

Post-traumatic Arthrofibrosis of the Knee Joint in a Patient with Familial Cutaneous Collagenoma and Recent ACL reconstruction with Hamstring Allograft

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Submitted: 15 Dec 2018; Accepted: 21 Dec 2018; Published: 31 Jan 2019

Abstract

Familial cutaneous collagenoma is a rare, autosomal dominant hereditary disease. These collagenous bundles are typically seen as hypopigmented, superficial skin lesions along the trunk and the upper limbs. We report a case in which a 31-year-old patient presented with a disproportionate growth of scar tissue in the knee joint after having almost non-existent flexion and continued pain for 3 months after ACL reconstruction despite aggressive rehabilitation with formal therapy. In this case, there was no organic cause of tissue overgrowth or reason to expect such a drastic change in the joint space so soon after surgery. Histopathology of tissue from the left knee showed dense fibroconnective tissue with myxoid change and granulation tissue. The overgrowth was diagnosed as a form of collagen disorder with increased amounts of irregularly arranged dense fascicular bundles of collagen which are similarly seen in familial cutaneous collagenoma. To the best of our knowledge, our case is the first reported case of post-traumatic arthrofibrosis in a patient with familial cutaneous collagenoma (FCC) in American literature.

Keywords: Familial Cutaneous Collagenoma, Arthrofibrosis, Left Knee

Introduction

Familial cutaneous collagenoma is a rare genetic disease inherited in an autosomal dominant fashion [1]. This term was first coined by Henderson in 1968 [2]. The characteristic findings and genetic pattern were also reported in 1979 in familial studies [3]. Typical characteristics of this condition include dense deposits of fibroconnective tissue giving the appearance of cutaneous papules and nodules throughout the body [1]. In addition to cutaneous findings, FCC has been shown to include fibrous involvement of the heart and blood vessel walls [3]. Biopsy of such lesions demonstrate disorder and increasing quantity of elastin, collagen, and proteoglycan. We present a case of post-traumatic arthrofibrosis of the left knee joint in a patient with familial cutaneous collagenoma and discuss it in the context of previously reported literature.

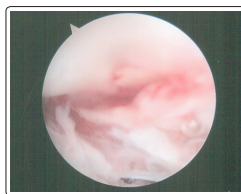
Case Presentation

A 31-year-old Korean female presented to clinic complaining of left knee pain. The patient reported injuring her knee two weeks prior to the visit while playing softball and running before she felt three pops followed by painful swelling. At this time, she noted pain along the anterior and medial aspect of her knee with associated swelling. Symptoms became worse when the patient attempted to bear weight as well as flex or extend her knee. The patient also reported catching, as well as instability. Imaging showed a large left knee joint effusion and an anterior cruciate ligament (ACL) tear was suspected. The patient followed up two weeks later after MRI imaging which showed a full

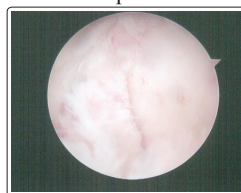
thickness tear of the ACL with bony edema pattern of the medial femoral condyle and medial tibial plateau consistent with pivot shift injury. Patient was positive for Lachman's and anterior drawer exams on physical exam, but there were limitations with range of motion due to pain and stiffness. Surgery was delayed for 3 weeks to restore preoperative full range of motion of the knee. Before scheduled surgery, the patient reinjured her knee. She felt her knee buckle and she noted increased pain, swelling, and stiffness. At this time, the medial collateral ligament (MCL) suffered a Grade 1 sprain. The procedure was performed as planned with preoperative range of motion being 0 to 100 degrees which was consistent with the patient's right knee. A standard anterolateral portal was made and diagnostic arthroscopy was performed starting with the suprapatellar pouch, lateral facet of patella, lateral gutter, patellofemoral articulation/tracking and the trochlear groove, medial gutter, medial joint followed by lateral joint. A limited synovectomy was performed in the medial and lateral compartments. The femoral tunnel guide and tibial guide pin were placed and secured and reaming of the tibial tunnel was achieved with full protection of all intra-articular structure. Intra-articular arthroscopic views showed anatomic graft placement without concerns for impingement, and with appropriate rigidity. Patient was doing well at her 2-week follow-up but noted some difficulty with range of motion and stiffness of her knee. At her 6 week follow up, she continued to use her brace with a single crutch to weight bear and had a flexion of 45 degrees. At this time, the patient mentioned that she had been diagnosed with a "collagen disease" as a child which affects scarring and healing. The only surgery that the patient had in the past was a previous shoulder operation which required a follow-up surgery secondary to scar tissue buildup according to the patient [4]. The patient is adopted and of

Korean descent and was unable to recall further details aside from showing us these hypertrophic scars on her shoulder and thighs from previous surgeries. At her 3-month follow-up the patient feels that she had regressed and heard “crackling noises” in her left knee after a particularly aggressive physical therapy session. She felt that she had almost regained her full range of motion until that therapy session where her physical exam at 3 months showed a 10 degree flexion contracture and flexion of only 20 degrees. At that time, an MRI was done which demonstrated a sizable cyclops lesion with diffuse scar tissue also noted in the suprapatellar notch. After full disclosure of the risks and benefits of the procedure, manipulation under anesthesia and knee arthroscopy were planned [Fig.1]. Preoperative range of motion was 10-20. A manipulation of the knee under anesthesia was first done to provide for easier scope positioning. Knee flexion was improved to 85 degrees. Arthroscopy necessitated extensive lysis of adhesions surrounding the ACL graft and medial and lateral gutters and removal of the cyclops lesion.

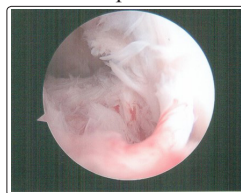
Further debridement from the lateral portal was necessary due to tissue overgrowth. There was exuberant sclerotic scar tissue in the suprapatellar notch which was also removed that allowed for significantly better patellar mobility.



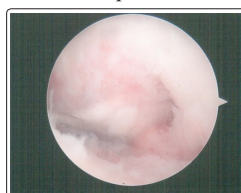
Scope 1



Scope 2



Scope 3



Scope 4

The amount of scar tissue noted 3 months status post ACL reconstruction was inconsistent with any routine scar tissue that would be present. It was even noted by the musculoskeletal radiologist who read the preoperative MRI that the amount of scar tissue noted would be consistent with overgrowth of at least a year or more. Tissue was sent to biopsy, and the observed arthrofibrosis was suspected to be a manifestation of a collagen disorder given the densely collagenized tissue with fibrocartilaginous metaplasia

consistent also seen in FCC.

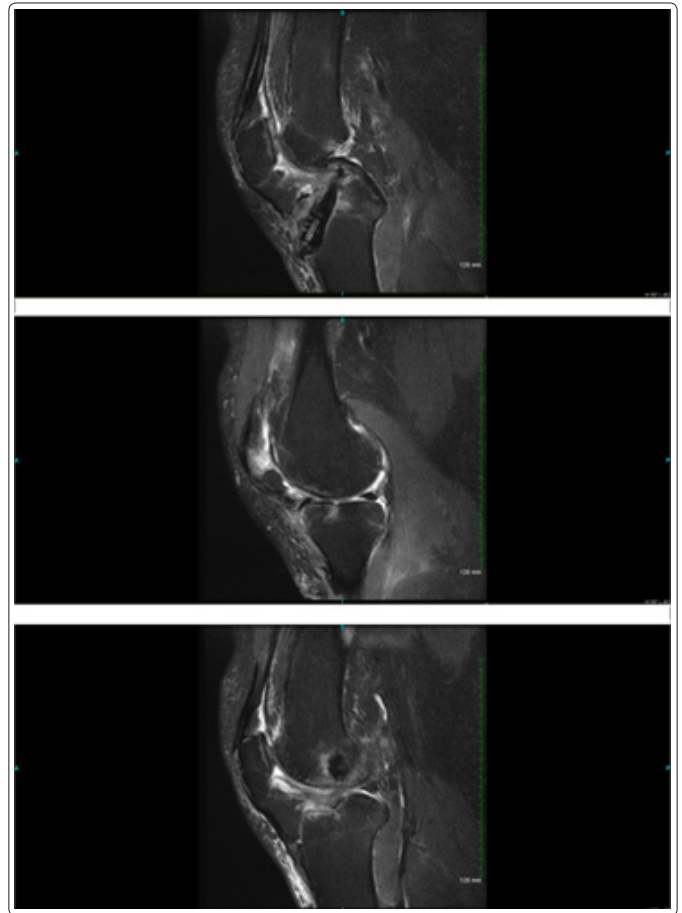


Figure 1: Left Knee MRI

Discussion

FCC is rare in nature and is reported in a very small subset of the population [Table 1]. Of these reported cases, none had involved manifestation in the knee joint. Aside from an autosomal dominant inheritance pattern, which is shared in our patient, the etiology is largely unknown. Patient evaluation should include a thorough physical examination to rule out other disorders with collagenoma such as tuberous sclerosis and Buschke-Ollendorf syndrome. In addition, our patient did not have ash-leaf macules nor osteopoikilosis during work-up and examination, lending to our confirmed diagnosis of FCC. Given the narrow scope of cases, conclusions of further complication are not unreasonable to draw [5]. With respect to the knee joint, the rich vascular supply including the inferior medial and lateral genicular arteries supplying the ACL via the fat pad is not an unreasonable target of blood vessel fibrosis seen in FCC, particularly after trauma [3]. A skin biopsy is necessary to confirm and distinguish between disorders of elastin, collagen, and proteoglycan [4]. In fibrosis, the collagen crosslinks and forms a pronounced alignment in a single direction. Our biopsy report included densely collagenized tissue as well as myxoid changes which fit the description of scar tissue in patients with FCC. Surgeons planning elective surgery on patients with FCC should consider the exuberant scarring and adhesions that may occur as a result of this autosomal dominant condition and they should counsel these patients accordingly.

Table 1: Reported Families of Familial Cutaneous Collagenoma (Adapted From Gurel et. al [7])

Author (Year)	Number of Cases	Onset	Location
Henderson <i>et al.</i> (1968)[2]	3	Postpubertal	Upper back
Uitto <i>et al.</i> (1979)[3]	6	15-19 years	Back and Chest
Phillips <i>et al.</i> (1999)[4]	2	Postpubertal	Upper back, abdomen
Boente <i>et al.</i> (2004)[5]	2	Prepubertal	Lower back, abdomen and upper arms
Amato <i>et al.</i> (2005)[6]	4	Postpubertal	Upper limbs, and abdomen
Gurel <i>et al.</i> (2007)[7]	2	Prepubertal	Lower back, abdomen

Conclusion

We report a rare instance of a patient with familial cutaneous collagenoma with post-traumatic arthrofibrosis of the left knee joint. The pathological features of biopsy and overgrowth of tissue in the vascular joint space of the knee and the patient’s history of FCC are consistent with a form of collagen disorder. In review, more experience with the subsequent expressions of this rare genetic disorder and any surgical outcomes is required as our case is novel

and thought-provoking in nature. To the best of our knowledge, this case is the first reported case of post-traumatic arthrofibrosis in a patient with FCC in American literature.

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