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Gluteal Fibrosis and Its Surgical Treatment

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Background: The objective of this study was to analyze the literature regarding the diagnosis, pathogenesis, and prevalence of gluteal fibrosis (GF) and the outcomes of treatment.

Methods: We searched PubMed, Embase, and Cochrane literature databases, from database inception to December 15, 2016. We used the following search terms including variants: "contracture," "fibrosis," "injections," "injections, adverse reactions," "gluteal," and "hip." All titles and abstracts of potentially relevant studies were scanned to determine whether the subject matter was potentially related to GF, using predefined inclusion and exclusion criteria. If the abstract had subject matter involving GF, the paper was selected for review if full text was available. Only papers including ≥10 subjects who underwent surgical treatment were included in the systematic analysis. Data abstracted included the number of patients, patient age and sex, the type of surgical treatment, the method of outcome measurement, and outcomes and complications.

Results: The literature search yielded 2,512 titles. Of these, 82 had a focus on GF, with 50 papers meeting the inclusion criteria. Of the 50 papers reviewed, 18 addressed surgical outcomes. The surgical techniques in these papers included open, minimally invasive, and arthroscopic release and radiofrequency ablation. Of 3,733 operatively treated patients in 6 reports who were evaluated on the basis of the criteria of Liu et al., 83% were found to have excellent results. Few papers focused on the incidence, prevalence, and natural history of GF, precluding quantitative synthesis of the evidence in these domains.

Conclusions: This study provided a systematic review of surgical outcomes and a summary of what has been reported on the prevalence, diagnosis, prognosis, and pathogenesis of GF. Although GF has been reported throughout the world, it requires further study to determine the exact etiology, pathogenesis, and appropriate treatment. Surgical outcomes appear satisfactory.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

Acquired gluteal fibrosis (GF) is a clinical condition characterized by contracture of the gluteal muscles including, in varying degrees, the gluteus maximus, medius, and minimus[1-47]. While the diagnosis of acquired GF relies primarily on clinical findings, poliomyelitis and other diseases, cerebral palsy, and neuromuscular disorders must initially be ruled out on clinical examination, and other hip abnormalities must be ruled out through imaging, in order for the diagnosis to be made. Patients diagnosed with GF present clinically with obligate external rotation and abduction of the hip when the hip joint is flexed, and with an awkward gait, particularly when running[1-47]. GF can lead to substantial functional limitations for children, which may limit their ability to attend school and to perform activities of daily living.

This condition was described as early as the 1970s. While many authors initially described the condition as rare, it has been found to be more prevalent, with descriptions from throughout Asia, Europe, Africa, and the U.S.[1-50]. Scully et al. recently reported cases of children diagnosed with GF in the U.S., noting that those unfamiliar with the condition may have difficulty recognizing and treating it[32].

This systematic review assesses the range of approaches to surgical treatment of GF and provides an overview of what is currently known regarding the diagnosis, pathogenesis, and prevalence of GF.

Materials and Methods

We performed a comprehensive literature search utilizing PubMed, Embase, and Cochrane databases, from database inception to December 15, 2016. The search was performed utilizing appropriate MeSH (medical subject heading) terms, including "contracture," "fibrosis," "injections,"

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"injections, adverse effects," "gluteal," and "hip," with variant terms included.

Papers with potentially relevant titles were selected for further review of the abstracts. If the abstract had subject matter involving GF, the paper was selected for review if full text was available. If the full text was in a language other than English, an appropriate translator with a medical degree was utilized. All reports that met the inclusion criteria were abstracted using a standardized instrument for information on etiology, pathogenesis, natural history, prevalence, or treatment of GF.

After the initial assessment for inclusion, we performed a more detailed analysis of papers addressing surgical outcomes. In the surgical-outcome analysis, inclusion criteria included a minimum of 10 patients treated operatively for a diagnosis of GF and a report of surgical outcomes. Data extracted from these articles included the number of patients, patient sex and age, the type of surgical treatment, the outcome measurement method, and outcomes and complications. The studies used a variety of tools for the measurement of outcomes, prohibiting a formal meta-analysis.

Results

The literature search yielded 2,512 titles. From these titles, 82 abstracts with a focus on GF were reviewed, of which 50 had full text available. Four of these reports were written in Chinese and were translated into English for our thorough review. Of the 50 reports, 18 had ≥10 patients who underwent surgical treatment with outcomes measured (Fig. 1). Overall, the heterogeneity in the study methodology, including the duration of follow-up, method of assessment of outcomes, and presentation of outcomes, made comparisons among the studies difficult. All studies assessed were retrospective, with Level-IV evidence. No randomized controlled trials were available.

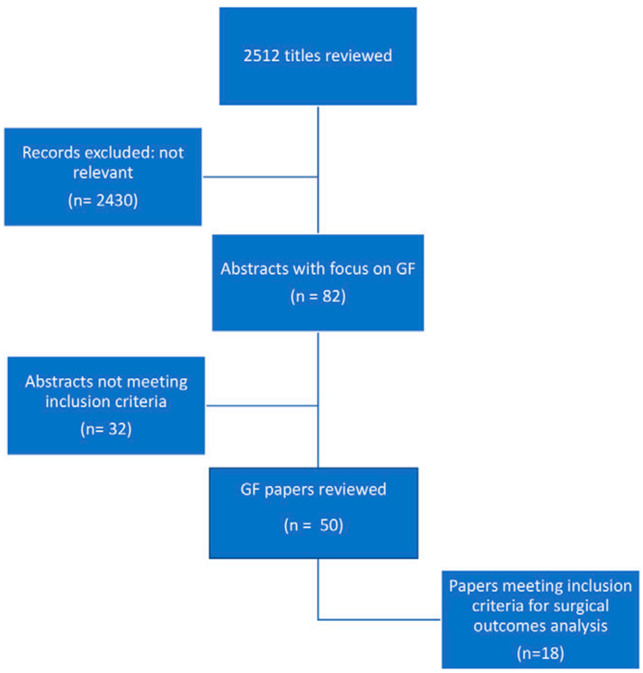

Fig. 1
Literature review search process.

Diagnosis

The reports reviewed did not propose a standard diagnosis or classification of GF, but each cited pathognomonic features of obligate abduction and external rotation contractures with attempt at hip flexion and an inability to squat with knees together, in the absence of other hip pathology or underlying neuromuscular diagnosis. These reports suggested that GF is characterized by hypertrophy of fibrotic tissue in the gluteal muscles that limits muscle excursion and, therefore, hip range of motion. The condition is mostly described as bilateral and frequently diagnosed in school-aged children[1,10,18,21,35,42,45]. Patients demonstrate a "frog-leg posture" because of the inability to adduct the hip in the sitting or squatting positions, with active and passive squat tests demonstrating the derangement in range of motion (Figs. 2 and 3)[5].

Of note, clinical diagnosis should not rest solely on the manifestation of frog-leg squatting, as other conditions can demonstrate this clinical finding[9]. Additional findings such as skin dimples over the buttocks, palpable contracture stripes, out-toeing gait, a positive Ober test, and leg-length discrepancies in conjunction with use of the squat test can assist with diagnosis, although the sensitivity and specificity of each of these tests for GF, have not, to our knowledge, been determined[5,21]. Authors often utilize combinations of these described clinical findings to grade the patients' GF as "mild," "moderate," or "severe," utilizing various classification schemes[22,47].

While the classification and diagnostic schemes rely on clinical findings, some authors have suggested a role for diagnostic imaging in confirming the diagnosis of GF. Cai et al. noted that 82.5% of patients with GF and 0% of controls demonstrated a hyperdense line on the ilium on radiographs corresponding to deformity in the posterior aspect of the ilium[4]. This line is hypothesized to come from the constant tension from the contracted gluteal muscles. Liu et al. demonstrated ultrasound findings of strips of hyperechoic muscle that had a sensitivity and specificity of 88.9% and 83.3%, respectively, while muscle thinning was found to have 92.6% sensitivity and 50% specificity[20]. In addition, Chen et al. demonstrated magnetic resonance imaging (MRI) features characteristic of GF including intramuscular atrophy and fibrotic cords that extend to the tendon, which were most commonly in the upper-third of the gluteus maximus[6].

Prevalence

While few studies remarked on prevalence, GF has been reported from around the world, including China, South Korea, India, Turkey, Iraq, England, Spain, Germany, Italy, Poland, Belgium, Uganda, and the U.S.[1-50]. Published estimates for GF generally range between 1% to 2.5% in affected populations, with prevalence seen as high as 13.9% of the pediatric population in some districts of Taiwan[5,10,23,44,46]. These reports suggest that high-prevalence areas are clustered in lower-resource areas.

Fig. 2
Active squat test for children with gluteal fibrosis.

Pathogenesis

The pathogenesis of GF has not been definitively determined. Several mechanisms have been hypothesized, including an idiopathic condition, a congenital collagen disorder, a genetic condition, an iatrogenic injury caused by sterile abscesses after intramuscular injections, compartment syndrome caused by large-volume intramuscular injection, or damage from neurotoxic intramuscular injections[1-50].

Despite the large variation in potential etiology and pathogenesis, nearly all studies cited injections as the cause of GF. However, in support of this contention, these studies referenced prior case reports that only hypothesized injections as the cause of GF. Moreover, in 45 of the 50 papers reviewed, the reported percentage of patients with a history of gluteal injections ranged from 51% to 100%. Only 2 of these studies examined whether injections had a causal role[10,17]. The 2 case-control studies assessed the odds of an injection history in GF cases compared with controls and found higher odds of gluteal injection among patients with GF (odds ratio [OR], 56) and a positive association between the frequency of injection and muscular fibrosis, with ORs ranging from 1.0 in the group with no injections to 69 in the group with the most injections (p value of <0.0001 for trend)[10,17]. In another case series of 310 patients, all were found to have a history of intramuscular injections, but again, no causal linkage was studied[44]. There were no data presented regarding other factors that may lead some patients who receive injections to develop GF, although hypotheses include fibrosis secondary to gluteal abscesses after unsterile injections and myoischemia post-injection due to injection volume[5,43].

Of the 50 papers reviewed, 6 noted patients who did not have a history of injections, and thus, each called for further study into the pathogenesis of GF[1,2,18,25,28,34]. Even authors who did find a high frequency of gluteal injections among their patients raised the question of whether an additional factor, such as the presence of an underlying collagen disorder, might be necessary for the development of GF. Supporting this concern was an apparent increased rate of keloids among the surgically treated patients, suggesting a predisposing collagen disorder[10,12,29,35,39,47]. One hypothesis is that affected patients have an underlying collagen disorder with a propensity for abnormal excessive fibrosis that is triggered by trauma. Supporting this theory are multiple studies with histological analysis demonstrating collagen fibrils with abnormal heterogeneity in diameter as well as excessive accumulation of collagen, particularly collagen types I and III, in the muscle[5,48-50]. Additionally, studies of muscle biopsies have demonstrated aberrant signaling pathways including the TGF-ß/Smad (transforming growth factor-beta/Smad) pathway as well as increased expression of TGF-ß1 and ß3, Smad4, and sphingosine-1-phosphate, factors involved in the stimulation of fibroblast and collagen proliferation, in the gluteal contraction bands[48-50]. Interestingly, similar findings have been noted with other fibrotic disorders, such as in keloid formation[48,49].

Outcomes of Nonoperative Therapy

The majority of reports regarding GF assessed the outcomes of patients who had surgical release of the gluteal contractures. Although nonoperative therapies were often mentioned for mild cases and as an adjunct to surgical treatment, only 1 study included the outcomes of nonoperative treatment of GF in its evaluation of outcomes[47]. In this study, nonoperative management, including 6 months of massage, physical therapy, shortwave diathermy, and the use of hot packs followed by active and passive exercises, was only effective for 52.9% of the patients with low-grade GF; even worse efficacy was demonstrated for higher grades of GF, with <10% effectively managed nonoperatively. While few studies have investigated the natural history of GF, there is a concern for the long-term effect of the altered hip range of motion on the surrounding joints. Increased stresses on the lumbar spine and knees may lead to the early onset of degeneration. Ye et al. reported that 90% of patients with GF had knee crepitus[42].

Surgical Treatment and Outcomes

The more frequently described and analyzed methods of management include a variety of surgical techniques involving intraoperative identification and release of fibrous bands

Fig. 3
Intraoperative photograph of passive gluteal fibrosis before release.

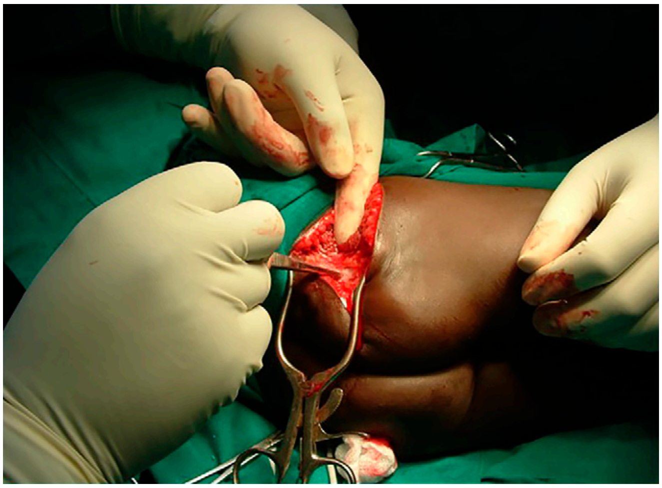

Fig. 4-A

Fig. 4-B

Figs. 4-A and 4-B Intraoperative photographs from 2 cases involving differing approaches to open surgical release for gluteal fibrosis (GF). **Fig. 4-A** Open release for the treatment of GF. **Fig. 4-B** Range of motion evaluated after surgical release.

to obtain normal hip range of motion[5,7,11,13-16,20-23,35,39,41,42,45-47]. In open surgical release of the fibrotic bands, the patient is placed in the lateral decubitus position. Incision placement varies, ranging from just posterior to the greater trochanter to directly over the buttock. Although the location and specifics of the incision vary (e.g., transverse versus longitudinal),

TABLE I Surgical Outcomes of Gluteal Fibrosis Release Utilizing the Scale of Liu et al.[21]*

Study	Operative Approach	No. of Participants	Age† *(yr)*	% Male	Follow-up† *(mo)*	Lost to Follow-up	Outcome	
							Excellent	Good
Chen and Yang (2015)[7]	Open release (sequential) + Z lengthening of gluteus maximus	20	13 (8-24)	60%	(12-60)	0	89%	11%
Fu et al. (2011)[14]	Open release (sequential)	50	8.9 (6-19)	44%	26 (12-24)	0	64%	30%
Fu et al. (2011)[14]	Arthroscopic (radiofrequency ablation)	52	9.2 (5-20)	42.3%	26 (12-24)	0	65%	23%
Liu et al. (2011)[20]	Open release (sequential) + Z lengthening of gluteus maximus	428	8 (5-15)	57.5%	15 (12-24)	0	93%	5%
Liu et al. (2008)[22]	Open release (sequential) + Z lengthening of gluteus maximus	286	8 (5-12)	53.1%	(3-24)	0	77%	18%
Xu et al. (2014)[41]	Open release (sequential) + Z lengthening of gluteus maximus	379	4 groups: 18.8, 20.5, 24.4, 28.4 (6-44)	41.4%	36 (12-60)	NR‡	60%	25%
Zhang et al. (2007)[45]	Open release (sequential) + Z lengthening of gluteus maximus	2,518	(5-30)	63.5%	24 (3-60)	0	90%	10%

*Liu et al.[21] scale of 4 levels of recovery. †The values are given as the mean, with the range in parentheses. ‡NR = not reported.

TABLE II Surgical Outcomes of Gluteal Fibrosis Release Utilizing Other Outcome Measurements

Study	Operative Approach	No. of Participants	Age* *(yr)*	% Male	Follow-up*† *(mo)*	Lost to Follow-up†	Measure of Outcome	Results
Chen et al. (1988)[5]	Open release	49	(5-58)	73.3%	NR	NR	Achievement of normal activity	100% achieved normal activity
Cui et al. (2008)[11]	Arthroscopic + radiofrequency ablation	86	16.3 (5-36)	41.9%	12.6 (6-18)	0	Scale including gait, range of motion, squat test	92% excellent, 6% good, and 2% fair; 100% satisfaction
Liu et al. (2000)[21]	Open release (sequential) + Z lengthening of gluteus maximus	1,280	13 (5-28)	56.3%	(3-24)	NR	Disappearance of preop. deficits and no recurrence for 3 mo	100% effective and 98.5% cured
Shen (1982)[35]	Open release	286	13 (5-23)	79.0%	(12-96)	30.1%	Squat test	100% demonstrated improvement
Ye et al. (2012)[42]	Open (minimally invasive) release	1,059	23 (8-43)	46.0%	30 (6-60)	0	Scale including gait, range of motion, squat test	100% excellent
Zhao et al. (2009)[47]	Open release (sequential)	158 (129 operative)	(4-17)	52.5%	58 (36-96)	8.1%	Improvement of hip range of motion and no additional operative intervention	Nonop. effective 39%; surgery effective 100% (83.7% excellent)
Ekure (2007)[13]	Open release	28	<12	44.4%	3	0	Hip range of motion, patient satisfaction	Hip flexion 80°-130°; 100% satisfaction
Gao (1988)[15]	Open release (gluteus maximus only)	27	8.5 (3-14)	55.6%	30 (3-108)	NR	Hip range of motion	93% with return of range of motion
Hang (1979)[16]	Open release (sequential)	28	(5-15)	89.3%	NR	0	Hip range of motion	Hip flexion 90°-120°; adduction full
Liu et al. (2009)[23]	Arthroscopic + radiofrequency ablation	108	23.7 (18-40)	52.8%	17.4 (7-42)	0	Hip range of motion	Mean hip flexion 110.2°; adduction 45.3°
Fernandez de Valderrama and Esteve de Miguel (1981)[39]	Open Z lengthening of gluteus maximus	71 (36 operative)	(1.5-15)	54.9%	(12-180)	0	Hip range of motion	89% with return of range of motion
Zhang et al. (2017)[46]	Arthroscopic + radiofrequency ablation	140	24.3 (19-43)	25.7%	38.9	2.9%	Harris hip score (HHS), patient satisfaction	HHS improved from a mean of 74.1 to 90.4 ($p < 0.05$); satisfaction 90.4%

*The values are given as the mean, with the range in parentheses. †NR = not reported.

after exposure of the musculature, each of the study descriptions follow a similar procedure (Figs. 4-A and 4-B)[5,7,13-16,20,21,35,39,41,42,45,47]. Full-thickness skin flaps are raised, exposing the gluteus maximus and notable fibrotic bands. The tensor fasciae latae, the gluteus maximus, the gluteus medius, and the gluteus minimus are sequentially released until examination indicates return of appropriate range of motion. Completion of release is determined by a negative Ober test and postoperative range of motion including hip adduction and internal and external rotation in flexion and extension. These surgical objectives can also be achieved with arthroscopy[11,14,23,46]. Postoperatively for both types of techniques (open and arthroscopic), the standard postoperative care involves immediate weight-bearing and full active range-of-motion exercises focusing on retaining maximum adduction with hip flexion.

Of the 18 papers with surgical outcomes, the patients seen with GF were largely adolescents or younger, although age ranged between 5 and 58 years; there was a slight predominance of male patients (Tables I and II). All of the studies reported effective results with the varying techniques, including arthroscopic, minimally invasive open release, open Z lengthening, and open complete transection of muscle. The range of follow-up was 3 to 180 months, with the majority of studies having a mean follow-up of >12 months.

In 6 of the 18 studies presenting surgical outcomes, the outcomes were assessed utilizing outcome criteria established by Liu et al. (Table III)[21]. In this rubric, outcomes are ranked as

TABLE III Liu et al. Criteria for Surgical Outcome Assessment[21]*

Excellent	Primary wound-healing, no positive GF signs, normal gait, no surgical complications, no LLD or pelvic tilt, full hip range of motion and strength, no recurrence by 6 months
Good	Primary wound-healing, no positive GF signs, normal gait, no surgical complications, LLD <1.5 cm, no pelvic tilt, nearly full functional hip (range of motion and strength), no recurrence by 6 months
Fair	Primary wound-healing, no positive GF signs, no surgical complications, LLD <2 cm, slight pelvic tilt, possible decreased hip range of motion or strength, no recurrence by 6 months
Poor	Primary wound-healing, presence of GF signs, surgical complications, LLD ≥2 cm, pelvic tilt, abnormal functional recovery of hip joint (range of motion and strength), possible recurrence

*LLD = leg-length discrepancy.

"excellent," "good," "fair," or "poor." These determinations are made on the basis of wound-healing, gait, pelvic tilt, hip strength and range of motion, and the presence or absence of positive GF signs, surgical complications, leg-length discrepancy, and recurrence. Of 3,733 patients in 6 reports who were treated surgically and evaluated using the criteria of Liu et al., 83% were found to have excellent results (Table I).

In the other 12 studies, outcomes were assessed by a variety of methods including clinical outcome scales, hip range of motion, the Harris hip score, and patient satisfaction (Table II). Generally, all of these studies demonstrated satisfactory outcomes with a return of range of motion for most patients and excellent patient satisfaction for all surgical techniques. In comparing open and arthroscopic release, the arthroscopic approach was found to involve a smaller incision, less tissue manipulation, less trauma, and less active bleeding/hematoma formation and was associated with fewer wound complications and faster recovery[11,14,23]. However, no significant differences in surgery duration, complications, clinical outcomes, or recurrence were found by Fu et al. in a study assessing the 2 approaches[14].

Complications with surgical management of GF ranged from 0% to 9.6% for arthroscopic techniques and 0% to 13.9% for open techniques (Table IV). Complications cited included

TABLE IV Complications with Surgical Management of Gluteal Fibrosis*

				Complication *(no.)*					
Study	Operative Approach	No. of Participants	No. (%) with Complications	Hematoma	Nerve Injury	Abductor Weakness	Infection	Recurrence	Other
Chen et al. (1988)[5]	Open	49	NR	NR	NR	NR	NR	NR	NR
Chen and Yang (2015)[7]	Open	20	0	0	0	0	0	0	0
Cui et al. (2008)[11]	Arthroscopic	86	0	0	0	0	0	0	0
Ekure (2007)[13]	Open	28	3 (10.7%)	0	1	0	2	0	0
Fu et al. (2011)[14]	Open	50	1 (2%)	0	0	0	0	1	0
Fu et al. (2011)[14]	Arthroscopic	52	5 (9.6%)	0	0	0	0	1	4
Gao (1988)[15]	Open	27	1 (3.7%)	1	0	0	0	0	0
Hang (1979)[16]	Open	28	2 (7.1%)	0	0	0	0	0	2
Liu et al. (2009)[23]	Arthroscopic	108	0	0	0	0	0	0	0
Liu et al. (2000)[21]	Open	1,280	54 (4.2%)	NR	NR	NR	NR	NR	NR
Liu et al. (2011)[20]	Open	428	16 (3.7%)	0	0	16	0	0	0
Liu et al. (2008)[22]	Open	286	16 (5.6%)*	NR	NR	NR	NR	NR	NR
Shen (1982)[35]	Open	286	0	0	0	0	0	0	0
Fernandez de Valderrama and Esteve de Miguel (1981)[39]	Open	71 (36 operative)	5 (13.9%)	1	0	0	0	1	3
Xu et al. (2014)[41]	Open	379	28 (7.4%)*	0	0	0	0	28	0
Ye et al. (2012)[42]	Open	1,059	3 (2.8%)	3	0	0	0	0	0
Zhang et al. (2017)[46]	Arthroscopic	140	12 (8.6%)	12	0	0	0	0	0
Zhang et al. (2007)[45]	Open	2,518	48 (1.9%)*	5	9	0	4	4	NR
Zhao et al. (2009)[47]	Open	158 (129 operative)	7 (5.4%)	4	0	0	2	0	1

*NR = not reported.

hematoma, sciatic nerve injury, abductor weakness, infection, and recurrence.

Discussion

This systematic review, including studies from across decades and a large number of case series, demonstrated that there remains a gap in our understanding of the prevalence, etiology, pathogenesis, and management of GF. While many studies cited injections as a cause of GF, rigorous research is necessary to verify injections as the cause and to determine if there is something particular about the injections or care processes that leads to GF. Additionally, it is important to assess for other factors involved in the development of GF, as many patients do not report an injection history. Is there a difference in collagen composition or a propensity for fibrosis in these patients as well that is triggered by trauma and leads to the development of GF? The answers to such questions will likely require genetic studies.

Furthermore, it is important to assess the natural history and outcomes for children who receive no treatment or undergo nonoperative management. Whether a surgical intervention is necessary or if nonoperative treatment would lead to similar long-term outcomes is not clear. We identified only 1 Level-IV study comparing nonoperative with operative management, and this study did not describe the method of treatment assignment or the rigor with which nonoperative management was attempted[47].

In contrast to this lack of literature regarding the nonoperative management of GF, there were a large number of studies assessing surgical management of GF, with 18 studies meeting the inclusion criteria for analysis[5,7,11,13-16,20-23,35,39,41,42,45-47]. There are several current methods of surgical intervention, and each method comes with a range of potential complications, from incomplete release to over-release with weakness or destabilization of the hip joint. Other short-term complications included some cases of hematoma and infection, while a few long-term complications, including nerve injury or recurrence, were reported as well. However, considering the aggregate of the surgical outcomes, the risk with surgical intervention was minimal. While the outcomes and complications were similar for both arthroscopic and open approaches, proponents of arthroscopic release believe that this surgical approach causes less trauma and allows for earlier rehabilitation, although Liu et al. noted an increased potential for incomplete release[23]. In fact, Fu et al. found that the arthroscopic approach was not reliable for severe cases[14]. Other proponents of an open approach suggest that open release is necessary to ensure avoidance of sciatic nerve damage as well as to obtain full therapeutic effect[5,7,20-22,35,41,45-47].

Limitations of this analysis of surgical outcomes for GF included the heterogeneity of the surgical approaches and methods of outcome assessment that were utilized. Despite the notable differences in methodology in each of the case series, the overall outcomes in these studies demonstrated the short-term success of surgical intervention. However, there is insufficient evidence to make clear recommendations regarding the prevention or treatment of GF. More rigorous studies are needed to confirm risk factors and pathogenesis, including case-control and prospective cohort studies, in addition to studies involving muscle biopsy and assessment of relevant collagen and genetic studies. Additionally, randomized controlled trials and/or prospective cohort studies involving both operative and nonoperative methods of treatment should be performed in order to provide clear recommendations regarding treatment.

In conclusion, intervention, whether nonoperative or operative, is necessary, as children with GF experience pain and activity limitation. There is concern that the increased stresses borne by adjacent structures may lead to further problems and early degeneration[42]. Over time, patients can develop leg-length discrepancy, pelvic obliquity, compensatory scoliosis, and, in severe cases, dislocation of the hip joint[14]. Not only does GF affect youths, but this disability has been found to limit those in the workforce, and GF has been used as a criterion for exemption from the military service in Taiwan[9]. Addressing GF early on is important to prevent a cascade of associated sequalae and disabilities. Future studies as outlined may lead to the prevention of GF, as well as improvement in the recognition and treatment of this condition. ■

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