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Case report

First case of chronic cell leukemia discovered incidentally in extra-saccular inguinal lymph node during laparoscopic bilateral inguinal hernia repair. Case report and literature review

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ABSTRACT

Introduction: Chronic cell leukemia discovered incidentally in extra-saccular inguinal lymph node during laparoscopic bilateral inguinal hernia repair is extremely rare.
Presentation of case: 62-year-old Romanian male presented at the outpatient general surgery clinic in April 2019 complaining of bilateral inguinal swelling that gradually increased in size mainly on right side and was diagnosed with bilateral inguinal hernia. During the laparoscopic repair of the hernia, a large lymph node in the left femoral canal was incidentally observed. Histopathologic, immunohistochemical, and flowcytometric evaluation of the excised specimen confirmed chronic lymphocytic leukemia/small lymphocytic lymphoma.
Discussion: Whole body CT showed supra and infra-diaphragmatic lymphadenopathy, and few small subsolid pulmonary nodules, possibly metastatic. Splenomegaly and pancreatomegaly were also noted, suggesting lymphomatoid infiltration.
Conclusion: There is need for cautious inspection and meticulous palpation of the inguinal area for any lymphadenopathy during routine inguinal hernia repair.

1. Background

Inguinal herniorrhaphy is one of the most commonly performed operations in the United States [1]. Nearly 20 million patients undergo inguinal hernia surgery every year worldwide [2]. Generally, hernia repair is curative therapy that improves the quality of life, relieves symptoms and prevents life-threatening complications [3].

Inguinal lymphadenopathy and malignancy discovered incidentally during hernia repair is rare. A study of 22,000 inguinal hernia repairs showed that 0.07% were found to have metastatic tumors after tissue pathology of the hernia sac, where 40% were of gastrointestinal origin, 20% ovary, 13% prostate, 13% mesothelioma and 13% from unknown origin [4]. In addition, the study identified only 2 cases of extra-saccular metastatic malignant lesions in the inguinal lymph node, and both were of prostate origin [4].

Chronic lymphocytic leukemia (CLL)/small lymphocytic lymphoma (SLL) is the most common leukemia among adults in the Western World [5]. CLL is a B-cell malignancy, follows an indolent course, and histologic transformation occurs in 2%–10% of patients to more aggressive lymphomas, such as diffuse large B-cell lymphoma or Hodgkin lymphoma [6,7]. The incidental finding of CLL/SLL in extra-saccular inguinal lymph node during inguinal hernia repair is extremely rare.

We present the first case of CLL/SLL diagnosed incidentally in extra-saccular inguinal lymph node during laparoscopic repair of bilateral inguinal hernia. We report this case in line with the updated consensus-based surgical case report (SCARE) guidelines [8]. In addition, we undertook a literature review of published cases of leukemia/lymphoma of extra-saccular lymph node discovered incidentally during inguinal or

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femoral hernia repair.

2. Case presentation

A 62-year-old Romanian male presented at the outpatient General Surgery clinic of our institution (Hamad General Hospital, largest tertiary facility in Doha, Qatar) in April 2019, complaining of bilateral inguinal swelling that gradually increased in size over time, mainly on the right side. There were no urinary symptoms, no fever, no weight loss, no change in bowel habits, and no other complaints. On physical examination, he had bilateral inguinal swellings that were reducible on both sides, with positive impulse on cough on both sides. His past medical history was significant for prostate cancer diagnosed 7 years back, for which the patient refused medical treatment, and was on herbal treatment. Past social, environmental, family and employment history were unremarkable. He did not smoke, never consumed alcohol and was not on long-term medications.

Laboratory work up showed WBC 8.9 $\times$ 10^3/μL, Hgb 11.8 g/dL, and platelets 102 $\times$ 10^3/μL. Total PSA was high at 39 ng/mL, whereas electrolytes, liver and renal function panels were within normal. The patient had a previous MRI of the pelvis and prostate that showed multiple enlarged lymph nodes in the bilateral common iliac region extending to the left para-aortic region. Regarding the prostate, there was right basal and mid gland peripheral zone corresponding to PI-RADS 5 with extra capsular extension, possible seminal vesicle invasion, and ill-defined smaller focal lesions in the left peripheral zone corresponding to PI-RADS 4. The patient was diagnosed with bilateral inguinal hernia based on clinical assessment and was scheduled for elective laparoscopic repair of bilateral inguinal hernia.

3. Surgical procedure

The patient underwent laparoscopic repair of bilateral inguinal hernia in October 2019. Intraoperatively, he was intubated (orotracheal) and placed in supine position, and under aseptic measures, a 10 mm supraumbilical port inserted by open technique, and another two 5 mm ports were inserted at the bilateral flank area. Inspection of the abdominal cavity using endo-camera showed a left side direct inguinal hernia (Fig. 1A), and a right side indirect inguinal hernia (Fig. 1B). The same steps of the procedure were undertaken on both sides of the inguinal hernia, where a senior consultant surgeon created a peritoneal flap starting 5 cm above the hernia canal at the level of the anterior superior iliac spine. The incision was then advanced to the medial side of the transverse plane through the upper 5 cm of the inguinal canal's inner ring and terminated at approximately 2 cm from the median ligament. A lower peritoneal flap was liberated until lateral visualization of the iliopubic tract and medial visualization of Cooper's ligament. The hernia sac was carefully dissected and the structures that were attached through the lower peritoneal membrane and vas deferens were seen and preserved.

While separating the structures in the left inguinal area, the surgeon incidentally observed a large lymph node in the femoral canal (2 $\times$ 3 cm, Fig. 2A and B). Excisional biopsy was undertaken for this large lymph node and it was sent to pathology. Then, a 15 $\times$ 13 cm ULTRA PRO advance mesh was inserted and fixed with tuckers. The peritoneal flap was closed with vicryl lock 3/0 suture, homeostasis was secured, ports were closed with J needle vicryl 3/0 suture, skin closed with monocryl 3/0 suture, and dressing was applied. The patient was smoothly extubated in the operating room and sent to the recovery suite in a stable condition.

4. Pathology

Histopathology examination showed that the lymph node was totally effaced by a predominantly diffuse lymphoid proliferation with scattered vague nodules. There were no definite residual germinal centers seen. Most of the cells were small lymphocytes with small nuclei, clumped chromatin and inconspicuous nucleoli. In the ill-defined nodules and the pseudo-follicular proliferation centers, there were larger lymphoid cells with prolymphocytes (Fig. 3A and B).

The small proliferating lymphocytes were B lymphocytes positive for CD45, CD20, BCL-2, CD5 and CD23, but negative for CD10, BCL6, Cyclin-D1 and SOX11. Ki-67 stained 25–30% of the lymphoid cellular nuclei (Fig. 4). Flowcytometry of the same lymph node showed monotypic B-cell population (approximately 55%) with cytoplasmic lambda light chain restriction and immunophenotypic profile of CLL/SLL with atypical partial down-regulation of CD23. Interphase fluorescence in situ hybridization (FISH) was also undertaken and was normal.

5. Follow-up

The postoperative course was unremarkable, and the patient was discharged on post-operative day 1. Regarding the post-operative follow up with the surgical team, the patient was seen 2 weeks later at the General Surgery outpatient clinic, and found to have a mild hematoma at the right hemi scrotum. Otherwise, the surgical wounds were healing well, with no swelling or discharge. The patient was reassured and urgently referred to the hematology clinic. As per request of the hematology team, a whole-body CT was done that showed supra and infradiaphragmatic lymphadenopathy, and few small subsolid pulmonary nodules, possibly metastatic (Fig. 5A, B and C). Splenomegaly and pancreatomegaly were also noted, suggesting lymphomatoid infiltration

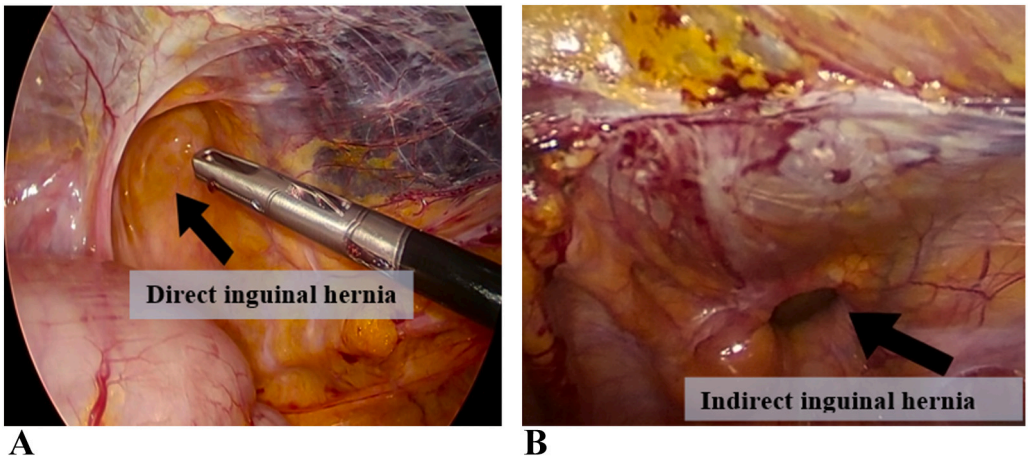

Fig. 1. Intraoperative findings showing: A) Left side direct inguinal hernia; and, B) right side indirect inguinal hernia.

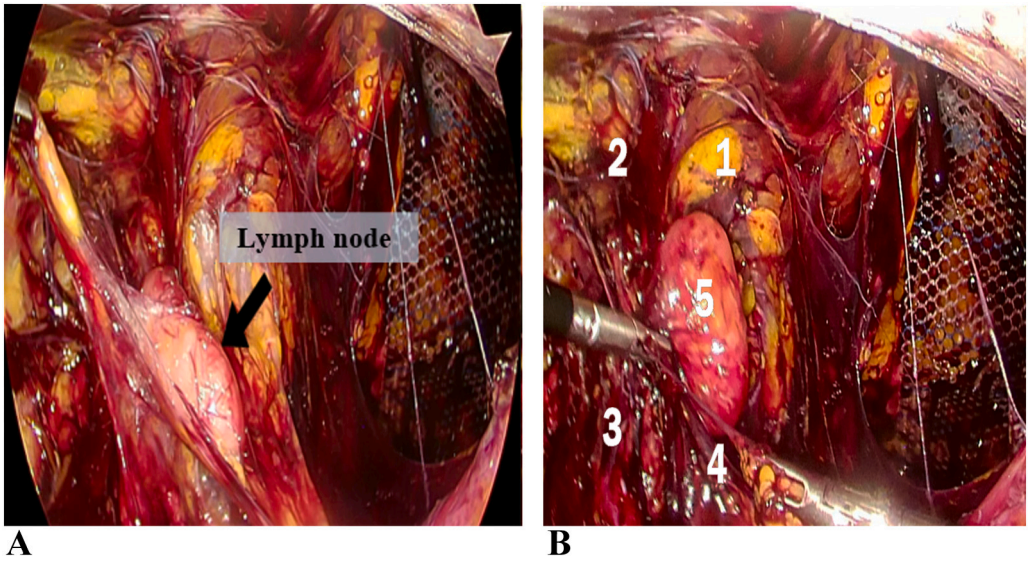

Fig. 2. Intraoperative findings showing: A) Left side direct inguinal hernia with lymph node lying in femoral canal measuring 2 × 3 cm; and, B) left side direct inguinal showing 1-Femoral ring, 2-Inferior epigastric vessel, 3-Testicular vessels, 4-Vas deferens, and, 5-Enlarged lymph node.

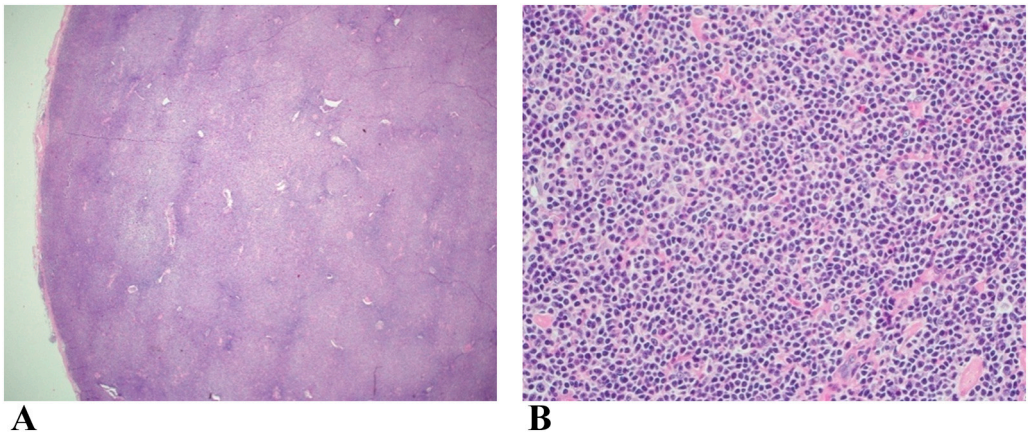

Fig. 3. Histopathology examination indicating chronic lymphocytic leukemia/small lymphocytic lymphoma and showing: A) lymph node expanded by proliferating small lymphocytes in vague nodular pattern; and, B) higher magnification of small proliferating lymphocytes (hematoxylin-eosin, original magnifications ×20 [A] and ×400 [B]).

(Fig. 6A and B). The patient was then discussed at the hemato-oncology multidisciplinary team (MDT) meeting and diagnosed as CLL/SLL, Binet stage 1B, Rai stage 2 with intermediate risk stratification and recommended for follow up with the hemato-oncology team. Subsequent to this, the patient was lost to follow up as he had travelled out of the country.

6. Discussion

CLL/SLL are the same disease entity in the 2016 revision of the World Health Organization (WHO) classification of lymphoid neoplasms [9]. Staging of CLL is mainly by clinical staging systems and treatment is according to staging [10,11].

In terms of demography, the average age of CLL/SLL patients is 72 years and it is more common in males [12]. Our patient was a slightly younger (62 years) male, in agreement with literature review we undertook (Table 1) that confirmed the male predominance and the age span ranged from 23 to 74 years.

As for clinical presentation, SLL classically presents with lymphadenopathy, hepatosplenomegaly, and/ or extra-nodal invasion; CLL presents traditionally with the incidental finding of an absolute number of monoclonal B lymphocytes $< 5 \times 10^9$/L in the peripheral blood [13]. About 15%–43% of patients present with β-symptoms (fever, night sweats, weight loss) at the time of diagnosis [14]. Our patient had an atypical presentation as his main complaint was bilateral inguinal swelling, otherwise he was completely asymptomatic.

The literature review (Table 1) suggested that our CLL/SLL case is extremely rare. To the best of our knowledge, this patient could be the first ever published case of chronic lymphocytic leukemia (small lymphocytic lymphoma) diagnosed in an extra-saccular inguinal lymph node incidentally during a laparoscopic repair of bilateral inguinal hernia. This highlights a deliberation in the literature as to whether all excised surgical specimens should be examined microscopically. Others reported that 1 in 1020 adult hernia sacs contained an unexpected abnormality and advised that such finding was too rare to recommend the routine examination of all specimens [15]. Conversely, some authors advocated that all hernia sacs should be sent for histopathological examination to exclude malignancy [16]. We sent our specimen for examination that led to the diagnosis of CLL/SLL.

As for investigations and imaging, the patient had a preoperative workup that showed a mild low platelet count in an otherwise normal complete blood picture, electrolytes, liver and renal function panels.

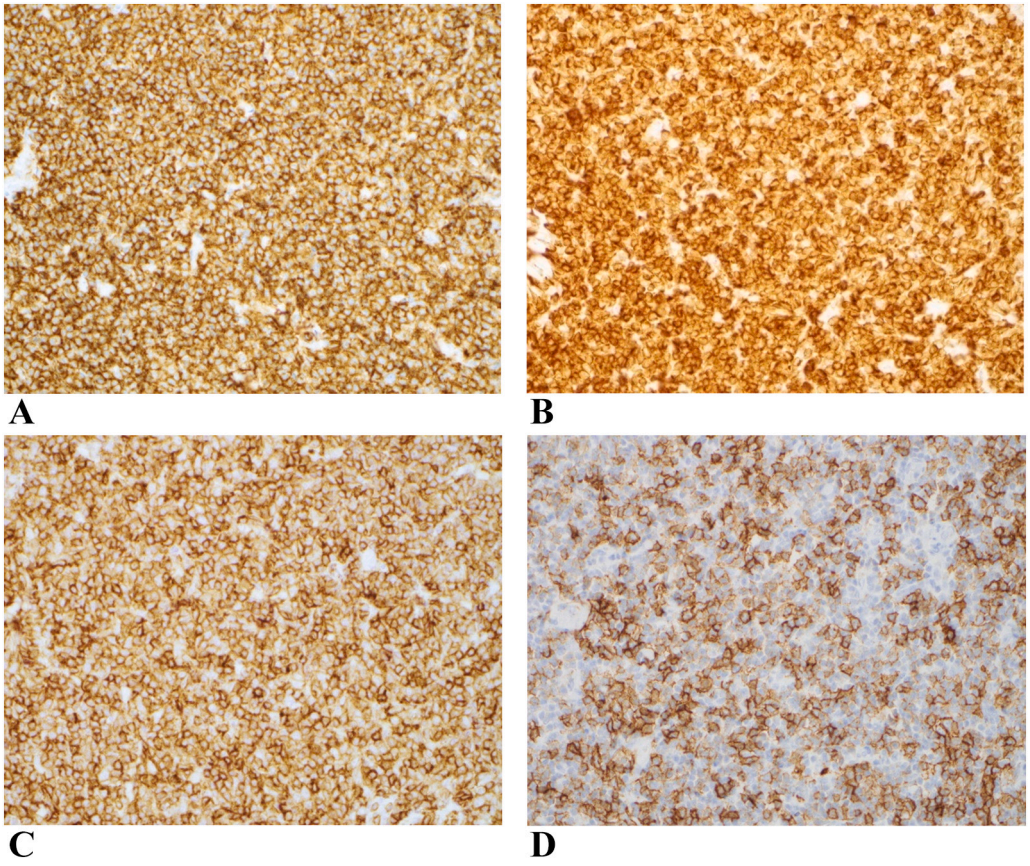

Fig. 4. Histopathology examination indicating chronic lymphocytic leukemia/small lymphocytic lymphoma and showing: A) lymphocytes positive for CD20; B) lymphocytes positive for BCL-2; C) lymphocytes positive for CD5; and, D) lymphocytes positive for CD23 (Immunohistochemical stains, original magnification ×400 [A, B, C and D]).

Previous MRI of the pelvis and prostate as workup for his prostate cancer showed multiple enlarged lymph nodes in the bilateral common iliac region extending to left para-aortic region. The same MRI of the prostate showed a right basal and mid gland peripheral zone corresponding to PI-RADS 5 with extra capsular extension, possible seminal vesicle invasion, and ill-defined smaller focal lesions in the left peripheral zone corresponding to PI-RADS 4.

Regarding the diagnosis, CLL/SLL requires histopathologic confirmation mainly by lymph node biopsy. Confirmation from an extra-saccular inguinal lymph node during laparoscopic inguinal hernia repair is extremely rare and only 2 other reports published in 1990 and 1982 discussed it (Table 1) [16,17]. Histologically, the lymph node shows diffuse effacement of its architecture with pale areas formed in a nodular pattern and darker areas composed of sheets of small lymphocytes with low mitotic activity [18]. In flowcytometry, CLL/SLL have an immunophenotypic pattern characterized by weak expression of surface immunoglobulin M and expression of CD5, CD23, CD19, CD79a, CD43, CD11c (weak), and CD200 [19]. Such findings agree with our case where the histopathology showed that the lymph node was totally effaced by a predominantly diffuse lymphoid proliferation with scattered vague nodules; and the flowcytometry depicted a monotypic B-cell population (approximately 55%) with cytoplasmic lambda light chain restriction and immunophenotypic profile of CLL/SLL with atypical partial down-regulation of CD23.

In terms of staging of CLL/SLL, a whole-body CT scan or better a whole-body PET/CT scan with contrast is required [20]. We agree, as for our case, whole body CT imaging showed supra and infra-diaphragmatic lymphadenopathy with splenomegaly and pancreatomegaly, and the patient was staged as Binet stage 1B, Rai stage 2 with intermediate risk stratification.

Regarding management, asymptomatic patients with early-stage disease can be followed up without any medical treatment except where there is evidence of rapid disease progression [21]. Reports suggest that there is no advantage of early initiation of treatment for the early stages of the disease [21]. The management of our case concurs with this view, as our hemato-oncology MDT recommended regular follow up of the patient by the hemato-oncology team.

7. Conclusion

We present the first published case of CLL/SLL diagnosed in an extra-saccular inguinal lymph node incidentally during laparoscopic repair of bilateral inguinal hernia. This extremely rare presentation of CLL/SLL suggests the need for cautious inspection and meticulous palpation of the inguinal area for any lymphadenopathy during routine inguinal hernia repair. Gently isolating the spermatic cord and vas deferens from the surrounding structures enhances the inspection and palpation of the surrounding tissues, in order to prevent missing any lymphadenopathy in the hernial sac or extra-saccular area. Where lymphadenopathy is encountered, excisional biopsy and full histopathologic, immunohistochemical, and flowcytometric evaluation of the specimen is critical. If CLL/SLL is confirmed, then appropriate full body imaging is warranted to stage the disease and detect its extent in order to guide management.

Consent

Due to the COVID-19 pandemic, written informed consent was not possible as it was deemed unethical that the patient travels to the hospital to sign the consent. Hence, informed verbal consent was obtained over the telephone from the patient after a through explanation of the

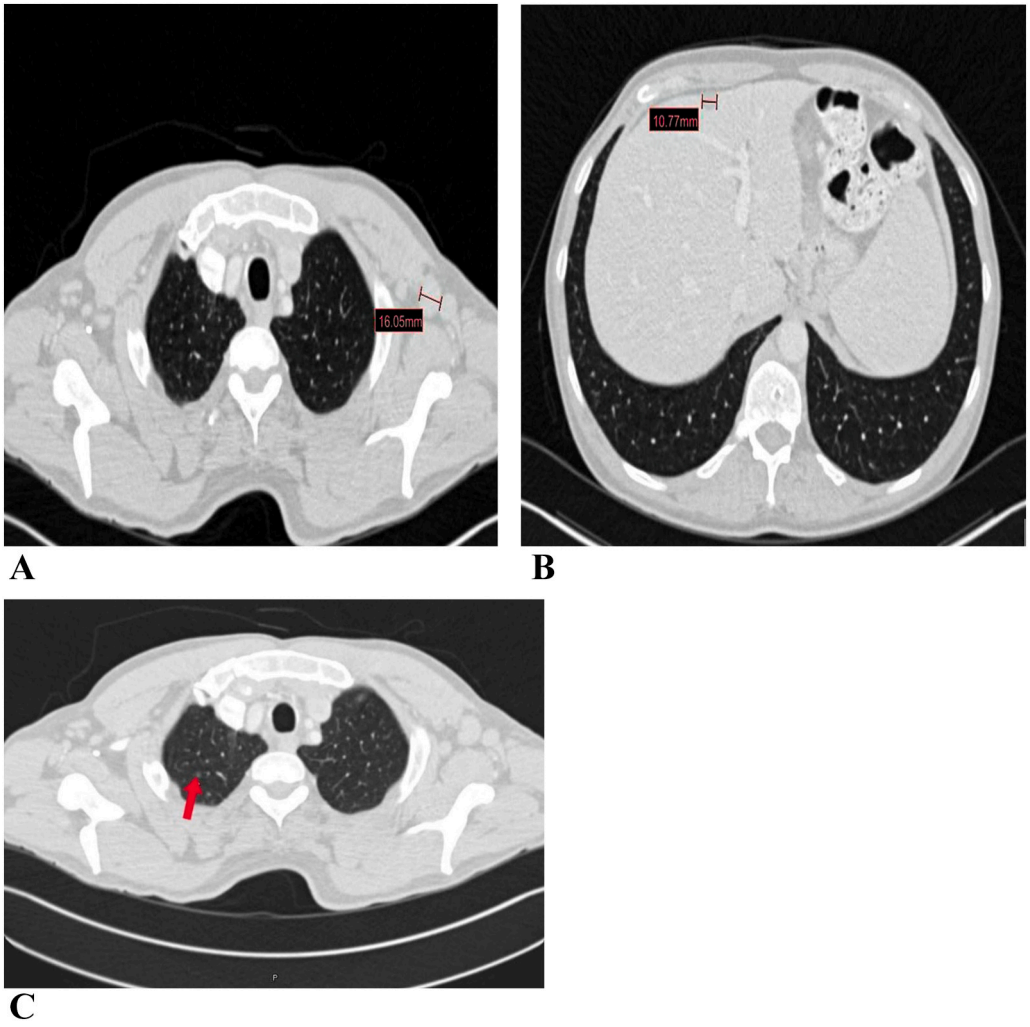

Fig. 5. Whole body CT (axial) with oral and IV contrast showing: A) supra-diaphragmatic lymphadenopathy; B) infra-diaphragmatic lymphadenopathy; and, C) few small subsolid pulmonary nodules.

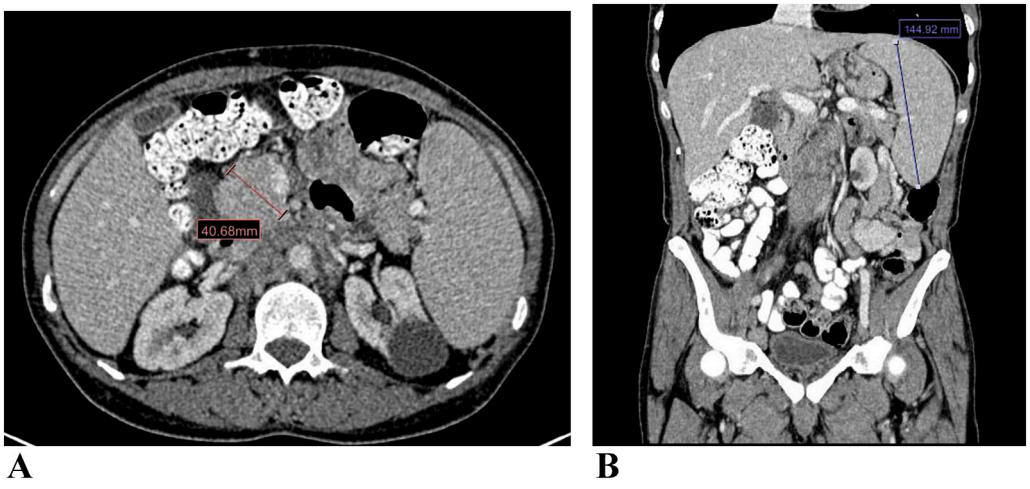

Fig. 6. Whole body CT showing: A) pancreatomegaly (axial view); and, B) splenomegaly (coronal view) suggesting lymphomatoid infiltration.

fact that his case will be published in a scientific journal without breaking his confidentiality or disclosing his identity and he happily agreed to do so; the discussion was witnessed by a co-author.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Table 1
Literature review: cases of leukemia/lymphoma of extra-saccular lymph node discovered incidentally during inguinal or femoral hernia repair.

Case	Age	Sex	Hernia site	Site of pathology	Surgical approach	Gross description	Diagnosis
Current case Qatar	62	M	Inguinal	Femoral canal	L	2 × 3 cm mass	Chronic lymphocytic leukemia/small lymphocytic lymphoma
Connelly [16]	50	M	Inguinal	—	O	—	Diffuse large cell lymphoma
1990	51	M	Inguinal	—	O	—	Follicular mixed cell lymphoma
USA	60	M	Inguinal	—	O	—	Follicular small cleaved cell lymphoma
	23	M	Inguinal	—	O	—	Lymphocytic predominance Hodgkin's disease, nodular L/H
	74	F	Unknown (inguinal or femoral)	—	O	—	Follicular mixed cell lymphoma
	40	F	Unknown (inguinal or femoral)	—	O	—	Follicular small cleaved cell lymphoma
	76	F	Unknown (inguinal or femoral)	—	O	—	Sclerosing diffuse large cell lymphoma
	58	M	Femoral	—	O	Lymph node	Diffuse large cell lymphoma
Geuna [17]	46	M	Inguinal	Spermatic cord	O	2 × 2 cm mass	Lymphosarcoma (nodular, mixed histiocytic, lymphocytic lymphoma)
1982 USA	58	M	Femoral	Femoral canal, internal opening	O	2 × 3 cm mass	Lymphosarcoma (nodular well differentiated)

L: laparoscopic; O: open; —: not reported.

Ethical approval

Approved by Medical Research Center, Hamad Medical Corporation reference number (MRC 04–21-751).

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Guarantor

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CRediT authorship contribution statement

Hamzah El Baba: data collection, data interpretation, writing the paper, review & editing. Ahmed Al Moudaris: review & editing. Hayan Abo Samra: data interpretation, review & editing. Layth Alateeg: review & editing. Walid El Ansari: study concept, data interpretation, writing the paper, review & editing. Mohammed Al-Yaseen review & editing. All authors read and approved the final version.

Declaration of competing interest

The authors declare that there is no conflict of interest that could be perceived as prejudicing the impartiality of the research reported.

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