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Case report

Nutrient vessel dysfunction can contribute to mucoid degeneration of the posterior cruciate ligament coexisting with lipoma arborescens: A case study

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ABSTRACT

Introduction: With the rapid improvement of magnetic resonance imaging (MRI), mucoid degeneration (MD) of the anterior cruciate ligament (MD-ACL) has become an established disease entity and mechanical factors, such as increased posterior tibial tilt and intercondylar notch impingement, have been proposed. However, symptomatic MD of the posterior cruciate ligament (MD-PCL) remains an orphan disease without any established etiology.

Presentation of case: A man in his 60s exhibited restricted range of motion with knee pain. MRI revealed PCL enlargement with high-signal intensity and tram-track appearance on T2-weighted sagittal images and lipoma arborescens (LA) in the suprapatellar pouch. On gadolinium-enhanced MRI, the distal PCL was not contrasted. Arthroscopy revealed an almost normal expanded appearance with partial loss of the envelope synovium. Debulking operation was performed. Pathological findings revealed intravascular thrombus formation in early lesions of MD, and intraligamentous vascular degeneration and severity of MD were proportional.

Discussion: ACL is susceptible to mechanical external forces from surrounding tissues because of its anatomical features that induce protease expression, resulting in MD-ACL with denatured large aggregating proteoglycans deposition. Conversely, occlusion of nutrient vessels within the ligament was observed in this case of MD-PCL. Coexisting LA likely provoked an inflammatory response with hypercoagulability, resulting in thromboembolism of the envelope synovial nutrient vessel.

Conclusion: MD-CL is a disease entity comprising multiple pathologies. Although symptomatic MD-ACL is mainly caused by mechanical factors with a relatively high morbidity rate, nutrient vessel dysfunction can contribute to symptomatic MD-PCL with coexisting LA in middle-aged adults with an extremely low morbidity rate.

1. Introduction

Mucoid degeneration of the cruciate ligament (MD-CL) is a condition in which excessive accumulation of mucoid substrates, represented by proteoglycans (PGs) [1]. PGs within cruciate ligament (CL) can be broadly divided into small leucine-rich proteoglycans (SLRPs) and large aggregating proteoglycans (LAPs) [2]. SLRPs are bound to collagen and maintain collagen fibers in regular alignment [2] with a half-life of 20 days [3]. In contrast, LAPs play a role in dispersing stress generated between collagen fibers and are unstable PGs with a half-life of only 2 days [3]. LAPs contain many negatively charged glycosaminoglycan chains attached to their core proteins. These LAPs attract and bind water and likely contribute to tissue expansion, which is a common feature of MD-CL [1,4–6]. Thus, MD-CL can be considered a pathological condition of denatured LAP deposition.

Denatured LAPs are constantly produced within CL, but under normal conditions, they do not accumulate within CL because they are constantly expelled from CL via lymphatic drainage activity [7,8]. The inability of denatured LAPs to drain out of the CL due to lymphatic dysfunction is thought to be one cause of MD-CL pathogenesis.

With the expanded use of magnetic resonance imaging (MRI) and improved recognition of MD-CL, the prevalence of MD of the posterior cruciate ligament (MD-PCL) based on MRI has increased from 0.1 % to 4.9 % [1,9]. For symptomatic MD of the anterior CL (MD-ACL), mechanical factors such as increased posterior tibial tilt and intercondylar notch impingement have been implicated [10,11]. However, symptomatic MD-PCL is rarely reported compared with MD-ACL in middle-

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Abbreviations

ACL	Anterior cruciate ligament
CL	Cruciate ligament
LA	Lipoma arborescens
LAP	Large aggregating proteoglycan
LC	Ligament cell
MD	Mucoid degeneration
MRI	Magnetic resonance imaging
PCL	Posterior cruciate ligament
ROM	Range of motion
SLRP	Small leucine-rich proteoglycan

aged adults, and its etiology is still much in dispute [4,6,12].

The prevalence of MD-CL is not significantly different in autopsy cases, with MD-ACL found in 62 % of cases and MD-PCL in 71 % [13,14]. Thus, the etiology of MD-PCL and MD-ACL is considered to be the same in the elderly, i.e., arising from age-related degeneration with progressive pathological conditions [4].

Regarding the affected side of MD-CL, some review articles on MD-PCL have reported MD-ACL complications, and the frequency of combined cases of MD-PCL with MD-ACL ranges from 45% [4] to 85 % [6]. However, review articles on MD-ACL have reported MD-PCL complications from 0% [1,5,12] to 12 % [15]. This indicates that the etiology that induces MD-PCL can simultaneously induce MD-ACL, but the etiology that induces MD-ACL does not always induce MD-PCL in middle-aged adults.

Furthermore, the present case was complicated with LA. This is significant as no MD-PCL case with LA has been reported. Therefore, considering whether the combination of MD-PCL and LA is merely coincidental or is caused by some type of interaction is necessary. Moreover, the effect of poliomyelitis on the present case needs to be discussed. This work has been reported in accordance with the SCARE criteria [16].

2. Presentation of case

The case is of a male in his 60s who suffered poliomyelitis in his left leg at the age of 2 years. In his 30s, he underwent a 35-mm lower leg lengthening procedure using callotasis. Notably, 25 years after the procedure, a painless left knee flexion restriction of 90° suddenly appeared without any trauma. Two weeks later, he visited the hospital due to a sudden worsening of symptoms, such as pain during weight bearing, knee extension restriction, joint swelling and severe night pain insomnia. The range of motion (ROM) of his left knee joint (extension/flexion) was −15°/70° at the consultation without any joint instability, other than mild muscle atrophy due to poliomyelitis and had no history of medication.

X-ray examination revealed no evidence of osteoarthritis and notch narrowing. MRI showed high-signal intensity with a low-signal structure along the long axis of the expanded PCL surface on T2-weighted sagittal images with tram-track appearance. Gadolinium-enhanced MRI (CE-MRI) was performed following an intravenous injection of ProHance® (Gadoteridol) 13 ml (0.2 ml/kg). A contrast effect was observed at the PCL proximal intercondylar femoral attachment side but not at the PCL tibial attachment side (Fig. 1). Based on the MRI images, the patient was diagnosed with MD-PCL, and arthroscopic surgery was performed 6 weeks after the onset of the ROM restriction.

Under general anesthesia, knee joint ROM is normal without any joint instability. The arthroscopic findings revealed that PCL was expanded, and yellow tissue was seen through the ligament surface. The synovium of PCL was missing on the surface in contact with ACL. The tension of PCL was good. After the anterolateral bundle was partially excised, the degeneration was severe deep inside, and the ligament surface layer remained apparently normal (Fig. 2). Regarding the posteromedial bundle, the degeneration was localized, and a debulking operation was performed.

Pathological findings in the early stage of the disease showed evidence of intravascular thrombus formation (Fig. 3(A)), and the intraligamentous vascular degeneration and the severity of MD were proportional.

Knee arthralgia disappeared on the day of the operation. ROM of the knee joint improved up to −5/130° within 3 days after the operation. Postoperative MRI scan obtained 1 year after surgery revealed decreased signal intensity and reduced ligament expansion with a contrast effect over the entire length of PCL on CE-MRI (Fig. 4). At present, five years after the operation, the patient has no clinical symptoms.

MRI showed a mass lesion in the suprapatellar pouch. During arthroscopic surgery, yellowish frond-like or papillary villi were noted

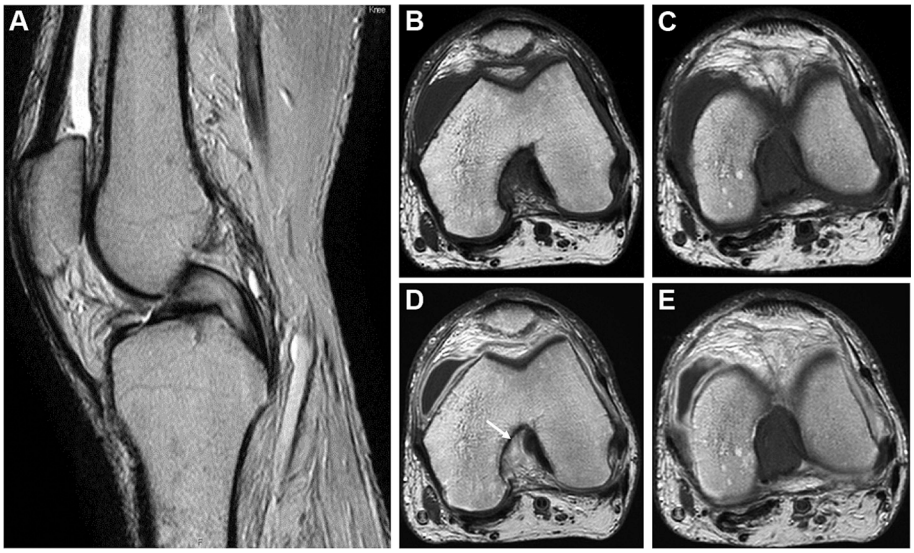

Fig. 1. Preoperative magnetic resonance imaging. Preoperative T2-weighted sagittal image showed tram-track appearance (A). Preoperative T1-weighted axial image (B, C). Preoperative T1-enhanced axial image showed contrast effect (arrow) at the posterior cruciate ligament (PCL) proximal intercondylar femoral attachment side (D), but not at the PCL tibial attachment side (E).

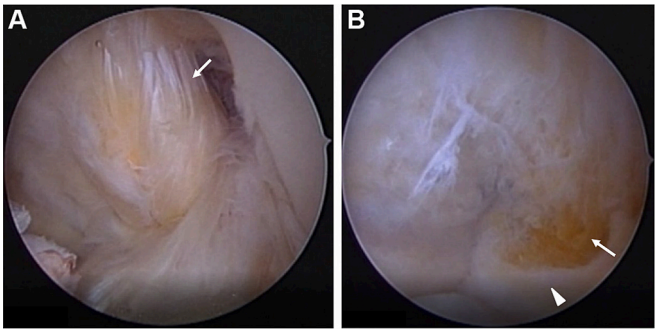

Fig. 2. Arthroscopic findings. The envelop synovium of posterior cruciate ligament (PCL) was partially missing (arrow) on the surface in contact with anterior cruciate ligament (A). The degeneration was strong in deep inside (arrow) and the surface layer of the ligament remained apparently normal properties (arrow head) in PCL anterolateral bundle (B).

in the suprapatellar pouch. Resection of the mass lesion was performed including the basal portion. Histological findings revealed that this mass lesion was lipoma arborescens (LA) (Fig. 5). On the healthy side, LA was not present on MRI.

3. Discussion

Ligament homeostasis is maintained by ligament cell (LC) homeostasis and affected by blood flow and mechanical stress [2,8].

Due to its anatomical features, ACL is susceptible to mechanical forces. Increased tension on the ACL induces inflammatory reactions through the production of various cytokines that induce extracellular matrix degradation by proteases, resulting in denatured LAP deposition [3]. In addition, external mechanical stress from the surrounding tissues triggered by a narrow intercondylar notch can easily disrupt nutrient vessels and lymphatic flow in the envelope synovium of ACL. These factors could explain why MD-ACL occurs more frequently than MD-PCL in middle-aged adults.

Conversely, MD-PCL is characterized by a high incidence of chondrocyte metaplasia caused by hypoxia [13], suggesting the involvement of reduced nutrient blood flow. In fact, occlusion of the nutrient vessels

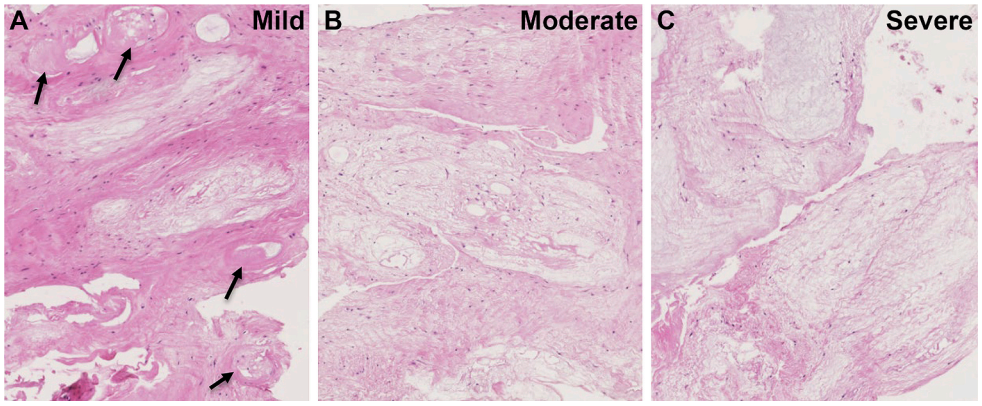

Fig. 3. Histological findings. Histological findings were classified into three stages depending on the degree of intraligamentous nutrient vessel degeneration and the severity of mucoid degeneration (MD). Vascular occlusion with thrombus formation or hyalinization (arrows) and focal island shaped MD on mild stage (A). Vascular structures have disappeared on the severe stage (C). (hematoxylin–eosin, ×200).

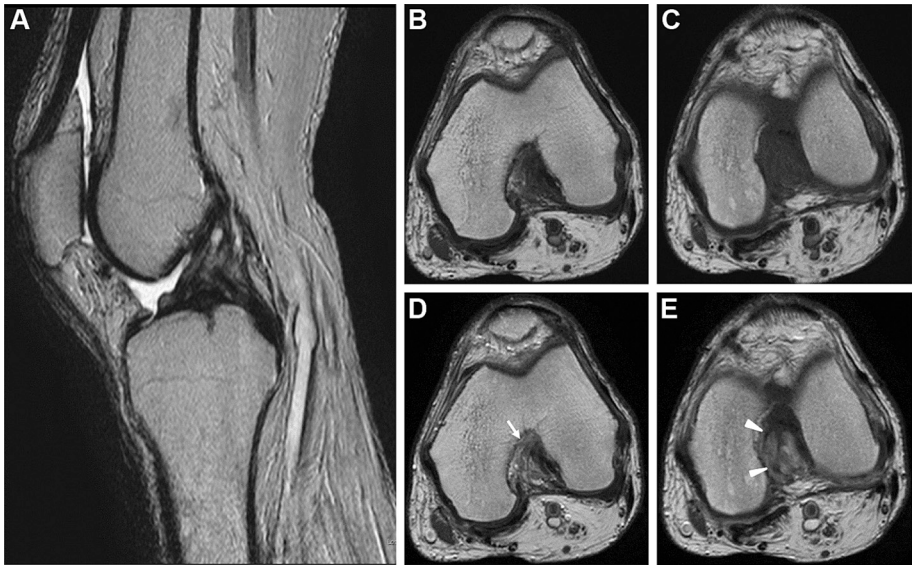

Fig. 4. Postoperative magnetic resonance imaging. Postoperative T2-weighted sagittal image showed a reduction of ligament expansion with normal ligament intensity (A). Postoperative T1-weighted axial image (B, C). Postoperative T1-enhanced axial image showed contrast effect at the posterior cruciate ligament proximal intercondylar femoral attachment side (arrow) (D) and tibial attachment side (arrow head) (E).

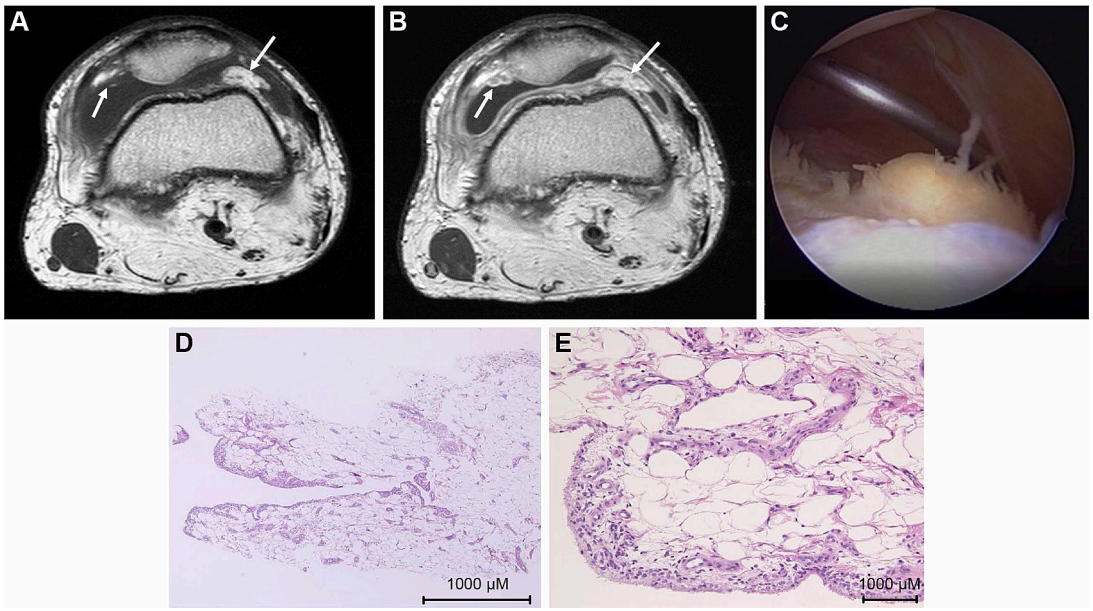

Fig. 5. Lipoma arborescens. Mass having villi-like protrusions with adipose tissue in the inside (arrow) (A; T1-weighted axial image, B; T1-enhanced axial image) composed of fatty proliferation with yellowish frond-like villi (C). Synovial thickening replaced by mature adipocytes and enlarged hyperemic capillaries with mature plasm cell infiltration (D, E). (hematoxylin–eosin, D × 200, E × 400).

within the ligament was observed in this case. Arthroscopic findings revealed that the superficial ligament has a normal component with an almost intact envelope synovium unaffected by mechanical external forces, and MD changes are more pronounced in the deep ligament, resulting in tram-track appearance on MRI [4] (Figs. 1 and 2). These findings indicate that the superficial ligament maintains its homeostasis by nutrient exchange and waste discharge directly between the joint fluid, although these are dysfunctional in the deep ligament. An exacerbating factor for the dysfunction of the intraligamentous nutrient and lymphatic vessels may be increased intraligamentous pressure, which is caused by the excessive deposition of denatured LAP, forming progressive lesions [4]. Debulking surgery [1] is thought to improve the circulatory dynamics of blood and lymph flow within the ligament by reducing pressure within the ligament, thus preventing the deposition of denatured LAP.

Furthermore, LC aging [17], along with the development of atherosclerotic lesions [18], may result in the same prevalence of MD-PCL and MD-ACL in the elderly [13,14]. As the factors that cause impaired intraligamentous blood flow can occur simultaneously in ACL and PCL, it is conceivable that MD-ACL may co-occur with MD-PCL.

As the etiopathogenesis of this case, the chronic mechanical irritation due to knee subclinical instability caused by poliomyelitis may have contributed to the simultaneous development of MD-PCL and LA. However, given the rapid onset of pain and limitation of ROM with nutrient vessel occlusion, this symptomatic MD-PCL case was ultimately concluded to have been caused by LA. No reports exist on other cases of MD-PCL caused by poliomyelitis. Coexisting LA probably provoked some inflammatory impact with hypercoagulability [19], resulting in thromboembolism of the envelope synovial nutrient vessel. When embolization occurs rapidly, the stimulation of nerve endings in the envelope synovium by increased levels of cytokines and neurotransmitters might cause sudden restrictions of ROM and acute severe pain. After the resection of the coexisting LA, the impact of the neurotransmitter disappeared, thereby immediately eliminating the symptoms of nerve irritation, such as pain and limited ROM.

McMonagle et al. reported that normal physical findings under anesthesia are a hallmark of MD-PCL [4]. Consistent with this report, restricted ROM under anesthesia was not observed in this patient. These findings indicate that the cause of restricted ROM in MD-PCL with LA is not a mechanical block but a neural factor, i.e., nociceptive reflexes.

However, there are cases with an envelope synovium lacking [1] and adjacent bone erosion with a narrow notch width in MD-ACL [11]. These pathological changes indicate that actual mechanical impingement with the surrounding tissue occurs in some cases with hypertrophied MD-ACL [1] affected by proinflammatory mediators [20].

4. Conclusion

As awareness of MD-PCL and LA increases and MRI technology advances, the incidence of MD-PCL with LA will likely increase, allowing clinicians to address and resolve several unanswered questions regarding these clinical entities.

Declaration of informed consent

Written consent for the publication of this case report and its accompanying images was acquired from the patient. There is no information in the submitted manuscript that can be used to identify the patient.

Ethical approval

According to our institution`s research ethics committee, ethical approval is not required for this case report. The committee has verified that the report adheres to standard clinical practice and does not involve experimental interventions or the need for additional data collection.

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Author contribution

The author confirms sole responsibility for the following: study conception and design, data collection, analysis and interpretation of results, and manuscript preparation.

Guarantor

Akira Inoue

Research registration number

The present article does not discuss a new surgical technique or examination method, but rather the etiology of the extremely rare symptomatic MD-PCL with LA, based on a combination of imaging, arthroscopic, and histopathologic findings.

Although this is the first article to discuss the etiology of symptomatic MD-PCL with LA, the surgical techniques and imaging methods used are part of the established standard of orthopedic treatment.

Therefore, this paper does not belong to the category of so-called "a first-in-man case". Thus, registration of Research Studies is not required.

Conflict of interest statement

None.

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References

[1] F. Lintz, N. Pujol, P. Boisrenoult, K. Bargoin, P. Beaufils, D. Dejour, Anterior cruciate ligament mucoid degeneration: a review of the literature and management guidelines, Knee Surg. Sports Traumatol. Arthrosc. 19 (2011) 1326–1333, https://doi.org/10.1007/s00167-011-1433-0.

[2] J. Parkinson, T. Samiric, M.Z. Ilic, J. Cook, C.J. Handley, Involvement of proteoglycans in tendinopathy, J. Musculoskelet. Neuronal Interact. 11 (2011) 86–93.

[3] T. Samiric, M.Z. Ilic, C.J. Handley, Large aggregating and small leucine-rich proteoglycans are degraded by different pathways and at different rates in tendon, Eur. J. Biochem. 271 (2004) 3612–3620, https://doi.org/10.1111/j.0014-2956.2004.04307.x.

[4] J.S. McMonagle, C.A. Helms, W.E. Garrett, E.N. Vinson, Tram-track appearance of the posterior cruciate ligament (PCL): correlations with mucoid degeneration, ligamentous stability, and differentiation from PCL tears, AJR 201 (2013) 394–399, https://doi.org/10.2214/AJR.11.7400.

[5] D. Bergin, W.B. Morrison, J.A. Carrino, S.N. Nallamshetty, A.R. Bartolozzi, Anterior cruciate ligament ganglia and mucoid degeneration: coexistence and clinical correlation, AJR 182 (2004) 1283–1287, https://doi.org/10.2214/ajr.182.5.1821283.

[6] S.L. Viana, J.L. Fernandes, J.L. Mendonça, F.M. Freitas, Diffuse intrasubstance signal abnormalities of the posterior cruciate ligament: the counterpart of the mucoid degeneration of the anterior cruciate ligament? A case series, JBR-BTR 91 (2008) 245–248.

[7] E. Solari, C. Marcozzi, D. Negrini, A. Moriondo, Lymphatic vessels and their surroundings: how local physical factors affect lymph flow, Biology 9 (2020) 463, https://doi.org/10.3390/biology9120463.

[8] W. Petersen, B. Tillmann, Blood and lymph supply of the posterior cruciate ligament: a cadaver study, Knee Surg. Sports Traumatol. Arthrosc. 7 (1999) 42–50, https://doi.org/10.1007/s001670050119.

[9] F. Draghi, C. Bortolotto, D.R. Coscia, M. Canepari, S. Gitto, Magnetic resonance imaging of degenerative changes of the posterior cruciate ligament, Acta Radiol. 58 (2017) 338–343, https://doi.org/10.1177/0284185116653280.

[10] Y.S. Youm, S.D. Cho, H.Y. Cho, S.H. Jung, Relationship between mucoid degeneration of the anterior cruciate ligament and posterior tibial slope in patients with total knee arthroplasty, Knee Surg. Relat. Res. 28 (2016) 34–38, https://doi.org/10.5792/ksrr.2016.28.1.34.

[11] J.H. Cha, S.H. Lee, M.J. Shin, B.K. Choi, Relationship between mucoid hypertrophy of the anterior cruciate ligament (ACL) and morphologic change of the intercondylar notch: MRI and arthroscopy correlation, Skeletal Radiol. 37 (2008) 821–826, https://doi.org/10.1007/s00256-008-0527-3.

[12] V. Pandey, C.P.S. Suman, S. Sharma, S.P. Rao, K.V.K. Kiran Acharya, C. Sambaji, Mucoid degeneration of the anterior cruciate ligament: management and outcome, Indian J. Orthop. 48 (2014) 197–202, https://doi.org/10.4103/0019-5413.128765.

[13] Y.D. Levy, A. Hasegawa, S. Patil, J.A. Koziol, M.K. Lotz, D.D. D'Lima, Histopathological changes in the human posterior cruciate ligament during aging and osteoarthritis: correlations with anterior cruciate ligament and cartilage changes, Ann. Rheum. Dis. 72 (2013) 271–277, https://doi.org/10.1136/annrheumdis-2012-201730.

[14] A. Hasegawa, S. Otsuki, C. Pauli, S. Miyaki, S. Patil, N. Steklov, et al., Anterior cruciate ligament changes in the human knee joint in aging and osteoarthritis, Arthritis Rheum. 64 (2012) 696–704, https://doi.org/10.1002/art.33417.

[15] D. Ventura, J.H. Nuñez, N. Joshi-Jubert, E. Castellet, J. Minguell, Outcome of arthroscopic treatment of mucoid degeneration of the anterior cruciate ligament, Clin. Orthop. Surg. 10 (2018) 307–314, https://doi.org/10.4055/cios.2018.10.3.307.

[16] C. Sohrabi, G. Mathew, N. Maria, A. Kerwan, T. Franchi, R.A. Agha, The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. Lond. Engl. 109 (5) (2023) 1136–1140.

[17] P. Kannus, M. Paavola, L. Józsa, Aging and degeneration of tendons, in: N. Maffulli, P. Renström, W.B. Leadbetter (Eds.), Tendon injuries, Springer, London, 2005, pp. 25–31, https://doi.org/10.1007/1-84628-050-8_4.

[18] G. Stubbs, J. Dahlstrom, P. Papantoniou, M. Cherian, Correlation between macroscopic changes of arthrosis and the posterior cruciate ligament histology in the osteoarthritic knee, ANZ J. Surg. 75 (2005) 1036–1040, https://doi.org/10.1111/j.1445-2197.2005.03610.x.

[19] A.R. Fraser, M.E. Perry, A. Crilly, J.H. Reilly, A.J. Hueber, I.B. McInnes, Lipoma arborescens co-existing with psoriatic arthritis releases tumour necrosis factor alpha and matrix metalloproteinase 3, Ann. Rheum. Dis. 69 (2010) 776–777, https://doi.org/10.1136/ard.2008.106047.

[20] G. Macchi, E. Stocco, C. Stecco, E. Belluzzi, M. Favero, A. Porzionato, et al., The infrapatellar fat pad and the synovial membrane: an anatomo-functional unit, J. Anat. 233 (2018) 146–154, https://doi.org/10.1111/joa.12820.