

Hip pain in an elite cyclist with Non-Hodgkin's Follicular Lymphoma: a case report

Melissa Belchos, BSc(Hons), DC¹

Varsha Kumar, BSc(Hons), DC, DACBR, FCCR(C)²

Carol Ann Weis, MSc, DC³

Objective: *We present a case of an elite cyclist that hesitated to follow the medical advice from her practitioners, as she was determined to train and compete resulting in delayed diagnosis and management of a rare hip pathology.*

Case presentation: *A 51-year old elite female cyclist had a history of years of hip pain with insidious onset. The chiropractor in this case observed a lack of response to treatment, and advised the patient to get an MRI with suspicion of a labral tear. She eventually agreed to further investigations and was diagnosed with Non-Hodgkin's follicular lymphoma and a labral tear.*

Summary: *Elite athletes are not immune to serious pathology. Chiropractors should be vigilant and ensure*

Douleur à la hanche chez un cycliste d'élite atteint d'un lymphome folliculaire non hodgkinien : un rapport de cas.

Objectif : *Nous présentons le cas d'une cycliste d'élite qui a hésité à suivre les conseils médicaux de ses praticiens, car elle était déterminée à s'entraîner et à participer à des compétitions, ce qui a retardé le diagnostic et la prise en charge d'une pathologie rare de la hanche.*

Présentation du cas : *Une cycliste d'élite de 51 ans avait des antécédents de douleurs à la hanche depuis des années; le début de ses douleurs avait été insidieux. Le chiropraticien a observé une absence de réponse au traitement et a conseillé à la patiente de subir un examen par IRM en soupçonnant une déchirure du labrum. La patiente a fini par accepter de subir des examens complémentaires. Un lymphome folliculaire non hodgkinien et une déchirure labrum ont été diagnostiqués.*

Résumé : *Les athlètes d'élite ne sont pas à l'abri d'une pathologie grave. Les chiropraticiens doivent*

¹ Division of Graduate Studies, Canadian Memorial Chiropractic College

² Diagnostic Imaging, Division of Graduate and Undergraduate Studies, Canadian Memorial Chiropractic College

³ Department of Graduate Education and Research, Canadian Memorial Chiropractic College

Corresponding author: Melissa Belchos, Canadian Memorial Chiropractic College 6100 Leslie Street, Toronto, ON M2H 3J

Tel: 416-482-2340

e-mail: mbelchos@cmcc.ca

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to investigate any patients with a lack of response to conservative management. Chiropractors should be aware of the risk of athletic patients that continue to train and compete when advised not to.

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KEY WORDS: cyclist, diagnosis, elite, hip, labrum, lymphoma, pain, chiropractic

Introduction

Elite athletes undergo substantial physical and mental demands in both their training and sport performance. A cross-sectional study of 518 cyclists reported that 85% of these athletes sustained one or more non-traumatic over-use injuries (with the most common sites including the neck, knees, groin, hands, and back) within the previous year.¹ Of the cyclists in that study, 31% sought medical treatment for their injuries, and on average, had symptoms that persisted for 3.7 months.¹ When the patient's symptoms are not fitting of a musculoskeletal pathology, further investigations are warranted. Whether the patient will follow through with the recommendations of a medical provider, is a separate challenge. Oftentimes, elite athletes may want to return to sport before they are advised to do so.² When athletes perceive their identities to be strongly linked to their sport, they may undergo grief and a loss of existence leading to depression, should they withdraw from sport due to injury.³ Consequently, the constant drive to continue and persevere may stand in the way of an athlete receiving a timely diagnosis or recovery.

Elite athletes are not immune to serious pathology such as infection or malignancies; therefore, chiropractors should be vigilant when patients are not responding to care or if there are signs and symptoms of sinister pathology. One such serious pathology to consider that spans multiple age groups, is lymphoma. Lymphoma is a lymphoid malignancy which typically develops in the lymph nodes.⁴ Hodgkin's or Non-Hodgkin's Lymphoma (NHL) should be suspected if a patient presents with generalized lymphadenopathy that is painless and persistent.⁴

être vigilants et s'assurer d'examiner tous les patients réfractaires à un traitement conservateur. Les chiropraticiens doivent être conscients du risque que représentent les patients sportifs qui continuent à s'entraîner et à participer à des compétitions alors qu'on leur a conseillé de ne pas le faire.

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MOTS CLÉS : cycliste, diagnostic, élite, hanche, labrum, lymphome, douleur, chiropratique

We present a case report of a 51-year old female elite cyclist with ongoing hip pain who was diagnosed with Non-Hodgkin's follicular lymphoma and a labral tear. The purpose of this report is to describe the challenges in diagnosing sinister pathology in athlete patients, and describe one such sinister pathology that chiropractors should consider when evaluating patients with hip pain.

Case presentation

Initial evaluation by the chiropractor

A 51-year old female elite cyclist presented to a chiropractor with a chief complaint of left adductor and hip pain in June of 2018. One month prior she had been on a three-day stage race where she developed a sudden onset of muscle cramping in her left thigh. The cramping started on the first day of the race, 60km into a 140km stage, while on a climb. She reported she had never experienced this symptom before and felt as though she had no power pushing off with her left leg, however she finished that stage of the race. That night she received a massage, and rested. The following day the athlete reported that her left leg was swollen, particularly on the anterior aspect of her thigh, compared to the right. The athlete was able to complete the second stage of 110km with discomfort, though she reported she had to change her race strategy by keeping her wattage low to prevent cramping of her leg and to avoid an "exploding" sensation. Following that stage, she once again, managed her pain with another massage. On the third stage, an uphill 8km time trial, she reported feeling that her left leg pain almost stopped her from finishing the event, but she pushed through to complete it. In

the weeks following this particular race, she managed her symptoms with massage, acupuncture, physiotherapy, and rest. The physiotherapist told the patient that she did not train sufficiently for the race, and that she should stretch. Three months after the initial onset of pain and cramping, she competed in a 24-hour race where she placed first and set a course record. She experienced pain in her left adductor muscles through the entire duration of the race, and performed self-massage for temporary relief.

Red flags (including but not limited to night sweats, unintentional weight loss, previous history of malignancy) were not identified by the chiropractor, and the systems review was normal. This patient had a medical history that included Reynaud's syndrome and eczema, but was otherwise healthy. She was not taking medications. Her family medical history included: maternal hypertension, Sjogren's syndrome, and arthritis; maternal grandmother with hypertension, ovarian cancer and dementia; maternal grandfather with stroke, high cholesterol, and hypertension; paternal colon cancer, hydrocephalus, and hypertension; paternal grandfather with stroke; and sister with uterine cancer. No other malignancies were noted in her family's medical history.

Physical examination

On examination when presenting to the chiropractor, observation revealed no visible swelling or abnormalities of the lower limbs. The femoral triangle on the left side was observed and palpated, with no abnormalities noted. Functional testing revealed a normal squat and quarter squat. Passive hip range of motion was normal bilaterally. Resisted hip flexion with adduction on the left was limited to 2/5 strength due to severe pain in her hip. Orthopaedic testing of the bilateral hips was within normal limits; including hip scour, log roll, FADDR (Flexion Adduction Internal Rotation), and FABER (Flexion Abduction External Rotation). Neurological examination of the lower limb was normal bilaterally. The patient was diagnosed with a left sided overuse adductor strain. She was advised to stretch her left adductors and quadriceps, as well as cease training for the interim.

Follow-ups

The remainder of the 2018 year until early 2019, the patient trained indoors and managed her pain with massage, physiotherapy, and acupuncture. In May of 2019, the pa-

tient returned to the chiropractor with continued pain and discomfort of her left adductor muscles. The patient reported that uphill road cycling would aggravate her pain significantly, but that she never seemed to experience a problem on her indoor bike trainer. The chiropractor noticed pes planus on the left, but no other observable abnormalities. Palpation of the pubic symphysis elicited pain, as well as palpation of the right sacroiliac joint. Passive knee flexion was limited on the left. No hip orthopaedic tests were positive. At this time, the patient was diagnosed with pubic symphysis irritation with sacroiliac joint (SIJ) dysfunction. The chiropractor's plan of management included heat, soft tissue therapy, and manipulative therapy to the right SIJ and pubic symphysis. On follow-up one week later, her pubic symphysis pain had improved but she started experiencing pain in her contralateral SIJ.

On a subsequent follow-up two weeks after her last appointment, the same treatment was applied. However, as the patient had experienced this pain for over a year, her symptoms were not completely resolving, and her pain was aggravated by uphill cycling, the chiropractor recommended she visit her family doctor for magnetic resonance imaging (MRI), querying a labral pathology. The patient did not follow-up with her medical doctor due to her denial that any serious pathology was present and the perception that she may be congesting the medical system. The patient also reported that she would like to continue to train and compete, and thus, participated in a 110km race one month later. At that race, she had horrible cramping and pain in her left anterior thigh at the midway point of the race. She spoke to her chiropractor via a phone call who mentioned the MRI again, but the patient did not follow-up with her medical doctor as she was leaving for Europe within a week's time. She travelled to Italy for a 2-week training camp, followed by another stage race in Norway with the following distances: 145km, 115km, and a 12km time trial with approximately 19,000ft of climbing in total. Upon her return from Europe, she reported a new, burning sensation in the anterior aspect of her left hip and her hip felt fatigued.

For the course of the next two months, the patient decided to follow a self-directed course of massage to the areas of chief complaint. She had seen many different providers for massage and physiotherapy since the pain began, but due to limited improvement of symptoms, she

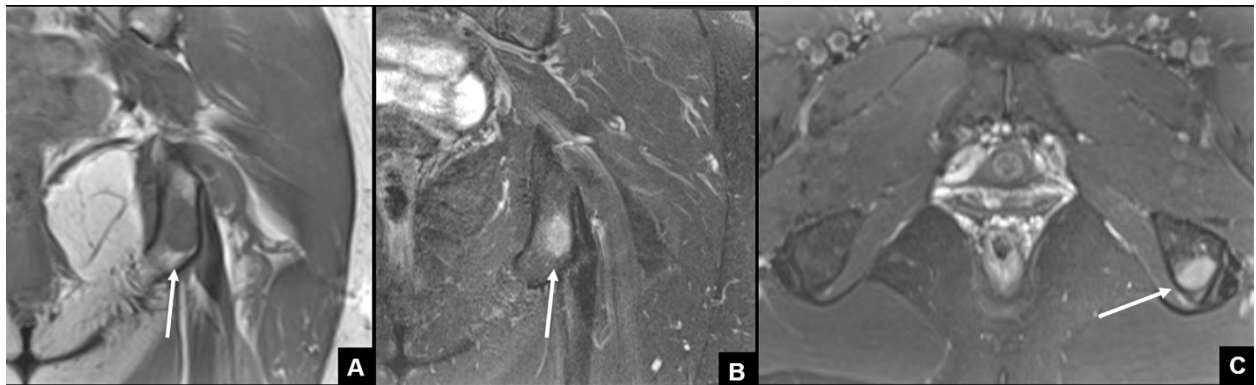


Figure 1.

T1-weighted (A) and T2-weighted FS (B) coronal left hip and T2-weighted FS axial pelvis (C) MRI sequences demonstrating hypointense T1 and hyperintense T2 FS signal signifying bone marrow infiltrative process in the left ischial tuberosity (arrows).

consulted with a sports medical doctor whom she had seen in the past for a different complaint. She was advised to rest, and an MRI of her left hip was requisitioned to rule out a labral tear.

Imaging findings

The MRI revealed a focal low T1-weighted signal, compared to the skeletal muscle, and high fat-saturated T2-weighted bone marrow signal change in the left ischial tuberosity measuring 2.5cm in the cephalad to caudad dimension. A small extraosseous, soft tissue extension is observed posterior to the ischial tuberosity (Figure 1). A

second lesion with the same signal characteristics was observed in the anteromedial acetabular wall of the left hip, extending halfway into the superior pubic ramus, with extension into the soft tissues anterior to the acetabular wall (Figure 2). These signal characteristics are consistent with an infiltrative process, of which a neoplastic condition such as metastatic disease or lymphoma is most likely. No pathological fractures within the pelvis or femurs was observed and no evidence of enlarged lymph nodes. Additionally, a tear of the anterosuperior labrum of the left hip was observed with an associated complete chondral tear at the anterior acetabular surface. There was no evidence

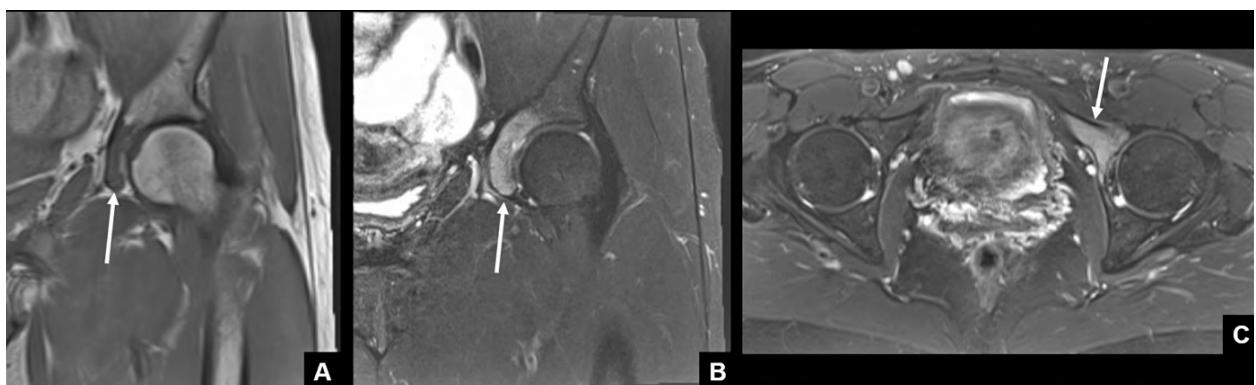


Figure 2

T1-weighted (A) and T2-weighted FS (B) coronal left hip and T2-weighted FS axial pelvis (C) MRI sequences demonstrating hypointense T1 and hyperintense T2 FS signal signifying bone marrow infiltrative process in the left anteromedial acetabulum and superior pubic ramus (arrows).



Figure 3.

Sagittal (A, B) and coronal (C) T2-weighted FS MRI sequences of the left hip demonstrating a tear of the anterosuperior labrum (solid arrows) with a complete chondral tear of the anterior acetabular cartilage (open arrow).

of a labral cyst or bone marrow edema within the adjacent bony attachment (Figure 3).

Given the concerns of a polyostotic condition, a bone scintigraphy scan was performed to assess for other areas of involvement. The whole-body bone scintigraphy demonstrated multiple areas of abnormal radionuclide uptake, most notably involving the right humeral head, the right 7th posterior rib and consistent with the lesions seen on the MRI, in the left ischial tuberosity and anteromedial acetabulum (Figure 4).

Diagnosis

The patient was referred to an oncologist who performed a thorough evaluation, including a fine needle core biopsy. She was subsequently diagnosed with Stage IV Non-Hodgkin's follicular lymphoma that was polyostotic in nature. A left sided anterosuperior labral tear was also noted.

Management

This patient underwent six cycles of four weeks of chemotherapy. During that time, she experienced significant fatigue but strived to walk every day, with an occasional bike ride. On follow-up approximately nine months after her diagnosis, she had a complete response to chemotherapy, with no trace of metabolic activity on her bone scintigraphy scans. The patient was advised she would require consistent monitoring and chemotherapy for the next three years. She began training with a cycling coach again, however sessions were managed and dosed



Figure 4.

Anterior (A) and posterior (B) bone scintigraphy scan of the whole body demonstrates abnormal hot spots at the right humeral head (arrow head), right 7th rib (solid arrow), left ischial tuberosity (open arrow) and left anteromedial acetabulum (thin arrow).

according to her abilities and goals. Presently, she is seeing an oncology physiotherapist to improve her overall functional strength. She aims to ride four to five times per week, complete strengthening exercises every other day, and stretches every day. Her fatigue remains to this day.

Discussion

Background and epidemiology

The majority of injuries sustained by elite cyclists are acute trauma, making up approximately 53% of all injuries in Tour de France athletes from 2010-2017.⁵ It has also been reported that these elite cyclists may experience strains and sprains (5%), overuse injuries (4%), or multiple injuries at once (6%).⁵ When a cyclist is presenting to a chiropractor with hip pain, pathologies that are intra-articular, extra-articular, or mimickers should be considered as differential diagnoses.⁶ Some differential diagnoses to consider in patients with hip pain include but are not limited to: labral tears, femoroacetabular impingement, iliopsoas tendinitis, adductor strain, greater trochanteric bursitis, arthritis, fracture, or malignancy.⁶ One such malignancy to consider is lymphoma.⁷ Lymphomas are generally classified into categories (Hodgkin's or Non-Hodgkin's), by clinical behaviour (indolent or aggressive), and by cell origin (B-cell or T-cell/Natural killer-cell). Lymphomas may rarely have bone involvement, but when they do, they are classified into the following categories: solitary primary bone lymphoma, polyostotic primary bone lymphoma, and disseminated lymphoma with secondary involvement of the bone.⁸ NHL typically develops in the lymph nodes rather than in bone, but can occur in almost any tissue.⁴

Follicular lymphoma is a more indolent form of NHL that presents with painless enlarged peripheral lymph nodes.⁴ The majority of NHL cases are B-cell origin (approximately 85-90%), and the remaining either NK or T cells. The patient in this case had a rare presentation, as she was diagnosed with B-cell NHL of a follicular type, with polyostotic bony lesions. The most common risk factor for developing NHL is immunosuppression.⁴ A history of NHL or other lymphoid cancers in close relatives increases the risk of NHL by 2-3 times.⁹

Clinical presentation

The presentation of NHL varies depending on subtype,

site of involvement, and presence of additional symptoms such as fever, night sweats and weight loss.⁴ Possible symptoms of bone marrow involvement include: recurrent infections, pruritus, anemia, bruising and bleeding.⁹ NHL should be suspected if a patient presents with generalized lymphadenopathy that is painless and persistent.⁴ Patients may also present with signs of bone marrow involvement, and splenomegaly.⁴ Some case reports have documented unilateral limb swelling as an additional sign of NHL.^{10,11} This particular symptom was present intermittently in this patient's symptomatology, however was attributed to a race-related injury. In order to achieve the correct diagnosis and staging of the disease, patients will undergo an array of tests including blood work, biopsies of lymph nodes and bone marrow, imaging, and immunophenotyping.⁴

Concurrently, this patient had an anterosuperior labral tear of her left hip. Labral tears are a frequent cause of groin and anterior hip pain.¹² The literature has shown that patients with labral tears may go undiagnosed for an average of greater than two years, as patients are often seen by multiple providers before a definitive diagnosis is obtained.¹² Labral tears are generally insidious in nature, with a possible inciting event of microtrauma.¹² In a study investigating patients with arthroscopically confirmed labral tears, the authors found that 92% of the patients had predominant local groin pain, whereas 52% had associated anterior thigh pain.¹² However, in our study, we cannot say if this patient's symptomatology was related to her labral tear or underlying pathology from her NHL.

Management

The management for NHL varies depending on the grade and stage of the disease.⁴ In early stages, radiation therapy, surveillance, immunotherapy or chemotherapy may be considered as treatment options.⁴ In later stages, surveillance, immunotherapy, prophylactic antiviral medicine, or immunotherapy may be considered.⁴ The patient in this case was treated with rituximab and bendamustine. Rituximab is used in conjunction with chemotherapy medications (such as bendamustine) to for increased survival.⁴ Additionally, in order to promote function, psychological wellbeing, independence and quality of life, exercise rehabilitation strategies are typically incorporated in lymphoma patients' plan of management.¹³ The athlete in this case continued conservative management

after her chemotherapy with a physiotherapist, to work on her strength and endurance. Although this patient did not seek further management from the chiropractor following her diagnosis, chiropractors can play a role in the management of these patients by promoting exercise and psychological wellbeing strategies in an interdisciplinary collaborative care approach.¹³ The literature has shown that NHL survivors who meet exercise guidelines report clinically important better health-related quality of life measures compared to those that do not meet exercise guidelines.¹³ Thus, exercise can be recommended and is safe for patients with lymphoma, as there are benefits to their cardiorespiratory fitness, fatigue, and physical functioning.¹³ The patient in this case was dedicated to walking every single day throughout her NHL treatment and afterwards. Once she was able to, she began cycling again. With respect to chiropractic care when managing patients with cancer, it is imperative to mention that spinal manipulative therapy and spinal mobilizations are contraindicated in patients with known spinal malignancy according to the World Health Organization.¹⁴

Delayed diagnosis

In patients with follicular lymphoma, the relative survival rates range from 93.6% at one year, to 61.6% at ten years.³ Prompt diagnosis is important for a more favourable prognosis.¹⁵ A patient's diagnosis may be delayed when they visit a larger number of healthcare providers or "practitioner shop". This occurs in patients with persistent symptoms, and those that disagree or are in denial of a diagnosis or treatment.¹¹ This practitioner shopping may hinder a single provider's ability to ensure effective and efficient treatment due to the lack of continuity of care and possible opposing health care views.^{11,16} The patient in this case expressed that she chose not to listen to the chiropractor, as she wanted to keep training and wanted to avoid interruptions to her competition schedule. She then chose to see her physiotherapist and massage therapist for treatment and advice. This led to a worsening of her symptoms, and a delayed diagnosis. The patient delayed consultation with both her family doctor and sports medicine physician, against the advice of the chiropractor, to continue her athletic pursuits. She did not want to give up on any opportunity to train or compete.

Athletes are known to push themselves to their limits physically and mentally¹⁷ and have various intrinsic (i.e.

achievement) and extrinsic (i.e. sponsorship) factors that contribute to their desire to pursue their sport.² Although these strategies can lead to athletic success, a balance must also be prioritized to avoid overtraining and injuries.¹⁷ Some athletes may go against the advice of their medical professionals and refuse to abstain from sport participation. For many athletes their identity is tied intimately to their sport, and abstaining from sport potentially leads to the fear of losing a major part of their life.³ Athletes may also be in denial of their injuries for fear of losing their ability to train and compete.³ Chiropractors can help manage this fear with patient education and interdisciplinary collaboration with psychotherapists or mental performance coaches.

Limitations

The major limitation of this report is by nature the type of report being presented. Case studies are considered a lower level of evidence with respect to the hierarchy of evidence.¹⁸ Hence, information such as rates, incidence and generalizability cannot be generated or inferred.¹⁸ However, case reports demonstrate novel patient presentations, which may act as a reminder to practitioners to consider these serious, but rare pathologies. Additionally, it is not possible for us to know the true source of the patient's pain – whether the labral pathology, the underlying lymphoma, both, or neither accounted for her symptoms.

Summary

A 51-year old elite female cyclist had a history of long-standing hip pain with insidious onset. Once she finally agreed to further investigations, she was diagnosed with NHL. The desire to continue to compete and train through injuries and pain is common in athletes, although it can lead to delays in appropriate diagnosis and management. This case is evidence of this scenario; the patient repeatedly denied advice to rest, and to seek further investigations. She sought multiple providers to manage her symptomatology. For various reason (loss of identity, sponsorship, etc.), athletes may avoid the advice of medical professional since they do not want to stop training or competing. The persistence of her medical professionals and her symptoms led to the eventual diagnosis of NHL. The patient has had a complete response to chemotherapy and is returning to sport in a gradual manner with the advice of a coach and her medical professionals. Chiro-

practitioners may play a role in referring for appropriate diagnosis, education, and exercise prescriptions for patients with NHL.

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