Surgical Release of Gluteal Fibrosis in Children Results in Sustained Benefit at 5-Year Follow-up

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Background: Gluteal fibrosis (GF) is a fibrotic infiltration of the gluteal muscles resulting in functionally limiting contracture of the hips and is associated with injections of medications into the gluteal muscles. It has been reported in numerous countries throughout the world. This study assesses the 5-year postoperative range of motion (ROM) and functional outcomes for Ugandan children who underwent surgical release of GF.

Methods: A retrospective cohort study of children who underwent release of GF in 2013 at Kumi Hospital in Eastern Uganda. Functional outcomes, hip ROM, and scar satisfaction data were collected for all patients residing within 40 km of the hospital.

Results: One hundred eighteen children ages 4 to 16 at the time of surgery were treated with surgical release of GF in 2013 at Kumi Hospital. Of those 118, 89 were included in this study (79.5%). The remaining 29 were lost to follow-up or lived outside the study's radius. Detailed preoperative ROM and functional data were available for 53 of the 89 patients. In comparison with preoperative assessment, all patients postoperatively reported ability to run normally ($P < 0.001$), sit upright in a chair ($P < 0.001$), sit while eating ($P < 0.001$), and attend the entire day of school ($P < 0.001$). Passive hip flexion ($P < 0.001$) improved when compared with preoperative measurements. In all, 85.2% (n = 75) of patients reported satisfaction with scar appearance as "ok," "good," or "excellent" 29.2% (n = 26) of patients reported back or hip complaints.

Conclusions: Overall, the 5-year postoperative outcomes suggest that surgical release of GF improves ROM and functional quality of life with sustained effect.

Level of Evidence: Level IV—case series.

Key Words: gluteal fibrosis, injection injury, pediatric hip contracture, fibrosis surgery, treatment outcome, Uganda

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Gluteal fibrosis (GF) is a fibrotic infiltration of the gluteal muscles that results in limited muscle excursion and reduced hip range of motion (ROM). GF is commonly associated with multiple gluteal intramuscular injections, the exact etiology of which is not well understood but is currently being explored.[1,2] Possible hypotheses include myotoxic effects of injected medications, sterile abscess formation that leads to scarring, chronic inflammatory responses to multiple injections, and possible underlying fibrotic disorder in certain populations.[3–7]

First described in the 1970s, GF has since been identified in Africa, Asia, Europe, and the United States.[2,6,8–16] Population estimates of GF across regions typically vary from 1% to 2.5%, but were recently estimated to be as high as 28.3% in Eastern Ugandan children.[1,8,10,13] A possible explanation for this discrepancy is the unusually high number of gluteal intramuscular injections given in certain regions.

Most cases of GF are bilateral and diagnosed in school-age children.[1,10] Patients with GF frequently present with difficulty squatting, trouble sitting upright in chairs, pain with long-distance walking, and abnormal gait (Fig. 1). They have obligate external rotation and abduction when the hips are actively or passively flexed.[1]

Treatment of GF ranges from physical therapy to surgical release, depending on severity. Surgical release is the only treatment demonstrated to restore near normal ROM for moderate or severe cases of GF.[12] Hip ROM has been demonstrated to improve immediately following surgery and up to 2 years postoperation.[13,17,18] Of the 18 studies published on surgical treatment of GF with >10 patients, all are under 5-year follow-up and most average 2 years.[19] The only study in Africa, has just 3 months follow-up.[2] Because surgery itself can create scarring, it is imperative to do longer term follow-up to determine if

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FIGURE 1. Patient with moderate gluteal fibrosis in squatting position and forward bend. A, Preoperative—significant adduction and internal rotation limitation and limited forward flexion. B, Postoperative—just a few days after surgery, obvious improvement in squatting position and forward bend excursion. Photo credit: Paul Ekellot.

there is sustained benefit from surgical release or if scarring recurs and motion again becomes limited. This study aims to assess the 5-year postoperative ROM and functional outcomes for Ugandan children who underwent surgical release of GF.

METHODS

This study was conducted with approval from the institutional review boards of the Mildmay Uganda Research and Ethics Committee and University of California, San Francisco and approved by the Uganda National Council for Science and Technology. One hundred eighteen patients who underwent surgery for GF between July 1, 2013 and July 9, 2013 at a surgical camp at Kumi Hospital in Kumi, Uganda were eligible to participate in this study. Patients were excluded if they (1) resided more than 40 km from Kumi Hospital, (2) had a radiographically confirmed hip abnormality that affects movement, or (3) had a known neurological abnormality that would be a confounder to assess outcome. Children were indicated for surgery if they displayed significantly reduced hip ROM and limitations in walking, running, squatting, or sitting.

A research team comprised of a physical therapist, social worker, and medical student visited patients and their parents at home or school to discuss the study with them and perform the 5-year postoperative assessment, if they agreed to participate (Appendix 1, Supplemental Digital Content 1, http://links.lww.com/BPO/A329). Measurements of passive and active hip flexion in neutral rotation, and prone hip internal and external rotation at 90-degree knee flexion were collected by the same physical therapist who did the original measurements in 2013. These measurements were compared with reported norms.[20] Hip adduction was assessed with Zhao 90-90 test.[21] Functional outcomes and scar satisfaction data were collected by an established set of questions asked verbally to the patient. Scar satisfaction questions were adapted from the Patient and Observer Scar Assessment Scale and Stony Brook Scar Evaluation Scale and included questions about scar appearance, symptoms, and overall satisfaction.[22–25] All patients had an assessment of leg length

discrepancy by a standing iliac crest height evaluation using measured blocks. All conversations were conducted in English or Ateso, the most common native language in Kumi region, in accordance with the patient's preference. Following in-person postoperative evaluation, a retrospective chart review was done to obtain the participants' preoperative ROM and functional assessment answers from 2013.

Data were analyzed in Microsoft Excel 2016. Demographic data were analyzed using a 1 sample t test or χ^2 goodness of fit. ROM measurements were compared using a paired 2-tailed t test. Qualitative preoperative and postoperative variables were compared using a McNemar test.

Description of Surgical Procedure

All surgeries were performed under general anesthesia, by 1 of 2 surgeons. There was mild variability in incision placement, but the general technique was as follows. The patient was positioned supine and prepped and draped from umbilicus to feet. The patient was then rolled somewhat laterally to allow exposure of the operative side. A 3- to 5-cm incision was made along the lateral one third of a line visualized between the posterior iliac spine and the greater trochanter, the skin incision being approximately in line with the direction of the gluteus maximus muscle fibers. There were often scars and puckering of the skin where the contracture was most severe. After incision of the subcutaneous tissues the fibrotic gluteal muscle (which appeared as a thick white sheet) was exposed as far possible anteriorly, posteriorly, superiorly, and inferiorly by blunt dissection. A short incision, 2 to 3 cm long, was made in the direction of the gluteus maximus fibers using electrocautery, and carefully carried through the fibrous tissue. Careful monitoring of the limb was undertaken for transmitted sciatic nerve stimulation from the cautery. The fibrotic tissue was usually more superficial than deep but may have included the entire thickness of the gluteus maximus muscle. In the deeper layers, blunt dissection was utilized until perineural fat was encountered. An instrument was then passed under the remaining fibers of gluteus maximus, keeping in the layer of fat superficial to the nerve, and the muscle and fibrotic tissue lifted away from the nerve. The sciatic nerve was not directly exposed as it was sufficiently protected with this technique. The dense fibrous tissue was then incised with cautery or scissors transverse to the skin incision and the normal direction of the gluteus muscle fibers (transecting the dense fibrous tissue until normal muscle was encountered in the deeper layers). Remaining intramuscular fibrotic bands were palpated and released. The hip was then forcibly flexed and adducted to release any remaining contracted tissue. A tearing or popping sound was usually elicited as the remaining fibrous bands gave way. If adequate ROM was not accomplished, further palpation and release was performed.

The fibrosis seen in our patients was primarily in the gluteus maximus. The tensor fascia lata was frequently found to be involved and released as indicated. Gluteus medius and minimus involvement is rare in this population, but when present would need release with a more extensile incision. The skin incision was closed with interrupted nylon suture. Postoperative physical therapy was instituted immediately after surgery, focused on hip adduction and flexion, and patients instructed in a home ROM program.

TABLE 1. Demographic Data

	Surgical Cohort	5-y Follow-up Cohort	*P*
Patients	118	89	NA
Age at surgery (y)	10.30 (2.54)	9.91 (2.44)	0.135
Sex			0.170
M	64	50	
F	54	39	
Surgeon			0.014
Surgeon 1	64	47	
Surgeon 2	48	40	
Other/unknown	6	2	

Age at surgery is reported as mean (SD) and compared using 1 sample t test. Sex and surgeon were compared using χ^2 goodness of fit.
F indicates female; M, male; NA, not applicable.

RESULTS

One hundred eighteen children ages 4 to 16 at the time of surgery were treated with surgical release of GF during a surgical camp in July 2013 at Kumi Hospital. Of those 118, 89 met inclusion criteria for this 5-year outcome study. Eighty-eight patients underwent bilateral release and 1 patient underwent unilateral release. Average age at surgery was 10.3 (SD 2.54) years. There was no significant difference in age at surgery ($P = 0.135$) and sex ($P = 0.170$) between the original surgical cohort and those sampled (Table 1). All patients (n = 89) reported injections into the gluteal muscles before developing GF and 23.6% (n = 21) reported injections in the 5 years following surgery, although the number of injections could not be reliably quantified. There was a statistical difference between the operating surgeon in the original surgical cohort and those sampled ($P = 0.014$). However, there was no statistical difference in functional outcomes between surgeons ($P < 0.001$).

ROM Outcomes

Postoperative ROM was collected for all 89 patients (Table 2). Preoperative ROM data from retrospective chart review for passive and active hip flexion was available for a maximum of 53 of the 89 patients. For

TABLE 2. Postoperative Range of Motion

	Average (deg.)
Hip flexion (active)	M: 97.2 F: 92.1
Hip flexion (passive)	M: 105.8 F: 101.3
Hip internal rotation	M: 37.0 F: 39.6
Hip external rotation	M: 49.5 F: 48.4

Postoperative range of motion collected for all 89 study participants (50 males, 39 females). Overall, 177 measurements were taken, comprised of 88 patients with bilateral gluteal fibrosis and 1 patient with unilateral gluteal fibrosis.
F indicates female; M, male.

TABLE 3. Preoperative vs. 5-y Postoperative Hip Flexion Comparison Using Paired 2-Tailed *t* Test*

	n	Minimum	Maximum	Average	*P*
Passive hip flexion	86	Pre: 10 Post: 46	Pre: 120 Post: 133	Pre: 63.3 Post: 102.6	<0.0001
Active hip flexion	105	Pre: 35 Post: 36	Pre: 100 Post: 129	Pre: 74.6 Post: 94.8	<0.0001

Hip range of motion measurements taken in 2013 ("pre") and 2018 ("post") and compared using a paired 2-tailed *t* test.

*This table only includes data for patients with both preoperative and postoperative measurements.

that smaller cohort, when compared with preoperative measurements, average active hip flexion improved from 74.6 (13.3) to 94.8 (15.3) degrees ($P < 0.001$) and average passive hip flexion improved from 63.3 (25.1) to 102.6 (15.5) degrees ($P < 0.001$) (Table 3). Hip adduction was measured in 58 patients. None were found to have a gap between the knees postoperatively per Zhao 90-90 test, despite having an abduction contracture preoperatively.

Functional Outcomes

Hundred percent of the 89 patients reported no postoperative limitations in ability to run, sit upright in a chair, sit on a bench, use the pit latrine/toilet, eat while sitting, attend a full day of school, and perform physical education. 98.8% (n = 88) of patients self-reported the ability to walk normally. The examiner determined that 94.4% (n = 84) had normal gait, 4.49% (n = 4) walked with Trendelenburg gait, and the remaining 1.12% (n = 1) walked with an abnormal non-Trendelenburg gait. Of those with abnormal gait, 2 were confirmed to have concomitant postinjection paralysis, an injection injury of the sciatic nerve causing paralytic foot deformity.

Preoperative functional data were available for 53 of the 89 patients. Preoperatively, normal ability to walk (96.2%), run (0.00%), sit upright in a chair (49.1%), sit on a bench (47.2%), use the pit latrine/toilet (8.00%), eat while sitting (11.5%), attend a full day of school (54.9%), and perform physical education (32.0%) was reported. Compared with 100% ability to perform all those activities postoperatively, each variable showed a statistically significant difference ($P < 0.001$) except for ability to walk normally ($P = 0.160$) (Table 4).

Scar Satisfaction

Average length of surgical scars was 54.9 mm. 85.2% (n = 75) of patients described satisfaction with scar appearance as "ok," "good," or "excellent." 95.5% (n = 84) reported that scar symptoms were "not at all troublesome," although 44.3% (n = 39) reported current or previous itchiness, 2.27% (n = 2) reported pain at incision site with certain positioning or pressure, and 5.88% (n = 5) reported stiffness, tightness, or numbness. 38.2% (n = 34) of scars were keloid. 98.9% (n = 88) of scars matched surrounding skin.

Other Outcomes

87.6% of patients had no leg length discrepancy (n = 78), 11.2% had a difference <2 cm (n = 10), and 1.12% had a difference >2 cm (n = 1). 29.2% (n = 26) of patients reported back or hip complaints. Lower back pain, especially with physical labor or prolonged bending, was the most common complaint (n = 18, 20.2%).

DISCUSSION

Although it has been reported worldwide, GF disproportionately affects patients in resource-limited countries. Because the apparent risk factors are similar to those of postinjection paralysis, the 2 conditions are often grouped as injection-induced injuries. From 2013 to 2015 in Kumi, Uganda, together they accounted for 30% of orthopaedic clinic complaints and 40% of MSK outreach visits among children. Of the 3339 patients seen in the outreach clinic for musculoskeletal complaints in this time frame, 1114 were diagnosed with GF.[1] This suggests that the prevalence is high enough to warrant more rigorously assessed treatment options and prevention strategies.[1] Many affected Ugandan patients are children and are functionally limited in their ability to walk, attend school, and participate in other culturally normative activities. Examples of specific limitations include inability to sit comfortably in chairs at school and significant hip pain during the daily walk to and from school, which is often longer than 30 minutes. Early and successful intervention has the potential to significantly impact their quality of life.

Overall, patients showed improved ROM with sustained effect in all categories for which we had both

TABLE 4. Functional Variable Comparison Using McNemar Test*

	n	Preoperative Normal	Preoperative Abnormal	Postoperative Normal	Postoperative Abnormal	*P*
Walks	52	50	2	52	0	0.16
Runs	52	0	52	52	0	<0.0001
Sits upright in chair	53	26	27	53	0	<0.0001
Sits on bench	53	25	28	53	0	<0.0001
Uses toilet	50	4	46	50	0	<0.0001
Eats while sitting	52	6	46	52	0	<0.0001
Attends school	51	28	23	51	0	<0.0001
Does PE	50	16	34	50	0	<0.0001

Preoperative (2013) and postoperative (2018) comparisons of ability to perform functional quality of life activities. Compared using a McNemar test, which allows for comparison of dichotomous traits in matched subjects.

*This table only includes data for patients with both preoperative and postoperative measurements.

preoperative and postoperative data. Passive and active hip flexion improved dramatically, but are still lower than what would be considered normal range.[20] This indicates that the surgery improves, but does not normalize ROM. However, all patients reported normal ability to walk, run, sit in a chair or bench, eat while sitting, use a pit-latrine style toilet, attend a full day of school, and perform physical education postoperatively. This improvement in functional outcomes is clinically significant. These improved outcomes are consistent with short-term follow-up results in Uganda and other regions.[2,6,7,19,21,26] This study presents a cohort with the longest average follow-up time (60 mo) in the literature and the only study in Africa that assesses outcomes > 3 months following surgery.[2,19]

Scar satisfaction has not been previously assessed in patients who underwent surgical release of GF. Although there was some concern initially that the scars would be considered unattractive, 85.2% of patients described their scars as "ok" or better, indicating that scar appearance is relatively well tolerated. However, there were 46 reports of current itching, pain, stiffness, tightness, or numbness at the scar. This suggests that the scar symptoms may be more troublesome than appearance. Importantly, all reported symptoms were nuisance symptoms only and did not affect activities of daily living or quality of life.

29.2% of patients acknowledged back or hip complaints when prompted. Because these complaints were often associated with prolonged bending, labor, and distance walking, it is difficult to assess whether they are related to the original GF, the surgical release, or other activities of daily life. Back and hip complaints in this age group and activity level may be found in patients with no history of GF at similar or higher rates.[27] This may be related to the relatively high amount of manual labor performed in this agriculturally based population. We do not have an unaffected group to compare to, which is a limitation of this study.

Given the retrospective nature of this study, there are inherent limitations. Although 89 patients of the original 118 patient cohort were able to be assessed in 2018, 17 patients were outside the study radius and 12 were lost to follow-up. However, a demographic analysis revealed no statistical difference between those included and excluded from the study. In addition, patients reported variable adherence to the postoperative physical therapy regimen, which may have affected their outcomes. A prospective study that tracks physical therapy adherence would be beneficial in determining its effect on ROM. In addition, we did not have preoperative hip x-rays on the patients to assess whether there was any concomitant hip joint pathology that could affect motion or pain symptoms. Although preoperative ROM measurements were conducted by the same physical therapist as the postoperative measurements, there may have been some inconsistency in technique, particularly with control of abduction with flexion measurements. Furthermore, the questionnaire used to evaluate scar satisfaction was based on the POSAS and Stony Brook scar assessment tools but adapted by the authors for use in children and has not been validated in Uganda. Finally, the normative hip ROM data are from an African American pediatric population in Philadelphia, which may not be representative of our population's normal motion.

In conclusion, this study demonstrated a statistically and clinically significant improvement in hip ROM and functional outcomes, with acceptable scar satisfaction, 5 years after surgical release of GF in this population of children from Eastern Uganda.

REFERENCES

1. Alves K, Penny N, Ekure J, et al. Burden of gluteal fibrosis and post-injection paralysis in the children of Kumi District in Uganda. *BMC Musculoskelet Disord*. 2018;19:343.
2. Ekure J. Gluteal fibrosis. A report of 28 cases from Kumi Hospital, Uganda. *East Central African J Surg*. 2007;12:144–147.
3. Scully WF, White KK, Song KM, et al. Injection-induced gluteus muscle contractures: diagnosis with the "reverse Ober test" and surgical management. *J Pediatr Orthop*. 2015;35:192–198.
4. Chung DC, Ko YC, Pai HH. A study on the prevalence and risk factors of muscular fibrotic contracture in Jia-Dong Township, Pingtung County, Taiwan. *Gaoxiong Yi Xue Ke Xue Za Zhi*. 1989;5:91–95.
5. Ko YC, Chung DC, Pai HH. Intramuscular-injection-associated gluteal fibrotic contracture and hepatitis B virus infection among school children. *Gaoxiong Yi Xue Ke Xue Za Zhi*. 1991;7:358–362.
6. Hang YS. Contracture of the hip secondary to fibrosis of the gluteus maximus muscle. *J Bone Joint Surg Am*. 1979;61:52–55.
7. Chen SS, Chien CH, Yu HS. Syndrome of deltoid and/or gluteal fibrotic contracture: an injection myopathy. *Acta Neurol Scand*. 1988;78:167–176.
8. Sun X. An investigation on injectional gluteal muscle contracture in childhood in Mianyang City. *Zhonghua Liu Xing Bing Xue Za Zhi*. 1990;11:291–294.
9. Fernandez de Valderrama JA, Esteve de Miguel R. Fibrosis of the gluteus maximus: a cause of limited flexion and adduction of the hip in children. *Clin Orthop Relat Res*. 1981;156:67–78.
10. Kotha VK, Reddy R, Reddy MV, et al. Congenital gluteus maximus contracture syndrome—a case report with review of imaging findings. *J Radiol Case Rep*. 2014;8:32–37.
11. Al Bayati MA, Kraidy BK. Gluteal muscle fibrosis with abduction contracture of the hip. *Int Orthop*. 2016;40:447–451.
12. Fu D, Yang S, Xiao B, et al. Comparison of endoscopic surgery and open surgery for gluteal muscle contracture. *J Pediatr Orthop*. 2011;31:e38–e43.
13. Liu YJ, Wang Y, Xue J, et al. Arthroscopic gluteal muscle contracture release with radiofrequency energy. *Clin Orthop Relat Res*. 2009;467:799–804.
14. Wang CX, Gong YS, Li SH, et al. Gluteal muscle contracture release for the treatment of gluteal muscle contracture induced knee osteoarthritis: a report of 52 cases. *Zhongguo Gu Shang*. 2011;24:594–596.
15. Zhang X, Ma Y, You T, et al. Roles of TGF-beta/Smad signaling pathway in pathogenesis and development of gluteal muscle contracture. *Connect Tissue Res*. 2015;56:9–17.
16. Liu G, Yang S, Du J, et al. Treatment of severe gluteal muscle contracture in children. *J Huazhong Univ Sci Technol Med Sci*. 2008;28:171–173.
17. Liu GH, Cao FQ, Yang SH, et al. Factors influencing the treatment of severe gluteal muscle contracture in children. *J Pediatr Orthop B*. 2011;20:67–69.
18. Rai S, Jin S, Meng C, et al. Arthroscopic release using F and C method versus conventional open release method in the treatment of gluteal muscle contracture: a comparative study. *BMC Musculoskelet Disord*. 2017;18:113.
19. Alves K, Katz JN, Sabatini CS. Gluteal fibrosis and its surgical treatment. *J Bone Joint Surg Am*. 2019;101:361–368.
20. Sankar WN, Laird CT, Baldwin KD. Hip range of motion in children: what is the norm? *J Pediatr Orthop*. 2012;32:399–405.

21. Zhao CG, He XJ, Lu B, et al. Classification of gluteal muscle contracture in children and outcome of different treatments. *BMC Musculoskelet Disord*. 2009;10:34.
22. Roques C, Teot L. A critical analysis of measurements used to assess and manage scars. *Int J Low Extrem Wounds*. 2007;6:249–253.
23. Fearmonti R, Bond J, Erdmann D, et al. A review of scar scales and scar measuring devices. *Eplasty*. 2010;10:e43.
24. Draaijers LJ, Tempelman FR, Botman YA, et al. The patient and observer scar assessment scale: a reliable and feasible tool for scar evaluation. *Plast Reconstr Surg*. 2004;113:1960–1965; discussion 6-7.
25. Singer AJ, Arora B, Dagum A, et al. Development and validation of a novel scar evaluation scale. *Plast Reconstr Surg*. 2007;120:1892–1897.
26. Ye B, Zhou P, Xia Y, et al. New minimally invasive option for the treatment of gluteal muscle contracture. *Orthopedics*. 2012;35:e1692–e1698.
27. Mwaka ES, Munabi IG, Buwembo W, et al. Musculoskeletal pain and school bag use: a cross-sectional study among Ugandan pupils. *BMC Res Notes*. 2014;7:222.