ISSN: 1584-9341

JOURNAL OF SURGERY

Jurnalul de Chirurgie

Volume 11, Issue 2

ISSN: 1584 - 9341

DOI: 10.7438/1584-9341

Journal of Surgery [Jurnalul de Chirurgie]

ISSN: 1584-9341

TABLE OF CONTENT

Journal of Surgery [Jurnalul de Chirurgie]
ISSN: 1584-9341

Journal of Surgery
[Jurnalul de Chirurgie]
Review Article
Open Access

Abdominal Complications of Typhoid Fever

Ketan Vagholkar*, Jimmy Mirani, Urvashi Jain, Madhavan Iyengar and Rahul Kumar Chavan
Department of Surgery, D Y Patil University School of Medicine, Navi Mumbai, India

Abstract

The natural history of typhoid fever poses both a diagnostic and a therapeutic challenge. Awareness of the clinical features of the primary presentation and of the complications are pivotal to early diagnosis. Typically, aggressive supportive care is all that is needed. However abdominal complications do occur and proper surgical care is required to lower morbidity and mortality.

Keywords: Typhoid fever; Abdominal complications; Treatment

Introduction

Typhoid fever is one of the most common enteric infections in the developing world. The infection starts with a febrile episode and if untreated eventually involves almost every system of the body with abdominal complications developing first. The initial treatment is predominantly medical and supportive in nature. A majority of the abdominal complications are surgical and early consultation with a surgeon should be considered. A high index of suspicion is essential. Therefore the attending clinician needs to be aware of the entire spectrum of clinical manifestations in order to avoid delay in diagnosis of complications. The paper discusses the clinical features, surgical complications and their management.

Clinical Evaluation

Typhoid fever is caused by *Salmonella Typhi* which is a gram negative non spore forming facultative anaerobic bacillus. The infective dose is usually 10^3 to 10^6 colony forming units. The mode of transmission is fecal - oral. The clinician needs to be aware of certain predisposing factors which render the patient more at risk for infection. Decreased stomach acidity by virtue of; young age (<1 year), antacid ingestion or achlorhydria especially associated with *H. pylori* infection is a major risk factor. A disruption in intestinal integrity either due to alteration of the intestinal flora usually caused by antibiotics or due to prior gastrointestinal surgery is also a risk factor. A detailed history is vital to diagnosing typhoid fever, instead of other common diseases such as malaria. Inadequacy of treatment can lead to the disease progression eventually manifesting with life threatening surgical complications that will require surgery. Abdominal complications are the most common. As these complications simulate many other abdominal conditions, a good history can alone differentiate the aetiology as typhoid fever and not something else. Fever is present in more than 75% cases followed by abdominal pain in 30–45% of cases. The incubation period varies from 3–21 days depending on the immunological status of the patients. Additional symptoms include headache, chills, cough, severe sweating, myalgia, malaise and diarrhoea. Physical examination reveals coated tongue, hepatosplenomegaly, a rash seen on the chest and abdomen described as rose spots, abdominal tenderness especially supra-umbilical.

Abdominal complications aren't the only issue with typhoid. 2–40% of patients may even exhibit neurologic manifestations such as meningitis, Guillain Barre syndrome, neuritis and various neuropsychiatric syndromes such as muttering delirium or coma vigil accompanied with the picking of the bed clothes [1,2].

Awareness of these clinical features can significantly help the attending clinician to arrive at a definitive diagnosis for the etiology of the fever as well as the abdominal complications.

Abdominal Complications

Small intestine

The organisms have a predilection for sites rich in cells belonging to the reticuloendothelial system [3]. This is typically seen in the Peyer's patches of the small intestine. There is significant hyperplasia followed by ulceration and necrosis of the Peyer's patches. This leads to significant bleeding eventually terminating into a perforation. This complication usually takes place in the third or fourth week of the disease. Since the terminal ileum is abound with Peyer's patches, multiple perforations in the ileum are common leading to high morbidity and a high mortality [4,5].

Patients invariably give a history of melena preceding the onset of abdominal pain. Abdominal signs are usually present eventually leading to board like rigidity of the abdomen. Due to severe peritonitis, the patient has a drastic fall in the urine output eventually passing into a state of septic shock. The time at which the condition is diagnosed and the rapidity with which surgical intervention is offered determine the outcome in such patients [5,6].

Rigorous and aggressive resuscitation is essential to ensure improvement in the urine output. Nasogastric aspiration and supportive intra venous fluids are usually accompaniments of the initial resuscitation. An abdominal x-ray in majority of times may be inconclusive. An ultrasound of the abdomen may reveal free fluid in the peritoneal cavity. A decision to perform exploratory laparotomy needs to be made as soon as the patient regains hemodynamic stability with good urine output [6].

The choice of surgical repair to be performed at laparotomy is the biggest dilemma to the surgeon [7]. An overall appraisal of the patient needs to be considered before taking a decision. For an isolated perforation, simple suturing may suffice. But if the size of

***Corresponding author:** Ketan Vagholkar, Professor, Department of Surgery, D Y Patil University School of Medicine, Annapurna Niwas, 229 Ghantali Road, Thane 400602, Maharashtra, India, Tel: +91 9821341290; E-mail: kvagholkar@yahoo.com

Received January 22, 2015; **Accepted** April 14, 2015; **Published** April 19, 2015

Citation: Vagholkar K, Mirani J, Jain U, Iyengar M, Chavan RK .Abdominal Complications of Typhoid Fever. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(1): 359-361 DOI:10.7438/1584-9341-11-2-1

the perforation is big, surrounding bowel edematous and the volume of peritoneal contamination significant then the chances of leakage of the sutured perforation are very high [4,6,7]. Leakage of the sutured perforation is associated with extremely high mortality. Therefore, in such a situation it would be a safe practice to exteriorize the ileum proximal to the sutured perforation [8]. This will significantly help in achieving control over the septic process especially in moribund patients.

If perforations are multiple, then resection of the entire segment is the method of choice. A primary anastomosis with proximal ileostomy is a safe choice as it prevents the chances of anastomotic dehiscence. The proximal ileostomy can be closed 12 weeks later [7,9].

Exteriorization is one of the safest and best options for typhoid enteric perforations. It reduces the chances of leakage thereby leading to re-laparotomy to a bare minimum. Majority of patients presenting with enteric perforation are severely moribund and nutritionally depleted at the time of surgery. Therefore, in a situation where the process of healing cannot be relied upon, exteriorization is the best lifesaving option for the patient.

Gall bladder

The gall bladder is also affected in typhoid fever developing acalculous cholecystitis [10,11]. Concurrent gall stones may worsen the problems predisposing to perforation of the gall bladder [10]. Various hypotheses have been put forward to explain the pathophysiology of acalculous cholecystitis in typhoid fever [11]. However the exact mechanism still remains uncertain. Endotoxin mediated injury seen in gram negative sepsis is one of the most proposed hypotheses. These mediators lead to biliary stasis which results in increased bile viscosity, sludge formation and mucocele of the gall bladder. This in turn results in functional or secondary mechanical obstruction of the cystic duct due to inflammation. The other hypothesis proposes increased abnormal permeability of the serous membranes along with capillary leakage as a result of direct invasion of the gall bladder wall by typhoid bacilli, thus leading to thickening of gall bladder wall and distension. In a select 10–15% of patients, if the cystic duct obstruction persists, the inflammatory process may progress to gangrene and perforation [10]. The commonest site is the fundus of the gall bladder. The treatment of acalculous cholecystitis is conservative. However, if perforation occurs, cholecystectomy has to be performed. Cases of concomitant ileal and gall bladder perforations have been reported [12]. This significantly adds to the mortality in such patients. These patients require early detection and prompt surgical intervention. Radiological investigations such as ultrasound and computed tomography have low specificity in detecting these complications. Hence, a high index of clinical suspicion can only help in early detection of such complications. The role of laparoscopy in typhoid enteric perforation is extremely limited. Extensive fluid collections, adhesions and distended bowel loops limit significantly the dexterity of laparoscopic instrumentation. Hence, laparoscopy is to be avoided in such situations [6].

Liver

Typhoid fever commonly results in a significant increase in liver size [13]. Enlargement of the liver leads to dysfunction of the hepatocytes which may be due to hyperplasia and hypertrophy of the reticulo-endothelial cells accompanied by hepatocyte damage induced by anti-pyretic medications [14,15]. The net result is cholestasis. There may be increase in levels of bilirubin, ALT, AST and GGT. PT/PTT may not be seriously altered. Treatment is predominantly supportive with administration of antibiotics such as quinolones which are the drug of choice in such cases [13,15].

Spleen

The spleen is a very important reticulo-endothelial organ and is typically greatly enlarged in typhoid fever [16]. Massive enlargement of the spleen causes increased stretching of the splenic capsule predisposing to either spontaneous rupture or increased susceptibility to rupture following minor trauma [17]. Left upper quadrant pain accompanied with severe pallor and shock should raise the suspicion of a ruptured spleen. Treatment is immediate laparotomy with splenectomy [17].

Pancreas

Pancreas is another organ which is affected in typhoid. Development of pancreatitis pre disposes to pancreatic abscess formation. The root of infection in pancreatic abscess may perhaps be due to infected bile reaching the pancreas by the pancreatic duct, hematogenous or lymphatic spread from the intestinal tract [18]. Treatment is usually conservative with excellent resolution with antibiotics and supportive care [19].

Conclusion

Typhoid continues to be febrile disease with significant surgical complications.

Awareness of the clinical features of this disease is pivotal in prompt diagnosis of the disease as well as its complications. A high degree of awareness and clinical suspicion is essential for early diagnosis of abdominal complications.

Prompt and aggressive treatment can only reduce the morbidity and mortality to a minimum.

Exteriorization is the best option for bowel perforations in moribund septic patients especially in children.

Conflict of interest

The authors have no conflict of interest to report.

Acknowledgements

We would like to thank Mr. Parth K Vagholkar for his help in typesetting the manuscript.

References

1. Datta V, Sahare P, Chaturved P (2004) Guillain Barre Syndrome as a complication of enteric fever. J Indian Med Assoc 102: 172-173.
2. Biswal N (1994) Neurological manifestations of typhoid fever in children. J Trop Pediatr 40: 190.
3. Uba AF, Chirdan LB, Ituen AM, Mohammed AM (2007) Typhoid intestinal perforation in children: a continuing scourge in a developing country. Pediatr Surg Int 23: 33-39.
4. Karmacharya B, Sharma VK (2006) Results of typhoid perforation management: our experience in Bir Hospital, Nepal. Kathmandu Univ Med J. (KUMJ) 4: 22-24.
5. Gedik KE, Girgin S, Tacylidiz IH, Akgun Y (2008) Risk factors affecting morbidity in typhoid enteric perforation. Langenbecks Arch Surg 393: 973-977.
6. Ukwenya AY, Ahmed A, Garba ES (2011) Progress in management of typhoid perforation. Ann Afr Med 10: 259-265.
7. Pandove PK, Moudgil A, Pandore M, Aggarwal K, Sharda D, et al. (2014) Multiple ileal perforations and concomitant choleycystitis with gall bladder gangrene as complication of typhoid fever. J Surg Case Rep 2014: rju070.
8. Shah AA, Wani KA, Wazir BS (1999) The ideal treatment of the typhoid enteric perforation-resection anastomosis. In Surg 84: 35-38.
9. Nuhu A, Dahwa S, Hamza A (2010) Operative management of typhoid ileal perforation in children. Afr J Paediatr Surg 7: 9-13.
10. Singh M, Kumar L, Singh R, Jain AK, Karande SK, et al. (2014) Gall bladder perforation: A rare complication of enteric fever. Int J Surg Case Rep 5: 73-75.
11. Singh S, Mohanty D, Singh D, Jain BK (2010) Enteric fever with gall bladder perforation. Trop Gastroenterol 31: 48-49.
12. Saxena V, Basu S, Sharma CL (2007) Perforation of the gall bladder following typhoid fever induced ileal perforation. Hong Kong Med J 13: 475-477.
13. Durrani AB (1995) Typhoid Hepatitis. J Pak Med Assoc 45: 317-318.

14. Pramoolsinsap C, Viranuvatti V (1998) Salmonella hepatitis. J Gastroenterol Hepatol 13: 745-750.

15. Khosla SN, Singh R, Singh GP, Trehan VK (1988) The spectrum of hepatic injury in enteric fever. Am J Gastroenterol 83: 413-416.

16. Ali G, Kamilli MA, Rashid S, Mansoor A, Lone BA, et al. (1994) Spontaneous splenic rupture in typhoid fever. Post grad Med J 70: 513-514.

17. Julia J, Canet JJ, Lacasa XM, Gonzalez G, Garau J (2000) Spontaneous spleen rupture during typhoid fever. Int J Infect Dis 4: 108-109.

18. Garg P, Parashar S (1992) Pancreatic abscess due to salmonella typhi. Postgrad Med J 68: 294-295.

19. Kune CA, Coster D (1972) Typhoid pancreatic abscess. Med J Aust 1: 417-418.

Journal of Surgery
[Jurnalul de Chirurgie]
Research Article
Open Access

High Resolution Magic Angle Proton Magnetic Resonance Spectroscopy (HRMAS) in Intact Sentinel Node Biopsy from Breast Cancer Patients: A New Diagnostic Tool!

Surender Kumar[1]*, Shailendra Kumar[1], Raja Roy[2], Ankita Singh Rathore[1], Madhu Mati Goel[3], Gaurav Agarwal[4] and Sandeep Kumar[1]

[1]*Department of Surgery, King Georges Medical University, Lucknow, India*
[2]*Center of Biomedical Magnetic Resonance, Sanjay Gandhi Postgraduate Institute of Medical Sciences, Lucknow, India*
[3]*Department of Pathology, King Georges Medical University, Lucknow, India*
[4]*Department of Endocrine Surgery, Sanjay Gandhi Postgraduate Institute of Medical Sciences, Lucknow, India*

Abstract

Introduction: The extent of axillary lymph node involvement is one of the most important prognostic markers in patients of breast cancer. However, axillary dissection is associated with significant morbidity. The intra-operative sentinel node biopsy (SNB) provides a basis for omitting the routine axillary clearance however; use of in-house frozen section histopathology is required in order to substitute later. We report the use of in vitro high resolution magic angle proton magnetic resonance spectroscopy (HRMAS) for assessing the axillary nodal status with increased sensitivity.

Methods: Freshly removed axillary lymph nodes (n=17) obtained during sentinel node biopsy from 17 different patients were bisected. One segment of the bisected node was snap frozen and transported to HRMAS laboratory and was blindly subjected to 400 MHz high resolution magic angle proton magnetic resonance spectroscopy. The other portion was sent for frozen section biopsy. The tissues specimens used for HRMAS analysis and remaining portion of bisected node were then formalin fixed, paraffin embedded and sent for histo-pathological examination in separate vials. The metabolic profiles of these nodes were correlated with the routine histo-pathogical findings.

Results: On histo-pathological examination, 7 nodes were found to be positive for metastasis were as 10 nodes were negative. The spectra of nodes (n=7) found to be positive for malignant cells were exclusively dominated by signals from choline, choline containing compounds and lactate in the spectral region of 3.2 ppm and 4.12 ppm respectively. Overall the sensitivity and specificity of HRMAS in the present study was 100%.

Conclusion: Metastatic and non-involved lymph nodes in breast cancer can be accurately distinguished based on its metabolic profile. The technique of high resolution magic angle proton magnetic resonance spectroscopy can be utilized in enhancing the sensitivity and specificity of sentinel node biopsy and may replace frozen section histopathology

Keywords: High resolution magic angle spectroscopy; Sentinel node; Breast cancer

Introduction

The extent of axillary lymph node involvement in breast cancer is a dominant indicator for systemic failure [1]. Therefore an adequate axillary lymph node dissection (ALND) along with mastectomy or breast conserving procedures is an established way to achieve a cure. However the procedure carries a significant morbidity in the form of sensory neuropathy, loss of shoulder mobility, shoulder pain and lymphedema of breast and arm [2].

Intra operative frozen section histopathology of the sentinel node (s) is an alternative to the standard axillary clearance however it has variable sensitivity of 60% to 95% [3,4]. Various methods such as immuno- staining for cytokeratin have been included along with frozen section histopathology to increase the sensitivity of the sentinel node biopsy [5].

The diagnostic workups using patho-morphological changes to some extent can be substituted by molecular diagnostics techniques. Molecular techniques of proteonomics, genomics and metabonomics have emerged as possible alternative or adjudicative to histomorphological tests [6-8]. The term metabonomics is defined as "the quantitative measurement of the dynamic multiparametric metabolic response of living system to pathophysiologic stimuli or genetic modification". Magnetic resonance spectroscopy has emerged out as one of the main techniques of metabonomics and has been widely used to asses the health risk of particular drug/toxins. The *in vitro* and *in vivo* application of magnetic resonance spectroscopy for the diagnosis and therapeutic monitoring of various medical and surgical conditions like the hydatid diseases, leishmaniasis, diagnosis of malabsorption syndrome and liver graft dysfunction have been described in the past [9-12]. The conventional technique of magnetic resonance spectroscopy [MRS] required separation of analytes by preparing time consuming and labour intensive tissue extracts. Furthermore, the specimen gets consumed in the process and is unavailable for histopathological examination. The technique of high resolution proton magic angle spinning (HRMAS) spectroscopy however is yet another advancement to analyze the metabolic profile of an intact specimen and has been used successfully to differentiate malignant breast tissue from adjacent normal tissue on the basis of metabolic finger prints [13]. The quick

***Corresponding author:** Surender Kumar, Department of General Surgery, King Georges Medical University, Lucknow, India– 226 003, Tel: +9190050112189; E-mail: dr_kukku@yahoo.co.in

Received July 17, 2014; **Accepted** January 03, 2015; **Published** January 09, 2015

Citation: Kumar S, Kumar S, Roy R, Rathore AS, Goel MM. High Resolution Magic Angle Proton Magnetic Resonance Spectroscopy (HRMAS) in Intact Sentinel Node Biopsy from Breast Cancer Patients: A New Diagnostic Tool!. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 363-367 DOI: 10.7438/1584-9341-11-2-2

processing time is an added advantage that prompted us to evaluate the value of HRMAS spectroscopy *vis-a vis* frozen section histopathology in sentinel node assessment from T_1/T_2 breast carcinoma patient in a pilot study.

Method

The study was carried out at the Department of General Surgery, CSM (King George's) Medical University, Lucknow and NMR Laboratory, Sophisticated Analytical Institute Facility, Central Drug Research Institute, Lucknow. The study was duly approved by the institutional ethical committee.

Sentinel node biopsy was performed in 17 patients by injecting blue dye (*iso-sulphan blue 1%*) in the subareolar region in patients planned for local wide excision for $T_{1/}T_2$ lesions with clinically negative axillae. None of the patients had received chemotherapy.

One largest blue node was taken for this study (Figure 1). The node was thoroughly washed in normal saline, bisected and one part was used for frozen section biopsy (Figure 2) and other was put into a cryogenic vial with a unique ID code. The vial was immediately snap frozen and carried in a liquid nitrogen container to the NMR laboratory facility (1.5 kms). The node was thinly sliced using a sharp surgical blade. A large slice about 2 to 3 mm thick and weighing 35 – 40 mg was put into 4 mm HRMAS rotor. The lymph node slice was assembled inside the rotor and 20 micro liter of D2O containing tri methyl silyl tetra deuteron propionic acid (TSP) was added as standard. The HRMAS experiments were carried out on a Bruker Avance 400 MHz FT NMR spectrometer equipped with 4mm ^{1}H and ^{13}C dual HRMAS with magic angle gradient at 4°C. The samples were spun at 4.0 KHz in order to keep rotation side bands out of the acquisition window.

One dimensional proton NMR spectra with water pre-saturation were acquired using NOESY pulse sequence with a mixing time of τm 100 milli seconds. Total relaxation delay of 3.99 sec was used using 8250.8 Hz spectral width, 128 transients with a total recording time of 9.44 minutes. The one dimensional CPMG pulse sequence with water pre-saturation using an echo time of 200 milli seconds was used in order to filter off short T2 lipid component. Each experiment took about 20 minutes and the spectra were available for study on the dedicated computer screen. Assignments of HRMAS spectra were done as per published data [14]. The HRMAS spectra were read by the NMR expert (RR) who was not aware of the histopathological diagnosis of the sample. He was asked to deliver and sign a report in the shortest possible time to mimic the per-operative frozen section histopathology scenario. It may be noteworthy that routine frozen section facility was not available in our institution at the time of study. Completion axillary dissection was carried out in all 17 subjects.

The tissue used for the study was retrieved from the HRMAS rotor it was then formalin fixed and was taken for histopathological examination by standard H & E staining. The histopathology results were generally available a week later. The remaining tissue slices were also formalin fixed and sent separately for histipathological examination.

Results

Seventeen lymph node specimens from 17 subjects undergoing sentinel node biopsy from T_1/T_2 clinically N0 breast carcinoma patients were taken for the study. Each patient in this group yielded only one worthwhile lymph node for the study. The mean size of the nodes used for HRMAS study was 0.68 cms. Of all the differences in the cellular metabolism detected the high peaks of the metabolites viz; choline (Ch) and choline containing compounds eg; phosphocholine and phosphatidyl choline in the region of 3.2 ppm in 7/17 slices were most prominently seen (Figure 3). The presence of lactate at 4.12 ppm suggesting a raised anaerobic metabolism was also seen in all these 7 nodes. Furthermore, high concentration of amino acids was also observed in all these nodes. A portion of stack plot of the CPMG spectra depicting the presence of amino acid; glycine is shown in Figure 4. In 10/17 slices that were examined using the same HRMAS experiment in a clear contrast did not show the above metabolites. The NMR laboratory results could be interpreted almost instantaneously and one was quick to point out the above described differences and overtly 2 types of spectra. The HRMAS study data print outs in all the specimens examined from time to time were available within 30 minutes of receiving the tissue from the operating room.

The tissue slices retrieved from the NMR rotor were subjected to histopathology using H & E stain. There was clear and unequivocal evidence of malignancy in 7/17 nodes and none in 10/17 nodes. The histopathology of the remaining lymph node tissue slices also corroborated with the core slice examined. Upon decoding and correlating the histomorphological data with HRMAS findings it was obvious that overall the technique of HRMAS was 100% sensitive and 100% specific in these experiments (Table 1).

Discussion

This study reports a rather new efficient method of detecting tumor in sentinel node biopsy specimen from early stage breast cancer patients. The *in-vitro* use of High Resolution Magic Angle Spectroscopy on intact lymph node slice within 30 minutes from the operating room may be an important alternative to frozen section histopathological examination. Molecular diagnostic markers are an emerging field in cancer diagnosis and prognostic predictions. The *in vitro* technique of intact tissue metabonomics using HRMAS studies was able to differentiate cancer from non-cancer in this small sample of lymph nodes.

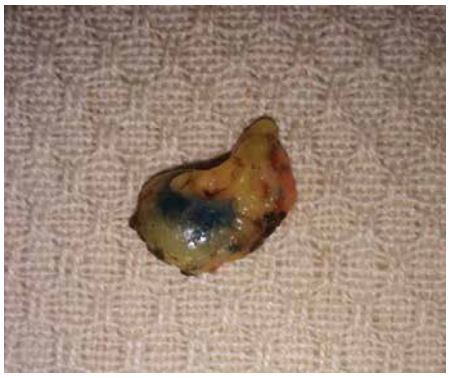

Figure 1: Extracted Blue Node

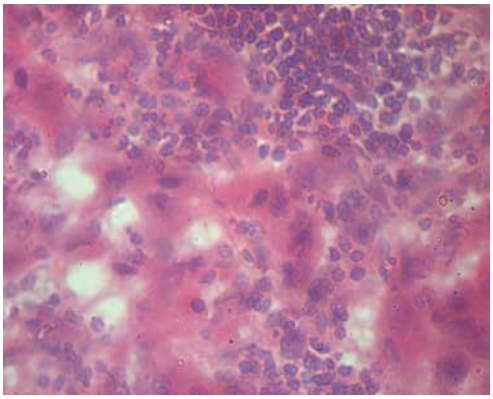

Figure 2: Frozen Section Biopsy of Involved Node.

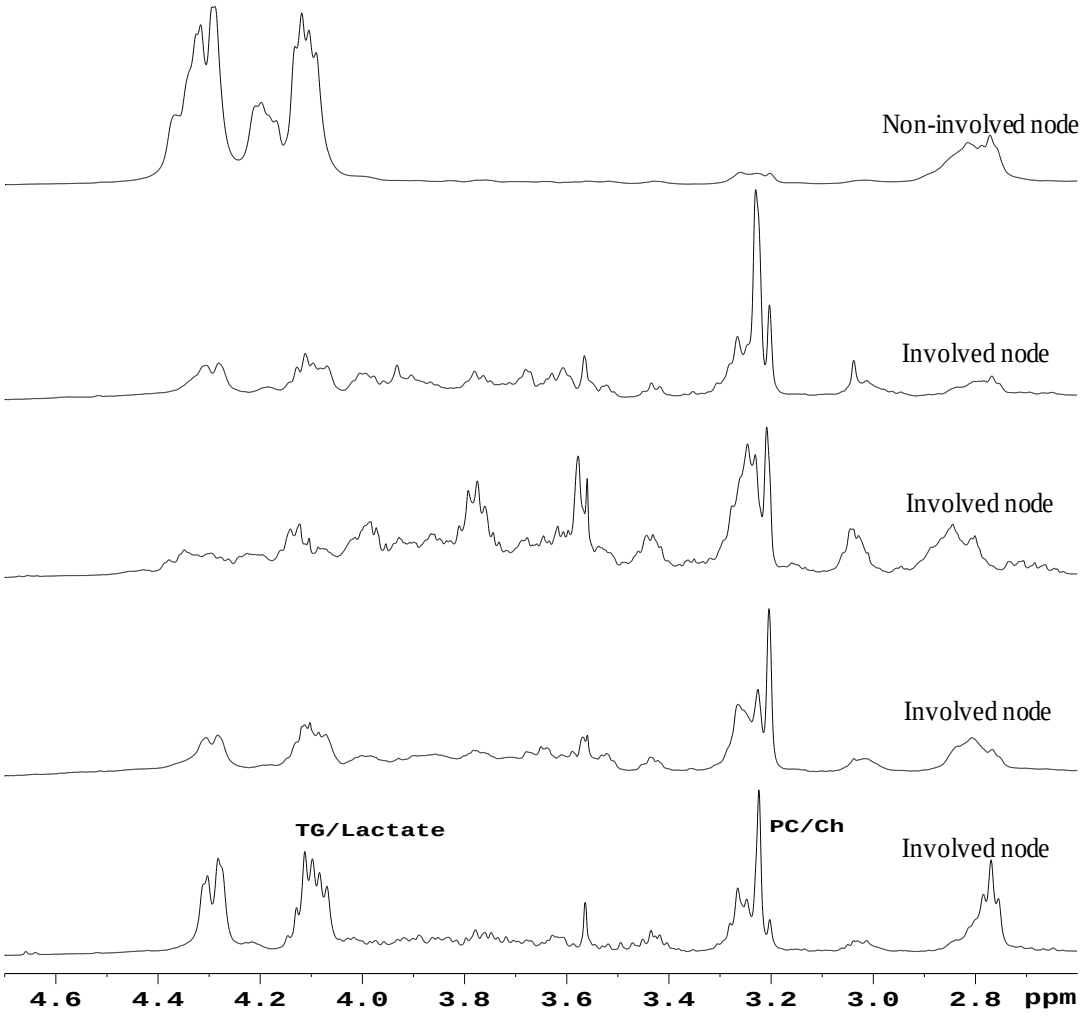

Figure 3: A Portion of 1D NOESY Stack Plot Spectra of Involved and Non-involved Lymph Nodes (PC- Phospocholine, Ch- Choline, TG- Tri Glycride)

Choline and choline containing compounds are present in more than one tumor type but these metabolites are characteristically absent in normal and benign tissues [15,16]. Identification of cancerous secondary in an axillary sentinel lymph node as opposed to a non affected node indeed demonstrated a practical use of this knowledge. Thus, of all the alterations in the metabolic profile detected by the HRMAS spectra, the region of 3.2 ppm demonstrated the most useful metabolite choline, choline containing compound (phosphocholine) and lactate at 4.12 ppm. Choline and its derivatives are one of the important building blocks of cell physiology and represent accelerated cell proliferation in the presence of malignancy. Similarly choline has been detected in breast cancer tissue extract and ex-vivo MRS studies of breast cancer [17,18]. The choline and choline containing compounds signal were reported to be less prominent in patients of breast cancer treated with neo adjuvant chemotherapy [19].

Choline and choline containing compounds were also reported from lactating breast tissue [13,20]. Whether axillary lymph nodes in lactating women also show high choline peaks is unknown. The state of lactation is associated with increased choline metabolism because of the need to nourish the newborn with large amounts of choline (supplied in the milk predominantly as phosphatidylcholine, phosphocholine, glycerophosphocholine, and free choline). This may limit the specificity of choline and choline containing compounds as discriminating metabolite in lactating breast carcinoma, a rarity, however.

The second important discriminating metabolite in this study was the lactate peaks at 4.12 ppm. In malignant cells the anaerobic metabolism of glucose is the major metabolic process and is thought to be responsible for the raised level of lactate [21].

The above described metabolites were indeed well known to occur in tissue extracts of cancers of several organs notably, breast, oral squamous cell carcinoma, urinary bladder and proste cancer, brain tumours etc. Most of the earlier studies were done on cellular extracts of tumour in the early nineties. The metabonomics of intact tissue slices referred to as *ex vivo in vitro* tissue metabonomics is a rather recent development. The HRMAS technique was used to diagnose primary breast tumour and showed a high correlation with histopathology. The breast carcinoma tissue and juxta cancer tissue free from cancer were distinguished on the basis of these spectra [22]. Choline, lactate and other metabolites were significantly elevated in the malignant tissue. This study reported from China claimed it as a new technique for the diagnosis of human breast cancer in addition to histopathology. However there is a limitation for the use of lactate as a sole marker of malignancy in the detached specimens as it may be produced anaerobically and give a false positive result. Combined presence of choline and lactate along with other metabolites like creatinine, beta-glucose, GPC, glycine, myo-inositol and taurine were suggested as a sum total marker of malignancy with greater degree of confidence or diagnostic sensitivity in tissue samples.

The clinical radiologists can also do metabolites estimation from their standard *in vivo* magnetic resonance imaging (MRI) machine by simply using software. Such an *in-vivo* HRMAS in which the patient

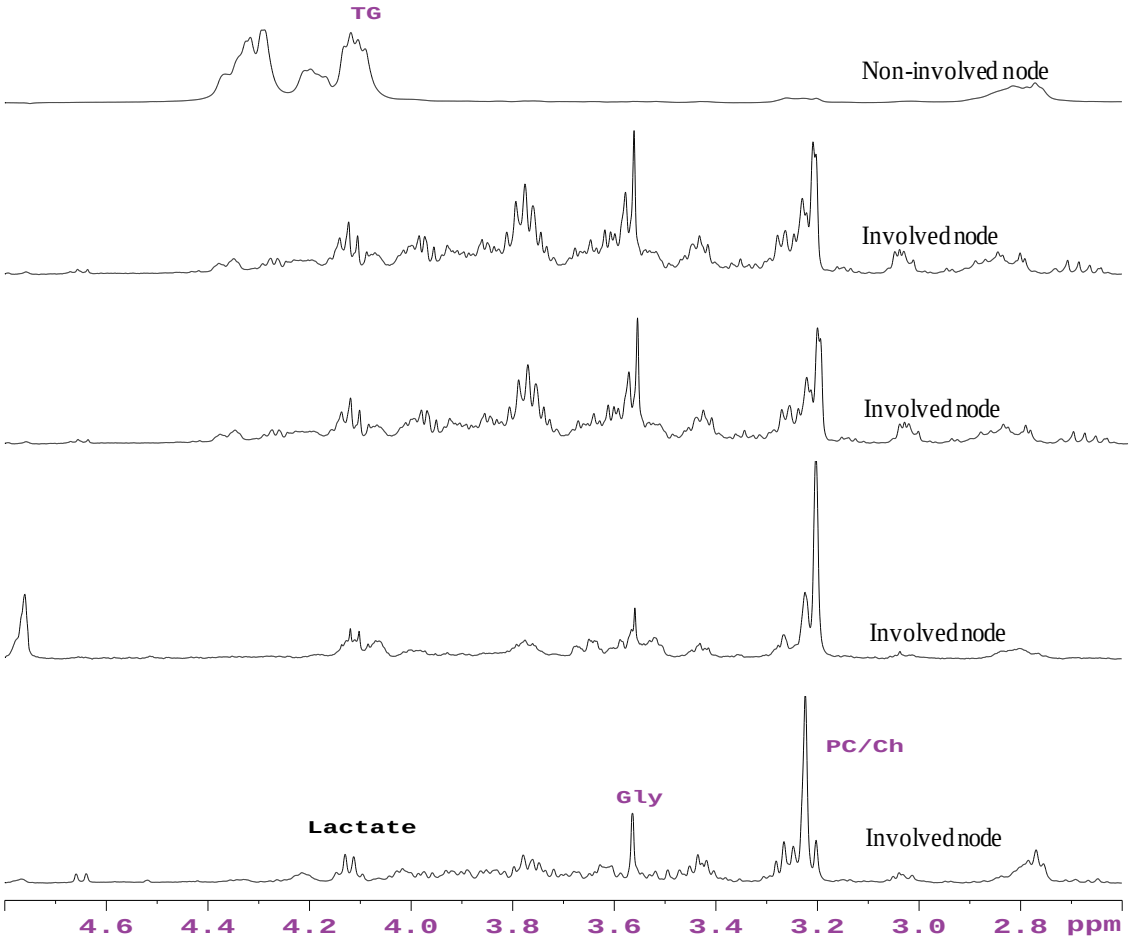

Figure 4: A Portion of CPMG Stack Plot Spectra of Various Lymph Nodes

Table 1: Test Results of HRMAS Compared with Histopathological Examination

	HPE +ve	HPE –ve	Total
HR MAS +ve	7	0	7
HR MAS -ve	0	10	10
Total	7	10	

Sensitivity: 100%

Specificity: 100%

Positive predictive value: 100%

Negative predictive value: 100%

herself goes into the MRI machine and the metabolic profile of the tumor is recorded with the help of special software has been evaluated in axillary nodes in breast cancer in 2 studies [23,24]. Firstly, in 35 nodes using the basis of choline signals alone the authors identified metastasis with sensitivity, specificity and accuracy of 82%, 100% and 90% [23]. In the second study, using the similar technique in 20 nodes the sensitivity, specificity and accuracy of 80%, 91%, and 88% were described. It was further noted in these studies conducted longitudinally on these patients that neo-adjuvant chemotherapy lowered the concentration of various metabolites e.g. choline, phosphocholine, etc. [24]. As these patients were undergoing chemotherapy and longitudinal assessment by MRI these describe a different subset of patients in whom rather large axillary lymph nodes were present as opposed to the non palpable nodes in the axillae of the patients that have been included in the present study. The ability of MRI to detect a sentinel node in clinically N0 axillae of early breast carcinoma has not been evaluated. The MRI along with *in vivo* MRS in clinically N0 axillae can theoretically be a tool for pre operative detection of malignancy in a sentinel node.

The molecular diagnosis of malignant change as opposed to histomorphological diagnosis is an emerging field of medical research. Metabnomics by MRS equipments are generally expensive commodity largely available in big public sector hospitals and corporate pharmaceuticals industries so far. There is relative paucity of trained man power in the field of MRS. Pharmacological companies and chemical industries routinely use MRS to test the purity of their products. Clinician's interest however, in this field has been tardy. An increasing interest and availability of the MRS equipment is being witnessed world over. The MRS or *in vivo* MRS and MRI as common facility can become cost effective I future with increasing usage.The *in-vitro* per-operative expeditious assessment HRMAS of an intact axillary lymph node slice or other tissues as a central facility for a number of hospitals in the vicinity of the HMRS facility can be promoted as a cost effective technology. Several other applications of *in- vitro* HRMAS studies include detection of micro-metastases, ability to detect residuals in the tumor bed and tumor margins, metabolites in tumor aspirate, exfoliated cells metabonomics, brush cytology specimens and, fluids like bronchial lavage, asicitic tap and nipple discharge. The initial fixed cost of the equipment though high is also likely to go down with widespread use in the future. Though at present the discriminating metabolites for a particular type of carcinoma or sarcoma are not available, the study designs like the present study offer a useful application of this technology within the present level of knowledge. This study was conducted to mimic the scenario of sentinel node assessment in the operating theatre. To this effect HMR spectroscopy emerged as an efficient and reliable method for the evaluation of the sentinel node as compared with routine histopathology. A larger sample size on a even higher frequency MRS (800 MHz) equipment may further enhance these results.

Acknowledgement

1. Financial assistance from ICMR New Delhi, India via grant no. 5 / 13 / 96 / 2003 – NCD III , IRIS No : 2003 – 04710

2. We are grateful to the NMR division of SAIF, CDRI, Lucknow for providing the NMR facility and thankful to MR HM Guniyal for recording the HR-MAS NMR spectra of the lymph node specimens.

References

1. Henderson IC, Patek AJ (1998) The relationship between prognostic and predictive factors in the management of breast cancer. Breast Cancer Res Treat 52: 261-288.

2. Duff M, Hill AD, McGreal G, Walsh S, McDermott EW, et al. (2001) Prospective evaluation of the morbidity of axillary clearance for breast cancer. Br J Surg 88: 114-117.

3. Tanis PJ, Boom RP, Koops HS, Faneyte IF, Peterse JL, et al. (2001) Frozen section investigation of the sentinel node in malignant melanoma and breast cancer. Ann Surg Oncol 8: 222-226.

4. Layfield DM, Agrawal A, Roche H, Cutress RI (2011) Intraoperative assessment of sentinel lymph nodes in breast cancer. Br J Surg 98: 4-17.

5. Zurrida S, Mazzarol G, Galimberti V, Renne G, Bassi F, et al. (2001) The problem of the accuracy of intraoperative examination of axillary sentinel nodes in breast cancer. Ann Surg Oncol 8: 817-820.

6. Bernini A, Spencer M, Frizelle S, Madoff RD, Willmott LD et al. (2000) Evidence for colorectal cancer micrometastasis using reverse transcriptase polymerase chain reaction analysis of MUC2 in lymph nodes. Cnacer Detect Prev 24: 72-79.

7. Liefers GJ, Tollenaar RA, Cleton-Jansen AM (1999) Molecular detection of minimal residual disease in colorectal and breast cancer. Histopathology 34: 385-390.

8. Cohn KH, Ornstein DL, Wang F, LaPaix FD, Phipps K, et al. (1997) The significance of allelic deletions and aneuploidy in colorectal carcinoma. Results of a 5-year follow-up study. Cancer 79: 233-244.

9. Garg M, Gupta RK, Prasad KN, Sikora SS, Pal L, et al. (2002) Fertility assessment of hydatid cyst by proton MR spectroscopy. J Surg Res 106: 196-201.

10. Gupta N, Goyal N, Singha UK, Bhakuni V, Roy R, et al. (1999) Characterization of intracellular metabolites of axenic amastigotes of Leishmania donovani by 1H NMR spectroscopy. Acta Trop 73: 121-133.

11. Bala L, Nagana Gowda GA, Ghoshal UC, Misra A, Bhandari M, et al. (2004) 1H NMR spectroscopic method for diagnosis of malabsorption syndrome: a pilot study. NMR Biomed 17: 69-75.

12. Singh HK, Yachha SK, Saxena R, Gupta A, Nagana Gowda GA, et al. (2003) A new dimension of 1H-NMR spectroscopy in assessment of liver graft dysfunction. NMR Biomed 16: 185-188.

13. Kvistad KA, Bakken IJ, Gribbestad IS, Ehrnholm B, Lundgren S, et al. (1999) Characterization of neoplastic and normal human breast tissues with *in vivo* (1) H MR spectroscopy. J Magn Reson Imaging 10: 159-164.

14. Sitter B, Sonnewald U, Spraul M, Fjösne HE, Gribbestad IS (2002) High-resolution magic angle spinning MRS of breast cancer tissue. NMR Biomed 15: 327-337.

15. Stanwell P, Gluch L, Clark D, Tomanek B, Baker L, et al. (2005) Specificity of choline metabolites for *in vivo* diagnosis of breast cancer using 1H MRS at 1.5 T Eur Radiol 15: 1037-1043.

16. Rutter A, Hugenholtz H, Saunders JK, Smith IC (1995) Classification of brain tumors by ex vivo 1H NMR spectroscopy. J Neurochem 64: 1655-1661.

17. Gribbestad IS, Fjösne HE, Haugen OA, Nilsen G, Krane J, et al. (1993) In vitro proton NMR spectroscopy of extracts from human breast tumours and non-involved breast tissue. Anticancer Res 13: 1973-1980.

18. Kumar M, Jagannathan NR, Seenu V, Dwivedi SN, Julka PK, et al. (2006) Monitoring the therapeutic response of locally advanced breast cancer patients: sequential *in vivo* proton MR spectroscopy study. J Magn Reson Imaging 24: 325-332.

19. Jagannathan NR, Kumar M, Seenu V, Coshic O, Dwivedi SN, et al. (2001) Evaluation of total choline from *in-vivo* volume localized proton MR spectroscopy and its response to neoadjuvant chemotherapy in locally advanced breast cancer. Br J Cancer 84: 1016-1022.

20. Stanwell P, Gluch L, Clark D, Tomanek B, Baker L, et al. (2005) Specificity of choline metabolites for *in vivo* diagnosis of breast cancer using 1H MRS at 1.5 T. Eur Radiol 15: 1037-1043.

21. Gribbestad IS, Fjösne HE, Haugen OA, Nilsen G, Krane J, et al. (1993) In vitro proton NMR spectroscopy of extracts from human breast tumours and non-involved breast tissue. Anticancer Res 13: 1973-1980.

22. Cheng LL, Chang IW, Smith BL, Gonzalez RG (1998) Evaluating human breast ductal carcinoma with High-Resolution Magic Angle Spinning Proton Magnetic Resonance Spectroscopy. J Magn Reson 135: 194-202

23. Yeung DK, Yang WT, Tse GM (2002) Breast cancer: *in vivo* proton MR spectroscopy in the characterization of histopathologic subtypes and preliminary observations in axillary node metastases. Radiology 225: 190-197.

24. Seenu V, Pavan Kumar MN, Sharma U, Gupta SD, Mehta SN, et al. (2005) Potential of magnetic resonance spectroscopy to detect metastasis in axillary lymph nodes in breast cancer. Magn Reson Imaging 23: 1005-1010.

Journal of Surgery
[Jurnalul de Chirurgie]
Case Report Open Access

Retrorectal Myxoid Fibrosarcoma: A New Entity

Mehdi Soufi[1]* and Abdelhamid Essadel[2]

[1]Division of Digestive Surgery, University Mohammed First, Oujda, Morocco
[2]Department of Surgery B, CHU Ibn Sina, Rabat, Morocco

Abstract

We report the case of a 40-year-old man who admitted to our department because of progressive increasing abdominal girth and weight loss. CT scan showed a huge retrorectal tumor. The mass was totally excised by laparotomy. At five years, the patient was doing well with disease free. Tumors occurring in the retrorectal space are rare; we report herein the first case of a myxoid fibrosarcoma of retrorectal space.

Keywords: Retrorectal tumor; Fibrosarcoma; Surgery

Introduction

The presacral space, which contains different types of embryonic tissue, is a potential site for several tumors. Primary fibrosarcoma is an exceptional retrorectal tumor. We report the case of a 40-year-old man with a huge retrorectal tumor treated successfully by surgery alone. This is the first report of a case of a myxoid fibrosarcoma of retrorectal space.

Case Report

A 40-year-old man was admitted to our department because of progressive increasing abdominal girth, asthenia and weight loss. On physical examination, the abdomen was distended owing to a huge, non-tender, palpable mass with no clear borders. Laboratory findings and tumor markers were within normal limits. A computed tomography scan revealed a bulky inhomogeneously enhancing retrorectal mass displacing completely the abdominal organs, with no clear signs of infiltration. T1- and T2-weighted magnetic resonance imaging showed a well-defined soft-tissue mass measuring 24 × 17 × 9.0 cm located in retrorectal space (Figure 1). At laparotomy, a giant mixte tumor that filled completely the retrorectal space was found. The mass was totally excised and the main difficulty was to guarantee clear margins sparing the rectum and the sacrum (Figure 2). The rectum was spared and no intestinal resection was necessary to have clear margins. No lymph node dissection was done.

The mass weighed 18 kg and appeared grossly multinodular, partially confluent with solid areas. The histopathological examination showed fibroblasts with myxoid stroma and a rich capillary network (Figure 3).

On immunohistochemical study, the tumor cells expressed vimentin, SMA and MUC 4 (Figure 3). There was no expression of desmin, CD34 and EMA. The diagnosis of a low grad fibromyxoid sarcoma was established. The postoperative course was uneventful. The patient was discharged one week after surgery. Five years later, the patient was free from recurrence.

Discussion

The true incidence of tumors occurring in the retrorectal (presacral) space is unknown, yet several retrospective series suggest that between one and six patients will be diagnosed annually in major referral centers [1]. The retrorectal space contains multiple embryologic remnants derived from a variety of tissues. Tumors that develop in this space are both macroscopically and histologically heterogeneous. Most lesions are benign, but malignant neoplasms are not uncommon. Solid lesions are more likely to be malignant than are cystic lesions. Neurogenic lesions typically arise from peripheral nerves and represent about 10% of retrorectal tumors [2]. These tumors include neurofibromas and sarcomas, neurilemomas, ependymomas, and ganglioneuromas. Fibromyxoid sarcoma is a rare soft tissue sarcoma usually located in the deep soft tissue in the groin or lower extremities. No case located in retrorectal space has been reported in literature review [2]. Thus, it is important to differentiate this tumor from other soft tissue tumors [3].

Fibrosarcoma occurs more commonly in men than in women. It can be diagnosed in patients of any age, but it is diagnosed more frequently in patients in the fourth or the fifth decade of life, as in our patient.

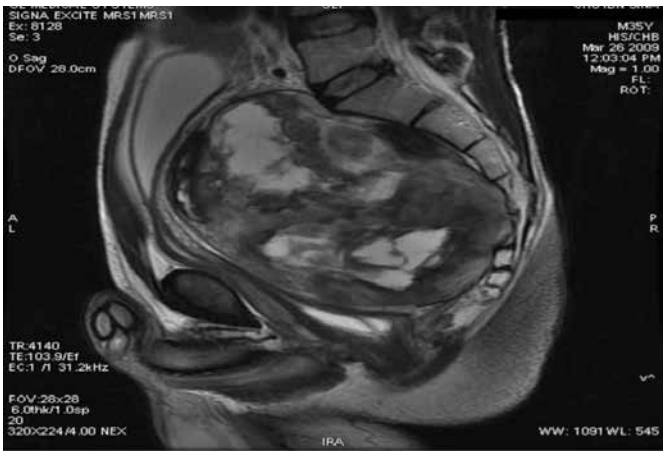

Figure 1: Magnetic resonance imaging showing a well-defined soft-tissue mass measuring 24 × 17 × 9.0 cm located in the retrorectal space.

***Corresponding author:** Mehdi Soufi, MD, Division of Digestive Surgery, University Hospital of Oujda, Faculty of Medicine, University Mohammed First, BP 4847; Oujda University, Oujda, Morocco, 60049, Tel: +212661212924; E-mail: drsoufimehdi@hotmail.fr

Received September 01, 2014; **Accepted** January 13, 2014; **Published** January 19, 2015

Citation: Soufi M, Essadel A. Retrorectal Myxoid Fibrosarcoma: A New Entity. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 369-370 DOI: 10.7438/1584-9341-11-2-3

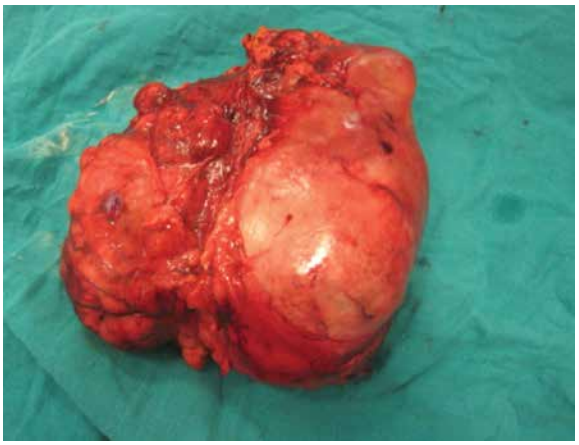

Figure 2: Resection Specimen.

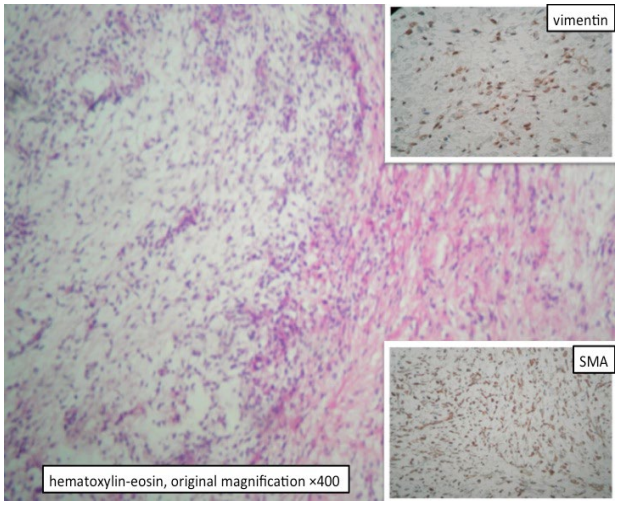

Figure 3: Histopathological examination showing fibroblasts with myxoid stroma and a rich capillary network (hematoxylin-eosin, original magnification ×400).

Symptoms of retrorectal tumors are often nonspecific and are related to the location and to the size of the lesion. The majority of benign cystic lesions are asymptomatic and usually discovered on routine rectal examination. Pain and neurologic dysfunction might be the presenting symptoms and are related to the route of the involved nerve.

Pelvic MRI is emerging as the most sensitive and specific imaging study of these tumors [4,5].

Almost all retrorectal tumors require surgical management [4]. The case described herein suggests that the dimension alone should not be considered as a contraindication for an aggressive surgical approach. From a technical point of view, clear margins of resection can be difficult to obtain for these tumors because of their proximity, attachment to, or often invasion of major anatomic structures. When a major volume tumor cannot be removed, aggressive resection of other organs must be done [6,7].

Histologically, these neoplasms demonstrated contrasting fibrous and myxoid areas, a swirling, whorled growth pattern (at least in part), and bland, deceptively benign-appearing fibroblastic spindle cells. The cellularity of these tumors is low with a rich capillary vascular network visible in myxoid areas [3]. In some cases, fibrosarcoma might be difficult to distinguish from other tumors such as a dedifferentiated liposarcoma or malignant fibrous histiocytoma. The presence of a storiform pattern and epithelioid type cells would support the diagnosis of a malignant fibrous histiocytoma.

Non-surgical treatment, such as radiation treatment and chemotherapy, might improve local control making the appearance of clinically evident metastatic disease less likely. Although adjuvant therapy has enhanced the chance of cure for retroperitoneal sarcomas, there are no studies for tumors of retrorectal space. In fact, chemotherapy for retrorectal sarcomas seems to be ineffective. Thus, further studies are necessary to clarify the role of adjuvant treatment for local control of these tumors [3].

Overall survival appears to be good if the resection is complete [7]. Indeed, our patient was disease free five years after surgery.

Conclusion

Myxoid fibrosarcoma of retrorectal space is rare. Surgery remains the key of treatment and might be of an aggressive approach in managing huge tumors.

Acknowledgement

All authors contributed to the realization of this manuscript.

Conflict of interest:

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

References

1. Hobson KG, Ghaemmaghami V, Roe JP, Goodnight JE, Khatri VP (2005) Tumors of the retrorectal space. Dis Colon Rectum 48: 1964-1974.
2. Glasgow SC, Birnbaum EH, Lowney JK, Fleshman JW, Kodner IJ, et al. (2005) Retrorectal tumors: a diagnostic and therapeutic challenge. Dis Colon Rectum 48: 1581-1587.
3. Evans HL (1993) Low-grade fibromyxoid sarcoma. A report of 12 cases. Am J Surg Pathol 17: 595-600.
4. Sagar AJ, Koshy A, Hyland R, Rotimi O, Sagar PM (2014) Preoperative assessment of retrorectal tumours. Br J Surg 101: 573-577.
5. Boscà A, Pous S, Artés MJ, Gómez F, Granero Castro P, et al. (2012) Tumours of the retrorectal space: management and outcome of a heterogeneous group of diseases. Colorectal Dis 14: 1418-1423.
6. Chéreau N, Lefevre JH, Meurette G, Mourra N, Shields C, et al. (2013) Surgical resection of retrorectal tumours in adults: long-term results in 47 patients. Colorectal Dis 15: 476-482.
7. Du F, Jin K, Hu X, Dong X, Cao F (2012) Surgical treatment of retrorectal tumors: a retrospective study of a ten-year experience in three institutions. Hepatogastroenterology 59: 1374-1377.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Accidental Corrosive Acid Ingestion Resulting in Isolated Pyloric Stenosis: A Rare Phenomenon

Lovekesh Kumar[1], Ashish Saxena[2]*, Mahendra Singh[3], Yuvraj Kolhe[2], Snehal K Karande[2] and Om Prakash Prasad[2]

[1]*Dept. of Surgery, Maulana Azad Medical College, New Delhi, India*
[2]*Dept. of Surgery, Hindu Rao Hospital, New Delhi, India*
[3]*Dept. of Surgery, AIIMS, Jodhpur, India*

Abstract

Accidental corrosive ingestion is a common occurrence in developing nations. In most of the cases these agents damage the oesophagus and stomach. Isolated injury of pylorus of stomach has a relatively low incidence. We report such a case of accidental ingestion of corrosive acid, which resulted in isolated stenosis of the pyloric antrum. The diagnosis was confirmed by Barium meal and endoscopy. The patient underwent gastro jejunostomy and recovered well after the surgery. A brief discussion of mechanism of corrosive injury, clinical features, investigations and management follows.

Keywords: Corrosive poisoning; Isolated pyloric stenosis; Barium meal; UGI endoscopy; Gastro jejunostomy

Introduction

Corrosive acid ingestion is a common source of morbidity in the developing world. The incident is especially higher in India due to unregulated sale of corrosive material in the market [1]. Children are particularly susceptible to the accidental exposure to such substances due to inadequate parental supervision and careless storing of these chemicals at homes [2]. Both acid and alkali when consumed, cause significant injury to the upper gastrointestinal tract. The extent of injury depends upon several factors such as nature of the offending agent, amount, concentration and duration of exposure [1]. Typically corrosive acid ingestion leads to local reaction, oesophageal damage and gastric injury in that order [3]. Isolated injury to the stomach resulting in pyloric stenosis is very rare, accounting to as little as 3.8% of all the cases of corrosive ingestion, as reported in literature [4]. We report such a case of corrosive acid poisoning that resulted in isolated pyloric stenosis, without any oesophageal damage necessitating a bypass procedure. A brief discussion upon clinical picture (Figures 1 and 2) investigations and management follows.

Case Report

An 11 year old girl child presented to us with the history of accidental corrosive ingestion 1 month back. She was admitted in a medical ward for 1 week and was managed conservatively. After discharge she was tolerating oral diet satisfactorily until 1 week back, when she started regurgitating the ingested food a few hours after intake. For the last 1 week she was having frequent episodes of vomiting, especially after food intake and was losing weight.

Upon arrival at the surgical facility she had an emaciated appearance. Abdomen was scaphoid with fullness appearing in epigastrium after food intake. Succussion splash was present and the stomach was found to be dilated upon ausculto percussion. However, no mass was palpable in epigastrium.

She underwent routine blood investigations which demonstrated anemia and hypoproteinemia. Barium meal study and upper GI endoscopy were undertaken which revealed the presence of pyloric stenosis. Oesophagus and proximal stomach were found to be normal in appearance.

A diagnosis of gastric outlet obstruction was made and patient was taken for a bypass procedure. Intra operatively gastric mucosa was found to be inflammed and pylorus was thickened with a narrowed lumen. A gastro jejunostomy was performed to bypass the obstruction. The patient made an uneventful recovery following the surgery. She was taking normal diet and gaining weight as noted during her last follow up visit.

Discussion

Corrosive injuries of upper gastrointestinal tract occur frequently in India. These result mostly from the ingestion of corrosive substances either accidentally or with suicidal intent. Hydrochloric acid is the most common cause of corrosive poisoning in India, due to its easy availability as a cheap toilet cleaner [5]. The oesophagus and the stomach bear the major brunt of injury. Almost one third of the cases develop cicatrization of the stomach [6].

Acids produce coagulative necrosis of the tissue, and form an eschar at the site of injury resulting in segmental or extensive stricture formation in the long run. In contrast alkalis produce penetrating or liquifactive necrosis [7]. Acid is more likely than alkali to impart injury to stomach [8]. The pathological process begins a few hours after corrosive ingestion in the form of small vessel thrombosis. It continues for one to two weeks beyond which bacterial infection along with inflammatory response and granulation tissue deposition dominates the pathological profile [9]. Healing process begins three weeks after the injury, leading to fibrosis and narrowing of lumen, ultimately resulting in stricture [10].

***Corresponding author:** Ashish Saxena, Department of Surgery, Hindu Rao Hospital, New Delhi, India- 110007, Tel: +91 9654249322; E-mail: docashish4u@gmail.com

Received November 03, 2014; **Accepted** February 06, 2014; **Published** February 12, 2015

Citation: Kumar L, Saxena A, Singh M, Kolhe Y, Karande SK, et al. Accidental Corrosive Acid Ingestion Resulting in Isolated Pyloric Stenosis: A Rare Phenomenon. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 371-373 DOI: 10.7438/1584-9341-11-2-4

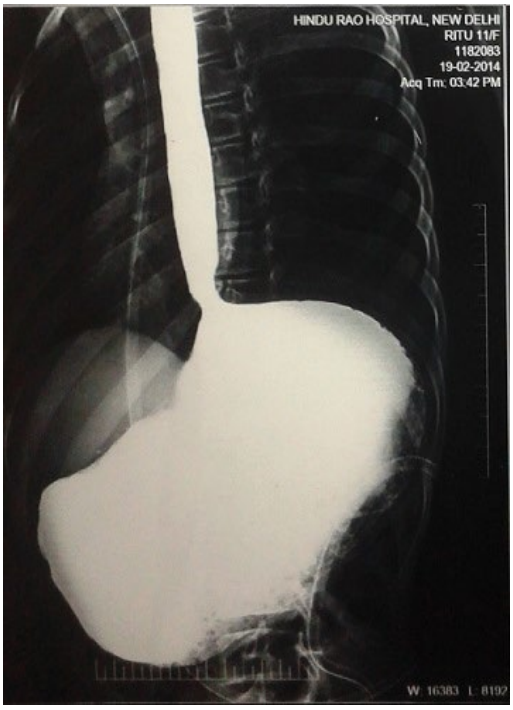

Figure 1: Barium meal study showing normal esophageal and gastric lining

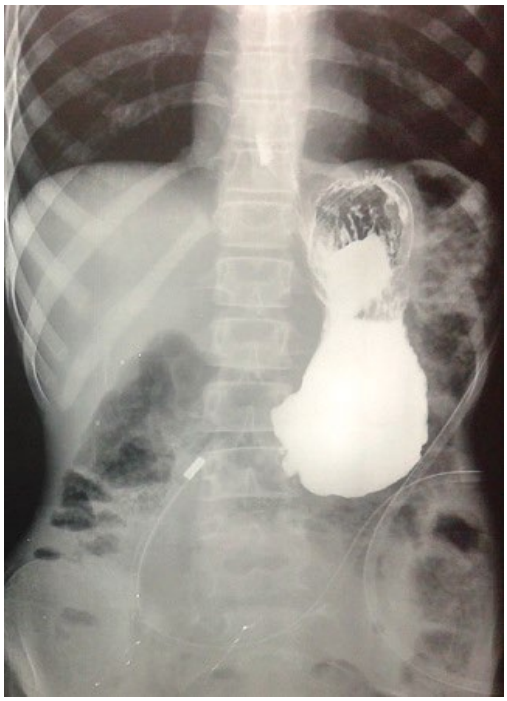

Figure 2: Barium meal study showing distended stomach with obstruction at pylorus Jejunostomy

Clinical features of the corrosive injury depend upon several factors such as nature; amount, concentration and duration of exposure of the offending agent [1]. The corrosives in powder and crystal form tend to adhere to the mouth and throat and produce maximum damage in these regions. The liquid agents pass rapidly through the esophagus and cause more damage to the sites of esophageal narrowing such as cricopharyngeal region, at the level of arch of aorta and lower esophageal sphincter [11].

The age old saying that "Acid licks the oesophagus and bites the stomach" still holds good [12]. Main reasons behind it being the relative resistance of oesophageal squamous epithelium to the acids, rapid passage through oesophagus, significant distensibility of stomach and acid induced reflex pyloric spasm. These factors prolong the contact period of acid with gastric mucosa and result in a myriad gastric deformity such as pyloric stenosis, antral stricture, hour glass stomach, or small contracted stomach [13]. In a study published by Ananthkrishnan et al., acid ingestion was found to be responsible for 82.6% of chronic gastric injuries, the majority of them constituted by pyloric stenosis [14].

Most of the patients with pyloric stenosis present within three months of ingestion of corrosive liquids, however symptoms are known to develop as late as one year after the injury [15]. These include feeling of fullness of stomach, nausea, and vomiting and weight loss, the features characteristic of gastric outlet obstruction (GOO). Associated features of GOO include severe dehydration and dyselectrolytemia.

Upon clinical examination fullness in the epigastrium is to be looked for, which is suggestive of underlying dilated stomach. Succusion splash and ausculto percussion are the hallmark clinical signs of GOO. Barium Meal and upper GI endoscopy constitute the radiological investigations required to establish the diagnosis. While a barium meal shows an over distended stomach with a narrowed pyloric lumen associated with delayed emptying, the endoscopy is vital in evaluating the mucosa of stomach and assessing the degree of lumen narrowing. The results of barium meal study and endoscopy determine the appropriate management.

The cases of partial obstruction can be managed by balloon dilation, endoscopic intra lesional steroid injection or pyloroplasty. On the other hand the complete gastric outlet obstruction is treated either by gastro jejunostomy or by gastric resection along with Bilroth I reconstruction [16]. As our patient was having complete obstruction of the pylorus she underwent gastro jejunostomy. Gastric resection was considered unsafe because of the presence of dense adhesions in the perigastric region.

Conclusion

Isolated pyloric stenosis following corrosive ingestion is a relatively uncommon entity. It is more frequently seen in the patients with acid ingestion. The features of gastric outlet obstruction manifest 3- 6 weeks after the ingestion of corrosive. Upper GI endoscopy and barium meal are the necessary investigations to establish the diagnosis. Surgical intervention in the form of pyloroplasty or gastro jejunostomy is the preferred treatment.

References

1. Lakshmi CP, Vijayhari R, Kate V, Ananthakrishnan N (2013) A hospital-based epidemiological study of corrosive alimentary injuries with particular reference to the Indian experience. Nat Med J India. 26: 31-36.
2. Urganci N, Usta M, Kalyoncu D, Demirel E (2014) Corrosive substance ingestion in children. Indian J Pediatr 81: 675-679.
3. Contini S, Scarpignato C (2013) Caustic injury of the upper gastrointestinal tract: a comprehensive review. World J Gastroenterol 19: 3918-3930.
4. Ciftci, AO, Senocak ME, Buyukpamukcu N, Hiçsönmez A (1999) Gastric outlet obstruction due to corrosive ingestion: incidence and outcome. Pediatr Surg Int. 15: 88-91.
5. Agarwal S, Sikora SS, Kumar A, Saxena R, Kapoor VK (2004) Surgical management of corrosive strictures of stomach. Indian J Gastroenterol. 23: 178-180.
6. Zargar SA, Kochhar R, Nagi B, Mehta S, Mehta SK (1989) Ingestion of corrosive acids. Spectrum of injury to upper gastrointestinal tract and natural history. Gastroenterology 97: 702-707.
7. Chibishev A, Simonovska N, Shikole A (2010) Post-corrosive injuries of upper gastrointestinal tract. Prilozi 31: 297-316.

8. Subbarao KS, Kakar AK, Chandrasekhar V, Ananthakrishnan N, Banerjee A (1988) Cicatrical gastric stenosis caused by corrosive ingestion. Aust N Z J Surg 58: 143-146.

9. Andreoni B, Biffi R, Padalino P, Marini A, Marzona L, et al. (1994) Artificial nutrition in the management of lesions caused by caustic ingestion. Chir Ital 46: 42-48.

10. Ionescu M, Tomulescu V, Gheorghe C, Popescu I (2000) [Post-caustic esophageal stenosis]. Chirurgia (Bucur) 95: 23-28.

11. Christesen HB (1993) Ingestion of caustic agents. Epidemiology, pathogenesis, course, complications and prognosis] Ugeskr Laeger 155: 2379-2382.

12. Marks IN, Bank S, Werbellof L, Farman J, Louw JH (1963) The natural history of corrosive gastritis: Report of five cases. Am J Dig Dis 8: 509-524.

13. Roy M Jr, Calonje MA, Mouton R (1962) Corrosive gastritis after formaldehyde ingestion: report of a case. N Engl J Med 266: 1248-1250.

14. Ananthakrishnan N, Parthasarathy G, Kate V (2010) Chronic corrosive injuries of the stomach-a single unit experience of 109 patients over thirty years. World J Surg 34: 758-764.

15. Hsu CP, Chen CY, Hsu NY, Hsia JY (1997) Surgical treatment and its long-term result for caustic-induced prepyloric obstruction. Eur J Surg 163: 275-279.

16. Poocharoen W (2008) Corrosive injury induced gastric outlet obstruction in children: A case report. Thai J Surg 29: 54-58.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Unexpected Outcome of a Floating Thrombus in the Ascending Aorta

Hassane Abdallah[1,2]*, Pierre-Emmanuel Noly[2] and Gebrine Elkhoury[1]

[1]Department of Cardiothoracic and Vascular Surgery, Cliniques Universitaires Saint-Luc, Brussels, Belgium
[2]Department of Cardiac Surgery, Montreal Heart Institute, Montreal, Canada

Abstract

We report the case of a 46-year-old woman, without any known systemic disease in the past, a history of car accident two months earlier with fracture of the left arm and the jaw. She presented to a local hospital, with prolonged precordial pain of ON/OFF occurrence, where acute coronary syndrome (ACS) was diagnosed and treated with conventional (aspirin, clopidegrel, β-blockers, heparin) treatment. She was then transferred to our hospital for cardiac catheterization.

Coronary angiography showed normal coronary arteries, a moving filling defect was visible in the ascending aorta. An immediate transesophageal echocardiography revealed a free floating mass attached to the left cusp of the aortic valve .and occluding the left main coronary trunk, a trace of aortic valve regurgitation. The patient was hemodynamically unstable. Urgent operation was carried on and resection of the mass was done.

Keywords: Ascending aorta; Floating thrombus; Coronary angiography; Cardiopulmonary bypass

Case Report

46-year-old female, without past medical history other than a car accident causing a left arm and a jaw fracture, presented a syncope lasting couple minutes, to get up later with no neurological sequel. She refused to go to the hospital and have passed the whole day shopping. On arrival back home, she had a severe precordial pain of ON/OFF nature that obliged her to go to a local hospital, where acute coronary syndrome (ACS) was diagnosed and treated with conventional treatment (aspirin, clopidogrel, nitroglycerin, and heparin). She was then transferred to our hospital for cardiac catheterization. On patient arrival, endotracheal intubation was performed, no abnormalities were found on examination of the heart, the electrocardiogram (ECG) showed ST-segment elevations in leads V2, V3, V4, V5, and V6. The supine chest X-ray showed mild pulmonary congestion with normal mediastinum. The systolic blood pressure was 90 mmHg, diastolic 60 mmHg; the pulse rate was 82 bpm. BUN and creatinine were normal. The total CK was 10000U/L A prompt coronary angiography was done which showed normal coronary arteries. A moving filling defect was visible in the ascending aorta (Figure 1). The patient was hemodynamically unstable and intraaortic balloon pump was inserted. As the patient's hemodynamic state deteriorated, conterpulsation was immediately terminated. A transesophageal echocardiography revealed a free floating mass (1,8 ×1,1cm) attached to the left cusp of the aortic valve that resulted in occlusion of the left main stem during diastole , severe septal and anterolateral wall hypokinesis, a trace of aortic valve regurgitation. The estimated left ventricular ejection fraction (LVEF) was 20%.

The patient was transferred immediately to the operation room. Through a median sternotomy, cardiopulmonary bypass (CPB) was instituted by means of cannulation of the ascending aorta and the right atrium. Anterograde and retrograde cardioplegia were used. The aortic root was transversely incised. A floating pedunculated mass attached to an atherosclerotic plaque on the left cusp of the aortic valve was found. The mass was in close proximity to the left main stem ostium, causing intermittent occlusion of the latter (Figure 2). The mass was excised and the aortotomy was closed directly with a running suture. The aortic cross clamp time was 30 minutes. Sinus rhythm reumed early after releasing the aortic cross clamp but it was impossible to wean CPB despite inotropic drugs and IABP support. Transesophageal echocardiography revealed septal and anterolateral wall akinesis. The LVEF was estimated to be less than 5%. A central extracorporeal membrane oxygenation (ECMO) was then installed and the patient was transferred to the intensive care unit. The immediate postoperative chest X-Ray showed moderate pulmonary congestion. On the second postoperative day, the chest X-Ray showed severe pulmonary edema. Cardiac contractility was evaluated by a daily echocardiography. On the seventh day, a better myocardial contractility was noted and the LVEF was estimated at 20%. However, attempts to wean the ECMO support were unsuccessful, despite the use of inotropic drugs and IABP. The patient was kept on ECMO support for eight more days and died of pulmonary hemorrhage awaiting a donor for cardiac transplantation.

Discussion

Floating thrombi in the aorta are a rare finding in the absence of any coagulation abnormality. They often represent a surgical emergency. This life threatening appears to be more common in female smokers in their fifth decade.

Atherosclerosis, dissection, trauma, malignancy and coagulopathies have been associated with aortic mural thrombi [1]. Intraluminal thrombus may be located in the ascending aorta, even without extensive atherosclerotic plaques [2].

In our patient, the origin of the aortic thrombus was atheromatous plaque/lesion located on left aortic valve cusp; the remaining cusps and the ascending aorta were intact/free from atherosclerosis. The base of the thrombus was pedunculated to an atherosclerotic plaque located on the left cusp of the aortic valve .The mass was in close approximity to the left main coronary trunk, causing intermittent occlusion of the latter.

***Corresponding author:** Hassane Abdallah, 10500 boulevard, de l'acadie 820 H4N2V4, Montreal, Canada, Tel: +14389893555; Fax: +15143383130; E-mail: abdallahhassane@hotmail.com

Received February 06, 2014; **Accepted** May 15, 2015; **Published** May 22, 2015

Citation: Abdallah H, Noly P, Elkhoury G. Unexpected Outcome of a Floating Thrombus in the Ascending Aorta. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 375-376 DOI:10.7438/1584-9341-11-2-5

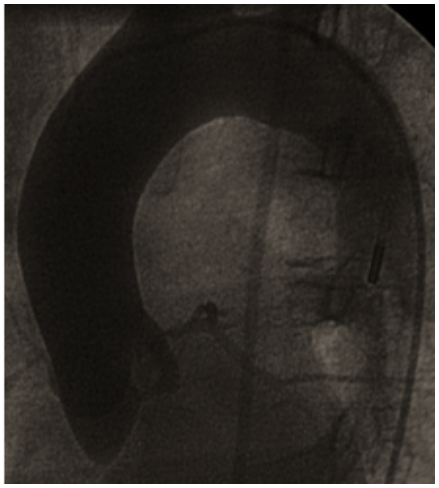

Figure 1: Left oblique view of coronary angiogram shows a filling defect was visible in the ascending aorta.

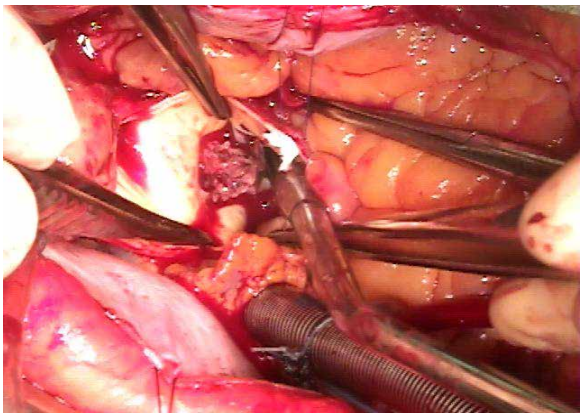

Figure 2: Left oblique view of coronary angiogram shows a filling defect was visible in the ascending aorta.

Thrombolysis has been suggested as a promising therapy for aortic thrombus [3,4] and in some cases heparin and oral warfarin have led to complete resolution in 3 months [5]. However, long-term anticoagulation for the complete resolution of a floating, friable thrombus carries unacceptable risk of partial lysis and distal embolization.

Conclusion

Despite aggressive medical and surgical treatments, consequences of a floating thrombus in the ascending aorta could be dramatic.

Walther et al. removed a thrombus from the aortic arch under hypothermic circulatory arrest, using retrograde perfusion through the femoral artery during extracorporeal circulation. In our patient, the thrombus was located in the first part of the ascending aorta. Therefore, we proceeded in routine way and we placed the arterial perfusion cannula in the proximal ascending aorta as for regular aortic valve replacement, an anterograde and retrograde cardioplegia were used, aortic root was transversely incised. After the thrombus had been excised, the aortic incision was sutured with a double suture lines.

References

1. Goueffic Y, Chaillou P, Pillet JC, Duveau D, Patra P (2002) Surgical treatment of nonaneurysmal aortic arch lesions in patients with systemic embolization. J Vasc Surg 36: 1186-1193.
2. Hausmann D, Gulba D, Bargheer K, Niedermeyer J, Comess KA, et al. (1992) Successful thrombolysis of an aortic arch thrombus in a patient after mesenteric embolism. N Engl J Med 327: 500-501.
3. Alaeddini J, Ilercil A, Shirani J (2000) Thoraco-abdominal aortic thrombosis and superior mesenteric artery embolism. Tex Heart Inst J 27: 318-319.
4. Blackshear JL, Jahangir A, Oldenburg WA, Safford RE (1993) Digital embolization from plaque-related thrombus in the thoracic aorta: identification with transesophageal echocardiography and resolution with warfarin therapy. Mayo Clin Proc 68: 268-272.
5. Walther T, Mochalski M, Falk V, Mohr FW (1996) Resection of a thrombus floating in the aortic arch. Ann Thorac Surg 62: 899-901.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Coverage Techniques in a "Crush Syndrome" Case with Extended Soft-Tissue Defect of the Shank

Laura Maria Curic*

Department of Plastic Surgery, USL 1 Umbria, Italy

Abstract

Introduction: The soft-tissue defects with bone exposal at the level of the shank raises issues in choosing the optimal surgical solution of coverage, since the locoregional muscular flaps assayed from this level are limited as number. In the introduction we present the muscular locoregional flaps most used in practice for solving the defects from the anterointernal region of the shank in the order of frequency.

Case report: We exemplify the therapeutical solutions applied in a case of crush syndrome of young men of 23 years old. A hemisolear medial flap, a flexor digitorum longus flap and a split-thickness skin graft were done. Kinethotherapy was part of the recovery treatment.

Results: Follow-up at 2 years confirmed very good functional outcomes with almost complete motor function of the lower limb.

Keywords: Lower limb trauma; Crush syndrome; Soft-tissue defect; Muscle transposition flap

Introducere

Traumatismele de strivire la nivelul extremităţilor, chiar dacă nu implică organele vitale, pot pune viaţa în pericol. "Crush syndrome" reprezintă manifestarea sistemică a distrugerii celulelor musculare şi eliberării conţinutului acestora în circulaţie, ducând la dereglarea metabolismului şi leziuni renale acute [1]. După reanimarea unui pacient cu o astfel de patologie, rămâne problema reconstrucţiei locale a defectului restant. Defectele de părţi moi cu expunere osoasă la nivelul gambei, în special cele pretibiale, rămân o provocare pentru chirurg, punând probleme în alegerea soluţiei chirurgicale optime de acoperire. Dificultatea este dată de mobilitatea redusă tegumentară a acestei regiuni şi de numărul redus de lambouri locoregionale (musculare, fasciocutanate) disponibile. De multe ori chirurgul este nevoit să apeleze la tehnici mult mai complexe de acoperire, cum ar fi lambourile perforante sau transferul liber microchirurgical [2].

Prezentare de caz

Prezentăm cazul unui pacient F.I., 32 ani, fără antecedente heredocolaterale şi personale semnificative, fumător cronic, internat în regim de urgenţă, acesta fiind victima unui traumatism major prin strivire provocat de căderea unei greutăţi cu masa de 3 tone de la înălţimea de aproximativ 1 m pe gamba dreaptă a acestuia. Prezenţa sindromului de strivire ("crush syndrome") presupune instalarea în timp relativ scurt a devitalizării musculare şi edemului important al gambei însoţit de semne ale stării de şoc traumatic şi hemoragic, ceea ce impune tratarea pacientului în etape, de către o echipă mixtă alcătuită din medici de ATI, ortopezi şi chirurgi plasticieni.

Prima etapă a fost reprezentată de primele 4 zile de internare ale pacientului în serviciul de ATI în vederea stabilizării hidroelectrolitice şi metabolice a acestuia şi prevenţia instalării complicaţiilor secundare stării de şoc. Examenul radiografic efectuat a evidenţiat prezenţa unei fracturi tip III C în 1/3 distală a gambei dreapte şi a unei fracturi de platou tibial extern drept. Examenul local al membrului pelvin drept a pus în evidenţă la inspecţie, după diminuarea edemului, atitudinea vicioasă a acestuia cu antepiciorul în varus, defect de părţi moi pe faţa antero-internă a gambei, cu expunerea marginii interne a tibialului anterior, a porţiunii externe a musculaturii din regiunea posterioară a gambei şi a tibiei pe toată lungimea sa cu vizualizarea focarului de fractură din 1/3 distală a gambei. Ţesuturile necrotice acopereau întreaga suprafaţă a defectului. S-a remarcat prezenţa multiplelor plăgi escoriate la nivelul regiunii posterioare a gambei şi coapsei, plagă transversală de 2 cm lungime la nivelul antepiciorului drept, sensibilitate normală şi puls prezent la pedioasă şi tibială posterioară. Mişcările din articulaţia genunchiului şi ale gleznei erau dureroase şi de amplitudine redusă. După reechilibrarea pacientului s-a intervenit chirurgical, practicându-se de către chirurgii ortopezi, reducerea sângerândă a fracturii din 1/3 distală a gambei şi stabilizarea acesteia prin montarea unui fixator extern (Figura 1), urmată de pansamente zilnice şi antibioterapie . În a 5-a zi postoperator pacientul a fost transferat în Clinica de Chirurgie Plastică şi Reconstructivă.

A doua etapă este reprezentată de următoarele 8 zile de internare a pacientului în clinica de Chirurgie Plastica, perioadă caracterizată printr-o stare generală bună a pacientului, stabil hemodinamic şi metabolic, afebril, cu tegumente şi mucoase palide. S-a intervenit chirurgical şi s-a practicat toaleta chirurgicală, excizia ţesuturilor necrotice, plastie cu lambouri musculare (hermisolear medial şi flexor lung de degete), plastie cu piele liberă despicată, drenaj, hemostază, sutură, pansament, imobilizare. S-a continuat în postoperator antibioterapia instituită pacientului după prima intervenţie chirurgicală, iar din a-2-a zi postoperator, când valoarea trombocitelor s-a normalizat s-a adaugat în schema terapeutica şi un anticoagulant .

Evoluţia postoperatorie favorabilă (stare generală bună, afebrilitate, scăderea valorilor markerilor strivirii musculare, normalizarea valorilor

***Corresponding author:** Laura Maria Curic, Department of Plastic Surgery, USL 1 Umbria, Via Luigi Angelini, 10, 06012 Città di Castello, Perugia, Italy, Tel: +39 075 862 8011; E-mail:lauracuric2004@yahoo.com

Received February 20, 2014; **Accepted** April 22, 2014; **Published** April 29, 2014

Citation: Curic LM. Coverage techniques in a "Crush Syndrome" case with extended soft-tissue defect of the shank. Journal of Surgery [Jurnalul de chirurgie]. 2015; 11(2): 377-380 DOI:10.7438/1584-9341-11-2-6 [article in Romanian]

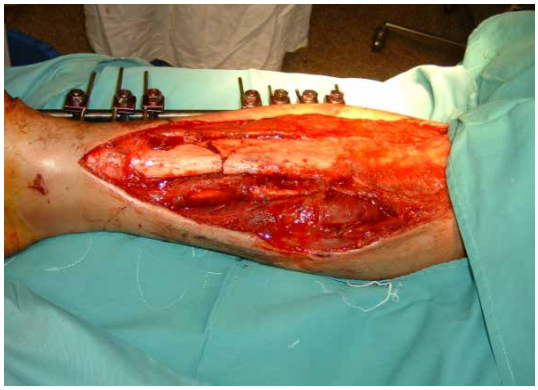

Figura 1: Defectul restant după excizia ţesuturilor devitalizate. Stabilizarea focarului de fractură tip III C cu fixator extern.

trombocitelor, hemoglobinei, hematocritului, hematiilor, pansament curat fără secreţii purulente, grefe bine integrate) a permis suprimarea drenului din loja posterioară profundă a gambei în a 6-a zi postoperator şi transferul bolnavului în Clinica de Ortopedie şi Traumatologie în a -8-a zi postoperator în vederea tratamentului chirurgical al fracturii de platou tibial extern.

Tehnica Operatorie

Intervenţia chirurgicală s-a efectuat sub rahianestezie. Abordul chirurgical s-a făcut pe la nivelul plăgii. S-au iventariat leziunile restante de strivire, s-a completat toaleta chirurgicală riguroasă cu soluţii antiseptice şi s-au excizat ţesuturile şi fasciile musculare devitalizate, necrotice prezente pe întreaga suprafaţă a plăgii (Figura 1).

Prioritatea în acest prim moment operator a fost acoperirea eficientă a focarului de fractură din ***1/3 distală a gambei***. Defectul tisular de la nivelul gambei fiind unul lung şi îngust s-a optat pentru prelevarea unui lambou hemisolear medial, care să fie avansat anterior, astfel încât să acopere regiunea medio-distală a gambei. Separarea muşchiului solear de gastrocnemian s-a făcut prin digitoclazie proximal iar distal s-a separat aponevroza solerului de cea a gastrocnemianului şi s-au secţionat fibrele musculare ale solearului în ½ medială pentru a le elibera de tendonul lui Achile. Fibrele au rămas intacte în ½ laterală, împiedicând astfel efectul de tracţiune exagerată asupra capătului distal al muşchiului (Figura 2). Partea distală eliberată s-a extras din plagă şi s-a continuat cu incizia fasciei intermusculare care separă compartimentul superficial al gambei de cel profund. Incizia acestei fascii în porţiunea laterală a permis evacuarea şi drenarea ulterioară a hematomului din loja profundă. Secţionarea ei în porţiunea medială s-a făcut cu grijă pentru a evita lezarea pachetului vasculo-nervos tibial posterior integru. S-a evidenţiat astfel lipsa totală a muşchiului flexor lung de degete în ***1/3 medie a gambei.*** S-a optat pentru utilizarea ½ inferioare a muşchiului ca lambou pediculat distal, utilizat frecvent în practică pentru acoperirea defectelor din porţiunea medio-distală a tibiei (Figura 2). Această ***particularitate a cazului*** **a** evitat utilizarea unui lambou de flexor lung de haluce, care, deşi, posedă un arc de rotaţie mai mare decât al flexor lung de degete, are dezavantajul că sacrifică artera peronieră. S-a continuat eliberarea feţei anterioare şi mediale a muşchiului solear până la ½ distanţei dintre maleola internă şi articulaţia genunchiului,locul de emergenţă a pediculul vascular secundar al solearului, care derivă din tibiala posterioară. Acesta s-a lasat pe loc, reprezentând de fapt atât pediculul vascular cât şi punctul de rotaţie al lamboului hemisolear medial inferior, care rotat, va acoperi defectul din porţiunea medio-distală (Figura 2). Pentru a favoriza avansarea anterioară a porţiunii superioare a muşchiului s-au practicat mai multe incizii longitudinale la nivelul aponevrozei lui superficiale şi secţionarea în evantai a ½ mediale a inserţiei proximale a muşchiului (Figura 3). Astfel acesta a acoperit ½ internă a gambei în porţiunea sa medie. S-a eliberat tibialul anterior de tibie, medial şi de ceilalţi muşchi ai compartimentului anterior al gambei, lateral şi s-a avansat anterior corpul muşchiului pentru a acoperi ½ externă a porţiunii medii şi distale a gambei (Figura 3). Nu s-a eliberat distal acest muşchi pentru a nu provoca un deficit funcţional în postoperator. S-au suturat solearul şi flexor lung de degete la tibialul anterior şi musculatura astfel avansată a fost acoperită ulterior cu grefă de piele liberă despicată prelevată de la nivelul regiunii antero-laterale a coapsei homolaterale (Figura 3 and 4).

În acest moment operator a rămas neacoperită ***1/3 proximală a gambei*** (Figura 3). Având în vedere existenţa la acest nivel a unor resturi tisulare suprajacente gambei, s-a acoperit zona cu o grefă de piele liberă despicată prelevată din aceeaşi regiune cu cea utilizată pentru acoperirea musculaturii (Figura1D). ***Această a doua particularitate a cazului*** ne-a oferit şansa evitării utilizării unui lambou gastrocnemian, cap medial, decizie care ar fi provocat în postoperator un dezechilibru funcţional la nivelul gambei.

După sutură şi pansament s-a imobilizat membrul pelvin pe o atelă Kramer posterioară. S-a interzis fumatul. După rezolvarea fracturii de platou tibial extern şi îndepărtarea atelei la 4 săptămâni şi a fixatorului extern la 3 luni, pacientul a urmat un program intens de kinetoterapie şi a fost urmărit ambulatorial periodic. Follow –up la 2 ani a pus în evidenţă un rezultat funcţional bun cu posibilitatea mersului în parametri normali.

Discuţii

Tehnicile de acoperire a defectelor de părţi moi cu expunere osoasă la nivelul gambei sunt reprezentate de lambourile locoregionale sau lambourile prelevate de la distanţă (transfer liber microchirurgical).

Lambourile locoregionale disponibile sunt *lambourile fasciocutanate, lambourile musculare* şi *lambourile pediculate perforante.*

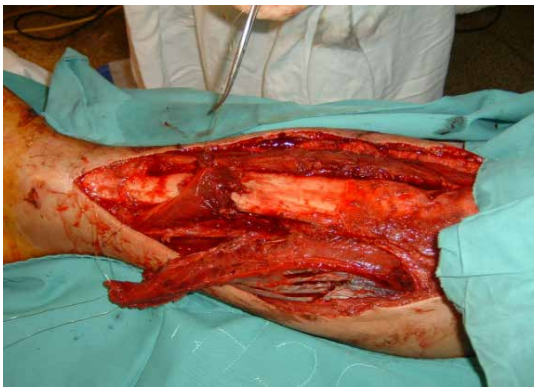

Figura 2: Lambou hemisolear medial inferior şi lambou muscular distal flexor lung de degete.

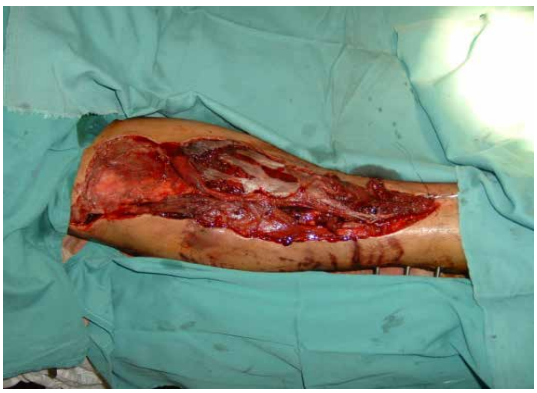

Figura 3: Incizii longitudinale ale aponevrozei superficiale a muşchiului solear şi avansarea anterioară a muşchiului tibial anterior.

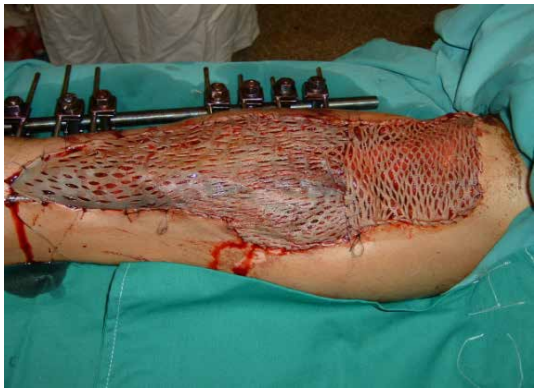

Figura 4: Plastie cu piele liberă despicată.

Lambourile fasciocutanate sunt reduse numeric datorită mobilităţii tegumentare reduse a acestei regiuni.

Cele mai utilizate ***lambouri musculare locoregionale*** pentru soluţionarea defectelor din regiunea antero –internă a gambei sunt, în ordinea frecvenţei:

1) **Lamboul gastrocnemian**

Gastrocnemianul este probabil cel mai utilizat muşchi pentru acoperirea gambei şi a genunchiului. Fiecare cap al muşchiului (medial şi lateral) poate fi mobilizat separat, fiecare posedând un pedicul vasculo-nervos propriu (artera surală) [3,4]. Capul medial permite acoperirea cu uşurinţă a 1/3 proximale a tibiei şi faţa antero-internă a genunchiului [3,4]. Prin creşterea arcului de rotaţie al muşchiului (incizii transverse multiple ale aponevrozei de pe faţa profundă a muşchiului şi secţionarea inserţiei proximale a tendonului) acesta poate acoperi defecte situate în 1/3 superioară a 1/3 medii a gambei [3,4].

2) **Lamboul solear**

Muşchiul solear se găseşte superficial în compartimentul posterior al gambei şi de cele mai multe ori nu este lezat în fracturile deschise ale 1/3 medii ale tibiei. Acoperirea cu ajutorul întregului muşchi este indicată în cazul unor defecate scurte şi ample din 1/3 medie a gambei , în timp ce acoperirea cu hemisolear medial e posibilă prin avansarea anterioară a lamboului şi este indicată în cazul unor defecte lungi şi înguste care interesează cresta tibială [3]. Lamboul solear cu bază distală nu este indicat în acoperirea 1/3 distale a gambei datorită existenţei a numeroase variaţii ale pediculului distal minor [4].

3) **Lambourile bazate pe flexorii gambei (flexorul comun de degete şi flexor lung de haluce)**

Aceste tipuri de lambouri musculare prezintă interes în acoperirea defectelor modeste ale 1/3 distale ale gambei [3]. Numai o ½ din muşchi poate fi prelevată şi folosită ca lambou pentru că cealaltă ½ e indispensabilă pentru a menţine viabilitatea acestuia [3,5]. Jumătatea inferioară a FLD (flexor lung de degete) (cea care se prelevă frecvent) se găseşte în raport intim cu tibia, cea ce face ca vascularizaţia ei să fie uşor influenţată de traumatism [3]. Acest muşchi este utilizat în special pentru acoperirea defectelor din porţiunea medio - distală a tibiei [3,5]. De cele mai multe ori FLH (flexor lung haluce) nu este lezat în traumatismele anterioare ale tibiei. Din acest motiv este mai frecvent utilizat decât FLD, are o rază de rotaţie amplă, dar are dezavantajul că pentru prelevarea lui trebuie sacrificată una din arterele principale ale gambei (artera peronieră) [3]. Este utilizat pentru acoperirea porţiunii infero-distale a tibiei [3,5].

4) **Lambourile bazate pe extensorii gambei (tibial anterior şi extensor lung de haluce)**

Aceste tipuri de lambouri musculare se utilizează mai puţin decât lambourile bazate pe musculatura flexoare. Prezintă interes pentru acoperirea unor defecte înguste şi lungi de la nivelul crestei tibiale, a cărei acoperire nu se poate face cu lambouri musculare flexoare [3]. Extensorul lung de haluce este indicat pentru acoperirea 1/3 distale a crestei tibiale, prin rotaţia ½ distale a muşchiului [3,5]. Lamboul muscular tibial anterior este un lambou puţin utilizat, în special datorită faptului că este un muşchi de mici dimensiuni şi inexpandabil [6]. Tibialul anterior (T.A.) este utilizat în special pentru acoperirea 1/3 medii a crestei tibiale [6], prin avansarea corpului muscular [3,5]. Funcţia acestui muşchi este vitală şi prelevarea lui duce la un deficit funcţional important. În plus, arcul de rotaţie al ½ distale este foarte redus. Din acest motiv cel mai bun mod de utilizare a acestui muşchi este avansarea anterioară a marginii corpului musular pentru a acoperi defecte lungi şi înguste ale crestei tibiale [6]. Acest procedeu evită instalarea unui deficit funcţional în postoperator, tibialul anterior fiind cel mai puternic flexor dorsal al piciorului, adductor şi rotator intern al acestuia [3].

Lambourile pediculate perforante, bazate pe artera tibială posterioară şi pe artera peronieră reprezintă o modalitate de tratament alternativă sigură, simplă şi fiabilă pentru acoperirarea defectelor pretibiale [2].

Lambourile liber transferate (lambouri fasciocutanate perforante, lambouri musculare) sunt frecvent utilizate pentru reconstrucţia posttraumatică a membrului pelvin [7]. Utilizarea unuia sau a celuilalt tip de lambou a rămas o problemă controversată în literatură [7]. Scopul principal al microchirurgiei reconstructive este obţinerea unui rezultat estetic şi funcţional optim cu o morbiditate minimă a regiunii donatoare [8]. Astfel, majoritatea articolelor retrospetive pledează pentru siguranţa mai mare a lambourilor libere fasciocutanate în reconstrucţia posttraumatică a membrului inferior [7,9], în timp ce alţii pledează pentru transferul liber de latissimus dorsi [10,11], cu o rată de supravieţuire a acestuia de 95%. Transferul liber microchirurgical presupune, însă: 1) o durată mare a intervenţiei datorită microanastomozelor vasculare, 2) necesitatea unei aparaturi de specialitate (microscop operator, instrumentar microchirurgical, material de sutură specific microanastomozelor) şi a personalului specializat şi antrenat cu aceste tehnici, 3) riscuri mai mari ale viabilităţii lambourilor şi 4) o rată de succes mai mică.

Concluzii

Metoda de reconstrucţie prezentată anterior reprezintă o metodă fiabilă de rezolvare a defectelor tisulare de la nivelul gambei, metodă care permite, atunci când este posibil, evitarea unor transferuri libere.

Avantajele metodei utilizate sunt: limitarea deficitului funcţional restant prin mobilizarea musculaturii din loja postero-internă combinată cu cea din loja antero-internă a gambei; oferă o bună masă musculară pentru acoperirea focarului de fractură; musculatura ofera un suport bun pentru grefare, cu integrare bună a acestora.

Dezavantajul tehnicii constă în disecţia minuţioasă şi deloc facilă a lambourilor amintite, cu secţionarea masei musculare care provoacă o sângerare secundară importantă, ceea ce impune de fiecare dată drenarea cavităţilor restante după transpoziţia musculaturii.

Conflict de interese

Autoarea nu declară niciun conflict de interese.

Bibliografie

1. Genthon A, Wilcox SR (2014) Crush syndrome: a case report and review of the literature. J Emerg Med 46: 313-319.
2. Shin IS, Lee DW, Rah DK, Lee WJ (2012) Reconstruction of pretibial defect using pedicled perforator flaps. Arch Plast Surg 39: 360-366.
3. Alain C (1996) Masquelet, Alain Gilbert. Atlante dei Lambi Cutanei nella Ricostruzione degli Arti. Edizione italiana a cura di M.Pizzetti. Presentazione di L. Perugia.Antonio Delfino Editore 118-162.

4. Masquelet AC, Mc Cullough C, Tubiana R (1996) Atlante delle Vie Chirurgiche d'Accesso dell'Arto Inferiore. Edizione italiana a cura di M .Pizzetti. Presentazione di L.Perugia. Antonio Delfino Editore.

5. Mathes J, Nahai F (1997) Reconstructive surgery Principles, Anatomy &Technique. United States of America; Philadeplia: Churchill Livingstone Inc.

6. Panse N, Sahasrabudhe P, Pande G, Chandanwale A, Dhongde R, et al. (2012) The split tibialis anterior muscle flap-A simple solution for longitudinal middle third tibial defects. Indian J Plast Surg 45: 53-57.

7. Sofiadellis F, Liu DS, Webb A, Macgill K, Rozen WM, et al. (2012) Fasciocutaneous free flaps are more reliable than muscle free flaps in lower limb trauma reconstruction:experience in a single trauma center. J Reconstr Microsurg 28: 333-340.

8. Ninkovic M, Voigt S, Dornseifer U, Lorenz S, Ninkovic M (2012) Microsurgical advances in extremity salvage. Clin Plast Surg 39: 491-505.

9. Chan JK, Harry L, Williams G, Nanchahal J (2012) Soft-tissue reconstruction of open fractures of the lower limb: muscle versus fasciocutaneous flaps. Plast Reconstr Surg 130: 284e-295e.

10. Knobloch K, Herold C, Vogt PM (2012) [Free latissimus dorsi flap transfer for reconstruction of soft tissue defects of the lower extremity]. Oper Orthop Traumatol 24: 122-130. [Article in German]

11. Hwang KT, Youn S, Kim JT, Lee SH, Ng SW, et al. (2012) Use of latissimus dorsi flap pedicle as a T-junction to facilitate simultaneous free fibular flap inset in lower extremity salvage. J Plast Reconstr Aesthet Surg 65: 517-520.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Anaplastic Thyroid Carcinoma or Thyroid Metastasis from Cholangiocarcinoma? A Case Report

Lidia Ionescu[1], Radu Dănilă[1]*, Mihaela Blaj[2], Mihai Savin[3], Carmen Vulpoi[4], Delia Ciobanu[5] and Daniel Timofte[1]

[1]*Department of General Surgery, University of Medicine and Pharmacy "Gr.T. Popa" Iasi, "St. Spiridon" Hospital, Iasi, Romania*
[2]*Intensive Care Unit, University of Medicine and Pharmacy "Gr.T. Popa" Iasi, "St. Spiridon" Hospital, Iasi, Romania*
[3]*Department of Radiology, "St. Spiridon" Hospital, Iasi, Romania*
[4]*Department of Endocrinology, University of Medicine and Pharmacy "Gr.T. Popa" Iasi, "St. Spiridon" Hospital, Iasi, Romania*
[5]*Department of Pathology, University of Medicine and Pharmacy "Gr.T. Popa" Iasi, "St. Spiridon" Hospital, Iasi, Romania*

Abstract

Anaplastic thyroid carcinoma presents as an extremely localy invasive neck mass while metastases in the thyroid are most commonly described as small, indolent, solitary nodules usually originating from kidney, breast, lungs and skin tumors. We report the case of a 74-year old male patient illustrating the difficulties of differential diagnosis between an anaplastic thyroid carcinoma and a thyroid metastasis of a peripheral cholangiocarcinoma in a cirrhotic patient diagnosed and operated for a locally advanced thyroid tumor. The history, clinical and imagistical features strongly pleaded for the diagnosis of anaplastic thyroid carcinoma presumably with liver metastases, also supported by the rapid recurrence following total thyroidectomy. Immunohistochemical tests showed a malignant carcinomatous proliferation with anaplastic prophile. Positive immunochemical staining for cytokeratin AE1/AE3, CK7 and negative CK20, AFP, CD15, CD30, CD5, TTF1 and thyroglobulin directed the possible diagnosis toward a secondary thyroid tumor from a peripheral cholangiocarcinoma. Immunohistochemical tests showed a malignant carcinomatous proliferation with anaplastic prophile. Positive immunochemical staining for cytokeratin AE1/AE3, CK7 and negative CK20, AFP, CD15, CD30, CD5, TTF1 and thyroglobulin directed the possible diagnosis toward a secondary thyroid tumor from a peripheral cholangiocarcinoma. A CT-guided percutaneous hepatic punction biopsy was planned but the patient presented an ischemic stroke with fatal outcome. In conclusion, in spite of surgical treatment the rapid recurrent thyroid cancer either primary or metastatic had a poor prognosis with fatal outcome mainly in the presence liver cirrhosis and cardio-vascular co-morbidities.

Keywords: Thyroid; Anaplastic carcinoma; Metastasis; Cholangiocarcinoma; Surgery

Introduction

Metastatic tumors in the thyroid gland occur in as many as 24% of subjects when examined at autopsy and most commonly primary tumors are located in the kidney, breast, lung, and malignant melanoma of the skin. Generally, a metastatic tumor in the thyroid gland presents as a solitary nodule that may be the initial evidence of disease or the first presentation of recurrent disease but more often there is a widespread metastatic disease present and the manifestations in the thyroid gland are clinically unimportant. Anaplastic carcinoma describes an undifferentiated malignancy derived from more well-differentiated thyroid follicular epithelium. In contrast to the generally indolent nature of differentiated thyroid carcinoma, anaplastic carcinoma represents one of the most aggressive human neoplasms, with a disease-specific mortality of at least 90%. Occasionally, it may be difficult to determine if the specimen represents metastatic disease or if it is originating from the thyroid gland, such as an anaplastic thyroid carcinoma [1].

We present a case illustrating the difficulties of differential diagnosis between an anaplastic thyroid carcinoma with liver metastases and a thyroid metastasis of a peripheral cholangiocarcinoma in a cirrhotic patient diagnosed and operated for a locally advanced thyroid tumor.

Case Report

A 74-year old male patient was referred to surgery from the endocrinology department for a thyroid tumor with compression signs. The patient was previously diagnosed with C virus liver cirrhosis, type 2 diabetes mellitus, arterial hypertension, ischemic heart disease and anemia. The patient reported a 4 month history of fatigue and weight loss and more recently (2 weeks) neck pain and enlargement of the anterior cervical region with dyspnoea and disphagia. A cervical lymph node biopsy previously performed in the ENT department revealed just a chronic nonspecific lymphadenitis. The physical examination showed a large, irregular, hard and fixed tumor of the right thyroid lobe with multiple laterocervical lymphadenopathies. The lab tests showed a normal thyroid function and calcitonine level, AFP, CEA and CA19-9 within normal range. Ultrasound of the thyroid described an extensive tumor of the right lobe with bilateral cervical lymphadenopathies. Thyroid scintigraphy revealed multiple areas of hypo and affixation of 99m Tc in both lobes. Computer tomography (CT) of the neck revealed the thyroid gland almost completely replaced by a 64/89/85 mm solid, inhomogeneous tumor, predominantly developed in the right lobe and deviating trachea, larynx and hypopharinx to the left. Right internal jugular vein was thrombosed without demarcation limit from the tumor and multiple laterocervical and superior mediastinum lymphadenopathies were present (Figure 1).

FNAC (fine needle aspiration cytology) showed a suspicious cytology-Bethesda V. Routine preoperative laryngoscopy was normal.

***Corresponding author:** Radu Danila, 3rd Surgical Unit, "St. Spiridon" Hospital, Bd. Independentei, No 1, 700111, Iasi, Romania, Tel: +40 (0) 232 217 781; E-mail: nr_danila@yahoo.com

Received March 13, 2014; **Accepted** May 20, 2014; **Published** May 27, 2014

Citation: Ionescu L, Dănilă R, Blaj M, Savin M, Vulpoi C, et al. Anaplastic Thyroid Carcinoma or Thyroid Metastasis from Cholangiocarcinoma? A Case Report. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 381-383 DOI:10.7438/1584-9341-11-2-7

Under the suspicion of thyroid malignancy the patient was operated and a total thyroidectomy with lymphadenectomy of the central and lateral compartments of the neck was performed. The frozen section exam revealed a massive malignant infiltration with marked pleomorphism. The final pathology described a massive malignant infiltration with marked pleomorphism and large areas of necrosis (Figure 2).

Immunohistochemical tests were performed but could not distinct between an anaplastic carcinoma with lymphoepithelial aspect and a metastatic carcinoma from a cholangiocarcinoma (Table I).

After 2 months he was readmitted with signs of local recurrence. CT showed a 90/86/82 mm tumor mass in right thyroid space with necrotic areas which compressed the carotid sheath, larynx, and esophagus. CT of the abdomen showed multiple nodules in the VI, VII, VIII segments of the liver, suggestive for liver metastases or peripheral cholangiocarcinoma. An exploratory laparoscopy with liver biopsy was intended but the patient suffered an ischemic stroke with grade II coma (extensive subarahnoidian hemorrhage.) and deceased 10 ten days later in the intensive care unit.

Discussion

The biological behaviour, clinical and imagistical features should normally allow an obvious distinction between an undifferentiated primary thyroid carcinoma and a secondary tumor in the gland. Whereas anaplastic thyroid carcinoma presents as an extremely localy invasive neck mass, metastases in the thyroid are most commonly described as small, indolent, solitary nodules. In a large series from the Mayo Clinic, the average size of the thyroid metastatic nodules was 3 cm [2]. The incidence of thyroid metastases secondary to any type of primary tumor is reported between 1.9-9.5% and surgical resection of the thyroid metastasis ranges between 0.02 and 1.4% [3-8]. The survival is poor, ranging between 1 and 12 months [9-12]. Most frequently the primary tumor is renal and only two cases of thyroid metastases secondary to cholangiocarcinoma were reported on Pubmed database [3,4].

In our case, the history, clinical and imagistical features strongly pleaded for the diagnosis of anaplastic thyroid carcinoma presumably with liver metastases, also supported by the rapid recurrence following total thyroidectomy. Immunohistochemical tests showed a malignant carcinomatous proliferation with anaplastic prophile. Positive immunochemical staining for cytokeratin AE1/AE3, CK7 and negative CK20, AFP, CD15, CD30, CD5, TTF1 and thyroglobulin raised the suspicion of a secondary thyroid tumor from a peripheral cholangiocarcinoma. This hypothesis was also supported by the diagnosis of macronodular liver cirrhosis and presumably a neoplasm in the segment VII. Although the tumoral markers for hepatoma or cholangiocarcinoma were within normal limits, immunohistochemistry tests raised the possibility of a metastatic thyroid tumor from a cholangiocarcinoma. Unfortunately, the patient presented a fatal stroke in the day when he was listed for a CT-guided percutaneous hepatic punction biopsy. This exploration would have enabled us to delineate with accuracy the relationship between the thyroid tumor and the liver nodules.

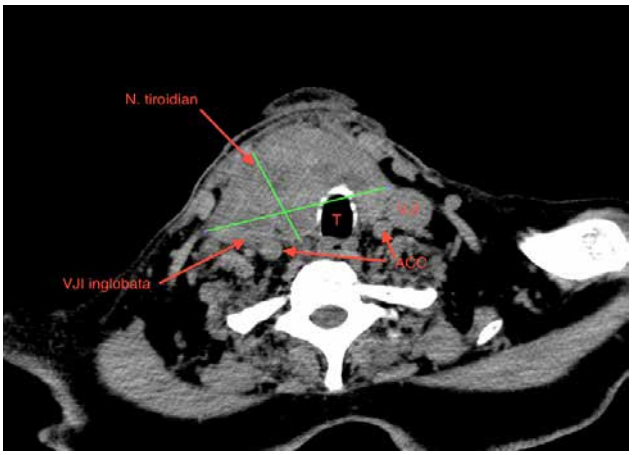

Figure 1: CT scan: Thyroid Gland Tumor with Internal Jugular Vein Invasion.

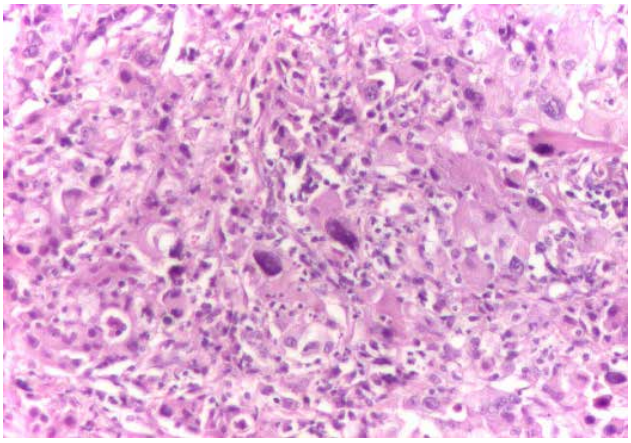

Figure 2: The Microscopic Examination (HE 100x): Pleomorphic Massive Malignant Infiltration.

Table I: Immune Histochemistry Tests.

Citokeratin CK AE1/AE3, CK7	Intense Positive
Citokeratin CK20	Negative
Thyroglobulin (TG), TTF1	Negative
AFP	Negative
Chromogranin, synaptophisin	Negative
LCA	Intense Positive in peritumoral lymphoid tissue but negative in the tumoral cells
CD15, CD30	Negative
S100	Negative

Conclusion

In conclusion, despite of surgical treatment the rapid recurrent thyroid cancer either primary or metastatic had a poor prognosis with fatal outcome mainly in the presence liver cirrhosis and cardio-vascular co-morbidities.

Conflict of interests

Authors have no conflict of interests to disclose

References

1. Burman KD, Ringel MD, Shmookler BM (2006) Miscellaneous and Unusual Types of Thyroid Cancer. In: Wartofsky L, Van Nostrand D (edt), Thyroid Cancer, A Comprehensive Guide to Clinical Management, Humana Press Totowa, New Jersey.
2. Hegerova L, Griebeler ML, Reynolds JP, Henry MR, Gharib H (2013) Metastasis to the thyroid gland: report of a large series from the Mayo Clinic. Am J Clin Oncol.
3. Czech JM, Lichtor TR, Carney JA, van Heerden JA (1982) Neoplasms metastatic to the thyroid gland. Surg Gynecol Obstet 155: 503-505.
4. Ericsson M, Biorklund A, Cederquist E, Ingemansson S, Akerman M (1981) Surgical treatment of metastatic disease in the thyroid gland. J Surg Oncol 17: 15-23.
5. Lin JD, Weng HF, Ho YS (1998) Clinical and pathological characteristics of secondary thyroid cancer. Thyroid 8: 149-153.
6. Papi G, Fadda G, Corsello SM, Corrado S, Rossi ED, et al. (2007) Metastases to the thyroid gland: prevalence, clinicopathological aspects and prognosis: a 10-year experience. Clin Endocrinol (Oxf) 66:565-571.
7. Nakhjavani MK, Gharib H, Goellner JR, van Heerden JA (1997) Metastasis to the thyroid gland. A report of 43 cases. Cancer 79: 574-578.

8. Mirallie E, Rigaud J, Mathonnet M, Gibelin H, Regenet N, Hamy A (2005) Management and prognosis of metastases to the thyroid gland. J Am Coll Surg 200(: 203-207.

9. Nixon IJ, Whitcher M, Glick J, Palmer FL, Shaha AR, et al. (2011) Surgical management of metastases to the thyroid gland. Ann Surg Oncol 8: 800-804.

10. Nakamura K, Nozawa K, Aoyagi Y, Ishihara S, Matsuda K, et al. (2011) A case report of thyroid gland metastasis associated with lung metastasis from colon cancer. Tumori 97: 229-232.

11. Park MH, Cho JS, Lee JS, Kim HK, Yoon JH (2012) Thyroid gland metastasis arising from primary liver cholangiocarcinoma: The first case report involving surgical operation. Int J Surg Case Rep 3: 78-81.

12. Bae WK, Shim HJ, Choi YD, Kim JW, Cho SH, et al. (2009) Severe hypothyroidism induced by thyroid metastasis of cholangiocarcinoma. Cancer Res Treat 41: 56-58.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Surgical Approach of Cervical Cancer Liver Metastases: Case Report

Nicolae Bacalbașa[1] and Irina Balescu[2]

[1]University of Medicine and Pharmacy "Carol Davila" Bucuresti, Romania
[2]"Ponderas" Hospital, Bucuresti, Romania

Abstract

Cervical cancer is an aggressive malignancy with a high tendency of invasion of the surrounding organs and also with capacity to give birth to metastases on both lymphatic and hematogenous routes. Cases who present distant metastases at the moment of diagnosis are generally referred to the medical oncologist than to the surgeon; however increasing reports on the benefit of liver surgery in non-colorectal non-neuroendocrine liver metastases have decreased the general reluctance to perform radical visa surgery on such cases. We present the case of a 53 years old female diagnosed with cervical cancer and liver metastases in which a radical resection was performed with good oncologic outcomes.

Keywords: Uterine cervical cancer; Radiation therapy; Liver metastases; Liver resection

Introduction

Cervical cancer still represents a major health problem with a reported annual incidence of 371.000 cases and a death rate of 190.000 women/year [1]. These data reflect the presence of a tumor with an aggressive behavior. The most important patterns of spread are local – through direct invasion into the surrounding viscera, lymphatic – responsible for the apparition of pelvic and para-aortic lymph node metastases and hematogenous. In patients with advanced cervical cancer metastases to the para-aortic lymph nodes they are usually secondary to those located in the pelvis, the frequency of positive para-aortic lymph nodes increasing with FIGO stage from 5% in FIGO stage IB1 to 30% in FIGO stage III [2,3]. Skip metastases direct to the inter aortico-caval lymph nodes with negative pelvic nodes are very rare. When studying the orderly process of nodal metastases in para-aortic lymph nodes there are studies which support a discontinuous metastatic dissemination. Gil Moreno et al. demonstrated that negative inframesenteric aortic lymph-nodes can be associated with positive infrarenal lymph nodes in about one third of patients with advanced cervical cancer [4].

When it comes to the presence of distant metastases by hematogenous spread, things are not so well standardized. The main locations of hematogenous metastases are bones, liver and lungs. The frequency of liver metastases reaches almost 3% and sometimes represents a contraindication for surgery. In cases with isolated liver metastases surgery might be tempted with good results [5].

Case Report

The 53 years old female presented for vaginal bleeding and pelvic pain. The local exam showed a large cervical tumor developed anteriorly, which was biopsied; the histopathological findings revealed a poor differentiated squamous cervical cancer. The patient was addressed to the oncology clinic and brachytherapy and external beam radiation therapy were performed. The computed tomography prior to surgery showed decrease in dimensions of the cervical tumor with a slight discontinuity of the demarcation line between the urinary bladder and the tumor, large pelvic and para-aortic lymph node metastases with a maximum diameter of 2.5 cm and a liver metastasis located in the 7th hepatic segment according to Couinaud's classification (Figures 1-3). Surgery was performed one month after completing the neo-adjuvant treatment. Intraoperatively an adherent to the urinary bladder tumor was found but with no tumoral invasion, so a radical hysterectomy en bloc with bilateral adnexectomy was performed. Lymphadenectomy included dissection of the pelvic groups – obturatory fossa, iliac group and abdominal ones- para-aortic groups – from the aortic bifurcation to duodenum (Figures 4-6). The inferior mesenteric artery was identified and completely dissected. Three liver metastases were also found in segments V, VI, VIII and were resected (Figure 7). The postoperative course was uneventful, the patient being discharged in the 8th postoperative day. Histopathological findings showed a moderate to poor differentiated squamous cell carcinoma.

Discussions

Although there are screening tests largely used worldwide in order to detect cervical cancer in an early stage of the disease, an important number of patients are diagnosed in advanced stages [1]. Once the patient is diagnosed with an advanced malignancy aggressive surgical approach represents the only way a good control of the disease can be obtained [6]. The aggressive biology of this tumor is demonstrated both by the local invasion of the surrounding organs and the capacity to metastazise through lymphatic or hematogenous ways. Local invasion of the surrounding viscera takes place in the moment when the compartimental borders, which are in fact natural barriers in front of the neoplastic process, are destroyed. When talking about cervical cancer the most important compartimental borders are represented by the peritoneal reflections from the urinary bladder to the uterus anteriorly and the reflection from the anterior rectal wall to the posterior surface of the uterus posteriorly [7]. In the moment when these barriers are destroyed tumoral invasion in the surrounding organs appears and multivisceral resections are needed in order to obtain a good local control of the disease [6,7]. In our case preoperative computed tomography showed a zone of possible tumoral invasion in the posterior wall of the urinary bladder but intraoperatively this was not found.

***Corresponding author:** Nicolae Bacalbașa, Dimitrie Racoviță Street, No 2, Bucuresti, Romania, Tel: +40 (0) 723 54 04 26; E-mail:nicolae_bacalbasa@yahoo.ro

Received October 2, 2014; **Accepted** November 10, 2014; **Published** November 17, 2014

Citation: Bacalbașa N, Balescu I. Surgical Approach of Cervical Cancer Liver Metastases: Case Report. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 385-387 DOI:10.7438/1584-9341-11-2-8

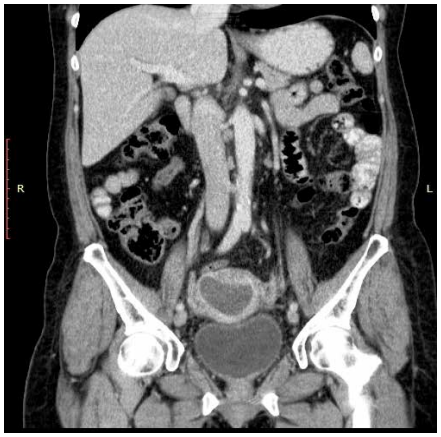

Figure 1: Large Inter-Aortico –Caval and Pelvic Adenopathies.

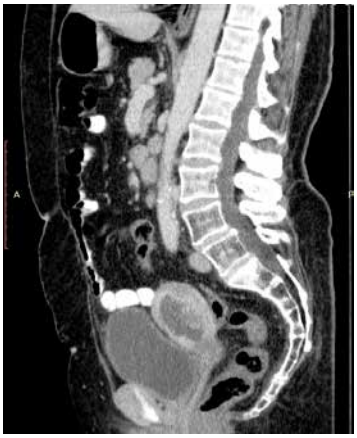

Figure 2: Cervical Tumor in close contact with the Posterior wall of the Urinary Bladder associated with Pre-Aortic Lymph Node.

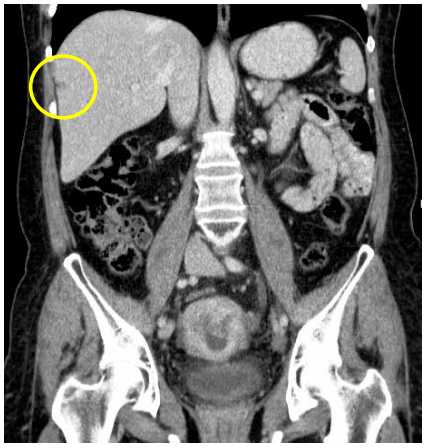

Figure 3: Liver Metastasis.

The lymphatic route represents the second pattern of spread in cervical cancer. Studies have shown that the presence of lymph node metastases, particularly para-aortic lymph node involvement associated with tumor stage is the most important prognostic factors [4,8]. Classically it was considered that the process of dissemination using the lymphatic channels is an orderly one, from the pelvic lymph nodes to the ipsilateral common iliac, inframesenteric and infrarenal aortic nodes. Recent studies have demonstrated that this pattern of spread isn't respected in all situations, cases with negative inframesenteric nodes and positive infra-renal nodes being reported [4,9]. In our case large lymph nodes were found both in pelvis and on the whole antero-lateral surface of the abdominal aorta from its' bifurcation to the duodenum. The important dimensions of these lymphadenopathies and the close contact with the great vessels makes them hard to be controlled by other methods than surgery. While pelvic node metastases can receive doses of 50-60 Gy to obtain an efficient control of the metastases, bulky para-aortic lymph nodes cannot be controlled through this procedure [10]. In cases presenting enlarged lymph nodes (>2 cm) an adequate dose of radiation to sterilize these tumoral masses would be unacceptable for the surrounding vessels or the spinal cord. Based on this concept important studies recommend surgery in order to remove bulky macroscopically positive lymph nodes associated with adjuvant chemo-irradiation for the possible remnant micrometastases [11-13].

While in cases with multiple bulky lymph nodes the most efficient therapeutic protocol is widely accepted as being aggressive surgical

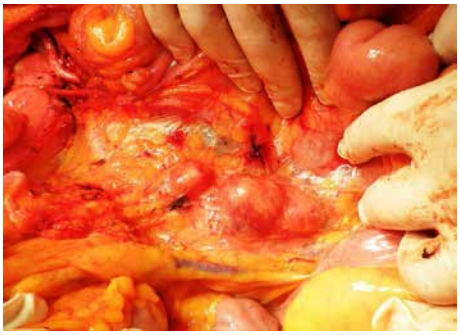

Figure 4: Large Preaortic Adenopathies situated from the Duodenum to the Aortic Bifurcation.

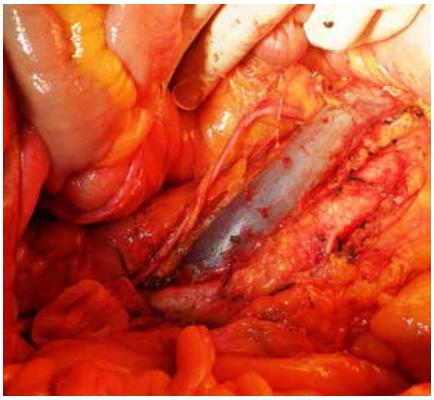

Figure 5: The final aspect after Inter-Aortico-Caval Lymph Node dissection.

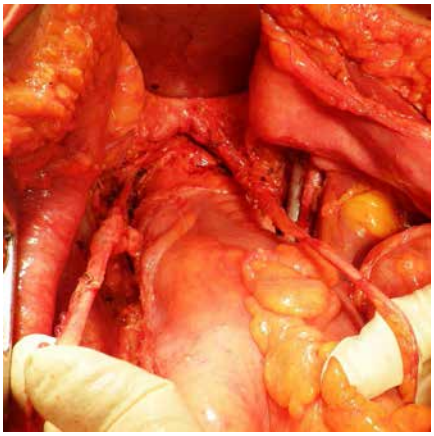

Figure 6: The final aspect of Pelvic dissection: the 2 Ureters and the Urinary Bladder are completely dissected.

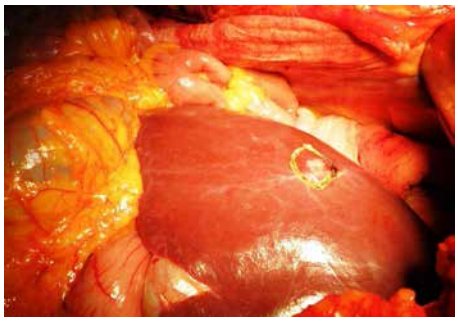

Figure 7: Liver Metastasis.

resection, when it comes to liver metastases from cervical cancer, things are not so clear. One of the main reasons for this fact is the lack of large randomized studies and the small number of patients included in the existing studies.

Liver metastases from cervical cancer are rarely seen, being reported in 2-3% of cases [5]. Based on the success reported in treating liver metastases from colo-rectal cancer or neuro-endocrine tumors, some authors tried to evaluate which is the role of liver resection in gynaecologic malignancies [14-17]. Main studies included patients with liver metastases from breast cancer or ovarian cancer and a benefit in terms of survival was demonstrated [17,18]. When searching the effect of surgery on liver metastases originating from other gynaecologic malignancies only few cases are reported (ranging 1-7 cases per series) [19-21].

Chi et al. evaluated the role of liver resection in metastatic gynaecologic malignancies on a group of 12 patients, with a median age of 60 years. Only 2 of the 12 cases presented metachronous liver metastases originating from cervical cancer. The conclusions of this study was that hepatectomy can be performed safely and prolong survival [20].

In a study conducted by Rene Adam et al. 45 patients with both uterine and cervical tumors were included. The rate of isolated liver metastases reached almost 71%. Most patients introduced in this study presented metachronous liver metastases, which were diagnosed after a disease free survival of 48 months. The 5 year overall survival rate was 35%, the only prognostic factor statistically significant being an R0 resection [22].

Kamel et al. reported a series of 87 patients with liver metastases from gynaecologic cancer. Only 3 cases were diagnosed with liver metastases from cervical cancer and although liver resection was performed, they reported a poorer 5 year overall survival than the cases who underwent the same type of surgery for liver metastases from ovarian cancer [23]. In our case the presence of 3 isolated liver metastases measuring between 1 and 2 cm with a perfectly normal remnant liver encouraged us to perform the 2 metastasectomies too in order to obtain an R0 resection

Conclusion

Cervical cancer remains an aggressive disease with multiple ways of spread; surgery seems to be the only way to control this lethal disease. While the therapeutic protocol for lymph node metastases is well standardized, things are not so clear established for the treatment of liver metastases. The main responsible factor for this deficit is the rarity of hepatic metastases from cervical cancer, reported series comprising few patients. Complete resection seems to be the only significant prognostic factor, although poorer rates of survival were reported (when compared to other gynaecologic malignancies). However, further studies on larger lots of patients are still needed.

Conflict of interest

Authors have no conflict of interest to disclose.

References

1. Parkin DM, Pisani P, Ferlay J (1999) Estimates of the worldwide incidence of 25 major cancers in 1990. Int J Cancer 80: 827-841.
2. Heller PB, Maletano JH, Bundy BN, Barnhill DR, Okagaki T (1990) Clinical-pathologic study of stage IIB, III, and IVA carcinoma of the cervix: extended diagnostic evaluation for paraaortic node metastasis--a Gynecologic Oncology Group study. Gynecol Oncol 38: 425-430.
3. Panici PB, Scambia G, Baiocchi G, Matonti G, Capelli A, et al. (1992) Anatomical study of para-aortic and pelvic lymph nodes in gynecologic malignancies. Obstet Gynecol 79: 498-502.
4. Gil-Moreno A, Magrina JF, Pérez-Benavente A, Díaz-Feijoo B, Sánchez-Iglesias JL, et al. (2012) Location of aortic node metastases in locally advanced cervical cancer. Gynecol Oncol 125: 312-314.
5. Gallup D, Glob. libr. women's med (2008) The Spread and Staging of Cervical Cancer. The Global Library of Women's Medicine.
6. Höckel M, Horn LC, Tetsch E, Einenkel J (2012) Pattern analysis of regional spread and therapeutic lymph node dissection in cervical cancer based on ontogenetic anatomy. Gynecol Oncol 125: 168-174.
7. Höckel M (2006) Ultra-radical compartmentalized surgery in gynaecological oncology. Eur J Surg Oncol 32: 859-865.
8. Leblanc E, Narducci F, Frumovitz M, Lesoin A, Castelain B, et al. (2007) Therapeutic value of pretherapeutic extraperitoneal laparoscopic staging of locally advanced cervical carcinoma. Gynecol Oncol 105: 304-311.
9. Michel G, Morice P, Castaigne D, Leblanc M, Rey A, et al. (1998) Lymphatic spread in stage Ib and II cervical carcinoma: anatomy and surgical implications. Obstet Gynecol 91: 360-363.
10. Fletcher GH (1984) Lucy Wortham James Lecture. Subclinical disease. Cancer 53: 1274-1284.
11. Potish RA, Downey GO, Adcock LL, Prem KA, Twiggs LB (1989) The role of surgical debulking in cancer of the uterine cervix. Int J Radiat Oncol Biol Phys 17: 979-984.
12. Cosin JA, Fowler JM, Chen MD, Paley PJ, Carson LF, et al. (1998) Pretreatment surgical staging of patients with cervical carcinoma: the case for lymph node debulking. Cancer 82: 2241-2248.
13. Kenter GG, Hellebrekers BW, Zwinderman KH, van de Vijver M, Peters LA, et al. (2000) The case for completing the lymphadenectomy when positive lymph nodes are found during radical hysterectomy for cervical carcinoma. Acta Obstet Gynecol Scand 79: 72-76.
14. Goéré D, Elias D (2008) Resection of liver metastases from non-colorectal non-endocrine primary tumours. Eur J Surg Oncol 34: 281-288.
15. Adam R, Chiche L, Aloia T, Elias D, Salmon R, et al. (2006) Hepatic resection for noncolorectal nonendocrine liver metastases. analysis of 1,452 patients and development of a prognostic model. Ann Surg 244: 524-535.
16. Reddy SK, Barbas AS, Marroquin CE, Morse MA, Kuo PC, et al. (2007) Resection of noncolorectal nonneuroendocrine liver metastases: a comparative analysis. J Am Coll Surg 204: 372-382.
17. Pockaj BA, Wasif N, Dueck AC, Wigle DA, Boughey JC, et al. (2010) Metastasectomy and surgical resection of the primary tumor in patients with stage IV breast cancer: time for a second look? Ann Surg Oncol 17: 2419-2426.
18. Thelen A, Benckert C, Jonas S, Lopez-Hänninen E, Sehouli J, et al. (2008) Liver resection for metastases from breast cancer. J Surg Oncol 97: 25-29.
19. Weitz J, Blumgart LH, Fong Y, Jarnagin WR, D'Angelica M, et al. (2005) Partial hepatectomy for metastases from noncolorectal, nonneuroendocrine carcinoma. Ann Surg 241: 269-276.
20. Chi DS, Fong Y, Venkatraman ES, Barakat RR (1997) Hepatic resection for metastatic gynecologic carcinomas. Gynecol Oncol 66: 45-51.
21. Elias D, Cavalcanti de Albuquerque A, Eggenspieler P, Plaud B, Ducreux M, et al. (1998) Resection of liver metastases from a noncolorectal primary: indications and results based on 147 monocentric patients. J Am Coll Surg 187: 487-493.
22. Adam R, Chiche L (2005) Chirurgie des me´tastases he´patiques de cancers non colo-rectaux, non-endocrines. Rapport présenté au 107e Congres francais de chirurgie 2005. Monographie de l'Association Francaise de Chirurgie. Paris
23. Kamel SI, de Jong MC, Schulick RD, Diaz-Montes TP, Wolfgang CL, et al. (2011) The role of liver-directed surgery in patients with hepatic metastasis from a gynecologic primary carcinoma. World J Surg 35: 1345-1354.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Incidental Discovery of an Esophageal Leiomyoma: Thoracoscopic Surgical Approach

Adrian Tudor[1], Ciprian Rosca[2]*, Vasile Bud[2], Cosmin Nicolescu[1], Bianca Tudor[3] and Calin Molnar[4]

[1]*University of Medicine and Pharmacy Târgu Mureş, Department M1, 1st Surgical Unit, Emergency County Hospital Târgu Mureş, Romania*
[2]*1st Surgical Unit, Emergency County Hospital Târgu Mures, Romania*
[3]*University of Medicine and Pharmacy Târgu Mureş, Department M2, Central Laboratory, Emergency County Hospital Târgu Mures, Romania*
[4]*University of Medicine and Pharmacy Târgu Mureş, Department M5, 1 st Surgical Unit, Emergency County Hospital Târgu Mureş, Romania*

Abstract

Background: Esophageal leiomyomas are the most common benign esophageal tumor, originating in the smooth muscle of the esophagus. Patients may accuse dysphagia, epigastric pain, but in 50% of cases are asymptomatic. Paraclinical exams used to highlight the esophageal tumor are esophageal and stomach barium swallow, esophagoscopy, chest CT scan, endoscopic ultrasonography. Thoracoscopic enucleation in recent years has gained many followers.

Case Report: We present herein the case of 43 years old patient admitted in our department for thoracic trauma; the CT scan revealed a tumor in the middle third of the esophagus suggestive for a leiomyoma. Upper GI endoscopy showed an extrinsic compression in the middle third of the esophagus, without mucosal lesions, and esophageal barium swallow showed a slight narrowing of the lumen at this level. Given the findings suggestive of a benign esophageal tumor, possible esophageal leiomyoma, thoracoscopic approach was chosen. We performed a thoracoscopic enucleation with uneventful postoperative follow-up. Histopathology confirmed the diagnosis of esophageal leiomyoma.

Conclusions: Thoracoscopic enucleation is a feasible method to treat esophageal leiomyomas.

Keywords: Esophageal benign tumor; Esophageal leiomyoma; Thoracoscopy; Thoracoscopic enucleation

Introduction

Benign tumors of the esophagus are rare, accounting for less than 10% of esophageal tumor [1]. In benign tumors, leiomyomas represent about two-thirds [2]. They are usually single tumors developed in the lower two thirds of the esophagus [3], originating in the smooth muscle of the esophagus. In most cases leiomyomas occur between 20 and 69 years, the peak incidence is in the fifth decade of life [4]. It occurs more frequently in men, male female ratio is 2:1. Symptoms are nonspecific, common causes are dysphagia and epigastric pain, but 50% of patients may be asymptomatic. The dimensions are between 1 and 29 cm, most of them being under 5 cm [5,6]. Surgical treatment by tumor extramucosal enucleation is a safe method in about 96% of esophageal leiomyomas [7]. We present a patient with esophageal leiomyoma discovered incidentally during investigations for thoracic trauma.

Case Report

We present a patient of 43 years, emergency admitted in 1st Surgical Unit Emergency County Hospital Targu Mures in January for thoracic trauma with fracture of the seventh and eighth ribs, left side arch and pulmonary contusion. Chest CT scan revealed an expansive mass (incidentaloma) in the middle third of the esophagus, with 60 x 55 mm, partially calcified (Figure 1).

Endoscopic exam showed an extrinsic compression of the esophagus in middle third, and no lesion on the esophageal mucosa. We mention that the patient reported no previous history of symptoms to be related to the presence of an esophageal tumor. The recover after thoracic trauma was uneventful and the patient was re admitted 2 months later for surgical treatment. The CT scan and upper gastrointestinal endoscopy revealed no changes from previous findings. Esophagogastric barium examination showed a slight narrowing in the middle third of the esophagus (Figure 2).

Given the suggestive findings of a benign esophageal tumor, probably esophageal leiomyoma, thoracoscopic approach was decided. The surgical procedure was performed, under general anesthesia with pulmonary selective intubation, the patient being positioned in the left lateral decubitus. Thoracoscopic ports were placed in intercostal spaces 9 and 6 on the anterior axillary line and intercostal spaces 7 and 5 on the posterior axillary line, respectively. We started by longitudinally dividing the mediastinal pleura over the esophageal tumor, then dividing the muscular fibers of the esophagus, progressively dissecting the tumor. The tumor had polylobate aspect, well defined, with 60×55 mm (Figure 3 and 4).

The tumor was completely dissected carefully not to injure the esophageal mucosa, by monopolar cautery, thermofusion device and ultrasonic dissector (Figure 5).

After enucleation of the tumor, esophageal mucosal integrity was verified by methylene blue dyne test on the nasogastric tube and nasogastric air insufflation (Figure 6). The esophageal muscular sheet and mediastinal pleura were then sutured interrupted absorbable stitches to prevent the development of esophageal pseudo diverticulum.

The tumor was extracted in a bag and a pleural drainage was performed.

***Corresponding author:** Ciprian Rosca, Aleea Constructorilor Street, No7, Ap. 13, Targu Mures, Romania, Tel: +40 (0) 744 51 78 23; E-mail: ciprian_rosca@yahoo.com

Received August 9, 2014; **Accepted** November 20, 2014; **Published** November 27, 2014

Citation: Tudor A, Rosca C, Bud V, Nicolescu C, Tudor B, et al. Incidental Discovery of an Esophageal Leiomyoma: Thoracoscopic Surgical Approach. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 389-391 DOI:10.7438/1584-9341-11-2-9

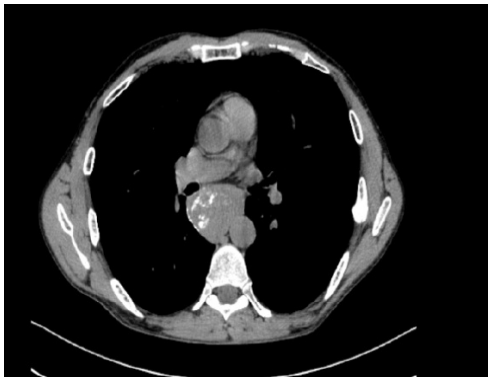

Figure 1: Esophageal Tumor CT image.

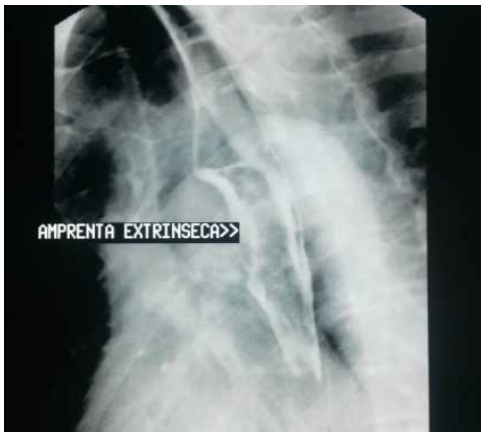

Figure 2: Esophagogastric Barium examination.

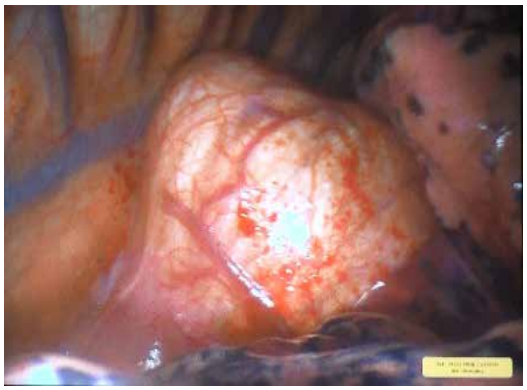

Figure 3: Intra Operative aspect of Esophageal Tumor.

The patient resumed a hydric diet from the first postoperative day and was discharged on day 7. Histopathological exam confirmed the diagnosis of esophageal leiomyoma (Figure 7).

Discussion

More than 90% of esophageal tumors are malignant. Esophageal leiomyoma is the most common benign esophageal tumor, the rest of benign tumors being extremely rare [8]. The majority develop in the muscular tunic of the esophagus, a part appearing in the muscularis mucosae [9]. Some authors consider that there is no direct relationship between tumor size and symptoms [10]. Other authors have found a correlation between symptoms and tumor size. Thus, at the mean tumor size of 5.3 cm, patients accused dysphagia, retrosternal pain, epigastric pain on palpation, regurgitation, dyspnea, weight loss [11]. In the case presented, the patient did not experience previously suggestive symptoms, the oesophageal tumor being incidentally discovered during investigations for thoracic trauma. Laboratory investigations used to highlight the esophageal tumor are esophageal-gastric barium swallow, esophagoscopy examination, computer tomography, endoscopic ultrasound. At the esophageal-gastric barium swallow appears a semilunar filling defect in the esophageal mucosa, the tumor is usually mobile with the swallowing of the barium [12-14]. The esophageal-gastric barium swallow revealed, in the presented patient, a slight narrowing in the middle third of the esophagus. Computerized tomography and endoscopic ultrasonography reveals the anatomical relationships of the tumor and differentiate from intramural and extrinsic lesions. Tomographic differentiation of esophageal leiomyomas neurofibromas, hemangiomas and other esophageal tumors is achieved with difficulty [10,13]. In the present case, chest CT scan showed an expansive process in the middle third of the esophagus, measuring 60 x 55 mm, partially calcified. Esophagoscopy is useful in

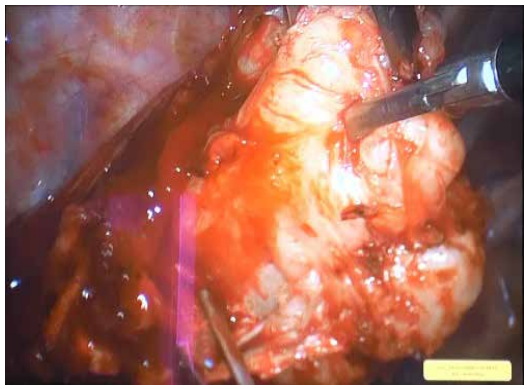

Figure 4: Dissecting the Tumor; to note the Polylobate aspect.

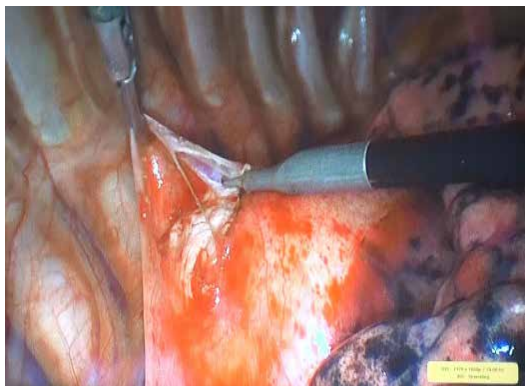

Figure 5: Cutting of the Muscular Tunic of the Esophagus.

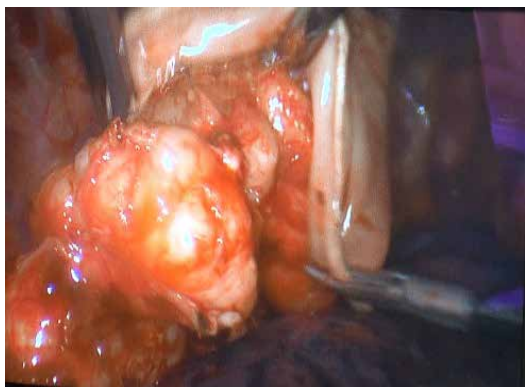

Figure 6: Tumor dissection and Enucleation.

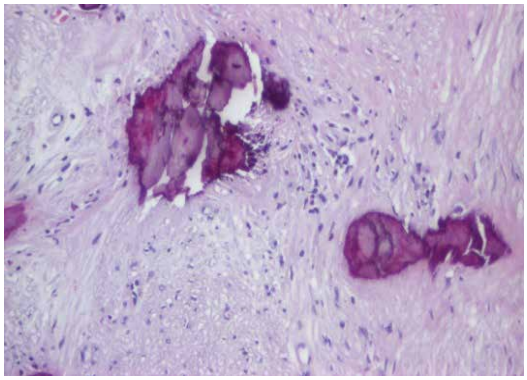

Figure 7: Histopathological aspect.

highlighting formations that protrude into the lumen and endoscopic biopsy may be beneficial in determining the nature of the tumor, but because of the risk of perforation, mediastinitis, most authors do not recommend it [6,11,15]. Preoperative endoscopic biopsy causes fibrosis between the tumor and submucosa, which increases the risk of mucosal perforation during tumor enucleation [16]. These tumors may into cyst degenerate, rarely can turn malignant [17]. There are authors who reported the discovery of a concomitant esophageal carcinoma and a leiomyoma [18]. The literature recommends surgery in symptomatic cases but also for asymptomatic cases, when the tumor is more than five centimeters, enlargement or ulceration of the mucosa [19,20]. There are authors who recommend non-surgical treatment in cases of asymptomatic or moderate symptoms, but radiological and endoscopic monitoring every 1-2 years [21]. Tumor removal can be done either by thoracotomy with esophageal resection or enucleation of tumor by thoracoscopic approach. Leiomyomas of the middle third of the esophagus are approached through right thoracotomy; tumors of the lower third require a left thoracotomy, while leiomyomas located near the gastro-oesophageal junction can be addressed through upper midline laparotomy. Esophageal resection is indicated for tumors over 8 cm, very adherent to the tumors mucosa or when there is extensive damage during mucosal dissection maneuvers [2,3]. Kent report even a thoracoscopic resection of leiomyoma measuring more than 8 cm [9]. During the last years, thoracoscopic approach of these tumors has gained many followers; Everitt made in 1992 the first thoracoscopic enucleation of an esophageal leiomyoma [22]. Thawatchai uses three points thoracoscopic approach [23]. In the present case we used four-point approach in 9 and 6 intercostal spaces, anterior axillary line, and the 7 and 5 intercostal spaces on the posterior axillary line. The advantages of thoracoscopic versus thoracotomy are: shortened hospitalization, reduced postoperative pain, quick re-expansion of the lung [24,25]. After enucleation of the tumor by blunt dissection and mucosal leak testing, myotomy is sutured with absorbable separate threads to prevent a pseudo diverticulum. According to some authors, suture of myotomy is not necessary [26], but most agree that the suture of muscular tunic is necessary to prevent protrusion of the mucosa [7,24-27].

Conclusion

Thoracoscopic enucleation is a feasible method for the treatment of esophageal leiomyomas with low morbidity rate and short hospital stay.

Conflict of interest

Authors have no conflict of interest to disclose.

References

1. Postlethwait RW, Lowe JE (1996) Benign tumors and cysts of the esophagus. In: Orringer MB, Zuidema GD (edt), Shackelford's surgery of the alimentary tract. WB Saunders, Pennsylvania.
2. Seremetis MG, Lyons WS, deGuzman VC, Peabody JW Jr (1976) Leiomyomata of the esophagus. An analysis of 838 cases. Cancer 38: 2166-2177.
3. Hatch GF, Wertheimer-Hatch L, Hatch KF, Davis GB, Blanchard DK, et al. (2000) Tumors of the esophagus. World J Surg 24: 401-411.
4. Lee LS, Singhal S, Brinster CJ, Marshall B, Kochman ML, et al. (2004) Current management of esophageal leiomyoma. J Am Coll Surg 198: 136-146.
5. Aurea P, Grazia M, Petrella F, Bazzocchi R (2002) Giant leiomyoma of the esophagus. Eur J Cardiothorac Surg 22: 1008-1010.
6. Priego P, Lobo E, Alonso N, Gil Olarte MA, Pérez de Oteyza J, et al. (2006) Surgical treatment of esophageal leiomyoma: an analysis of our experience. Rev Esp Enferm Dig 98: 350-358.
7. Bonavina L, Segalin A, Rosati R, Pavanello M, Peracchia A (1995) Surgical therapy of esophageal leiomyoma. J Am Coll Surg 181: 257-262.
8. Zhang W, Xue X, Zhou Q (2008) Benign esophageal schwannoma. South Med J 101: 450-451.
9. Kent M, d'Amato T, Nordman C, Schuchert M, Landreneau R, et al. (2007) Minimally invasive resection of benign esophageal tumors. J Thorac Cardiovasc Surg 134: 176-181.
10. Karagülle E, Akkaya D, Türk E, Göktürk HS, Yildirim E, et al. (2008) Giant leiomyoma of the esophagus: a case report and review of the literature. Turk J Gastroenterol 19: 180-183.
11. Mutrie CJ, Donahue DM, Wain JC, Wright CD, Gaissert HA, et al. (2005) Esophageal leiomyoma: a 40-year experience. Ann Thorac Surg 79: 1122-1125.
12. Sidhu R, Sood BP, Kalra N, Vajpae K, Joshi K, et al. (2002) Imaging features of esophageal leiomyomatosis: a case report. Clin Imaging 26: 293-295.
13. Jang KM, Lee KS, Lee SJ, Kim EA, Kim TS, et al. (2002) The spectrum of benign esophageal lesions: imaging findings. Korean J Radiol 3: 199-210.
14. Pujol J, Parés D, Mora L, Sans M, Jaurrieta E (2000) Diagnosis and management of diffuse leiomyomatosis of the oesophagus. Dis Esophagus 13:169-171.
15. Jiang G, Zhao H, Yang F, Li J, Li Y, et al. (2009) Thoracoscopic enucleation of esophageal leiomyoma: a retrospective study on 40 cases. Dis Esophagus 22: 279-283.
16. Cattaneo SM, Yang SC (2004) Benign tumors of the esophagus. In: Yang SC, Cameron DC (edts) Current Therapy in Thoracic and Cardiovascular Surgery, Philadelphia, Pennsylvania.
17. Nagashima R, Takeda H, Motoyama T, Tsukamoto O, Takahashi T (1997) Coexistence of superficial esophageal carcinoma and leiomyoma: case report of an endoscopic resection. Endoscopy 29: 683-684.
18. Mizobuchi S, Kuge K, Matsumoto Y, Yokoyama Y, Ookawauchi K, et al. (2004) Co-existence of early esophageal carcinoma and leiomyoma: a case report. Jpn J Clin Oncol 34: 751-754.
19. Zuccaro G Jr, Rice TW (1999) Tumors of the esophagus. In: Brandt LJ (edts), Clinical practice of gastroenterology. Churchill Livingstone, Philadelphia.
20. Fleischer DE, Haddad NG (1999) Neoplasms of the esophagus. In: Castell DO, Richter JE, (edts) The Esophagus, (3rd edn). Lippincott, Williams and Wilkins, Philadelphia.
21. Glanz I, Grünebaum M (1977) The radiological approach to leiomyoma of the oesophagus with a long-term follow-up. Clin Radiol 28: 197-200.
22. Everitt NJ, Glinatsis M, McMahon MJ (1992) Thoracoscopic enucleation of leiomyoma of the oesophagus. Br J Surg 79: 643.
23. Akaraviputh T, Chinswangwatanakul V, Swangsri J, Lohsiriwat V (2006) Thoracoscopic enucleation of a large esophageal leiomyoma using a three thoracic ports technique. World J Surg Oncol 4: 70.
24. van der Peet DL, Berends FJ, Klinkenberg-Knol EC, Cuesta MA (2001) Endoscopic treatment of benign esophageal tumors: case report of three patients. Surg Endosc 15: 1489.
25. Bardini R, Asolati M (1997) Thoracoscopic resection of benign tumours of the esophagus. Int Surg 82: 5-6.
26. Hennesey TPJ, Cushieri A (1992) Tumors of the oesophagus. In: Hennesey TPJ, Cushieri A (edts), Surgery of the oesophagus, Butterworth-Heinemann, London.
27. Lee LS, Singhal S, Brinster CJ, Marshall B, Kochman ML, et al. (2004) Current management of esophageal leiomyoma. J Am Coll Surg 198: 136-146.

Journal of Surgery
[Jurnalul de Chirurgie]

Case Report Open Access

Myelolipoma: A Rare Adrenal Incidentaloma

Debajyoti Pathak, Anshuman Tiwari, Subhobroto Das, Sandip Halder and Nilanjan Panda*

Department of Surgery, R.G Kar Medical College, Kolkata, India

Abstract

Background: Myelolipoma is a rarely encountered, adrenal incidentaloma diagnosed on the basis of its radiological features.

Aim: To describe a rare presentation with dual pathology.

Case presentation: Our patient, a 40-year-old lady presented with menorrhagia along with a large palpable uterine fibroid. Abdominal CECT detected a large, eight centimeter, left adrenal myelolipoma. After other possibilities were ruled out she underwent left adrenalectomy. Histopathology of the specimen revealed features of myelolipoma. She had an uneventful recovery and doing well now at six months follow up.

Conclusion: We present herewith a case of this uncommon tumour with dual pathology and discuss the clinical radiological and pathological features of adrenal myelolipoma.

Keywords: Incidentaloma; Adrenal; Myelolipoma

Introduction

Myelolipomas are rare, benign tumors composed of mature adipose tissue and hematopoietic elements (myeloid and erythroid cells) [1]. We describe a case of dual pathology where adrenal incidentaloma was successfully treated.

Case Report

A 40-year-old lady was referred to surgical outpatient clinic with an ultrasonologically detected adrenal mass, while being investigated for menorrhagia with a large pelvic mass. She did not have any symptoms related to the adrenal mass and was normotensive. Further biochemical investigations directed to adrenal pathology revealed a normal urinary VMA level, serum electrolytes and cortisol levels. The initial ultrasound scan suggested a well-defined, hyperechoic SOL in the left suprarenal region (94 x 80 mm) suggestive of a lipomatous tumour, along with a large intrauterine fibroid (119 x 96 mm). Subsequently, CECT of the abdomen revealed a large, well defined, mildly & heterogeneously enhancing mass lesion showing attenuation value of fat, involving left adrenal gland, suggestive of myolipoma (Figures 1 and 2). The right suprarenal was normal. In view of the large size of the uterine tumour and the fact that menorrhagia was under control, she underwent left adrenalectomy only in the first sitting, through a modified chevron incision. Macroscopical cut section of the specimen showed homogenous yellow surface with reddish streaking. Histopathology of the specimen revealed features of myelolipoma. She had an uneventful recovery and doing well now at six months follow up.

Discussion and Conclusion

Edgar von Gierke first described this lesion in the adrenal in 1905 but it was named, "myelolipoma", by Charles Oberling [2,3]. The adrenal gland is the most common site, but myelolipomas also (rarely) occur in extra-adrenal sites (14% of myelolipomas are extra-adrenal [4]) including the pelvis, mediastinum, retroperitoneum, and paravertebral region, as an isolated soft tissue mass [5].

It is usually hormonally inactive, and found in 0.08 to 0.2% of autopsy series [6,7] but comprise up to 15% of adrenal incidentalomas with the increasing use of noninvasive imaging [8] and account for 2.6% of all primary adrenal tumours [9]. Myelolipomas, affect both sexes equally and usually occur during fifth and seventh decades of life [10]. Adrenal myelolipomas are in the majority of cases unilateral. However, they can also be bilateral [11]. Adrenal myelolipomas may be found coincidentally with other lesions in the adrenal glands, such as adenomas and less commonly with pheochromocytoma or metastases. These cases are described as "collision tumours" [12].

Adrenal myelolipomas vary in size, from several millimetres to more than 30 cm, and usually in the range of 2-10 cm in diameter [1,13]. The term giant myelolipoma is preferred when the size exceeds 8 cm [14], as seen in our case.

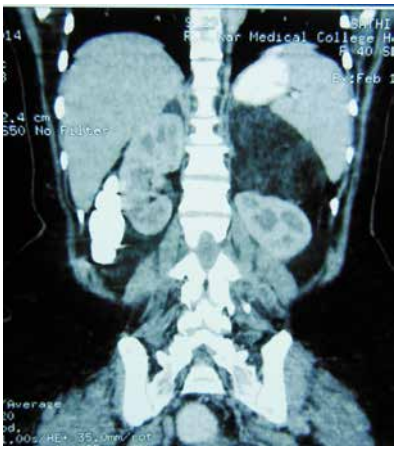

Figure 1: CT Coronal Image.

***Corresponding author:** Nilanjan Panda, Department of Surgery, R.G. Kar Medical College, P318 B.C.I.T road, Kankurgachi, Kolkata, 700054, West Bengal, India, Tel: +919748774942/+913323629007; E-mail: npanda05@aol.com

Received March 15, 2014; **Accepted** April 22, 2014; **Published** April 29, 2014

Citation: Pathak D, Tiwari A, Das S, Halder S, Panda N. Myelolipoma: A Rare Adrenal Incidentaloma. Journal of Surgery [Jurnalul de chirurgie]. 2015; 11(2): 393-395 DOI:10.7438/1584-9341-11-2-10

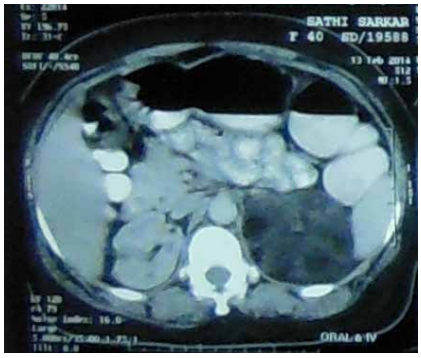

Figure 2: CT Axial Image.

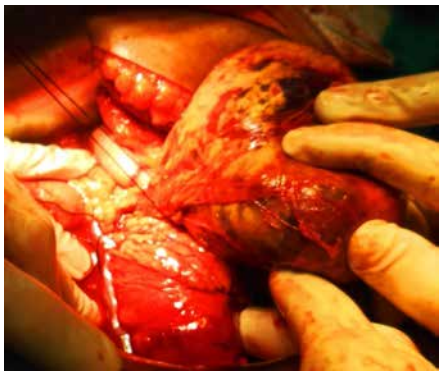

Figure 3: Intraoperative Aspect.

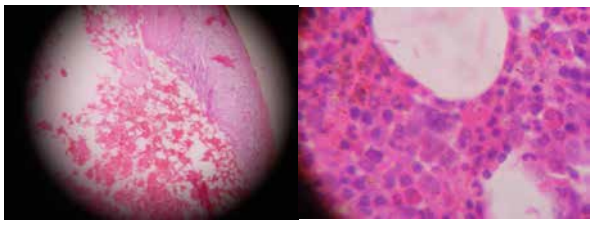

Figure 4: Histopathology (HE).

The exact pathogenesis of myelolipoma is not clear at present. Most theories involve the development and differentiation of either ectopic adrenal rests or hematopoietic stem cell rests in response to a triggering stimulus, particularly endocrine dysfunction [15]. Other researchers found clonal cytogenetic abnormalities, which suggested it was of neoplastic pathogenesis [16]. Chromosomal translocations (3;21) (q25;p11) detected in myelolipomas and in benign lipomatous neoplasia seen in patients with acute myelogenous leukemia or myelodysplastic syndrome suggest the origin of this tumor to be bone marrow, and may indicate that myelolipoma is derived from erroneously transferred erythroid cells [17].

Adrenal myelolipomas, which are usually asymptomatic and non-functioning, are generally importantpurposes [19]. The most frequent symptoms of large myelolipomas are non-specific abdominal pain, hematuria, renovascular hypertension and other symptoms secondary to the mechanical compression of adjacent organs [18-22]. Although rare, surgical emergencies such as retroperitoneal hemorrhage may also be encountered [21].

Adrenal myelolipomas are generally hormonally inactive, although there are case reports of their association with overproduction of adrenal hormones. They have been associated with overproduction of dehydroepiandrosterone- sulphate (DHEAS), congenital adrenal hyperplasia caused by 21-hydroxylase deficiency, congenital adrenal 17 α-hydroxylase deficiency, Cushing disease, Conn's syndrome, adrenal insufficiency, and pheochromocytoma [23-28].

The extensive use of abdominal CT-scan and magnetic resonance imaging has led to a dramatic increase in incidentally discovered adrenal masses that have also been called adrenal incidentalomas [29]. Radiological imaging typically reveals a well-circumscribed mass with a heterogeneous appearance due to the varying proportions of fat within the mass. Adipose tissue is characterized by low attenuation on CT imaging (i.e., – 25 to – 100 Hounsfield units). On MRI, fat displays high signal intensity on T1-weighted images whereas the myeloid component of these tumors has a T2-weighted signal. Contrast enhancement with CT scan or MRI will vary depending on the composition of the mass. Soft tissue components enhance whereas adipose tissue does not [30]. Calcification is present in a minority of cases on CT. Because of their characteristic appearance ono CT, adrenal myelolipomas if small can usually be diagnosed without intervention and followed radiographically. Extra-adrenal myelolipomas, however, are more difficult to diagnose preoperatively because they are easily confused with several malignancies. If a definite diagnosis is needed, a fine-needle biopsy is indicated either under US or CT guidance.

Grossly, myelolipoma is a solitary circumscribed mass ranging in size from a few centimeters to 27cm [31]. The tumor is usually spherical to ovoid, well circumscribed, sometimes surrounded by a pseudo-capsule. The cut surface typically has a variegated appearance, with areas of greasy-appearing soft yellow tissue alternating with irregular areas of dark red-brown friable tissue, as we found in our case. Microscopically, the tumor is composed of a variable admixture of mature adipose tissue with islands and nests of hematopoietic elements of different percentages. The cellularity of hematopoietic precursors is variable and the three hematopoietic cell lineages (granulopoietic, erythropoietic and megakaryocytic) are present. In some cases, areas of infarction, hemorrhage, and rarely foci of calcification are noted [32]. Immunohistochemical staining and molecular testing is of no clinical or histological benefit.

When the diagnosis of myelolipoma is considered, it should be differentiated from other fat containing retroperitoneal tumors including retroperitoneal lipomas, retroperitoneal liposarcoma, extra-renal angiomyolipoma, extramedullary hematopoietic 'tumors', retroperitoneal leiomyosarcoma, primary or metastatic adrenal malignancy and teratomas [9,33,34].

Once adrenal myelolipoma is diagnosed, regular follow-up with sonography or CT is recommended and surgery is reserved for symptomatic cases. Some studies suggest surgical intervention for symptomatic tumors, growing tumors, or tumors larger than 10 cm to reduce the risk of developing abdominal pain or life-threatening hemorrhage [35]. From the reviewed papers, 17% cases whose tumor size was greater than 6 cm experienced spontaneous rupture. Therefore, elective surgery can prevent more severe symptom presentation and life-threatening progression and can allow accurate diagnosis in patients with tumors larger than 6 cm [33]. Castillo et al. advocated laparoscopic adrenalectomies for myelolipoma. [36,37]

In our case the tumour was almost 10 cm and presence of another large tumour in the pelvis made the case unusual and required early intervention to prevent rupture or haemorrhage and to reduce the confusion of double pathology.

Conflict of interests

Authors have no conflict of interests to disclose.

References

1. Federle M, Anne V (2004) Adrenal Myelolipoma. In: Federle M, Diagnostic Imaging: Abdomen, (1st edn), Amirsys, Salt lake.

2. Gierke E (1905) Uber Knochenmarksgewebe in der Nebenniere. Beitr Pathol Anat 7:311-325.

3. Oberling C (1929) Les formation myelo-lipomateuses. Bull Assoc Fr Etud Cancer 18:234-246.

4. Hakim A, Rozeik C (2014) Adrenal and extra-adrenal myelolipomas - a comparative case report. J Radiol Case Rep 8: 1-12.

5. Kammen BF, Elder DE, Franker DL, Siegelman ES (1998) Extraadrenal myelolipoma: MR imaging findings. AJR Am J Roentgenol 171: 721-723.

6. Manassero F, Pomara G, Rappa F, Cuttano MG, Crisci A, et al. (2004) Adrenal myelolipoma associated with adenoma. Int J Urol 11: 326-328.

7. Tamidari H, Mishra AK, Gupta S, Agarwal A (2006) Catecholamine secreting adrenal myelolipoma. Indian J Med Sci 60: 331-333.

8. Anagnostis P, Karagiannis A, Tziomalos K, Kakafika AI, Athyros VG, et al. (2009) Adrenal incidentaloma: a diagnostic challenge. Hormones 8:163-184.

9. Lam KY, Lo CY (2001) Adrenal lipomatous tumours: a 30 year clinicopathological experience at a single institution. J Clin Pathol 54: 707-712.

10. Meaglia JP, Schmidt JD (1992) Natural history of an adrenal myelolipoma. J Urol 147: 1089-1090.

11. Cha JS, Shin YS, Kim MK, Kim HJ (2011) Myelolipomas of both adrenal glands. Korean J Urol 52: 582-585.

12. Otal P, Escourrou G, Mazerolles C, Janne d'Othee B, Mezghani S, et al. (1999) Imaging features of uncommon adrenal masses with histopathologic correlation. Radiographics 19: 569-581.

13. Ramchandani P (2011) Adrenal Myelolipoma Imaging. Medscape Reference.

14. Akamatsu H, Koseki M, Nakaba H, Sunada S, Ito A, et al. (2004) Giant adrenal myelolipoma: report of a case. Surg Today 34: 283-285.

15. Hunter SB, Schemankewitz EH, Patterson C, Varma VA (1992) Extra-adrenal myelolipoma: a report of two cases. Am J Clin Pathol 97: 402-404.

16. Bishop E, Eble JN, Cheng L, Wang M, Chase DR, et al. (2006) Adrenal myelolipomas show non-random X-chromosome inactivation in hematopoietic elements and fat: support for a clonal origin of myelolipomas. Am J Surg Pathol 30: 838-843.

17. Chang KC, Chen PI, Huang ZH, Lin YM, Kuo PL (2002) Adrenal myelolipoma with translocation (3;21)(q25;p11). Cancer Genet Cytgenet 134: 77-80.

18. Boudreaux D, Waisman J, Skinner DG et al. Giant adrenal myelolipoma and testicular interstitial cell tumor in a man with congenital 21-hydroxylase deficiency. Am J Surg Pathol 3: 109-123.

19. Wilhelmus JL, Schrodt GR, Alberhasky M et al (1981) Giant adrenal myelolipomas: Case report and review of the literature. Arch Pathol Lab Med 105: 532-535.

20. O'Daniel-Pierce ME, Weeks JA, McGrath PC (1996) Giant adrenal myelolipoma. Southern Med J 89: 1116-1118.

21. Goldman HB, Howard RC, Patterson AI (1996) Spontaneous retroperitoneal hemorrhage from giant adrenal myelolipoma. J Urol 155: 639.

22. Vierna J, Laforga JB (1994) Giant adrenal myelolipoma. Scand. J Urol Neph 28: 301-304.

23. Hisamatsu H, Sakai H, Tsuda S, Shigematsu K, Kanetake H (2004) Combined adrenal adenoma and myelolipoma in a patient with Cushing's syndrome: case report and review of the literature. Int J Urol 11: 416-418.

24. Skucas J (2006) Adrenals. In: Advanced Imaging of the Abdomen. (1st edn) Springer, London.

25. Oliva A, Duarte B, Hammadeh R, Ghosh L, Baker RJ (1988) Myelolipoma and endocrine dysfunction. Surgery 103: 711-715.

26. Miyazaki Y, Yoshida M, Doi J (1990) [A case of adrenal myelolipoma associated with adrenogenital syndrome]. Hinyokika Kiyo 36: 35-39. [Article in Japanese]

27. Ukimura O, Inui E, Ochiai A, Koljima M, Watanabe H (1995) Combined adrenal myelolipoma and pheochromocytoma. J Urol 154: 1470.

28. Goldman S, Kenney P (2006) The adrenal gland. In: LEE J, et al. Computed body tomography with MRI correlation. (4th edn), Lippincott Williams & Wilkins, Philadelphia.

29. Mansmann G, Lau J, Balk E, Rothberg M, Miyachi Y, et al. (2004) The clinically inapparent adrenal mass: update in diagnosis and management. Endocr Rev 25: 309-340.

30. Cox A, Offman LS, Merrimen LOJ, Kew A, Norman WR (2010) Bilateral renal sinus myelolipomas. Can Urol Assoc J 4: E164-E168.

31. Shapiro JL, Goldblum JR, Dobrow DA, Ratliff NB (1995) Giant bilateral extraadrenal myelolipoma. Arch Pathol Lab Med 119: 283-285.

32. Olobatuyi FA, MacLennan GT (2006) Myelolipoma. J Urol 176: 1188.

33. Hsu SW, Shu K, Lee WC, Cheng YT, Chiang PH (2012) Adrenal myelolipoma: a 10-year single-center experience and literature review. Kaohsiung J Med Sci 28: 377-382.

34. Zhao J, Sun F, Jing X, Zhou W, Huang X, et al. (2014) The diagnosis and treatment of primary adrenal lipomatous tumours in Chinese patients: A 31-year follow-up study. Can Urol Assoc J 8: E132-E136.

35. Patel VG, Babalola OA, Fortson JK, Weaver WL (2006) Adrenal myelolipoma: report of a case and review of the literature. Am Surg 72: 649-654.

36. Castillo OA, Vitagliano G, Cortes O, Sa´nchez-Salas R, Arellano L (2007) Laparoscopic adrenalectomy for adrenal myelolipoma. Arch Esp Urol 60: 217-221.

37. Yamashita S, Ito K, Furushima K, Fukushima J, Kameyama S, et al. (2014) Laparoscopic versus open adrenalectomy for adrenal myelolipoma. Annals of Medicine and Surgery 3: 34-38.