”long thoracic nerve entrapment”.mp OR ”long thoracic nerve injury”.mp OR ”long thoracic nerve lesion”.mp OR ”long thoracic nerve neuropathy”.mp OR ”long thoracic nerve palsies”.mp OR ”long thoracic nerve palsy”.mp OR ”long thoracic neuropathy”.mp OR ((”long thoracic nerve*”.mp) ADJ3 (”pals*”.mp OR ”lesion*”.mp)) OR ”serratus anterior muscle palsy”.mp OR ”serratus anterior muscle paralysis”.mp OR ”serratus anterior palsy”.mp OR ”serratus anterior paralysis”.mp OR (”serratus anterior”.mp AND (exp ”Paralysis”/ OR ”palsy”.mp OR ”paralysis”.mp OR ”palsies”.mp OR ”paraly*”.mp)) OR ”trapezius muscle palsy”.mp OR ”trapezius muscle paralysis”.mp OR ”trapezius palsy”.mp OR ”trapezius paralysis”.mp OR ”trapezial palsy”.mp OR ”trapezial paralysis”.mp OR ((”trapezius muscle”/ OR ”trapezius”.mp OR ”trapezial”.mp OR ”Accessory Nerve”/ OR ”accessorius”.mp OR ”accessory nerve”.mp OR ”accessory nerves”.mp OR ”Eleventh Cranial Nerve”.mp OR ”Eleventh Cranial Nerves”.mp OR ”Nerve XI”.mp) AND (exp ”Paralysis”/ OR ”palsy”.mp OR ”paralysis”.mp OR ”palsies”.mp OR ”paraly*”.mp))) NOT ((exp “Case Report”/ OR “case”.ti) NOT (“Review”/ OR “review”.ti OR “Clinical Study”/ OR exp “Clinical Trial”/ OR “trial”.ti OR “RCT”.ti))	1107 321 meeting abstracts	549 319 meeting abstracts

Web of Science	ti=(”Scapula Alata” OR ”Scapular winging” OR ”Scapula winging” OR ”Scapular wing*” OR ”Scapula wing*” OR ”winged scapula” OR ”winged scapula*” OR ”winging scapula*” OR ”winging of scapula” OR (scapula* NEAR3 (wingi* OR winge*)) OR ”long thoracic nerve block” OR ”long thoracic nerve entrapment” OR ”long thoracic nerve injury” OR ”long thoracic nerve lesion” OR ”long thoracic nerve neuropathy” OR ”long thoracic nerve palsies” OR ”long thoracic nerve palsy” OR ”long thoracic neuropathy” OR ((”long thoracic nerve*”) NEAR3 (”pals*” OR ”lesion*”)) OR ”serratus anterior muscle palsy” OR ”serratus anterior muscle paralysis” OR ”serratus anterior palsy” OR ”serratus anterior paralysis” OR (”serratus anterior” AND (”palsy” OR ”paralysis” OR ”palsies” OR ”paraly*”)) OR ”trapezius muscle palsy” OR ”trapezius muscle paralysis” OR ”trapezius palsy” OR ”trapezius paralysis” OR ”trapezial palsy” OR ”trapezial paralysis” OR ((”trapezius muscle” OR ”trapezius” OR ”trapezial” OR ”Accessory Nerve” OR ”accessorius” OR ”accessory nerve” OR ”accessory nerves” OR ”Eleventh Cranial Nerve” OR ”Eleventh Cranial Nerves” OR ”Nerve XI”) AND (”Paralysis” OR ”palsy” OR ”paralysis” OR ”palsies” OR ”paraly*”))) NOT ((TS=(“Case Report”) OR TI=(“case”)) NOT TI=(“Review” OR “review” OR “Clinical Study” OR “Clinical Trial” OR “trial” OR “RCT”))	323 42 meeting abstracts	133 33 meeting abstracts
Cochrane Library	(“Scapula Alata” OR “Scapular winging” OR “Scapula winging” OR “Scapular wing*” OR “Scapula wing*” OR “winged scapula” OR “winged scapula*” OR “winging scapula*” OR “winging of scapula” OR (scapula* NEAR/3 (wingi* OR winge*)) OR “long thoracic nerve block” OR “long thoracic nerve entrapment” OR “long thoracic nerve injury” OR “long thoracic nerve lesion” OR “long thoracic nerve neuropathy” OR “long thoracic nerve palsies” OR “long thoracic nerve palsy” OR “long thoracic neuropathy” OR ((“long thoracic nerve*”) NEAR/3 (“pals*” OR “lesion*”)) OR “serratus anterior muscle palsy” OR “serratus anterior muscle paralysis” OR “serratus anterior palsy” OR “serratus anterior paralysis” OR (“serratus anterior” AND (“palsy” OR “paralysis” OR “palsies” OR “paraly*”)) OR “trapezius muscle palsy” OR “trapezius muscle paralysis” OR “trapezius palsy” OR “trapezius paralysis” OR “trapezial palsy” OR “trapezial paralysis” OR ((“trapezius muscle” OR “trapezius” OR “trapezial” OR “Accessory Nerve” OR “accessorius” OR “accessory nerve” OR “accessory nerves” OR “Eleventh Cranial Nerve” OR “Eleventh Cranial Nerves” OR “Nerve XI”) AND (“Paralysis” OR “palsy” OR “paralysis” OR “palsies” OR “paraly*”))):ti,ab,kw	27 4 meeting abstracts	15 0 meeting abstracts
Emcare (OVID version)	(”Scapula Alata”.mp OR ”Scapular winging”.mp OR ”Scapula winging”.mp OR ”Scapular wing*”.mp OR ”Scapula wing*”.mp OR ”winged scapula”.mp OR ”winged scapula*”.mp OR ”winging scapula*”.mp OR ”winging of scapula”.mp OR (scapula*.mp ADJ3 (wingi*.mp OR winge*.mp)) OR ”long thoracic nerve block”.mp OR ”long thoracic nerve entrapment”.mp OR ”long thoracic nerve injury”.mp OR ”long thoracic nerve lesion”.mp OR ”long thoracic nerve neuropathy”.mp OR ”long thoracic nerve palsies”.mp OR ”long thoracic nerve palsy”.mp OR ”long thoracic neuropathy”.mp OR ((”long thoracic nerve*”.mp) ADJ3 (”pals*”.mp OR ”lesion*”.mp)) OR ”serratus anterior muscle palsy”.mp OR ”serratus anterior muscle paralysis”.mp OR ”serratus anterior palsy”.mp OR ”serratus anterior paralysis”.mp OR (”serratus anterior”.mp AND (exp ”Paralysis”/ OR ”palsy”.mp OR ”paralysis”.mp OR ”palsies”.mp OR ”paraly*”.mp)) OR ”trapezius muscle palsy”.mp OR ”trapezius muscle paralysis”.mp OR ”trapezius palsy”.mp OR ”trapezius paralysis”.mp OR ”trapezial palsy”.mp OR ”trapezial paralysis”.mp OR ((”trapezius muscle”/ OR ”trapezius”.mp OR ”trapezial”.mp OR ”Accessory Nerve”/ OR ”accessorius”.mp OR ”accessory nerve”.mp OR ”accessory nerves”.mp OR ”Eleventh Cranial Nerve”.mp OR ”Eleventh Cranial Nerves”.mp OR ”Nerve XI”.mp) AND (exp ”Paralysis”/ OR ”palsy”.mp OR ”paralysis”.mp OR ”palsies”.mp OR ”paraly*”.mp))) NOT ((exp “Case Report”/ OR “case”.ti) NOT (“Review”/ OR “review”.ti OR “Clinical Study”/ OR exp “Clinical Trial”/ OR “trial”.ti OR “RCT”.ti))	394	60

Academic Search Premier (EBSCOhost)	TI(”Scapula Alata” OR ”Scapular winging” OR ”Scapula winging” OR ”Scapular wing*” OR ”Scapula wing*” OR ”winged scapula” OR ”winged scapula*” OR ”winging scapula*” OR ”winging of scapula” OR (scapula* N3 (wingi* OR winge*)) OR ”long thoracic nerve block” OR ”long thoracic nerve entrapment” OR ”long thoracic nerve injury” OR ”long thoracic nerve lesion” OR ”long thoracic nerve neuropathy” OR ”long thoracic nerve palsies” OR ”long thoracic nerve palsy” OR ”long thoracic neuropathy” OR ((”long thoracic nerve*”) N3 (”pals*” OR ”lesion*”)) OR ”serratus anterior muscle palsy” OR ”serratus anterior muscle paralysis” OR ”serratus anterior palsy” OR ”serratus anterior paralysis” OR (”serratus anterior” AND (”palsy” OR ”paralysis” OR ”palsies” OR ”paraly*”)) OR ”trapezius muscle palsy” OR ”trapezius muscle paralysis” OR ”trapezius palsy” OR ”trapezius paralysis” OR ”trapezial palsy” OR ”trapezial paralysis” OR ((”trapezius muscle” OR ”trapezius” OR ”trapezial” OR ”Accessory Nerve” OR ”accessorius” OR ”accessory nerve” OR ”accessory nerves” OR ”Eleventh Cranial Nerve” OR ”Eleventh Cranial Nerves” OR ”Nerve XI”) AND (”Paralysis” OR ”palsy” OR ”paralysis” OR ”palsies” OR ”paraly*”))) NOT TI(“Case”)	129	28
Total		2864 367 meeting abstracts	1669 352 meeting abstracts

Appendix S2. Integrated quality criteria for review of multiple study designs (ICROMS) for non-controlled before after studies.

DIMENSION	SPECIFIC CRITERIA
1 Clear aims and justification	**A Clear statement of the aims of the research?**
	B Rationale for number of pre- and post-intervention points or adequate baseline measurement
	C Explanation for lack of control group
2 Managing bias in sampling or between groups	**C Justification for sample choice**
3 Managing bias in outcome measurements and blinding	E Protection against detection bias: Blinded assessment of primary outcome measures
	F Reliable primary outcome measures
4 Managing bias in follow-up	C Incomplete outcome data addressed
5 Managing bias in other study aspects	A Protection against detection bias: Intervention unlikely to affect data collection
	D Attempts to mitigate effects of no control
6 Analytical rigour	C Analysis sufficiently rigorous/free from bias
7 Managing bias in reporting/ethical considerations	A Free of selective outcome reporting
	B Limitations addressed
	C Conclusions clear and justified
	D Free of other bias
	E Ethics issues addressed

Appendix S2 Table describing the dimensions and specific criteria of the Integrated quality criteria for review of multiple study designs (ICROMS) risk of bias (RoB) assessment tool for cohort studies[18]. Seven dimensions with thirteen criteria specific for cohort studies were used in the present study. Every specific criteria can be answered with either 'Yes (2 points)', ' Unclear (1 point)' or 'No (0 points)'. This tool was designed with several mandatory criteria (highlighted bold) and with a minimum score of 22 (for non-controlled before after studies) to be included. However, the present review included all studies independent of the ICROMS score and reported the RoB for every study with the rationale that the RoB could be taken into account when weighting study results while excluding studies with high or medium RoB would result in the loss of possibly valuable information.

Appendix S3. Definitions and study outcomes of included studies

Study (year)	Shoulders (n)	Diagnostic criteria (%EMG proven)	Anteflexion		Abduction		Shoulder scores			Persistent winging	
			Pre	Post	Pre	Post	Scores	Pre	Post	Assessment method	Degree persistent winging (n)
Connor (1997)	11	- Clinical assessment (posterior periscapular pain, shoulder muscle fatigue and difficulty with arm elevation, marked scapular winging) - EMG(91%)	110	175	-	-	VAS ASES	8.2	3.0 71	-Clinical examination (Not specified)	-None (7) -Mild(3) -Full recurrence(1)
Warner (1998)	8*	- Clinical assessment (painful scapular winging characterized by prominence of inferior tip of scapula and loss of protraction of scapula during shoulder elevation, no flexion above 120°, not further specified) - EMG(53%)	97	150	-	-	-	-	-	-Clinical examination (Not specified)	-None(7)
Perlmutter (1999)	16	- Clinical assessment (marked scapular winging, loss strength of shoulder, difficulty with activities and daily living, not further specified) - EMG(100%)	87	158	-	-	CS	36	92	-Clinical examination (Not specified)	-None (12) -Mild (2) -Marked (2)
Noerdlinger (2002)	15	- Clinical assessment (prominent scapular winging, weakness of arm abduction, fatigue with overhead activities, not further specified) - EMG(73%)	-	156	-	-	ASES	-	63	-Clinical examination (Not specified)	-Negligible (6) -Mild (7) -Moderate (2)
Steinmann (2003)	9	- Clinical assessment (shoulder range of motion, pain, shoulder strength, visible protrusion scapula during elevation, not further specified) - EMG(100%)	90	144	-	-	ASES	-	67.4	-Clinical examination (Not specified)	-None(6) -Slight(3)

Appendix S3. Definitions and study outcomes of included studies (continued)

Surgical (Serratus anterior palsy)

Study (year)	Shoulders (n)	Diagnostic criteria (%EMG proven)	Anteflexion		Abduction		Shoulder scores			Persistent winging	
			Pre	Post	Pre	Post	Scores	Pre	Post	Assessment method	Degree persistent winging (n)
Galano (2008)	11	- Clinical assessment (not explicitly specified) - EMG(91%)	158	164	-	-	VAS ASES	5.0 53.3	2.9 63.8	-Clinical examination (Not specified)	-None (11)
Tauber (2008)	12	- Clinical assessment (range of motion, degree of pain, shoulder strength and presence of scapular winging, evaluated in active forward flexion against resistance at horizontal level, not further specified) - EMG(100%)	89	171*	86	161*	VAS CS	7.6 41	1* 85.4*	-Clinical examination (Not specified)	-None(11) -Slight(1)
Streit (2012)	26	- Clinical assessment (painful winging of scapula) - EMG(100%)	112 (36)	149 (24)*	-	-	VAS ASES	7.7 (1.6) 28 (13.8)	3 (2.6)* 67 (19)*	-Clinical examination (Not specified)	-None(21) -Recurrent(5)
Chalmers (2015)	24	- Clinical assessment (not explicitly specified) - EMG(71%)	114 (37)	155 (34)	-	-	*VAS* *ASES* *SST*	5.7 (2.6) 42 (24) 4.4 (3.6)	1.0 (3.0) 59 (29) 6.8 (4.3)	-Clinical examination (Not specified)	-Winging resolved(22) -Winging not resolved (2)
Li (2017)	28	- Clinical assessment (limited range of motion, weakness affected limb, scapular winging aggravated in forward flexion or on a wall in push-up motion, not further specified) - EMG(100%)	100 (12)	159 (16)*	86 (13)	154 (16)*	VAS ASES	5.2 (0.6) 48.8 (4.2)	3.5 (0.4)* 63.0 (3.8)	-Clinical examination (Not specified)	-No recurrence(28)

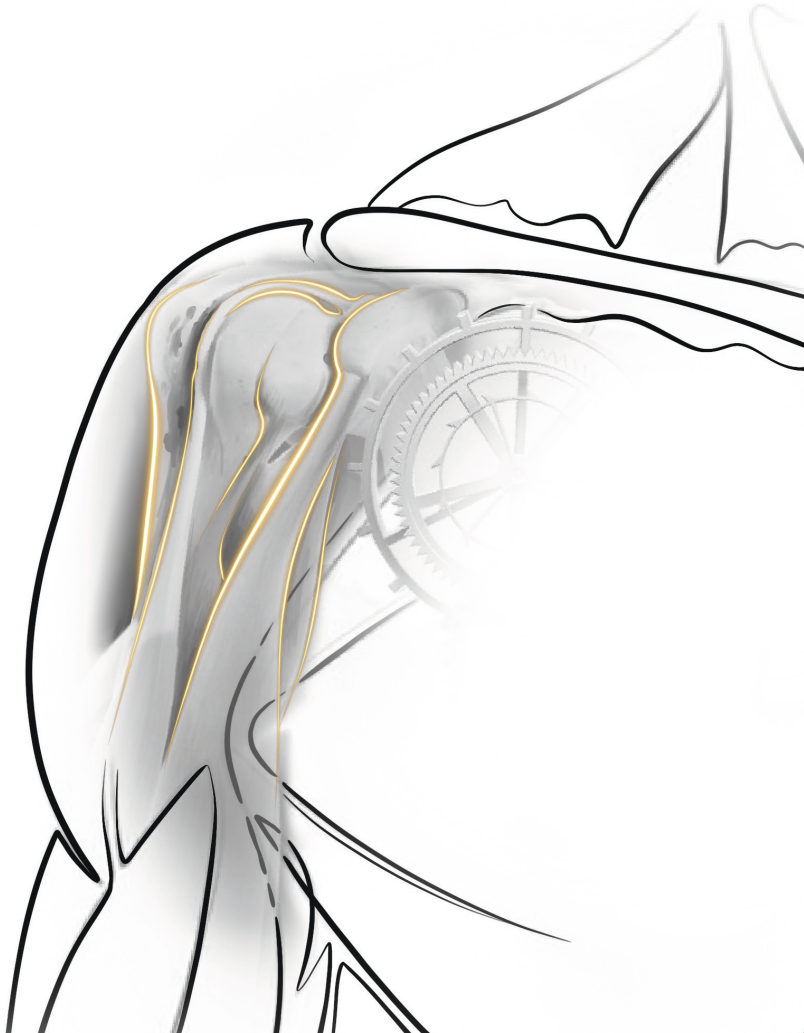
Appendix S3. Definitions and study outcomes of included studies (continued)

Surgical (Trapezius palsy)

Study (year)	Shoulders (n)	Diagnostic criteria (%EMG proven)	Anteflexion		Abduction		Shoulder scores			Persistent winging	
			Pre	Post	Pre	Post	Scores	Pre	Post	Assessment method	Degree persistent winging (n)
Bigliani (1996)	22	- Clinical assessment (not explicitly specified) - EMG (*unclear*).	-	-	-	-	ASES (pain score)	0.9	4.3	-Clinical examination (Not specified)	-None (13) -Slight (6) -Unclear (3)
Romero (2003)	12	- Clinical assessment (not explicitly specified) - EMG (33%)	-	-	-	-	CS	-	73.5 (median)	NR	NR
Teboul (2004)	7	- Clinical assessment (not explicitly specified)	-	-	-	120 (40)	-	-	-	NR	NR
Galano (2008)	6	- Clinical assessment (not explicitly specified) - EMG(100%)	142 (34)	151 (36)	-	-	VAS ASES	7.0 33.3	2.3 64.6	-Clinical examination (Not specified)	-None (6)
Amroodi (2018)	11	- Clinical assessment (obvious atrophy of the trapezius muscle, not further specified) - EMG(100%)	122 (10)	154 (7)*	80 (17)	148 (9)*	VAS ASES	7.8 (0.9) 33 (8.0)	1.6 (1.0)* 82 (5.7)*	NR	NR

All the values indicate the mean with standard deviation (SD) unless state otherwise; ^{0} = Degrees, EMG= Electromyography, Pre= Pre-operative, Post= Post-operative, AF = Anteflexion in degrees; Abd = Abduction in degrees; * = significant ($p<0.05$); VAS = Visual Analogue Scale for pain; ASES = American Shoulder Elbow Score; SST = Simple shoulder test; CS = Constant Score; SSV = Subjective Shoulder Value; DASH = Disability of the Arm, Shoulder and Hand Score, NR= Not reported.

Conservative (Serratus anterior palsy)			
Study (year)	**Diagnostic criteria (%EMG proven)**	**Persistent winging**	
		Assessment method	**Degree persistent winging (n)**
Goodman (1975)[26]	- Clinical assessment (pain, fatigue, limitations of active motion of shoulder, winging at rest, with elevation and against resistance, muscle strength, not further specified) - EMG (67%)	- Clinical examination (Not specified)	- None (3) - Persistent (9)
Foo (1983)[27]	- Clinical assessment (not explicitly specified)	- Clinical examination (Not specified)	- None (15) - Slight (5)
Kaupilla (1996)[28]	- Clinical assessment (not explicitly specified) - EMG (100%)	- Clinical examination (Not specified)	- None or minimal (7) - Partial (12) (during muscle test; pushing against wall) - Winging in rest (7)
Pikkarainen (2012)[13]	- Clinical assessment (winging ≤3cm, small limitation in ROM (flexion ≥125^{0})). - EMG (100%)	- Clinical examination (posterior displacement of medial border of scapula from posterior thorax in centimeters at, 90^{o} unresisted flexion)	- None (29) - Mild (1) - Moderate (4) - Severe (3)
Vastamaki (2015)[25]	- Clinical assessment(winging >3cm) - EMG (100%)	- Clinical examination (posterior displacement of medial border of scapula from posterior thorax in centimeters at, 90^{o} unresisted flexion)	- None (38) - Persistent (17), on average 3.4 centimeter
Ng (2021)[8]	- Clinical assessment (onset of scapular winging, history of pain, subsequent clinical progression, resultant disability, scapular winging was graded by the scapular winging severity score, not further specified) - EMG (100%) - Trauma cohort: EMG + clear trauma episode before winging - Neuralgic amyotrophy cohort: EMG + spontaneous winging after physical exertion or viral illness	- Clinical examination (not specified)	- Persistent winging traumatic cohort (26) - Persistent winging neuralgic amyotrophy cohort (25)



Chapter 3

Long-term outcomes of pectoralis major transfer for scapular winging due to long thoracic nerve palsy: results after a median follow-up of 17 years.

Timon H. Geurkink[1], Perla J. Marang-van de Mheen[1 2], Tim Robbers[1], Jochem Nagels[1], Rob G.H.H. Nelissen[1]

1. *Department of Orthopaedics, Leiden University Medical Centre, Postbus 9600, 2300 RC, Leiden, the Netherlands.*
2. *Safety & Security Science and Centre for Safety in Healthcare, Delft University of Technology, Delft, The Netherlands*

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ABSTRACT

Background:
A pectoralis major (PM) transfer is a viable treatment option for patients with scapular winging due to long thoracic nerve (LTN) palsy not responding to nonsurgical management. However the long-term outcomes remain unknown. Therefore, the purpose of this study was to evaluate the long-term outcome of shoulder function (i.e. minimum follow-up of 10 years) and quality-of-life (QoL) of patients treated for scapular winging due to LTN palsy with a PM transfer.

Methods:
This observational cohort study included 15 Patients (16 shoulders) who underwent PM transfer, using an tendoachilles allograft, between 1995 and 2012. Shoulder forward flexion and abduction were analyzed preoperatively, one year after surgery and at the final follow-up. SF-36 component scores (physical component summary (PCS) and mental component summary (MCS)) were used to evaluate QoL.

Results:
Shoulder forward flexion and abduction measured in degrees improved from 86 (SD 14.5) and 82 (SD 33.8) preoperatively to 140 (SD 27.3) and 138 (31.3) one year postoperatively. After a median follow-up of 17 years, mean shoulder functions were slightly lower than at one year postoperatively but still better than preoperative function, i.e. forward flexion 121 (SD 41.9) and abduction 122 (SD 44.5). The mean PCS score at the final follow-up was 41.9 (SD 9.7) and the mean MCS score was 49.9 (SD 12.5). Better shoulder function at final follow-up was significantly associated with higher QoL in terms of PCS scores ($p = 0.023$), but not MCS scores ($p = 0.287$)

Conclusion:
The results of the present study indicate that PM transfer augmented with an achilles tendon allograft for scapular winging due to LTN palsy leads to functional improvements that persist in long term. These functional improvements likely translate to better QoL based on their association.

INTRODUCTION

Scapular winging is a rare condition of the shoulder girdle, characterized by the abnormal protrusion of the scapula relative to the thorax during forward flexion of the arm[1]. This abnormal scapular movement pattern frequently results from long thoracic nerve (LTN) palsy, resulting in weakness of the serratus anterior (SA) muscle with subsequent inability to fixate the scapula to the thorax during shoulder motion[2-5]. In clinical practice, patients report limited forward flexion due to the inability to fixate the scapula against the thorax, with subsequent overuse or overcompensation of other shoulder girdle muscles, resulting in more rapid muscle fatigue and pain[5-8].

The majority of patients with scapular winging due to LTN palsy have complete functional recovery within two years with nonsurgical management[9]. Therefore, patients with LTN palsy should be treated nonsurgically (e.g. prevention of overuse, physical therapy) for at least this period[10]. In case of insufficient functional recovery at 2 years, surgical intervention through a pectoralis major (PM) tendon transfer can be considered. PM transfer surgery aims to restore scapulothoracic stabilization by transferring the PM muscle to the scapula as a substitute for the loss in SA muscle function, thereby restoring shoulder function[11].

A recent systematic review with a mean follow-up of three years, showed that patients with scapular winging due to LTN palsy significantly improved in function and pain after PM transfer[12]. These short-term outcomes indicate that PM transfer is a viable option for patients not responding to nonsurgical management. However, data on long-term outcomes and their impact on quality-of-life (QoL) are lacking, even though this is important information for patient counselling and expectation management. The present study aimed to report the long-term improvement in shoulder function, i.e. range of motion(after a minimum follow-up of 10 years) after PM transfer in patients with scapular winging due to LTN palsy and gauge the impact on QoL by examining the association of QoL with function measures.

MATERIALS AND METHODS

Study design and participants

This was a retrospective cohort study of all consecutive patients with scapular winging due to LTN palsy who underwent PM transfer surgery at the department of Orthopaedics, Leiden University Medical Center (LUMC, The Netherlands) between January 1995 and December 2012 to ensure a minimal follow-up duration of ten years. Ethical board approval was not deemed necessary by the local medical ethical research committee (G20.123) due to the nature of the research and its minimal risk to participants. All patients over 18 years of age who underwent PM transfer for LTN palsy proven by EMG were eligible for inclusion. The senior author (RN) initially diagnosed the condition through clinical examination, involving visual inspection for winging during forward flexion or while pushing against a wall in a push-

up position. The diagnosis was subsequently confirmed by EMG. Exclusion criteria were previous surgery to the affected shoulder, language barrier and lack of informed consent.

Out of 17 eligible patients (18 shoulders, 1 patient had bilateral pathology), in total 16 patients (17 shoulders) were included in the study (Figure 1). All of these patients had limited range of motion (ROM) of the affected shoulder joint and had no recovery after nonsurgical management (i.e. physical therapy aimed at restoring shoulder function) for more than 24 months. All surgeries were performed by a single surgeon (RN). 1 Patient was lost to follow-up due to death, leaving 15 patients (16 shoulders) available for analysis (Figure 1).

Surgical procedure

During surgery, patients were positioned in a semi-lateral decubital position. Surgical exposure was performed by an anterior (deltopectoral) approach, and a posterior approach to the angulus inferior of the scapula. Through the deltopectoral approach, the three parts of the PM muscle were identified: the clavicular, the sternocostal and the abdominal part. The sternocostal part was released from the humerus, carefully identifying the pectoral nerves in the interval between the clavicular and sternocostal parts. Then the second incision was made at the level of the angulus inferior (about 4-5 cm long). A muscle split was done of the latissimus dorsi overlaying the angulus inferior of the scapula, identifying the bony lateral edge of the angulus inferior of the scapula, subperiostal stripping of the atrophied SA muscle and proximal part of the subscapular muscle with cautery, exposing the bony part of the angulus inferior. A 6 mm drill hole was made, which is slightly enlarged with a bone nibbler to accommodate the tunneling of the achilles tendon allograft. The allograft was first weaved with its wide proximal part around the the sternocostal part of the PM, such to create a tube of pectoral muscle and allograft. Next, a tunnel was made from the deltopectoral window to the posterior thorax to the level of the angulus inferior. The sternocostal PM-allograft construct was passed from anterior to posterior to the angulus inferior of the scapula. This construct was pulled from ventral to dorsal through the bony hole, with the long sutures back to the deltopectoral surgical wound, thus tensioning the pectoral muscle fixation to the scapula. The allograft was fixed onto itself using non-resorbable sutures just proximal from the bony hole in the scapula.

Data collection and outcome measures

Individual patient records were independently evaluated by two of the authors (TG, TR) to extract patient demographics at the time of surgery and clinical data (i.e. ROM of the shoulder) both preoperatively and one year after surgery. All patients were contacted by e-mail or telephone and invited to return for long-term (final) follow-up examinations of their shoulder function at the Laboratory for Kinematics and Neuromechanics (LUMC, the Netherlands) between January and July 2022. Two independent researchers (TG, TR), who were not involved in their surgery, conducted a comprehensive assessment of long-term outcomes through the following measurements: shoulder ROM (i.e. shoulder forward flexion and -abduction measured in degrees), VAS scores for pain (VAS no pain: 0), Constant

Shoulder (CS) Score, Disability of the Arm, Shoulder and Hand (DASH) Questionnaire, the SF-36 Health Survey Questionnaire to assess the health-related QoL.

Statistical analysis

Data were described using descriptive statistics, using means, standard deviation (SD) and 95% confidence intervals (CI) for parametric data and medians and interquartile range (IQR) for non-parametric data, unless stated otherwise. Numbers and percentages were used for categorical data. Shoulder function (i.e. active ROM) over time (preoperatively, 1 year after surgery and at final follow-up) was analyzed using repeated-measures ANOVA to measure within subject effects. Summary scores of the SF-36 (Mental Component Summary (MCS) and Physical Component Summary (PCS)) were calculated by standardizing the SF-36 scores against Dutch population means, standard deviations, and factor coefficients (i.e. a MCS and PCS score of 50 corresponds to the mean score for the general Dutch population)[13]. SF-36 scores from our study were compared with US- and Dutch population norm data, and with the mean scores of five other common shoulder pathologies (i.e. anterior glenohumeral instability, complete reparable rotator cuff tear, adhesive capsulitis, glenohumeral osteoarthritis, and impingement syndrome)[14, 15]. The association between the QoL summary scores (i.e. MCS and PCS) and shoulder function (i.e. active shoulder forward flexion at last follow-up) at final follow-up was analyzed with linear regression analysis. All statistical analyses were performed using the statistical package SPSS version 25.0 (IBM, Armonk, NY, USA).

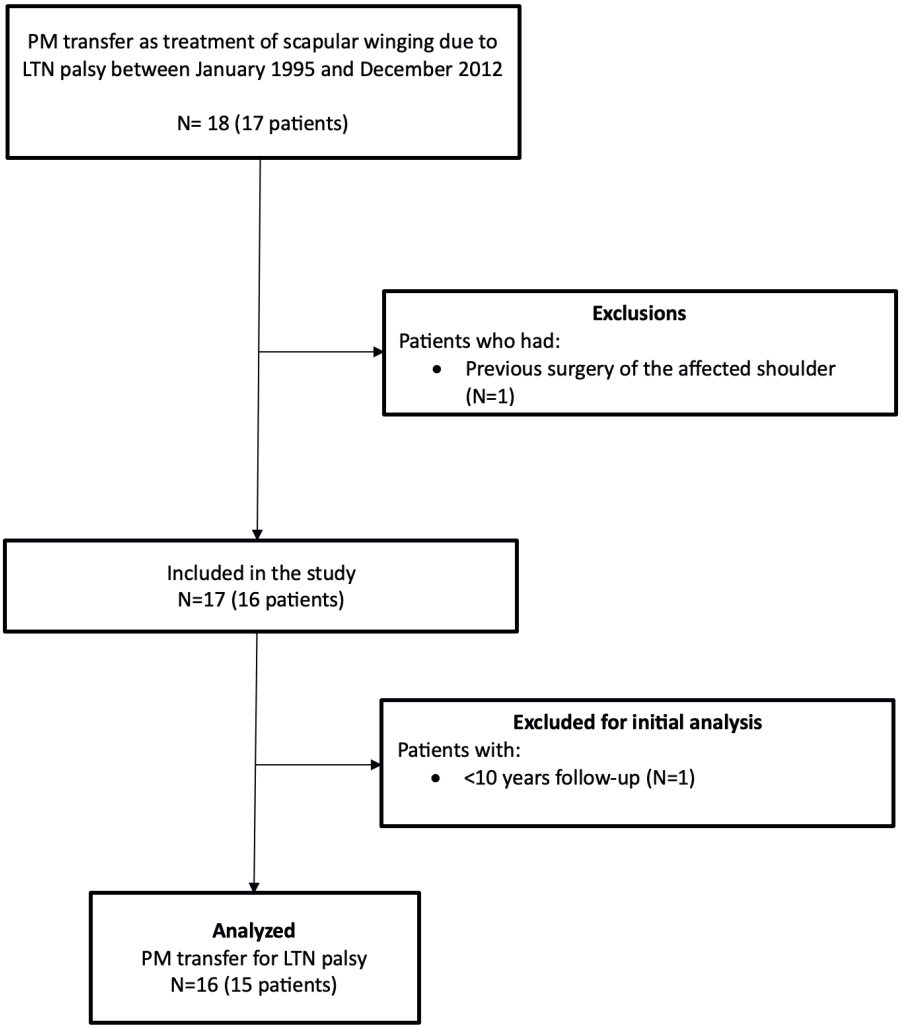

Figure 1. Flow chart of study selection. PM, Pectoralis Major; LTN, Long Thoracic Nerve; N, number of shoulders.

RESULTS

Patients had a median age of 33 years (range 18-50) at the time of surgery and ten (63%) were female. Six patients had a traumatic origin of their LTN palsy, the cause was idiopathic in five patients (including the patient with bilateral winging), and four were iatrogenic (i.e. mastectomy, axillary dissection, first rib resection (2)). The dominant arm was affected in nine (56%) patients. The median time to final follow-up was 17 years (range 10-25).

At final follow-up, patients reported a median VAS pain score of 2.0 (IQR 0.7-5.4) at rest and 2.9 (IQR 1.1-5.9) during movement. The median Constant Score of the affected arm was 68 (IQR 42-91) compared with 89 (IQR 80-97) of the unaffected arm. The median DASH score was 30 (IQR 15-38). Residual scapular winging was observed in five patients (33%) at final follow-up. Twelve patients (80%) would recommend the surgical procedure to friends and all patients were happy with the cosmesis. No postoperative complications were reported. None of the patients underwent any other shoulder surgery following the PM transfer.

Functional outcome scores of individual patients are presented in Table 1. Forward flexion improved from 86 (SD 14.5) preoperatively to 140 (SD 27.3) 1-year postoperatively, while abduction increased from 82 (SD 33.8) to 138 (31.3) respectively (Figure 2). After a median follow-up of 17 years, mean shoulder functions were slightly lower than at 1 year but still better compared to preoperative function, i.e. forward flexion 121 (SD 41.9) and abduction 122 (SD 44.5) respectively (Figure 2). For both shoulder forward flexion and abduction, repeated measures ANOVA showed a significant effect of treatment over time ($P<0.001$). Three patients (patients 5,6 and 14 in Table 1) had deterioration of their good one-year postoperative result at final follow-up, which was even worse than preoperative forward flexion.

SF-36 scores at final follow-up are shown in Table 2. The Role-Physical domain score was the lowest (mean: 48.4; SD 41.3) and the Mental Health domain had the highest score (mean 78.5; SD 17.5). The mean PCS score (41.9; SD 9.7) was considerably lower than US- and Dutch general population norm data, but higher than the mean PCS scores of other common shoulder pathologies (Table 2). The mean MCS score (49.9; SD 12.5) was comparable to general population means and MCS scores of other shoulder pathologies. Better shoulder forward flexion function at final follow-up was significantly associated with higher QoL in terms of PCS scores (Beta = 0.131, p = 0.023), but not MCS scores (Beta = 0.085, p = 0.287).

Table 1. Functional outcome scores of individual patients.

Patient	*Age at time of surgery(years)*	*Follow-up time (years)*	*Forward flexion*			*Abduction*			*Residual winging at final follow-up*
			Pre-OK	*1 year*	*Last Follow-up*	*pre-OK*	*1year*	*Last Follow-up*	
1	31	16	70	160	170	110	160	170	No
2	37	25	100	180	180	90	180	180	No
3	26	24	80	110	100	75	125	100	Yes
4	40	22	90	160	130	100	170	170	Yes
5*	18	22	90	160	80	60	160	90	No
6	26	20	70	105	50	50	100	40	Yes
7	35	19	50	150	60	60	130	90	Yes
8	23	20	90	170	150	60	170	130	No
9*	22	18	80	100	100	60	100	100	No
10	26	10	95	100	100	60	80	80	Yes
11	38	16	100	140	140	110	130	140	No
12	50	14	70	130	160	60	150	160	No
13	21	14	95	140	120	50	140	130	No
14	46	13	90	120	70	70	100	50	No
15	34	13	100	170	170	170	170	170	No
16	38	12	100	140	150	130	130	150	No
Mean (SD)			86 (14.5)	140 (27.3)	121 (41.9)	82 (33.8)	138 (31.3)	122 (44.5)	

SD, standard deviation.

* = patient with bilateral winging.

Table 2. SF-36 scores from the present study in comparison to US- and Dutch general population norms and five common shoulder pathologies.
SD, standard deviation; PM, pectoralis major

Scales/Summaries	US general population norms (SD)	Dutch general population norms (SD)	Anterior glenohumeral instability (SD)	Complete reparable rotator cuff tear (SD)	Adhesive capsulitis (SD)	Glenohumeral osteoarthritis (SD)	Impingement syndrome (SD)	Scapular winging (after PM transfer) (SD)
Physical Functioning	84.2 (23.3)	83.0 (22.8)	71.3 (21.6)	56.9 (25.8)	67.2 (22.4)	57.5 (33.6)	62.9 (23.2)	68.7 (22.5)
Role-Physical	81.0 (34.0)	76.4 (36.3)	24.7 (36.4)	26.8 (39.3)	34.5 (37.0)	41.0 (45.4)	29.8 (38.1)	48.4 (41.3)
Bodily Pain	75.2 (23.7)	74.9 (23.4)	36.8 (22.8)	29.9 (19.3)	37.6 (20.6)	36.6 (22.7)	35.0 (16.5)	63.9 (30.0)
General Health	72.0 (20.3)	70.7 (20.7)	72.0 (20.6)	67.9 (24.7)	70.2 (25.1)	71.7 (20.2)	70.4 (21.2)	62.2 (20.7)
Vitality	60.9 (21.0)	68.6 (19.3)	55.4 (21.7)	50.2 (22.2)	52.1 (24.1)	58.5 (23.8)	55.8 (23.6)	60.3 (23.9)
Social Functioning	83.3 (22.7)	84.0 (22.4)	62.2 (28.5)	57.1 (31.9)	74.1 (29.2)	72.9 (33.8)	65.0 (26.9)	70.3 (30.6)
Role-Emotional	81.3 (33.0)	82.3 (32.9)	66.2 (43.2)	58.8 (44.5)	70.9 (42.1)	69.6 (42.9)	62.6 (43.6)	60.4 (47.5)
Mental Health	74.7 (18.1)	76.8 (17.4)	68.2 (19.3)	67.2 (22.1)	72.7 (21.8)	73 (20.1)	70.1 (21.5)	78.5 (17.5)
Physical component summary	50.0	50.0	38.2 (9.0)	34.7 (9.2)	37.6 (8.8)	36.4 (11.8)	36.6 (8.3)	41.9 (9.7)
Mental component summary	50.0	50.0	48.1 (21.2)	47.2 (12.6)	51.0 (12.6)	52.2 (11.4)	49.1 (12.6)	49.9 (12.5)

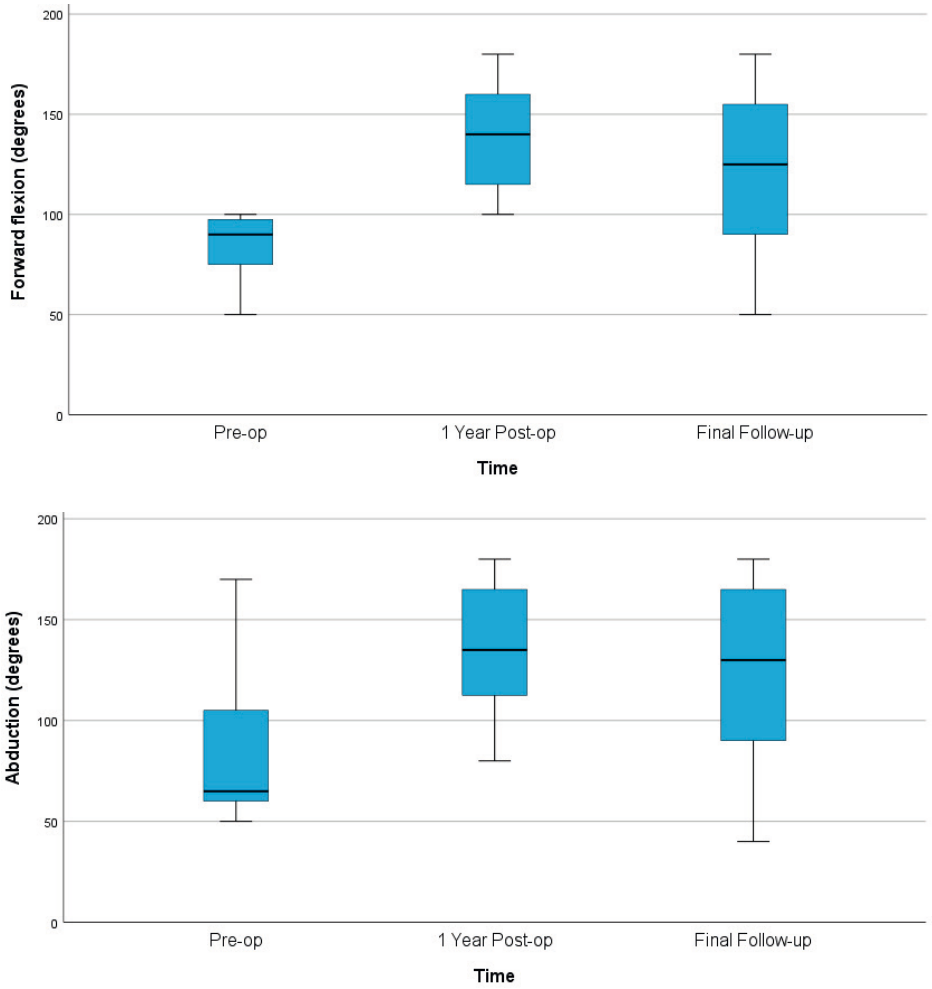

Figure 2. Boxplots showing shoulder function (i.e., forward flexion and abduction) before and after PM transfer for LTN palsy. Boundaries of the boxplots indicate the 25th and 75th percentile. Wiskers below and above indicate the minimum and maximum of the variables. The median is indicated with the black line. Pre-op, preoperative; Post-op, postoperative; PM, Pectoralis Major; LTN, Long Thoracic Nerve.

DISCUSSION

This is the first study to describe long-term improvement in shoulder function in a cohort of patients with LTN palsy treated with PM transfer. Results of the present study demonstrate that the shoulder function of patients with scapular winging due to LTN palsy that were unresponsive to nonsurgical management substantially improved after PM transfer and that the functional improvements were still present in the majority of patients after a median follow-up of 17 years. These functional improvements likely translate into improvement in QoL as better shoulder function was associated with better PCS scores. Another new finding of this study is that, despite significant improvements in shoulder function after PM transfer, scapular winging still impacts the long-term QoL. On average, lower QoL was observed our cohort than for the general Dutch population, indicating that these patients still experience greater limitations in physical functional and role participation, but scores in the mental domain did not differ.

A PM transfer to compensate for the loss of SA function caused by LTN palsy was first described by Tubby in 1904. Since then, several studies described different surgical techniques for attaching the PM either directly to the scapula or after augmenting and lengthening with various allografts or autografts. Studies using an augmented and lengthened tendon graft most commonly used a fascia lata autograft[2, 5, 7, 16-18]. The use of a graft provides the ability to control the length of the PM and lessen the risk of putting too much stress on the transfer. In the present study, we used an achilles tendon allograft to avoid the morbidity of the donor site and reduce intra-operative time of graft harvesting. To the best of our knowledge, only Chalmers et al. have previously published results of a small patient cohort (n = 10) with scapular winging due to LTN palsy that underwent PM transfer with an achilles allograft[19].

Irrespective of the chosen technique, a recent systematic review reported significant improvements in shoulder function, pain scores, and shoulder scores after PM transfer for LTN palsy after a median follow-up of 32 months, indicating that PM transfer is an excellent option for patients not recovering after nonsurgical management[12]. However, no studies that were included in this review described the long-term outcomes, with the longest mean follow-up being approximately 5 years[3]. Long-term outcomes are, however, particularly important in this specific patient population as most patients are relatively young at the time of disease onset. Information on long-term improvement of shoulder function and associated impact on QoL are useful for both orthopedic surgeons and patients during counselling, thus for better handling of patient expectations.

The mean VAS pain scores in the present study (i.e. 2.0 at rest and 2.9 during motion, 10 being excruciating pain) after a median follow-up of 17 years were comparable to postoperative VAS pain-scores found by other studies (mean; 2.9; range 1-4)[3-5, 19-21]. Mean Constant scores at final follow-up were, however, lower: 69 in the present study in comparison with scores between 82 and 85 reported by three other studies[4, 7, 22]. Also, the recurrence of scapular winging at

final follow-up was slightly higher in our study (33%) in comparison to other studies (mean 16%, range 0-60%)[12]. In the present study, forward flexion increased by a mean of 50 degrees one year after surgery, which is comparable to the pooled results presented in a systematic review (mean improvement: 47 degrees)[12]. After a median follow-up of 17 years shoulder forward flexion was on average 20 degrees lower compared to the first postopyear, which was still substantially better than preoperative shoulder function, indicating that functional improvements persist in the long run.

It has been suggested previously to evaluate the effect of surgical intervention by measuring the functional ROM, i.e. the minimum required ROM to complete all tasks in daily living, as this is easier to interpret for both patient and surgeon[23]. Namdari et al. concluded that to successfully complete all tasks of daily living, approximately 120° forward elevation, 130° abduction and 60° external rotation in 90° abduction is needed[24]. When interpreting our results in this manner, none of the patients had a functional ROM preoperatively, while eleven patients (68%) had a functional RoM 1-year postoperatively and nine patients (56%) still had a functional RoM at final follow-up. This indicates that even after a median follow-up of 17 years, a substantial part of patients still have sufficient shoulder functionality to perform all daily life activities.

Several factors might explain the relatively high recurrence rate of scapular winging in comparison to other studies and decrease in function when comparing long-term results to 1 year postoperative function. First, the tendon graft may have ruptured. Secondly, it is also possible that stretching of the graft might decrease shoulder function and lead to recurrence of winging over time. Furthermore, it is possible that patients developed other shoulder pathology limiting shoulder function as a consequence of altered shoulder kinematic patterns due to the PM transfer. Lastly, shoulder function is known to decline with age[25]. The loss in function after a median follow-up of 17 years may be partially attributable to "healthy ageing"[26].

Our study has some limitations: first, the retrospective study design comes with inherent flaws such as missing data. For that matter, only data on shoulder function were available both pre- and postoperatively, so we could not compare other outcome measures with its baseline value to more directly assess improvement in e.g. pain scores or QoL. Secondly, it is a small case series, although this is representative of the rarity of this type of shoulder pathology both in clinical practice and the literature. Thirdly, no postoperative imaging (e.g. by MRI) was performed to visualize the tendon-bone connection, so that we cannot ascertain that the tendon transfer remained intact, or that patients had developed other shoulder pathology. Lastly, it is possible that scapular winging in some of the patients (e.g. the patient with bilateral scapular winging) was caused by a more extensive and complex peripheral nervous system disorder (e.g.neuralgic amyotrophy), which could have affected functional outcomes[27]. Strengths of the study are its length of follow-up, which to the best

of our knowledge is the longest follow-up for PM transfer reported in the literature, and its minimal loss to follow up.

CONCLUSION

The results of the present study indicate that PM transfer with an achilles tendon allograft augmentation for persistent scapular winging due to LTN palsy results in functional improvements that persist long-term. These functional improvements likely translate to better QoL based on their association. Despite the functional improvements, the present study also showed that scapular winging due to LTN palsy had a substantial impact on the QoL of patients after PM transfer, with scores in the physical domain lower than for the general Dutch population. The present study adds information on the long-term outcomes of PM transfer for LTN palsy which can be used to inform patients during a shared decision-making process with their physician in clinical practice.

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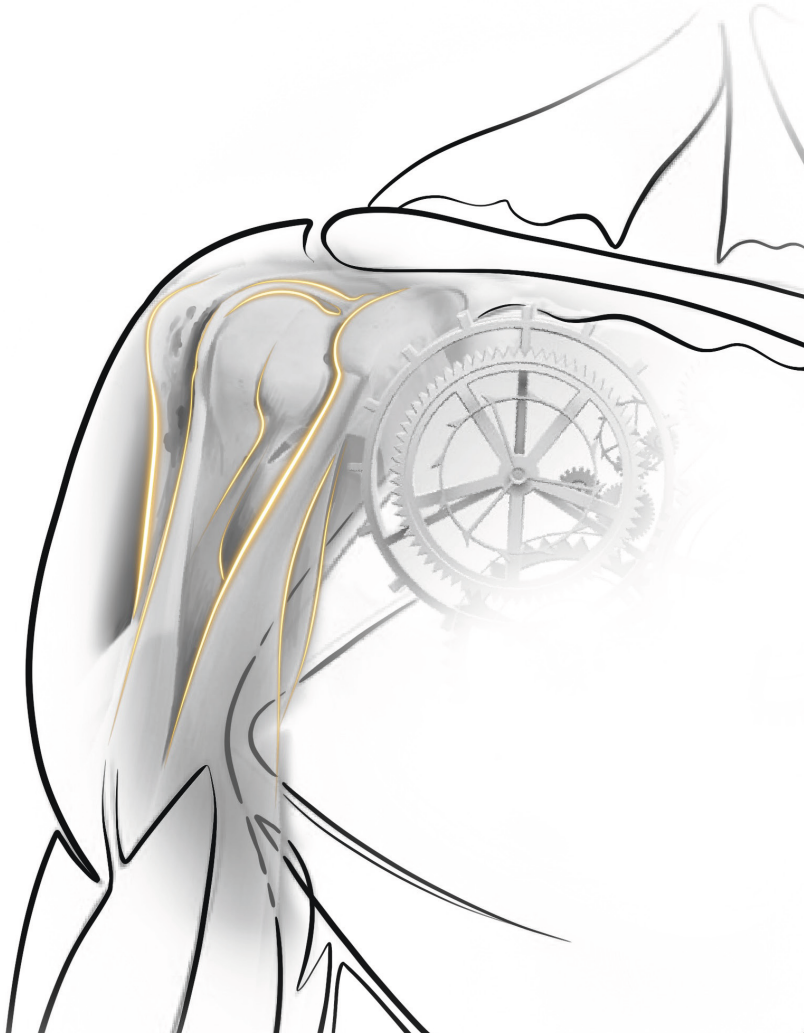

Chapter 4

Ageing and joint position sense of the asymptomatic shoulder: an observational study

Timon H. Geurkink[1,2], MD , Celeste L. Overbeek[1,2], MD, Perla J. Marang-van de Mheen, PhD[3], Jochem Nagels[1,2], MD, Rob G.H.H. Nelissen[1,2], MD, PhD, Jurriaan H. de Groot, PhD[2].

1. Department of Orthopaedics, Leiden University Medical Center, Postzone J-11-R, PO box 9600, 2300RC Leiden, the Netherlands
2. Laboratory for Kinematics and Neuromechanics, department of Orthopaedics and Rehabilitation, Leiden University Medical Center, Leiden, the Netherlands
3. Department of Biomedical Data Sciences, Medical Decision Making, Leiden University Medical Center, Leiden, the Netherlands.

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ABSTRACT

Purpose:
This study aimed to quantify the extent to which age was associated with joint position sense (JPS) of the asymptomatic shoulder as measured by joint position reproduction (JPR) tasks and assess the reproducibility of these tasks.

Methods:
120 Asymptomatic participants aged 18-70 years each performed 10 JPR-tasks. Both contralateral and ipsilateral JPR-tasks were evaluated on accuracy of JPR under active- and passive conditions at two levels within the shoulder forward flexion trajectory. Each task was performed three times. In a subgroup of 40 participants, the reproducibility of JPR-tasks was assessed one week after initial measurement. Reproducibility of JPR-tasks was evaluated by both reliability (intra-class correlation coefficients (ICC's)) and agreement (standard error of measurement (SEM)) measures.

Results:
Age was not associated with increased JPR-errors for any of the contralateral or ipsilateral JPR-tasks. ICC's ranged between 0.63 and 0.80 for contralateral JPR-tasks, and from 0.32 to 0.48 for ipsilateral tasks, except for one ipsilateral task where the reliability was similar to contralateral tasks (0.79). The SEM was comparable and small for all JPR-tasks, ranging between 1.1 and 2.1.

Conclusion:
No age-related decline in JPS of the asymptomatic shoulder was found, and good agreement between test and re-test measurements for all JPR-tasks as indicated by the small SEM.

Keywords: Shoulder, Ageing, Proprioception, Joint Position Sense, Joint Position Reproduction, Reproducibility.

INTRODUCTION

The shoulder, the glenohumeral (GH) joint in particular, contributes to the exceptional mobility of the arm. However, because of its extensive mobility, it is also inherently an unstable joint and, therefore, susceptible to injury[1]. To maintain joint stability during movement and prevent injury, the glenohumeral joint relies heavily on a coordinated interplay between its dynamic (e.g. muscles) and static stabilisers (e.g. labrum, ligaments, and capsule)[1]. A crucial factor for a well-coordinated interplay between these stabilisers is proprioception[2]. Proprioception is defined as "our perception of joint movement and positioning in space in the absence of visual feedback" [3, 4]. It is regulated by i) the cumulative proprioceptive input of mechanoreceptors within muscles, tendons, ligaments, joint capsules and skin, and ii) the central processing of this proprioceptive input in the central nervous system[5, 6]. Together, the peripheral mechanoreceptors and central information processing ensure adequate motor responses from shoulder stabilisers and consequently joint stability during movement[2].

Proprioception includes several subdomains such as joint positioning sense (JPS), kinaesthesia, sense of change in velocity and sense of force[7]. Various measurement methods have been developed to test these subdomains specifically, of which JPS is most commonly used to measure proprioception in a clinical setting[8, 9]. JPS can be evaluated with various joint position reproduction (JPR) tasks. JPR-tasks can be assessed under active- or passive conditions and may involve either ipsilateral (i.e. the same arm is used for position reproduction) or contralateral (i.e. opposite arm is used for position reproduction) tasks[6, 10]. The peripheral proprioceptive input and central processing of this input depend on the type of JPR-task. For example, muscle spindles and Golgi tendon organs are considered the most important mechanoreceptors for active JPR-tasks, while cutaneous mechanoreceptors (e.g. Pacini and Meisner's corpuscles) play a more dominant role in passive JPR-tasks[6]. Ipsilateral JPR-tasks have a memory component as the same arm is used for both the reference- and reproduction position so that participants must use their memory to accurately reproduce the reference position. Contralateral JPR-tasks require interhemispheric communication as the opposite arm is used for position reproduction[11]. Therefore, studies should assess a combination of different JPR-tasks to provide a comprehensive overview of JPS.

In existing literature, it is suggested that JPS declines with age[12, 13], thereby jeopardizing joint stability and increasing the risk of shoulder injury[2]. Several physiological changes that occur with ageing might affect JPS, such as a decline in the number of mechanoreceptors, decreased mechanoreceptor sensitivity, and degenerative changes of the central nervous system[14-17]. Previous studies that evaluated the effect of age on JPS have, however, reported conflicting results and predominantly focussed on the lower extremities[12, 18-22]. Only two previous studies evaluated the association between ageing and JPS in the asymptomatic shoulder, both suggesting that JPS declines with age[10, 23]. However, these studies only evaluated a selected subset of active- or passive ipsilateral JPR-tasks, thereby not providing a comprehensive overview of JPS for the ageing shoulder, and also did not assess the reproducibility of JPR-

tasks. This study therefore aimed to provide a more comprehensive overview of the extent to which age was associated with JPS of the shoulder in an asymptomatic population. We hypothesised that there would be an age-related decline in JPS. As a secondary aim, we explored the reproducibility of JPR-tasks over time in a subgroup of participants.

METHODS

Participants

In this observational study we evaluated JPS of the shoulder in asymptomatic participants between the age of 18 and 70 years. The participants were recruited through advertising in the Leiden University Medical Center (LUMC) public areas and snowballing methods between May 2018 and January 2019 (Figure 1). To ensure an equal distribution of participants across different age ranges, we recruited 30 participants within each of the following age categories: 18-31, 32-45, 46-58, and 59-70 years old. The exclusion criteria were previous shoulder complaints (i.e. participants who received medical attention for a shoulder complaint or experienced shoulder complaints >1 week), no full range of motion during physical examination, pregnancy, a history of malignancy, previous shoulder fracture, previous shoulder surgery, neurologic or muscle disease, diabetes mellitus, electronic implants, or insufficient Dutch language skills. All measurements were conducted at the laboratory for Kinematics and Neuromechanics (LUMC, the Netherlands). The institutional medical ethical board (METC Leiden-Den Haag-Delft) approved this study (protocol number: P18.028) and written informed consent was obtained from all study participants.

Measurement set-up

All measurements were performed using a 3D-electromagnetic motion analysis device (Flock of Birds (FoB); Ascension Technology, Milton, VT, USA). This validated motion device is frequently used to quantify shoulder motion and can accurately (error margin is approximately 2 millimetres) assess the position of the upper limbs in the 3 dimensional space[24, 25].

During all measurements, participants were seated in the FoB with their torso upright against the back of a chair. Seven wired sensors were placed on the participant in a standardised way by the investigator using either straps with adhesive tape (manubrium sterni and bilaterally on the flat craniolateral surface of the acromion) or hook-and-loop closures (bilaterally posteriorly on the distal part of the humerus and bilaterally on the dorsal side of the distal forearm). One additional sensor was attached to a stylus to digitise twenty-four-bony landmarks identified by palpation and create a 3D bone model specific for each participant[24, 26].

Experiment design

JPS was assessed using multiple JPR-tasks in the trajectory of shoulder forward flexion. JPR is widely accepted and one of the most commonly used methods for measuring proprioception through the accuracy of position reproduction (JPR-error, i.e. the difference between a

predetermined reference position and the reproduction of this position) in the absence of visual feedback[4, 27]. In the present study, the position of the wrist (i.e. the projection of the centre of the processus styloideus radii and the processuss styloideus ulnae) was used to estimate JPR-error. JPR-error was defined as the absolute difference in height (in centimetres (cm)) between the wrist's reference- and reproduction position on the y-axis of the FoB system.

A combination of ipsilateral and contralateral JPR-tasks were conducted because of the difference in central processing for these tasks as described above[6]. For ipsilateral JPR-tasks, the arm of a blindfolded participant is brought (either actively by the participant or passively by the investigator) to a predefined reference position for at least three seconds and the participant is asked to remember this position. Then, the arm is returned to the starting position. Subsequently, the investigator requests the participant to reproduce the predefined position (again either actively or passively) with the same arm. For contralateral JPR-tasks, the arm of a blindfolded participant is brought to a predefined reference position (again either actively or passively) and the arm stays in this position. Thereafter, the investigator requests the participant to reproduce the predefined position with the contralateral arm.

As it has been shown that JPR-error varies with the level of shoulder forward flexion[28], we tested JPS at two different levels of shoulder forward flexion: i) a low position (i.e. approximately 50 degrees of shoulder forward flexion) and ii) a high position (i.e. approximately 90 degrees of shoulder forward flexion). For each position, participants had to perform five different types of JPR-tasks as the peripheral proprioceptive input differs for active- and passive JPR-tasks[11]: i) contralateral active-active reproduction, ii) contralateral passive-active reproduction, iii) ipsilateral active-active reproduction, iv) ipsilateral passive-passive reproduction, and v) ipsilateral passive-active reproduction. Each JPR-task was performed three times, so each participant performed 30 JPR-tasks in total.

During all measurements the participants were blindfolded and did not wear clothes covering the shoulder to avoid proprioceptive input of the skin. The participants were instructed to keep their elbows straight during all JPR-tasks. All measurements were conducted by four investigators, who had received extensive training before study start. To minimise the effect of arm dominance, the arm to be tested was determined using computer-generated block randomisation in blocks of two. Furthermore, to reduce learning effects, the participants did not receive feedback regarding their JPR accuracy and the sequence of tasks was randomised to minimise the impact of muscle fatigue[29].

When evaluating the association between age and JPS, it is essential that the outcome measure is reproducible. Therefore, we also assessed whether JPR-tasks could be reproduced over time. For a subgroup of participants, JPR-tasks were assessed twice by the same investigator, one week after the first assessment (see sample size justification below). We assumed that one week was short enough to avoid any significant changes within the participants and/or investigator affecting study measurements.

All data were analysed using custom-made software in MATLAB (2021b release, The Mathworks Inc. Natick, Massachusetts, USA).

Statistical analysis

Before our study, a power analysis using G*power Version 3.0.10[30] was conducted to estimate the sample size needed. Based on an alpha of 0.05 and a power of 0.80 it was estimated that 111 participants were required for an effect size of 0.3 with regression analysis. Accounting for approximately 10% loss of data, 120 participants were recruited. For reproducibility analysis, it is advised to recruit at least 30 participants[31]. We increased the sample size for reproducibility analysis to 40 participants to account for potential loss of data.

All statistical analyses were performed using the statistical package SPSS version 25.0 (IBM, Armonk, NY, USA). Parametric continuous data were described using means, standard deviation (SD) and 95% confidence intervals (CI) and nonparametric data were expressed in medians and interquartile ranges (IQR). Numbers and percentages were presented for categorical data.

For each of the 10 JPR-tasks, a linear mixed model analysis was used to evaluate the association between JPR-error and age (in years) to account for the three repeated task measurements. We modelled covariance with an unstructured covariance structure. Repetition (repetition 1, 2, and 3) was included as the repeated factor, and we adjusted for sex (male/female), BMI (kg/m^2) and sports hours per week which were included as fixed factors. Measurements in which the reference position deviated > 20 degrees from the actual target value were excluded from the analysis. A Bonferroni correction was applied to correct for multiple testing[32] which set the P-value to indicate statistical significance on less than 0.005 (α=.05/10).

To estimate the reproducibility of JPR-tasks, both reliability (intra-class correlation coefficients (ICC)) and agreement (standard error of measurement (SEM)) measures were calculated[33]. The mean JPR-error of the initial- and re-test measurements in the subgroup of participants was used to determine JPR-task reliability over time, and quantified by the ICC from a two-way mixed model with absolute agreement[34]. The following classification was used to interpret ICC values: 0.0-0.5, poor reliability; 0.5-0.75 moderate reliability; 0.75-0.9, good reliability; 0.9-1.0 excellent reliability[31]. A well-known disadvantage of ICC is that a lack of variability among the sampled participants may result in misleadingly low ICC values[31]. Therefore, the ICC was supplemented by the SEM, calculated for each JPR-task using the following formula: $SEM = SD \times \sqrt{(1-ICC)}$[33]. Here, SD reflects the pooled standard deviation from the initial and re-test measurements.

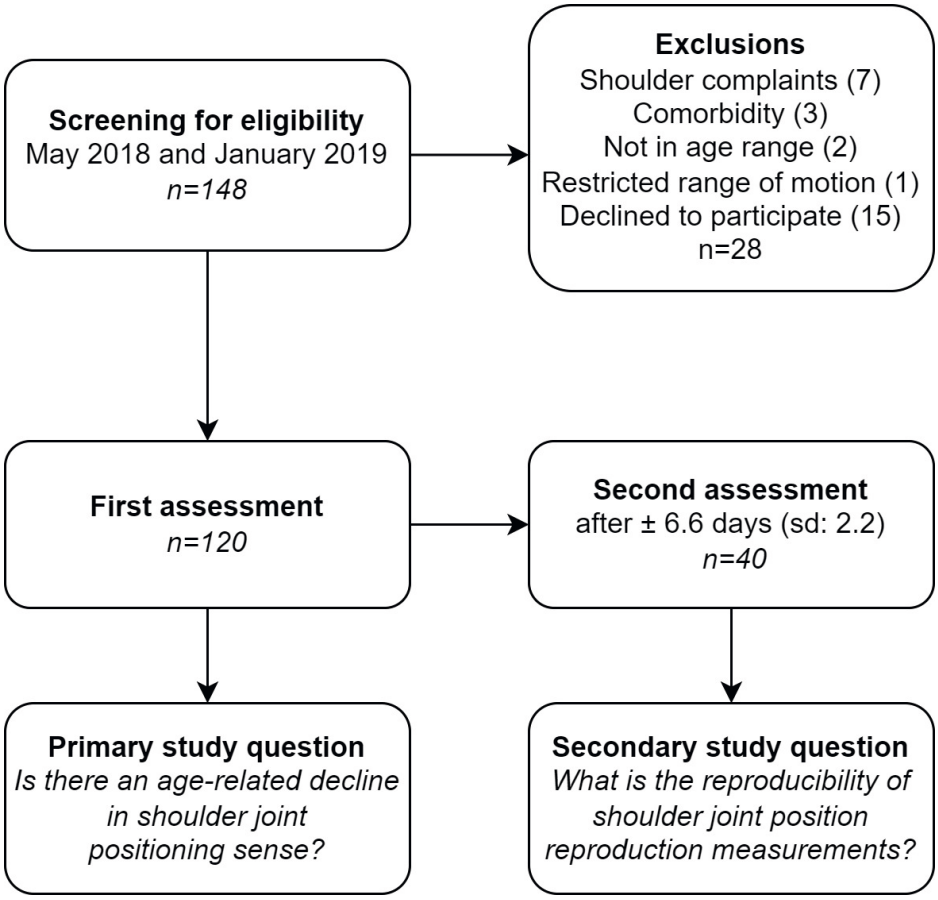

Figure 1. Flowchart of participant inclusion.

RESULTS

In total, 120 participants participated in the study with a mean age of 44 years (SD: 14.9). The majority were female (56%) and right-hand dominant (92%). Other baseline characteristics can be found in Table 1.

Table 2 shows the association of age with JPR-error in each of the contralateral and ipsilateral reproduction tasks. Age was not significantly associated with JPR-errors for any of the contralateral JPR-tasks ($p > 0.005$). Similar results were found for the ipsilateral JPR-tasks, with only one of the ipsilateral JPR-tasks approaching statistical significance (task: Passive-Active; Low, estimate: 0.066 (95%CI:[0.020-0.112], $p = 0.005$).

A subgroup of 40 (33%) participants had their JPR-tasks re-assessed by the same assessor, after a mean of seven days (SD: 2.2). These participants had a mean age of 44 years (SD: 15.4),

23 (58%) were male and 38 (95%) were right-hand dominant. For contralateral reproduction tasks, the ICC's ranged between 0.63 (task: Active-Active; Low) and 0.80 (task: Passive-Active; Low)(Table 3). For ipsilateral reproduction tasks, the ICC's were considerably lower (ranging between 0.32 and 0.48), except for the passive-active task in low position (0.79). The SEM was comparable for all JPR-tasks, ranging from 1.1 to 2.1 (Table 3).

Table 1. Participant characteristics

	Asymptomatic participants
	n=120
Age, years (mean, sd)	44 (14.9)
Female (n, %)	67 (56)
Right side dominance (n, %)	110 (92)
Dominant side assessed (n, %)	60 (50)
BMI (mean, sd)	24 (3.7)
Profession (n, %)	
Unemployed (n, %)	12 (10)
With upper limb activity below shoulder level (n, %)	99 (82.5)
With upper limb activity above shoulder level (n, %)	9 (7.5)
Sports	
No sports (n, %)	15 (12.5)
Sports with upper limb activity below shoulder level (n, %)	53 (44.2)
Sports with upper limb activity above shoulder level(n, %)	52 (43.3)
Hours/ week	3.8 (2.8)
Self-reported general health	
Excellent (n, %)	31 (25.8)
Very good (n,%)	49 (40.8)
Good (n,%)	39 (32.5)
Fair (n,%)	1 (0.8)
Bad (n,%)	0 (0)
Constant Shoulder score dominant arm (median, IQR)	96 (93, 100)
Constant Shoulder score non-dominant arm (median, IQR)	95 (92, 100)
VAS for pain at rest 0-100 (median, IQR)	0 (0, 3)
VAS for pain during movement 0-100 (median, IQR)	1 (0, 3)
VAS for daily functioning 0-100 (median, IQR)	0 (0, 3

Table 2. Association of age and other independent variables with JPR-error for all contralateral and ipsilateral JPR-tasks.

Contralateral JPR-tasks

Task & Position	*Active-Active, High*		*Active-Active, Low*		*Passive-Active, High*		*Passive-Active, Low*	
	Estimate (95CI)	P-value	Estimate (95CI)	P-value	Estimate (95CI)	P-value	Estimate (95CI)	P-value
Intercept	0.593 (-1.953-3.139)	0.645	0.085 (-3.763-3.933)	0.965	2.419 (-2.066-6.904)	0.287	4.189 (-1.211-9.590)	0.127
Age (years)	0.014 (-0.011-0.040)	0.271	0.010 (-0.027-0.046)	0.595	0.015 (-0.030-0.060)	0.512	-0.013 (-0.066-0.041)	0.644
Sex*	-0.457 (-1.172-0.258)	0.208	-0.141 (-1.159-0.877)	0.785	-0.618 (-1.862-0.626)	0.327	-0.090 (-1.563-1.382)	0.903
BMI (kg/m^2)	0.078 (-0.025-0.180)	0.136	0.128 (-0.030-0.287)	0.112	0.081 (-0.104-0.266)	0.389	0.133 (-0.090-0.356)	0.240
Sports (hours/week)	-0.008 (-0.136-0.120)	0.900	-0.044 (-0.219-0.130)	0.614	0.026 (-0.199-0.251)	0.817	0.009 (-0.255-0.273)	0.946

Ipsilateral JPR-tasks

Task & Position	*Active-Active, High*		*Active-Active, Low*		*Passive-Passive, High*		*Passive-Passive, Low*		*Passive-Active, High*		*Passive-Active, Low*	
	Estimate (95CI)	P-value	Estimate (95CI)	P-value	Estimate (95CI)	P-value	Estimate (95CI)	P-value	Estimate (95CI)	P-value	Estimate (95CI)	P-value
Intercept	3.348 (1.367-5.330)	0.001	4.747 (1.445-8.048)	0.005	5.402 (2.803-8.002)	<0.001	3.195 (0.933-5.457)	0.006	0.434 (-2.708-3.576)	0.785	4.694 (0.066-9.322)	0.047
Age (years)	0.002 (-0.018-0.022)	0.842	0.018 (-0.014-0.051)	0.264	-0.026 (-0.052- -0.000)	0.048	0.024 (0.001-0.046)	0.040	0.000 (-0.031-0.032)	0.978	0.066 (0.020-0.112)	0.005
Sex*	-0.194 (-0.739-0.351)	0.483	-0.430 (-1.336-0.475)	0.347	-0.049 (-0.769-0.672)	0.894	-0.248 (-0.874-0.378)	0.434	-0.258 (-1.138-0.623)	0.563	-0.690 (-1.966-0.585)	0.286

Table 2. Association of age and other independent variables with JPR-error for all contralateral and ipsilateral JPR-tasks. (continued)

Ipsilateral JPR-tasks												
Task & Position	*Active-Active, High*		*Active-Active, Low*		*Passive-Passive, High*		*Passive-Passive, Low*		*Passive-Active, High*		*Passive-Active, Low*	
BMI (kg/m^2)	-0.028 (-0.106-0.050)	0.480	-0.093 (-0.228-0.042)	0.176	-0.026 (-0.131-0.078)	0.615	-0.038 (-0.130-0.055)	0.421	0.168 (0.043-0.294)	0.009	-0.092 (-0.279-0.094)	0.329
Sports (hours/week)	-0.081 (-0.182-0.020)	0.115	0.022 (-0.142-0.187)	0.787	-0.081 (-0.209-0.047)	0.213	-0.055 (-0.166-0.056)	0.330	0.032 (-0.126-0.190)	0.692	0.091 (-0.139-0.321)	0.433

Results of linear mixed model analysis: A p-value <0.005 was considered statistically significant.

* Male is reference. Estimates in 10^{-2} m.

Abbreviations: JPR = Joint Position Reproduction, CI = Confidence Interval

Table 3. Reproducibility of contralateral and ipsilateral JPR-tasks.

Contralateral JPR-tasks				
Task & Position	*Active-Active, High*	*Active-Active, Low*	*Passive-Active, High*	*Passive-Active, Low*
Reliability (ICC)	0.716	0.632	0.752	0.804
95% CI	0.451-0.853	0.245-0.821	0.514-0.873	0.612-0.901
Agreement (SEM)	1.1	1.5	1.5	1.7

Ipsilateral JPR-tasks						
Task & Position	*Active-Active, High*	*Active-Active, Low*	*Passive-Passive, High*	*Passive-Passive, Low*	*Passive-Active, High*	*Passive-Active, Low*
Reliability (ICC)	0.400	0.389	0.419	0.480	0.316	0.794
95% CI	-0.143-0.684	-0.229-0.694	-0.119-0.698	0.039-0.723	-0.284-0.637	0.606-0.892
Agreement (SEM)	1.4	1.9	1.4	1.2	2.1	2.1

Abbreviations: JPR= Joint Position Reproduction; ICC= Intra-Class Correlation Coefficient; CI= Confidence Interval; SEM= Standard Error of Measurement (in centimetres)

DISCUSSION

The present study showed that higher age was not associated with a decline in JPS of the shoulder, contrary to our initial hypothesis. The ICC's suggested moderate-to-good reliability over time for contralateral JPR-tasks but lower (poor) reliability for ipsilateral JPR-tasks, except for the passive-active task in low position which had similar good reliability. However, the SEM was comparable and low for all JPR-tasks, indicating good agreement between test and re-test measurements.

Two previous studies evaluated the association between age and JPS in the asymptomatic shoulder[10, 23]. Both studies suggested there was an age-related decline in JPS, but the reported differences in JPR-error between younger and older participants were small (range: 1-4 degrees of shoulder forward flexion) and the clinical relevance of these differences can be questioned[20]. Additionally, the results of these studies must be interpreted with caution since they had several methodological limitations and were limited to a relatively small number of participants (40 and 44 participants respectively). For instance, Zuckerman et al. only performed one measurement for every JPR-task, which is considered insufficient for JPR[10]. Echalier et al. did perform multiple measurements for each JPR-task, using the mean value across measurements for analysis. However, an overall mean value does not adequately convey proprioceptive information since the variance of JPR measurements is lost when using only a mean value[6]. Importantly, both studies only evaluated a selected subset of JPR-tasks, thereby not providing a complete overview of JPS for the ageing shoulder.

Several other factors could explain why we did not find an age-related decline in JPS. First, it is possible that JPS is not primarily affected by ageing itself. Instead, it may reflect age-related changes in cognitive functions[22] (e.g. deficits in memory) or is the consequence of reduced physical activity with ageing[19, 20]. Rikli et al. previously suggested that physical activity level is more important for maintaining proprioception than age[35] which might explain our findings as the participants in our study were relatively active (mean duration of sports activities was 4 hours per week). Secondly, a decline in JPS could be present only in individuals older than 70 years of age. Yang et al. showed that a decline in proprioceptive acuity of the ankle joint was most prominent beyond the age of 75[36] whereas the participants in our study were considerably younger. Third, an age-related decline may not be present in the shoulder forward flexion trajectory. Contrary to shoulder abduction or rotation, shoulder forward flexion is almost completely within the visual field and the participants may be more skilled and experienced with such tasks as most daily activity movements are performed in front of the body[11, 37]. Lastly, an age-related decline in proprioception may be absent in JPS, but could be present in other subdomains of proprioception (e.g. kinaesthesia).

The pathophysiology of shoulder disorders is considered multifactorial[38]. A decline in proprioception could contribute to the development of shoulder pathology as it leads to instability of the shoulder joint[2]. Previous studies have demonstrated that there is an

association between proprioceptive deficits and shoulder disorders, such as rotator cuff disease, shoulder instability, frozen shoulder and subacromial pain syndrome[4, 39-42]. It is unknown whether proprioceptive deficits are the cause or the result of shoulder pathology[2]. Recent evidence shows that deficits in proprioception have a negative influence on rehabilitation processes and may predict poor surgical outcomes, thereby showing its clinical importance[43]. For that matter, proprioception may be targeted to treat shoulder pathology which highlights the need for future studies to further investigate the role of shoulder proprioception. To further explore the role of proprioception in shoulder pathology, it is first necessary to understand its natural course in healthy individuals. The present study therefore adds to existing literature that there seems to be no age-related decline of JPS in healthy individuals, thereby providing reference for future research.

With regard to reproducibility of JPR tasks we found substantially lower ICC's for most ipsilateral JPR-tasks, but comparable and low SEM. The lower ICC's for ipsilateral JPR-tasks in comparison to contralateral JPR-tasks can be understood by the fact that ipsilateral JPR-tasks result in smaller JPR-errors, which may be explained by the lack of need for interhemispheric communication (i.e. ipsilateral JPR tasks do not require interhemispheric communication as the same arm is used for both the reference and reproduction position) that might reduce the accuracy of a JPR-task[11]. Ipsilateral JPR-tasks may lead to lower ICC's as the intra-individual variability is relatively high compared to a low population variability (participants generally accurately reproduced the reference position, i.e. had good JPS), even when the intra-individual variability is very small (see Appendix A for test-retest plots). Rather than telling something about reproducibility, the low ICC's for ipsilateral JPR-tasks may merely indicate that JPR is not able to discriminate between individuals in such a homogenous population (i.e. asymptomatic participants). The latter illustrates the necessity to evaluate the reproducibility of outcome measures with both reliability (ICC) and agreement (SEM) measures[33].

The strengths of the present study are its large sample size and the application of a variety of JPR-tasks and thus its extensive evaluation of JPS in the shoulder. While most studies only assess JPS by one specific JPR-task, we measured both contralateral and ipsilateral JPR-tasks under both active and passive conditions, thereby providing a more comprehensive overview of shoulder JPS in the asymptomatic population. However, some limitations should be noted. First, we only measured JPS in the shoulder forward flexion trajectory. Therefore, we cannot conclude whether our results are generalizable to other movement trajectories of the shoulder (e.g. abduction and/or rotation movements). Second, it is also possible to perform contralateral JPR tasks with a memory component (i.e. contralateral remembered matching), where the reference arm is returned to the starting position before position reproduction with the opposite arm. However, we did not perform contralateral remembered matching tasks within the present study. Thirdly, we cannot rule out the presence of selection bias due to the fact that participants were recruited via advertisements, which may result in a selected group of participants (e.g. a relatively active group with special interest in shoulder functioning) so

that the results do not necessarily apply to the general healthy adult population. Furthermore, we included participants based on clinical assessment and did not rule out asymptomatic pathologies through radiological examination. Hence, participants with asymptomatic shoulder pathology may have been included in the present study. Lastly, we did not include participants beyond the age of 70 and it is possible that a proprioceptive decline mainly occurs above the age of 70 years.

CONCLUSION

Using a 3D-electromagnetic motion analysis device for measuring JPS in the shoulder flexion trajectory, we found no age-related decline in JPS for the asymptomatic shoulder. Furthermore, the comparably low SEM for all JPR tasks indicated good agreement between test and re-test measurements. Future studies are needed to confirm our findings and further explore the role of proprioception in shoulder pathology.

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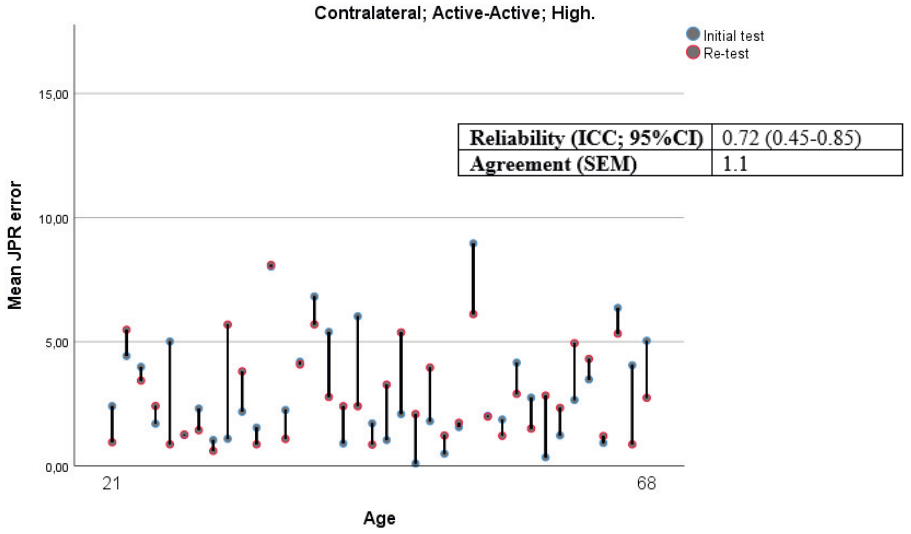
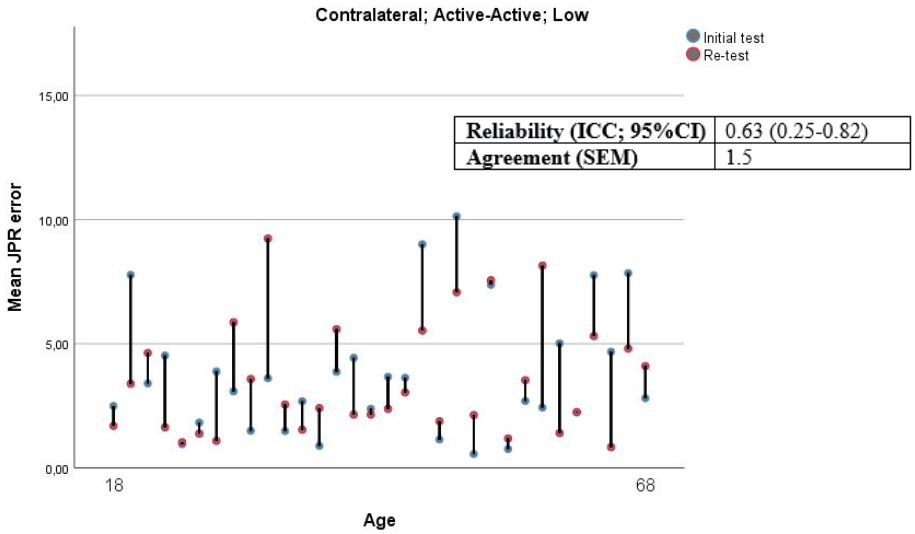
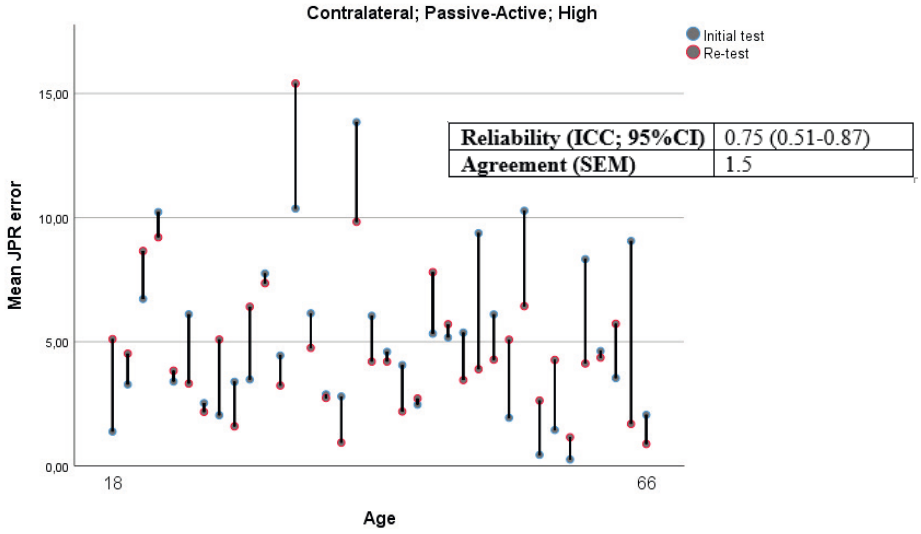
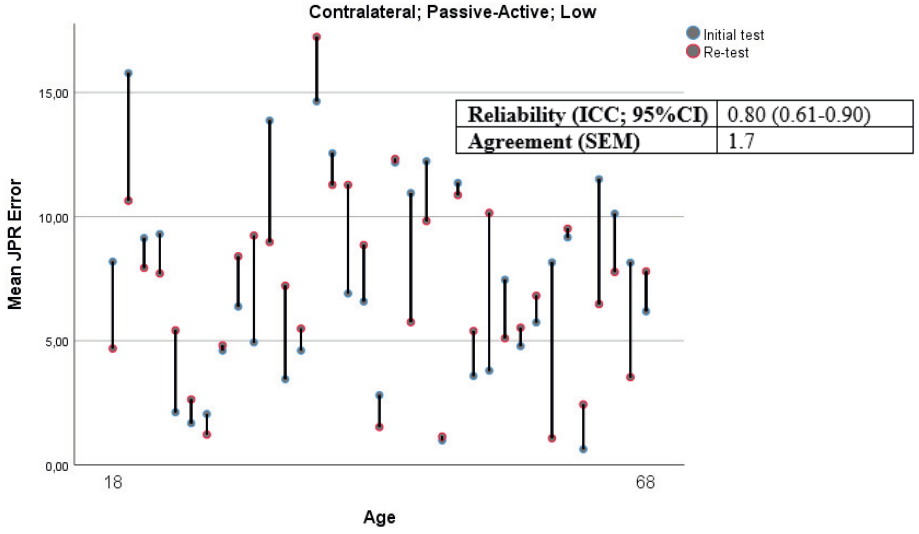
Appendix A. Test re-test plot and corresponding ICC and SEM for all JPR tasks.

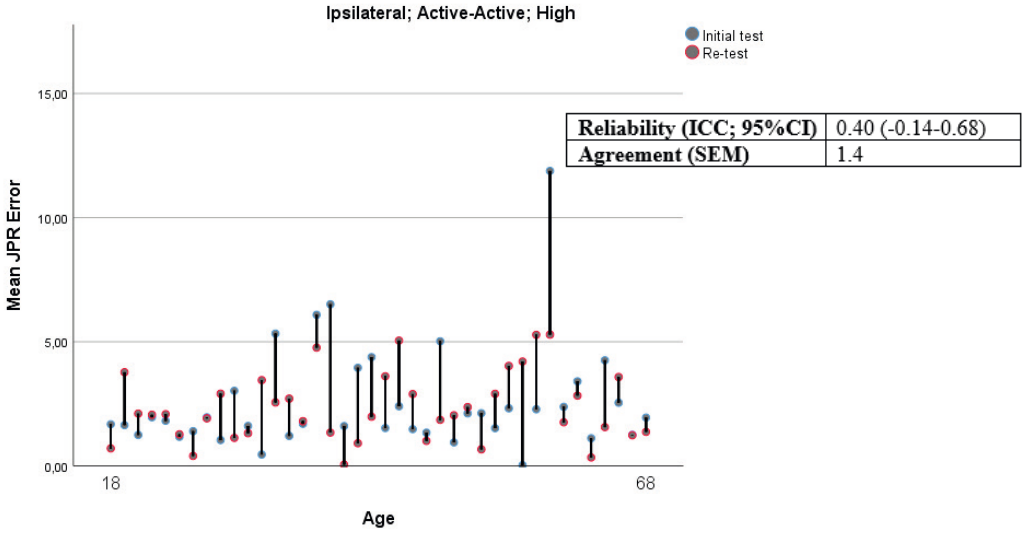
Mean JPR error of the initial test and re-test of all participants in the subgroup analysis. Each vertical line corresponds to a participant. The blue disks indicate the mean JPR error of the initial test and the red disks represent the mean JPR error of the re-test. The black line shows the difference in mean JPR error between the initial test and re-test.

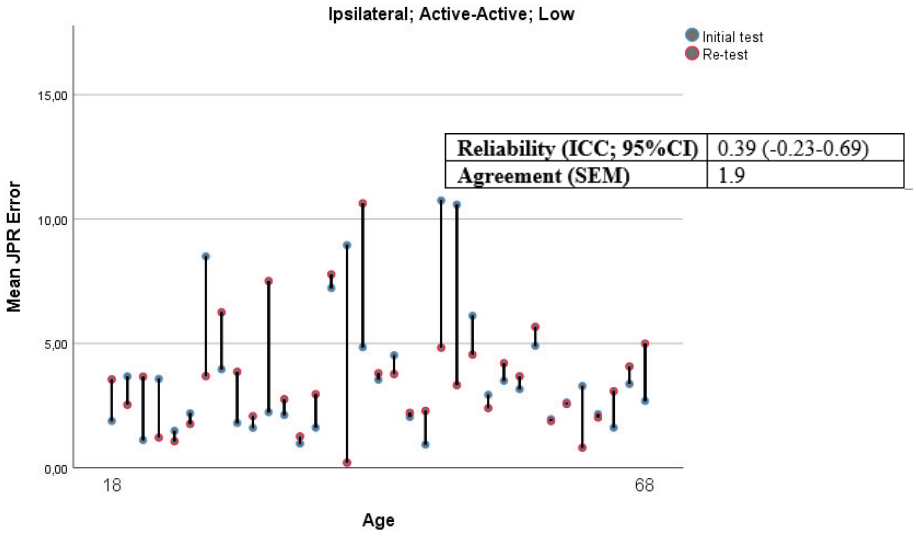


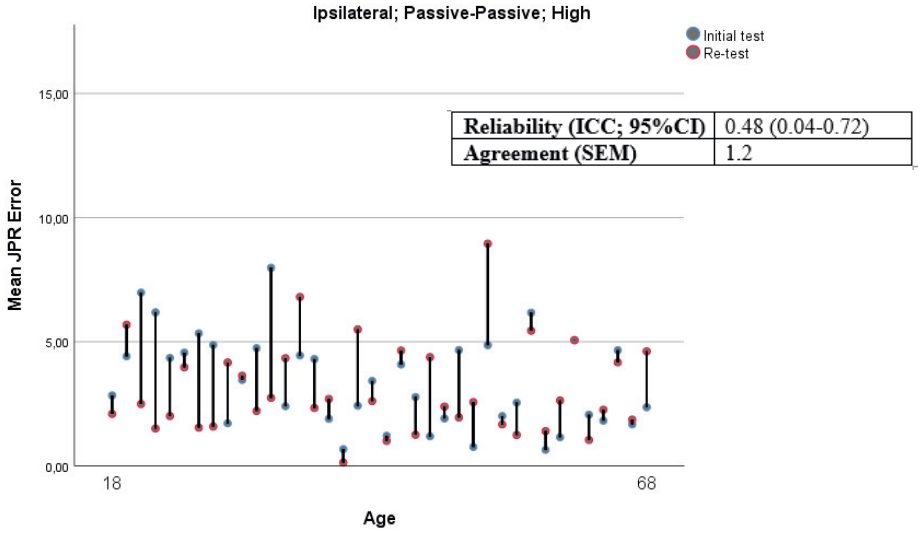


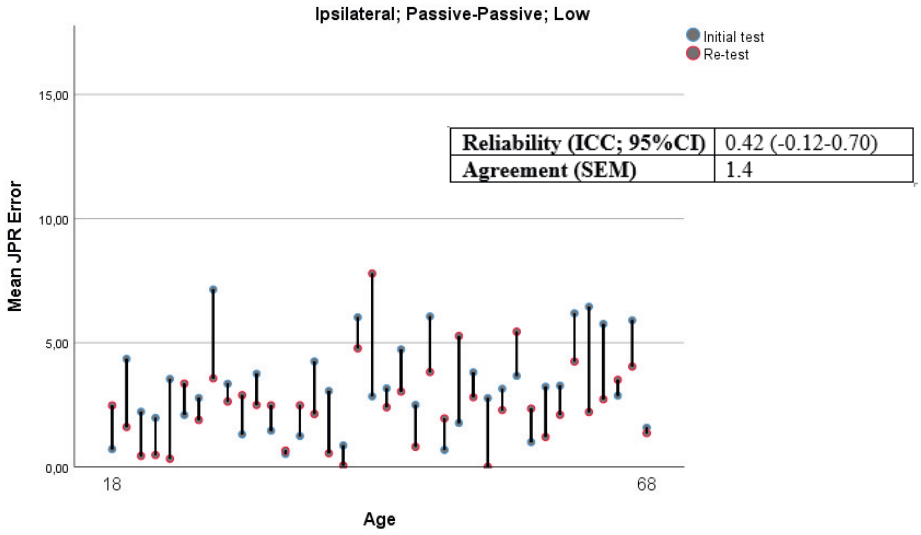

Contralateral; Passive-Active; High
Initial test
Re-test
Mean JPR error
15,00
10,00
5,00
0,00
18
66
Age
Reliability (ICC; 95%CI) 0.75 (0.51-0.87)
Agreement (SEM) 1.5


Contralateral; Passive-Active; Low
Initial test
Re-test
Mean JPR Error
15,00
10,00
5,00
0,00
18
68
Age
Reliability (ICC; 95%CI) 0.80 (0.61-0.90)
Agreement (SEM) 1.7


Ipsilateral; Active-Active; High
Initial test
Re-test
Reliability (ICC; 95%CI) 0.40 (-0.14-0.68)
Agreement (SEM) 1.4
Mean JPR Error
15,00
10,00
5,00
0,00
18
68
Age


Ipsilateral; Active-Active; Low
Initial test
Re-test
Reliability (ICC; 95%CI) 0.39 (-0.23-0.69)
Agreement (SEM) 1.9
Mean JPR Error
15,00
10,00
5,00
0,00
18
68
Age


Ipsilateral; Passive-Passive; High
Initial test
Re-test
Reliability (ICC; 95%CI) 0.48 (0.04-0.72)
Agreement (SEM) 1.2
Mean JPR Error
15,00
10,00
5,00
0,00
18
68
Age


Ipsilateral; Passive-Passive; Low
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Re-test
Reliability (ICC; 95%CI) 0.42 (-0.12-0.70)
Agreement (SEM) 1.4
Mean JPR Error
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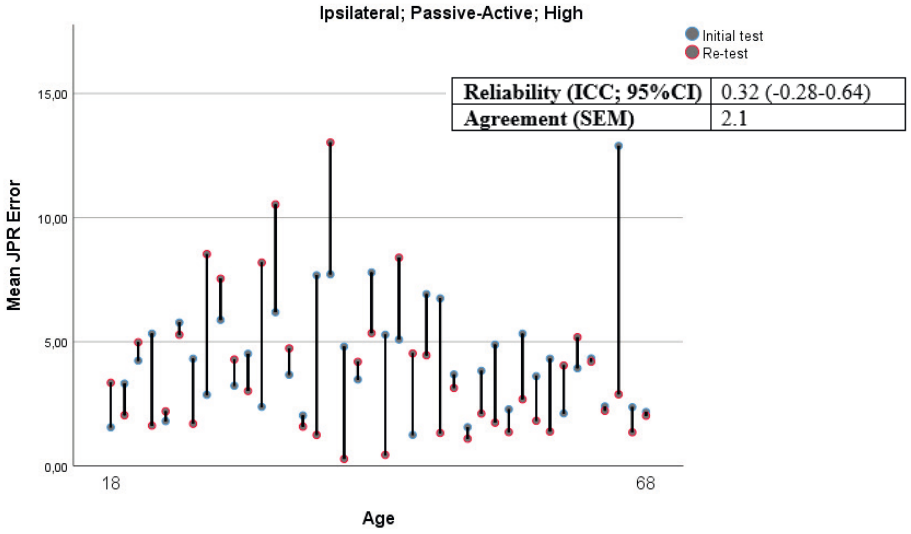
10,00
5,00
0,00
18
68
Age


Ipsilateral; Passive-Active; High
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Re-test
Reliability (ICC; 95%CI) 0.32 (-0.28-0.64)
Agreement (SEM) 2.1
Mean JPR Error
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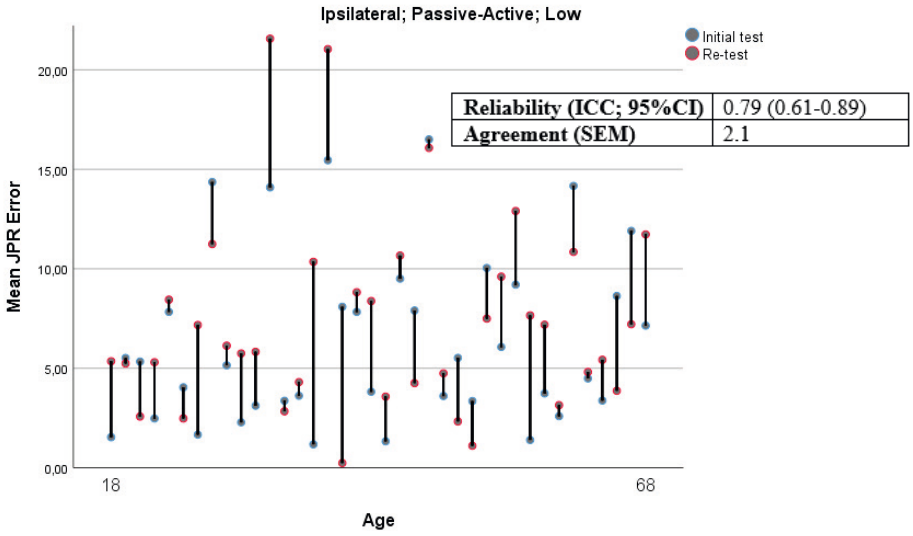
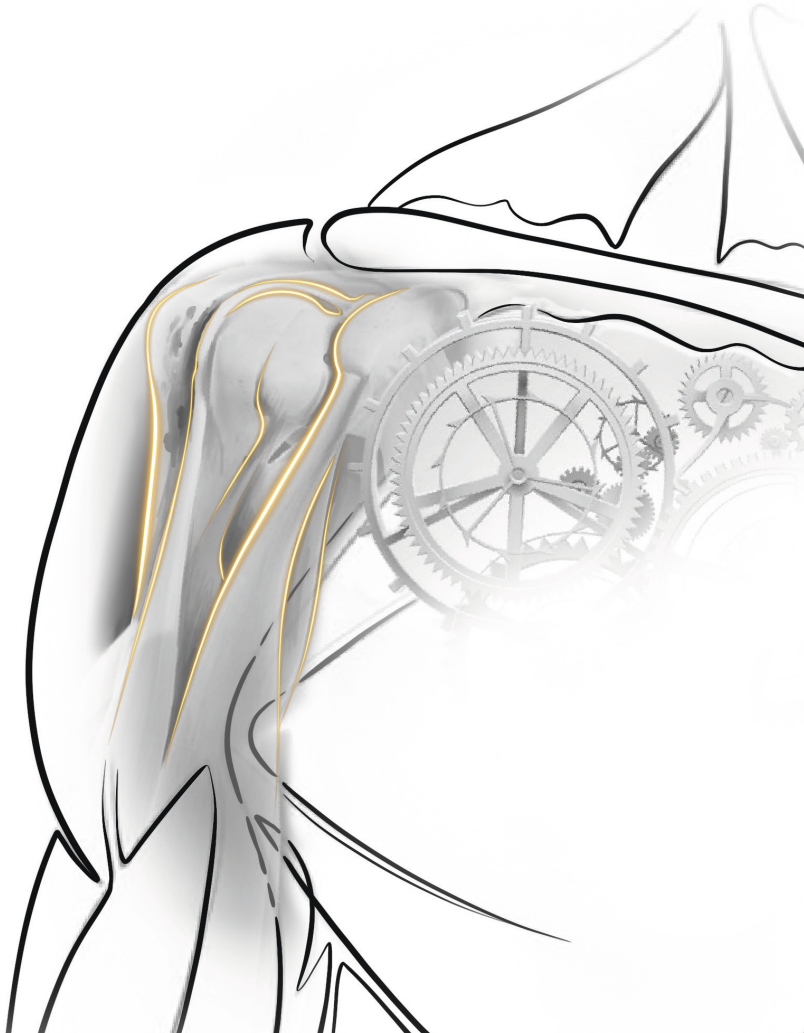
10,00
5,00
0,00
18
68
Age


Ipsilateral; Passive-Active; Low
Initial test
Re-test
Reliability (ICC; 95%CI) 0.79 (0.61-0.89)
Agreement (SEM) 2.1
Mean JPR Error
20,00
15,00
10,00
5,00
0,00
18
68
Age



Chapter 5

The relationship between publication of high-quality evidence and changes in the volume and trend of subacromial decompression surgery for patients with subacromial pain syndrome in hospitals across Australia, Europe and the United States: A controlled interrupted time series analysis.

Timon H. Geurkink, MD[1,2], Leti van Bodegom-Vos, PhD[2], Jochem Nagels, MD[1], Susan Liew, MD[3], Pieter Stijnen, PhD[4], Rob G.H.H. Nelissen, MD, PhD[1], Perla J. Marang-van de Mheen, PhD[2]

1. *Department of Orthopaedics, Leiden University Medical Centre, Postbus 9600, 2300 RC, Leiden, the Netherlands.*
2. *Department of Biomedical Data Sciences, Medical Decision Making, Leiden University Medical Centre, Leiden, the Netherlands.*
3. *Department of Orthopaedic Surgery, Alfred Hospital, Melbourne, Australia.*
4. *Department of Management Information and Reporting, University Hospital Leuven, Leuven, Belgium.*

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ABSTRACT

Aims:
To evaluate the extent to which publication of high-quality randomised controlled trials (RCTs) in 2018 was associated with a change in volume or trend of subacromial decompression (SAD) surgery in patients with subacromial pain syndrome (SAPS) treated in hospitals across various countries.

Methods:
Routinely collected administrative data of the Global Health Data@work collaborative were used to identify SAPS patients who underwent SAD surgery in six hospitals from five countries (Australia, Belgium, Netherlands, United Kingdom, United States) between 01/2016 and 02/2020. Following a controlled interrupted time series design, segmented Poisson regression was used to compare trends in monthly SAD surgeries before (01/2016 - 01/2018) and after (02/2018 - 02/2020) publication of the RCTs. The control group consisted of musculoskeletal patients undergoing other procedures.

Results:
A total of 3.046 SAD surgeries were performed among SAPS patients treated in five hospitals; one hospital did not perform any SAD surgeries. Overall, publication of trial results was associated with a significant reduction in the trend to use SAD surgery of 2% per month (Incidence rate ratio (IRR) 0.984 [0.971-0.998]; $P = 0.021$), but with large variation between hospitals. No changes in the control group were observed. However, publication of trial results was also associated with a 2% monthly increased trend (IRR 1.019 [1.004-1.034]; $P = 0.014$) towards other procedures performed in SAPS patients.

Conclusion:
Publication of RCT results was associated with a significantly decreased trend in SAD surgery for SAPS patients, although large variation between participating hospitals existed and a possible shift in coding practices cannot be ruled out. This highlights the complexities of implementing recommendations to change routine clinical practice even if based on high-quality evidence.

INTRODUCTION

The subacromial pain syndrome(SAPS) is an umbrella diagnosis that accounts for up to 70% of cases with shoulder pain[1]. Although most SAPS patients are treated non-operatively, a substantial part undergoes subacromial decompression(SAD) surgery[2]. High-quality randomised controlled trials(RCTs), however, found no significant improvement in pain or function after SAD surgery in SAPS patients compared with nonoperative management and placebo surgery[3-12]. Moreover, SAD surgery carries a risk of harm for patients and contributes to increased resources[11, 13]. Therefore, SAD surgery for SAPS is considered low-value care, a term used to refer to treatment or tests where there is little or no benefit for patients or more potential harm than benefit, and a strong international recommendation has been formulated against its use[14]. Multiple studies previously investigated trends in worldwide use of SAD surgery for SAPS[13, 15-21]. Decreasing trends have been reported in various countries, such as the Netherlands, Finland, Scotland and the United States(US), but increasing trends were observed in Australia, the United Kingdom(UK) and the US[13, 15-22]. No studies have examined trends in SAD surgery beyond 2017, whereas two high-quality RCTs were published in 2018 that may have impacted routine clinical practice[4, 6].

Exploring how publication of high-quality evidence may influence clinical decision-making in routine clinical practice has received limited attention in orthopaedic literature[23]. Timely implementation of evidence is of vital importance for both healthcare providers and patients, as performing low-value procedures does not provide the patient with the best treatment and contributes to rising healthcare costs[24]. The studies by Beard- and Paavola et al. were the first two placebo-controlled trials and formed the foundation for the strong international recommendation against SAD surgery by a panel assembled by the British Medical Journal[14]. It is, however, unknown to what extent publication of these RCTs has changed previous trends in SAD surgery in daily practice. Therefore, the aim of this study was to evaluate the extent to which publication of these high-impact RCTs in 2018 was associated with changes in the absolute volume or trend in monthly SAD surgeries in hospitals from different countries.

METHODS

Study Design

A controlled interrupted time series(ITS) design was used, which is a powerful quasi-experimental approach to evaluate effects of an intervention implemented at a clearly defined time point[25-29] and previously shown to give concordant results as those from a cluster RCT[30]. By comparing the trend before and after intervention, the intervention effect can be estimated by a change in absolute level and/or change in trend[26]. A change in trend represents a gradual change in daily practice following an intervention, whereas a change in level constitutes a more abrupt effect[31]. Given the importance of the two trials published in 2018 for subsequent recommendations, we used the publication month of the first published RCT(01/2018) as

the intervention time. We compared the volume of monthly SAD surgeries before(01/2016–01/2018), with that after the intervention(02/2018–02/2020).

Pseudonymised patient data from the Global Heath Data@Work (GHD@Work) collaborative were used, in which hospitals from various countries share their experiences and compare their outcomes using routinely collected administrative admission data. Data on clinical admissions and day case surgeries) were used for patients from six hospitals in five countries (Australia, Belgium, Netherlands, UK, US). Participating hospitals (Appendix A) are large academic medical centres, that are likely comparable with regard to their (complex) patient population. Within the collaborative, diagnoses and procedures were combined into groups and comorbidities in the Elixhauser comorbidity index[32], which were matched across countries to reconcile the different coding systems being used, as done in previous studies[33].

Patients and definitions

The study population included all patients aged 18+ years with a primary or secondary diagnosis potentially indicating SAPS, who underwent surgery in participating hospitals between 01/2016 and 02/2020. We excluded data from 03/2020 onwards as the number of surgeries was likely affected by the COVID-19 pandemic which would violate one of the key assumptions for the ITS (i.e. the intervention occurred independently of other changes over time)[26]. SAD procedures were identified using a combination of diagnosis and procedure codes. First, all clinical patient admissions and day case surgeries with a possible SAPS diagnosis were selected based on their primary or secondary diagnosis, using the following ICD-10 codes: M75.1-Rotator Cuff Syndrome, M75.2-Bicipital Tendinitis, M75.3-Calcific tendinitis of shoulder, M75.4-Impingement syndrome of shoulder, M75.5-Bursitis of shoulder. Within this patient selection, we selected those with SAD procedure codes. As hospitals from different countries used different coding systems for procedures, these were harmonized across countries to reconcile the differences between coding systems used. To ensure that we would capture local coding practices, we asked experts from participating hospitals to verify the diagnostic and procedure codes that were used to identify this patient group before seeing the results, or that some codes were not used, incorrect or missing (Appendix B).

As control group, we included all other patients likely to be treated by orthopaedic surgeons for musculoskeletal problems to control for potential confounding effects (e.g. other interventions/events occurring during the study period affecting surgery volumes such as a new hospital policy)[34]. The control group was represented by all patients who underwent a procedure within the ICD-10 clusters 'Diseases of the musculoskeletal system and connective tissue disease' or 'Injury and poisoning' (MSK clusters; Appendix B), excluding SAPS patients, as these clusters will capture most musculoskeletal patients.

It is possible that a change in performed SAD surgeries is accompanied by a shift towards other procedures, either a true change or merely in coding practice among clinicians, for instance if they have strong beliefs that SAD surgery may benefit their patients. Therefore, a

sensitivity analysis was carried out to examine changes in performed procedures within the following groups: 1) Any other performed orthopaedic procedure in SAPS patients (SAPS–Other procedures) reflecting a possible shift in procedure coding. Since patients with SAPS as a secondary diagnosis could undergo procedures to treat e.g. cardiac comorbidity, we only included patients within the beforementioned MSK clusters. 2) SAD surgeries in patients with any other diagnosis code than SAPS (NonSAPS–SAD) reflecting a possible shift in diagnosis coding.

Statistical analysis

First, monthly volumes of admissions and procedures were examined for every hospital to gauge the size of the hospital and the musculoskeletal department for 1) all patients, 2) patients within the MSK clusters, and 3) volume of procedures. Parametric continuous data were described using means, standard deviation (SD) and 95% confidence intervals (CI) and nonparametric data were expressed in medians and interquartile ranges. Categorical data were presented by numbers and percentages.

A segmented Poisson regression model with random intercept for hospital was used to assess changes in level and/or trend of monthly volume of SAD surgeries before (25 Data points) and after (25 Data points) publication of the first RCT[31]. A separated controlled design was used to compare the intervention group with the control group[34]. The same analysis was done for each individual hospital and for the sensitivity analyses. The Kolmogorov-Smirnov test was used to ensure our data followed a Poisson distribution, and robust standard errors to safeguard against any mild violations of regression assumptions[35]. Negative binomial regression was used for over-dispersed count data.

The following equation was used to estimate the changes in level and/or trend associated with publication of the high-quality RCTs (the intervention): $Y_t = \beta_0 + \beta_1$*Time(months) + β_2*Intervention + β_3*Time after intervention + e_t. Here, Y_t is the number of procedures, β1 estimates the pre-intervention trend, while β2 estimates the change in level directly following the intervention and β3 indicates the change in trend following the intervention. A random intercept was included to take into account between-hospital differences in the volume of surgeries, reflecting e.g. different hospital size.

We evaluated stationarity using the augmented Dicky-Fuller and KPSS tests, tested for first order autocorrelation using the Durbin-Watson test and higher order autocorrelations and/or seasonality using (partial) autocorrelation function plots. In case of non-stationarity, data were differenced. No autocorrelation or seasonality was found in the time series. Stata Version 17.1 (Stata-Corp LLC, USA) was used for analysis. Significance was established at $P < 0.05$.

RESULTS

Hospital monthly volumes in patients and procedures are presented in Table 1. A total of 3.046 patients undergoing SAD procedures in six hospitals across five countries were included, with 1.601 performed before and 1.445 after publication of the RCTs. One hospital did not perform any SAD surgeries during this period and thereby did not contribute to further analysis. Characteristics of patients undergoing SAD surgery are shown in Table 2, showing considerable variation across hospitals. For instance, patients were older in one US hospital, whereas patients less often had comorbidities and were less often treated in day case surgery in the Australian hospital. The readmission rate varied between 0.1% and 4.4%.

Figure 1 shows wide variation in volume of SAD surgeries (indicated by the data points), reflecting the different size of hospitals and/or musculoskeletal departments. Adjusting for clustering of patients within hospitals, there was no significant trend in volume of SAD surgeries before publication of the RCTs (Incidence rate ratio (IRR): 1.006 [0.996-1.017]; $P = 0.221$). Publication of the RCTs was not associated with an abrupt change in volume (IRR: 0.943 [0.824-1.079]; $P = 0.393$) but was significantly associated with a change in trend towards 2% fewer SAD surgeries on average per month (IRR: 0.984 [0.971-0.998]; $P = 0.021$), i.e. 18% fewer surgeries per year (0.984^{12}). Within the control group, there was no significant pre-publication trend (IRR: 1.000 [0.992-1.007]; $P = 0.939$) and no significant association between publication of the RCTs with any changes in level (IRR: 0.998 [0.936-1.063]; $P = 0.940$) or trend (IRR: 1.002 [0.993-1.012]; $P = 0.645$) (Figure 1).

Given the wide variation in volumes of SAD surgeries (Table 1), we also examined the trends for individual hospitals as there may have been contrasting trends that could level out in an overall analysis (Figure 2). This analysis showed that the association with a changing trend towards reduced volume of SAD surgeries was shown for 4 of 5 hospitals, albeit only significant in the Australian (IRR: 0.948 [0.911-0.987]; $P = 0.009$) and Belgium (IRR: 0.968 [0.939-0.999]; $P = 0.041$) hospitals. One US hospital showed a significantly increasing pre-publication trend (IRR: 1.020 [1.004-1.036]; $P = 0.017$) with publication of the RCTs not associated with any significant change in level or trend, i.e. it continued to increase. In the control group, volumes of procedures increased in the Australian hospital before publication of the RCTs (IRR: 1.002 [1.000-1.005]; $P = 0.026$). Publication of the RCTs was associated with a significant change in level (IRR: 0.931 [0.885-0.978]; $P = 0.004$), but not with a changing trend i.e. it continued to increase *(IRR:1.001 [0.988-1.004]; $P = 0.406$)*. No significant associations with changes in level and/or trend were found for the other hospitals (Appendix C).

Potential shifts towards other procedures

The results from the sensitivity analysis are shown in Figure 3. Within the SAPS-Other group, there was a significantly decreasing overall trend of about 2% per month before publication of the RCTs (IRR:0.985 [0.982-0.989]; *$P < 0.001$).* Publication of the RCTs was not associated

with a significant change in level (IRR: 1.037 [0.938-1.147]; *P = 0.474)* but was associated with significant increase of 2% per month in other procedures within SAPS patients (IRR:1.019 [1.004-1.034]; *P* = 0.014). The most frequently performed procedures within the SAPS-Other group included repair of shoulder tendon, excision of shoulder tendon and replacement of the shoulder joint. When examining this further within individual hospitals, the association with an increased trend of other procedures within SAPS patients was seen in 4 of 5 hospitals, although significance was only reached in the UK (IRR: 1.049 [1.013-1.085]; *P* = 0.007) and one US hospital (IRR: 1.031 [1.001-1.063]; *P* = 0.042)(Appendix C).

Within the NonSAPS-SAD group, there was no significant overall pre-publication trend (Figure 3). Publication of the RCTs was associated with a significant change in level (IRR:1.329[1.179-1.497]; *P <0.001*), but not with any significant changes in trend (Figure 3). For individual hospitals, the numbers of performed procedures for the NonSAPS-SAD were low (data not shown).

Table 1. Hospital monthly volumes of patients* and procedures between 01/2016 – 02/2020.

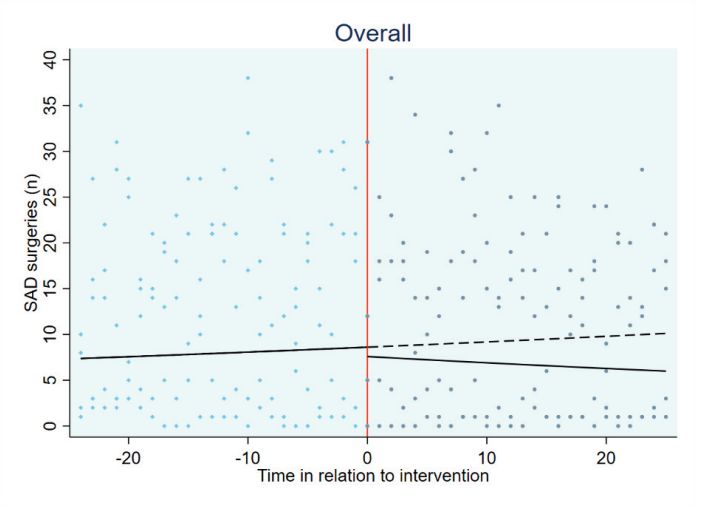
	Australia	*Belgium*	*The Netherlands*	*United Kingdom*	*United States (1)*	*United States (2)*
Median monthly volume of patients (IQR)	9.448 (9.072-9.779)	10.251 (10.046-10.634)	3.641 (3.532-3.797)	12.398 (11.968-12.880)	15.835 (14.618-16.370)	5.646 (5.272-5.967)
Median monthly volume of patients within MSK clusters** (IQR)	1.842 (1.773-1.907)	1.160 (1.110-1.235)	345 (326-364)	1.703 (1.644-1.780)	2.530 (2.383-2.616)	684 (634-750)
Median monthly volume of procedures (IQR)	7.882 (7.628-8.133)	6.705 (6.492-7.082)	948 (903-999)	8.963 (8.559-9.344)	6.788 (6.453-7.064)	2.399 (2.302-2.480)
Median monthly volume of procedures within MSK clusters** (IQR)	1.464 (1.419-1.538)	1.039 (991-1104)	145 (135-155)	1.348 (1.305-1.417)	1.221 (1.145-1.291)	591 (550-616)
Median monthly volume of subacromial decompressions (IQR)	3 (1-4)	17 (12-24)	0	22 (18-27)	1 (0-1)	17 (14-21)

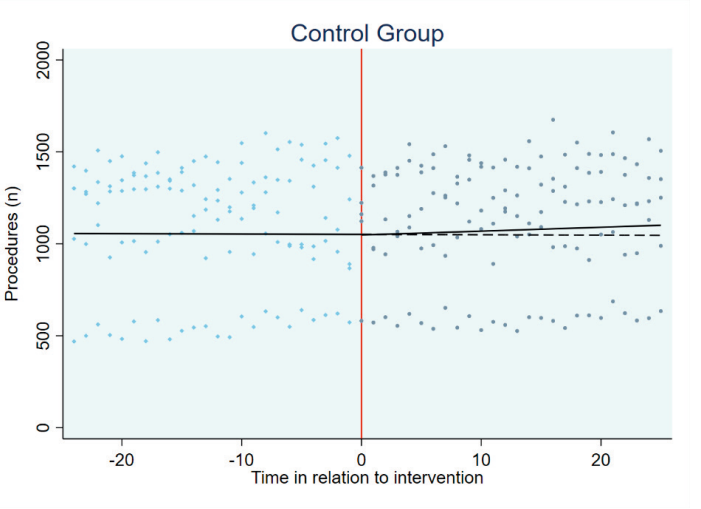
*Includes clinical admissions and day case surgeries. **Monthly volume of patients within the ICD-10 clusters: 'Diseases of the musculoskeletal system and connective tissue disease' and 'Injury and poisoning', capturing most musculoskeletal (MSK) patients. Abbreviations: IQR= interquartile range.

Table 2. Characteristics of patients undergoing subacromial decompression*.

	Australia (n=145)	***Belgium*** (n=907)	***United Kingdom*** (n=1102)	***United States (1)*** (n=45)	***United States (2)*** (n=847)
Mean age (SD)	58,8 (11,9)	55,9 (10,6)	57,2 (10,5)	62,3 (12,2)	55,8 (12,8)
% Female	48,3%	56,3%	48,9%	57,8%	40,4%
*Comorbidities***					
_% ≥1 Comorbidities	23,4%	56,9%	65,7%	57,8%	53,0%
_% Diabetes Mellitus	9,0%	6,2%	13,4%	17,8%	10,7%
_% Hypertension	2,8%	19,6%	27,9%	44,4%	32,1%
_% Obesity	0%	37,2%	35,6%	8,9%	5,8%
_% Pulmonary	0,7%	5,1%	15,3%	13,3%	10,3%
Median number of comorbidities (IQR)	0 (0-0)	0 (0-1)	0 (0-1)	1 (0-2)	0 (0-1)
Median LOS (IQR)	1 (1-1)	1 (0-1)	0 (0-0)	2 (0-7)	0 (0-0)
% Day case surgeries	1,4%	50,6%	85,7%	46,7%	100%
Readmission rate***	2,8%	3,1%	2,5%	4,4%	0,1%

*No subacromial decompressions were performed in the Dutch hospital. ** According to the Elixhauser Comorbidity index, only the most prevalent comorbidities are shown. ***Readmission rate within 30 days after discharge. Abbreviations: SD= Standard Deviation, LOS= Length of Stay, IQR= Interquartile Range.



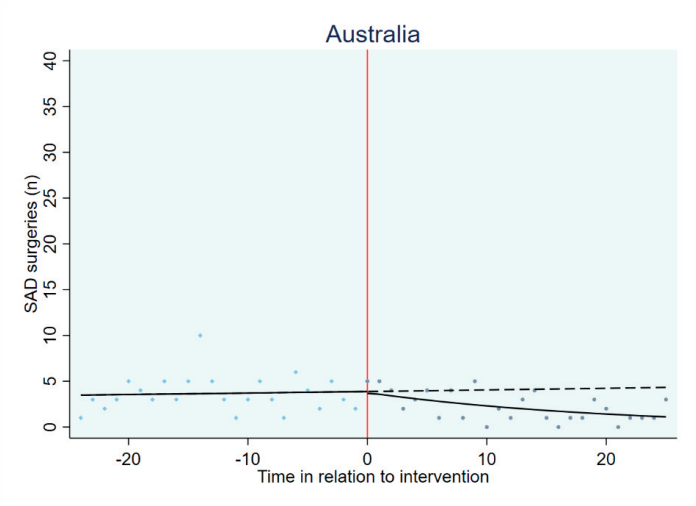


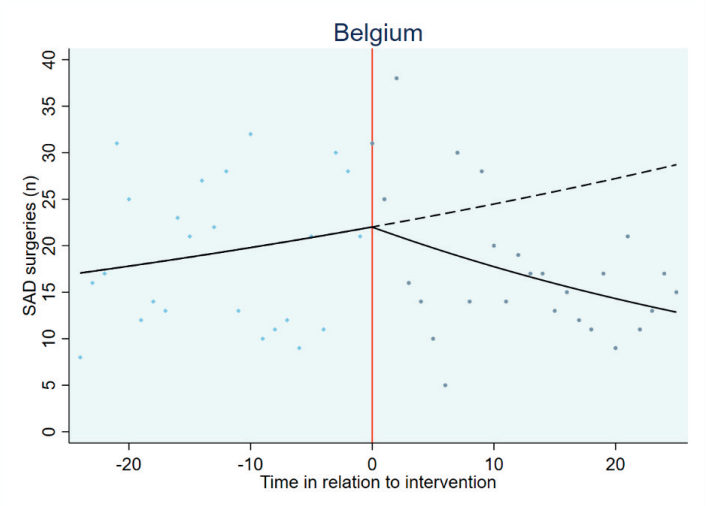
	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	1.006 (0.996-1.017)	*0.221*
Level change (β2)	0.943 (0.824-1.079)	*0.393*
Trend change (β3)	0.984 (0.971-0.998)	***0.021***
Constant (β0)	8.046 (2.3-28.3)	*0.001*

	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	1.000 (0.992-1.007)	*0.939*
Level change (β2)	0.998 (0.936-1.063)	*0.940*
Trend change (β3)	1.002 (0.993-1.012)	*0.645*
Constant (β0)	1051 (771-1432)	*<0.001*

Figure 1. Outcomes overall SAPS group and control group.

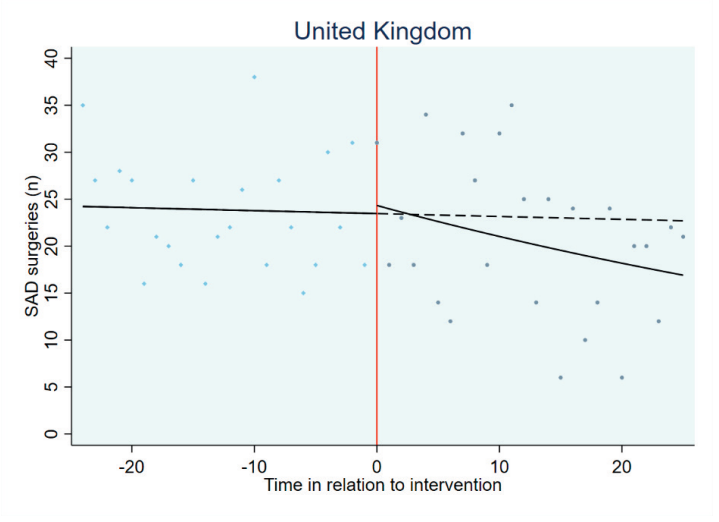
Figure 1. shows the fitted trend lines and regression coefficients, adjusted for clustering of patients within hospitals, of the number of monthly SAD surgeries (left; SAPS group) and other orthopaedic procedures (right; control group) before and after publication of the RCTs (time in relation to intervention represented in *months*). The dashed lines represent the fitted trend lines post-intervention as if the RCTs had not been published. Significant values are presented in bold. Abbreviations: SAD= subacromial decompression, IRR= incidence rate ratio, 95%CI= 95% Confidence Interval.

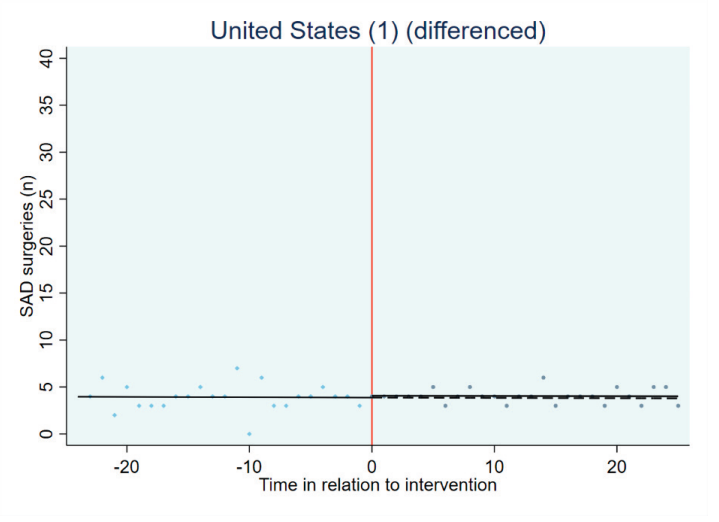




	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	1.004 (0.982-1.027)	*0.704*
Level change (β2)	0.969 (0.590-1.590)	*0.901*
Trend change (β3)	0.948 (0.911-0.987)	***0.009***
Constant (β0)	3.9 (2.8-5.3)	*<0.001*

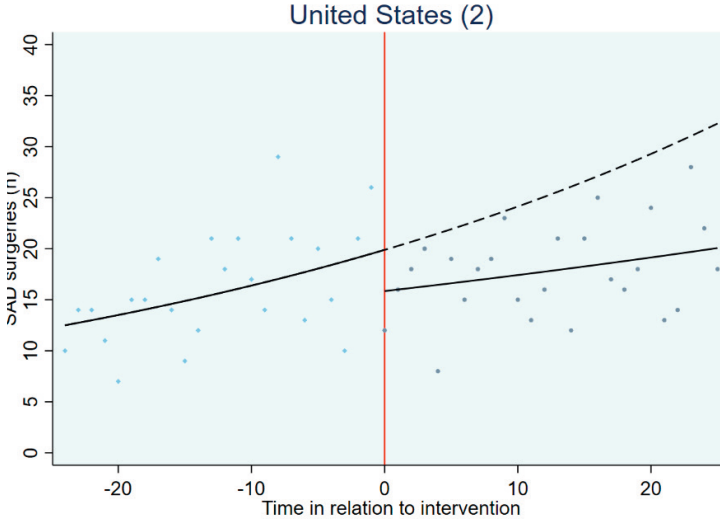
	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	1.011 (0.990-1.032)	*0.319*
Level change (β2)	0.999 (0.617-1.618)	*0.997*
Trend change (β3)	0.968 (0.939-0.999)	***0.041***
Constant (β0)	22.0 (16.5-29.5)	*<0.001*





	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	0.999 (0.984-1.013)	*0.857*
Level change (β2)	1.037 (0.738-1.457)	*0.836*
Trend change (β3)	0.987 (0.965-1.009)	*0.250*
Constant (β0)	23.5 (19.1-29.0)	*<0.001*

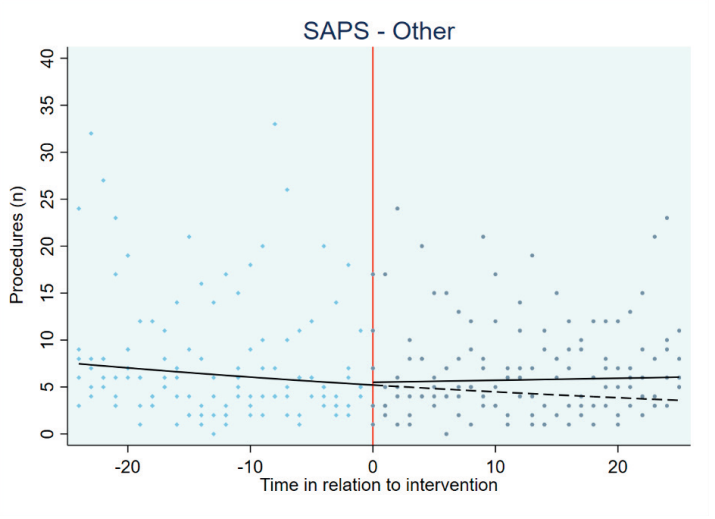
	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	0.999 (0.984-1.015)	*0.991*
Level change (β2)	1.050 (0.826-1.334)	*0.691*
Trend change (β3)	1.000 (0.982-1.019)	*0.973*
Constant (β0)	3.9 (3.204.8)	*<0.001*

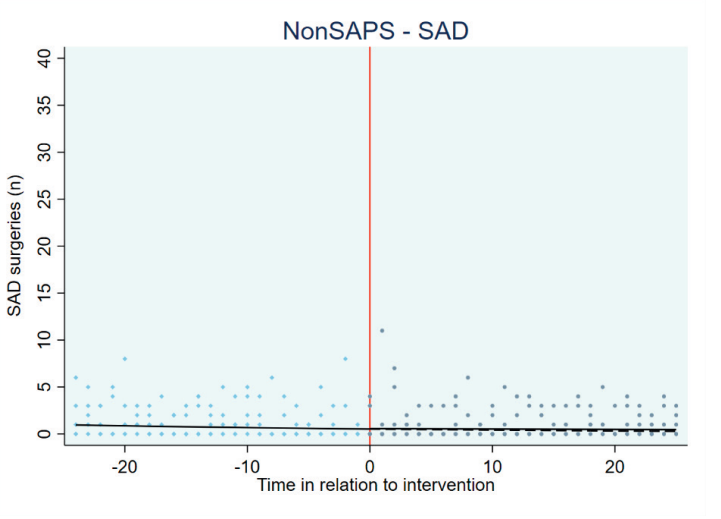


	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	1.020 (1.004-1.036)	***0.017***
Level change (β2)	0.797 (0.585-1.085)	*0.150*
Trend change (β3)	0.990 (0.970-1.011)	*0.342*
Constant (β0)	19.9 (15.5-24.4)	*<0.001*

Figure 2. Outcomes SAPS group- individual hospitals.

Figure 2 shows the fitted trend lines and regression coefficients of the number of monthly SAD surgeries within the individual hospitals before and after publication of the RCTs (time in relation to intervention represented in *months*). The dashed lines represent the





	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	0.985 (0.982-0.989)	***<0.001***
Level change (β2)	1.037 (0.938-1.147)	*0.474*
Trend change (β3)	1.019 (1.004-1.034)	***0.014***
Constant (β0)	5.3 (2.9-9.6)	*<0.001*

	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	0.976 (0.946-1.006)	*0.112*
Level change (β2)	1.329 (1.179-1.497)	***<0.001***
Trend change (β3)	1.016 (0.965-1.068)	*0.549*
Constant (β0)	0.4 (0.1-2.5)	*0.350*

Figure 3. Outcomes SAPS-Other group and NonSAPS-SAD group.

Figure 3 shows the fitted trend lines and regression coefficients, adjusted for clustering of patients within hospitals, of the number of monthly procedures within the SAPS-Other group (left) and NonSAPS-SAD group (right) before and after publication of the RCTs (time in relation to intervention represented in *months*). The dashed lines represent the fitted trend lines post-intervention as if the RCTs had not been published. Significant values are presented in bold.

Abbreviations: SAD= subacromial decompression, IRR= incidence rate ratio, 95%CI= 95% Confidence Interval.

DISCUSSION

The present study has shown that publication of the high-quality RCTs by Beard- and Paavola et al. in 2018[4, 6] was associated with a significantly reduced overall trend in use of SAD surgery of on average 2% per month (i.e. 18% per year), although the effect varied between hospitals. This association with a reduced trend in SAD surgery was shown for 4 of the 5 hospitals, albeit significant only in the Australian and Belgium hospitals, and was not seen in the control group. Sensitivity analysis showed that publication of the RCTs was also associated with a concurrent 2% monthly increased trend towards other procedures within SAPS patients and with an abrupt increase in volume of SAD surgeries in the Non-SAPS group.

The strength of the present study is that we used a controlled ITS design, a strong quasi-experimental design, that can estimate the effects of an intervention in a natural experimental setting with the control group taking into account any other interventions influencing the volume of surgeries[31]. Furthermore, all diagnosis and procedure data were harmonized to reconcile differences between coding systems. The hospitals were large academic centres, which provided a unique opportunity to evaluate the effect of evolving evidence on daily practice across different countries. Limitations of our study include the use of administrative data which could be subject to both over- or under-coding of patient characteristics such as more comorbidities, where for instance US hospitals may have higher occurrence of comorbidities due to financial incentives associated with coding. However, reimbursement of health services in Australia also depends on clinical coding, yet showed lower frequency of comorbidity in the current study. Particularly since we examined volumes of SAD surgery without adjusting for differences in patient-mix, this is unlikely to explain our results. Secondly, it is important to note that the study findings are only based on limited number of hospitals. As each centre was a large academic hospital, the included hospitals are broadly comparable but may differ from other(non-academic) hospitals in the selected countries, thus limiting the generalizability of our results to academic hospitals. Thirdly, no data on outpatient visits were available for analysis making it impossible to explore changes in the percentage of SAPS patients receiving SAD surgery. However, since the main outcome of interest was the volume of SAD surgeries which are performed as a day case surgery or require a hospital admission, it seems unlikely to have affected our results. Lastly, other interventions than the publication of the RCTs (e.g. payment policy- or guideline changes occurring around the same time) may have influenced clinician behaviour with regard to SAPS patients. However, we are unaware of other interventions during the period of interest and discussion among collaborating hospitals also did not suggest any simultaneous interventions.

Comparison with Literature

To our knowledge this is the first study that evaluates whether publication of the two placebo-controlled RCTs on treatment for SAPS in 2018 were associated with a change in existing trends in SAD surgery in hospitals from different countries. Various studies have investigated trends in earlier time periods when other RCTs showing on the effectiveness of SAD surgery

were published[13, 18, 22]. A Finnish study reported a declining trend in volume of SAD surgery starting in 2007, but this was two years after the RCT by Haahr et al.[7] was published, so that it is unclear whether the decline was associated with publication of that RCT or something else. In the UK, a slight decrease in the number of SAD surgeries was observed after 2011/2012, two years after publication of the RCTs by Henkus- and Ketola et al. and also the starting year of the CSAW trial which eventually led to the publication by Beard in 2018[4, 8, 9, 16]. A Scottish study found a decline in the use of SAD surgery starting in 2017, but this was one year before the RCT by Beard was published[22] and therefore unclear whether the decline is associated with publication of this RCT or due to the rising tide phenomenon[36]. Lastly, a decreasing trend was observed in the Netherlands, following a clinical practice guideline implementation in 2012 that advocated against SAD surgery, but lack of data for the period before guideline implementation made evaluation impossible[20]. Results of the present study therefore add to this literature that a change in trend is associated with publication of high-quality evidence.

Two studies describing decreasing trends in SAD surgeries showed a simultaneous increase in other procedures(e.g. rotator cuff surgery, acromioclavicular-joint excision), suggesting a shift in coding patterns[19, 22]. Our sensitivity analyses also showed that publication of the RCTs was associated not only with a change towards a reduced trend in use of SAD surgery in SAPS patients, but also with an increased trend in other procedures among SAPS patients, and an abrupt increase in the use of SAD surgery for Non-SAPS patients. Therefore, only evaluating the total number of SAD surgeries could create a distorted picture how research results affect daily practice, if a decline of a surgical procedure is accompanied by a shift in coding practices rather than not performing the procedure at all.

Interpretation and Clinical Implications

The results of this study suggest that publication of high-quality RCTs can change clinical practice. Even though statistical significance does not equal clinical relevance, we believe our results are relevant because of the strong recommendation against the use of SAD surgery for SAPS, so that every reduction in the use of this low-value care procedure is important. However, we cannot rule out the possibility that there has been a concurrent shift in coding practice given that publication of the RCTs was associated both with an overall 2% reduction in trend in SAD surgeries but also a 2% increase in other procedures among SAPS patients. Rather than a reduction of care providing no benefit for patients, it may indicate substitution towards other surgical procedures. The use of a control group provided stronger evidence to support the publication of the RCTs really causing the observed changes in trends. We also showed large variation in effect between hospitals from various countries, suggesting that the uptake of evolving evidence differs significantly between healthcare providers potentially influenced by different reimbursement for healthcare services. Additionally, SAPS is an umbrella diagnosis, covering a large heterogeneous group of shoulder problems with unknown aetiology and despite high-quality evidence showing no benefit of SAD surgery for SAPS patients, clinical guidelines remain unclear on the best alternative(non-surgical) treatment[14]. This leaves the clinicians with uncertainty about the best alternative treatment

and might introduce action bias, the general preference for active over passive treatment in clinical decision-making[37, 38]. All of these factors highlight the complexities of implementing such international recommendations in daily practice even if based on strong evidence, and more research is needed to understand which factors influence the uptake of evidence to change clinical practice towards reducing low-value care and to improve quality of care.

The presented case of SAD surgery for SAPS can be viewed as an example to explore the relationship between evolving evidence and changes in clinical practice in various countries. Similar study designs can be used to evaluate and monitor the effect of clinical guidelines or research evidence on daily practice for other procedures considered to have no or little benefit for patients. Reducing low-value care is of vital importance to protect patients from harm and to lower the financial burden on healthcare systems. International campaigns have been launched that aim to improve the quality of care by reducing low-value care. Quick dissemination of new evidence into clinical practice is in line with these international campaigns and can be done in the context of collaboratives, which are considered an effective approach to shared learning and improvements in the quality of care[39]. Our results illustrate the value of such collaboratives to compare clinical practice and to use observed variation as a starting point to enable improvements in quality of care.

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APPENDIX A: Hospitals participating in the Global Health Data @ Work (GHD@Work) collaborative.

Country	Hospital
Australia	Alfred Health Hospital Melbourne
Belgium	University Hospital Leuven
The Netherlands	Leiden University Medical Centre (LUMC)
United Kingdom	University Hospital Coventry
United States (1)	Keck Medical Center of the University of Southern California
United States (2)	Hackensack Meridian Health

Appendix B: Used diagnosis & procedure codes and CCS groups.

1. Used diagnosis and procedure codes to identify subacromial decompression (SAD) surgery per coding system.

Used diagnosis codes:

ICD-10 Code	*Description*
M75.1	Rotator cuff syndrome
M75.2	Bicipital tendinitis
M75.3	Calcific tendinitis of shoulder
M75.4	Impingement syndrome of shoulder
M75.5	Bursitis of shoulder

In combination with any of the following procedure codes:

A. ACHI (Australia)

ACHI CODE	Description
47969	TENOSYNOVECTOMY.
48900	SHOULDER, excision of coraco-acromial ligament or removal of calcium deposit from cuff or both.
48903	SHOULDER, decompression of subacromial space by acromioplasty, excision of coraco-acromial ligament and distal clavicle, or any combination.
48906	SHOULDER, repair of rotator cuff, including excision of coraco-acromial ligament or removal of calcium deposit from cuff, or both.
48909	SHOULDER, repair of rotator cuff, including decompression of subacromial space by acromioplasty, excision of coraco-acromial ligament and distal clavicle, or any combination.
48936	SHOULDER, synovectomy of, as an independent procedure
48948	SHOULDER, arthroscopic surgery of, involving any 1 or more of: removal of loose bodies; decompression of calcium deposit; debridement of labrum, synovium or rotator cuff; or chondroplasty.
48951	SHOULDER, arthroscopic division of coraco-acromial ligament including acromioplasty
48954	SHOULDER, arthroscopic total synovectomy of, including release of contracture when performed
48960	SHOULDER, reconstruction or repair of, including repair of rotator cuff by arthroscopic, arthroscopic assisted or mini open means; arthroscopic acromioplasty; or resection of acromioclavicular joint by separate approach when performed
50104	JOINT, synovectomy

B. ICD10-PCS (Belgium and United States)

ICD10-PCS Code	Description
0RBG0ZZ	Excision of Right Acromioclavicular Joint, Open Approach
0RBH0ZZ	Excision of Left Acromioclavicular Joint, Open Approach
0RBG4Zz	Excision of Right Acromioclavicular Joint, Percutaneous Endoscopic Approach
0RBH4ZZ	Excision of Left Acromioclavicular Joint, Percutaneous Endoscopic Approach
0RBJ0ZZ	Excision of Right Shoulder Joint, Open Approach
0RBK0ZZ	Excision of Left Shoulder Joint, Open Approach
0RBJ4ZZ	Excision of Right Shoulder Joint, Percutaneous Endoscopic Approach
0RBK4ZZ	Excision of Left Shoulder Joint, Percutaneous Endoscopic Approach
0RNJ0ZZ	Release Right Shoulder Joint, Open Approach
0RNK0ZZ	Release Left Shoulder Joint, Open Approach
0RNJ4ZZ	Release Right Shoulder Joint, Percutaneous Endoscopic Approach
0RNK4ZZ	Release Left Shoulder Joint, Percutaneous Endoscopic Approach
0RTG0ZZ	Resection of Right Acromioclavicular Joint, Open Approach
0RTH0ZZ	Resection of Left Acromioclavicular Joint, Open Approach
0MB10ZZ	Excision of Right Shoulder Bursa and Ligament, Open Approach
0MB20ZZ	Excision of Left Shoulder Bursa and Ligament, Open Approach
0MB14ZZ	Excision of Right Shoulder Bursa and Ligament, Percutaneous Endoscopic Approach
0MB24ZZ	Excision of Left Shoulder Bursa and Ligament, Percutaneous Endoscopic Approach
0MB90ZZ	Excision of Right Upper Extremity Bursa and Ligament, Open Approach
0MBB0ZZ	Excision of Left Upper Extremity Bursa and Ligament, Open Approach
0MB94ZZ	Excision of Right Upper Extremity Bursa and Ligament, Percutaneous Endoscopic Approach
0MBB4ZZ	Excision of Left Upper Extremity Bursa and Ligament

C. OPCS (United Kingdom)

OPCS Code	Description
O29.1	Subacromial decompression
T62.1	Total excision of bursa
Y52.8	Other specified approach to organ through other opening
Y76.7	Arthroscopic approach to joint
W84.4	Endoscopic decompression of joint
W84.8	Other specified therapeutic endoscopic operations on other joint structures

D. CBV (the Netherlands)

CBV Code	Description
338100	SCHOUDER- ACROMIONRESECTIE- PARTIEEL
338100A	SCHOUDER - ACROMIONRESECTIE -PARTIEEL -LINKS
338100B	SCHOUDER - ACROMIONRESECTIE -PARTIEEL -RECHTS
338100C	SCHOUDER -ACROMIONRESECTIE
338100D	SCHOUDER -ACROMIONRESECTIE VLGS NEER VIAARTROSCOPIE
338100E	SCHOUDER -ACROMIONRESECTIE-PARTIEEL VIAARTROTOMIE
338100F	SCHOUDER - ACROMIONRESECTIE -PARTIEEL VIA ARTROSCOPIE
338100L	SCHOUDER -ACROMIONRESECTIE-TOTAAL -LINKS
338100R	SCHOUDER -ACROMIONRESECTIE-TOTAAL -RECHTS
338114	SCHOUDER -NEER-ACROMIONPLASTIEK
338114A	SCHOUDER -NEER-ACROMIONPLASTIEK -LINKS
338114B	SCHOUDER -NEER-ACROMIONPLASTIEK -RECHTS
338146	SCHOUDER-ARM- EXCISIE LIGAMENT
338146C	SCHOUDER-ARM- EXCISIE LIGAMENT CORACO-ACROMIAL
338149	SCHOUDER - CAPSULOTOMIE
338149B	SCHOUDER - DECOMPRESSIE SUBACR.SUPRASPIN.PEES-SCOPIE
338149C	SCHOUDER - NETTOYAGE MBV ARTROSCOPIE
338170	SCHOUDER-ARM- EXCISIE AFW. SPIER-PEES-FASCIE-BURSA
338170C	SCHOUDER-ARM- RESECTIE SPIER-PEES-FASCIE-BURSA -LINKS
338170D	SCHOUDER-ARM- RESECTIE SPIER-PEES-FASCIE-BURSA -RECHTS
338170F	SCHOUDER-ARM- RESECTIE SPIER-PEES-FASCIE-BURSA
338179A	SCHOUDER -INGREPEN BURSA

2. CCS groups within the ICD-10 clusters 'Diseases of the musculoskeletal system and connective tissue disease' & 'Injury, poisoning and certain other consequences of external causes' ("MSK Clusters")

ICD-10 Cluster	*CCS Group*	*Description*
Diseases of the musculoskeletal system and connective tissue disease	*54*	*Gout and other crystal arthropathies*
	201	*Infective arthritis and osteomyelitis (except that caused by tuberculosis or sexually transmitted disease)*
	202	*Rheumatoid arthritis and related disease*
	203	*Osteoarthritis*
	204	*Other non-traumatic joint disorders*
	205	*Spondylosis; intervertebral disc disorders; other back problems*
	206	*Osteoporosis*
	207	*Pathological fracture*
	208	*Acquired foot deformities*
	209	*Other acquired deformities*
	210	*Systemic lupus erythematosus and connective tissue disorders*
	211	*Other connective tissue disease*
	212	*Other bone disease and musculoskeletal deformities*

2. CCS groups within the ICD-10 clusters 'Diseases of the musculoskeletal system and connective tissue disease' & 'Injury, poisoning and certain other consequences of external causes' ("MSK Clusters") (continued)

ICD-10 Cluster	*CCS Group*	*Description*
Injury, poisoning and certain other consequences of external causes	*225*	*Joint disorders and dislocations; trauma-related*
	226	*Fracture of neck of femur (hip)*
	227	*Spinal cord injury*
	228	*Skull and face fractures*
	229	*Fracture of upper limb*
	230	*Fracture of lower limb*
	231	*Other fractures*
	232	*Sprains and strains*
	233	*Intracranial injury*
	234	*Crushing injury or internal injury*
	235	*Open wounds of head; neck; and trunk*
	236	*Open wounds of extremities*
	237	*Complication of device; implant or graft*
	238	*Complications of surgical procedures or medical care*
	239	*Superficial injury; contusion*
	240	*Burns*
	241	*Poisoning by psychotropic agents*
	242	*Poisoning by other medications and drugs*
	243	*Poisoning by nonmedicinal substances*
	244	*Other injuries and conditions due to external causes*

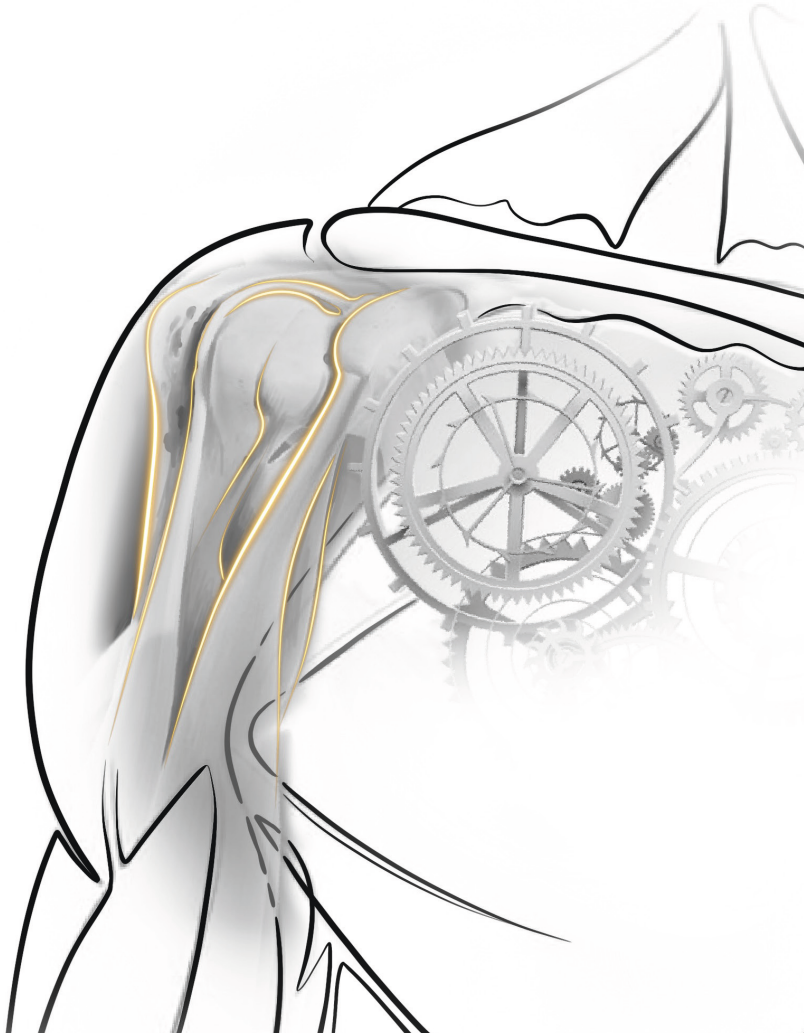
Appendix C. Additional results.

1. **Regression coefficients of the control group per health care provider.**

	Australia		**Belgium**		**United Kingdom**		**United States (1) (differenced)**		**United States (2) (differenced)**	
	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	1.002 (1.000-1.005)	***0.026***	1.001 (0.997-1.006)	*0.507*	1.002 (0.999-1.005)	*0.177*	1.002 (0.971-1.034)	*0.909*	0.995 (0.967-1.023)	*0.719*
Level change (β2)	0.931 (0.885-0.978)	***0.004***	1.002 (0.920-1.092)	*0.960*	1.036 (0.969-1.108)	*0.300*	1.053 (0.534-2.078)	*0.881*	0.992 (0.577-1.705)	*0.976*
Trend change (β3)	1.001 (0.988-1.004)	*0.406*	0.997 (0.992-1.003)	*0.300*	0.996 (0.992-1.000)	*0.072*	0.998 (0.957-1.040)	*0.920*	1.009 (0.972-1.048)	*0.649*
Constant (β0)	1507 (1453-1562)	*<0.001*	1042 (976-1113)	*<0.001*	1345 (1276-1417)	*<0.001*	179 (106-303)	*<0.001*	106 (75-150)	*<0.001*

2. **Regression coefficients of the SAPS-Other group per health care provider.**

	Australia		**Belgium**		**United Kingdom**		**United States (1)**		**United States (2)**	
	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*	IRR (95%CI)	*P-value*
Trend pre intervention (β1)	0.994 (0.969-1.020)	*0.636*	0.987 (0.970-1.004)	*0.124*	0.979 (0.956-1.004)	*0.099*	0.980 (0.955-1.006)	*0.137*	0.983 (0.956-1.010)	*0.205*
Level change (β2)	0.995 (0.581-1.704)	*0.985*	1.032 (0.725-1.469)	*0.863*	0.997 (0.556-1.787)	*0.992*	1.304 (0.847-2.009)	*0.228*	0.884 (0.479-1.633)	*0.695*
Trend change (β3	0.996 (0.958-1.035)	*0.819*	1.008 (0.983-1.034)	*0.535*	1.049 (1.013-1.085)	***0.007***	1.031 (1.001-1.063)	***0.042***	1.030 (0.990-1.071)	*0.141*
Constant (β0)	3.822 (1.568-3.780)	*<0.001*	15.389 (12.063-19.631)	*<0.001*	2.867 (1.928-4.264)	*<0.001*	4.839 (3.433-6.819)	*<0.001*	4.495 (2.874-7.031)	*<0.001*



Chapter 6

Substantial variation in decision making to perform subacromial decompression surgery for subacromial pain syndrome between orthopaedic shoulder surgeons for identical clinical scenarios: A case-vignette study.

Timon H. Geurkink MD[1,2], Perla J. Marang-van de Mheen PhD[2], Jochem Nagels MD[1], Ronald N. Wessel MD[3], Rudolf W. Poolman MD PhD[1], Rob G.H.H. Nelissen MD PhD[1], Leti van Bodegom-Vos MD PhD[2]

1. *Department of Orthopaedics, Leiden University Medical Centre, Postbus 9600, 2300 RC, Leiden, the Netherlands.*
2. *Department of Biomedical Data Sciences, Medical Decision Making, Leiden University Medical Center, Leiden, the Netherlands.*
3. *Department of Orthopaedics, Sint Antonius Hospital, Nieuwegein, the Netherlands.*

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ABSTRACT

Purpose:
To provide further insight into the variation in decision-making to perform subacromial decompression (SAD) surgery in patients with subacromial pain syndrome (SAPS) and its influencing factors.

Methods:
Between November 2021 and February 2022, we invited 202 Dutch Shoulder and Elbow Society members to participate in a cross-sectional Web-based survey including four clinical scenarios of SAPS patients. Scenarios varied in patient characteristics, clinical presentation, and other contextual factors. For each scenario, respondents were asked (1) to indicate whether they would perform SAD surgery, (2) to indicate the probability of benefit of SAD surgery (i.e., pain reduction), (3) to indicate the probability of harm (i.e., complications) and (4) to rank the five most important factors influencing their treatment decision.

Results:
A total of 78 (39%) respondents participated. The percentage of respondents who would perform SAD surgery ranged from 4 to 25% among scenarios. The median probability of perceived benefit ranged between 70 and 79% across scenarios for respondents indicating to perform surgery compared with 15 to 29% for those indicating not to perform surgery. The difference in median probability of perceived harm ranged from 3 to 9% for those indicating to perform surgery compared with 8 to 13% for those indicating not to perform surgery. Surgeons who would perform surgery mainly reported patient-related factors (e.g., complaint duration and response to physical therapy) as the most important factors to perform SAD surgery, whereas surgeons who would not perform surgery mainly reported guideline-related factors.

Conclusion:
Overall, Dutch orthopaedic shoulder surgeons are reluctant to perform SAD surgery for SAPS patients. There is substantial variation among orthopaedic surgeons regarding decisions to perform SAD surgery for SAPS even when evaluating identical scenarios, where particularly the perceived benefit of surgery differed between those who would perform surgery and those who would not. Surgeons who would not perform SAD surgery mainly referred to guideline-related factors as influential factors for their decision, whereas those who would perform SAD surgery considered patient-related factors more important.

INTRODUCTION

Subacromial pain syndrome (SAPS) is the most frequent diagnosis given to patients presenting with shoulder pain[1, 2]. Most SAPS patients are treated nonsurgically (e.g., glucocorticoid injections, physical therapy) but subacromial decompression (SAD) surgery can be performed when patients are not responding to nonsurgical treatment[3, 4]. SAD surgery is intended to reduce pain and improve shoulder function, but adverse effects may also occur (e.g., no pain reduction, infection, thromboembolism or frozen shoulder)[2, 5]. When considering surgery, orthopaedic surgeons must therefore carefully weigh the potential benefits of surgery against its potential harms[6].

Recent high-quality experimental studies found that SAD surgery provides no significant improvement in pain or functionality in SAPS patients when compared with placebo surgery or nonsurgical management[4, 7-9], whereas it still carries a risk of harm to patients. On the basis of these studies, a panel assembled by the British Medical Journal formulated a strong recommendation against SAD surgery for SAPS[2] and it is considered "low-value care" –a term referring to procedures with little or no benefit or more potential harm than benefit to patients. Nevertheless, SAD surgery is still frequently performed. In the United Kingdom, the United States and Australia increasing trends in SAD surgery for SAPS have even been reported[10-12]. Moreover, in the Netherlands, approximately 10.000 SAPS patients underwent SAD surgery in 2016[13]. Consequently, several initiatives have been launched to further reduce the use of SAD surgery for SAPS worldwide. In the Netherlands, activities such as clinical guideline changes are undertaken to reduce the use of SAD surgery, and in 2020, there was a withdrawal of reimbursement through a policy change (i.e., active disinvestment) by one of the large health care insurers.

To be effective, such initiatives to reduce low-value care procedures should address factors influencing surgeons' decisions to perform surgery. Previous studies showed large variation between surgeons in their clinical decision-making to perform surgery, but little is known about the factors that contribute to this variation[14-16]. These factors include differences in patient characteristics, surgeon characteristics, surgeons' perception of benefit and/or harm of surgical intervention, and surgeons' knowledge and interpretation of guidelines and financial constraints [6, 15, 17]. This study aimed to provide further insight into the variation in decision-making to perform SAD surgery for patients with SAPS and its influencing factors. Our hypothesis was that there would be substantial variation in clinical decision-making between individual orthopaedic surgeons.

METHODS

Study design

Between November 2021 and February 2022, we conducted a cross-sectional Web-based survey including four clinical scenarios among Dutch orthopaedic shoulder surgeons to

examine the variation in clinical decision-making to perform SAD surgery for SAPS. The study protocol (No. N20.127) was presented to the Medical Ethical Committee of Leiden University Medical Center(METC-LDD), which waived the need for ethical approval under Dutch law. All results are reported according to the Checklist for Reporting of Survey Studies(CROSS)[18].

Setting

From January 2020 onwards, one of the four largest Dutch health care insurers launched an active disinvestment initiative for SAD surgery in SAPS patients. This specific health care insurer decided to partially withdraw reimbursement for this procedure by contracting 30% fewer procedures than the preceding year in each hospital, based on (inter)national guidelines. This active disinvestment strategy was examined within the survey as one of the possible factors influencing clinical decision-making of orthopaedic surgeons regarding surgical treatment of SAPS.

Study population

All 202 members of the Dutch Shoulder and Elbow Society (DSES) were invited to participate in the survey, which was sent on November 30, 2021. Members of the DSES are either orthopaedic shoulder surgeons or orthopaedic residents with a specific interest in shoulder surgery. At the time our study, approximately 40 to 50 members were actively participating in DSES meetings. All members received a link for the survey by e-mail from the DSES. Two reminder e-mails were sent to all members after three weeks and six weeks respectively. Eligible participants were orthopaedic surgeons and orthopaedic surgery residents who, on average, treated at least one SAPS patient per month. To prevent multiple submissions by one respondent, the survey could only be filled in once for every unique IP address. Participation was voluntary and anonymous. Participants were asked to further disseminate the survey to colleagues involved in treating SAPS patients.

Survey development

Qualtrics software (Qualtrics, Provo, UT) was used to develop the survey and to perform data collection. A pilot study was carried out among fifteen individuals (i.e., orthopaedic surgeons, residents and researchers) to test the survey. The first part of the survey requested demographic information, including age, sex, current function (i.e., orthopaedic surgeon or resident), area of interest within orthopaedics, type of hospital (i.e., academic teaching hospital, non-academic teaching hospital, non-academic non-teaching hospital, independent treatment center), and the number of SAPS patients seen per month. The second part of the survey consisted of four hypothetical but realistic clinical scenarios regarding the treatment of a SAPS patient, each followed by four questions (described later; Figure 1). The last part of the survey investigated awareness and attitude of the respondents toward the active disinvestment strategy by the health care insurer described earlier by use of 7-point Likert scales. The translated survey can be found in Appendix S1.

Clinical scenarios

Four hypothetical clinical scenarios describing SAPS patients were developed to study the variation in clinical decision-making to perform SAD surgery (see Figure 1). The clinical scenarios consisted of a short paragraph and varied regarding patient characteristics, clinical presentation, the outcomes of imaging tests, and other contextual factors (e.g., reimbursement status of SAD surgery). The clinical scenarios were developed by multiple orthopaedic surgeons (JN, RW, RP, RN) to ensure these were realistic for clinical cases seen in orthopaedic practice, but they were deliberately created such that there may be variation in decision-making regarding whether to perform surgery or not.

Four questions accompanied each clinical scenario. The first question explored the decision whether or not to perform SAD surgery in the patient described in the clinical scenario. The second and third questions queried the probabilities of perceived benefit (i.e., pain reduction) and harm (i.e., complications) of SAD surgery on a scale from 0 to 100%. Finally, respondents were asked to select and rank the five most important factors affecting their clinical decision-making to perform SAD surgery or not. These factors could be selected from a predefined list (Appendix S1) and included patient-related factors (e.g., characteristics of patients and their clinical presentation), guideline-related factors such as whether surgical treatment was indicated, and other contextual factors such as the reimbursement status of SAD surgery in the hospital where the patient was treated. Partially filled-in surveys were included in the analysis if at least one clinical scenario was completed.

Statistical analysis

Parametric continuous data were described using means, standard deviations (SD) and 95% confidence intervals (CI), whereas nonparametric data were expressed in medians and interquartile ranges (IQRs). Numbers and percentages were used to present categorical data. First, the proportion of respondents who decided to perform SAD surgery was calculated for each clinical scenario to indicate the variation in clinical decision-making between scenarios. We then explored the association between respondent characteristics (i.e., age (per year), sex (female vs male), function (orthopaedic surgeon vs resident), years of experience as an orthopaedic surgeon and resident combined, type of hospital (teaching vs non-teaching) and the number of SAPS patients seen per month) and the decision to perform surgery or not across all clinical scenarios, using univariate logistic regression analysis with generalized estimating equations to adjust for clustering of scenarios within respondents. Factors with $P < .20$ were included in multivariate analysis to assess their independent effects. Because orthopaedic residents will inherently have less experience which may affect their decision-making, a sensitivity analysis was performed in which responses from residents were excluded.

Second, we evaluated whether the decision to perform SAD surgery or not was influenced by the perceived probabilities of benefit and harm for each of the scenarios. This was performed using a logistic model that included the logarithmically transformed benefit-harm (BH) ratio (i.e., the probability of perceived benefit divided by the probability of perceived harm) as an

independent variable to predict the probability of surgery and no surgery for each respondent per clinical scenario. These predicted probabilities were plotted against the BH ratio. Using these plots, we identified the break-even point, that is, the value of the BH ratio at which the predicted probabilities of performing SAD surgery and performing no SAD surgery were equal. Assuming that surgeons decide to operate when the perceived benefits outweigh the perceived harms, the probability of performing surgery can be expected to exceed the probability of not performing surgery when the BH ratio is greater than 1.

Finally, we used descriptive statistics to evaluate which factors were most important for the decision to perform SAD surgery or not, as well as the perceived effect of the active disinvestment strategy on clinical decision-making. Stata software (Version 17.1; StataCorp, College Station, TX) and SPSS software (version 20.0; IBM, Armonk, NY) were used for analysis. Significance was established at $P < .05$.

RESULTS

Of 202 invited members of the DSES, 78 (39%) participated in the study. Fourteen (18%) respondents did not complete the first clinical scenario, thus leaving 64 (82%) respondents for analysis. Of these, 57 (89%) completed all questions. The respondents who did not complete the first clinical scenario did not differ in demographic characteristics from the group included in the analysis (data not shown). Among respondents, 52 (81%) were orthopaedic surgeons and 12 (19%) were orthopaedic residents. Respondents had a mean age of 45 years (SD 9.4) and most (80%) were men. Most respondents (55%) worked in a non-academic teaching hospital. Table 1 shows the baseline characteristics of the respondents.

Decision to perform surgery

The decision to perform surgery varied among the four clinical scenarios. In the first clinical scenario ("58-year-old construction worker"), 8 respondents (13%) would perform SAD surgery. In the second ("48-year-old woman with previous SAD") and third ("51-year-old painter") clinical scenarios, thirteen respondents (22%) and fourteen respondents (25%), respectively, would perform SAD surgery, whereas only two respondents (4%) would perform surgery in the fourth clinical scenario ("36-year-old volleyball player"). None of the respondent characteristics was associated with the decision to perform SAD surgery (using $p < .20$ as the threshold (Appendix S2)) so that multivariate analysis was not conducted. The results remained the same when responses of orthopaedic residents were excluded in the sensitivity analysis (See Appendix S2).

Perceived benefits and harms of surgery

The median probabilities of perceived benefit across respondents varied from 15% to 36% among clinical scenarios, and the median probabilities of perceived harm ranged from 8% to 10%. An interesting finding was that the the median probability of perceived benefit for respondents who decided to perform SAD surgery ranged from 70% to 79% among clinical

scenarios compared with a range from 15% to 29% for those deciding not to perform surgery. Much smaller differences were observed in the probabilities of perceived harm, ranging from 3% to 9% among clinical scenarios for surgeons deciding to perform surgery and 8% to 13% for those who would not perform surgery (Table 2).

The BH ratio was significantly associated with the decision to perform surgery (OR 8.2 95%CI 3.7-18.1, P <.001). The break-even point of the BH ratio (i.e., the value of the BH ratio at which the predicted probabilities of surgery and no surgery were equal (50%)), was 32 and 33 for scenario 2 ("48-year-old woman with previous SAD") and 3 ("51-year-old painter") respectively (Figure 1). Thus, only when the perceived benefit is 32 or 33 times higher than the perceived harm will the predicted probability of receiving SAD surgery for SAPS exceed 50%. The break-even point was not calculated for the other scenarios given that few surgeons would perform surgery in these scenarios.

Factors influencing decision to perform surgery

Table 3 provides a list of the factors ranked as most important by respondents in their decision to perform surgery or not for each scenario. Among the respondents who decided to perform surgery, "the duration of complaints", "effectiveness of subacromial infiltration", "the outcomes of imaging tests", and "response to targeted physical therapy" were most frequently reported as the factors that were the most important for the decision to perform SAD surgery. In contrast, among respondents who decided not to perform surgery "surgical treatment not indicated", "the outcomes of imaging tests" and "other nonsurgical treatment better" were most frequently reported as factors important for the decision. The presence or absence of reimbursement (i.e., reimbursement status) of SAD surgery was scarcely reported as an important factor (range: 3-9% among clinical scenarios). Only 51% of the respondents were familiar with the active disinvestment strategy for SAD surgery by the health care insurer, of whom 18% could name the specific insurer implementing this strategy. Respondents who decided to perform SAD surgery for at least one clinical scenario reported that there was insufficient evidence to stop reimbursement for SAD surgery in SAPS patients (median 3,

IQR 2-3) on a Likert scale from 1 (absolutely insufficient evidence) to 7 (absolutely sufficient evidence), whereas surgeons deciding not to perform surgery believed that there was sufficient evidence (median 5, IQR 3-6).

Table 1. Characteristics of respondents who participated in study.

	Orthopaedic surgeons	Orthopaedic residents
N (%)	52 (81)	12 (19)
Mean age (SD)	48 (7.7)	32 (2.9)
% Female	15	33
Mean years' experience (SD)	12 (7.1)	*NA*
Median year of residency (IQR)	*NA*	5 (4-5)
Area of interest		
_% Shoulder	98	58
_% Elbow	58	25
_% Wrist and hand	27	0
_% Spine	0	0
_% Hip	14	33
_% Knee	31	67
_% Ankle and feet	4	8
_% Sport	29	25
_% Traumatology	39	58
_% Paediatrics	6	0
Type of hospital		
_% General-Teaching	50	75
_% General-Non teaching	31	8
_% Academic	2	17
_% Private	10	0
_% Other	8	0
Median SAPS patients per month (IQR)	48 (25-74)	10 (3-28)

IQR, interquartile range; NA, not applicable; SAPS, subacromial pain syndrome; SD, standard deviation; N, number.

Table 2. Probabilities of perceived benefit (i.e. pain reduction) and harms (i.e. complications) of subacromial decompression surgery for each clinical scenario, stratified by the decision to perform surgery.

	Clinical scenario							
	Scenario 1. 58-year-old construction worker		Scenario 2. 48-year-old woman with previous subacromial decompression		Scenario 3. 51-year-old painter		Scenario 4. 36-year-old volleyball player	
Surgery	**No**	**Yes**	**No**	**Yes**	**No**	**Yes**	**No**	**Yes**
N (%)	56 (87)	8 (13)	47 (78)	13 (22)	43 (75)	14 (25)	55 (96)	2 (4)
Median probability of benefit (IQR)	25 (10-50)	75 (70-88)	29 (10-50)	70 (50-81)	20 (9-39)	79 (50-81)	15 (6-39)	71 (70-71)
Median probability of harm (IQR)	9 (4-17)	9 (7-10)	10 (5-19)	5 (4-12)	13 (8-20)	8 (4-14)	8 (3-15)	3 (1-4)

IQR = Interquartile Range, N= number.

Table 3. Most important factors for the decision to perform surgery or not for each clinical scenario, stratified by the decision to perform surgery.

		Clinical scenario			
		Scenario 1. 58-year-old construction worker	**Scenario 2. 48-year-old woman with previous subacromial decompression**	**Scenario 3. 51-year-old painter**	**Scenario 4. 36-year-old volleyball player**
Treatment decision	**Subacromial decompression**	*(n=8; 13%)*	*(n=13; 22%)*	*(n=14; 25%)*	*(n=2; 4%)**
		- >1 Year complaints (n=6) - Shape acromion (Bigliani type 3) (n=6) - Reduction of complaints after subacromial infiltration (n=5) - Unable to work (n=5)	- Imaging: patrial supraspinatus rupture, no signs of bursitis (n=10) - > 6 Months complaints (n=6) - Reduction of complaints after subacromial infiltration (n=6) - Specific tests: empty can, Hawkins test and cross-chest test all positive (n=5)	- > 1 Year complaints (n=11) - Imaging: bursitis-like abnormalities, partial supraspinatus tendon rupture (n=8) - Progressive complaints (n=6) - No effect of exercise therapy on complaints (n=5)	- Professional volleyball player (n=2) - No effect of physical therapy on complaints(n=2)
	No subacromial decompression	*(n=56; 87%)*	*(n=47; 78%)*	*(n=43; 75%)*	*(n=55; 96%)*
		- Surgical treatment not indicated (n=41) - Imaging: no abnormalities on ultrasound/imaging (n=31) - Nonsurgical treatment better (n=30) - Complaints bilateral (n=15)	- Surgical treatment not indicated (n=36) - Imaging: patrial supraspinatus rupture, no signs of bursitis (n=26) - Nonsurgical treatment better (n=23) - Female, 48 years (n=11)	- Surgical treatment not indicated (n=25) - Imaging: bursitis-like abnormalities, partial supraspinatus tendon rupture (n=24) - Comorbidities of patient (n=19) - Nonsurgical treatment better (n=17)	- Surgical treatment not indicated (n=39) - Imaging: No abnormalities (n=35) - Nonsurgical treatment better (n=27) - Female, 36 years (n=25)

*Only the two most important factors were shown owing to the low fraction of surgeons deciding to perform subacromial decompression surgery in this clinical scenario.

Scenario 1: 58-year-old construction worker

A 58-year-old construction worker visits your outpatient clinic. He complaints about a gradually developed pain in both shoulders (left > right). Initially, he only experienced pain when working above shoulder level, but currently he cannot work anymore. When he lifts his arms above shoulder level, pain arises on the left side which radiates to the lateral side of his upper arm. He has experienced pain complaints for over a year, despite regular use of NSAID and a long trajectory of physiotherapy (>10 treatments) with a physical therapist. The general practitioner has already given subacromial infiltration twice, which gave a short-term but significant reduction in symptoms.

The physical examination shows a full range of motion. There is a slight loss of strength and a painful arc on the left. The Hawkins test is positive. The cross-chest test is negative. Radiological and ultrasound imaging shows no abnormalities except for a Bigliani type 3 acromion. Your hospital's leading insurer has announced that they will only reimburse 70% of surgical treatments for SAPS compared to the previous year. The department has indicated that the 70% ceiling is almost reached and that surgical intervention may no longer be reimbursed.

Scenario 2: 48-year-old woman with previous subacromial decompression

A 48-year-old woman visits your outpatient clinic because of chronic (>6 months) shoulder complaints on the left side. The pain worsens with overhead activities and she experiences a loss of strength. She has had exercise therapy for several weeks, but with minimal effect. She experienced the same symptoms 1.5 years ago on her right shoulder and recognizes the symptoms from that period. Back then, subacromial decompression surgery resulted in her getting rid of the complaints. At that time, she already experienced some symptoms on her left shoulder and the orthopedic surgeon indicated that the left shoulder possibly might be next. Currently, the pain symptoms are such that she cannot sleep, despite using many painkillers. She is also unable to play tennis, which is her hobby. She would like to undergo the same surgery as for her right shoulder, because this helped really well. The general practitioner has already given a subacromial infiltration, which gave short-term relief of her symptoms.

During physical examination there is antalgic restriction of shoulder movement. There is a painful arc. The empty can, Hawkins and cross-chest tests are all positive. The X-shoulder shows no abnormalities. The MRI scan shows apart from a partial supraspinatus tear no other abnormalities. The main healthcare insurer for your hospital has indicated that they will no longer reimburse the surgical treatments for SAPS, as they consider this not meeting the current standards of science and practice.

Scenario 3: 51-year-old painter

A 51-year-old painter visits your outpatient clinic with long-term pain complaints in his right shoulder. The pain started after a fall while playing soccer. He has visited your outpatient clinic several times in the past year. Previously, you saw bursitis-like abnormalities and a partial supraspinatus tendon rupture on imaging and diagnosed him with SAPS, which was treated with physical therapy and pain medication. The pain complaints by now have lasted for more than a year and limit him in his work and hobbies. Since recently, he also wakes up at night due to pain. He takes a lot of painkillers (NSAIDs) and would like to get rid of the pain. He has now received physical therapy for more than a year, but the pain remains. Subacromial injections work very well, but the pain keeps coming back. The patient tells you his colleague had the same complaints, for which he was treated by subacromial decompression surgery. His colleague was able to return to work soon after surgery and the patient asks if this would be a possibility for him as well.

During physical examination you see antalgic restriction of shoulder movement and a painful arc is present. The empty can and Hawkins tests are both positive. The cross-chest test is negative. Additional imaging shows no changes compared to the previous year. The patient has smoked all his life (30 pack years) and is treated by the cardiologist for Angina Pectoris. The healthcare insurer reimburses both surgical and non-surgical management.

Scenario 4: 36-year-old volleyball player

A 36-year-old woman has been experiencing pain complaints in the front of her right shoulder since 6 months. She is a professional volleyball player and the season has just ended. She mainly experiences pain symptoms when serving and smashing the ball. Despite her pain complaints, she has continued playing sports. This went reasonably well with painkillers and after a subacromial injection from the general practitioner. There has been no clear traumatic moment. She did go straight to the physical therapist, but physical therapy had no effect. Meanwhile the pain is such that she is impaired in performing daily activities and cannot play sports anymore. The last two weeks things seem to go slightly better (end of the season). She wants to be fit for the new season as soon as possible. Her physical therapist has contacted you and does not know what else he can do, he suggests surgery.

During physical examination, the range of motion of the right arm is slightly reduced compared to the left arm. There is a low painful arc and the Hawkins test is positive. The cross-chest test is negative. Additional imaging shows no abnormalities. The main healthcare insurer for your hospital has announced that they only reimburse 30% of surgical treatments for SAPS compared with the previous year. The departmental management has indicated that the 30% ceiling has almost been reached and that surgery possibly may not be reimbursed.

Figure 1. Description of clinical scenarios included in study.

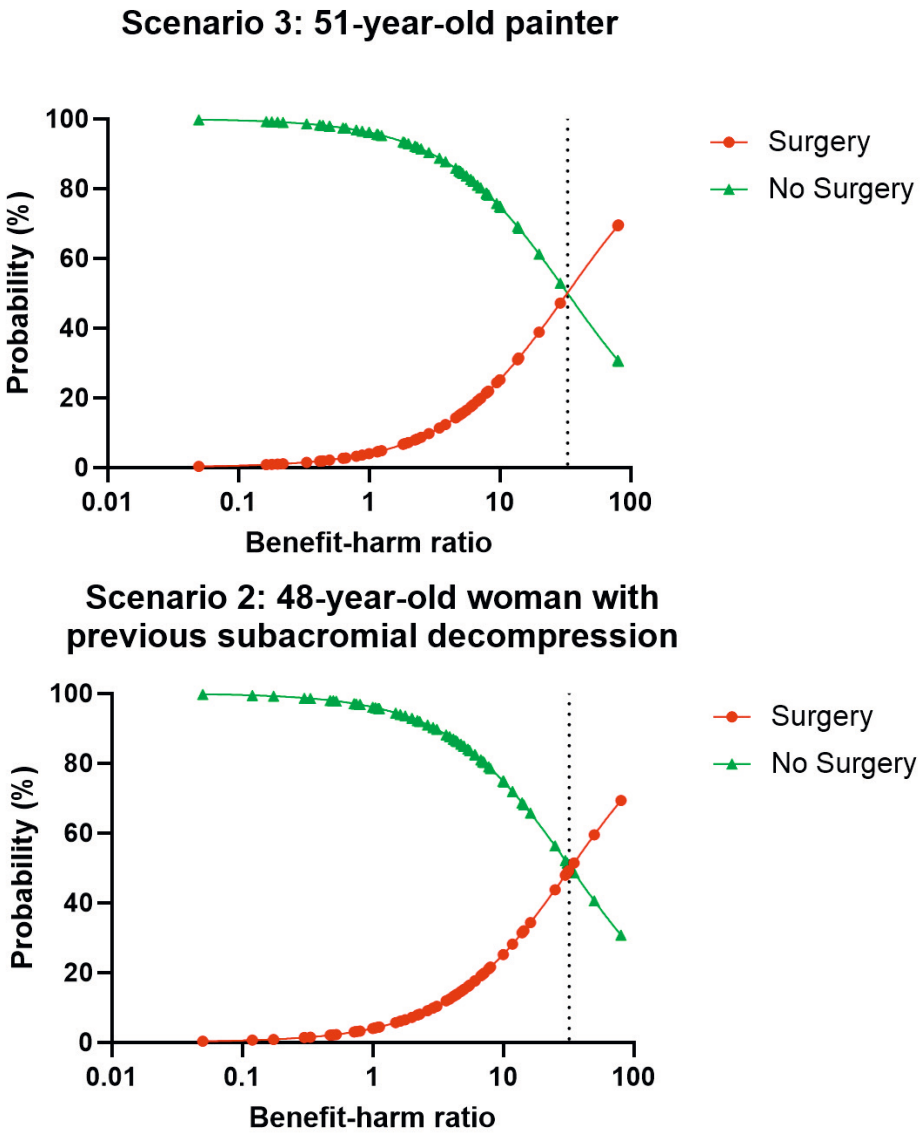

Figure 2. Predicted probability of subacromial decompression (SAD) surgery and no SAD surgery versus the benefit-harm ratio for two clinical scenarios. The dotted line represents the break-even point (i.e., the value of the benefit-harm ratio at which the predicted probabilities of performing SAD surgery and the predicted probability of not performing SAD surgery were equal [50%]): 32 for clinical scenario 2 "48-year-old woman with previous subacromial decompression" and 33 for clinical scenario 3 "51-year-old painter".

DISCUSSION

Consistently with our hypothesis, this study showed that there was substantial variation in the decision-making to perform surgery for SAPS between orthopaedic shoulder surgeons. Overall, the respondents were reluctant to perform SAD surgery as shown by the high break-even points indicating that the perceived benefit of SAD surgery had to substantially outweigh the harm before most of respondents decided to perform surgery. The decision to perform SAD surgery seemed to depend particularly on differences in the perceived benefits of surgery rather than differences in the perceived harms. Additionally, surgeons who decided to perform SAD surgery mainly reported patient-related factors to be among the most important factors, whereas surgeons who decided not to perform surgery mainly reported factors related to current clinical guidelines.

The overall reluctance to perform SAD surgery for SAPS in this study is in line with current evidence, the Dutch national guideline of the Netherlands Orthopaedic Association, and the clinical practice guideline recommendation by the British Medical Journal panel[2, 4, 7, 19]. Consistently with this, Veen et al. previously reported a decreasing trend in the use of SAD surgery for SAPS in the Netherlands, but still approximately 7% of the patients with a SAPS diagnosis underwent surgery in 2016[13]. Decreasing trends have also been reported in various other countries such as Scotland and Finland[20, 21], but increasing trends have been described for Australia, the United Kingdom and the United States[10, 11, 22]. The previously described conflicting trends highlight the need for studies exploring factors that might drive decisions to perform SAD surgery for SAPS despite the presence of high-quality evidence showing no benefit.

This study explored the variation in clinical decision-making to perform surgery for a low-value care procedure such as SAPS by using clinical case-vignettes. Previous studies examined the variation in clinical decision-making for various other surgical interventions (e.g., rotator cuff repair and gastrointestinal surgical procedures)[6, 17, 23]. These studies not only showed substantial variation in the clinical decision-making to perform surgery between surgeons[17, 24, 25], but also showed that this variation occurred within surgeons over time when the scenarios remained identical[23]. This finding suggests that the decision to perform surgery may depend on the subjective clinical judgment of a surgeon at a specific time point, but it is unknown what factors may have influenced the change in a surgeons' judgment over time. Sacks et al. studied how general surgeons' judgment regarding the likelihood of benefit and harm of surgery influenced their decision to perform surgery[6]. They reported that surgeons were more likely to perform surgery when their perceived benefit of surgery was high and their perceived likelihood of harm was low. The results of our study add to this literature that variation in the decision to perform surgery mainly seems to result from differences in perceived benefit rather than harm. Similar findings were reported by a nonsurgical study that evaluated the variation in transfusion decisions (i.e., red blood cell transfusion) within the intensive care unit[26].

It is unclear which factors drive differences in perceived benefit of surgery. Dunn et al. found that orthopaedic surgeons who performed a high volume of rotator cuff repair procedures had higher expectations of the surgical intervention than those who performed a low volume of procedures[17]. Therefore, it might reflect that these surgeons value their own experience higher than evidence from guidelines and the literature[27, 28]. However, inadequate judgement of perceived benefit may also result from cognitive biases in decision-making, such as the tendency of clinicians to overestimate the benefits and underestimate the harms of interventions (i.e., impact bias)[29]. Training surgeons to make them more aware of the influence of cognitive bias on their decision-making may help to improve this[30]. It is also possible that surgeons first decide to perform surgery in a particular clinical scenario and subsequently match their assessment of potential benefit and harm with their decision[31], which would suggest that we need to study factors influencing the decision to perform surgery rather than differences in perceived benefit. Finally, it is hypothesized that the variation in perceived benefit might be the result of different weighing of factors in the clinical scenarios[26], which is consistent with our results showing differences in factors reported as most important between surgeons who would perform surgery and and those who would not perform surgery.

Previous literature has shown that factors such as patient characteristics, scientific evidence, clinical guidelines, and financial constraints are important factors in the decision-making process regarding surgery[32]. In this study, we found that surgeons who would not perform surgery mainly reported factors related to current clinical guideline recommendations whereas surgeons who would perform SAD surgery reported the importance of clinical benefits . Wright et al. have previously proposed that the paucity of evidence, the controversy around evidence, and a lack of awareness or acceptance of evidence may cause differences in the interpretation and acceptance of clinical guidelines and evidence[33]. Consistently with this, we found that respondents who would perform SAD surgery for at least one clinical scenario, reported that there was insufficient evidence to stop reimbursement for SAD surgery for SAPS whereas surgeons who would not perform surgery indicated that there was sufficient evidence to justify such an initiative. It is interesting to note that orthopaedic surgeons more often decided to perform surgery in the clinical scenarios in which the patients had a partial supraspinatus tear (i.e., scenarios 2 and 3). A possible explanation may be that patients with a high grade partial-thickness tear were excluded in previous randomized trials, which surgeons might have taken as an indication that these patients might still benefit from SAD surgery[19]. Additionally, surgeons who would perform SAD surgery weighed patient-related factors more heavily in their decision, indicating a different interpretation of the guidelines or that they might find it difficult to abstain from surgical treatment in case of(long-lasting) patient complaints[34]. The latter would suggest that action bias[35], that is, the general preference for active over passive treatment in clinical decision-making, might also play a role. Unfortunately, there is no consensus on what alternative treatment is best for SAPS patients, leaving the clinicians with uncertainty, which might further contribute to action bias[36].

Knowing when to perform surgery is considered a critical skill for a surgeon[6, 28]. Our study findings highlight the complexity of reducing the use of a low-value care procedure in daily practice even if based on strong recommendations. Ultimately, decisions to perform surgery (or not) are not only based on objective evidence but also based on subjective clinical judgment which in turn depends on surgeons' personal experiences and beliefs. Changing clinician behavior is therefore considered an extremely complex process[37]. Most interventions that aim to reduce low value-care, such as guideline changes or the withdrawal of reimbursement, address only objective evidence but not the subjective clinical judgment of surgeons. Therefore, it is unlikely that such solitary interventions will be effective. The results of this study highlight the necessity for multifaceted interventions that address objective evidence, but also target surgeons' personal beliefs and perceptions because these will be more likely to have an effect.

The strength of this study is that we used the same clinical scenarios for every respondent to study the variation in clinical decision-making for SAD surgery and its influencing factors. This allowed for a better evaluation of differences in decision-making between orthopaedic surgeons rather than these decisions being influenced by differences in (complexity of) patients. Whereas most studies have only investigated the influence of patient-related factors[6, 26], we also included guideline-related factors and contextual factors in the clinical scenarios. Doing so provides a more complete understanding of the factors contributing to variation in surgical decision-making. Furthermore, respondents could fill out the survey anonymously, which likely improves respondents' willingness to give honest rather than socially desirable answers.

Limitations

Limitations of our study include the low response rate (39%). There will be inactive members in any professional society, who will not be likely to respond to a questionnaire. However, it is also possible that members had an interest in shoulder and elbow surgery, but did not frequently see and treat SAPS patients and therefore did not responded, given that the survey stipulated that at least one SAPS patient should be treated per month to participate. Because only 40 to 50 DSES members were actively participating in meetings and there were 64 respondents who participated in the present study, it seems that our study is likely representative of surgeons frequently performing shoulder surgery or treating sufficient numbers of SAPS patients. Second, common method bias may have influenced our results because both the independent and dependent variables were part of the same questionnaire[38]. To reduce the likelihood of this occurring, we varied the factors that could be chosen across scenarios, varied the question types, and used different wordings within the questions that included a Likert scale. Finally, because we conducted this survey among orthopaedic surgeons and residents in the Netherlands, the results of our study may not be generalizable to other countries, given that other factors influencing decisions to perform SAD surgery may be more relevant in other settings. However, the overarching finding of variation in clinical decision-making being particularly influenced by differences in perceived benefit

and not perceived harm to patients will likely also apply to other countries given results of other studies[26].

CONCLUSION

Overall, Dutch orthopaedic shoulder surgeons are reluctant to perform SAD surgery in SAPS patients. There is substantial variation among orthopaedic surgeons regarding decisions to perform SAD surgery for SAPS even when evaluating identical scenarios, where particularly the perceived benefit of surgery differed between those who would perform surgery and those who would not. Surgeons who would not perform SAD surgery mainly referred to guideline-related factors as influential factors for their decision, whereas those who would perform SAD surgery considered patient-related factors more important.

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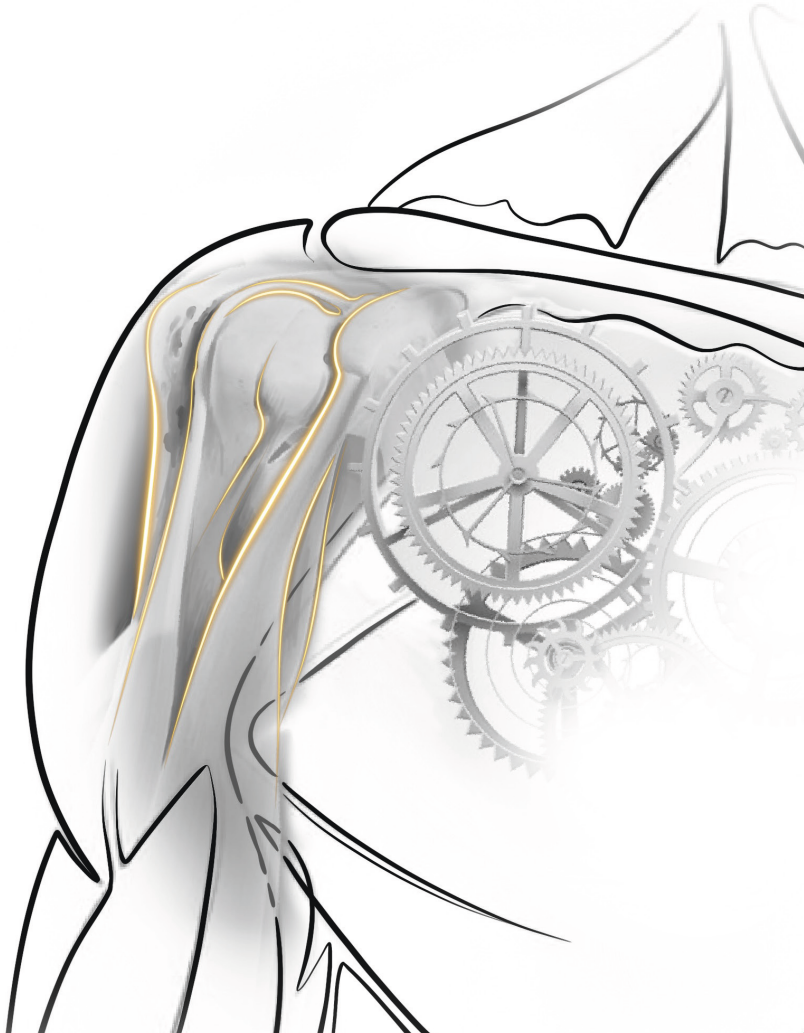

Chapter 7

Impact of Active Disinvestment on Decision-Making for Surgery in Patients With Subacromial Pain Syndrome: A Qualitative Semi-Structured Interview Study Among Hospital Sales Managers and Orthopedic Surgeons

Timon H. Geurkink M.D[1,2], Perla J. Marang-van de Mheen PhD[2], Jochem Nagels MD[1], Rudolf W. Poolman MD, PhD[1], Rob G.H.H. Nelissen MD, PhD[1], Leti van Bodegom-Vos PhD[2]

1. *Department of Orthopaedics, Leiden University Medical Center, Postbus 9600, 2300 RC, Leiden, the Netherlands.*
2. *Department of Biomedical Data Sciences, Medical Decision Making, Leiden University Medical Center, Leiden, the Netherlands.*

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ABSTRACT

Background:
Withdrawal of reimbursement for low-value care through a policy change, i.e., active disinvestment, is considered a potentially effective de-implementation strategy. However, previous studies have shown conflicting results and the mechanism through which active disinvestment may be effective is unclear. This study explored how the active disinvestment initiative regarding subacromial decompression surgery for subacromial pain syndrome (SAPS) in the Netherlands influenced clinical decision-making around surgery, including the perspectives of orthopedic surgeons and hospital sales managers.

Methods:
We performed 20 semi-structured interviews from November 2020 to October 2021 with ten hospital sales managers and ten orthopedic surgeons from twelve hospitals across the Netherlands as relevant stakeholders in the active disinvestment process. The interviews were video-recorded and transcribed verbatim. Inductive thematic analysis was used to analyse interview transcripts independently by two authors and discrepancies were resolved through discussion.

Results:
Two overarching themes were identified that negatively influenced the effect of the active disinvestment initiative for SAPS. The first theme was that the active disinvestment represented a "Too small piece of the pie" indicating little financial consequences for the hospital as it was merely used in negotiations with healthcare insurers to reduce costs, required a disproportionate amount of effort from hospital staff given the small saving-potential, and was not clearly defined nor enforced in the overall healthcare insurer agreements. The second theme was "They [healthcare insurer] got it wrong", as the evidence and guidelines had been incorrectly interpreted, the active disinvestment was at odds with clinician experiences and beliefs and was perceived as a reduction in their professional autonomy.

Conclusion:
The two overarching themes and their underlying factors highlight the complexity for active disinvestment initiatives to be effective. Future de-implementation initiatives including active disinvestment should engage relevant stakeholders at an early stage to incorporate their different perspectives, gain support and increase the probability of success.

Key Messages:

Implications for policy-makers

- Based on the results of this study, the effectiveness of an active disinvestment initiative seems to be largely dependent on the support for active disinvestment by relevant stakeholders.
- To gain support for active disinvestment and improve the probability of success, policy-makers should actively engage relevant stakeholders early on in the development of the disinvestment strategy.
- In specific, active disinvestment initiatives must have sufficient saving-potential and a required effort from hospital staff that is proportionate to the financial impact, need to be clearly defined and enforced in overall hospital agreements, and be supported by evidence and guidelines.

Implications for the public

The withdrawal of reimbursement for low-value procedures through a policy change, i.e., active disinvestment, is considered a potentially effective but underused strategy to reduce low-value care. This study found that the effectiveness of such initiatives seems to be largely dependent on the support for active disinvestment from relevant stakeholders (e.g., hospital sales managers and orthopedic surgeons). Therefore, policy-makers should engage relevant stakeholders early on in the development of an active disinvestment initiative to improve the possibility for success. Furthermore, several specific factors were identified within this study that may contribute to the limited support for active disinvestment by relevant stakeholders.

INTRODUCTION

Healthcare costs have increased drastically worldwide over the past several decades and are expected to continue rising in the coming years[1]. This rise in healthcare costs forces policy-makers to explore solutions to ensure good quality of care while working with limited financial resources[2, 3]. One potential solution to solve this challenge is to reduce low-value care, i.e., services for which there is little evidence of benefit for patients or that cause more harm than benefit (e.g., risk of complications, psychological distress, treatment burden and financial loss)[4]. Currently, it is estimated that approximately one-third of all medical spending is related to low-value care[5].

Choosing Wisely (CW) is an international campaign launched to open the discussion on low-value care and develop interventions to reduce overuse[6-8]. However, the literature merely shows a slight decline or unchanged trends in low-value care following such CW campaigns[9, 10]. Smaller reductions in the use of low-value care are associated with the release of CW recommendations than for a policy change eliminating reimbursement as shown recently for low-value use of vitamin D screening[11]. Therefore, withdrawal of reimbursement through a policy change –or active disinvestment– has been suggested as a promising alternative[12]. Active disinvestment has been associated with substantial reductions in low-value care and is considered an effective but underused de-implementation strategy[11, 13]. However, it is also considered a very complex strategy, influenced by various potentially complicating factors (e.g. level of support for disinvestment among clinicians and policy-makers), which make successful disinvestment a complex undertaking[12, 14, 15].

Given that less than half of the disinvestment initiatives have been successful until now[16], more research is needed to further explore and understand the complex mechanism through which active disinvestment may have an effect on reducing low-value care[16-18]. Theoretical frameworks that may facilitate understanding how active disinvestment influences (clinical) decision-making of different stakeholders for specific interventions are lacking but needed to guide future active disinvestment initiatives[18, 19]. Therefore, we investigated how the active disinvestment initiative of subacromial decompression (SAD) surgery for subacromial pain syndrome (SAPS) in the Netherlands influenced clinical decision-making around surgery, including perspectives of hospital sales managers and orthopedic surgeons, to increase our understanding on how active disinvestment initiatives may exercise their effect on clinical decision-making.

METHODS

Study Design

A qualitative study was conducted with semi-structured interviews among both hospital healthcare sales managers as well as orthopedic surgeons treating SAPS patients. We used a qualitative research approach as this provides more in-depth insights into processes that

numerical data cannot capture and is able to fully explore the perspectives of relevant stakeholders[20]. All results were reported according to the COnsolidated criteria for REporting Qualitative research(COREQ) checklist[21].

Setting

The Netherlands has a private-public financed healthcare system, with mandatory standard private healthcare insurance for all Dutch citizens from healthcare insurance companies and optional additional insurance (e.g., special dental care)[22]. The insurance market is dominated by four large insurers who together have a total market share of approximately 85%[23]. Most insurers operate nationally, but market shares vary per region and each region has a different market leader[23]. The government has given healthcare insurers an essential role in quality assurance by allowing them to selectively contract healthcare providers (e.g., hospitals) and specific interventions[24]. This selective contracting by healthcare insurers aims to reduce overall healthcare costs and improve the hospitals' quality of care. Periodically (mostly each year), healthcare insurers negotiate with hospitals (through their hospital sales managers) on prices and volumes of specific interventions. The latter gives the healthcare insurer the opportunity to apply active disinvestment initiatives for low-value care interventions and thereby reduce their costs.

Description of Intervention

SAD surgery for SAPS is considered a low-value care intervention as high-quality literature found no overall clinical benefit of surgical treatment for SAPS compared to non-operative treatment[25-28]. Nevertheless, in 2016 still approximately 10.000 patients underwent SAD surgery for SAPS in the Netherlands[29]. Therefore, one of the four largest Dutch healthcare insurers introduced an active disinvestment initiative from January 2020 onwards to reduce SAD surgery in SAPS patients. This healthcare insurer considered 80% of all currently performed surgical procedures for SAPS to be low-value care. To reduce the use of this low-value procedure, this insurer decided to contract 30% fewer surgical procedures for SAPS from each contracted hospital compared with the number of procedures in the previous year. The insurer informed hospitals about this specific active disinvestment by email and during the annual healthcare contract negotiations with hospital sales managers.

Participant selection

As the active disinvestment initiative of the healthcare insurer primarily targeted hospitals, we approached a purposive sample of 25 different relevant stakeholders working withing these hospitals (i.e., hospital sales managers and orthopedic surgeons) to participate in the semi-structured interviews. In the Netherlands, hospital sales managers form the direct link between hospitals and healthcare insurers. They are responsible for making financial arrangements on reimbursement of healthcare services provided to patients by a hospital. In addition, they are accountable for communicating healthcare insurers' policy changes within the hospital, including active disinvestment initiatives. Therefore, they are considered key players in making the process of active disinvestment work in daily practice. The orthopedic (shoulder) surgeons

treating SAPS patients were interviewed as they are ultimately responsible for clinical decision-making together with the patient.

We purposively sampled participants from different types of hospitals, i.e., academic and non-academic teaching and non-teaching hospitals or independent treatment centers, and different geographical regions because the impact of active disinvestment may vary significantly between types of hospitals and regions, depending on which part of their patients is insured by the healthcare insurer applying the active disinvestment initiative. The relevant stakeholders were recruited from the authors' professional network. All stakeholders were invited to participate and received information about the interview by email. Twenty of the 25 contacted stakeholders (80%) agreed to participate. One orthopedic surgeon did not agree to be interviewed due to a lack of time, while four approached stakeholders (i.e., one hospital sales manager and three orthopedic surgeons) did not respond to the invitation, despite several reminders by email.

Data collection

Given their different role in the decision-making process, separate interview guides were created for hospital sales managers and orthopedic surgeons (Appendix A). All interviews started with the question whether the participant was familiar with the active disinvestment for SAD surgery among SAPS patients from the particular healthcare insurer. The interviewer (TG) explained the active disinvestment initiative if they were unfamiliar with this. From this point, the interviews for hospital sales managers included the following topics: (i) the negotiation process between healthcare insurers and healthcare providers, (ii) the attention given to the active disinvestment initiative for SAPS during these negotiations, (iii) the consequences of this active disinvestment for the hospital, and (iv) the perceived effect of this active disinvestment on clinical decision-making. The interviews with orthopedic surgeons covered the following topics: (i) their treatment strategy for SAPS, (ii) the surgeons' perspectives about the active disinvestment initiative and (iii) the perceived effect of the active disinvestment on clinical decision-making. Potentially relevant factors (related to organizational context or individual professional) that might influence how the active disinvestment worked were taken from a study by van Dulmen et al. evaluating barriers and facilitators to reduce low-value care, and added to the interview guide as topics to discuss during the interview[30]. Participants were actively stimulated to say everything that came to mind and share their experiences and opinions. At the end of the interview, all participants had the opportunity to provide additional feedback.

Since the COVID-19 pandemic hindered face-to-face contact all semi-structured interviews were conducted and video-recorded (after verbal consent was obtained) via secured video calls (Microsoft Teams) by the same interviewer between November 2020 and October 2021. The interviewer (TG, male), a physician with additional qualitative interviewing training, did not have an established relationship with the participants before the interview nor was involved in clinical care. Two pilot interviews were conducted with one orthopedic surgeon and one

hospital sales manager to test relevance and refine the interview questions. Because the pilot interviews did not result in significant changes in the interview guide, both interviews were included in the analysis. The interviews continued until data saturation was reached, defined as at least three consecutive interviews revealing no new insights[31]. The median interview duration was 34 minutes (interquartile range: 30-39 minutes). During the interviews, the interviewer took notes to direct further questioning. Repeat interviews were not conducted and transcripts were not returned to the participants for comment or correction. Participants did not receive any financial compensation for their time.

Data analysis

All interviews were transcribed verbatim and entered into ATLAS.ti (version 7.0). TG and LB verified transcript accuracy. Interview transcripts were analysed using inductive thematic analysis. Inductive thematic analysis was applied to increase our understanding how the active disinvestment strategy may exercise its effect on clinical decision making in daily practice, thereby contributing to further development of theory rather than testing an existing theory. Thematic analysis is a flexible approach that identifies patterns within qualitative data, which is especially useful for describing processes that lack an existing theoretical framework[32]. After familiarizing with the data, initial codes were identified and a coding tree was developed into which the data was assigned. All interviews were independently coded by two authors (TG, LB). Discrepancies were discussed until a consensus was reached. Coded text segments were searched for and grouped into overarching themes by TG and LB. Overarching themes were defined as a group of factors that might influence how active disinvestment would affect clinical decision-making on SAD surgery for SAPS. The analysis of overarching themes was iterative and continuous throughout data collection. The overarching themes were inspected and discussed by TG, PM and LB for recurring themes and influencing factors on the disinvestment process until consensus was reached. Participants did not receive the results of the analyses and were not asked to provide feedback on the findings.

Ethical approval

The study protocol (protocol number: N20.127) was presented to the Medical Ethical Committee of the Leiden University Medical Center (METC-LDD, Code 058, Leiden, the Netherlands) who waived the need for ethical approval under Dutch law.

RESULTS

Ten hospital sales managers and ten orthopedic surgeons from twelve different hospitals were interviewed, with data saturation achieved after respectively nine- and eight interviews. The healthcare insurer applying the active disinvestment initiative had the largest market share in three (25%) hospitals. Most (55%) interviewed participants worked in non-academic teaching hospitals. Three hospital sales managers (30%) and four orthopedic surgeons (40%) were familiar with the active disinvestment initiative for SAPS patients prior to the interview. Descriptive characteristics of the respondents are shown in Table 1.

Table 1. Baseline characteristics of study participants.

Participant characteristics	Hospital Sales Managers *(n=10)*	Orthopedic surgeons *(n=10)*
Mean Age (SD)	46 (8.1)	50 (7.8)
% Female	50	20
Mean years of experience as orthopedic surgeon (SD)	*NA*	13 (7.2)
Mean number of SAPS patients per week (SD)	*NA*	13 (9.1)
Hospitals		
Academic	1	1
Non-academic Teaching	5	6
Non-academic Non-teaching	3	3
Independent treatment center	1	0
Healthcare insurer market leader in hospital	3	3
Familiar with active disinvestment for SAPS patients	3	4

Thematic analysis resulted in the identification of two overarching themes which negatively influenced the support for the active disinvestment of SAD surgery for SAPS patients. The first theme was that the active disinvestment represented a 'Too small piece of the pie' for the hospitals. Particularly hospital sales managers stated that the active disinvestment initiative for SAPS i) had little financial consequences for the total hospital budget, ii) was only part of the negotiation process in the sense of that healthcare insurance companies used it merely to lower the overall pricing of the hospital's overall contract agreement, iii) required too much effort from hospital staff to accomplish only a slight reduction in overall costs and iv) was not clearly defined. For these reasons the active disinvestment did not influence hospital-level decision making and information regarding the active disinvestment was not communicated within the hospital to orthopedic surgeons (see Figure 1).

The second overarching theme was 'They[the healthcare insurer] got it wrong'. This theme was mainly highlighted by orthopedic surgeons who disagreed with the active disinvestment by the healthcare insurer. More specifically, the surgeons reported that the active disinvestment initiative was i) the result of misinterpretation of scientific evidence and clinical guidelines, ii) at odds with physician experience and beliefs and iii) reduced the professional autonomy of clinicians. With regard to physician experience and beliefs, the general obligation as a physician to provide care was highlighted, as well as that SAD surgery could still be beneficial for specific patients and should therefore remain as a treatment option. For these reasons the active disinvestment did not influence patient-level decision making (see Figure 1).

Besides these two overarching themes, several other contextual factors were identified that also influenced the effectiveness of the active disinvestment, such as a lack of communication between relevant stakeholders, patient preferences, fear of losing revenues and simultaneous other interventions that rewarded rather than penalized hospitals for not performing specific procedures. The overarching themes, subthemes and contextual factors are described in the following section, with representative quotes supporting each theme shown in Table 2. Although most participants indicated they were not against active disinvestment initiatives, we did not identify any facilitating factors for the active disinvestment initiative to work as intended to reduce SAD surgery for SAPS.

TABLES

Table 2. Representative quotations supporting each theme.
SM indicates hospital sales manager, OS indicates orthopedic surgeon**.** *The ZE&GG program is a program for the evaluation and appropriate use of care from Zorginstituut Nederland. Within this program, hospitals work together with healthcare insurers and the Dutch government in order to actively reduce the use of low-value care. The hospitals receive a financial incentive for their participation.

"Too small piece of the pie"		
Subtheme	*Quote number*	*Representative quotations*
Little financial consequences for the hospital budget	1	"I mean: there is so much money involved. So let's then focus on the big groups, on the mass and on the good things. This incentive to me comes across as a specialization within a specialization within a sub-specialization." (SM3)
	2	"Look, with the healthcare insurer we negotiate about 100 million euros. Yes, you know, then you are not going to talk about one specific diagnosis treatment combination, that does not happen." (SM8)
	3	"I am not going into details, because that would go beyond what I am allowed to do. I cannot describe the details of such an agreement here. But it had no real financial impact on the total of the agreement" (SM4)
	4	"According to me is that not our large group [of patients] but I don't know that myself. In other words, then you will not notice that at all, if you only have a fraction of [patients insured by] [Name insurer]. (OS 2)
Only part of the negotiations	5	"Sometimes it seems as if you have a lot of conversations, where there is only push to knock off a little of the overall price" (SM4)
	6	"Because you have to realize that during the negotiation this is only one part of the total amount on the table. In the end, after exchanging a lot of arguments, you come to an agreement with each other, which is often painful on all fronts. You have to leave certain things to be able to come to an agreement. This is also to the case for the healthcare insurer. They too cannot achieve everything from their dreamed mandate. (SM4)

Table 2. Representative quotations supporting each theme. (continued)

"Too small piece of the pie"		
Only part of the negotiations	7	"It is often under one big insurance ceiling, so then you get sort of a waterbed effect. So if you say: we want you to do this much fewer procedures and if you don't entirely exclude that from the contract, so take it out from the ceiling in a separate financial agreement, then it may be possible that you can fill it up with for instance acute care or birth care. So the only way a healthcare insurer can say that you cannot do it anymore and we are going to enforce it, is that they cut out the entire block and really make it a separate part of the agreement. With a clear ceiling on the procedure so that it is really impossible to reimburse it anymore. (SM6)
	8	"There is often discussion about this during negotiations, with a lot in plus and a lot in minus, then these die individually so to speak and you come to an overall deal, 30, 40, 50, -60 million, in which everything is quasi intertwined." (SM7)
	9	"And with that (with the active disinvestment) you are actually forced to stop performing that procedure as you don't get paid for it. That works I think only limited because what you do is that you often agree on one big pocket of money in which you apply the reduction. But if you secretly keep performing that procedure and instead do not perform something else where you have not talked about, then you will still receive the money but for the wrong things." (SM1)
Too much effort for a slight reduction in costs	10	"You are talking about a legitimate 420 people, that is a very small number [of patients] and if you also take off the ones with the two other indications then that leaves so few, that yes, is it really worth the effort to spend time on it?" (OS 9)
	11	"And let's then mainly talk about the good things and not about a few niches which will take 80 percent of the energy, but only account for 2% of the costs." (SM3)
	12	"At the moment that you would you want to take steps towards efficiency, then you are looking for the big hits. Because you have to look at it this way, the commitment of people and resources is probably just as much to improve a certain procedure where we do only a small amount, compared- to where we do large amounts ..... But more from the perspective: how do I invest my resources that are limited, human capacity too, to monitor [this]. (SM1)
	13	"All the topics that are now on the orthopedics agenda. Yes, these have passed here in the last few years. Still I am forced to catch up on a certain indexation and to make a plan. For which I actually need to hire someone, who would write than plan for me. Whilst we are already doing this.... you are almost forced to go along in that flow. While in the end it often doesn't lead to the intended goal, or that goal has already been achieved. It involves a lot of effort." (SM10)

Table 2. Representative quotations supporting each theme. (continued)

"Too small piece of the pie"		
Active disinvestment not clearly defined	14	"But what we find very difficult about that information from [Name healthcare insurer] to which you just referred. That is that they indeed say: yes, 80 percent [of these procedures] you should no longer- do. But you don't really know what 100 percent is. That I find very difficult as you only see what you do, maybe that is already very little, that you have left already that 20 percent [of the procedures that is of value to the patients]. .....So yes, what has actually been your baseline measurement?" (SM5)
	15	"We expect that next year you only do one-third of this [number of procedures]. What kind of discussion is that?" (SM9)
	16	"If a healthcare insurer would really shut it down with us and would say: you just cannot reimburse this [procedure] anymore, because it is not meaningful and that has been proven in so much literature. You should not do this anymore. Then we would send out a very clear signal [to the clinicians] you are not allowed to do this anymore." (SM6)
"They got it wrong"		
Subtheme	Quote number	*Representative quotations*
Misinterpretation of scientific evidence and clinical guidelines	17	"Yes, I'm fine with that [active disinvestment strategy by insurer] as long as they are well-founded and that's where it goes wrong. They don't have the knowledge, of course they have some medical advisors, but they don't have substantive knowledge to enter a discussion with me. They don't have the clinical experience with these patients and can't interpret scientific research. (OS 1)
	18	"Regardless of the situation, I don't think the overall trend is good. Maybe it's typical doctors reasoning but they [healthcare insurers] can't gauge the true value of the science. They are good with numbers and at negotiating but not in valuing scientific research and its relation with clinical practice." (OS 8)
	19	"And certainly the SAPS complaint, that is such a diverse group of patients so that you can hardly draw any conclusions about SAPS treatment in general." (OS 2)
	20	"They [healthcare insurer] include all kinds of things under the SAPS diagnosis.....So medically speaking, those are all very different things. But the healthcare insurer, the layman, includes everything under the same as if it is one big umbrella diagnosis, covering everything." (OS 4)

Table 2. Representative quotations supporting each theme. (continued)

"They got it wrong"		
Misinterpretation of scientific evidence and clinical guidelines	21	"I know that they have been working on that for a long time, to stop reimbursing this care, however, they classify everything under one diagnosis. I think that you can't do that, an acromioclavicular resection is suddenly a part of SAPS. That's not in our guideline, that's just not right." (OS 5)
	22	"In line with this, my biggest fear is that they will also stop reimbursement for cuff repair as they will say it is the same. But it's not the same. They classify everything under the SAPS syndrome, one collective term for every diagnosis. Yes, before you know it you can't perform any surgery anymore. Not that that's what this is about, but I think it's a worrying development." (OS 8)
	23	"Guidelines remain guidelines and it is not the case that this means that these interventions must absolutely not be performed and it is also not said that it's a medical error when you perform this procedure. Therefore, I don't understand why this policy is used by the healthcare insurer" (OS 7)
At odds with physician experience and beliefs	24	"When they say: it is not allowed anymore, it will no longer be reimbursed. Yes, then I'll use a different [reimbursement] code as I still have a patient who is crying out in pain. I have a general duty of care to help the patient. It is very strange that they say that I can't do this. They should not be able to do this. They also have a general obligation to provide care (OS 9)
	25	"When I notice that my treatments don't have any effect, than I'm not going to offer it. The majority of patients that we treat are eventually happy and have less pain. I don't care whether this is the result of surgery, an injection or some explanation. If I can treat patients well with conservative treatment then I will be happy to do so. But I still think there is a role for surgical treatments. (OS 2)
	26	"But when there is a persistent problem and the patient has had adequate treatment for at least one year, our physical therapists cannot do anything anymore, the diagnosis is confirmed on echo and a subacromial injection provides temporary pain relief, and everything points towards that direction [subacromial pain syndrome]. Yes, then I think you should still have the option to perform surgery. (OS 5)
	27	"When I think there is a medical indication, then that's it and then I'm going to do that. I can still justify this for myself from the medical evidence. But on the other hand, it's so artificial to say: we stop reimbursement. Look, orthopedic surgeons are smart enough to use another code [diagnosis and procedure code combination*]. So yeah, this is not a good way to regulate something at all." (OS 7)
Reduced professional autonomy	28	"Well, I think that the healthcare insurer should not take the place of the doctor..... I know for sure that we, orthopedic surgeons, are not waiting for healthcare insurers that tell us what we should and shouldn't do" (OS 7)

Table 2. Representative quotations supporting each theme. (continued)

"They got it wrong"		
Reduced professional autonomy	29	"Speaking for myself, I have the feeling that they think that we want to fool someone or to get financial gain out of something. The more subacromial decompressions we do, a simple procedure, the more money we earn. But nobody, at least none of my colleagues, thinks about this while doing consultations at the outpatient clinic." (OS 6)
	30	"I think that the incentive should be initiated by the professional association..... In the end, I think that clinicians should decide and that the treatment policy should not be determined by the health insurer." (OS 8)
	31	"You get people in the right direction more quickly if they are intrinsically motivated, instead of extrinsic matters. I think that the social control in the Netherlands is also large enough. At a certain point, you know from colleagues if they still do it [procedures] or if they do it a lot and you come across them and you will see their patients for a second opinion. So, I think the circuit also works well and you don't want to be seen as the one who is still performing these operations. I think everyone has his own pride in that. I think that this might work better than a healthcare insurer interfering in this, as they are not seen as a partner. And then I'm putting it mildly." (OS 1)
Contextual factors		
Subtheme	*Quote number*	*Representative quotations*
Lack of communication between relevant stakeholders	32	"What I sometimes find rather peculiar about [name healthcare insurer], they are, in my opinion, very good at just throwing things over the fence..... If you really want to change something, then start a conversation." (SM3)
	33	"We are never told about this by sales or the medical manager, that we need to change certain things in our working procedure." (OS 2)
	34	"Within our group [orthopedic surgeons] we have discussed this extensively and I also formulated a response [for the healthcare insurer], which was checked by the others. ..... The stupid thing is, I don't hear anything about it. I know that this now also is a point of concern nationally, so that it will also be tackled nationally after that discussion in the shoulder elbow working group. But I haven't heard back what's going to happen now, whether the care we provide will be reimbursed or not." (OS 5)
Patient preferences	35	"People who have had the [same] surgery in the past, had good results on that side and now have similar pain or shoulder complaints on the other side. They want surgery." (OS 3)

Table 2. Representative quotations supporting each theme. (continued)

Contextual factors		
Patient preferences	36	"By the way, then people will sometimes just go to Belgium. And that will subsequently also be reimbursed by the insurers. So there are patients who bypass the system."(SM4)
Fear of losing revenues	37	"But the projects [reducing low-value procedures] don't run themselves as the surgeons are financially rewarded for their volume [of performed procedures]. Therefore, at the moment you remove volumes, they will not be the first ones applauding for you" (SM2)
	38	"We are a hospital in which incomes and honoraria are highly correlated with production. So yes, I dare to state that these kind of desired movements will not be helped by the way in which hospitals like ours work and that such incentives could be very tricky in that context." (SM5)
	39	"I understand that there are still clinics where they [surgeons] just do everything: hips, knees, shoulders, and that subacromial pain leads to a subacromial decompression..... Because they make their living with this. An arthroscopy will give you a lot of money. So if you just do a bursectomy or a Neer acromioplasty, that will yield a lot of money."(OS 9)
Simultaneous other interventions	40	"For example, there is now also the ZE&GG program*, we actively started with this last year..... Last year this was a bit more noncommittal and you could see this in the varying degree of involvement between departments. This year we are going to make it mandatory as there are financial agreements linked to this for the hospitals." (SM3)
	41	"I'm not familiar with the example [active disinvestment strategy for SAPS], that's how I should phrase it. However, the general tendency and the conversations, we are constantly working on this. Patients a day shorter. They used to spend three days in a room after surgery, now only two or sometimes one. So in general, we are already working on similar trajectories."(SM8)
	42	"Then we're talking again about the ZE&GG agenda. The hospitals get a certain additional compensation for increases in wages and collective labor agreement, and for that they have agreed to actively participate with that ZE&GG agenda ..... So in that sense this works with an incentive. Apparently, the hospitals were interested and have agreed with this. Because they get their money, but also do something in return. We all get it. I think this might be relevant for you to consider, to what extent would incentives work better than disincentives."(SM1)

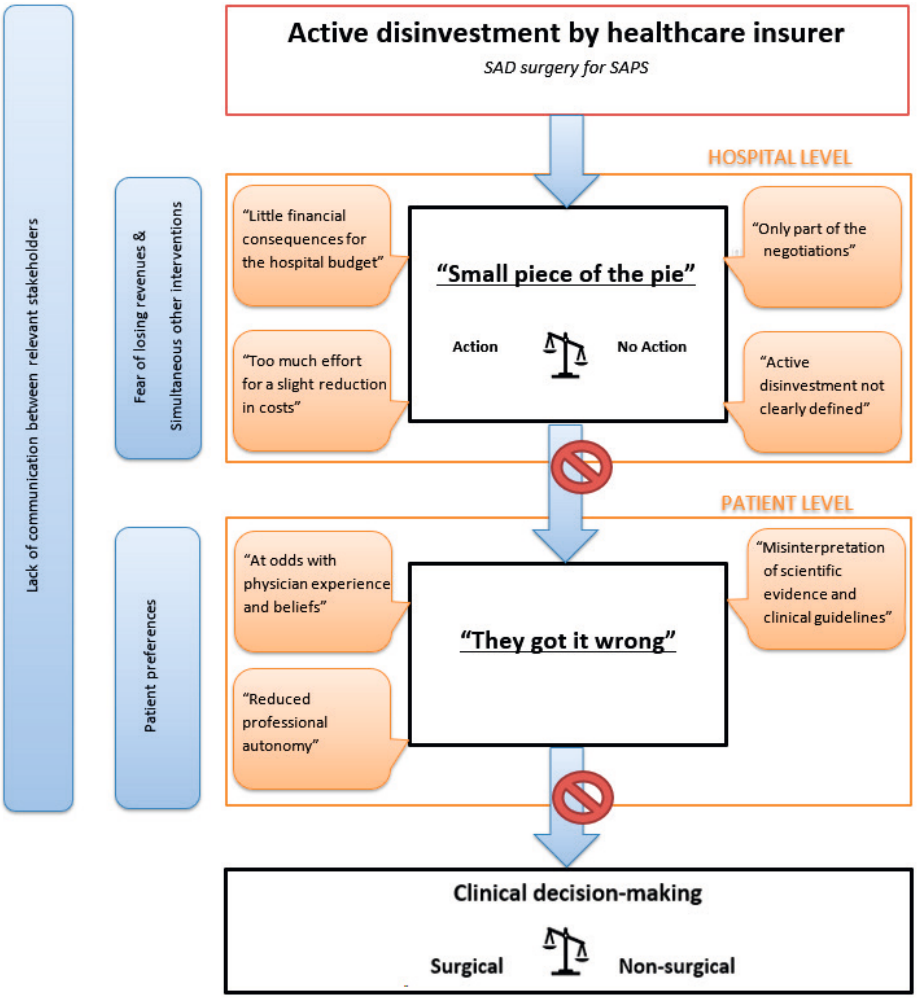

Figure 1. Factors negatively influencing the effectiveness of the active disinvestment initiative on clinical decision-making.

Too small piece of the pie

Little financial consequences for the hospital budget

Both hospital sales managers and orthopedic surgeons stated that SAD surgery for SAPS reflects an insignificant part of the total care provided by the hospital. As a result, the active disinvestment for one specific procedure represented little financial value compared to the overall costs of care provided by the hospital (Quote 1-2). The active disinvestment initiative in its current form was considered to have no financial consequences for the hospitals' budget, especially in hospitals where this healthcare insurer only had a small market share (Quote 3-4).

Only part of the negotiations
Hospital sales managers mentioned that the active disinvestment was only a tiny part of the negotiation process between the healthcare insurer and hospitals. They believed it merely aimed to lower the price of the overall contract agreement rather than explicitly reducing the number of performed surgeries for SAPS patients (Quote 5-7). At the end of the negotiation process, a contract is drawn up that includes the total volume of procedures (not only SAD for SAPS, but one overall agreement containing all procedures within the hospital) together with an overall price (Quote 8). Within this agreement, the active disinvestment no longer receives any particular attention, thereby leaving the possibility to perform SAD surgery for SAPS and receive reimbursement for it (Quote 9).

Too much effort for a slight reduction in costs
Related to the first sub-theme that SAD surgery for SAPS had little financial consequences for the hospital, both hospital sales managers and orthopedic surgeons believed the amount of effort they had to put into reducing SAD surgery was disproportionate to the saving-potential (Quote 10-11). They also indicated that efforts by hospital staff to reduce low-value care procedures are likely to be the same for low-volume procedures as high-volume procedures. From an efficiency perspective, hospital staff should therefore better focus on high-volume procedures (Quote 12). In addition, hospitals often already have initiatives that aim to reduce the use of low-value care procedures so that such initiatives from healthcare insurers lead to unnecessary duplication of work (Quote 13).

Active disinvestment not clearly defined
A final sub-theme was that the active disinvestment was unclear (e.g., the use of relative outcome measures without adequate baseline measurement), not specific enough and still allowed to perform surgery for SAPS as part of the surgeries were still reimbursed (Quote 14-15). Hospital sales managers argued that surgeons would only stop performing SAD surgery for SAPS when there would be no reimbursement at all (Quote 16).

They got it wrong

Misinterpretation of scientific evidence and clinical guidelines
Orthopedic surgeons felt the healthcare insurer had misinterpreted the existing scientific evidence and clinical guidelines on which the active disinvestment was based. In general, they supported the reduction of low-value care. Still, they highlighted that healthcare insurers often lack the knowledge, skills and clinical experience to correctly interpret the scientific evidence and guidelines, so that active disinvestment initiatives cannot be based on their interpretation (Quote 17-18). Given that SAPS is an umbrella diagnosis covering a heterogeneous group of etiologies with different treatment needs, they felt that too many diagnoses and procedure codes were included in this particular active disinvestment. As consequence, the active disinvestment did not correctly reflect the SAPS population for which surgery is or is not appropriate, nor which surgical procedures were not appropriate for these

patients (Quote 19-22). In addition, clinical guidelines aim only to guide clinical decision-making, and do not dictate treatment for specific patient groups. After all, it is the health professional's decision to decide on an individual patient's treatment given the specific input of clinical information of an individual patient in conjunction with the clinical experience of the orthopedic surgeon. Thus, surgeons felt that clinical guidelines should not be used to formulate active disinvestment initiatives(Quote 23).

At odds with physician experience and beliefs
Orthopedic surgeons argued that withholding treatment options resulting from active disinvestment initiatives, is at odds with their general obligation to provide care (Quote 24). They also declared that healthcare insurers have a similar obligation, to reimburse care needed by the patient, which is also violated by this active disinvestment. In addition, surgeons disagreed with the active disinvestment initiative as they believed that some patients could still benefit from surgery, often based on previous individual experience (Quote 25-26). They were not convinced that such a disinvestment initiative would result in a reduction of surgery for SAPS as surgeons would still decide to perform surgery when deemed appropriate by changing their coding practices rather than not doing the surgery anymore (Quote 27).

Reduced professional autonomy
Orthopedic surgeons argued they had extensive training to weigh different treatment options appropriately, to best care for their patients. Applying such active disinvestment initiatives, the healthcare insurer intervenes in the physician's work by limiting clinical treatment options and thereby diminishes their professional autonomy. The surgeons mentioned that healthcare insurers should not take over the role of physicians (Quote 28) and felt that such initiatives expose an underlying mistrust of healthcare insurers in the professional autonomy of physicians (Quote 29). In general, they stated that healthcare insurers should not initiate such initiatives, but that these should be initiated by the orthopedic professional association (Quote 30-31).

Contextual factors

Four contextual factors were identified that influenced the effectiveness of the active disinvestment initiative for SAPS. First, both hospital sales managers and orthopedic surgeons mentioned the lack of communication between relevant stakeholders. More communication was needed between the healthcare insurer and hospitals as sales managers needed additional explanation about the active disinvestment by the healthcare insurer (Quote 32). Increased communication was also required among the relevant stakeholders within hospitals as orthopedic surgeons indicated not being informed about the active disinvestment by neither the sales managers nor the healthcare insurer (Quote 33-34). Second, patient preferences may persuade orthopedic surgeons to perform surgery for SAPS (Quote 35-36). Third, fear of losing revenues was suggested as a contextual factor as orthopedic surgeons may have financial benefit from performing surgery, which might reduce the impact of the active

disinvestment (Quote 37-39). Striking was that these were only suggested to apply to others (e.g., sales managers about orthopedic surgeons, or orthopedic surgeons working in general hospitals about surgeons working in ITCs) and therefore it remains unclear whether this really affects active disinvestment. Finally, simultaneous other interventions (either national or within hospitals) aiming to reduce low-value care may also influence how well the active disinvestment will work (Quote 40-41). Some of these interventions financially rewarded hospitals for not performing low-value care procedures anymore. Sales managers thought that such initiatives would be more successful than active disinvestment as this would be more motivating for hospitals (Quote 42).

DISCUSSION

The present study showed that two overarching themes negatively influenced the impact of the active disinvestment regarding SAD surgery for SAPS in the Netherlands, as both hospital sales managers and orthopedic surgeons did not support the active disinvestment from the healthcare insurer. Particularly hospital sales managers felt it represented a "Too small piece of the pie" where it was merely used in negotiations to reduce costs but had little financial consequences for the hospital budget while requiring a lot of effort, and was not clearly defined nor enforced in the overall agreements between healthcare insurers and hospitals. As a result, they did not communicate the information on the active disinvestment initiative to orthopedic surgeons. Additionally, orthopedic surgeons felt "They got it wrong" as the active disinvestment had incorrectly interpreted the evidence and guidelines, was at odds with physicians' experiences and beliefs, and perceived it as a reduction in professional autonomy. As a result, it did not affect their clinical decision-making regarding surgical or non-surgical treatment of these patients. Contextual factors that influenced the impact of the active disinvestment were lack of communication between stakeholders, others being afraid to lose revenue, patient preferences and other simultaneous interventions.

A strength of this study is that we investigated how this active disinvestment exercises its effects in daily practice from both organizational and clinical perspectives. As we interviewed all study participants within two years after the active disinvestment was put into place, the results of the present study represent the topical opinions and experiences of key players involved in this process. Our findings add to existing theories on the effect of active disinvestment, which was suggested as a promising alternative to reduce low-value care, regarding the various factors through which the impact in daily practice may be considerably reduced. Limitations of our study include the fact that we did not interview SAPS patients themselves, even though they are important stakeholders in clinical decision-making who are likely to be affected by the active disinvestment initiative[12]. In that context, it is relevant to note that the preferences of patients were identified as a contextual factor influencing the clinical decision process and indirectly also as factor influencing the effect of the active disinvestment because patients bypass the systems to get their preferred SAD surgery (see Quote 36). Secondly, since we only investigated the active disinvestment initiative for one

specific procedure in a Dutch healthcare setting, the results are not necessarily generalizable to other contexts as other factors may be relevant in different circumstances because such initiatives are deemed context-specific[14]. On the other hand, the overarching themes may still apply, i.e., that it should have financial consequences for a hospital to make it worth their effort and that evidence on which it is based should be correctly interpreted, for which it is essential to engage clinicians with relevant expertise. Thirdly, we recruited participants from our professional networks to ensure a diverse sample. It is possible that our professional network does not adequately reflect the views of all sales managers and orthopedic surgeons from the Netherlands. For example, stakeholders who strongly disagree with such active disinvestment initiatives may have been more willing to participate. Furthermore, no orthopedic surgeons from ITCs agreed to participate, while it has been suggested that non-teaching hospitals deliver more low-value care[33]. We did include participants from non-teaching hospitals as well as a sales manager from an ITC, who will likely capture the main views from ITCs although there may be some context-dependent differences. Additionally, our results may have been biased as the active disinvestment initiative started during the COVID pandemic, so that the active disinvestment may have received less attention from relevant stakeholders within the hospital. Since we provided a clear explanation of the active disinvestment initiative to participants not familiar with the active disinvestment, we do not believe that this had a significant influence.

Many studies have been published on priority setting and resource allocation in healthcare and de-implementation strategies for low-value care procedures[34, 35]. Hardly any studies have, however, previously evaluated the outcome of an active disinvestment initiative on low-value care[11, 12]. Despite its potentially powerful effect, only few initiatives have shown to result in actual disinvestment[12]. More frequently, active disinvestment initiatives are preliminary terminated. Rotteveel et al. evaluated commonalities between factors influencing the outcomes of active disinvestment initiatives in five recent cases in the Netherlands[12]. Consistent with the results of the present study, they found that the degree of support from relevant stakeholders largely determined the success of an active disinvestment initiative. Rotteveel et al. mainly evaluated the active disinvestment initiatives from a macro-level policy-makers perspective (e.g. governmental institutions, health insurers) and concluded that policy-makers should search for interventions for which there is support from relevant stakeholders when applying an active disinvestment initiative. The present study therefore adds evaluating an active disinvestment initiative from a more meso/micro-level perspective (highlighting e.g., local institutional factors[36]) and identified several factors at meso/micro-level that contributed to the limited support for active disinvestment from relevant stakeholders. This adds to our understanding how support for active disinvestment initiatives by relevant stakeholders is needed to affect clinical practice, rather than only issuing an active disinvestment by policy-makers without any additional strategies targeting behavioural factors.

Healthcare providers frequently disagree on how disinvestment initiatives should be prioritized as more than one low-value care practice is often considered suitable for

disinvestment. Previous studies on priority setting and de-implementation strategies in healthcare state that the prioritization of such initiatives should also be based on the potential financial impact (i.e. cost-saving potential), which is consistent with basic economic theory principles[37,38]. Our findings are in line with this, as we found that there was no support for the active disinvestment initiative because it had too little financial consequences for the hospital budget given the small number of SAPS patients for most hospitals. Hence, they highlighted the importance of looking for bigger hits with more significant saving potential proportional to the required effort from hospital staff. The importance of a proportional ratio between financial impact and required effort (e.g., from hospital staff) is also described by Conrad et al. who explored the effect of monetary incentives on healthcare quality improvement[39]. They concluded that larger financial incentives are more likely to result in improved quality of care and cover the costs of additional efforts and care process changes[39]. They also suggested that financial penalties may elicit an even stronger response due to "loss aversion". However, lack of financial impact may contra wisely limit its effectiveness as it will not motivate healthcare providers to change behaviour nor cover the costs of additional efforts. Furthermore, sales managers related the lack of financial impact of the active disinvestment to it only being a partial reimbursement stop and the active disinvestment being unclear (e.g., the use of relative outcome measures) and not specifically enforced. Consequently, orthopedic surgeons could still perform surgery for SAPS and receive reimbursement. It is possible that active disinvestment initiatives would only work in situations with a complete reimbursement stop, as was the case with the successful Vitamin D reimbursement stop in Canada[11]. Such a complete reimbursement stop makes it impossible to circumvent the active disinvestment. Additionally, it may be that an absolute performance target would have worked better than the relative performance measure used in the active disinvestment regarding SAPS as it is known from literature that these have better incentive properties than relative performance targets[39]. A difficulty of relative performance targets is that they need adequate baseline measurements, which make clear what proportion of care is of low-value. However, in the active disinvestment regarding SAPS hospitals only knew how many surgical procedures they performed but not whether those were low-value care. Consequently, they didn't have a clear view of their improvement potential.

Orthopedic surgeons felt the healthcare insurer had misinterpreted existing scientific evidence and guidelines on treatment in SAPS patients even though literature suggests that disinvestment initiatives should be based on the strength of evidence supporting the lack of effectiveness[37]. The construction of evidence in a disinvestment context is a very complex process as the results of scientific evidence are often not "black or white" and subject to between-subject variation in interpretation. Hodgetts et al. stated that selection and interpretation of evidence in a disinvestment decision is necessarily framed such that it better fits the disinvestment initiative[40]. Therefore, they highlighted the need for physician engagement within this process as they can add vital nuance to the debate on what evidence counts in a disinvestment decision and avoid any misinterpretations arising from this 'fitting it in the disinvestment initiative'. Additionally, policy-makers often present their disinvestment

initiatives as being "black or white" which leaves little room for clinical judgment (it is either low-value care or not) even though it is more nuanced in clinical practice[41]. Although sometimes there may be clear-cut candidates for disinvestment initiatives, i.e., interventions that are entirely ineffective, these are generally scarce as most interventions will have at least some effect or in some situations, as otherwise they would likely have been abandoned already[41]. Orthopedic surgeons believed that the active disinvestment initiative did not adequately distinguish in the heterogeneous group of etiologies that make up SAPS patients and felt that some patients could still benefit from surgery, which implies that they do not see SAPS as a good disinvestment candidate(i.e. may not entirely be low-value care). Although the concept of low-value care is well-known, a clear definition of what constitutes low-value care is missing as well as who decides what constitutes low-value care, which may depend on the perspective taken[42]. Hence, different stakeholders may have different views on what constitutes low-value care for their situation, as also found in our interviews. In this case, the healthcare insurer decided that most surgery for SAPS patients is low-value care and believed that costs related to this procedure could be saved or should be allocated to other procedures providing more value. The orthopedic surgeons, however, stated that it was not as clear-cut as some patients may benefit from surgery. The latter highlights the tension in perspectives between physicians that want to do as much as possible for their patients and healthcare policy-makers that need to make trade-offs in priority setting in the context of scarcity in healthcare spending.

There are important implications for future active disinvestment initiatives based on the results from this study, increasing our understanding how active disinvestment initiatives may or may not exercise their effect. The first is that an active disinvestment initiative initiated from a macro-level perspective needs to go together with additional strategies for implementation at micro-level. A crucial step for this implementation at micro-level is to create support from relevant stakeholders[43], with the present study identifying several specific factors that may inhibit stakeholders' support. Although the present study investigated only the perspectives of hospital sales managers and orthopedic surgeons, support from other relevant stakeholders (e.g., patients, general public) also have been shown to be essential for successful active disinvestment[12, 44-46]. Gaining support of all relevant stakeholders is, however, extremely difficult as there are often contrasting viewpoints so it will be very complex to design an active disinvestment initiative incorporating all of these views[41]. In our study, most participants were not against active disinvestment. Still, they highlighted several reasons why they should have been involved from the start in a policy change to (partially) stop reimbursement e.g., to ensure correct interpretation of scientific evidence and prioritize initiatives with the most significant cost-saving potential. Such early engagement of relevant stakeholders and transparency of the designing process will therefore create a more nuanced strategy that will enhance the degree of support, thus increasing the possibility of successful active disinvestment[41, 47]. The necessity of stakeholder engagement also has been emphasized in various other studies on e.g., priority setting and other de-implementation strategies[8, 43].

Another implication is that contextual factors will affect the impact of any active disinvestment initiative, such as fear of losing revenues and patient preferences as found in the present study. As a policy change to stop reimbursement will not influence such factors, active disinvestment initiatives should always be paired with other initiatives appealing to the more intrinsic motivation of clinicians such as clinical decision support, performance feedback, patient-oriented educational materials and other interventions that aim to change clinician behaviour[48-50]. Therefore, future "top-down" policy changes, such as an active disinvestment initiative, should always be combined with "bottom-up" (e.g., physician-oriented) co-interventions in order to maximize its effectiveness and increase to possibility for success[51, 52]. Additionally, future active disinvestment initiatives must be aligned with pre-existing theories, such as basic economic theory, and consider theoretical frameworks on e.g., priority setting and/or de-implementation[35, 53] as their implications largely overlap. Future research should further explore the effectiveness of active disinvestment initiatives, while taking into account these co-interventions, incorporate the perspectives of patients and develop more specific theoretical frameworks to facilitate understanding how active disinvestment influences (clinical) decision-making. Additionally, future studies must focus on creating a shared view on low-value care and the process around active disinvestment, so that all stakeholders have a uniform perspective in approaching this concept and can start working on initiatives to reduce low-value care.

CONCLUSION

In conclusion, this study showed that two overarching themes negatively influenced the support for and effect of the active disinvestment regarding SAD surgery for SAPS. Hospital sales managers in particular felt it represented a "Too small piece of the pie" while orthopedic surgeons believed "They got it wrong". Future active disinvestment initiatives should engage all relevant stakeholders at an early stage to gain support, ensure correct interpretation of the evidence and clear definition of the targeted procedures and should target low-value procedures that have sufficient saving-potential to increase the possibility of success

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Appendix A. Semi-structured interview guides.

1. Hospital sales managers.

Welcome
Welcome participant and thank participant for participation in interview.

- Verbal consent for participation in study.
- Short personal introduction.
- Characteristics participant

Introduction
- Research about de-implementation of low-value care.
- Research question: effectiveness of an active disinvestment strategy for subacromial pain syndrome.
- Goal interview: to explore how this de-implementation strategy influenced decision-making around surgery from perspectives of relevant stakeholders.

Major topics
- Explanation disinvestment strategy
- Negotiation process between insurers and healthcare providers.
- Attention given to active disinvestment strategy during negotiations.
- Consequences of active disinvestment strategy for hospital.
- Perceived effect of active disinvestment strategy on clinical decision-making

Closing
- Other relevant things to discuss?
- Additional thoughts and/or questions?
- Other relevant stakeholders?

Thank respondent and finish interview

2. Orthopedic surgeons.

Welcome
Welcome participant and thank participant for participation in interview.

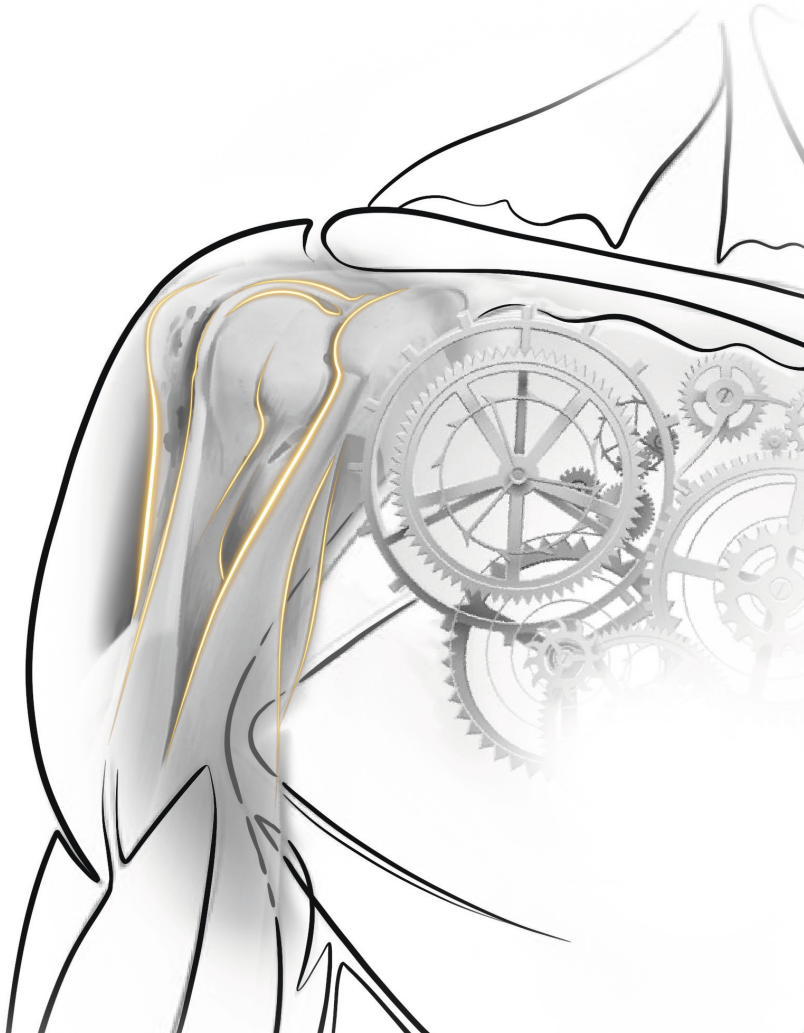
- Verbal consent for participation in study.
- Short personal introduction.
- Characteristics participant

Introduction
- Research about de-implementation of low-value care.
- Research question: effectiveness of an active disinvestment strategy for subacromial pain syndrome.
- Goal interview: to explore how this de-implementation strategy influenced decision-making around surgery from perspectives of relevant stakeholders.

Major topics
- Explanation disinvestment strategy
- Treatment strategy for SAPS.
- Perspectives about active disinvestment strategy.
- Perceived effect of active disinvestment strategy on clinical decision-making.
- Other relevant factors.

Closing
- Other relevant things to discuss?
- Additional thoughts and/or questions?
- Other relevant stakeholders?

Thank respondent and finish interview



Chapter 8

Thesis Summary and Future Perspectives

Clinical decision-making is a complex and ever-evolving process that integrates clinical expertise and scientific evidence to ensure optimal patient care. When diagnosing shoulder complaints, clinicians often strive to pinpoint a specific anatomical substrate to target treatment effectively. However, in most cases, the exact anatomical cause of shoulder complaints remains unclear and the complaints have a multifactorial origin with complex interrelationships among the contributing factors[1]. Clinicians therefore face several challenges when diagnosing and treating shoulder complaints both with well-defined pathological entities (e.g. neurogenic scapular winging) and those lacking clear anatomical origins (e.g. Subacromial Pain Syndrome (SAPS)) and this thesis aims to contribute to understanding the factors that shape and guide clinical decision-making in the management of shoulder pain and motion syndromes. In this chapter, the findings of the studies that were conducted in this thesis are summarized and placed in the context of system thinking. System thinking tries to make sense of complexities in the world by looking at problems as a whole, focusing on the interrelationships between components rather than splitting them up into parts[2]. Aristotle already described this concept over 2.000 years ago, stating "the whole is greater than the sum of its parts"[3]. Improving only one part does not necessarily result in an improvement of the whole system. It seems particularly well-suited to apply system thinking to the management of shoulder complaints as there are only few linear anatomical causes and complaints relationships, and mostly a complex variety of factors contributing to the shoulder complaints.

SUMMARY OF MAIN FINDINGS

Chapters 2 and 3 focused on a rare shoulder disorder with a well-defined pathological entity that has a clear causal relationship with the complaints, i.e. neurogenic scapular winging. Due to its rarity, clinicians may not always recognize this condition leading to misdiagnosis and inappropriate treatment strategies and most scientific studies have small sample sizes that may not be generalizable to the general population or rely heavily on expert opinion[4]. The systematic review of current evidence on both surgical (i.e. tendon transfer surgery) and non-surgical management of neurogenic scapular winging in **Chapter 2** revealed that a significant proportion (i.e. 22-79% after a median follow-up of 72 months) of the non-surgically managed patients experience persistent winging[5]. Additionally, tendon transfer surgery was shown to be an effective surgical treatment for patients unresponsive to non-surgical management as data pooling showed that patients improved significantly after surgery in active range of motion, had lower pain scores, and substantial improvements in subjective shoulder scores (i.e. American Shoulder and Elbow Surgeons and Constant Score) after a median follow-up of six years. The systematic review also highlighted that data on long-term clinical outcomes was still lacking, which is particularly important for this patient population as most patients with neurogenic scapular winging are relatively young at disease onset. To address this knowledge gap, **Chapter 3** evaluated outcomes after a minimum follow-up of 10 years of patients with neurogenic scapular winging due to long thoracic nerve (LTN) palsy who underwent pectoralis major (PM) transfer augmented with an achilles tendon allograft[6].

Our study is the first to show that functional improvements in these patients persisted for the majority (81%) of patients and were associated with a better quality-of-life. However, we also found a relatively high recurrence rate (33%) in our case series, possibly caused from graft stretching over time or presence of more extensive and complex peripheral nervous system disorders (e.g. neuralgic amyotrophy)[7]. The results of these studies offer valuable insights to guide clinical decision-making, highlighting a potential treatment option for persistent symptomatic scapular winging. If surgical treatment is discussed, the patient as a whole (i.e. holistic) should be taken into account, honoring personal wishes and needs as well as expectations on outcome while also taking into account patient-related factors such as lifestyle, comorbidities, and psychosocial influences.

Clinical decision-making for most (non-traumatic) shoulder complaints is, however, more complicated, primarily due to the absence of a linear single anatomical cause and consequently a clear treatment target. In recent years, research has increasingly recognized that musculoskeletal complaints often do not align with anatomical changes observed by imaging modalities (e.g. radiograph, ultrasonography, magnetic resonance imaging)[8, 9]. Such changes may simply reflect physiological processes related to ageing[10-15]. This realization has prompted a shift in focus beyond purely anatomical considerations towards other factors such as, psychosocial aspects, pain sensitization, movement complexity and proprioception[15-18]. In other words, the patient should be considered as a whole, i.e. an interconnected system, where numerous of complex factors and interactions (e.g. pain sensitization) influence shoulder complaints, and where improvement of only one component does not necessarily result in improved outcomes for the patient. In this context, proprioceptive decline is also a relevant example, which is associated with several shoulder disorders, including rotator cuff disease, frozen shoulder, and subacromial pain syndrome (SAPS[19-21]). Interestingly, it is also associated with poor rehabilitation- and surgical outcomes, highlighting its clinical importance[21-23]. Reflecting on findings we found with ageing (e.g muscle atrophy, molecular changes of muscle, decline in proprioception) it is essential to comprehend the normal dynamic variability in the system, like the natural decline of proprioception with increasing age in healthy individuals, before considering a deviation of proprioception and its role in shoulder pathology or treatment[10-15]. In **Chapter 4** we therefore quantified the extent to which age was associated with joint position sense in 120 asymptomatic participants between 18 and 70 years old and found no age-related decline in the asymptomatic shoulder[14]. This serves as a valuable benchmark, as proprioceptive deficits seem to be associated with disease rather than that there is an interrelationship with normal ageing. It remains uncertain whether these deficits are a cause or consequence of shoulder pathology and our findings underscore the need for further exploration into the role of proprioception and its interaction with other factors to better understand shoulder complaints, its potential as a treatment target and its ability to predict poor treatment outcomes- ultimately aiding clinicians in making more informed decisions[23].

The subacromial pain syndrome (SAPS) is a complex shoulder disorder with an unclear etiology. Formerly known as impingement syndrome, it was once thought to result from impingement of the rotator cuff tendons and subacromial tissues by the coracoacromial arch suggesting a single cause that we could treat by surgery. This linear cause-disease relationship turned out to be an overly simplistic view not reflecting the true etiology of SAPS, as evidenced by high-quality evidence showing that subacromial decompression surgery provides no clinically meaningful improvement in pain, shoulder function or quality-of-life[24]. As a result, it is now more appropriately described as a pain syndrome, where shoulder complaints are considered within a broader context. The unclear etiology of SAPS complicates clinical decision-making, making it challenging to identify specific treatment targets and creating uncertainty regarding the most effective treatment strategy. Adding to the complexity of which treatment strategy should be chosen is that also multiple factors outside the patient with complaints may influence treatment decisions. For instance, some clinicians may not be aware of the strong scientific evidence against the effectiveness of SAD surgery, resulting in continued belief in the historical perspective. The timely adoption of evidence-based practices is essential to provide patients the most optimal state-of-the-art treatment as continuing ineffective treatments not only deprives patients of optimal care but also risks potential harm as well as increases healthcare expenditure[25]. **Chapter 5** showed how the publication of two high-quality randomised controlled trials led to a notable decrease in SAD surgeries for SAPS patients, underscoring the potential for new evidence to influence the context of clinical decision-making. However, there was significant variation among participating hospitals and a potential shift in coding practices, suggesting that more factors than only the publication of evidence are needed to change clinical decision-making. Thus, treatment decision-making could also be seen as a complex system with multiple interconnected factors. An improvement in one of these factors, like available high-quality evidence, does not automatically lead to better clinical decision-making, as it is also influenced by other elements such as surgeon as well as patient beliefs on potential effect of a treatment.

Chapter 6 further examined factors that drive the decision-making of orthopedic surgeons regarding SAD surgery for SAPS patients in the context of a wider system, using identical patient scenarios so that treatment decisions are primarily influenced by surgeon factors such as beliefs, personal preference, and their weighing of the scientific evidence. Previous studies have shown significant variability in surgical decision-making for identical clinical scenarios both among different surgeons and within individual surgeons over time, suggesting that the clinical decision-making process is not only influenced by patient-related factors but also by a much broader context (e.g. surgeon preferences)[26-29]. Consistent with this, significant disparities among orthopedic surgeons in their decision-making to perform SAD surgery for SAPS was shown in Chapter 6, which mainly arose from differences in the perceived benefit of SAD surgery. Additionally, patient-related factors, such as long symptom duration, progressive complaints, failure of physical therapy, and patient expectations also played a significant role in the decision to perform surgery. This suggests that surgeons (and also patients), may have unrealistic expectations of SAD surgery and struggle to refrain from

surgery when patients have persistent symptoms or when alternative treatments have failed. Furthermore, action bias, i.e. the general preference to take some kind of (surgical) action rather than to refrain from surgical treatment, might also play a role, particularly when there is uncertainty on the best treatment strategy[30, 31]. The results of this study therefore underscore the variety of interrelated factors that influence treatment decisions, and that to improve this system as a whole it is not enough to target just one component such as publication of objective evidence and/or clinical guidelines but we need multifaceted (de-implementation) strategies that will improve multiple components and their interactions. To effectively change clinical decision-making it is not sufficient to provide sound clinical evidence; it is crucial to target behavioural surgeon- and patient factors such as surgeons' beliefs, perceptions and patient expectations and how they interact or even compete with the clinical evidence and guidelines.

Numerous de-implementation strategies have been launched to steer clinical decision-making away from low-value care procedures, with varying degrees of effectiveness[32, 33]. Among these, active disinvestment strategies (i.e. reimbursement withdrawal) are considered the most effective but also most challenging to implement, with less than half of the initiatives proving successful to date[34, 35]. To understand the underlying mechanisms of how these initiatives may influence clinical decision-making, these should also be considered in the wider system in which they are implemented. In **Chapter 7**, we therefore interviewed orthopaedic surgeons and hospital sales managers as important stakeholders influencing decision making around SAD surgery to examine the impact of an active disinvestment strategy conducted by one of the largest healthcare insurers in the Netherlands to reduce the use of SAD surgery for SAPS. The views of these stakeholders will shed light on several factors in the wider healthcare system that determine whether the intervention will reach its intended effect, for which support by these stakeholders is needed. Our findings show that hospital sales managers were not motivated to alter hospital practices as the disinvestment strategy was not integrated into overall hospital agreements, offered limited saving potential, and required a disproportional amount of effort from hospital staff. Similarly, orthopaedic surgeons did not adjust their clinical decision-making in response to the disinvestment strategy, as they believed it misinterpreted scientific evidence and guidelines and diminished their professional autonomy. Thus, the result of this study strengthened our understanding of how various stakeholders in the healthcare system determine the final impact of active disinvestment strategies. Top-down disinvestment strategies initiated from a macro-level perspective, such as by governmental institutions or a healthcare insurer, are unlikely to affect clinical decision-making without first ensuring support by crucial stakeholders. It must therefore be accompanied by additional de-implementation strategies, targeting the whole system including local and institutional stakeholders, to increase the likelihood of successful disinvestment.

A holistic approach in clinical decision-making deviates from classical "linear" thinking and is more aligned with modern system thinking, offering a valuable framework for enhancing

clinical decision-making and improving patient care in the context of shoulder disorders. The multifaceted nature of managing these conditions illustrates the complex interplay of numerous factors influencing clinical decision-making, such as patient factors, clinician factors, the ever-evolving scientific evidence and clinical guidelines, and the role of healthcare organizations and governmental policies. By viewing clinical decision-making as part of a complex interrelated system rather than a series of isolated issues (e.g. MRI "changes"), the holistic approach facilitates system thinking and thus stimulates a complete understanding of the factors that influence impact on patient outcomes and treatment decisions. The latter encourages collaboration among all stakeholders involved within the healthcare system. Embracing a system-oriented approach can help clinicians to better navigate the complexities around the treatment of any complex disorders. By recognizing the broader context of each patient's condition, clinicians can make more informed decisions, ultimately leading to more effective, patient-centered care and better outcomes for individuals suffering from shoulder disorders, which, in turn, benefits society as a whole.

FUTURE PERSPECTIVES

Concerning shoulder pain syndromes and motion disorders, clinical science provided evidence to slowly change clinical practice, although still a knowledge gap exists on what the best clinical decision tree is for a specific patient. The latter implicates, that clinicians are operating under a certain level of uncertainty, although this is often not well recognized[36]. To advance our knowledge, future research on shoulder pain and motion disorders should include the earlier discussed holistic patient assessment, which considers the interaction of a multitude of patient factors such as physiological ageing, proprioception, movement complexity, and pain sensitisation alongside traditional anatomical evaluations. This approach should focus on the complexity of interconnections of these elements rather than treating isolated components, as doing so may not improve patient outcomes. Chapter 4 provides a foundational reference for future research which could explore particularly how proprioception in interaction with other factors relates to shoulder disorders. Similarly, the role of motor complexity in shoulder complaints warrants further investigation. The complexity of repetitive movement trajectories, serves as an indicator of motor redundancy, reflecting the overall "health" of the motoric system[15]. Decreased motor complexity has already been linked to SAPS, highlighting its potential clinical significance[37]. However, as with proprioception, it remains unclear whether this is a causal factor or a result of the condition. Even more the interaction with other factors such as pain sensitisation should be considered to understand shoulder pain and symptoms. Previous studies already showed an association between shoulder pain and sensitisation of the nervous system, i.e. an increased responsiveness to pain stimuli due to altered somatosensory perceptions[38]. This pain sensitisation may contribute to the chronicity of shoulder pain and complicate treatment strategies. Gaining a deeper understanding of such systemic factors and their interrelationship will aid clinical decision-making and could potentially reveal new treatment targets.

Future research should also incorporate factors beyond patient-related aspects, including clinician-related factors, healthcare organisational dynamics and governmental healthcare policies. By examining how clinicians bias towards evidence and personal preference based on past outcomes shape treatment decisions, we can gain insight into the underlying mechanisms that perpetuate their persistent use of low-value care procedures at group level. For instance, action bias can drive clinicians to opt for more aggressive (e.g. surgical) interventions even when evidence suggests otherwise. Gaining a deeper understanding of such bias will guide the development of targeted interventions that reshape clinical decision-making and promote evidence-based practice by reframing in-action into an active monitoring strategy[39]. Moreover, it is essential to establish a shared understanding of low-value care, as stakeholders- including patients, clinicians, healthcare organisations and policymakers- often have different perspectives shaped by their own assessment of costs, benefits and harms[31]. Lastly, a deeper exploration of how healthcare organisational dynamics and policy changes, such as resource allocation, impact clinical decision-making is crucial for improving treatment outcomes in shoulder pathology. Understanding the interplay between the available resources and clinical practice may help identify barriers to implementing evidence-based treatments and reveal opportunities for optimising healthcare expenditures. By integrating these broader factors into future research, we can better navigate the complexities of clinical decision-making in shoulder disorders, ultimately enhancing patient-centered approaches and improving treatment outcomes for individuals with shoulder complaints.

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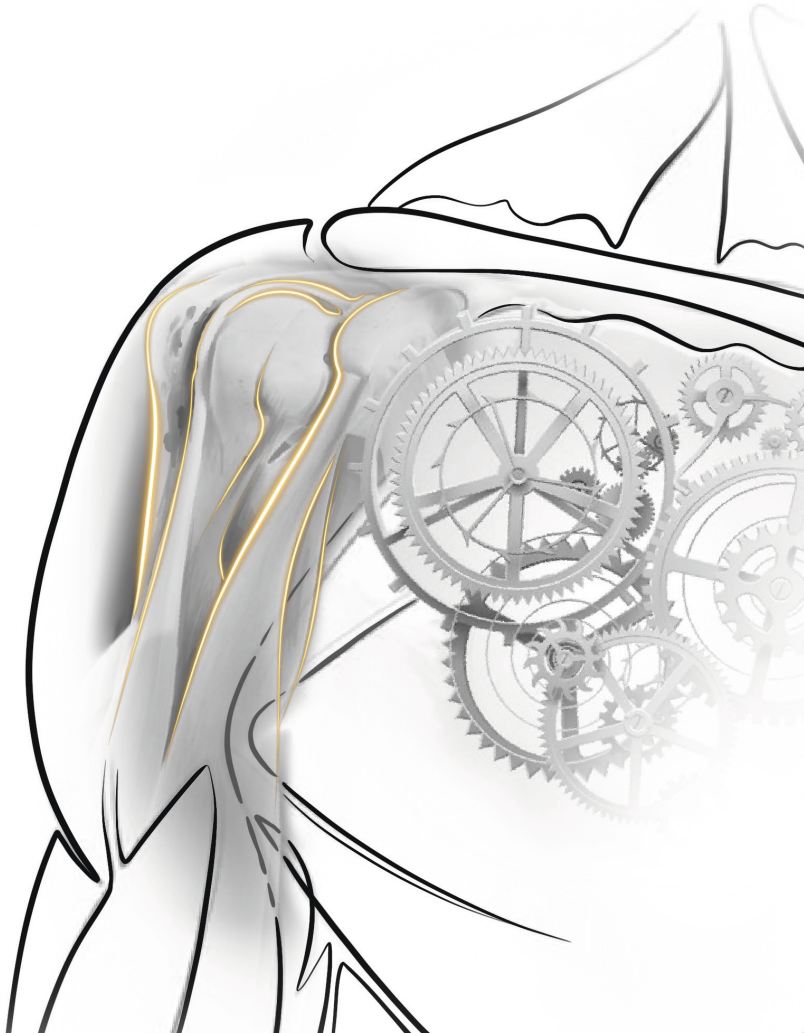


Chapter 9

Layman's Summary in Dutch (Samenvatting in het Nederlands).

Klinische besluitvorming is een complex en dynamisch proces waarin de klinische expertise van de arts en wetenschappelijk bewijs samenkomen om de patiënt de best mogelijke zorg te bieden. Bij het stellen van een diagnose bij schouderklachten proberen artsen vaak een anatomisch substraat te identificeren om een gerichte behandeling toe te kunnen passen. In veel gevallen blijft echter de exacte oorzaak van schouderklachten onduidelijk en blijken de klachten het resultaat van meerdere onderling verbonden en soms nog onbekende factoren. Artsen staan daarom voor verschillende uitdagingen bij het diagnosticeren en behandelen van schouderklachten, zowel bij aandoeningen met een duidelijke etiologie (bijvoorbeeld neurogene scapula alata) als bij aandoeningen zonder duidelijke etiologie (bijvoorbeeld het subacromiaal pijn syndroom (SAPS)). Dit proefschrift richt zich op het beter begrijpen van de factoren die de behandeling van schouderpijn en bewegingssyndromen beïnvloeden. Het doel is om inzicht te bieden in de factoren die artsen nodig hebben om weloverwogen beslissingen te nemen bij de behandeling van schouderklachten, zowel wanneer de oorzaak duidelijk is als wanneer deze onbekend blijft.

Samenvatting resultaten proefschrift.

De eerste twee studies van dit proefschrift richten zich op een zeldzame schouderaandoening met een duidelijk anatomisch substraat, namelijk neurogene scapula alata. Dit is een aandoening waarbij het schouderblad abnormaal af staat van de rug ("vleugel scapula") ten gevolge van een zenuwbeschadiging. Omdat de aandoening weinig voorkomt is er weinig wetenschappelijke informatie beschikbaar over de uitkomsten van zowel niet-chirurgische (bijvoorbeeld fysiotherapie of brace-behandeling) als chirurgische behandelingen (een spierpeestranspositie), wat de klinische besluitvorming bemoeilijkt. In **Hoofdstuk 2** werd daarom een systematisch literatuuronderzoek uitgevoerd om de resultaten van niet-chirurgische en chirurgische behandeling van een neurogene scapula alata te bundelen. Uit zes studies (kwaliteit score artikelen: laag tot middelmatig) bleek dat een aanzienlijk percentage (22-79%) van de niet-chirurgisch behandelde patiënten aanhoudende klachten blijven ervaren na een mediane follow-up van 6 jaar. Daarnaast toonde deze studie aan dat een spierpeestranspositie een effectieve chirurgische behandeling is voor patiënten die niet reageren op niet-chirurgische therapieën. Zeventien geïncludeerde studies (met kwaliteitscores variërend van laag tot middelmatig), waarin de scapula alata door middel van een peestranspositie chirurgisch werd behandeld, lieten zien dat deze patiënten na een mediane postoperatieve follow up van vier jaar lagere pijnscores hadden dan voor de operatie, en zowel de schouderfunctie als functionele scores (zoals Constant-Murley score en American Shoulder and Elbow Surgeons score) verbeterden.

Uit dit literatuuronderzoek bleek ook dat er weinig bekend is over de lange termijn uitkomsten van chirurgische behandeling bij neurogene scapula alata. Dit heeft echter belangrijke implicaties voor de klinische besluitvorming, aangezien de aandoening zich doorgaans op relatief jonge leeftijd manifesteert bij patiënten. Om deze kenniskloof te dichten, evalueerde **Hoofdstuk 3** de lange termijn uitkomsten van een pectoralis major peestranspositie bij

patiënten met een scapula alata als gevolg van uitval van de nervus thoracicus longus met een minimale follow-up van 10 jaar. Deze observationele studie met 15 patiënten liet zien dat bij de meerderheid van de patiënten (81%) de postoperatieve functionele verbeteringen op de lange termijn behouden bleven, en deze verbeteringen waren geassocieerd met een hogere kwaliteit van leven. Er werd echter ook een relatief hoog recidiefpercentage (33%) van scapula alata waargenomen, wat mogelijk verklaard kan worden door achteruitgang van de tonus van de pectoralis major spiergraft in de loop van de tijd, of de aanwezigheid van meer uitgebreide zenuwuitval, wat niet voldoende kan worden gecompenseerd door alleen de pectoralis major.

Klinische besluitvorming voor de meeste schouderklachten is echter een stuk complexer, voornamelijk door het ontbreken van een duidelijk anatomisch substraat, waardoor er ook een gebrek aan een specifiek behandeldoel bestaat, anders dan pijn vermindering. In de afgelopen jaren is steeds duidelijker geworden dat musculoskeletale klachten vaak niet corresponderen met afwijkingen die gevonden worden bij beeldvorming, zoals echografie of MRI. Hierdoor is de focus van wetenschappelijk onderzoek verschoven, waarbij naast anatomische factoren ook andere elementen zoals bijvoorbeeld psychosociale invloeden, pijnsensitisatie en proprioceptie worden meegenomen in het diagnostisch traject. Dit benadrukt hoe belangrijk het is de patiënt als geheel te beschouwen, waarbij vele verschillende factoren invloed hebben op schouderklachten. Verminderde proprioceptie wordt bijvoorbeeld al geassocieerd met diverse schouderaandoeningen zoals frozen shoulder en SAPS, en wordt daarnaast ook gezien als mogelijke negatieve voorspeller voor de uitkomsten van chirurgie of revalidatie. Om de rol van dergelijke factoren bij het ontstaan en behandelen van schouderklachten beter te begrijpen, is het belangrijk om eerst de normale variatie in een gezonde populatie te onderzoeken. **Hoofdstuk 4** kwantificeerde daarom de mate waarin leeftijd geassocieerd was met een verminderde proprioceptie in 120 gezonde vrijwilligers tussen de 18 en 70 jaar. De proprioceptie werd gemeten door middel van 10 verschillende armbewegingstaken, waarbij gewrichtspositie en reproductie daarvan werden geëvalueerd. In deze studie werd geen leeftijdsgebonden achteruitgang in de asymptomatische schouder waargenomen. Een verminderde proprioceptie lijkt dus geassocieerd te zijn met schouderpathologie en niet het gevolg van een normaal verouderingsproces. De data van deze gezonde controle groep kunnen als een benchmark voor toekomstig onderzoek beschouwd worden, om de rol van proprioceptie bij het ontstaan en behandeling van schouderaandoeningen te onderzoeken.

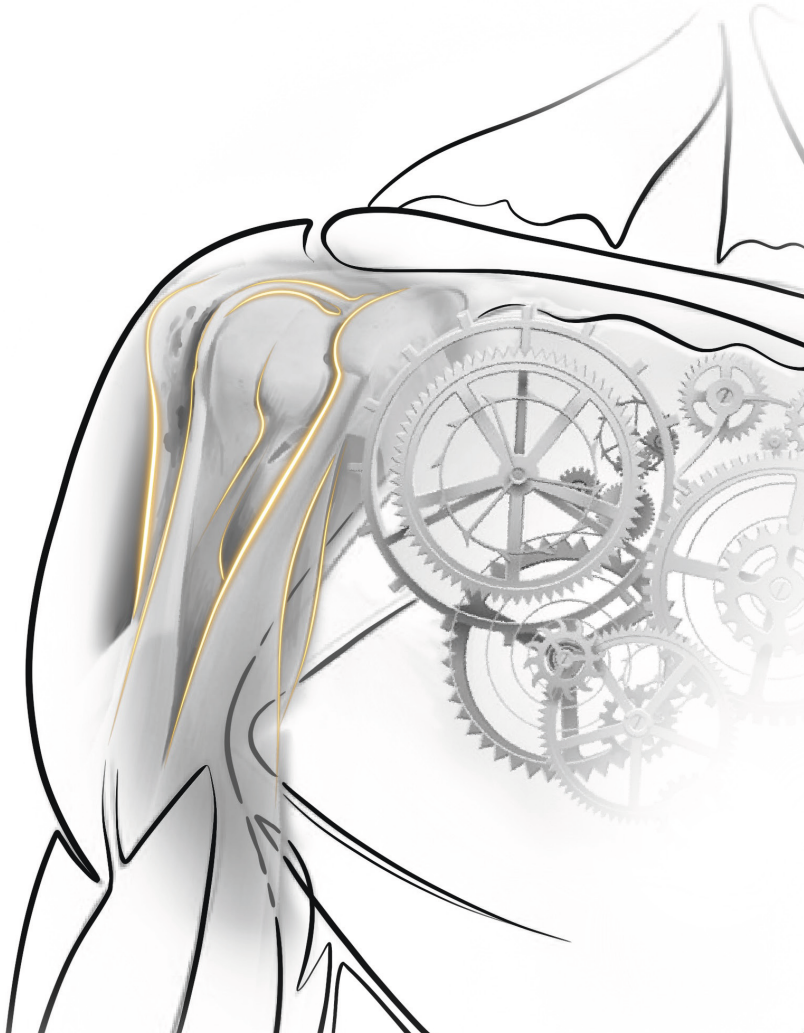
SAPS is een complexe schouderaandoening met onduidelijke etiologie. Lange tijd werd gedacht dat patiënten met SAPS pijn ervaarden door inklemming van de schouderspieren onder het acromion ("impingement"), wat suggereerde dat er een oorzaak was die met een operatie (een subacromiale decompressie) kon worden behandeld. In 2018 lieten twee gerandomiseerde studies echter zien dat deze operatie niet leidt tot betere resultaten bij de behandeling van SAPS, waardoor deze ingreep als niet zinvol wordt beschouwd en er sterke aanbevelingen tegen het gebruik ervan zijn opgenomen in (inter)nationale richtlijnen. De onduidelijke etiologie van SAPS bemoeilijkt de klinische besluitvorming, aangezien erg geen eenduidige effectieve behandeling beschikbaar is. Dit leidt ertoe dat sommige artsen vasthouden aan

verouderde strategieën, zoals de subacromiale decompressie, ondanks de slechte effectiviteit op groepsniveau. Onderzoek naar de mate waarin wetenschappelijk onderzoek wordt geïntegreerd in de klinische praktijk is van belang, omdat ineffectieve behandelingen niet alleen nadelig zijn voor patiënten, maar ook leiden tot onnodige zorgkosten en langere (onnodige) wachtlijsten voor patiënten die wel een chirurgische behandeling nodig hebben. **Hoofdstuk 5** onderzocht daarom wat het effect was van de publicaties van de twee grote gepubliceerde onderzoeken uit 2018 op het aantal uitgevoerde subacromiale decompressies in zes ziekenhuizen uit vijf verschillende landen gedurende de periode van 2016 tot 2020. De publicatie van de onderzoeken was geassocieerd met een significante afname van 2% per maand in het aantal uitgevoerde subacromiale decompressies. Er was wel een aanzienlijke variatie tussen de deelnemende ziekenhuizen. Desondanks illustreert deze studie het potentieel van wetenschappelijke publicaties om de klinische beslissingen van artsen te beïnvloeden.

In **Hoofdstuk 6** werd verder onderzoek gedaan naar de factoren die bijdragen aan de besluitvorming van Nederlandse orthopeden bij de keuze om wel of geen subacromiale decompressie uit te voeren bij patiënten met SAPS. Hierbij werden vier identieke patiëntscenario's gebruikt welke werden voorgelegd aan 52 orthopeden en 12 orthopeden in opleiding, zodat de behandelbeslissingen niet werden beïnvloed door verschil in patiëntfactoren maar voornamelijk door factoren rondom de chirurg, zoals hun persoonlijke overtuiging, voorkeuren en interpretatie van de wetenschap. In deze studie vonden we grote verschillen in de besluitvorming van orthopeden met betrekking tot het wel of niet uitvoeren van een subacromiale decompressie. Deze verschillen waren voornamelijk een gevolg van uiteenlopende opvattingen over het te verwachten effect van de operatie. Daarnaast speelden verschillende interpretaties van het belang van andere factoren een belangrijke rol bij de beslissing om een operatie uit te voeren, zoals een lange duur van klachten, het falen van conservatieve behandeling (zoals fysiotherapie) en de wens van de patiënt. Dit suggereert dat chirurgen (en mogelijk ook patiënten) onrealistische verwachtingen hebben van een subacromiale decompressie operatie en moeite hebben om van een operatie af te zien wanneer er langdurige klachten bestaan en niet-operatieve behandelingen falen. De resultaten van dit onderzoek laten zien dat alleen de aanwezigheid van goed wetenschappelijk onderzoek onvoldoende is om de klinische besluitvorming te veranderen. Het is ook noodzakelijk om gedragsmatige factoren van zowel artsen als patiënten te beïnvloeden, zoals hun persoonlijke overtuigingen, verwachtingen en de manier waarop zij wetenschappelijk bewijs interpreteren en toepassen.

Er zijn veel verschillende strategieën om de klinische besluitvorming te beïnvloeden en het gebruik van ineffectieve behandelingen te ontmoedigen. De-investeringsstrategieën, zoals het stopzetten van de vergoeding van niet effectieve behandelingen, worden in de wetenschappelijke literatuur als het meest effectief beschouwd, maar het is onduidelijk of zulke interventies daadwerkelijk de besluitvorming van artsen beïnvloeden. Om het gebruik van subacromiale decompressies als behandeling voor SAPS verder te ontmoedigen, is een van de grootste zorgverzekeraars van Nederland in 2020 met een de-investeringsstrategie

gestart, waarbij een aanzienlijk deel van de subacromiale decompressies bij SAPS niet langer wordt vergoed. In **Hoofdstuk 7** werd met semi-gestructureerde interviews onderzocht hoe deze de-investeringsstrategie de klinische besluitvorming van clinici beïnvloedt. Tien zorgverkopers en tien orthopeden uit twaalf verschillende ziekenhuizen in Nederland werden hiervoor geïnterviewd. Uit de resultaten bleek dat de kans op succes van een dergelijke de-investeringsstrategie grotendeels afhankelijk is van de steun van relevante stakeholders voor de strategie. De zorgverkopers in deze studie waren niet gemotiveerd om het ziekenhuisbeleid aan te passen, omdat de strategie niet werd geïntegreerd in de uiteindelijke contractafspraken met het ziekenhuis, weinig financiële impact had en een onevenredige hoeveelheid inspanning van het ziekenhuispersoneel vergde. Eveneens kon de strategie niet rekenen op de steun van orthopeden, omdat zij vonden dat er onvoldoende wetenschappelijk bewijs was ter ondersteuning van de strategie en dat deze hun professionele autonomie ondermijnde. De resultaten van deze studie tonen daarom aan dat een de-investeringsstrategie, geïnitieerd vanuit een top-down perspectief, niet succesvol zal zijn zonder de steun van relevante stakeholders. Dergelijke strategieën moeten daarom gepaard gaan met aanvullende interventies die zich richten op het hele systeem om de kans op een succes te vergroten.



Appendices

Acknowledgements

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Curriculum Vitae

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Curriculum Vitae

Timon Hermanus Geurkink was born on July 11th, 1993 in Zevenaar, The Netherlands, and spent most of his youth in Babberich. After graduating from Stedelijk Gymnasium Arnhem in 2011, he started his medical education at the University Medical Center Utrecht (UMCU). His love for sports from a young age led to an interest in musculoskeletal conditions, which he sought to explore more through research. This caused him to work with Dr. B.C.H. van der Wal during his medical study, where he became involved in research on 3D implants for hip and shoulder surgeries, resulting in multiple publications. In 2019, he started his PhD at the department of orthopaedics at the Leiden University Medical Center (LUMC), working under the supervision of Dr. P.J. Marang-van de Mheen and Prof. Dr. R.G.H.H. Nelissen. The results from his research are presented in this thesis. After gaining experience in musculoskeletal medicine, he decided in 2023 to broaden his medical knowledge by working as a resident not in training in the departments of cardiology and pulmonology at Diakonessenhuis Utrecht. In 2024, he was accepted into the general practice residency program at the UMCU, a new chapter that reflects his ambition to embrace the broader spectrum of healthcare.