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Abdominal Complications of Typhoid Fever

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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 10^9 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 neuropyschiatric 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 [3,5,6].

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...
the perforation is big, surrounding bowel edematous and the volume of peri toneal 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 perforation. 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 mucocle 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.

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

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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-pathological 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 proteomics, genomics and metabolomics have emerged as possible alternative or adjunctive to histomorphological tests [6-8]. The term metabolomics is defined as “the quantitative measurement of the dynamic multiparametric metabolic response of living system to pathophysiological stimuli or genetic modification”. Magnetic resonance spectroscopy has emerged out as one of the main techniques of metabolomics and has been widely used to assess 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
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 T1/T2 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 T1/T2 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 'H and '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 histopathological examination.

**Results**

Seventeen lymph node specimens from 17 subjects undergoing sentinel node biopsy from T1/T2, 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 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.
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].

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 3: A Portion of 1D NOESY Stack Plot Spectra of Involved and Non-involved Lymph Nodes (PC- Phospocholine, Ch- Choline, TG- Tri Glycride)](image-url)
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. Metabonomics 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, ascitic 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 an 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.

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Retrorectal Myxoid Fibrosarcoma: A New Entity

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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.

Keywords: Retrorectal tumor; Fibrosarcoma; Surgery

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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

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Volume 11 • Issue 2 • 3

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.

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

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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 gastrojejunostomy 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; Gastrojejunostomy

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 anaemia and hypoproteinaemia. 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 inflamed and pylorus was thickened with a narrowed lumen. A gastrojejunostomy 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].

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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

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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. Succussion 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.

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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 esophagus 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].
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Unexpected Outcome of a Floating Thrombus in the Ascending Aorta

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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, presented with a syncpe 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 (aspirin, clopidogrel, β-blockers, heparin) treatment. 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, the pulse rate was 82 bpm. BUN and creatinine were normal. The total CK was 10000U/L. A prompt coronary angiography; 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 coronary trunk, 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 resumed 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 approximation to the left main coronary trunk, causing intermittent occlusion of the latter.

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 with a syncpe 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 (aspirin, clopidogrel, β-blockers, heparin) treatment. 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, 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, contrpulsion was immediately terminated. A transesophageal echocardiography revealed a free floating mass 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 resumed early after releasing the aortic cross clamp but it was impossible to wean CPB despite inotropic drugs and IABP support.
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 antegrade and retrograde cardiopleagia were used, aortic root was transversely incised. After the thrombus had been excised, the aortic incision was sutured with a double suture lines.

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

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Abstract

Introduction: The soft-tissue defects with bone exposure 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 therapeutic solutions applied in a case of crush syndrome of young men of 23 years old. A hemisoleal medial flap, a flexor digitorum longus flap and a split-thickness skin graft were done. Kineatherapy 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

Introduction

Traumatismele de strivire la nivelul extremităților, chiar dacă nu implică organele vitale, pot pune viața în pericol. “Crush syndrome” reprezintă manifestarea sistemică a distrugerii celulelor musculare și eliberării conținutului acestora în circulație, ducând la dereglaarea metabolismului și leziunii renale acute [1]. După reanimarea unui pacient cu o astfel de patologie, rămâne problema reconstrucției locale a defectului restant. Defectele de părți moi cu expunere osoasă la nivelul gambei, în special cele prețiove, rămân provocarea principală pentru chirurgii, punând probleme în alegerea soluției chirurgicale optime de acoperire. Dificultatea este dată de mobilitatea redusă a acestui nivel și 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 și personale semnificative, fumător cronicit, internat în regim de urgență, acesta fiind victima unui traumatism major prin strivire provocat de căderea unei greutăți cu masa de 3 tone de la nivelul regiunii posterioare a gambei și coapsei, plagă transversală de 2 cm lungime la nivelul antepiciorului drept, sensibilitate normală și reducerea sângerării a fracturii din 1/3 distală a gambei și stabilitarea acesteia prin montarea unui fixator externd (Figura 1), urmată de pânsamentul zilnic și antibioterapie. În a 5-a zi postoperator pacientul a fost transferat în Clinica de Chirurgie Plastica și Reconstrucțivă.

Prima etapă

A doua etapă este reprezentată de următoarele 8 zile de internare a pacientului în clinica de Chirurgie Plastică, perioadă caracterizată prin saltă a indicatorului de recuperare a pacientului la nivelul regiunii traumatizate, stabilit hemodinamic și metabolic, afibril, cu temperaturi normale și modificări de tranzit. S-a intervenit chirurgical și s-a practicat toaleta chirurgicală, excizia știrea ultimelor necrotozice, plastie cu lambouri musculare (hermisolear medial și flexor lung de degete), plastie cu piele liberă despicată, drenaj, hemostază, sutură, pansament, imobilizare. S-a continuat în postoperator antibioterapia instituită pacientului după prima intervenție chirurgicală, iar din a-2-a zi postoperator, când valoarea trombocitelor s-a normalizat s-a adaugat în schema terapeutică și un anticoagulant.

Evoluția postoperatorie favorabilă (stare generală bună, afibrilitate, scăderea valorilor markerilor strivirii musculare, normalizarea valorilor
trombocitelor, hemoglobinii, hematocritului, hematiiilor, pansament curat fără secreții purulente, grefe bine integrate) a permis suprimarea drenului din loja posterioară profundă a gambei în a 6-a zi postoperator și transferul bolnavului în Clinica de Ortopedie și Traumatologie în a 8-a zi postoperator în vederea tratamentului chirurgical al fracturii de platou tibial extern.

**Tehnica Operatorie**

Intervenția chirurgicală s-a efectuat sub rahianestezie. Abordul chirurgical s-a făcut pe la nivelul plăgii. S-au inventariat leziunile restante de strivire, s-a completat toaleta chirurgicală riguroasă cu soluții antiseptice și s-au excizat țesuturile și fasciile musculare devitalizate, necrotice prezentate pe întreaga suprafață 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 și ingust s-a optat pentru prelevară unui lambou hemisolear medial, care să fie avansat anterior, astfel încât să acopere regiunea medio-distală a gambei. Separaarea mușchiului solear de gastrocnemian s-a făcut prin digitoclasie proximal iar distal s-a separat aponevroza solerului de cea a gastrocnemianului și s-au secționat fibrele musculare ale solearului în 1/3 medială pentru a le elibera de tendonul lui Achile. Fibrele au rămas intarcte în 1/3 laterală, împiedicând astfel efectul de tracțiune exagerat asupra capătului distal al mușchiului (Figura 2). Partea distală eliberată a extras din plăsgă și s-a continuat cu incizia fasciei intermusculare care separă compartimentul superficial al gambei de cel profund. Incizia acestei fascii în poziția laterală a permis evacuarea și drenarea ulterioară a hematomului din loja profundă. Secționarea ei în poziția medială s-a făcut cu grijă pentru a evita lezarea pachetului vasculo-nervos tibial posterioar. S-a evidențiat astfel lipsa totală a mușchiului pentru a acoperi 1/3 externă a porțiunii mediei și acoperirea musculaturii (Figura 1D). Această a doua particularitate a cazului ne-a oferit șansa evitării utilizării unui lambou gastrocnemian, care, deși, posedă un arc de rotație mai mare decât al flexorului distal al mușchiului, decizia care ar fi provocat în postoperator un dezechilibru funcțional la nivelul gambei.

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

**Discuții**

Tehnicile de acoperire a defectelor de părți moi cu expunere osoasă la nivelul gambei sunt reprezentate de lambourile locoregionale sau lambourile pediculate perforante. Lambourile fasciocutanate, lambourile muscular și lambourile pediculate perforante s-au făcut cu grijă pentru a evita lezarea pachetului vasculo-nervos tibial anterior. S-a evidențiat astfel lipsa totală a mușchiului pentru a acoperi 1/3 externă a porțiunii mediei și acoperirea musculaturii (Figura 1D). Această a doua particularitate a cazului ne-a oferit șansa evitării utilizării unui lambou gastrocnemian, care, deși, posedă un arc de rotație mai mare decât al flexorului distal al mușchiului, decizia care ar fi provocat în postoperator un dezechilibru funcțional la nivelul gambei.

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**Figura 1:** Defecțul restant după excizia țesuturilor devitalizate. Stabilizarea focarului de fractură tip III C cu fixator extern.

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

**Figura 3:** Incizii longitudinale ale aponevrozei superficiale a mușchiului solear și avansarea anterioară a mușchiului tibial anterior.
Lambourile fasciocutanate sunt reduse numeric datorită mobilității tegumentare care reduc aceste regiuni.

Cele mai utilizate lambouri musculare locoregionale pentru soluționarea defectelor din regiunea antero-interna a gambei sunt, în ordinea frecvenței:

1) Lamboul gastrocnemian

Gastrocnemianul este cel mai utilizat mușchi pentru acoperirea gambei și a gemenului. Fiecare cap al mușchielor (medial și lateral) poate fi mobilizat separat, fiecare posedând un pedicul vascular-nervos propriu (artera surală) [3,4]. Capul medial permite acoperirea cu ușurință a 1/3 proximale a tibiei și fața antero-internă a gemenului [3,4]. Prin creșterea arcului de rotație al mușchielor (încălzit în transvers transversale ale aponevrozei de pe fața profunză a mușchielor și secționarea inserției proximale a tendonului) acesta poate acoperi defecte situate în 1/3 superioară a 1/3 medi a gambei [3,4].

2) Lamboul solear

Mușchiul solear se găsește superficila în compartimentul posterior al gambei și de cele mai multe ori nu este lezat în fracturile deschise ale 1/3 medi ale tibiei. Acoperirea cu ajutorul întregului mușchi este indicată în cazul unor defecțiuni scurte de la 1/3 medi a gambei, în timp ce acoperirea cu hemisolear medial e posibilă prin avansarea antero-interna a lamboiului și este indicată în cazul unor defecte lungi și înguste care interesează cresta tibială [3]. Lamboul solear cu bază distală nu este indicat în acoperirea 1/3 distale a gambei datorită existenței a numeroase variatii ale pediculului distal minor [4].

3) Lambourile bazate pe flexorii gambei (flexorul comun de degete și flexor lung de halucu)

Aceste tipuri de lambouri musculare prezintă interes în acoperirea defectelor modulo de la 1/3 distal ale gambei [3]. Numai o ½ din mușchi poate fi prelevată și folosită ca lambou pentru a acoperi fața centrală a gambei, a cărei acoperire nu se poate face cu lambouri musculare flacare [3]. Extensorul lung de halucu este indicat pentru acoperirea 1/3 distale a crestei tibiale, prin rotația ½ distală a mușchielor [3,5]. Lamboul muscular tibial anterior este un lambou puțin utilizat, în special datorită faptului că este un mușchi de mici dimensiuni și de minimă flexibilitate [6]. Tibialul anterior (T.A.) este utilizat în special pentru acoperirea 1/3 medi a crestei tibiale [6], prin avansarea corpului muscular [3,5]. Funcția acestui mușchi este vitală și prelevarea lui duce la un deficit funcțional important. În plus, arcul de rotație al ½ distal este foarte redus. Din acest motiv, cel mai bun mod de utilizare a acestui mușchi este acoperirea antero-internă a gambei și a tacticilor musculare pentru a acoperi defecte lungi și înguste ale crestei tibiale [6]. Acest procedeu evită instalairea unui deficit funcțional în postoperator, tibialul anterior fiind cel mai puternic flexor dorsal al piciorului, adductor și rotator intern al acestuia [3].

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

Lambourile liber transferate (lambouri fasciocutanate perforante, lambouri musculare) sunt frecvent utilizate pentru reconstrucția posttraumatică a membrului pelvin [7]. Utilizarea unei sau a celorlalte tip de lambou a rămas un problema controversată în literatură [7]. Scopul principal al microchirurgiei reconstructive este obținerea unui rezultat estetic și funcțional optim cu o morbiditate minimă a regiunii donatoare [8]. Astfel, majoritatea articolelor retrospective pledează pentru siguranța mai mare a lambourilor liber transferate. Microchirurgia reconstructivă reprezintă o modalitate de tratament alternativă pentru defectele mari a membrelor inferioare și de la membrele superioare [9].

Concluzii

Metoda de reconstrucție prezentată anterior reprezintă o metodă fiabilă de rezolvare a defectelor tisulare din regiunea antero-interna a gambei, utilizând unele lambouri musculare. Acestea sunt observații care permit, atunci când este posibil, evitarea transferului liber. Avantajele metodei utilizeate sunt: limitarea deficitului funcțional restant prin mobilizarea pe volumul muscularului din loja postero-interna combinată cu cea din loja antero-interna a gambei; oferă o bună masă musculară pentru acoperirea focarului de fractură; musculatura oferă un suport bun pentru grefarea, cu integraj bună a acestora. Dezavantajul tehnicii constă în scăderea mișcării și deloc facilă a lambourilor amintite, cu secționarea maselor musculare care provoacă o săngereare secundară importantă, ceea ce impune de fiecare dată drenarea cavitatei restante după transplantarea musculaturii.

Conflict de interese

Autoarea nu declară niciun conflict de interese.

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

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Abstract

Anaplastic thyroid carcinoma presents as an extremely locally 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 imagiological features strongly pleaded for the diagnosis of anaplastic thyroid carcinoma presumably with liver metastases, also supported by the rapid recurrence following total thyroidecmy. Immunohistochemical tests showed a malignant carcinomatous proliferation with anaplastic prohile. Positive immunochemical staining for cytokeratin AE1/AE3, CK7 and negative CK20, AFP, CD15, CD30, CDS, 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 prohile. 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. 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.

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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

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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 subarachnoid 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 locally 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 profile. Positive immunohistochemical 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 puncton biopsy. This exploration would have enabled us to delineate with accuracy the relationship between the thyroid tumor and the liver nodules.

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.

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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.

| Cytokeratin       | Intense Positive |
|-------------------|------------------|
| Cytokeratin CK20  | Negative         |
| Thyroglobulin (TG), TTF1 | Negative    |
| AFP               | Negative         |
| Chromogranin, synaptophysin | Negative |
| LCA               | Intense Positive in peritumoral lymphoid tissue but negative in the tumoral cells |
| CD15, CD30        | Negative         |
| S100              | Negative         |
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Surgical Approach of Cervical Cancer Liver Metastases: Case Report

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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 liver 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 adnexitomy 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 metastasize 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.

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

Citation: Bacalbaşa N, Balescu I. Surgical Approach of Cervical Cancer Liver Metastases: Case Report. Journal of Surgery [Jurnalul de chirurgie] 2015; 11(2): 385-387 DOI: 10.7438/1584-9341-11-2-6
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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
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 colorectal 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 metastat ed 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 metastatous liver metastases originating from cervical cancer. The conclusions of this study was that hepatectomy can be performed safely and prolong survival [20].

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 [22].

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.

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Incidental Discovery of an Esophageal Leiomyoma: Thoracoscopic Surgical Approach

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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 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 tumors [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. Esophagogastroduodenoscopy 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 60x55 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.
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 esophageal 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
highlighting formations that protrude into the lumen and endoscopic biopsy may be beneficial in determining the nature of the tumour, 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 thorascopic 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 thorascopic resection of leiomyoma measuring more than 8 cm [9]. During the last years, thorascopic approach of these tumors has gained many followers; Everitt made in 1992 the first thorascoscopic enucleation of an esophageal leiomyoma [22]. Thawatchai uses three points thorascopic 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 thorascopic 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
Thorascoscopic 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.

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Myelolipoma: A Rare Adrenal Incidentaloma

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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 [13]. The term giant myelolipoma is preferred when the size exceeds 8 cm [14], as seen in our case.

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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

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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, 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 on 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 27 cm [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 angiomylolipoma, 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.

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