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Original Article

Intraligamentous synovial osteochondroma of the ligamentum teres: a series of 14 cases

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Abstract: Background: The ligamentum teres (LT) is covered by synovium. It acts as a stabilizer of the hip and as such it has been compared to the ACL of the knee joint. Pathologic changes occur in the LT with aging and osteoarthritis (OA), including degeneration, occasional chondroid metaplasia, and synovial chondromatosis are well-recognized in the literature. However, there are no reports of intraligamentous synovial osteochondroma occuring in the LT. Methods: We reviewed the pathology reports of 542 osteoarthritic femoral arthroplasty specimens between January 2016 and December 2018. The LT was examined histologically in 55 cases because it was abnormal on gross examination. Results: A single synovial osteochondroma, ranging in size from 0.4-1.7 cm in diameter, was present in the body of the LT in 14 cases (9 males; 5 females, aged 34 to 81 years), representing 2.6% of 542 arthroplasty cases. Ten of the osteochondromas had bone marrow fat without hematopoietic elements, 1 had hematopoietic elements, and 3 had no marrow among the bony trabeculae. Radiographically, all cases had moderate to severe osteoarthritis with no mention of an abnormality of LT. Conclusion: To our knowledge, this is the first report of intraligamentous synovial osteochondroma in the LT in osteoarthritis patients undergoing hip arthroplasty. It provides further support for microscopic examination of arthroplasty specimens for histologic abnormalities. Further prospective study is needed to determine if this lesion contributes adversely to the development or progression of osteoarthritis and if it is a reactive or neoplastic process.

Keywords: Ligamentum teres, intraligamentous synovial osteochondroma, osteochondroma, osteoarthritis

Introduction

The ligamentum teres (LT) is a short, triangular connective tissue structure at the top of the hip joint. The base of the LT arises from the sides of the acetabular notch where it blends with the transverse acetabular ligament, and the apex attaches to the fovea capitis on the medial aspect of the femoral head [1-3]. The LT is composed of a mixture of collagen types I, III, IV and V interspersed with blood vessels, nerves, and adipose tissue, and is covered by a single layer of synoviocytes [4, 5]. The LT has structural anatomy and functions that are similar to those of the anterior cruciate ligament of the knee joint [4, 6]. It plays a significant role in stabilization of the hip [1, 4, 7] and protects the femoral head from subluxation.

The LT undergoes a variety of degenerative changes associated with aging and osteoarthritis (OA) in which all structures in the hip joint, including the articular cartilage, ligaments, and subchondral bone are affected. Microscopic tears, mucoid and fibromatous degeneration, fatty change of the LT, and chondroid metaplasia of fovea capitis were reported by Sampatchalit et al. [8] in a study of LT of 11 cadaveric hips that had been frozen, thawed for 36 hours without fixation, MRI-scanned, fixed in formalin for 3 days, and then decalcified for 8 days. The authors had no clinical information on these individuals other than age, which ranged from 60-95 years, and sex (8 females and 3 males) [8]. Dehao et al. [5] described osteochondroid and chondroid metaplasia in 3 of 11 LTs removed at arthroplasty for femoral

Table 1. Osteoarthritis Research Society International (ORSI) working group grading and staging systems

Grade Assessment		Stage Assessment	
Grade	Key Features	Stage	% Involvement (Surface, area, volume)
G0	Surface intact, cartilage morphology intact	S0	No OA features seen
G1	Surface intact	S1	<10%
G2	Surface discontinuity	S2	10-25%
G3	Vertical fissure (cleft)	S3	25-50%
G4	Erosion: Cartilage matrix loss of superficial layer and mid zone	S4	>50%
G5	Denudation: Sclerotic bone or reparative tissue, including fibrocartilage within denuded surface. Microfracture, with repair limited to bone surface		
G6	Deformation: Bone remodeling (more than osteophyte formation only). Includes: microfracture with fibrocartilaginous and osseous repair extending beyond the bone surface		

fracture (8 cases) or OA (3 cases). Metaplasia was more prevalent in patients with more severe degenerative changes. The focus of this study was to describe the microstructure of the ligamentum teres of the hip, with special emphasis on the presence of osteochondroid and chondroid metaplasia since they had not been described histologically in detail, making it unclear whether these were bone fide examples of synovial osteochondromas of the ligamentum teres. Thus, to our knowledge, osteochondroma has not been reported in the LT, and no cases were included in a recent review of the literature of the hip [9] or in a comprehensive review of MRI studies of the LT [10].

Here, we report the histologic, clinical, and radiologic features of 14 cases of synovial osteochondroma within the body of LT identified in a systematic histopathologic review of patients undergoing arthroplasty for osteoarthritis of the hip over a 3-year period in a tertiary medical center.

Materials and methods

The senior author (BFB) diagnosed 2 cases of intraligamentous synovial osteochondroma of the LT within a 2-week period in 2016. This prompted a retrospective and prospective review of our surgical pathology database over a 3-year period from January 2016 to December 2018 for femoral heads that had been submitted with an attached LT from patients undergoing arthroplasty for degenerative joint disease. We found 542 arthroplasty cases submitted for histologic analysis, of which 55 had the LT submitted for histologic examination mainly because they had areas of nodularity or firmness identified during gross examination; a few were submitted randomly with no significant changes grossly. Fourteen of these 55 cases had a diagnosis of osteochondroma of the ligamentum teres made in formalin-fixed, acid decalcified, paraffin-embedded, 4 μm thick, hematoxylin and eosin (H&E)-stained sections. The histologic features of H&E sections from the associated decalcified, paraffin-embedded femoral heads were reviewed. The severity of degenerative joint disease was assessed histologically by applying the Osteoarthritis Research Society International (ORSI) working group grading and staging systems, which are based on the depth and extent of cartilage involvement, respectively [11] (**Table 1**). We correlated these data with the clinical and radiologic findings.

Results

A single synovial osteochondroma was found within the body of the ligamentum teres in 14 of the 55 cases (9 males; 5 females, aged 34-81 years). The osteochondromas were identified grossly as a single, small nodule in 13 cases. They varied in size from 0.4-1.7 cm in diameter, and one case had only a focus of firmness. None of these lesions had been identified on routine radiographs before surgery and no loose bodies were identified. Histologically, these nodules were well-circumscribed and consisted of mainly woven trabecular bone covered by cellular viable cartilage, with overlying synovium, features typical of synovial osteochondroma (**Figure 1A-C**). The bone was

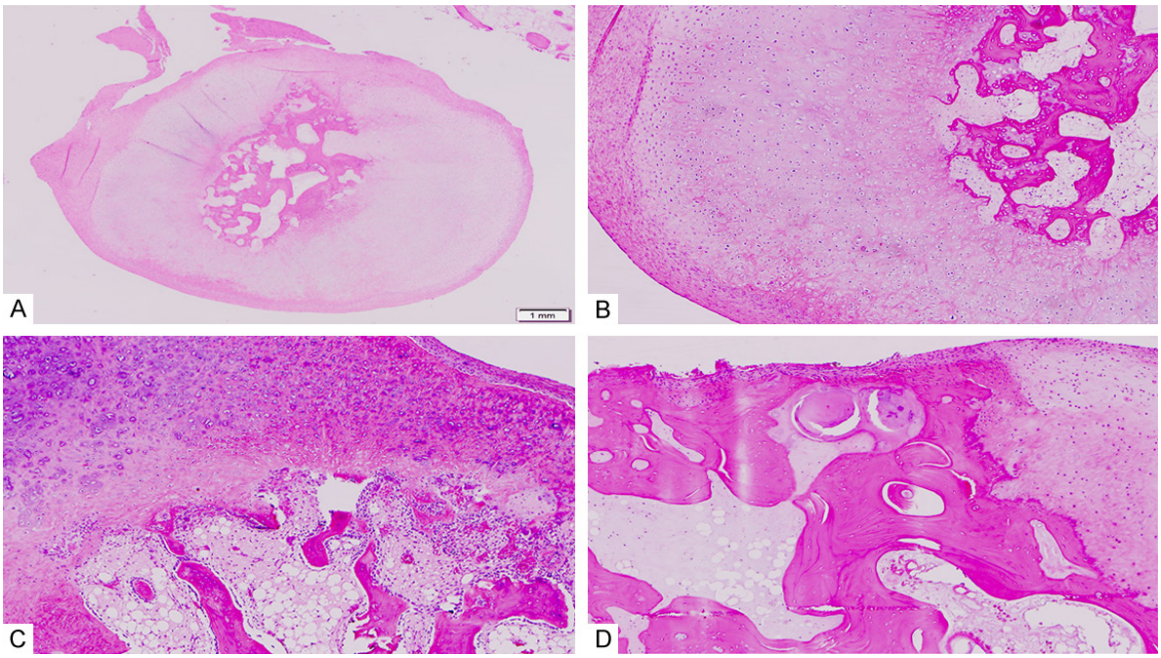

Figure 1. Synovial osteochondromas of the ligamentum teres. (A and B) Low (H&E ×20) and higher (H&E ×40) magnification images of an osteochondroma with cellular cartilage, central ossification, and a covering rim of fibrous tissue. Chondrocytes and fibroblastic cells in the marrow cavity are viable, but osteocyte lacunae are empty, indicative of osteonecrosis, presumably due to inadequate blood supply. (C) (H&E ×40) Osteochondroma with cellular cartilage and a covering of fibrous tissue with a thin outer layer of synoviocytes. Central, mostly viable, remodeling bone with active osteoclastic resorption and osteoblastic new bone formation and fatty marrow. (D) (H&E ×40) Eburnated surface of femoral head from same case as (C), with subchondral fractures repair, and osteonecrosis.

necrotic in one case (**Figure 1B**), but it was viable in most cases, with active osteoclastic resorption and new bone formation (**Figure 1C**). Ten specimens had bone marrow fat with no marrow hematopoietic elements, one had marrow hematopoietic elements, and three had no marrow among the bony trabeculae. Twelve of the 14 cases had mild to moderate hyperplasia of the joint synovium and four of these had associated mild to moderate focal chronic inflammation in the synovial tissues. Of the remaining 41 LT specimens, 2 had a focus of reactive chondroid metaplasia within the LT with no associated synovial covering, one had synovial chondromatosis present on the femoral head articular surface and in the joint cavity, and one had calcium pyrophosphate crystal deposits. The other 37 LT specimens had non-specific reactive changes, including mild to moderate synovial hyperplasia, fibrosis, mild focal chronic inflammation, focal calcification and bony detritus.

Twelve of the 14 synovial osteochondroma cases had primary and 2 had secondary OA. One patient had developmental dysplasia of the hip and the other had post-traumatic arthritis. Radiologically, all of the cases had moderate to severe arthritic changes without any mention of abnormality of the LT. In three cases, avascular necrosis of the femoral head was diagnosed histologically with viable overlying cartilage. Most of the cases had histologically advanced grade OA with varying degrees of eburnation of the articular cartilage (**Figure 1D**) and underlying subchondral cyst formation and microfracture repair (**Figure 2A**, **2B**). Eight cases were ORSI grade G5, 5 were G6, and 1 was G4, with minimal loss of articular cartilage. In addition, 11 cases had extensive loss of 50% or more of the articular cartilage (ORSI Stage 3 & 4) and 3 had <50% loss (Stage 2; **Table 2**).

Discussion

To our knowledge this is the first definitive report of osteochondroma of the ligamentum teres (LT) occurring in 14 of 55 hip arthroplasty cases over a 3-year period as solitary discrete nodules submitted for histologic examination. This represents 2.6% of 527 arthroplasty specimens from OA patients and 25.5% of the sub-

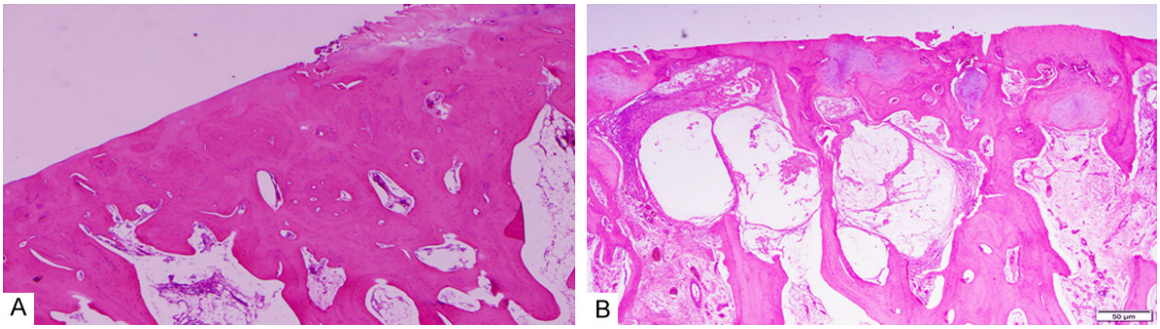

Figure 2. Femoral heads with advanced grades of osteoarthritis. A. G5 degenerative joint disease with complete loss of articular surface (H&E ×20). B. G6 degenerative joint disease with complete loss of articular surface, subchondral microfracture, and subchondral cyst.

Table 2. Clinicoradiologic and histologic correlation

Case No.	Age	Sex	Radiologic findings	Histologic Grade	Histologic Stage
1	67	F	Advanced DJD with osteophytes and avascular necrosis	G4	S2
2	66	F	Post traumatic moderate DJD with osteophytes	G6	S4
3	70	M	Advanced DJD	G5	S3
4	61	F	Advanced DJD	G5	S2
5	65	M	Advanced DJD	G6	S4
6	66	F	Advanced DJD	G5	S3
7	73	F	Advanced DJD	G5	S3
8	81	F	Advanced DJD with complete erosion of femoral head	G5	S4
9	34	F	Flattening of the femoral head and superior aspect of the acetabulum with severe DJD, developmental dysplasia of the hip and avascular necrosis	G5	S4
10	70	F	Advanced DJD	G5	S3
11	55	M	Advanced DJD	G5	S2
12	60	F	Advanced DJD	G6	S4
13	71	M	Advanced DJD with osteophytes	G6	S4
14	59	M	Loss of the superior lateral joint space in the hip joint with contiguous bony sclerosis. Subchondral cysts are present. Large marginal osteophytes arise from the lateral and medial aspects of the acetabular margin and also from the lateral and medial aspects of the femoral head and neck. Findings are consistent with moderately severe degenerative changes.	G6	S4

mitted LT specimens. The osteochondromas were not suspected clinically, and despite one being 1.7 cm in diameter, none was detected during routine radiographic examination, which would be difficult, given the complexity of the bones at the hip. Tumors or tumor-like lesions of the LT are extremely rare. We are aware of only 2 reported cases: a giant cell tumor in a 46-year-old woman with an 18-month history of groin, thigh, and buttock pain [12], and a fibromyxoid pseudotumor in a 29-year-old man with a 2-year history of right hip pain [13]. All of our arthritic patients had hip pain, but we do not know whether the osteochondromas of the LT contributed to the pain.

The LT and ACL share many structural, anatomic, and functional characteristics [4, 6], including showing degenerative changes during aging and in OA, disorganization of collagen fibers, mucoid and chondroid metaplasia, calcification, and synovial chondromatosis [14-19].

Ligament-derived stem cells with adipogenic, osteogenic, and chondrogenic potential have been isolated from human ACLs [20-22] and other ligaments [20], and were more often found in ACLs of older than younger patients [22], although they had similar multilineage differentiation potentials. Hasegawa et al. [23], hypothesized that chondrocyte-like cells prolif-

erate from abnormally differentiated progenitor cells in degenerating ACLs. Wagenbrenner et al. [24] isolated mesenchymal stromal cells (MSCs) from the LTs of five patients who underwent total arthroplasty and documented that these cells had adiopogenic and osteogenic differentiation potential but the chondorogenic differentiation potential was not confirmed. Further, they did not do a quantitative measure of this differentiation potential [24] and we found no studies reporting the density of ligament-derived stem cells in the LT.

OA affects every structure in the joint and causes degeneration of ligaments, triggered by inflammatory mediators and cytokines secreted by synovial cells and chondrocytes [25]. We do not know whether the osteochondromas started in the LT as a neoplastic or reactive process or whether they developed before the degeneration of articular cartilage or avascular necrosis occurred, or as a result of the joint pathology. Blankenbaker et al. [26] reported partial LT tears in 116 patients undergoing preoperative MR arthrography and hip arthroscopy, but none had intraligamentous osteochondroma. More recently, tears but no osteochondromatous lesions, were reported in 14-45% of LTs in a comprehensive review of reports of 1456 MRI examinations to determine the diagnostic performance of MRI and MR arthrography for depicting LT lesions [10]. Because osteochondromas have not been reported previously in the LT, they could have been present in some cases, but not recognized. MRI was not done in most of our cases, and is not medically indicated preoperatively in most patients with arthritis.

The purpose of this study is to report that osteochondromas can be detected in the LT by careful gross and histologic examination. In many hospitals, arthroplasty specimens are examined grossly to confirm features of OA, and no histologic sections are examined. The cost effectiveness of examination of these specimens has been questioned [27-30], mostly in relatively small studies, but histologic analysis can lead to diagnosis of unsuspected pathology, including calcium pyrophosphate crystal disease, primary and metastatic tumors, pigmented villonodular synovitis (tenosynovial giant cell tumor), and synovial chondromatosis [27, 31-34]. The largest study to date reported that the cost of histologic analysis was <0.5% of the medical cost of the procedure [35].

We cannot tell whether ostoechondromas of LT contributed adversely to the development of OA, occurred independently, or are an end-stage manifestation of OA, or a neoplastic process. This would have required pre-operative MRI and other imaging studies, coupled with careful histologic examination, karyotype analysis, and molecular genetic testing of resected specimens. A limitation of current pathology practice is that arthroplasty specimens are typically decalcified in acid-containing solutions. Thus, we were unable to perform FISH analysis, targeted RNA sequencing, or next generation sequencing studies, which require decalcification in EDTA, to determine whether synovial osteochondromas of the LT are neoplastic.

Disclosure of conflict of interest

None.

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References

[1] Martin RL, McDonough C, Enseki K, Kohreiser D, Kivlan BR. Clinical relevance of the ligamentum teres: a literature review. Int J Sports Phys Ther 2019; 14: 459-467.

[2] Bardakos N and Villar R. The ligamentum teres of the adult hip. J Bone Joint Surg Br 2009; 91: 8-15.

[3] Perumal V, Woodley SJ and Nicholson HD. Ligament of the head of femur: a comprehensive review of its anatomy, embryology, and potential function. Clin Anat 2016; 29: 247-255.

[4] O'Donnell JM, Devitt BM and Arora M. The role of the ligamentum teres in the adult hip: redundant or relevant? A review. J Hip preserv Surg 2018; 5: 15-22.

[5] Dehao BW, Bing TK and Young JLS. Understanding the ligamentum teres of the hip: a histological study. Acta Ortop Bras 2015; 23: 29-33.

[6] Wenger D, Miyanji F, Mahar A and Oka R. The mechanical properties of the ligamentum teres: a pilot study to assess its potential for improving stability in children's hip surgery. J Pediatr Orthop 2007; 27: 408-410.

[7] Cerezal L, Kassarjian A, Canga A, Dobado MC, Montero JA, Llopis E, Rolón A and Pérez-Carro L. Anatomy, biomechanics, imaging, and management of ligamentum teres injuries. Radiographics 2010; 30: 1637-1651.
[8] Sampatchalit S, Barbosa D, Gentili A, Haghighi P, Trudell D and Resnick D. Degenerative changes in the ligamentum teres of the hip: cadaveric study with magnetic resonance arthrography, anatomical inspection, and histologic examination. J Comput Assist Tomogr 2009; 33: 927-933.
[9] van der Valk MR, Veltman ES, Assink J and Veen MR. Synovial chondromatosis of the hip, a case report and literature review. J Orthop 2019; 16: 249-253.
[10] Shakoor D, Farahani SJ, Hafezi-Nejad N, Johnson A, Vaidya D, Khanuja HS, Eng J and Demehri S. Lesions of ligamentum teres: diagnostic performance of MRI and MR arthrography a systematic review and meta-analysis. Am J Roentgenol 2018; 211: W52-W63.
[11] Pritzker KP, Gay S, Jimenez SA, Ostergaard K, Pelletier JP, Revell PA, Salter D and Van den Berg WB. Osteoarthritis cartilage histopathology: grading and staging. Osteoarthritis Cartilage 2006; 14: 13-29.
[12] Singh P, Constable L and O'Donnell J. Arthroscopic excision of a giant-cell tumour of the ligamentum teres. J Bone Joint Surg Br 2009; 91: 809-811.
[13] McLawhorn AS, Bansal M, Swensen S, Schneider R, Kelly BT and Sink EL. Fibromyxoid pseudotumor of the ligamentum teres treated with fresh osteochondral allograft. Skeletal Radiol 2014; 43: 541-546.
[14] Loeser RF, Goldring SR, Scanzello CR and Goldring MB. Osteoarthritis: a disease of the joint as an organ. Arthritis Rheum 2012; 64: 1697-1707.
[15] Hasegawa A, Otsuki S, Pauli C, Miyaki S, Patil S, Steklov N, Kinoshita M, Koziol J, D'Lima DD and Lotz MK. Anterior cruciate ligament changes in the human knee joint in aging and osteoarthritis. Arthritis Rheum 2012; 64: 696-704.
[16] AlAqeel MA and Al-Ahaideb A. Synovial osteochondroma originating from the synovium of the anterior cruciate ligament. BMJ Case Rep 2013; 2013: bcr2012008218.
[17] Mio K, Tamura K and Nemoto K. Intraligamentous synovial chondromatosis of the anterior cruciate ligament: a case report. JBJS Case connect 2014; 4: e4.
[18] Majima T, Kamishima T and Susuda K. Synovial chondromatosis originating from the synovium of the anterior cruciate ligament: a case report. Sports Med Arhtrosc Rehabil Ther Technol 2009; 1: 6.
[19] Ashir A, Li WX, Shirazian H, Chang DG and Chan EY. Intraligamentous synovial chondromatosis of the anterior cruciate ligament. Skeletal Radiol 2020; 49: 645-650.
[20] Lee KJ, Clegg PD, Comerford EJ and Canty-Laird EG. Ligament-derived stem cells: identification, characterisation, and therapeutic application. Stem Cells Int 2017; 2017.
[21] Huang TF, Chen YT, Yang TH, Chen LL, Chiou SH, Tsai TH, Tsai CC, Chen MH, Ma HL and Hung SC. Isolation and characterization of mesenchymal stromal cells from human anterior cruciate ligament. Cytotherapy 2008; 10: 806-814.
[22] Lee DH, Ng J, Kim SB, Lee KM and Han SB. Effect of donor age on the proportion of mesenchymal stem cells derived from anterior cruciate ligaments. PLoS One 2015; 10: e0117224.
[23] Hasegawa A, Nakahara H, Kinoshita M, Asahara H, Koziol J and Lotz MK. Cellular and extracellular matrix changes in anterior cruciate ligaments during human knee aging and osteoarthritis. Arthritis Res Ther 2013; 15: R29.
[24] Wagenbrenner M, Heinz T, Horas K, Jakuscheit A, Arnholdt J, Herrmann M, Rudert M, Holzapfel BM, Steinert AF and Weißenberger M. The human arthritic hip joint is a source of mesenchymal stromal cells (MSCs) with extensive multipotent differentiation potential. BMC Musculoskelet Disord 2020; 21: 297.
[25] Goldring MB and Otero M. Inflammation in osteoarthritis. Curr Opin Rheumatol 2011; 23: 471-478.
[26] Blankenbaker DG, De Smet AA, Keene JS and del Rio AM. Imaging appearance of the normal and partially torn ligamentum teres on hip mr arthrography. Am J Roentgenol 2012; 199: 1093-1098.
[27] Lin MM, Goldsmith JD, Resch SC, DeAngelis JP and Ramappa AJ. Histologic examinations of arthroplasty specimens are not cost-effective: a retrospective cohort study. Clin Orthop Relat Res 2012; 470: 1452-1460.
[28] Lawrence T, Moskal JT and Dliduch DR. Analysis of routine histological evaluation of tissues removed during primary hip and knee arthroplasty. J Bone Joind Surg Am 1999; 81: 926-931.
[29] Kocher MS, Erens G, Thornhill TS and Ready JE. Cost and effectiveness of routine pathological examination of operative specimens obtained during primary total hip and knee replacement in patients with osteoarthritis. J Bone Joint Surg Am 2000; 82: 1531.
[30] Meding JB, Ritter MA, Jones NL, Keating EM and Faris PM. Determining the necessity for routine pathologic examinations in uncompli-

cated total hip and total knee arthroplasties. J Arthroplasty 2000; 15: 69-71.
[31] Billings SD, Wurtz LD, Tejada E and Henley JD. Occult sarcoma of the femoral head in patients undergoing total hip arthroplasty: a report of two cases. J Bone Joint Surg Am 2000; 82: 1536.
[32] Vadivelu R, Kar N, Margetts M and Abdul-Cader A. Non-Hodgkin's lymphoma as an unexpected diagnosis in a hip arthroplasty. Arch Orthop Trauma Surg 2002; 122: 532-534.
[33] Lauder AJ, Cheatham SA and Garvin KL. Unsuspected non-Hodgkin's lymphoma discovered with routine histopathology after elective total hip arthroplasty. J Arthroplasty 2004; 19: 1055-1060.
[34] DiCarlo EF and Klein MJ. Comparison of clinical and histologic diagnoses in 16,587 total joint arthroplasties: implications for orthopedic and pathologic practices. Am J Clin Pathol 2014; 141: 111-118.
[35] Holbrook HS, Plate JF, Langfitt MK, Lang JE and Shields JS. Cost analysis of sending routine pathology specimens following total joint arthroplasty in the age of bundled payments. Surg Technol Int 2017; 31: 182-188.