Lymphoma Diagnosed at Inguinal Hernia Repair

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Abstract

Tumors presenting in the inguinal hernia sac are considered to be extremely rare, with the more common neoplasms metastasizing from the gastrointestinal tract, ovary and prostate. We report the case of Mantle cell lymphoma identified in the inguinal hernia sac following hernia repair. While the hernia sac appeared normal to the surgeon, evaluation by the pathologist showed subtle gross irregularities, with subsequent histologic and immunochemical diagnosis of Mantle cell lymphoma. Twelve previous cases of a lymphoma diagnosed during hernia repair have been described in the English literature. This is the first report of Mantle cell lymphoma found in the hernia sac. This case illustrates the value of routine microscopic evaluation of hernia sacs found from inguinal/femoral herniorrhaphies, as it may be the primary presentation of an asymptomatic metastatic lymphoma. Additionally, it underscores the importance of the surgeon's role in screening hernia sacs if the practice of submitting only macroscopically abnormal specimens for microscopic evaluation is adopted.

Introduction

Mantle cell lymphoma accounts for 5-10% of all lymphomas, with a median age of 65 years at diagnosis. At the time of diagnosis, the lymphoma is typically found diffusely throughout the lymphoid tissue, and may be found in other tissues including the intestinal tract, skin and breast. While this type of lymphoma is responsive to chemotherapy, the nature of the disease tends to be aggressive, with a median survival of 3 years.[1] We present the case of a Mantle cell lymphoma found upon inguinal hernia repair, in which routine histologic evaluation of the hernia sac affected this patient's medical management. While the pathologist did note irregularity of the hernia sac, no macroscopic abnormality was observed intraoperatively. For this patient, routine histologic evaluation of the inguinal hernia sac allowed for the diagnosis and treatment of an aggressive lymphoma that may have otherwise remained undiagnosed.

Case Report

A previously healthy 55-year-old Chinese male presented with right groin pain and progressive swelling in the inguinal region of several months duration, with no prior history of hernia, heavy lifting or trauma. He reported no other symptoms including fatigue, night sweats or weight loss. Past medical history was notable only for a 15 pack-year history of smoking and hyperlipidemia for which he was taking atorvastatin. Past surgical history was positive only for wrist surgery. Family history was negative for any malignancy. Vital signs were within normal limits with a BMI of 25.2 and physical examination was notable only for a right-sided reducible inguinal hernia.

An inguinal hernia repair operation was subsequently scheduled, and was performed successfully using a polypropylene mesh plug, with no complications. The surgeon did not note any abnormalities of the hernia sac at the time of operation. The patient went home on the same day and recovered uneventfully.

The hernia sac, measuring 3.8 x 2.3 x 0.8 cm, was grossly described as an irregular portion of membranous, pink-tan tissue. Figure 1a demonstrates a low power hematoxylin/eosin (H&E) view of hernia sac connective tissue with massive lymphocytic invasion. Demonstrated here is the junction between the tumor/lymphoma and the underlying normal tissue it has invaded. Figure 1b depicts a closer view with H&E stain of the lymphocytes (a monotonous sheet of small round blue cells with scant cytoplasm), with a thick walled blood vessel in the upper right. Figure 1c shows positive immunostaining for CD 20 staining (brown) in a background stain of blue. Immunostaining was also positive for BCL-2 and cyclin D1. CD5 was weakly positive. Immunostains were negative for CD3, CD4, CD8, CD10, CD23 and BCL-6. These findings are consistent with a diagnosis of Mantle cell lymphoma.

Subsequent CT of the abdomen and pelvis with contrast demonstrated a 2.8 cm enhancing liver lesion at the dome of the right lobe, suspicious for tumor. There were also several smaller nonspecific nodules scattered throughout the liver. Retroperitoneal adenopathy and adenopathy around the celiac axis were noted. Specifically, there were several lymph nodes measuring up to 2.3 cm surrounding the celiac artery and a 5.1 cm ill-defined mass surrounding the infrarenal inferior vena cava.

CT scan of the chest showed multiple hilar, mediastinal and cardiophrenic nodes, as well as a 4.9 cm mediastinal mass. Positron-Emission Tomogram (PET) scan demonstrated multiple active sites of lymphoma from the neck to the inguinal region--including the supraclavicular areas, neck, mediastinum, stomach and retroperitoneum. The patient was started on a chemotherapeutic regimen of doxorubicin, cyclophosphamide, rituxamab and vincristine.

Discussion

According to the National Center for Health Statistics, over 700,000 inguinal hernia repairs are performed annually in the United States.[2] The vast majority of inguinal and femoral hernia sacs from these repairs are unremarkable upon routine histologic evaluation. In a study of over 22,000 inguinal hernia repairs at the Mayo Clinic, 0.07% were found to have metastatic tumors, with colon cancer being the most commonly implicated tumor. Forty percent were of gastrointestinal origin, 20% ovary, 13% prostate, 13% mesothelioma and 13% from unknown origin. The most common presenting symptoms were an inguinal mass and abdominal or groin pain.[3]

Overall, less than 0.5% of hernia sacs contain primary or metastatic tumors.[4] Kassan et al, in evaluating 1,020 inguinal and femoral hernias, questioned the cost-effectiveness of sampling macroscopically normal hernia sacs, reporting that only 3 specimens (0.098%) showed abnormal pathology while appearing normal at the time of operation. In a review of the literature, these authors also concluded that in the rare case of a malignant tumor, 73.3% were identified macroscopically.[5] Nicholson et al, in evaluating patients from 1950-1988, arrived at a similar conclusion that macroscopically normal hernia sacs did not warrant histologic evaluation.[3] Matthyssens analyzed routine specimens in general surgical procedures — specifically hemorrhoidectomies, cholecystectomies, appendectomies and inguinal hernia repairs between 1993 and 2002. In these cases, 1%

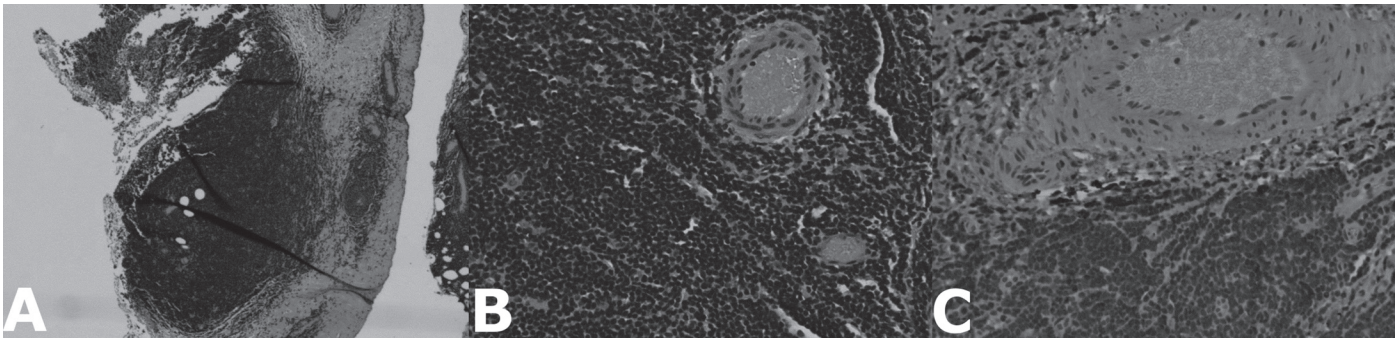

Figure 1.— Inguinal hernia sac with H&E staining under low power (a), high power (b), and CD20 immunostaining (c).

Table 1.— Lymphomas Mimicking or Presenting with Femoral or Inguinal Hernias

Author/Yr	Age/Sex	Hernia	Location	Gross Description	Final diagnosis
	55/M	Inguinal	Hernia sac	Irregular membranous, pink-tan tissue	Mantle cell lymphoma
Geuna/1982	46/M	Inguinal	Spermatic cord	2x2 cm mass	Lymphosarcoma (nodular, mixed histiocytic, lymphocytic lymphoma)
	58/M	Femoral	Femoral canal, internal opening	2x3cm mass	Lymphosarcoma (nodular well differentiated)
Kassan/1986	?	Femoral	?	Lymph node, grossly abnormal	non-Hodgkin's lymphoma
Connelly/1990	67/F	Femoral	?	Lymph nodes of "focal flesh-like areas"	Diffuse large cell lymphoma
	50/M	Inguinal	?	?†	Diffuse large cell lymphoma
	51/M	Inguinal	?	?†	Follicular mixed cell lymphoma
	60/M	Inguinal	?	?†	Follicular small cleaved cell lymphoma
	23/M	Inguinal	?	?†	Lymphocytic predominance Hodgkin's disease, nodular L/H
	40/F	*	?	?†	Follicular small cleaved cell lymphoma
	74/F	*	?	?†	Follicular mixed cell lymphoma
	76/F	*	?	?†	Sclerosing diffuse large cell lymphoma
Moller/1994	48/M	Inguinal	Spermatic cord	Nodular 3-cm tumor	High grade B cell lymphoma polymorphic centroblastic type

*Diagnosed initially as having an inguinal or femoral hernia; †Lymph nodes collectively described as light tan and homogenous, ranging 1.0 -7.0 cm

of hemorrhoidectomies, 0.4% of cholecystectomies and 0.1% of appendectomy specimens had unexpected malignant/premalignant findings on histologic evaluation but each of these had macroscopic findings suggestive of these abnormalities.[6] None of the 2000 hernia sac specimens had any gross or histologic abnormalities. They suggested that routine histologic examination in the absence of any gross abnormalities could be omitted. The US College of American Pathologists' statement in 1996 also recommended selective surgical specimen examination—reflecting on current trends of cost containment via practice guidelines, as well as the aim for more responsible and evidenced-based use of diagnostic testing.[7]

Older literature has advocated histologic evaluation of all hernia sacs, with many authors supporting routine examination to avoid overlooking an occult malignancy.[8-10] Roslyn et al, in 1200 inguinal and femoral hernorraphies from 1972-1978 noted that tumors of the hernia sac were often not diagnosed until pathologic evaluation, and argued for the need to microscopically examine all hernia sacs.[10] There is also evidence that a higher index of suspicion may be necessary in the context of enlarged lymph nodes found upon hernia sac evaluation. Connelly et al reviewed twelve patients with enlarged lymph nodes associated or presenting as inguinal or femoral hernias, and reported 7 of these patients as having non-Hodgkin's lymphoma and 1 with Hodgkin's lymphoma.[11] The authors cited special processing requirements and potential of misdiagnosis as reasons for having a higher index of suspicion for lymphoreticular disease when evaluating enlarged lymph nodes during hernia repair. Finally, Guena et al reported 2 cases of lymphosarcoma found upon hernia repair operation, and noted that the extreme variability in presentation of lymphosarcoma called for sampling of lymph nodes found during the course of a hernia operation.[12]

Twelve cases of lymphoma diagnosed at inguinal/femoral hernia repair have been described previously. Mean age of these patients was 54 years (range 23-76) with male predominance. The lymphomas presented as a unilateral groin mass, with most patients presenting asymptomatically. A summary including the present case is provided in Table 1. Primary malignant spermatic cord tumors often present as inguinoscrotal hernias, with primary spermatic lymphomas usually presenting either as a tumor in the groin or upper part of the scrotum.[13] In a review of 11 cases of spermatic cord lymphomas, Moller reports that three cases were initially misdiagnosed as hernias.[14] Our literature review includes primary spermatic cord lymphomas that presented as inguinal or femoral hernias and were discovered upon herniorrhaphy. It does not include spermatic cord tumors presenting as scrotal masses.

In summary, we report the case of Mantle cell lymphoma discovered incidentally at inguinal hernia repair. Literature review

indicates that occult malignancies diagnosed from routine histologic evaluation of inguinal and femoral hernia sacs is a rare occurrence and gross examination of the specimen would likely have identified theses malignancies. However, this patient's hernia sac was characterized by very subtle irregularities that were only detected upon histologic evaluation. This case supports routine histologic evaluation of hernia sacs. However, in cases where only selective microscopic evaluations are performed, the surgeon should meticulously inspect all hernia sacs and submit specimens with even subtle irregularities. As lymphoreticular disease can present variably and includes a broad differential of benign and malignant processes, the specimen should be submitted to pathology if any suspicious lymph nodes are encountered or if lymphoreticular pathology is already suspected. Finally, if routine histologic evaluation of all inguinal and femoral hernias is not feasible due to cost concerns a limited microscopic evaluation by pathology on grossly normal appearing sacs would reduce cost without compromising the identification of occult malignancy.

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