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Hand

Tendon Transfers to Improve Grip and Pinch in Patients with Sporadic Inclusion Body Myositis

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Background: Sporadic inclusion body myositis (sIBM) is a rare and slowly progressive skeletal muscle disease that can cause hand dysfunction, which is a major source of disability. Tendon transfers have been reliably used to improve function in other neuromuscular settings. Given that sIBM patients often present with flexion impairments and mostly functioning extensors, we investigated the potential opportunity for tendon transfer surgery to improve hand dysfunction in sIBM patients.

Methods: We conducted a scoping review for studies of sIBM and tendon transfers, extracted descriptions of hand function and surgical technique, and recorded results in terms of hand function. We also conducted an institutional review board–approved survey with 470 participants to determine baseline patient-reported function and to determine participant perceptions and expectations for tendon transfer surgery to improve hand function in sIBM.

Results: We identified three published case reports on tendon transfers in sIBM patients with subjectively improved grip and pinch strength, but standardized measures of hand function or quality-of-life were not reported. Within the surveyed cohort, half of participants reported that they would consider surgery, yet only 8% had been referred to a hand surgeon. Fifty four percent of participants reported that they would consider surgery if there would be 1–2 years of benefit after surgery. All participants who would consider surgery also had significant upper extremity disability.

Discussion: Tendon transfer surgery has the potential to improve quality-of-life for sIBM patients, and there is significant patient interest in this approach. To objectively assess its efficacy, we propose conducting a surgical trial. *(Plast Reconstr Surg Glob Open 2023; 11:e5418; doi: 10.1097/GOX.0000000000005418; Published online 17 November 2023.)*

INTRODUCTION

Sporadic inclusion body myositis (sIBM) is a type of idiopathic inflammatory myopathy, a rare and slowly progressive skeletal muscle disease that typically affects patients between 45 and 70 years of age. This disease is associated with unique clinical features and uncertain etiology, as it is unknown if sIBM is primarily an autoimmune or degenerative myopathy.[1,2] Further, sIBM typically leads to disability without significantly increased mortality.[1] Symptoms typically present in middle or late age (>40 years),[2] with initial weakness in the quadriceps and/or long finger flexors,[1] eventually leading to difficulty standing up, increased risk of falling, and loss of pinch and grip strength. Another common presenting symptom is dysphagia.[3] Most sIBM patients would require a cane or wheelchair within their lifetime.[2] Diagnosis of sIBM is based on a combination of clinical and pathological features. Patients typically present with a unique clinical weakness pattern that includes chronic, progressive

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knee extension, and/or finger flexion weakness over the course of a minimum of 12 months. Patients can present with a normal or elevated creatine kinase level. Muscle biopsies can reveal pathological features of sIBM, which include endomysial infiltrates, rimmed vacuoles, and protein aggregates (amyloid or other proteins). To further support an sIBM diagnosis, patients are tested for serum anticytosolic 5-nucleotidase 1A antibody. However, the sensitivity and specificity of this serological test were 45% and 96%, respectively.[4–6]

During a clinical examination, patients with sIBM would display a unique hand weakness pattern. Grip and pinch impairments result from weakness of the extrinsic finger flexors [flexor digitorum profundus/superficialis (FDP/FDS) and flexor pollicus longus (FPL)].[7,8] Because the intrinsic muscles are relatively preserved, patients weakly grasp and pinch objects through thumb adduction and metacarpophalangeal flexion.[2] Volar forearm atrophy is also a key feature in sIBM because the finger and wrist extensor muscles are relatively spared until late into the disease. Hand dysfunction is subjectively reported to be a major source of disability for individuals with sIBM. To date, there are currently no Food and Drug Administration–approved treatments to halt or slow the rate of disease progression. Therefore, there is a significant unmet need to improve the quality-of-life for these patients. Given the unique pattern of finger and wrist flexion impairments with relative sparing of the extensors, we investigated the potential opportunity for tendon transfer surgery to improve hand dysfunction in sIBM patients. Tendon transfers have been safely and reliably used to improve function in neuromuscular and neurological settings, including Charcot-Marie-Tooth disease,[9–11] brachial plexus lesions,[12–14] Hirayama disease,[15] and stroke.[16]

Tendon transfer is a versatile tool in reconstructive surgery that takes advantage of muscle redundancy or expendability to restore muscle-tendon units (MTU). Tendon transfers were first used in 1882 to restore ankle dorsiflexion in children with weakness secondary to polio, and throughout the mid-1900s, several advances were made to advance tendon transfers within the upper extremity (UE).[16] The success of tendon transfer surgery depends on matching donor and recipient MTU force vectors and excursion. It is important to note that one tendon should be limited to one function. Furthermore, a donor MTU will lose one grade of strength on the Medical Research Council scale for muscle strength after transfer. However, this is balanced by the viscoelastic force (resistance to stretch of the MTU), line of tension, and MTU synergism. Additionally, the force of an MTU is proportional to the muscle cross-sectional area at rest; thus, certain muscles in the arm are more amenable to transfer due to a higher relative strength, such as the brachioradialis (BR). An ideal transfer imitates normal tendon insertion and tension. Multiple techniques, such as the Pulvertaft weave, achieve this goal via interweaving the donor and recipient tendons. After 2-4 weeks of immobilization post-transfer, the patient can begin mobilization.

Given the potential promise of tendon transfers to improve hand function, we conducted a scoping review and conducted an international survey to understand perceptions, current function, and expected function after a tendon transfer surgery in sIBM patients.

Takeaways

Question: Can tendon transfer surgery improve hand function for patients with sIBM?

Findings: Our scoping review revealed three publications describing outcomes of tendon transfers in sIBM patients. We surveyed 232 participants, and 54% would consider surgery if it provided 1-2 years of benefit. This goal is feasible; tendon transfers have been beneficial for at least 2 years for ideal surgical candidates.

Meaning: Tendon transfers show promise for improving hand function in sIBM patients.

METHODS

Scoping Review

Data Sources and Study Selection

The databases of PubMed, CINAHL, and MEDLINE were searched from inception to December 2022 for studies of inclusion body myositis and tendon transfers using the terms "inclusion body myositis" and "tendon transfer." These terms were searched together. Three investigators (C.H., A.R.B., S.B.) independently reviewed and assessed studies within the year 2023. For our search for relevant studies, we excluded review articles, articles written in a language other than English, articles with unavailable full text, articles that were not of the original article type (commentary, letter, etc), and articles not associated with sIBM or tendon transfer. In total, three studies were included in this study (Fig. 1).

Data Extraction and Synthesis

Standardized data abstraction was used to extract surgical technique and measures of hand function. The Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) reporting guideline was followed.[17]

Main Outcomes and Measures

The main outcome was improvement in hand function [defined as improvement in patient-reported outcome measures (PROMs), range-of-motion, pinch/grip strength, or muscle grade]. Information on indications, surgical approach, and tendon transfer techniques with postoperative protocol was also recorded.

Survey

An anonymous, institutional review board–approved survey was created to understand the perceptions and expectations that participants have of tendon transfer surgery in regard to improving hand function in sIBM. (**See appendix, Supplemental Digital Content 1,** which displays the patients myositis journey and burden of disease survey. **http://links.lww.com/PRSGO/C878**.) The survey was distributed worldwide to members of

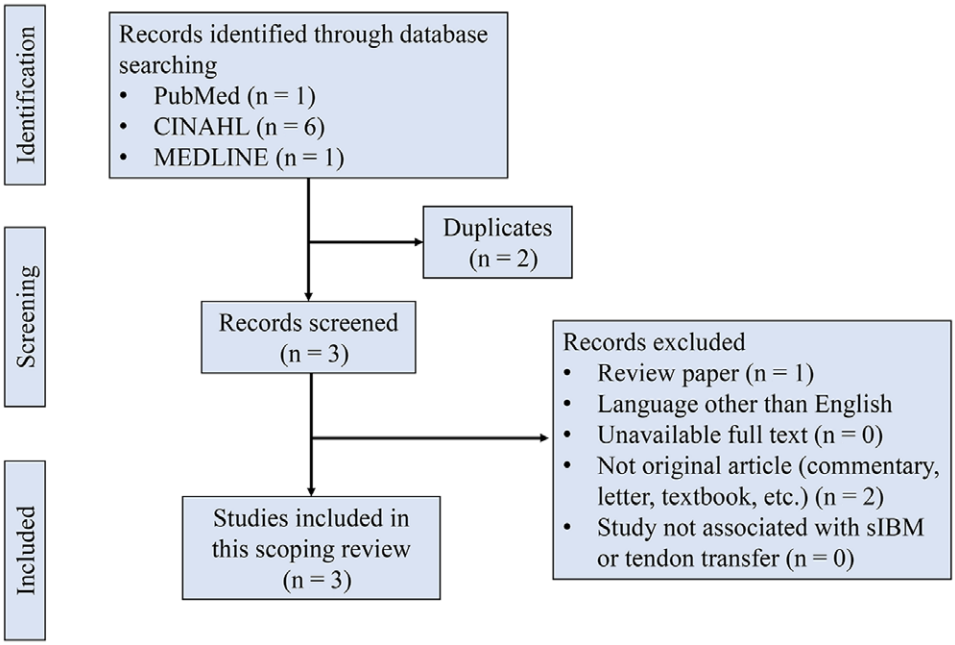

Fig. 1. Scoping review procedure and inclusion and exclusion criteria.

Myositis Support and Understanding (MSU), a patient advocacy group. The survey was piloted by a smaller subset of MSU members before being sent to the entire study population. Data were collected in a de-identified manner through REDcap. Over 4 weeks, 470 participants with a self-reported diagnosis of myositis completed the survey.

The survey collected demographic and clinical information from participants. Specifically, participants with sIBM were asked if they would consider tendon transfer surgery, and if so, how long of a benefit would justify the surgery. PROM metrics included PROMIS UE activities of daily living (ADL) difficulty, PROMIS self-efficacy (scores: emotions, 44.9 and symptoms, 41.2), and PROMIS physical function (score: 29.2). We also assessed if a simple query about subjective UE function (rate your upper extremity function compared with a completely normal upper extremity on a scale of 0%–100%) could help triage patients, and we defined this as the Single Assessment Numeric Evaluation (SANE) score.[18]

Statistical Analysis

Data were analyzed using the chi-squared test for categorical variables or the *t* test for continuous variables when appropriate. Multivariable regression analysis was used to identify factors associated with increased preference surgery and to compare outcome metrics. A *P* value of less than 0.05 was considered statistically significant.

RESULTS

Scoping Review

Our scoping review resulted in the identification of three case reports associated with sIBM and tendon transfer that were published in 2002, 2020, and 2021.[19–21] In all three cases, the indications for tendon transfer included limitations in grip or pinch strength that affected activities of daily living. All patients had subjective improvement in gross hand function, and in two cases, this was sustained for at least 2 years (Table 1). In all patients, wrist extensors were used to reconstruct finger flexors. Specific transfers included (1) BR to FDP and extensor carpi radialis longus (ECRL) to FPL transfers, (2) BR to FPL and ECRL to FDP, and (3) ECRL to FPL and ECU to FDP. Postoperatively, patients were immobilized for 4 weeks before starting hand therapy. Standardized measures of hand function or quality of life were not reported.

Table 1. Demographic and Surgical Information from Scoping Review

Case	Tendon Transfer Performed	Age (y)	Sex	Outcome	Sustained Time of Improvement
Waclawik and Rao[20]	BR to FPL and ECRL to FDP	70	Male	Improved finger flexion function with the ability to pinch and hold objects	2 years
Belward et al[21]	ECRL to FPL and ECU to FDP	56	Male	Pinch and grip improved from 0 to 0.5 kg pinch and 1 kg grip	Not mentioned
Thompson et al[19]	ECRL to FPL BR to FDP	71	Male	Thumb opposition improved from −4.0 cm to being able to contact the index fingertip	2 years

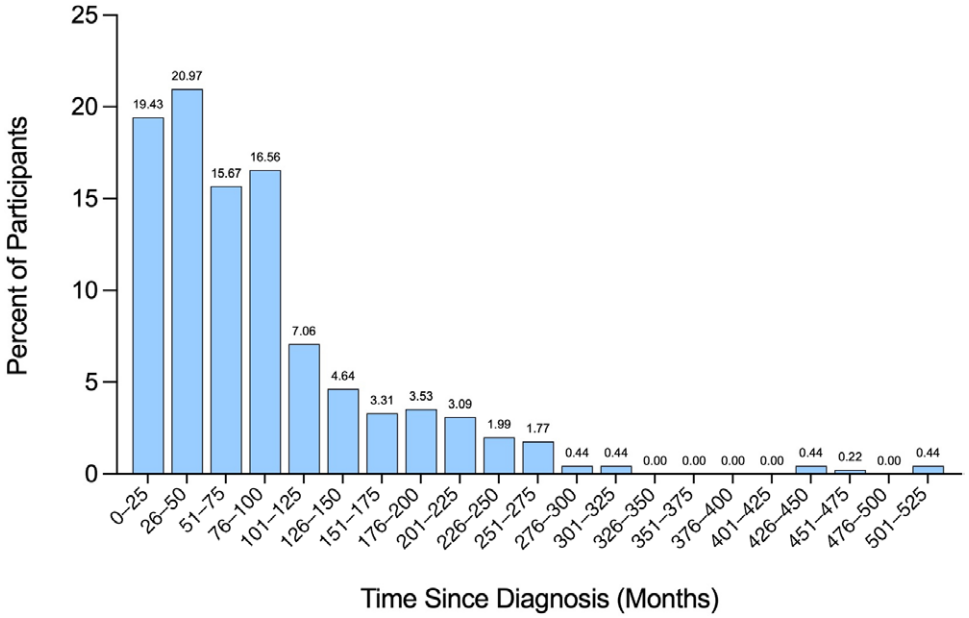

Fig. 2. Frequency of participant-reported time since sIBM diagnosis. Mean time since diagnosis is 86 ± 71.6.

Survey

Of the 470 participants who completed the survey, 232 participants reported a diagnosis of sIBM. Approximately 1200 individuals have self-reported a diagnosis of sIBM within MSU, demonstrating a survey response rate of 19.3%. The age distribution and time to diagnosis (median 86 months) of the sIBM participants are within a range similar to those reported previously and experienced in clinical practice. Of the 232 patients with sIBM, 193 patients completed the surgical questionnaire and the associated PROMs (Fig. 2 and Table 2).

Fifty three percent of sIBM participants would consider surgery. There was no significant difference regarding age, sex, race, or time to diagnosis between those who would consider surgery versus those who would not consider surgery (Table 3). There was a significant difference ($P = 0.041$) in SANE scores between those considering surgery compared with those who were not considering surgery. There were more participants considering tendon transfer surgery who reported the lowest interval of SANE scores (0–10) (worse dysfunction) than participants who were not considering tendon transfer surgery. The median SANE score for participants considering tendon transfer surgery was lower (30–40) than for participants who were not considering tendon transfer surgery (40–50) (Fig. 3). Of the participants who were open to surgery, 8% had seen a hand surgeon; of participants who were not considering surgery, only 3% had seen a hand surgeon. There was no difference between the two groups for the PROMIS self-efficacy (emotions and symptoms) and physical function scores. For patients who were considering surgery, 40% of participants expected 1–2 years of benefit, 47% expected 2+ years of benefit, and 13% of participants expected 12 months or less of benefit to consider surgery (Table 2).

The SANE score was self-reported by survey participants and is positively correlated with established outcome measures: PROMIS physical function [OR, 0.161 (95% CI, 0.135–0.188); $P \leq 0.001$] and PROMIS self-efficacy (symptoms) [OR, 0.061 (95% CI, 0.026–0.097); $P = 0.001$]. SANE Scores from 61 to 100 were significantly associated with an increased PROMIS self-efficacy (symptoms) score (Table 4).

DISCUSSION

In this study, we found that many survey participants with sIBM, especially those who reported more severe UE dysfunction, would consider tendon transfer if it could improve their quality of life. Our scoping review demonstrated a paucity of published evidence on the utility of tendon transfers in improving grip and pinch in sIBM. Tendon transfers are commonly used in orthopedic and plastic surgery to treat MTU dysfunction, but there are only three documented cases in which tendon transfers were not conducted in patients with sIBM.

The clinical features of sIBM are similar to those associated with a high median nerve injury, which is a condition for which tendon transfers are routinely performed. In sIBM, tissue equilibrium exists such that the tissue bed is mature and lacks obstacles to tendon gliding, such as scar tissue and edema. Surgery could include a combination of tendon transfers, depending on the exact pattern of weakness, with the goal of restoring composite grasp and tip pinch to improve lifting, carrying, and fine dexterity. A weak FPL muscle could receive a transfer from the BR, ECRL, or pronator teres muscles. A weak FDP muscle

Table 2. Demographic Characteristics and Surgical Considerations among Survey Participants

		SD
Total participants	193	
Age*		
40-49	3 (1.6)	
50-59	22 (11.4)	
60-69	65 (33.7)	
70-79	80 (41.4)	
80-89	22 (11.4)	
90-99	1 (0.5)	
Female*	91 (47.2)	
White*	183 (94.8)	
Time since diagnosis (mo)	86	71.6
UE ADL difficulty*	185 (95.9)	
Seen hand surgeon?*	11 (6)	
SANE score*		
0–10	16 (8.3)	8.3
11–20	28 (14.6)	14.6
21–30	27 (14.1)	14.1
31–40	29 (15.1)	15.1
41–50	23 (12.0)	12
51–60	20 (10.4)	10.4
61–70	18 (9.4)	9.4
71–80	17 (8.8)	8.8
81–90	10 (5.2)	5.2
91–100	4 (2.1)	2.1
Consider surgery*	102 (52.8)	
How long should benefit last to consider surgery? (N = 102)*		
<3 months	1 (1)	
3 to <6 months	3 (2.9)	
6 to <12 months	9 (8.8)	
1–2 years	41 (40.2)	
>3 years	48 (47.1)	
PROMIS self-efficacy (emotions)	44.9	8.1
PROMIS self-efficacy (symptoms)	41.2	6.4
PROMIS physical function	29.2	7.5

*Values listed are the number of participants with percentage in parentheses.

could receive transfers from the BR, ECRL, or ECU muscles. The ideal muscle to transfer is one that is grade 5 within the Medical Research Council scale for muscle strength. If tendon transfers are not an option, a free muscle graft can be considered, because this is the case with severe cases of Volkmann ischemic contracture, which has a clinical presentation similar to that of sIBM in that patients experience restricted digit and wrist flexion.[22]

In terms of approach, a modified Burkhalter transfer with end-to-side coaptation using a Pulvertaft weave would allow for range-of-motion and improvement in strength. The surgeon should set the tendon tension so that it mimics natural resting muscle length; tendons that are over- or under-tensioned will yield inefficient interactions between myosin and actin filaments, thus reducing the strength of the MTU. Surgeons should take advantage of muscle synergism, which, for example, can exist between wrist extension and finger flexion. This can allow for ease of retraining and improvements in strength in patients. Lengthy periods of immobilization are contraindicated because sIBM is a catabolic myopathy. In this case, "prehabilitation" may improve functional capabilities in patients in preparation for surgery, alongside early range-of-motion and strengthening that begins 2–3 weeks after surgery. Based on our previous clinical experience with tendon transfer surgery and the results of our scoping review, we suggest 2–3 weeks of total immobilization, 4 weeks of active range-of-motion, and 4 weeks of night splinting to protect the tendon junctures. A progressive plan with occupational therapy will assist in maintaining strength of muscles not involved in sIBM. Lastly, we recommend that the surgeon should avoid techniques that would limit joint range-of-motion, such as joint fusion or opponensplasty. One exception may be with the use of thumb interphalangeal joint fusion, because this could be a valuable strategy to facilitate tip pinch in patients who do not have a good donor for FPL restoration and who have preserved thenar muscles.

Based on the limited published reports and our clinical experience, we suspect that the patients with sIBM who would experience successful results after tendon transfer are those who have forearm finger and thumb flexor weakness with preserved finger, thumb, and wrist extensor strength and who are willing to participate in hand therapy before and after surgery. Although sIBM progresses slowly over several years for most patients, a subset of patients are fast progressors. Patients' disease progression can be identified by the history of their progression rate. Additionally, peripheral flow cytometry can identify fast progressors by those who have T-LGL clonal expansion—greater clonality is associated with a faster rate of progression.[23] These fast progressors should not be offered surgery because the benefit will be limited once the forearm extensor muscles are involved. Furthermore, all patients will be evaluated on a case-by-case basis to determine if they are good candidates for tendon transfer.

Although sIBM is a progressive disease without treatment, tendon transfer surgery can function as a "palliative" surgery that can improve quality-of-life. If tendon transfer is performed on ideal surgical candidates, patients may experience benefits for up to 2 years (or longer).[19,20] Importantly, our survey of patients aligns with this and shows that 54% would consider surgery if there were 1–2 years of benefit. Given the outcomes reported with the literature and given our clinical experience with sIBM patients, 1–2 years of benefit following tendon transfer surgery would be an achievable goal. In addition, patients who were considering surgery had significant UE disability as measured with the PROMIS UE ADL difficulty score, suggesting that there is objective opportunity for improvement in patient quality-of-life. Although over half of surveyed participants would consider surgery, only 8% were referred to a hand surgeon.

We also identified that a simple SANE score (rating from 0% to 100%) was strongly correlated with PROMIS metrics and could be used to triage patients for hand surgery referral by neurologists and rheumatologists who often take care of these patients.[18] Introducing this perspective to neurologists, rheumatologists, and patients may be helpful in improving education and increasing referrals to hand surgeons to discuss surgical options.

Table 3. Demographic and Clinical Characteristics and Outcomes of Participants and Preference for Surgery

	Would Consider Surgery	Would Not Consider Surgery	*P**
Total	102	90	
Age†			0.321
40–49	2 (2)	1 (1.1)	
50–59	15 (14.7)	7 (7.8)	
60–69	37 (36.3)	27 (30.0)	
70–79	39 (38.2)	41 (45.6)	
80–89	9 (8.8)	13 (14.4)	
90–99	0 (0)	1 (1.1)	
Female†	46 (45.0)	44 (48.9)	0.599
White†	5 (4.9)	5 (5.6)	0.839
Time since diagnosis (mo)†	78.2 (6.3)	95.4 (8.7)	0.112
UE ADL difficulty†	100 (98.0)	84 (93.3)	0.103
Seen by a hand surgeon?†	8 (7.8)	3 (3.3)	
SANE score			0.041
0–10	4	12	
11–20	16	12	
21–30	17	10	
31–40	13	16	
41–50	14	8	
51–60	15	5	
61–70	11	7	
71–80	5	12	
81–90	5	5	
91–100	1	3	
PROMIS self-efficacy (emotions)†	44.3 (7.3)	45.6 (9.0)	0.283
PROMIS self-efficacy (symptoms)†	41.0 (6.3)	41.4 (6.5)	0.682
PROMIS physical function†	29.1 (7.6)	29.5 (7.5)	0.705

*Chi-square *t* test.
†Values listed are the number of participants with percentage in parentheses.

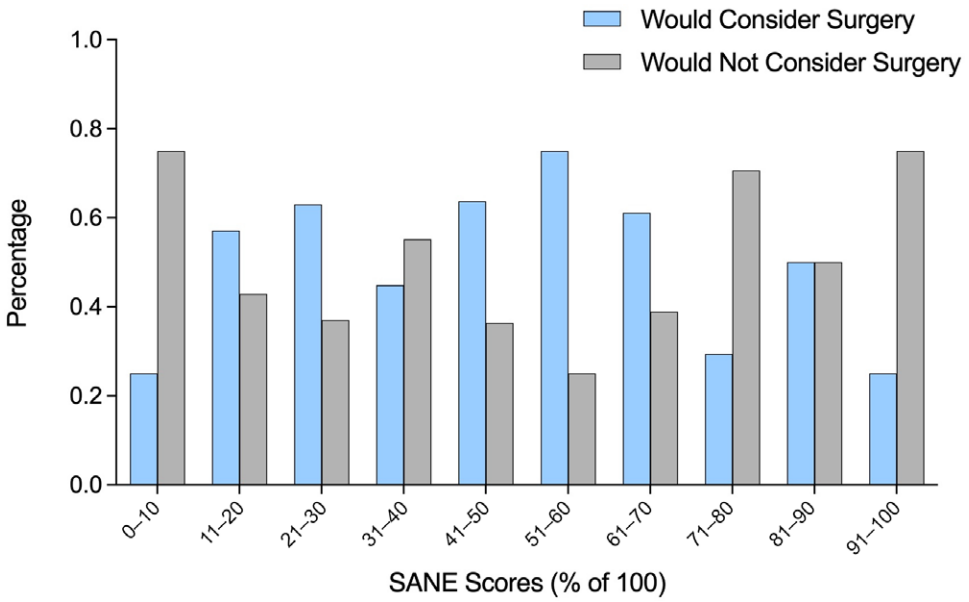

Fig. 3. SANE scores of participants considering tendon transfer surgery. Higher SANE scores indicate higher levels of physical function.

The results of this study provide support for conducting a surgical trial to assess if tendon transfers reliably reduce morbidity from hand dysfunction in sIBM. The impetus for conducting a trial to assess the efficacy of tendon transfers to improve hand function in sIBM is not only from neurologists and neuromuscular physicians caring for these patients, but also from patients and patient organizations, such as MSU. However, one challenge in

Table 4. Multivariable Models Demonstrating the Correlation between SANE Scores and PROMIS Scores

SANE Score	PROMIS Physical Function		PROMIS Self-efficacy (Symptoms)		PROMIS Self-efficacy (Emotions)	
	Effect Estimate*†	*P*	Effect Estimate*†	*P*	Effect Estimate*†	*P*
0–10	Reference		Reference		Reference	
11–20	1.02 (−2.476 to 4.517)	0.567	0.333 (−2.979 to 3.645)	0.843	−1.7 (−5.707 to 2.307)	0.405
21–30	2.35 (−1.147 to 5.847)	0.187	0.95 (−2.348 to 4.247)	0.572	0.74 (−3.9 to 4.045)	0.971
31–40	6.644 (3.331 to 9.956)	<0.001	2.649 (−0.48 to 5.779)	0.097	−0.101 (−3.863 to 3.661)	0.958
41–50	7.802 (4.447 to 11.157)	<0.001	3.135 (−0.024 to 6.296)	0.052	0.361 (−3.439 to 4.161)	0.852
51–60	9.243 (5.939 to 12.548)	<0.001	2.765 (−0.371 to 5.903)	0.084	0.368 (−3.422 to 4.158)	0.849
61–70	9.791 (6.463 to 13.119)	<0.001	3.589 (0.451 to 6.725)	0.025	1.754 (−2.026 to 5.535)	0.362
71–80	11.461 (8.141 to 14.781)	<0.001	3.358 (0.214 to 6.502)	0.036	−0.454 (−4.208 to 3.298)	0.812
81–90	15.778 (12.349 to 19.207)	<0.001	7.325 (4.054 to 10.597)	<0.001	2.126 (−1.768 to 6.021)	0.284
91–100	18.506 (14.905 to 22.107)	<0.001	9.29 (5.914 to 40.665)	<0.001	3.07 (−1.021 to 7.16)	<0.001

SANE Score		
	OR‡§	*P*
PROMIS physical function	0.161 (0.135 to 0.188)	<0.001
PROMIS self-efficacy (emotions)	−0.021 (−0.048 to 0.006)	0.125
PROMIS self-efficacy (symptoms)	0.061 (0.026 to 0.097)	0.001

*The values are given as the estimated effect of a SANE score with the 95% CI in parentheses. The model used is the Poisson model.
†Positive effects signify similar outcome scores between PROMIS and the SANE score.
‡Ratios greater than 1 are considered positively correlated with the SANE score and ratios less than 1 are considered negatively correlated. The model for the OR is the logit model.
§The values are given as the OR with the 95% CI in parentheses.

conducting a surgical trial will be in appropriately assessing "benefit." Various outcome measures have previously been used to track the progression of the disease and hand dysfunction, including muscle strength [manual muscle testing, quantitative muscle testing, handheld dynamometry, physical function (ie, quickDASH, IBM-FRS), and the Jebsen-Taylor Hand Function Test].[7,24] Although IBM-FRS is a reliable and validated measure, this scale has a much lower correlation than expected with finger flexor strength, and an even lower correlation with grip strength.[8,25] However, a modified IBM-FRS with the addition of a patient-reported outcome UE scale (IBM-PRO) showed a significantly greater correlation to both pinch and grip strengths,[25,26] and may be a better alternative to IBM-FRS alone. In this study, we were also able to establish a baseline for this population of patients using the PROMIS UE ADL difficulty, PROMIS self-efficacy, and PROMIS physical function scores. Future studies will likely need to use multiple PROMs to appropriately assess the effects of surgery and provide prognostic guidance to patients.

Limitations of our scoping literature review are inherent to the rarity of sIBM and the paucity of literature about tendon transfers for sIBM. Because this was also a survey-based study, we are unable to confirm diagnoses and patient data. Misdiagnosis is common, and selection bias is possible given the methodology. However, our study consists of a large sample size and large data distribution. Data of this survey are also limited by the cross-sectional design.

CONCLUSIONS

Given the limited amount of standardized data on tendon transfer surgery for patients with sIBM, yet the reported patient preference for a surgical option for management of disease progression, we propose conducting a surgical trial with systematic assessment of hand function that includes a hand examination, pinch and grip dynamometry, and PROMs.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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