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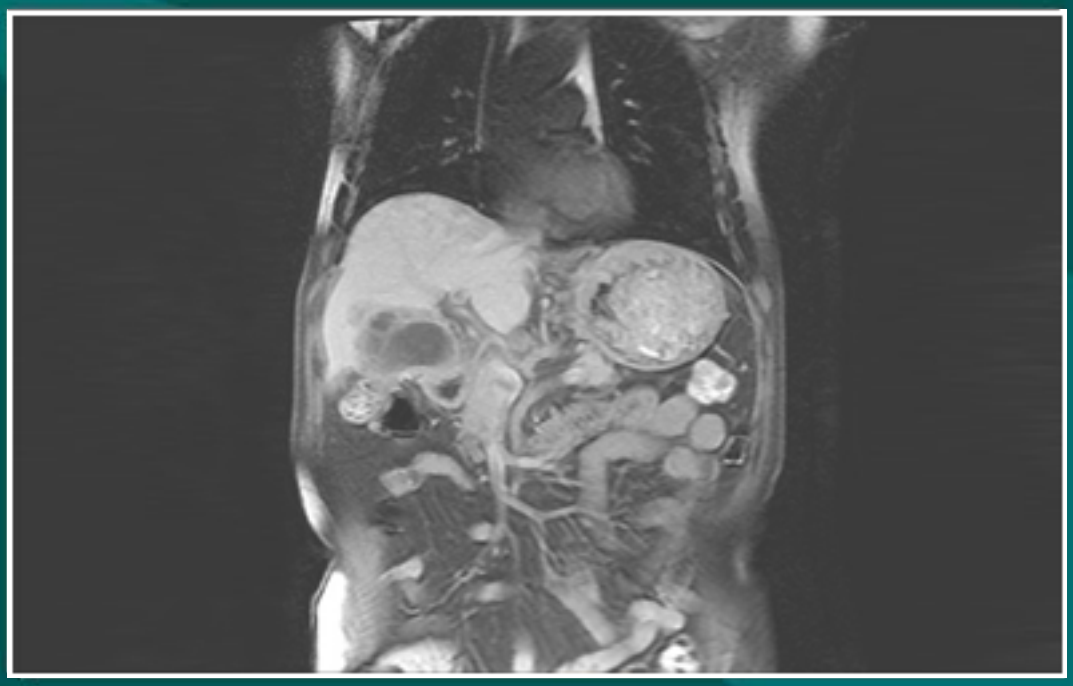

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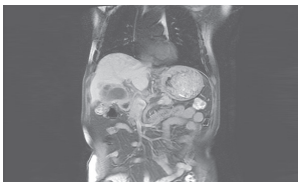

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Medullary thyroid carcinoma: Management and complexities of postoperative follow-up

Mirza Faraz Saeed, Nida Fatima Sakrani, Isam Mazin Juma, Abbas Ali

ABSTRACT

Introduction: Medullary thyroid cancer is one of the less common types of thyroid malignancies as compared to the more frequent papillary and follicular types and constitutes 4% of thyroid cancer. Serum tumors markers are a significant part of diagnosis and postoperative follow-up. Although calcitonin is an apparent marker, levels of carcinoembryonic antigen (CEA) can also be used. In patients that are surgically managed, calcitonin levels can begin to rapidly decline within the first postoperative hour. Case Report: This paper presents a case of 37-year-old Indian female treated for medullary thyroid carcinoma and followed-up over a year. Fine-needle aspiration cytology (FNAC) biopsy was reported as highly suspicious of malignancy. The patient underwent a near total thyroidectomy and the following histopathology report confirmed medullary carcinoma. Conclusion: This paper highlights the importance of biochemical follow-up of calcitonin level, following surgical resection in cases of medullary thyroid carcinoma as a part of postoperative care.

Keywords: Calcitonin, Fine-needle aspiration cytology (FNAC), Medullary thyroid carcinoma, Postoperative care

How to cite this article

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INTRODUCTION

The thyroid tissue is known to be one of the most radiation-sensitive tissues in the body, with excess cancers occurring at doses as low as 100 mGy [1, 2]. Of note, American statistics report that medical radiation accounts for nearly half of the radiation exposure experienced by the population [2]. Medullary thyroid cancer is one of the less common types of thyroid malignancies as compared to the more frequent papillary and follicular types [3] and constitutes 4% of thyroid cancer [1]. Chances of survival are much improved and notably optimistic when the malignancy is detected early.

This subtype of cancer is a neuroendocrine tumor and comprises the parafollicular C cells. The malignant C cells release an increased level of calcitonin that is medically useful as a tumor marker used in diagnosis and follow-up

[1]. Medullary cancer is infamous due to its involvement in the multiple endocrine neoplasia type 2 (MEN2) syndromes. However, the sporadic, nonhereditary type is actually responsible for 80% of cases [1]. The tumor can present as a neck mass and this is most common, or with symptoms pertaining to increased hormone levels similar to other tumors composed of neural crest cells. Tumor secretion of calcitonin, calcitonin gene related peptide can cause diarrhea or facial flushing in patients with advanced disease, while occasionally the secretion of ACTH (corticotropin) can manifest as Cushing's syndrome [1]. When present as part of the MEN2 combination the patient may suffer accordingly with hyperparathyroidism, a pheochromocytoma or a marfanoid habitus, mucosal neuromas, and intestinal ganglioneuromatosis [1].

This report pertains to a case of early stage medullary thyroid carcinoma that developed in a lady with previous radiation exposure and was successfully operated upon. In this discussion, we dissect the process of postoperative follow-up and the dilemma of the increasing tumor marker, calcitonin following such definitive treatment.

CASE REPORT

A 37-year-old Indian female nurse, with a five-year history of without any radiation exposure while working as orthopedic surgical staff, presented to our surgical clinic in July 2012 with a self-discovered nodule in the right lobe of her thyroid gland. Further questioning revealed weight loss, a history of gestational hypothyroidism treated with levothyroxine and a significant family history with a father currently in remission after treatment for an oropharyngeal malignancy.

Following appropriate examination, appropriate investigations were undertaken including a fine-needle aspiration cytology (FNAC) biopsy. The FNAC was reported as highly suspicious of malignancy. FNAC features showed several small and large clusters of elongated spindle-shaped cells. Hyperchromatic, pleomorphic nuclei with scant cytoplasm. Glassy pink amyloid-like material was also present, suggestive of medullary thyroid carcinoma. Consequently, the patient underwent a near total thyroidectomy on 24th July 2012 and the histopathological diagnosis confirmed our suspicions of malignancy and further diagnosed a containing medullary carcinoma, staged as T1N0M0 with no nodular involvement. Computer tomography scans of the chest, abdomen and pelvis were done after the surgery ruled out any metastasis.

Between 2013 and 2014, the patient was seen at regular follow-up visits and neck ultrasounds, as well as calcitonin and CEA levels were repeated. The increasing size of the small residual thyroid tissue left from 1 cm in 2013 to 1.8 in 2014 was noted along with rising CEA and calcitonin levels as depicted in the (Figure 1 and Figure 2).

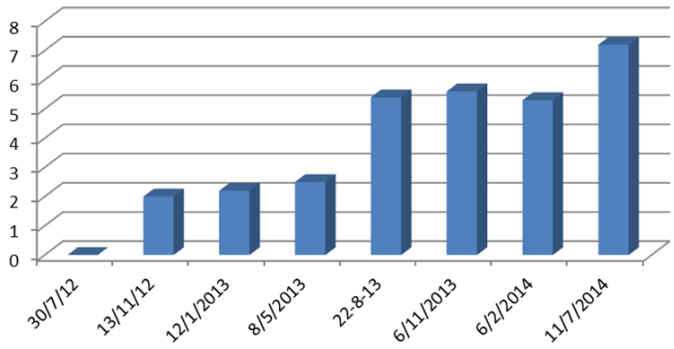

Figure 1: Rising trend of calcitonin in postoperative period.

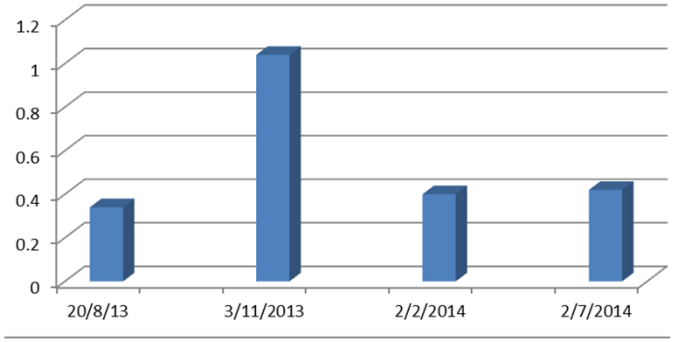

Figure 2: Rising trend of carcinoembryonic antigen in postoperative period.

DISCUSSION

Initial Steps

Multiple sources list the same process of diagnosis as was followed by our team which comprises clinical examination, followed by a neck ultrasound and fine needle aspiration. There is controversy regarding the use of calcitonin measurement when there is a high suspicion and is not carried out in the United States because of the high frequency of falsely high serum calcitonin values and the accuracy of FNAC [1]. Sonographic imaging is used to determine lymph node metastasis, and when detected then further imaging is used to stage the tumor. Imaging techniques include neck computed tomography (CT), chest CT, tri-phasic liver CT with contrast or liver magnetic resonance imaging (MRI) with contrast [1].

Following pathological confirmation, serum tumor markers become significant, and while calcitonin is the apparent one, carcinoembryonic antigen (CEA) is also of significance. Advanced imaging techniques discussed above are also to be employed when the basal preoperative calcitonin level is >400 pg/mL since this implies a high risk of metastatic disease [1] Postoperatively, these results allow the clinician to decide on the prognosis and postoperatively indicate biochemical cure.

To complete the process, the patient must be screened for the other manifestations of the MEN2 syndromes with measurement of calcium levels for hyperparathyroidism and plasma fractionated

metanephrines for pheochromocytoma, 4-urinary fractionated metanephrines and catecholamine's [1, 4]. Appropriate imaging can be additionally used if any of these are suspected.

As with all neoplasms treatment varies according to the stage of disease. Table 1 gives the details on the TNM staging adopted by The Union for International Cancer Control (UICC) and the American Joint Committee on Cancer (AJCC) [1]. Our patient was classified as Stage 2 and the optimum treatment was offered through a subtotal thyroidectomy.

The Evasiveness of Follow-Up

The case reported presents the question of when a postoperative rise in calcitonin should be considered as significant and when to provide further treatment. It is important to note that there are alternative causes for a rise in calcitonin. False-positive calcitonin results may be obtained in patients with hypercalcemia, hypergastrinemia, neuroendocrine tumors, renal insufficiency, papillary and follicular thyroid carcinomas, goiter, and chronic autoimmune thyroiditis [5]. Drugs that can cause a false positive result include omeprazole, beta blockers and glucocorticoids [5].

Kebebew et al, followed a 104 patients over an average of eight years and found through multivariate analysis that age and stage of disease are independent prognostic factors, meaning that younger age and an earlier stage of disease allow greater survival [6]. Pelizzo discovered the same after a 37-year follow-up of a 157 patients but also add extent of surgery as a prognostic factor [7]. This implies that a more advanced tumor requires more extensive surgery and both relate to poor prognosis. Furthermore, Kebebew et al., report that patients with postoperative hypercalcitoninemia without clinical or radiologic evidence of residual tumor after apparently curative surgery may enjoy long term survival, but elevated calcitonin can be attributed to occult disease [6].

The recommended method of postoperative follow-up is through measurement of serum calcitonin and CEA after two to six months, and normal serum level of these hormones indicates a biochemical cure [4, 8]. Successful surgery and a biochemical cure have been associated, with less than 4% risk of recurrence during follow-up [3]. In patients that are surgically cured, calcitonin levels begin to rapidly decline within the first postoperative hour [8]. However, it is important to correctly time the measurement since calcitonin levels in some patients can sometimes take several months to normalize [8].

Table 1: Pathological TNM criteria

	Thyroid carcinoma
STAGE 1	Less than 2 cm in diameter without evidence of disease outside of the thyroid gland.
STAGE 2	Between 2 and 4 cm without evidence of extra thyroidal disease.
STAGE 3	Greater than 4 cm, or level VI nodal metastases or microscopic extra thyroidal invasion regardless of tumor size.
STAGE 4	Any distant metastases, or lymph node involvement outside of level VI, or gross soft tissue extension.

If a high calcitonin level is detected six or more months following surgery than residual disease should be suspected [8]. However, considering the risks and difficulty of further and sometimes-radical neck surgery, it is then required to decide the degree of residual disease. Calcitonin values which are detectable but less than 150 pg/mL indicate persistent loco-regional disease in the neck [8]. To locate this disease, neck ultrasound and further imaging including CT scan and MRI scan should be used [8]. High-resolution ultrasonography is preferred because it is noninvasive, sensitive, and relatively inexpensive, and moreover, it can be used in acquiring a cytological diagnosis through FNAC [4].

It is recommended that in non-specialized institutions, only cases of overt disease without metastasis should undergo neck surgery [8]. If, on the other hand, macroscopic disease is not found through imaging and the calcitonin level remains constant then annual follow-up is required. This is done through a neck ultrasound and calcitonin measurement at sixth month intervals, with particular emphasis on determining the doubling time, which is a strong indicator of survival [3, 4]. The American thyroid association states that when there is no anatomic evidence of disease, despite detectable serum creatinine, the best option is observation [4]. Furthermore it recommends that, in the case of nonhazardous loco-regional disease, immediate intervention is of unknown benefit and such lymph nodes may be observed or undergo re-operative compartmental dissection of image or biopsy-positive compartments [4].

Further when calcitonin levels are higher than 150 pg/mL, distant metastasis is more likely and extensive imaging including neck and chest CT scan, liver MRI scan, bone scan, spine and pelvis MRI scans or PET/CT scan are required [4]. Although it is not the focus of this report, a brief mention regarding the treatment options for metastatic disease is necessary. Palliative treatments include surgery, hepatic embolization, external beam radiation therapy, and percutaneous interventions. Active treatment is recommended for lesions in vital locations, such as brain metastases, impending or active central nervous system compression, airway compromise, symptomatic lesions, hormonal secretion, and impending or active fracture of a weight bearing bone [4]. The management of patients with metastatic disease outside the neck remains controversial [9].

CONCLUSION

Postoperative follow-up and further management consist of measuring calcitonin levels, with the key number being at 150 pg/ml. Elevated biochemical levels of calcitonin should be correlated with evidence of macroscopic disease. Further management would range from follow-up in cases of elevated biochemical levels only to implementation of palliative care in cases of evidences of metastatic disease in addition raised calcitonin levels.

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Author Contributions

Mirza Faraz Saeed – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Nida Fatima Sakrani – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Isam Mazin Juma – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Abbas Ali – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Penetrating neck trauma in children causing aerodigestive tract injury: A case presentation

Aram Baram, Fahmi Kakamad, Fitoon Yaldo, Twana Kareem

ABSTRACT

Introduction: Pediatric traumatic esophageal perforation has high morbidity and mortality rates. It is most commonly iatrogenic in nature with penetrating trauma account for only 0.5% of cases. We report a case of pediatric penetrating trauma to the neck (zone two) causing both through and through cervical esophageal injury and laceration of the posterior wall of the trachea. Case Report: A six-year-old boy suffering from penetrating neck trauma due to blast explosion presented to us with severe respiratory distress and drooling of saliva. Diagnostic workup revealed both tracheal and esophageal injury. Immediate primary repair was done for both the organs. Postoperatively, he developed ARDS which treated conservatively with good outcome. Conclusion: High index of suspicion is always required not to miss any injuries especially esophageal injuries, bearing in mind whenever you have one injury searching for others is rationale. Tracheostomy tube whenever applied in children needs more meticulous care than adult population.

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INTRODUCTION

Pediatric penetrating neck traumatic has high morbidity and mortality rates [1]. Pediatric esophageal perforation is most often due to iatrogenic instrumentation, foreign body impactions and rarely trauma. External penetrating trauma is infrequent [2, 3]. While no definite data available for pediatric cervical esophageal perforation in current literature.

The signs and symptoms of early esophageal injury can be vague and nonspecific. The clinical presentation depends on the cause, location of the injury, size of the perforation, degree of contamination, length of time elapsed after injury, and presence of associated injury. Pain is the most common symptom (71%), followed by fever (51%), dyspnea (24%), and crepitus (22%) [4]. Signs and symptoms of upper air way obstruction are not recognized features of esophageal injury and whenever present they may indicate accompanying airway injury [5].

Plain chest and neck radiographs may show pneumomediastinum, subcutaneous emphysema, pleural effusions, and hydropneumothorax but they are not

sensitive enough to exclude the diagnosis of esophageal perforation. Contrast study remains the "gold standard" for excluding the diagnosis of an esophageal perforation [2]. Computed tomography scan and esophagoscopy are other diagnostic modalities that can be used when contrast radiography failed to demonstrate perforation despite of high clinical suspicion [4, 6].

Surgery has been the traditional and preferred treatment which aims to restore continuity, elimination of septic focus, provides adequate drainage, augments host defenses and maintains adequate nutrition [4].

Cervical esophageal perforation caused by blast injury is a rare presentation. We report a case of through and through cervical esophageal perforation accompanied by minor upper airway injury (aerodigestive tract injury) caused by shell injury in a six-year-old boy.

CASE REPORT

A six-year-old boy, victim of blast injury, was referred to our tertiary center seven hours after the accident. Tracheostomy has been done there as an urgent procedure for relieving upper airway obstruction. He sustained sharp nail injury to the left side of the neck, in zone II and outlet was to the right side of the back over the scapula. He was conscious but in distress, dyspneic, there was swelling, tenderness with subcutaneous emphysema all over the neck, more on left side. Chest examination showed decreased air entry on the right side. Abdominal and vascular examinations were unremarkable. His oxygen saturation (SPO_2) was 95% with 10 liters oxygen, pulse rate 130 beats/minute, temperature 38.5°C, blood pressure 90/60 mmHg, respiratory rate 60 cycles/minutes. Focused assessment with sonography for trauma (FAST) showed mild right side pleural collection. Chest and neck radiographies showed right upper opacity and subcutaneous emphysema. Computed tomography (CT) scan revealed fractured right first rib with apical hematoma, and right side hemothorax with pneumomediastinum. Patient was transferred to intensive care unit (ICU), sedation with 1 mg midazolam and 20 µg fentanyl done, right side tube thoracostomy was inserted which drained 200 cc of blood. After stabilization of the condition, gastrografin swallow was done which showed contrast leak at the mid-cervical esophagus (Figure 1).

Under general anesthesia with nasogastric tube, exploration was done via left longitudinal incision, ipsilateral internal jugular vein was found to be transected with through and through injury to the esophagus (Figure 2). The vein was ligated and the perforations in the esophagus was repaired using 3.0 vicryl in single layer, the right side perforation repaired from within the lumen and the left side perforation by 4 interrupted stiches. Postoperatively, nasogastric tube feeding was started with antibiotic coverage. In the second postoperative day, the tracheostomy tube blocked, resulted in aspiration and desaturation.

The child did not respond to conservative management. Rigid bronchoscopy was performed. Pus was found filling both major bronchi, bronchial lavage was done. Saturation was improved. Few hours later the patient condition deteriorated again, saturation decreased, chest examination revealed bilateral coarse crackles and chest-X-ray showed pictures in favor of pulmonary edema.

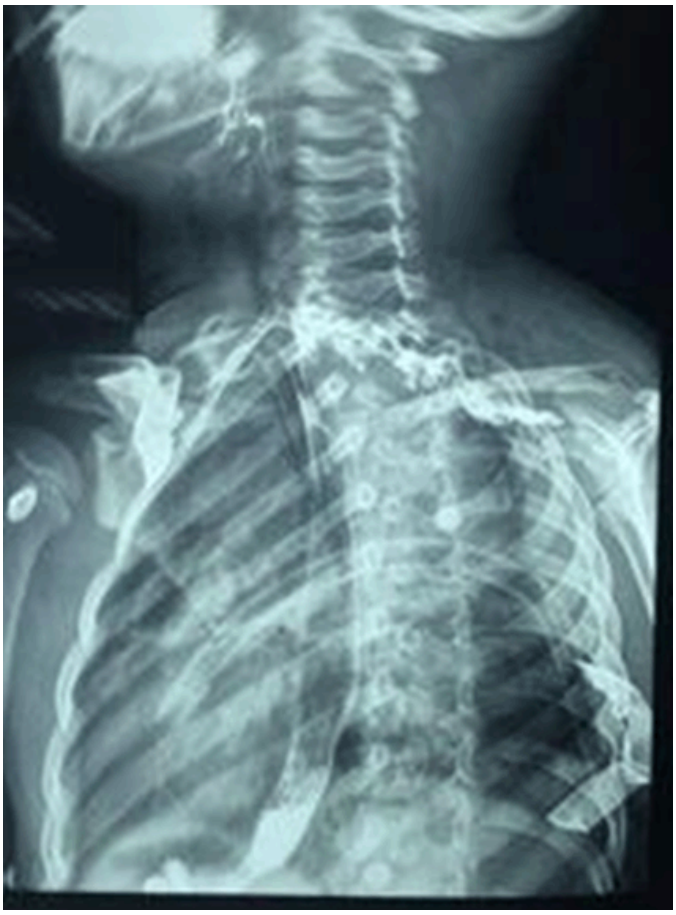

Figure 1: Gastrografin swallow showing contrast leak at the mid-cervical esophagus.

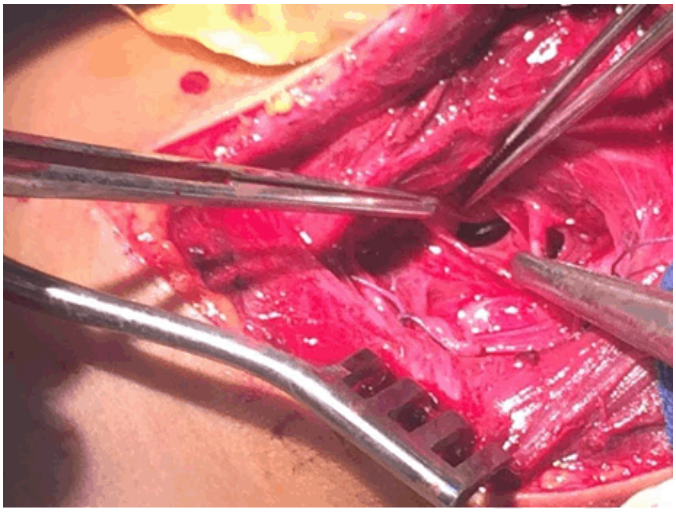

Figure 2: Showing through and through esophagus injury.

Supportive treatment was started; patient was put on continuous positive airway pressure (CPAP). He was weaned from CPAP four days later.

Water soluble contrast study showed no leak at seventh postoperative day. Oral intake started. Tracheostomy tube was removed at 10th postoperative day and he was discharged from hospital a day later.

DISCUSSION

Among all perforations of the alimentary tract, perforations of the esophagus are considered the most dire and life-threatening especially in children [2]. These perforations are:

(i) more often iatrogenic, (ii) more likely to occur within the cervical esophagus, and (iii) not generally associated with an underlying malignancy. Chest pain, fever, tachypnea, and/or tachycardia with subcutaneous emphysema are common features which were found in our case too [2].

Esophageal perforation is a surgical emergency associated with high morbidity and mortality. Consensus regarding the appropriate management of this life-threatening condition is lacking [7]. The reported mortality from treated esophageal perforation is 10–25%, when therapy is initiated within 24 hours of perforation and it is 40–60% when the treatment is delayed. The reason for this multifold increase in mortality is due to the unique anatomical configuration and location of the esophagus, which allows bacteria and digestive enzymes easy access to the mediastinum, leading to the development of severe mediastinitis, empyema, sepsis, and multiple organ dysfunction syndromes [7]. After immediate resuscitations and multisystem support, the child was taken to the operating room for cervical esophageal exploration.

The most common type of esophageal perforation is iatrogenic (approximately 60% in most series), usually as part of endoscopic therapy for stricture or achalasia. Barogenic or Boerhaave syndrome make up about 15–30% of cases, with trauma, foreign body ingestion, and operative injury accounting for most of the remaining benign perforations [8]. Penetrating injury as a causal factor is one of the rare causes [8]. To our knowledge, there is no reported case in literature about blast injuries causing through and through cervical esophageal perforation with minor upper airway trauma.

In our case, first complaint was confused with tracheal injury for which he underwent tracheostomy in the first hospital, and it was one of the morbidity factors as it is well known that tracheotomy tube occlusion is a common problem, occurring at a rate of up to 72% of premature and newborn children and, less frequently, at a rate of up to 14%, in children one year and older which also emphasizes the need for meticulous monitoring for pediatric patients to avoid such disastrous events [9]. Our case developed suffocation and aspiration from tracheostomy tube obstruction despite had been in intensive care unit with continuous nursing care.

The shell tract in this patient was also unusual being its inlet from zone II on the left side, and its outlet from the right side of the back, causing fracture of the right first rib with neither neurological deficits nor vascular injuries. Intraoperatively, the edges were healthy, no debridement was needed, and repair was done after good irrigation by normal saline by single layer using absorbable suture material.

CONCLUSION

High index of suspicion is always required not to miss any injuries especially esophageal injuries. Tracheostomy tube whenever applied in children needs more meticulous care than adult population.

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Author Contributions
Aram Baram – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Fahmi Kakamad – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Fitoon Yaldo – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Twana Kareem – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Perforated gangrenous cholecystitis with concurrent *Clostridium perfringens* bacteraemia masquerading as adenomyomatosis of the gallbladder: A case report

Natalie LY Ngu, Alexander Olaussen, Jessica Wong, Hayden Snow, Mark Cullinan, Paul J. Sitzler

ABSTRACT

Introduction: *Clostridium perfringens* (*C. perfringens*) is an unusual cause of bacteraemia in the healthy, immunocompetent host. Similarly, acalculous cholecystitis is rare in the absence of critical illness or preceding trauma. We present, to the best of our knowledge, the first documented case of concurrent *C. perfringens* bacteraemia and acalculous cholecystitis in a previously well human. Case Report: An apparently healthy 56-year-old male was presented with sepsis of unknown origin, and was treated for a respiratory infection and incidentally found to have gallbladder mural thickening on a computed tomography (CT) chest scan. An abdominal ultrasound (USG) demonstrated adenomyomatosis of the gallbladder, without evidence of acute cholecystitis or gallstones. The initial blood sample hemolyzed, however, subsequent specimens showed raised inflammatory markers, neutrophilia and thrombocytopenia. The patient continued to deteriorate clinically and biochemically. At 30 hours from presentation, blood cultures demonstrated a *C. perfringens* bacteraemia and intravenous antibiotics were commenced. Following these findings and the development of right upper quadrant abdominal pain, biliary sepsis was suspected and the patient taken to theatre. During laparoscopic cholecystectomy, a perforated and gangrenous gallbladder was identified and the intraoperative cholangiogram demonstrated no retained stones. Acute gangrenous cholecystitis was confirmed on histopathology. Postperatively, the patient recovered quickly and was discharged with oral antibiotics. Conclusion: We present a case of acalculous cholecystitis in a patient with an unusual clinical presentation and lack of positive imaging findings. In this setting, the need for definitive surgical intervention and clinical suspicion of cholecystitis was recognized with the finding of *C. perfringens* bacteraemia despite imaging suggesting adenomyomatosis. This case highlights that acalculous cholecystitis can occur in patients without risk factors, and can be complicated by atypical bacteraemia, even in previously healthy individuals.

Keywords: Acalculous cholecystitis, adenomyomatosis, Bacteraemia, *Clostridium perfringens*

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INTRODUCTION

C. perfringens is a potentially dangerous gram positive anaerobic rod, causing disease through toxin release. The incidence has been estimated to be less than 2 per 100,000 [1] and mortality rates have been reported between 27% and 44% [1]. Recognized sources of *C. perfringens* bacteraemia include wound contamination and iatrogenic bowel leakage, and can be complicated by hemolysis and overwhelming infection if treatment is delayed [2].

Acute cholecystitis commonly presents with a syndrome of right upper quadrant abdominal pain, fever and leukocytosis, and is usually attributed to gallstones [3]. Common organisms associated with cholecystitis include *Escherichia coli*, *Klebsiella* spp. and *Enterococcus faecalis* [4]. Acalculous cholecystitis has a similar clinical picture but in the absence of gallstones. It accounts for 2–15% [5] of acute cholecystitis cases and often presents in critically ill patients or with severe systemic stress e.g., trauma, major surgery, shock or burns [6]. Acalculous cholecystitis is more frequently associated with *Pseudomonas*, Staphylococci including methicillin-resistant *Staphylococcus aureus*, *Enterobacter*, *Bacteroides* spp. and fungi [4].

We present a diagnostic dilemma of concurrent acalculous cholecystitis inaccurately identified on ultrasound and *C. perfringens* bacteraemia in a previously healthy individual, with implications for future diagnosis.

CASE REPORT

A 56-year-old male presented to the emergency department with a history of one day of severe chest and epigastric pain, fevers and rigors. His medical history included ischemic heart disease, peptic ulcer disease and gastroesophageal reflux disease.

On examination, he was febrile (39.4°C), demonstrated tachycardia of 110 beats per minute, tachypnea of 26 breaths per minute, an oxygen saturation of 92% on one litre of supplemental oxygen and was normotensive. His chest and abdominal examinations were normal.

An initial blood sample was hemolyzed prior to analysis. Subsequent blood tests revealed a mild neutrophilia, raised C-reactive protein (CRP) and thrombocytopenia (Table 1). All other blood levels including liver function tests were normal. Since a chest X-ray demonstrated no abnormality, a CT pulmonary angiogram was performed for suspected pulmonary embolism, however, the only salient findings were gallbladder wall thickening and a small volume of pericholecystic fluid. An abdominal USG suggested adenomyomatsis of the gallbladder without features of cholecystitis. Gallstones were not visualized on either scan. Empirical intravenous antibiotics (ceftriaxone 1 g and azithromycin 500 mg) were commenced.

The patient continued to be febrile overnight with mild right upper quadrant abdominal pain developing. The following morning, the CRP had increased and the platelet count had fallen further (Table 1). Preliminary blood cultures taken at time of presentation, demonstrated a *C. perfringens* bacteraemia at 30 hours from presentation, and antibiotics were changed to intravenous tazobactam/piperacillin 4.5 g and clindamycin 600 mg. This positive blood culture result and the location of abdominal pain greatly increased suspicion for biliary sepsis, and the patient was taken to theatre. Intraoperatively, a perforated and gangrenous gallbladder was removed laparoscopically and a cholangiogram demonstrated no filling defects. The patient received a single pack of pooled, irradiated platelets, due to refractory bleeding from the gallbladder fossa. Postoperatively, the thrombocytopenia had improved and the patient was discharged with oral antibiotics (amoxicillin/clavulanate 875/125 mg). Acute gangrenous cholecystitis without gallstones was confirmed on histopathology (Figure 1). At a two-week follow-up appointment, all blood abnormalities had resolved to within normal range (Table 1).

DISCUSSION

This case represents a clinical dilemma given the unusual presentation and difficult pathway to diagnosis. The diagnosis of acalculous cholecystitis was finally made based on intraoperative findings of a perforated and gangrenous gallbladder, the normal cholangiogram and the subsequent histopathologic analysis of the specimen. As outlined in Table 2, the commonly

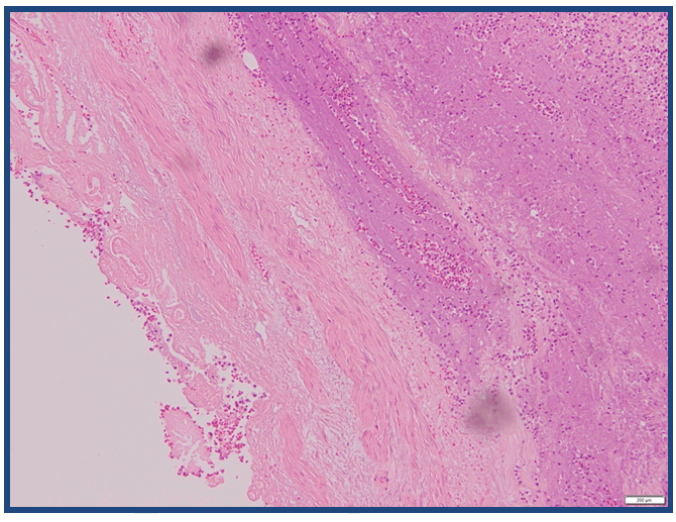

Figure 1: Necrotic gallbladder wall with area of mural suppuration (lumen on left), (H&E stain, x40).

Table 1: Relevant blood test results

	Day 1	Day 2	Day 3	Day 15	Reference Interval
Hemoglobin (g/L)	144	131	114	132	128–175
White Cell Count (x 10^9/L)	11.40	13.79	14.97	6.95	3.90–12.70
Platelets (x10^9/L)	106	82	110	174	150–396
C-Reactive Protein (mg/L)	26	208	273	NA	0–5
Neutrophils (x10^9/L)	9.26	11.75	13.02	3.67	1.90–8.00
Bilirubin (umol/L)	11	14	10	6	≤21
ALT (units/L)	17	13	43	28	≤40
GGT (units/L)	14	16	17	20	≤62
ALP (units/L)	62	60	68	84	30–110
Lipase (units/L)	15	NA	NA	NA	10–70

Abbreviations: NA Not available

Table 2: Organisms commonly associated with diagnosis [4, 5, 10]

Calculous Cholecystitis	Acalculous Cholecystitis	Emphysematous Cholecystitis
Escherichia coli, *Klebsiella* spp., *Enterococcus faecalis*	*Pseudomonas* *Staphylococci* (including MRSA) *Enterobacter* *Bacteroides* spp. Fungi.	*Clostridium* spp.

Abbreviations: MRSA Methicillin-Resistant *Staphylococcus aureus*

associated organisms vary between calculous, acalculous and emphysematous cholecystitis. Although *Clostridium* spp. has been associated with secondary emphysematous acalculous cholecystitis in critically ill patients or following antibiotics [3], no features suggestive of emphysematous acalculous cholecystitis including air in the gallbladder lumen or wall were seen on imaging in this case. In addition, the absence of a preceding physiological stress or immunocompromise is unusual in the development of both acalculous cholecystitis and *C. perfringens* bacteraemia.

Although no other case of acalculous cholecystitis and *C. perfringens* bacteraemia has been reported in human literature, there is a similar published case of a pig with undifferentiated sepsis, a necrotic gall bladder without gallstones on autopsy and hemolysis of initial blood samples [6]. A human case of *C. perfringens* bacteraemia with clinical suspicion of a biliary source, was attributed to gallstones, which were confirmed on endoscopic retrograde cholangiopancreatography [7]. These examples highlight the atypical presentation in our patient and the need to consider all clinical features when faced with a similar diagnostic dilemma.

The significance of the patient's thrombocytopenia remains unclear, however, a corresponding phenomenon has been reported in a retrospective cohort study of 93 patients with *C. perfringens* [1]. Additionally, hemolysis of an initial blood sample has been reported in another case of *C. perfringens* bacteraemia [8]. This may be attributed to toxin release [9]. However further exploration of these two associations is needed.

CONCLUSION

C. perfringens bacteraemia is a rare and potentially serious condition. Acalculous cholecystitis can occur despite abdominal ultrasonography suggesting an

alternative diagnosis. This case highlights that acalculous cholecystitis can occur in patients without risk factors, and can be complicated by atypical bacteraemia, even in previously healthy individuals.

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Author Contributions
Natalie LY Ngu – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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Hayden Snow – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published
Mark Cullinan – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published
Paul Sitzler – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

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Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Vancomycin-associated neutropenia in hemodialysis patient with sepsis

Esraa Shukri Altawil, Fadi Saleh Aljamaan

ABSTRACT

Introduction: Vancomycin-induced neutropenia is a rare but serious adverse reaction that appears to occur with prolonged use with a minimum of seven days. In most cases, it has been reported to resolve spontaneously upon discontinuation of vancomycin. Case Report: A 53-year-old male admitted as a case of septic arthritis complicated with sepsis in which vancomycin induced prolonged neutropenia occurred after nine days of therapy and was associated with toxic serum level. Conclusion: Vancomycin induced neutropenia may occur at any dose or duration of therapy, but generally it is associated with prolonged use for more than seven days. Our findings suggest that clinicians should be cautious about it as it can occur at any time in the course of treatment, and may last for longer time especially in hemodialysis patients in addition to being linked to the toxic serum level. All patients who receive vancomycin for more than seven days should have white blood cells count and differential counts monitored weekly. Discontinuation of the drug appears to be prudent as soon as this hematological abnormality detected.

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INTRODUCTION

Vancomycin-induced neutropenia, defined as an absolute neutrophil count (ANC) less than 1000 cells/mm^3, is a rare but serious adverse reaction has been reported to occur at rates of 2–12% [1, 2]. In 1960, shortly after the introduction of the drug, Dangerfield et al. published the first two case reports [1].

The onset of neutropenia does appear to occur with prolonged treatment duration with at least seven days and mostly occurring at 20th day after therapy initiation [3]. Although the mechanism of action of the neutropenia is controversial, several theories have been proposed including an immunological mechanism with an IgG or IgM immune-mediated hypersensitivity reaction [4]. Evidence to support this hypothesis includes studies examined the bone marrow of patients with vancomycin induced neutropenia which reveal granulocyte-specific antibodies [5, 6]. Another suggested mechanism is direct toxicity to the bone marrow. This has been supported by hypo-cellular marrow with maturation arrest on bone marrow biopsy [7].

In all of the cases, neutropenia has been reversible and resolve spontaneously upon withdrawal of vancomycin.

Improvement generally occurs within a few days of vancomycin discontinuation [2], but, in patients who are undergoing hemodialysis, prolonged neutropenia for up to 4 weeks has been observed [8].

CASE REPORT

A 53-year-old male, a known case of diabetes mellitus and hypertension for 20 years, presented with ischemic heart disease for the last 12 years, and chronic kidney disease for the last five years. He was admitted to the hospital as a case of left shoulder septic arthritis as a complication of intra-articular steroid injection for tendonitis, started empirically on vancomycin 1 g IV every 24 hours and clindamycin 600 mg IV q 8 hr. Two days later, blood culture revealed methicillin sensitive staphylococcus aureus (MSSA) at which both antibiotics stopped and cloxacillin 2 g IV every 4 hr was initiated.

Throughout his hospital stay he developed acute on top of chronic renal failure requiring hemodialysis. On day-25 he developed hypotension during dialysis session, associated with decreased level of consciousness (LOC), which mandated transfer to intensive care unit. Furthermore, he developed acute respiratory distress syndrome (ARDS) requiring mechanical ventilation in addition to severe circulatory failure all of that due to septic shock. The patient's intravenous (IV) antibiotics were escalated to meropenem 1 g IV q 12 hr and vancomycin 1 g IV q 24 hr (target trough level 10–15) with full septic screen sent.

The daily vancomycin trough level continued to be supra-therapeutic (range 17–28 µg/mL), given patient's poor renal function, mandating several times of dose holding. Despite the maximum therapeutic care provided, patient's condition started to deteriorate manifested by severe hemodynamic instability demanding maximum doses of vasopressors. On day-31, his blood gases analysis was consistent with severe hypoxemia in addition to lactic acidosis. Laboratory results showed neutropenia with ANC 200 cells/mm^3 (baseline was 6,200 cells/mm^3), in which vancomycin stopped after total duration of nine days (cumulative dose received 5 g), and filgrastim (G-CSF) 300 µg SC started. Patient remained on meropenem and cloxacillin resumed over. Blood culture showed *Pseudomonas aeruginosa* for which colistin was added (loading dose of 9 million units IV followed by maintenance dose 2 million units IV every 8 hr). The neutrophil count continued to fall to < 100 cells /mm^3 reaching undetectable amount even after he received seven doses of G-CSF (Figure 1). On day-40, the patient had worsening multiorgan failure and was declared for comfort care ending with cardiorespiratory arrest.

DISCUSSION

Several published case reports demonstrated neutropenia as an adverse event associated with

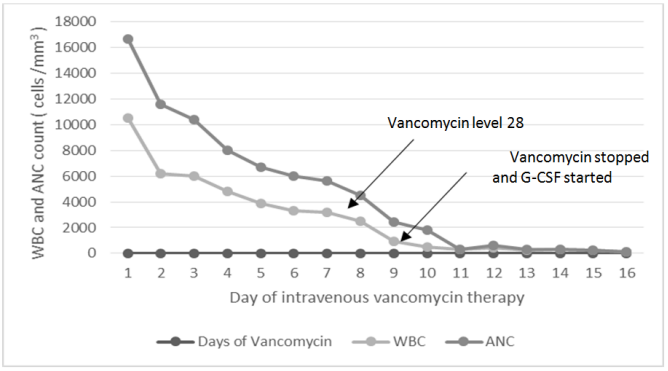

Figure 1: Total white blood cell count and absolute neutrophil count for the patient, G-CSF 300 µg subcutaneous was initially administered on day-9 on daily basis (total seven doses).

vancomycin therapy, nevertheless, to date there are no published prospective studies assessing this important clinical complication. Neutropenia may occur at any dose, but generally with the prolonged vancomycin use for more than seven days. To our knowledge, this is the first case report addressed vancomycin induced neutropenia in Saudi Arabia.

Many of the cases reported included patients receiving concomitant drugs also known to cause neutropenia, resulting in uncertainty with the influence of vancomycin. Based on the Naranjo probability scale, the association of vancomycin with neutropenia was considered as probable in our case. In addition to delayed onset, vancomycin induced neutropenia has always been reported as reversible, with complete resolution taking place in 2–5 days which is not the case. Factors, other than drug related, including sepsis and hemodialysis could be the cause of our patient's prolonged neutropenia.

Latest American Society of Health-System Pharmacists and the Infectious Diseases Society of America (ASHP/IDSA) guidelines did not indicate the need of frequent monitoring of leukocyte count while receiving vancomycin [9]. Although there are no guidelines or recommendations exist for the use of G-CSF in vancomycin-induced neutropenia, several case reports have shown successful treatment of vancomycin-induced neutropenia with G-CSF while continuing use of vancomycin [4]. It is crucial that patients undergoing prolonged vancomycin therapy remain under close monitoring with at least once weekly laboratory testing for blood counts.

At present, it is uncertain whether patients with history of vancomycin-associated neutropenia would develop the same response if re-challenged with vancomycin. Koo et al. has reported a successful case where a patient who recovered from vancomycin induced neutropenia restarted on a five days course and at a lower dose without recurrence of neutropenia. In most circumstances, clinicians prefer to use an alternative agent rather than resuming the offending agent. Further studies are needed to conclude the possibility of neutropenia with repeat exposure to vancomycin [10].

CONCLUSION

Vancomycin induced neutropenia may occur at any dose, but generally with the prolonged vancomycin use for more than seven days. Our findings suggest that clinicians should be cautious that vancomycin-associated neutropenia can occur at any time in the course of treatment, and can be linked to the toxic serum level. All patients who receive vancomycin for more than seven days should have white blood cell and differential counts monitored weekly. Discontinuation of the drug appears to be prudent as soon as this hematological abnormality detected.

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Author Contributions

Esraa S. Altawil – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Fadi S. Aljamaan – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Salmonella spondylitis in an immunocompetent non-sickle cell patient

Zaid B. Al Jebaje, Andrew Zhao, Mohammed Samannodi, Mohammed Al-Sofiani, Michael Hocko

ABSTRACT

Introduction: Osteomyelitis caused by *Salmonella* is rare, especially in immunocompetent patients, as it typically occurs in patients with sickle cell anemia, HIV, corticosteroid use, or any other conditions that can compromise the immune system. The objective of this report is to establish *Salmonella* as a potential causative agent in the differential diagnosis of osteomyelitis for clinicians. Case Report: We report a 25-year-old male who does not have any history of sickle cell anemia, corticosteroid use, or HIV who presented with back pain and constitutional symptoms suggestive of an ongoing infection. Laboratory workup, X-ray, magnetic resonance imaging, and fluoroscopic biopsy of the affected area helped establish the diagnosis and guide treatment options. The condition of patient improved after extended course of antibiotics. Conclusion: Patients with *Salmonella* spondylitis typically present with back pain and fever. Initial diagnostic workup should include complete blood count (CBC), C-reactive protein (CRP), and erythrocyte sedimentation rate (ESR) levels. X-ray can be used for initial imaging but MRI will better visualize the full extent of the infection while guided needle biopsy with cultures will distinguish the organism and its antimicrobial susceptibilities. Extended antibiotic coverage for 6–12 weeks is often needed and recommended.

Keywords: Immunocompetent, Osteomyelitis, *Salmonella*, Spondylitis

How to cite this article

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INTRODUCTION

Salmonella is a gram-negative bacillus that is most commonly associated with gastrointestinal infections worldwide and typhoid/paratyphoid fevers in less developed countries. In addition, *Salmonella* can also rarely cause spondylitis, defined as osteomyelitis of the spine with destruction of the vertebral bodies, but it is associated with only 0.5% of all osteomyelitis cases [1]. The majority of said cases are frequently associated with patients with immunocompromised states, such as malignancies, long-term corticosteroid use, and sickle cell anemia. We report a case of a 25-year-old

immunocompetent non-sickle cell African-American male with chronic lower back pain diagnosed as *Salmonella* non-typhoidal spondylitis.

CASE REPORT

A 25-year-old African-American male presented to the Sisters of Charity hospital with complaints of progressively worsening lower back pain for six months. The pain started as mild back ache that did not have significant effect on the man's daily activities but gradually worsened, causing the patient to seek medical help. He visited multiple local healthcare facilities where he was diagnosed with muscle strain and treated with non-steroidal anti-inflammatory drugs (NSAIDs) and muscle relaxants.

During the course of his progressive pain, the man started to notice increasing back stiffness and difficulty in walking. Two days prior to presentation, he also started experiencing fever, chills and generalized tiredness. The patient denied any recent trauma, weight loss, nausea, vomiting or diarrhea, or urinary symptoms. He also denied consumption of unpasteurized milk or undercooked poultry. Patient denies recently interacting with any known sick contacts. He had a negative past medical and surgical history and any history of illicit drug use or owning pet animals.

On examination, the patient had a temperature of 39.0°C, pulse rate 98 BPM, blood pressure 125/80 mmHg and respiratory rate 16 breaths/minute. On musculoskeletal examination, he had mild midline lumbar spine and para-spinal tenderness. Spinal range of movement examination revealed mild pain and stiffness on flexion. Hips were stable and straight leg raising test was negative bilaterally. Neurological examination and remainder of physical examination was unremarkable in its entirety.

Initial laboratory blood work were drawn and results returned as followed: hemoglobin 12.1 g/dL, hematocrit 37.1%, white blood cells count $8.6x10^3$/ul, platelets $157x10^3$/ul , C-reactive protein 108 mg/L and erythrocyte sedimentation rate 66 mm/hr. Renal and liver function tests were unremarkable, coagulation profile within normal range, and sickle cell screen negative. Hemoglobin electrophoresis was also normal. Cultures of blood, stool and midstream urine later returned negative. Serum anti-nuclear antibody (ANA), perinuclear anti-neutrophil cytoplasmic antibodies (pANCA) and cytoplasmic anti-neutrophil cytoplasmic antibodies (cANCA) were all negative. HIV testing results returned negative as well and CD4 count was within normal limits.

X-ray of the lumbar spine was negative for any bony erosion or soft tissue abnormality. Magnetic resonance imaging (MRI) scan of the lumbar spine showed enhanced abnormal signal involving L4 and L5 anteriorly with thickening of anterior longitudinal ligament and sparing of disc space a picture likely consistent with infectious spondylitis (Figure 1).

The patient underwent a fluoroscopy guided needle aspiration and biopsy of L4/L5 space and received intravenous ceftriaxone 2 g daily. The biopsy results returned positive for *Salmonella* non-typhoid group, sensitive to ceftriaxone and ciprofloxacin.

Basing the working diagnosis on spondylitis secondary to a *Salmonella* non-typhoid infection, our decision was to continue the patient on intravenous

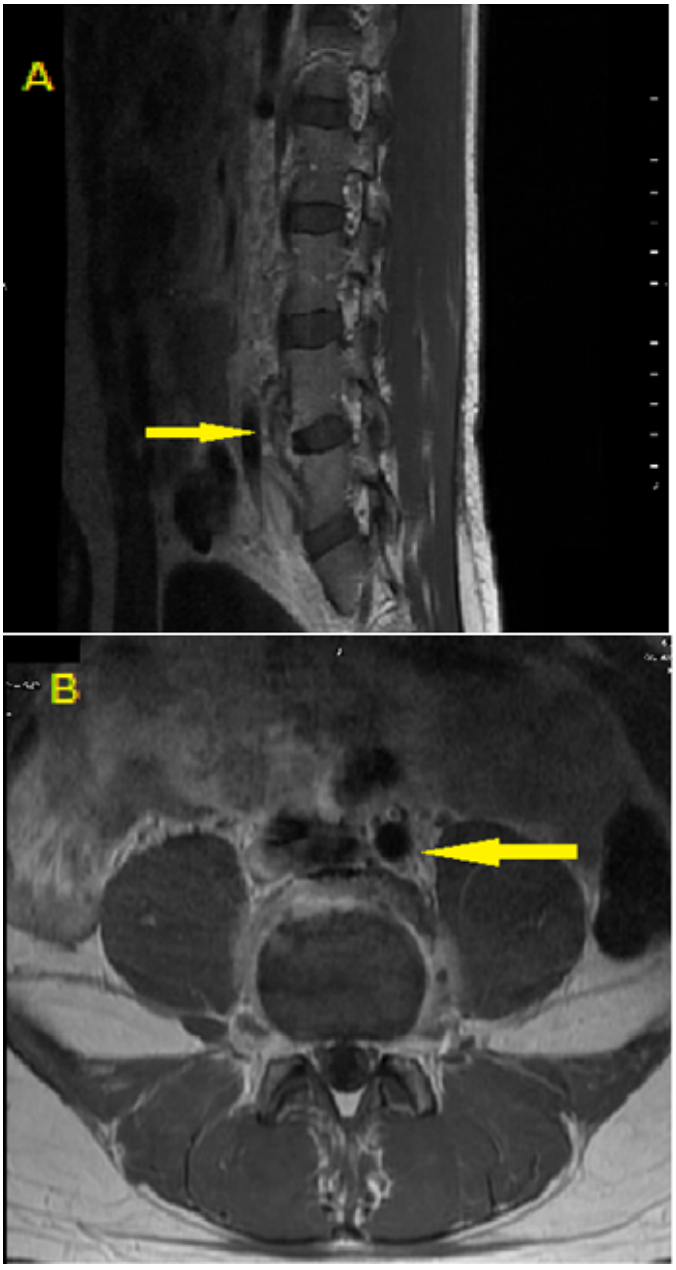

Figure 1: Magnetic resonance imaging scan showing (A) sagittal, and (B) axial sections of the lumbar spine with 15 cc of ProHance. The disc spaces does not show any narrowing while L4/L5 junction shows morrow edema with enhancement of anterior endplates and thickening of anterior longitudinal ligament; overall picture is suggestive of disc sparing infectious spondylitis.

ceftriaxone for six weeks, followed by another six weeks of oral ciprofloxacin. During his recovery, he also wore a thoracolumbar brace for spinal immobilization. His clinical presentation and inflammatory markers started improving within several days. The patient was discharged to a physical rehabilitation facility with a peripherally inserted central line in place for intravenous antibiotic treatment. After three weeks of antibiotic therapy, his ESR was down to 14 mm/hr and CRP 2.2 mg/L. To date, he has finished his antimicrobial course and has remained asymptomatic.

DISCUSSION

Salmonella has been most commonly associated with gastrointestinal infections but it also holds the distinction of causing osteomyelitis in immunocompromised patients. Sickle cell disease (SSD) particularly stands out as the increased risk of *Salmonella* osteomyelitis in SSD patients has been well documented in medical literature. The accepted explanation of the underlying pathophysiology involves intravascular sickling of the bowel leading to ischemic infarction and development of vulnerable areas in the bowel for bacterial entry. This is then amplified by SSD patients' susceptibility for hyposplenism at a young age, causing a sub-functional immune system, and microvascular occlusions, leading to bone infarcts and necrosis [2, 3]. However, as demonstrated in this case, immunocompetent individuals can still rarely suffer from *Salmonella* osteomyelitis. Clinicians should strive for earlier diagnosis, stabilization, and medical/surgical intervention in these cases because of the mortality in patients with vertebral osteomyelitis [4].

The patient involved in this report suffered from progressive lower back pain, back stiffness, difficulty walking, and constitutional symptoms (fever, chills, and fatigue) later on. In the review by Santos and Sapico, fever and back pain were the main complaints in patients with *Salmonella* vertebral osteomyelitis, with fever present in 87% of cases and back pain 92% [5]. Review of current literature also supports this finding as an overwhelming majority of case reports also mention back pain and recurrent fevers as their patients' chief complaints [1, 4, 6–8]. At the same time, patients commonly did not recall any previous history of typhoid fever or gastroenteritis (as several weeks may have passed).

Clinicians are also recommended to pay close attention to the laboratory results of any patients presenting with lower back pain and fever because reliance on the white blood cell count alone might not be as helpful in diagnosis of *Salmonella* spondylitis. The patient involved in this report himself did not have an elevated white cell count but did show elevated C-reactive protein and erythrocyte sedimentation rate levels, which is once again consistent with past cases [1, 4–9]. However, a complete blood cell count should still be ordered in concurrence with ESR and CRP levels as patients can have other underlying conditions that require medical attention.

In terms of imaging modalities, the initial X-ray of thoracic/lumbar spine can help establish the presence of any abnormalities in the bone and soft tissue. Common findings in spondylitis include osteolysis, end plate erosions, and narrowing of the disk space. However, MRI scan provides better visualization of the spread of the infection and involvement of the soft tissues, neural column, and bone structure. Afterwards, patients should undergo needle biopsy, either fluoroscopic or computed tomography guided, in order to definitively diagnose the involving organism prior to starting antibiotics. This step is paramount as treatment options can vary widely with different suspecting organisms, Amritanand reported a patient that was started on antituberculous therapy because her biopsy did not yield enough tissue but her condition continued to worsen [7].

As stated above, antibiotic therapy should be initiated based on the organism and susceptibilities identified via cultures. Various studies have advocated different lengths of the treatment courses, ranging from 4–12 weeks [2–9]. A literature review by Grados demonstrated recurrence rates that were two to three times higher in patients who received antibiotics for four to eight weeks when compared to those who were treated for 12 weeks or longer [10]. The presented patient underwent a total of 12 weeks with ceftriaxone during the initial sex weeks and then ciprofloxacin for the reminder. Given the significant mortality associated with this condition, an extended antibiotic course of at least six to eight weeks should be considered along with close monitoring of both symptoms and inflammatory markers and a low threshold to reevaluate and restart therapy if symptoms return.

CONCLUSION

Although *Salmonella* osteomyelitis is extremely rare in immunocompetent patients, it should still be part of the differential diagnosis for patients with spondylitis. Patients presenting with fever and back pain should be evaluated for spondylitis. Initial blood work should include complete blood count (but anticipate that white blood cell count might not be elevated, C-reactive protein, and erythrocyte sedimentation rate. Diagnostic modalities include the initial spinal X-ray followed by spinal magnetic resonance imaging if indicated. Fluoroscopy or computed tomography guided needle aspiration with subsequent culture should be performed for definitive diagnosis. Patients with *Salmonella* spondylitis should receive antibiotics for at least six weeks with recommendations for 12 weeks or more.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Electrical storm: Is right ventricular pacing dangerous?

Daniel Mark Cooper, Kathleen M. Kennedy

ABSTRACT

Introduction: Electrical storm is commonly defined as the occurrence of three or more episodes of ventricular tachycardia (VT) or ventricular fibrillation (VF) in 24 hours. Patients with an implantable cardiac defibrillator (ICD) are at increased risk of electrical storm due to history of decreased ejection fraction and/or sustained VT/VF. In addition to medical treatment of electrical storm in ICD patients, special consideration should be given to ICD reprogramming to optimize hemodynamics by increasing basic pacing rate and maintaining atrioventricular as well as interventricular synchrony. If possible, anti-tachycardia pacing rather than repeated shocks can reduce sympathetic tone. Unnecessary right ventricular pacing may worsen left ventricular function by desynchronizing the ventricles and is generally avoided. Case Report: We present a case of a 67-year-old white male with severe electrical storm due to polymorphic ventricular tachycardia (PMVT) that was dramatically brought under immediate control by forced right ventricular pacing. Subsequent continuous right ventricular pacing helped suppress any ventricular tachycardia recurrence until catheter ablation was performed eight months later. Conclusion: When confronted with patients with refractory electrical storm, we propose programming a faster right ventricular pacing rate.

Keywords: Electrical storm, Implantable cardiac defibrillator, Polymorphic ventricular tachycardia, Right ventricular pacing, Ventricular desynchronization

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INTRODUCTION

Electrical storm is commonly defined as the occurrence of three or more episodes of ventricular tachycardia or ventricular fibrillation in 24 hours [1]. Patients with an implantable cardiac defibrillator (ICD) are at increased risk of electrical storm due to history of decreased ejection fraction and/or sustained VT/VF. In addition to medical treatment of electrical storm in ICD patients, special consideration should be given to ICD reprogramming to optimize hemodynamics by increasing basic pacing rate and maintaining atrioventricular as well as interventricular synchrony. If possible, anti-tachycardia pacing rather than repeated shocks can reduce

sympathetic tone. Unnecessary right ventricular pacing may worsen left ventricular function by desynchronizing the ventricles and is generally avoided.

We present a case of severe electrical storm due to PMVT that was dramatically brought under immediate control by forced right ventricular pacing.

CASE REPORT

A 67-year-old white male was admitted to a community hospital after developing syncope while driving. Previous history of myocardial infarction three years before, with subsequent coronary artery bypass graft x3 (internal mammary artery bypass to the left anterior descending artery, vein grafts to right coronary and circumflex). One year after, he was hospitalized for congestive heart failure (CHF) with 25% ejection fraction prompting the placement of a dual chamber ICD. Coronary angiography showed patent grafts, with distal native vessel disease. He remained on carvedilol, ramipril, aspirin, eplerenone, bumetanide. Patient experienced no shocks for two years until this syncopal episode.

Initial evaluation in the emergency room showed episodes of polymorphic ventricular tachycardia (PMVT), (Figure 1). Blood pressure was 157/68 mmHg, respirations 16, pulse 60 bpm, oxygen saturation 95% on room air. Electrocardiogram showed atrial pacing at 60 bpm with atrioventricular delay of 240 msec and intrinsic R wave with deep inferior Q waves T wave inversions in leads 1, L, V3-V6. QTC interval was 440 msec with subsequent tracings unchanged (Figure 2). Chest X-ray showed no CHF. Interrogation of ICD revealed seventeen episodes of ventricular tachycardia with rates from 240–300 bpm, correctly detected and shocked. Laboratory included potassium 3.7 (normal value 3.5–5.3 mmol/L), magnesium 1.9 (normal value 1.6–2.3 mg/dL), BUN 40 (normal value 5–25 mg/dL) and creatinine 1.3 (normal value 0.5–1.4 mg/dL). BNP was 132 (normal value <100 pg/mL). Initial troponin was 0.20, at two hours 0.60 and a 12 hour peak of 3.4 (normal value < 0.08 ng/mL).

Management included a bolus of 300 mg IV amiodarone followed by another 150 mg and a subsequent 1 mg/min drip. Lidocaine IV was given in boluses and titrated up to 4 mg/min drip. A total of 10 mg IV metoprolol was administered (in addition to his oral carvedilol). Four milligrams of magnesium and 20 mEq of potassium were given intravenously and sedation with IV lorazepam 0.5 mg in repeated doses. Respiratory status remained stable. Patient denied angina pectoris. Despite all measures, rapid PMVT continued.

Six hours after admission, the patient was transferred by helicopter to our hospital. Upon arrival to our Cardiac Unit, IV midazolam was given for sedation and lidocaine was stopped. Potassium and magnesium levels were 3.9 and 2.5 respectively. The episodes of PMVT persisted (Figure 3). Echocardiogram revealed a moderate size left ventricular posterior aneurysm and an ejection fraction of 25%. Implantable cardiac defibrillator was interrogated revealing 71 episodes of ventricular tachycardia correctly detected and shocked since his syncope. Implantable cardiac defibrillator was reprogrammed from a rate of 60 to 70 bpm, however PMVT reoccurred. Between PMVT episodes, he remained atrial paced with ventricular sensed rhythm. Subsequently, within approximately ten minutes, the atrioventricular delay was shortened from 250–140 msec (Table 1) which resulted in right ventricular pacing with an atrial and ventricular paced rhythm (Figure 4). PMVT immediately ceased.

Electrical storm completely subsided and the next steps taken within the first hour were an esmolol drip titrated up to 100 μg/kg/min, and continuation of intravenous amiodarone.

The next day, cardiac catheterization showed patent grafts with distal small vessel disease and extensive

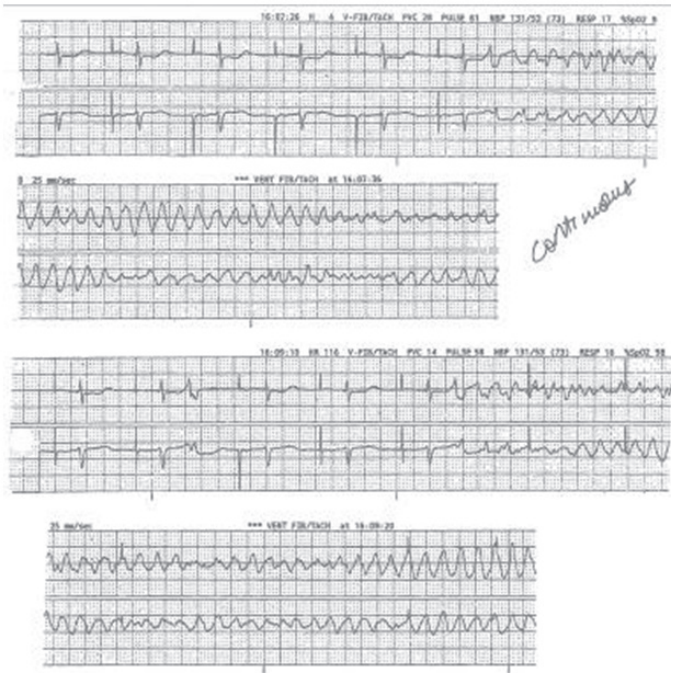

Figure 1: Rhythm strips (after treatment with intravenous amiodarone and lidocaine) demonstrate atrial paced rhythm 60 bpm with QT interval 440 msec degenerating into polymorphic ventricular tachycardia.

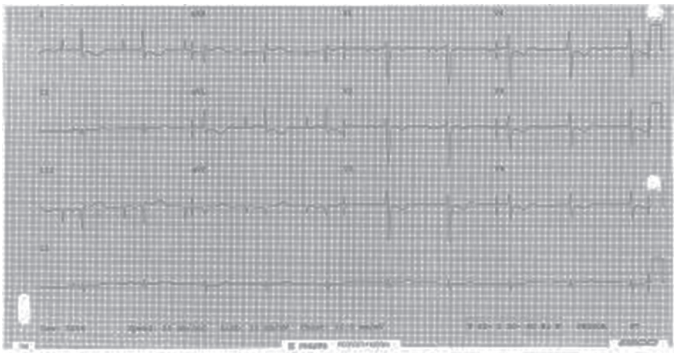

Figure 2: Electrocardiogram on admit to cardiac unit showing atrial pacing at 60 bpm with QT of 480 msec.

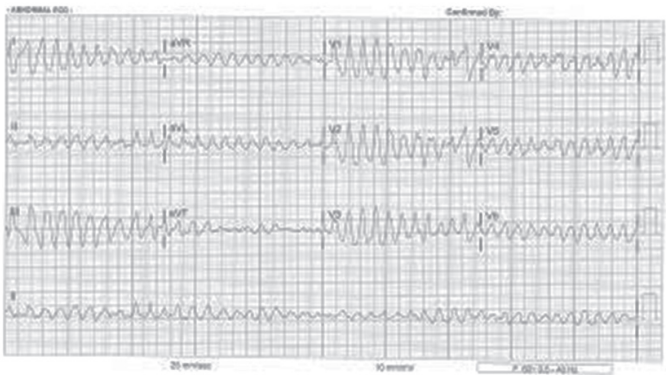

Figure 3: Electrocardiogram during one of the arrhythmia episodes showing polymorphic ventricular tachycardia with rate of 300 bpm.

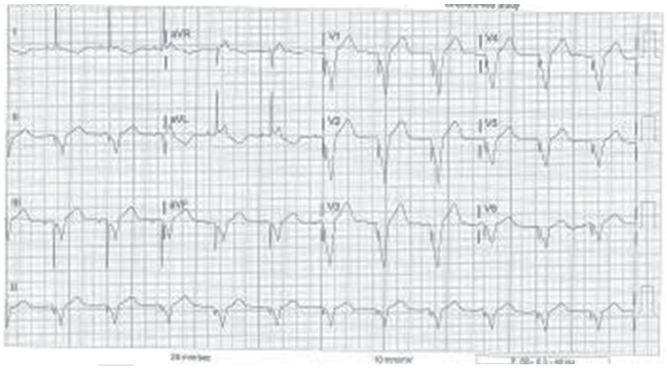

Figure 4: Follow-up electrocardiogram after ICD reprogramming showing forced right ventricular pacing after atrioventricular delay was shortened, leading to atrioventricular pacing at 70 bpm with no recurrence of polymorphic ventricular tachycardia.

Table 1: Implantable cardioverter defibrillator (ICD) programmed parameters

ICD Parameters	At Initial Admission	After Reprogramming
Basic Pacing Rate	60 bpm	70 bpm
Atrioventricular delay	250 milliseconds	140 milliseconds
Atrioventricular pacing	25% atrial paced, <1% ventricular paced	99% atrial paced, 93% ventricular paced
Ventricular tachycardia zone	Not programmed	160 bpm
Ventricular fibrillation zone	200 bpm	200 bpm

collateral circulation from septal branches. No culprit vessels were found. A noninvasive electrophysiology study showed no inducible ventricular tachycardia. Medications at discharge included amiodarone, carvedilol, ramipril, eplerenone, furosemide, potassium and dabigatran.

Follow-up at two months revealed a stable status, NYHA class 2 and unchanged ejection fraction 25% despite right ventricular pacing 100%. Implantable cardiac defibrillator interrogation demonstrated no ventricular tachycardia and amiodarone was reduced to 100 mg daily. Eight months after the admission, elective radiofrequency ablation with substrate modification of the left ventricular posterior scar was performed successfully. 3D bipolar and unipolar voltage maps within posterior basal wall scar, guided the radiofrequency ablation of the fractionated potentials using a Biosense Webster Navistar Thermocool catheter. Subsequently, right ventricular pacing was discontinued by extending atrioventricular delay. A two year follow-up confirmed the patient remained free of ventricular tachycardia.

DISCUSSION

Electrical storm is estimated to occur in approximately 10% of ICD patients and is associated with unfavorable long-term prognosis [2]. Acute treatment of electrical storm in ICD patients includes beta blockers, amiodarone, benzodiazepines, and electrolyte management. Potential causes of recurrent ventricular tachycardia in patients with an ICD include myocardial ischemia, drug and electrolyte induced QT prolongation. It is essential to rule out device malfunction from acute lead dislodgement, sensing of electrical noise triggering anti-tachycardia pacing or shocks. Furthermore, VT/VF in ICD patients may be a pacing related phenomenon where short-long-short sequences may initiate reentry and indicate a need for a change in pacing mechanism [3].

Catheter ablation of an arrhythmogenic focus has been used for treatment of electrical storm in ICD patients [4]. However, most of the experience is with monomorphic ventricular tachycardia [5]. Furthermore, ablation for hypotensive polymorphic ventricular tachycardia can be challenging. Thus, alternative options for acutely ceasing an incessant PMVT must be sought.

Recently, a multicenter randomized trial has compared elective ventricular tachycardia ablation versus escalated medical therapy in patients with ICD, ischemic cardiomyopathy and recurrent ventricular tachycardia. The findings showed a significant lower rate of death and ventricular tachycardia in the catheter ablation group [6].

In patients with left ventricular dysfunction with an ejection fraction < 35% and a wide QRS, left ventricular pacing can improve clinical status by optimizing hemodynamics with interventricular synchrony. Even though resynchronization and early left ventricular depolarization has helped improve ventricular function in patients with heart failure and left ventricular delay, changing the activation sequence of the ventricle by pacing also has potential electrophysiological effects which can be proarrhythmic [3].

We hypothesize, in our patient with incessant PMVT and a left ventricular aneurysm, that early depolarization by pacing the right ventricle was beneficial. Right ventricular pacing may delay activation of the left

ventricular aneurysm, which may have rendered the surrounding ventricle refractory or less susceptible to activation from the aneurysmal region. Override of the left ventricular arrhythmogenic focus, which we believed to be the left ventricular aneurysm, redirected depolarization and essentially desynchronized his ventricles. Unnecessary right ventricular pacing has been shown to diminish cardiac function over time and exacerbate heart failure in patients with structural heart disease [7, 8]. This case however, raises the question of whether right ventricular pacing can in fact be therapeutic for a subset of patients with ICD and left ventricular aneurysm as the focus of ventricular tachycardia. During the eight month period from our patient's original presentation until his therapeutic catheter ablation, he remained consistently atrioventricular pacing (right ventricular activation) and clinically stable with a NYHA Class 2 functional status. Although it is unclear whether permanent right ventricular pacing is a long-term solution, early success with no recurrent ventricular tachycardia and a stable ejection fraction would suggest it can at least be utilized for temporary stabilization.

In our patient, ventricular tachycardia ablation was successful eight months after the electrical storm and after stabilization with right ventricular pacing. However, if ablation had not initially been successful, management could have included left ventricular aneurysm resection or even cardiac transplantation [9]. Given the patient's history of ischemic cardiomyopathy, congestive heart failure, and low ejection fraction, consideration could also have been given to upgrade his ICD to a biventricular device. However, if our hypothesis of left ventricular depolarization as the trigger for the PMVT was correct, biventricular pacing might result in aggravation of the tachyarrhythmia [10].

This case is an example of how the counterintuitive notion of ventricular desynchronization by forced right ventricular pacing and late left ventricular depolarization can be therapeutic in the acute suppression of electrical storm. The prevention of short to medium term electrical storm recurrence in the subset of patients with PMVT and left ventricular arrhythmogenic focus can also be postulated. PMVT may have been mediated by chronic ischemia and reentry circuits in the aneurysmal infarct region. The fact that the episodes were suppressed by forced right ventricular pacing would support some contribution of reentry within the aneurysm as opposed to just ischemia, although it is not possible to sort this out definitely.

CONCLUSION

We hypothesize that altering the electrical depolarization and ventricular activation sequence by right ventricular pacing prompted 'electrophysiological isolation' of the aneurysm and immediate resolution of the incessant polymorphic ventricular tachycardia (PMVT), as if suddenly turning off a switch. Therefore, when confronted with patients with refractory electrical storm, we propose programming a faster right ventricular pacing rate.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Role of immunohistochemistry in metastatic clear cell variant of follicular thyroid carcinoma: A case report

Yash Pradeep Vaidya, Rajan Vaithianathan, Ramanathan Manickam, Dhananjay Kotasthane

ABSTRACT

Introduction: Clear cell variant of follicular thyroid carcinoma with synchronous bony metastasis and a normal thyroid stimulating hormone level, is an extremely rare condition. Case Report: A 55-year-old male was presented to us with a painful swelling in the right arm. The biopsy showed clear cell adenocarcinoma, raising concerns for a metastatic renal cell carcinoma. Computed tomography scan of abdomen failed to show any renal lesions. A detailed physical examination revealed a small nodule of the right thyroid lobe. Fine needle aspiration cytology (FNAC) of the nodule was reported as follicular neoplasm. A right hemithyroidectomy and the subsequent completion thyroidectomy showed clear cell type of follicular thyroid carcinoma. Immunohistochemistry for thyroglobulin (Tg) further confirmed the diagnosis. Conclusion: Clear cell variant of follicular thyroid carcinoma is a very rare condition, hence a high index of suspicion is essential for diagnosis. The importance of performing a detailed physical examination cannot be more emphasized as small thyroid lesion like in this case can be easily missed, leading to a delay in diagnosis.

Keywords: Clear cell variant, Follicular thyroid carcinoma, Thyroglobulin, Thyroidectomy

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INTRODUCTION

Follicular thyroid carcinoma is the second most common type of thyroid cancer, comprising around 10–15% of thyroid malignancies. It has been histologically classified into oncocytic and clear cell types by the World Health Organization [1]. Follicular thyroid carcinoma with clear cell change is rare, and only some cases have been reported in literature [2]. This can be a diagnostic problem as metastatic clear cell carcinoma to the bones is usually attributed to a renal primary. Moreover, the most common cause for metastatic lesion in the thyroid is also renal cell carcinoma [3]. This clinical scenario may prove very challenging in arriving at an accurate diagnosis and for optimum management.

CASE REPORT

A 55-year-old male initially presented with pain and difficulty in moving his right arm after a fall. No significant past medical or family history was noted. On examination, he had a hard, 6x4 cm swelling over the upper third of the humerus. X-ray of the right humerus showed a soft tissue mass and a lytic lesion causing pathological fracture with varus angulation. Magnetic resonance imaging scan showed an intramedullary lesion in the bone with soft tissue extension, suggestive of a primary bone malignancy (Figure 1).

An open biopsy of the lesion revealed clear cell adenocarcinoma, probably of renal origin (Figure 2). However, immunohistochemistry staining using vimentin, CD 10, cytokeratin and S100 were equivocal. Computed tomography scan of abdomen was normal (Figure 3). On further examination, a small nodule was palpable in the right lobe of the thyroid. Thyroid function tests were within normal limits. Ultrasound of the neck demonstrated a 2x1 cm, hypoechoic nodule in the right lobe. Ultrasound guided FNAC was suggestive of a follicular neoplasm. A U-slab was applied for the fracture in right humerus.

The patient underwent an initial right hemithyroidectomy. Preoperatively, the patient was explained in detail about the need for this procedure as well as for completion thyroidectomy if malignancy was confirmed. Histopathology proved it to be clear cell variant of follicular thyroid carcinoma, with tumor cells invading into vascular spaces, features very similar to the biopsy from the right humerus (Figure 4). He subsequently underwent completion thyroidectomy to remove the left lobe and this also showed similar features.

Immunohistochemistry was strongly positive for thyroglobulin and negative for prior mentioned markers, thus confirming the lesion to be a thyroid primary rather than a metastatic lesion from the kidneys.

The patient made an uneventful recovery, with no symptoms of hypocalcaemia. He was later referred to the regional Radiation Oncology Centre for radioactive iodine (RAI) treatment and planned for a RAI scan (both thyroid and whole body). We plan to monitor the patient regularly with serum thyroglobulin levels.

DISCUSSION

Most follicular thyroid carcinomas are asymptomatic and diagnosed late. Thyroid scintigraphy usually shows a cold nodule. They typically present as solitary lesions, and very rarely involve both the lobes. Distant metastasis is common when there is capsular or vascular invasion [1]. Follicular thyroid carcinoma commonly spreads by the hematogenous route. Around 10–15% of patients have distant metastasis, occurring more often in tumors larger than 2 cm size [4]. The most common sites of distant metastases are the bones (with typical lytic lesions)

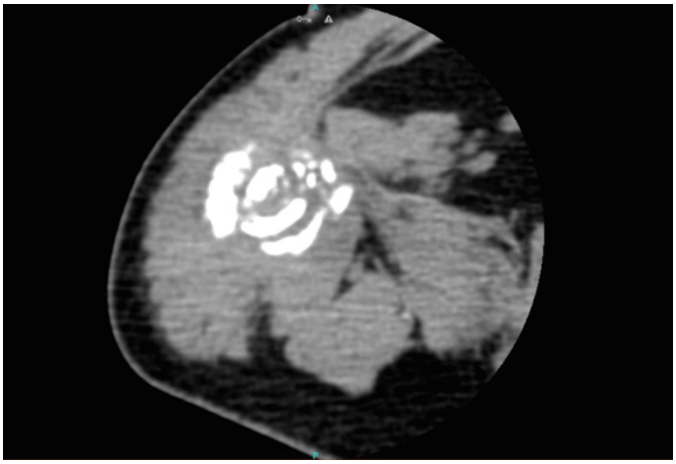

Figure 1: Magnetic resonance imaging section of the proximal humerus showing intramedullary lesion with pathological fracture.

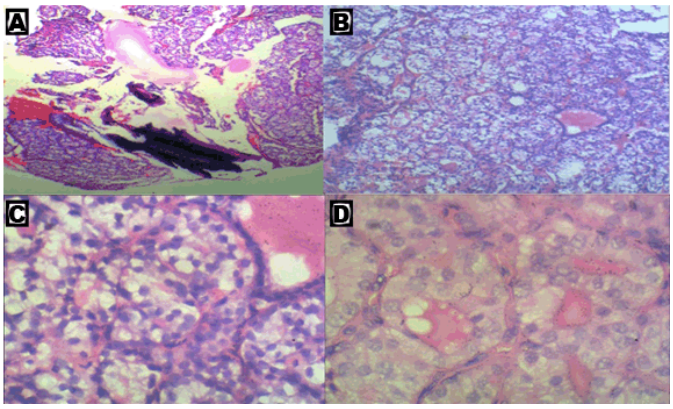

Figure 2: Histopathology of the bone biopsy (A) Bone fragments with surrounding fibrocollagenous tissue, (B) Metastatic tumor deposits, (C) Tumor cells arranged in solid nests with round dense nuclei, and (D) Alveolar cells with vacuolated cytoplasm suggestive of adenocarcinoma with clear cell changes.

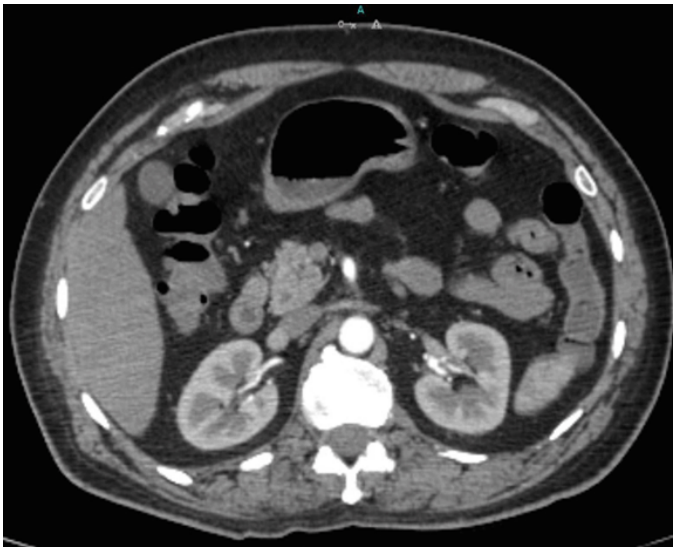

Figure 3: Computed tomography scan of abdomen showing normal kidneys.

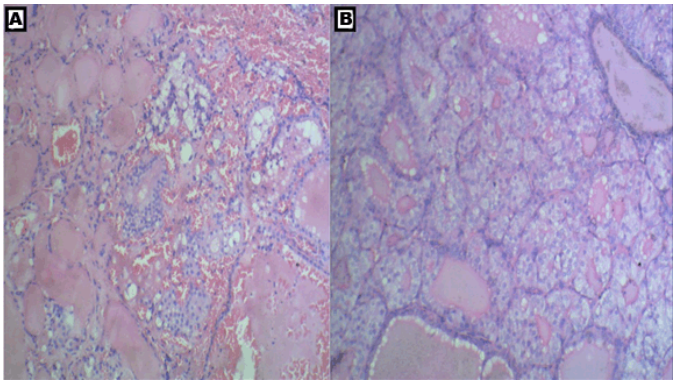

Figure 4: Histopathology of the thyroid specimen (A) Tumor cells with extensive invasion into vascular spaces, and (B) Similar appearance to the bone lesion, showing clear cell changes.

and lungs followed by the brain, liver, bladder and skin [5]. The incidence of metastatic disease is higher in the oncocytic variant than the clear cell type [6].

The clear cell appearance has been attributed to either accumulation of vesicles or glycogenation, thyroglobulin accumulation and hypertrophy of the Golgi complex due to excessive thyroid stimulating hormone stimulation [1, 7, 8]. Presence of clear cell features in follicular thyroid carcinoma very rare in patients with a normal thyroid stimulating hormone value as noted in this patient.

Almost 25–30% patients with renal cell carcinoma have distant metastasis at the time of diagnosis [9]. Hence this is usually the first differential diagnosis in such cases.

When metastatic lesions in the bone and the thyroid exhibit clear cell changes, it becomes difficult to differentiate metastatic renal cell carcinoma from a primary clear cell follicular thyroid carcinoma [10]. In such cases, immunohistochemistry plays an important role in clinching the origin of the tumor cells, as we found in our case. Clear cell renal cell carcinoma usually stains positive for vimentin, cytokeratin antibodies (AE1/AE3), CD10, renal cell carcinoma marker (RCCM), PAX2, PAX8, and carbonic anhydrase IX (CAIX), and stains negative for cytokeratins (CK7, CK 20), high-molecular-weight cytokeratin (HMWCK), CD 117 and parvalbumin [11].

CONCLUSION

Clear cell variant of follicular thyroid carcinoma is a very rare condition, hence a high index of suspicion is essential for diagnosis. Metastatic carcinomas exhibiting clear cell changes are commonly mistaken for a spread from a renal cell carcinoma . The importance of performing a detailed physical examination cannot be more emphasized as small thyroid lesion like in this case can be easily missed, leading to a delay in diagnosis. Immunohistochemistry is the definitive way of differentiating clear cell carcinomas arising from thyroid and the kidneys.

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Author Contributions

Yash Pradeep Vaidya – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Rajan Vaithianathan – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ramanathan Manickam – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Dhananjay Kotasthane – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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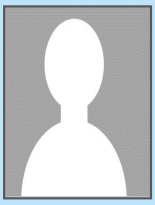

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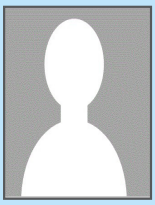

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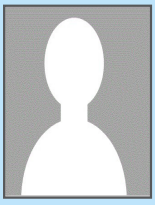

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Umbilical metastasis as a primary presentation in carcinoma rectum: A case report

Syed Altaf, Kapil Dev, Jaiprakash Gurawalia, Shiva Kumar

ABSTRACT

Introduction: Sister Mary Joseph nodule refers to periumbilical metastatic lesion and is an indicator of advanced intra-abdominal malignancy. It can be smooth, non-ulcerated or ulcerated and necrotic mass with or without blood, mucinous, serous or purulent discharge. It can appear before the diagnosis of the primary lesion or during or after the definitive treatment. Case Report: A 39-year-old man presented with a two-month history of painful swelling over the umbilicus. On workup, umbilical nodule was diagnosed as metastatic adenocarcinoma with a primary lesion in mid rectum. Conclusion: Sister Mary Joseph nodule is a thumbprint of disseminated and advanced disease that requires an aggressive combined treatment in every individual instance and bears poor prognosis.

Keywords: Carcinoma rectum, Metastatic adenocarcinoma, Sister Mary Joseph nodule

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INTRODUCTION

Metastatic umbilical lesion also known as Sister Mary Joseph nodule (SMJN) is secondary to a primary malignancy of any viscera. So far reported cases of SMJN can be categorized as metastatic from gastrointestinal malignancies (35–65%), genitourinary tract (12–35%), unknown sites (15–30%) and those from the lung and breast (3–6%) [1]. Depending on the characteristics of the primary tumor inside, it can be fissured or ulcerated and secreting serous, mucinous, purulent or bloody discharge [1, 2]. Umbilical metastasis from intra-abdominal visceral malignancies is a form of SMJN. It can be a presenting feature of undiagnosed malignancy. Here, we present a case of umbilical metastasis in carcinoma rectum as a primary presentation.

CASE REPORT

A 39-year-old male presented with a two-month history of swelling over the umbilicus with pain abdomen and anorexia. On examination, the patient was asthenic and dehydrated. His general physical examination revealed mild anemia and pedal edema without any lymphadenopathy. On systemic examination, the

abdomen was mildly distended with an approximately 2x2 cm umbilical swelling, without any other visible lump, scar or sinus (Figure 1). On abdominal palpation, a hard, tender umbilical swelling was felt that was arising from the anterior abdominal wall. There was no organomegaly, succession splash or fluid thrill in the abdomen. The rest of the systemic examination did not reveal any abnormality. There was no abnormality on digital per rectal examination. The investigations revealed a low hemoglobin of 7 g/dL and other hematological investigations were within normal limits. Serum CEA level was 15.68 ng/mL. Ultrasound of the abdomen revealed an isoechoic to hyperechoic lesion in anterior abdominal wall of size 4x3.5 cm with minimal ascites. The liver, pancreas, gallbladder and kidneys were normal and there was no para-aortic lymphadenopathy. Computed tomography scan of the abdomen revealed noncircumferential thickening of upper rectum with mesorectal lymphadenopathy. There was a heterogeneous mass lesion in anterior abdominal wall with multiple deposits over the omentum and peritoneum with minimal free fluid (Figure 2). Colonoscopy revealed a proliferative lesion in mid rectum which was confirmed by histopathology as mucinous adenocarcinoma (Figure 3). Fine needle aspiration cytology (FNAC) of the umbilical nodule done showed metastatic deposits of mucinous adenocarcinoma and a diagnosis of Sister Mary Joseph's nodule was made (Figure 4). Patient receiving palliative chemotherapy with regimen FOLFOX-IV and was on regular follow-up till six months.

Figure 1: An umbilical swelling of size 2x2 cm.

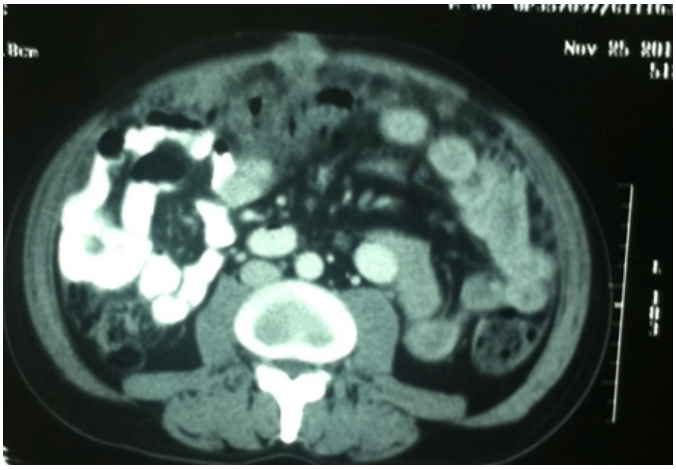

Figure 2: Contrast-enhanced computed tomography scan of abdomen (cross section view) showing a heterogeneous mass lesion in anterior abdominal wall at the level of umbilicus.

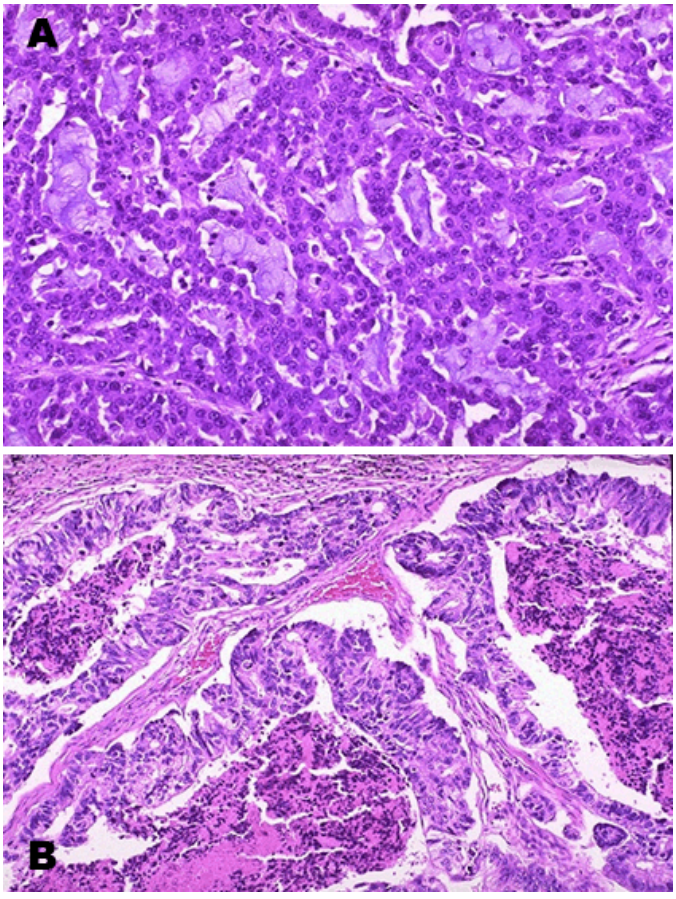

Figure 3: (A, B) Microscopic sections showing rectal mucosa, malignant cells with glandular pattern with lining being pseudostratified with individual cells having vesicular nucleus, prominent nucleoli with moderate amount of cytoplasm (H&E stain, x400).

DISCUSSION

The term "Sister Joseph's nodule" was proposed by a British Surgeon, Sir Hamilton Bailey for the umbilical metastasis of an abdominal cancer in 1948. This

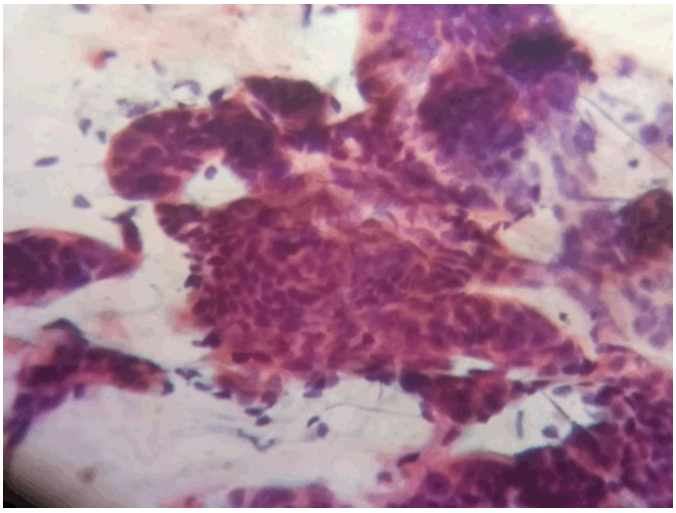

Figure 4: Fine-needle aspiration cytology smear showing a metastatic deposits of mucinous adenocarcinoma (H&E stain, x400).

uncommon pathology has numerous nascent etiologies. Three-fourths malignant umbilical tumors resemble to a "Sister Joseph's nodule" [2].

The incidence of cutaneous metastases from various malignancies reported in a range from 1% to up to 9%, according to autopsy reports. Merely, 10% out of them are presented as an umbilical metastasis [3]. Among all malignant umbilical lesions, 88% are metastatic; the rest are primary skin tumors [2]. Approximate 1–3% of intra-abdominal malignancies, including gastrointestinal and genitourinary may present with umbilical metastasis during the whole course of the disease. Among them, most of the patients 88% metastasized to umbilicus, and the rest to the skin [1, 3]. In patients with a history of known malignancies, as a recurrence either a solitary or synchronous lesion with another systemic dissemination of the disease is a common presentation. The SMJN as an initial presentation of any primary malignancy is reported in up to 30% of the cases, whereas the remainder presents in patients with known history of malignancy [3, 4]. Galvan et al. reported based on a large review of 407 patients over a period of three decades (1966–1997) that 14.6% of umbilical metastatic lesions have colorectal cancers as the primary source of metastasis [5]. Females are predominantly known to have these lesions [2]. Some case reports of SMJN published, as a first presenting feature in rectal cancer [6].

The common primary cancers metastasize to umbilicus are the gastrointestinal (35–65%), and genitourinary (12–35%) tract followed by rarely, hematological malignancies, lung or breast cancers may be in 3–6% of cases. After a thorough investigation of a patient with a metastatic umbilical lesion, 15–30% of patients remain with unknown primary [1, 2]. Most common histology of SMJN is reported adenocarcinoma followed by squamous cell carcinoma, melanoma or sarcoma [7].

The pathogenesis of umbilical metastasis is yet not well understood, various theories, including

1) Direct spread of the tumor cells over the surface of tumor spread of anterior peritoneum and invasion to umbilical tissue which is most accepted,
2) Lymphatic dissemination via the axillary, inguinal, para-aortic, internal mammary and external iliac lymph nodes;
3) Spread along the embryological remnants; and
4) Hematogenous spread, etc. [2, 3].

The rich arterial supply and venous network flowing cranially and caudally from the umbilicus with lymphatic drainage systems including pelvic and para-aortic lymph nodes favors the deposition of circulating cancer cells to the umbilicus. Traumatic violation of the anterior abdominal wall circulation during invasive procedures like diagnostic laparoscopy or operation for sterilizations especially in women, is also known to be a risk factor for higher dissemination of cancer cells to the anterior abdominal wall [2–4].

Clinically may present as the first presentation of underlying malignancy or during the progression of known cases of cancer [6, 8, 9]. The differential diagnosis of umbilical nodule includes neoplastic or non-neoplastic lesions such as Paget's disease, angioma, umbilical adenoma (raspberry tumor), umbilical hernia, endometriosis, hypertrophic scar, umbilical granuloma, pilonidal sinus, mycosis psoriasis, and eczema [2, 6]. Tissue diagnosis in the form of fine-needle aspiration cytology is acceptable to establish the diagnosis [10].

Presence of umbilical metastasis usually runs with poor prognosis due to advanced cancer with widespread metastases [2, 3, 6]. However, multimodality therapy including surgery and adjuvant therapy may improve the survival in some patients having good performance status [3]. Rarely, the surgical resection with negative margins with or without reconstruction of abdominal wall defect in isolated umbilical metastatic is a satisfactory curative treatment option, but chemotherapy is usually the mainstay of the treatment [3, 6]. It is dubious to get cured an umbilical metastasis in patients with disseminated cancer. However, systemic chemotherapy like FOLFOX based regimen should be considered as it sometimes gives good response in colorectal cancers. Overall, the prognosis of umbilical metastasis is poor with median survival only about one year [11].

CONCLUSION

Sister Mary Joseph nodule is an unusual indicator of occult visceral and other malignancies which can be diagnosed promptly only if physicians are made aware of this rare clinical presentation. It generally signifies disseminated advanced disease with poor prognosis for which aggressive combined modality treatment is required in every individual instance.

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Author Contributions

Syed Altaf – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Kapil Dev – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Jaiprakash Gurawalia – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Shiva Kumar – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Severe rhabdomyolysis induced renal failure after influenza vaccination in a patient on statins therapy

Shu-Hua Chen, Cheng-Jui Lin, Tung-Ying Chen

ABSTRACT

Introduction: There are various causes leading to acute rhabdomyolysis, and the most common potentially lethal complication of rhabdomyolysis is acute kidney injury. Influenza vaccination-associated rhabdomyolysis with acute renal failure is rarely reported in literature. There are few data regarding kidney biopsy results from rhabdomyolysis induced renal failure after influenza vaccination. Case Report: A 55-year-old male with chronic kidney disease stage 3a with hyperlipidemia under rosuvastatin control, who was admitted due to severe rhabdomyolysis induced renal failure after influenza vaccination. Kidney biopsy revealed acute tubular necrosis (ATN) and acute interstitial nephritis (AIN) superimposed on focal segmental glomerulosclerosis. Conclusion: Our experience illustrates the dual pathologic findings of ATN and AIN, both induced by influenza vaccine in a patient with chronic kidney disease on statin therapy.

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INTRODUCTION

There are several causes of rhabdomyolysis, including trauma, exertion, lipid-lowering agents, infection, electrolyte imbalance and toxins. Injury of skeletal muscle leads to release of intramuscular contents, including creatine phosphokinase and myoglobin, often resulting in acute kidney injury. Acute kidney injury secondary to rhabdomyolysis is fairly common, representing about 13–50% of all cases of rhabdomyolysis [1–3]. There are few reports of influenza vaccination-induced rhabdomyolysis in patients under treatment with lipid-lowering agents. Our report presented a patient, whom had chronic kidney disease and hyperlipidemia under statin therapy, developed severe rhabdomyolysis after an influenza vaccination.

CASE REPORT

A 55-year-old male with history of chronic kidney disease stage 3a and hyperlipidemia with baseline

creatinine 1.5 mg/dL two years ago. He was admitted to our hospital with symptoms of severe muscle pain and weakness. He took rosuvastatin 5 mg per day for hyperlipidemia for over one year. The patient received trivalent influenza vaccine (Sanofi Pasteur) five days prior to admission. He suffered from a single episode of fever and several times of diarrhea on the following four days. Subsequently, he developed general muscle soreness and weakness.

Initial physical examination showed, blood pressure 91/61 mmHg, temperature 36°C, and pulse rate 84/min. Heart sounds were normal and the chest was clear to auscultation. He had dry mouth, poor skin turgor, dark urine, and tenderness of bilateral extremities, but normal muscle power. Laboratory data revealed hemoglobin 16.6 g/dL, platelets 214000/mm^3, white blood cell count 23400/mm^3, serum creatinine phosphokinase 17121 IU/L (normal levels <300), creatinine 3.5 mg/dL, blood urea nitrogen 36 mg/dL, aspartate aminotransferase 200 IU/L, alanine aminotransferase 69 IU/L, uric acid 13.2 mg/dL, sodium 136 mEq/L and potassium 4.0 mEq/L. Urine analysis disclosed high specific gravity 1.024, low pH value 5.0, protein 3+, trace glucosuria, trace ketone body, occult blood 3+, RBC 13 cells/HPF, WBC 6 cells/hpf. Daily urine protein *loss was 2,413 mg/day*. Other serologic tests: hepatitis B and C were negative; Anti-nuclear antibodies, p-ANCA, and c-ANCA were negative. Serum complement level was normal.

After admission, rosuvastatin was discontinued. He was treated with aggressive intravenous fluid supplement and sodium bicarbonate alkalization therapy. Serum creatine kinase rose to a peak level of 77075 IU/L on the third day of admission (Figure 1). Urine myoglobin test was positive on the sixth day of admission. Concurrently, deterioration of renal function was noted (highest serum creatinine 12.2 mg/dL on 12th day of admission) and oliguria (daily urine amount < 200 ml) was noted as well (Figure 2). Temporary hemodialysis was total of six sessions between 3rd day and 14th day of admission. The level of serum creatine kinase and creatinine decreased gradually on the following days with intensive treatment. Renal biopsy was performed on the 17th day of admission, when daily urine amount was more than 2 liters. Histopathology, revealed the presence of advanced focal segmental glomerulosclerosis (FSGS) (Figure 3), acute tubular necrosis (ATN) (Figure 4) and allergy-associated acute interstitial nephritis (AIN) (Figure 5). Serum creatinine was 2.3 mg/dL on the 52th day.

DISCUSSION

Seasonal influenza vaccine contains inactivated viruses and is administered annually to protect against the influenza. According to Taiwan Centers for Disease Control and Prevention vaccination guidelines, influenza vaccines are recommended for everyone ages six months and older. Trivalent influenza vaccines protect against

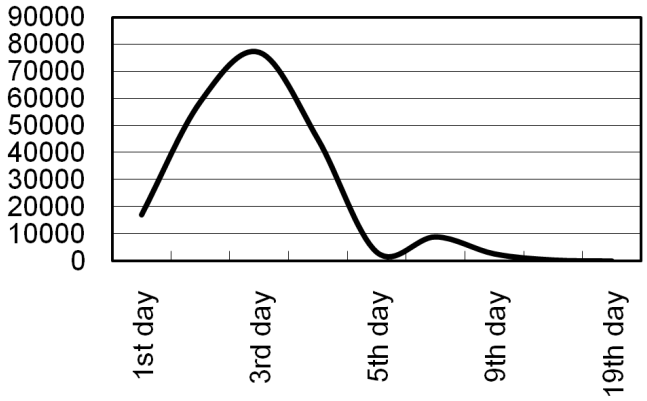

Figure 1: Serum creatine kinase (IU/L) levels during hospitalization.

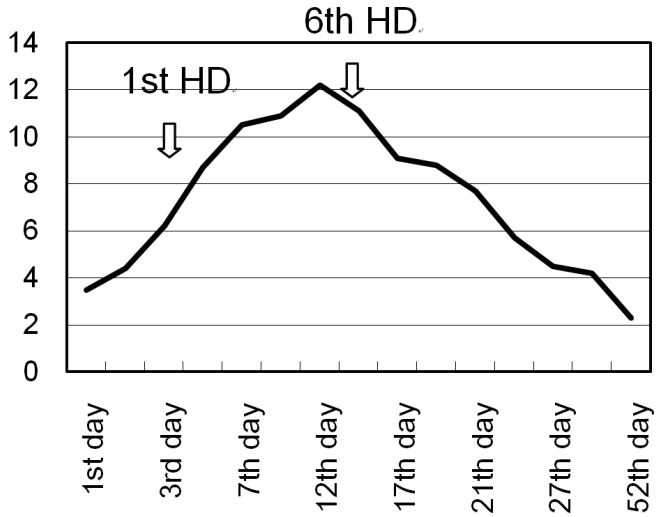

Figure 2: Serum creatinine (mg/dL) levels during hospitalization.

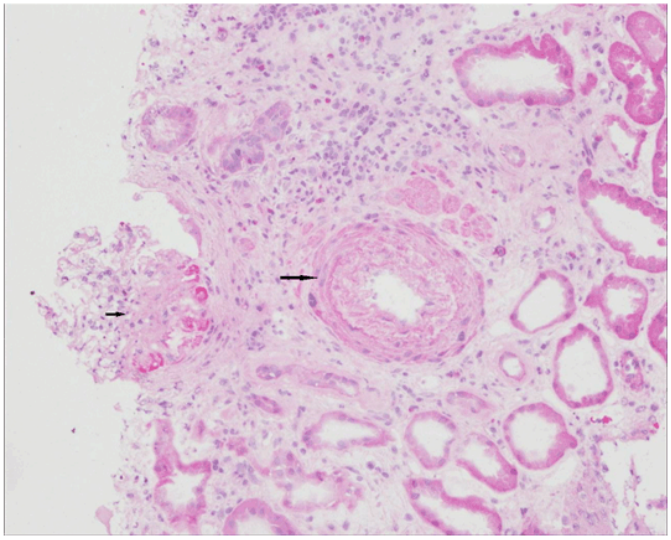

Figure 3: This glomerular tissue showing focal segmental sclerosis (short arrow) and mesangial proliferation on renal biopsy specimen. Vascular tissue revealed intimal fibrosis, media proliferation and hyaline arteriosclerosis (long arrow) (H&E stain, ×200).

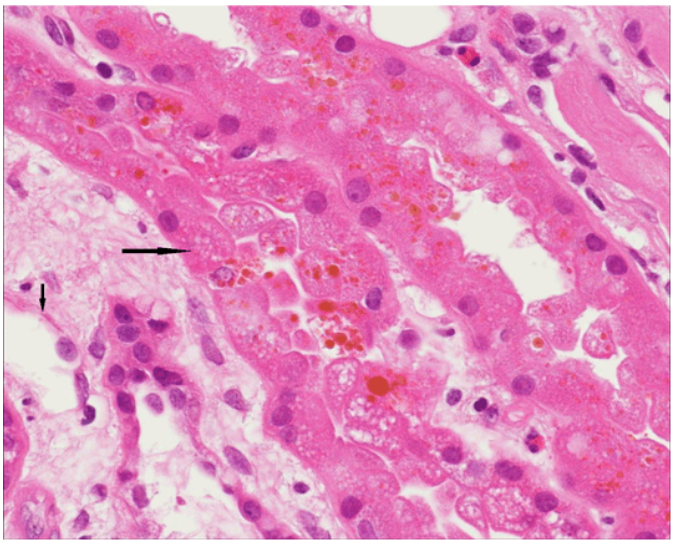

Figure 4: A photomicrograph of renal biopsy showing acute tubular necrosis. There are flattening or focal loss of the renal tubule epithelial cells, loss of epithelial brush border (short arrow), partial occlusion of tubular lumen by cellular debris, tubular epithelial cell vacuolization and tubular dilation (long arrow) (H&E stain, ×400).

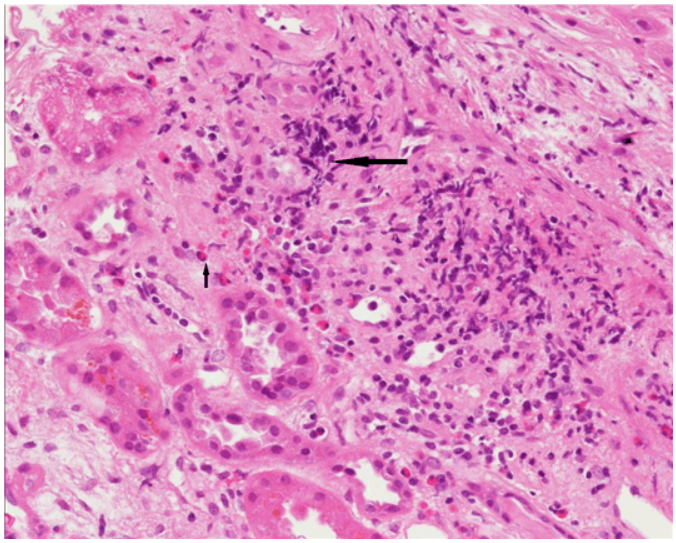

Figure 5: Renal biopsy specimen showing acute interstitial nephritis. Interstitial tissue was found with edema and inflammatory changes, containing eosinophils (short arrow) and mononuclear cells (long arrow) (H&E stain, x200).

two influenza A strains, H1N1 and H3N2, and one influenza B strain. Mild side effects from the influenza vaccination include transient soreness, redness or swelling at the injection site, nausea, low-grade fever and headache. Rare but serious side effects were anaphylactic shock, difficult breathing, swelling around the eyes or lips, wheal, temporary thrombocytopenia, high fever and neurologic symptoms, such as Guillain–Barre syndrome [4].

In addition to lipid-lowering agents and virus infection, there are several reports about rhabdomyolysis triggered by seasonal influenza vaccination [5–9]. Raman et al. found the first report of acute renal failure secondary to rhabdomyolysis following an influenza vaccination in a renal transplant recipient who had been on simvastatin and cyclosporine A therapy [5]. Novati et al. presented one case on statin therapy with acute renal failure twelve days after influenza vaccination, which consisted with acute tubulointerstitial nephropathy on urine analysis [9]. From our report, this patient developed severe rhabdomyolysis and acute allergic reactions with influenza vaccination. The renal pathological findings also demonstrated FSGS, ATN and AIN. The cause of rhabdomyolysis includes statin therapy as well as influenza vaccination. Statin may exert toxic effects on skeletal muscle, but whose overall incidence is typically <0.1% of patients receiving statin therapy [10]. Despite the widespread use of statin, the mechanism of statin-induced myopathy remains controversial and poorly understood. We know that, the ubiquitin-proteasome–dependent proteolytic pathway (UP pathway) is responsible for the breakdown of long-lived myofibrillar proteins (e.g., actin, myosin and troponin) in skeletal muscle, which releases of myoglobin. Statin may upregulate skeletal muscle gene expression of UP pathway [11]. Mechanisms of rhabdomyolysis induced acute kidney injury are renal vasoconstriction secondary to excessive leakage of extracellular fluid into the damaged muscle cells, formation of intratubular myoglobin casts that induce intra-tubular obstruction and direct toxin-related tubular damage [12]. In this case, no myoglobin cast was found in renal tubules on renal biopsy, which could be disappearing of myoglobin cast on the recovery of oliguria. Moreover, this patient had a previous history of chronic kidney disease (CKD) stage 3a, and was diagnosed as focal segmental glomerulosclerosis (FSGS) by kidney pathology. This can explain the patient's susceptibility for developing rhabdomyolysis induced renal failure after vaccination. Acute interstitial nephritis is often associated with use of drugs or infection, and is likely mediated through allergic mechanisms. In this patient, influenza vaccination may explain this hypersensitivity reaction. Although, influenza vaccination-associated rhabdomyolysis is rare, our report provides clinician a warning about serious complication in patients with renal impairment and statin therapy, that intend to be vaccinated against common viruses.

CONCLUSION

In summary, rhabdomyolysis induced renal failure may contribute to the morbidity and mortality during hospitalization. We report a case developing severe rhabdomyolysis-induced renal failure with dual pathologic finding after an influenza vaccination. Kidney pathology demonstrates that acute tubular necrosis and

acute interstitial nephritis may happen after an influenza vaccination. This report should caution clinicians about this potentially serious complication especially for those needed influenza vaccination with renal function impairment and on lipid-lowering medicine.

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Author Contributions

Shu-Hua Chen – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Cheng-Jui Lin – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Tung-Ying Chen – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Synchronous dual malignancy of papillary carcinoma thyroid and squamous cell carcinoma tongue: A case report

Tej Prakash Soni, Sajal Goel, Lalit Mohan Sharma, Anil Kumar Gupta, Shantanu Sharma, Ravindra Gothwal

ABSTRACT

Introduction: Patients with thyroid carcinoma have increased risk for development of second malignancy either synchronous or metachronous. We are reporting a rare case of synchronous squamous cell carcinoma oral tongue and papillary thyroid carcinoma. Case Report: A 60-year-old smoker male was presented with the complaints of non-healing ulcer (2x2 cm) at right anterolateral tongue since six months. Excision biopsy of tongue ulcer was reported as pT 0.8x0.8 cm, moderately differentiated squamous cell carcinoma. Wide local excision of tongue lesion with right modified neck dissection was done. Histopathology was reported as no residual primary tongue tumor seen, 1 out of 28 lymph nodes showed metastatic deposits of squamous cell carcinoma. Unexpectedly another one lymph node was positive of metastatic thyroid papillary carcinoma. Total thyroidectomy with central compartment neck dissection and left modified neck dissection was done. Histopathology was reported as pT 2x1 cm, papillary carcinoma right lobe of thyroid, 7 out of 32 lymph nodes showed metatstatic paillary carcinoma thyroid. Although exact cause of second malignancy is unknown but field cancerization, genetic predisposition, history of prior radiation or chemotherapy, improved survival can be attributed for increased incidence of second cancer. Conclusion: Double malignancies require multidisciplinary management approach.

Keywords: Carcinoma tongue, Papillary thyroid carcinoma, Synchronous

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INTRODUCTION

Double malignancy now comprises the sixth most common cancers and it makes 16% of all incident cancers [1]. Head and neck is a common site for synchronous

malignancy owing to field cancerization with use of tobacco but the co-existence of tongue and thyroid primaries are rare [1]. Incidental cervical lymph node metastasis from papillary thyroid carcinoma, detected in lymph node dissection done for separate head neck primary cancer is extremely rare. A case of synchronous papillary thyroid carcinoma and squamous cell carcinoma tongue is being reported with review of literature.

CASE REPORT

A 60-year-old smoker male was presented with the complaints of non-healing ulcer over right side tongue since six months. On examination there was an ulcer (1x1 cm) at right anterolateral tongue and palpable ipsilateral submandibular lymph node size 2x2 cm. Excision biopsy of tongue ulcer was reported as pT 0.8x0.8 cm, moderately differentiated squamous cell carcinoma, depth 0.5 mm, no lymphovascular or perineural invasion, cut margins free of tumor. Fine needle aspiration cytology (FNAC) from right cervical lymph node was reported as metastatic squamous cell carcinoma. Metastatic work-up was done.

All routine blood investigations, X-ray chest, ultrasound-whole abdomen were within normal limits. Wide local excision of tongue lesion with right modified neck dissection was done. Histopathology was reported as no residual primary tongue tumor seen, 1 out of 28 lymph nodes showed metastatic deposits of squamous cell carcinoma with no perinodal spread (Figure 1), 3 nodes showed epithelioid granulomas reaction, 1 lymph node showed a focus of follicles filled with colloid and lined by ovoid cells with optically clear nuclei suggestive of metastatic thyroid papillary carcinoma (Figure 2). Computed tomography scan neck showed an enhancing nodular lesion in right lobe of thyroid with large ring calcification measuring 2.9x1.7x1.8 cm in size with surgical changes in overlying cutaneous and subcutaneous tissues of neck (Figure 3). Total thyroidectomy with central compartment neck dissection and left modified neck dissection was done. Histopathology was reported as pT 2x1 cm, papillary carcinoma right lobe of thyroid, 7 out of 32 lymph nodes showed metastatic papillary carcinoma thyroid, maximum nodal size 2 cm, no extracapsular extension or lymphovascular invasion. Finally, he was diagnosed as a case of synchronous carcinoma anterior tongue pT1pN1M0 (stage III) and papillary carcinoma thyroid pT2pN1bM0. Expert opinion was sought from physician to rule out active tuberculosis. He received adjuvant external beam radiation to face/neck region to dose of 60Gy in 30 fractions over six weeks duration. He tolerated the treatment well. Whole body I-131 scan showed residual functioning thyroid tissue in thyroid bed. He received single dose of 150 mCi of I-131 orally. He was started on tablet levothyroxine 100 μg once a day and tablet calcium 500 mg twice a day.

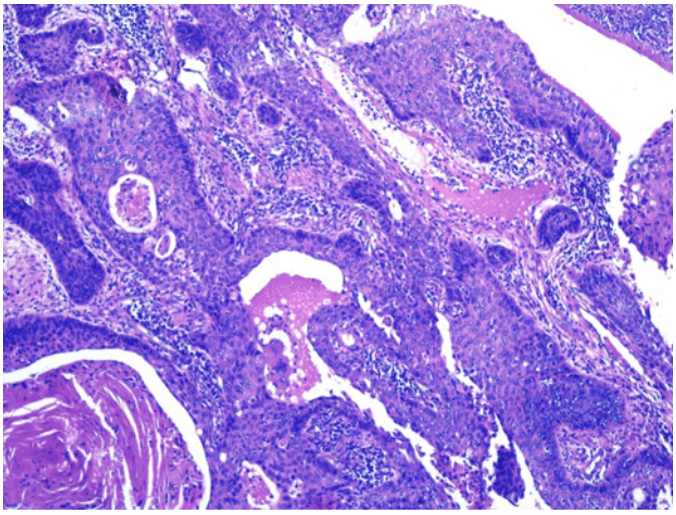

Figure 1: Lymph node metastasis by squamous cell carcinoma (H&E stain, x100).

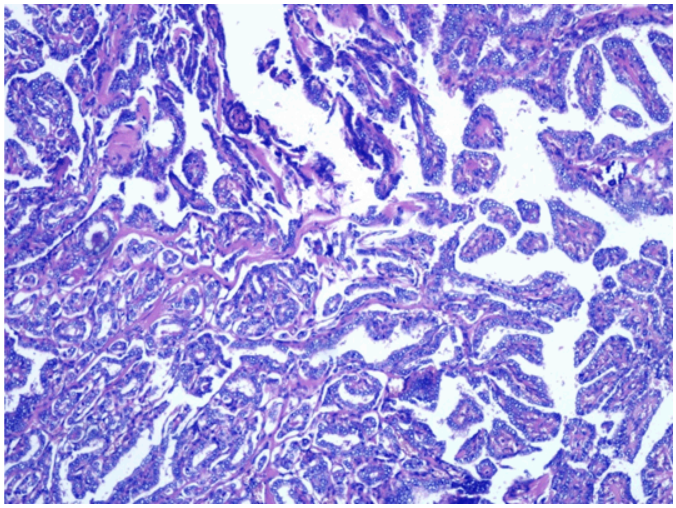

Figure 2: Lymph node metastasis by papillary carcinoma thyroid (H&E stain, x100).

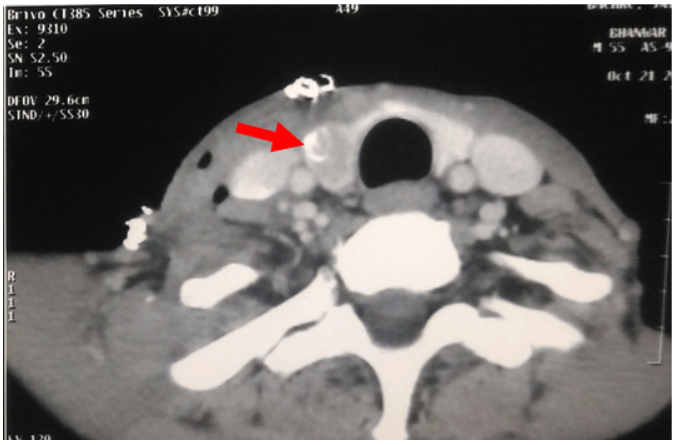

Figure 3: Axial view of contrast enhanced computed tomography scan of neck showing an enhancing nodular lesion (size 2.9x1.7x1.8 cm) in right lobe of thyroid with large ring calcification.

DISCUSSION

Thyroid cancer is the fifth most common cancer in females and accounts for 1% of all cancers. Incidence of carcinoma thyroid has increased significantly in last two decades. Surveillance epidemiology and end results database also reveals a 2.4-fold rise in thyroid cancer incidence from 3.6 per 100,000 in 1973 to 8.7 per 100,000 in 2002 consisting primarily of papillary thyroid carcinoma [2]. Wide use and availability of ultrasound and other imaging techniques contributes at least partly of this rise in incidence of carcinoma thyroid. Papillary thyroid cancer is the most common type of thyroid cancer and represents about 75% of all thyroid malignancies. Cervical lymph node metastasis is common in papillary thyroid cancer (ranging from 15–65% cases) and it is associated with a significant probability for loco-regional recurrence [3]. Lymphatic metastasis from papillary thyroid cancer occurs to neck lateral nodal groups of levels II, III, IV and central (level VI) lymph nodes. Thyroid cancer patients have an increased risk of developing a second cancer as either synchronous or metachronous [4]. In a pooled analysis of primary carcinoma thyroid patients from 13 registries, Sandeep et al. [4] found a 30% increase in risk of second cancers including salivary gland, kidney, prostate, skin, breast, brain, myeloma, leukemia, and non-Hodgkin lymphoma. On another side primary head neck cancer patients also have increased incidence (2–3% per year) of development of second primary cancer [5]. Barnes [6] found 0.7% of patients with squamous of carcinoma cell carcinoma of the head and neck cancers had lymphatic metastasis of papillary thyroid carcinoma. Singh J et al. [7] also reported a similar case of synchronous double malignancy of thyroid carcinoma and tongue carcinoma.

The exact mechanism of double malignancy is unknown. The predisposing risk factors for dual malignancy include field cancerization, tobacco and alcohol, genetic predisposition (Li–Fraumeni syndrome and Beckwith–Weidemann syndrome, Cowden syndrome), history of prior external radiation, radioiodine ablation or chemotherapy, environmental risk factors and improved survival [8].

Field cancerization also promotes transformation of an existing precancerous lesion into a malignancy and causes multifocal and second cancer in individual sites. Carcinogen exposure creates multiple genetic abnormalities in the whole tissue region and explores development of many foci of malignant transformation.

Thyroid cancer is associated with genetic mutations and abnormalities in tumor suppressor genes and cell cycle proteins which activates development of second cancers. Mutation of CHEK2 protein and RET genes are associated with an increased risk of thyroid, breast, prostate, and colon cancers [9].

CONCLUSION

In conclusion, incidence of second cancer has increased substantially. For early detection of second primary malignancy in head neck region complete and thorough examination and investigations including contrast enhanced computed tomography scan and pan-endoscopy are warranted. Squamous cell carcinoma of oral cavity along with synchronous papillary thyroid carcinoma is a rare situation. In such cases of double malignancy, multidisciplinary management and team work is required with the consensus of surgical oncology, radiation oncology, pathology, nuclear medicine and radiology experts. Oral cancers require aggressive treatment on priority as thyroid carcinoma has excellent prognosis. Metastatic workup followed by wide excision of primary oral cancer, total thyroidectomy and neck node dissection is required for adequate staging, proper diagnosis and management.

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Author Contributions

Tej Prakash Soni – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published
Sajal Goel – Acquisition of data, Drafting the article, Final approval of the version to be published
Lalit Mohan Sharma – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published
Anil Kumar Gupta – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published
Shantanu Sharma – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published
Ravindra Gothwal – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Hypoxemia in minimally invasive esophagectomy for squamous cell carcinoma of lower esophagus

Pak Lun Lam, Lam Viet Trung

ABSTRACT

Introduction: Minimally invasive esophagectomy for esophageal cancer is the surgical treatment-of-choice, and comprises both thoracoscopy and laparoscopy. The risk of hypoxemia arises during one-lung ventilation in the thoracoscopy stage, especially for patients with reduced lung function, and thus poses significant threat to the surgical outcome. This case report aims to demonstrate one possible solution to minimize hypoxemia in minimally invasive esophagectomy. Case Report: A 55-year-old Southeast-Asian male, with a history of chronic smoking, was presented with dysphagia. Diagnosis of squamous cell carcinoma was made, and minimally invasive esophagectomy was performed. Hypoxemia occurred during the thoracoscopic stage, and the oxygen saturation was stabilized with introduction of positive end-expiratory pressure of 5 cmH2O to reduce atelectasis, and tilting the operating table to the ventilated lung to improve perfusion by gravity. Conclusion: Minimally invasive esophagectomy may be complicated by hypoxemia during one-lung ventilation. One possible solution, as demonstrated in this case report, is the provision of a 5 cmH2O positive end-expiratory pressure, and approaching the thoracoscopic stage in a lateral decubitus position with the operating table tilted to the ventilated lung.

Keywords: End-expiratory pressure, Esophagectomy, Hypoxemia

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INTRODUCTION

Esophageal cancer is the seventh common cause of cancer death globally [1]. The mainstay of treatment for esophageal cancer is esophagectomy. Minimally invasive esophagectomy has become more commonly practiced due to a more rapid recovery and shorter hospital stay [2]. However, the thoracoscopic approach to esophagectomy requires one-lung ventilation, and this leads to a risk of hypoxemia during operation [3].

This case report aims to demonstrate one possible solution to minimize hypoxemia in minimally invasive esophagectomy, which is the provision a 5 cmH_2O positive end-expiratory pressure, and approaching the thoracoscopic stage in a lateral decubitus position with the operating table tilted to the ventilated lung.

CASE REPORT

A 55-year-old Southeast-Asian male presented with progressive dysphagia and significant weight loss over the

last six months. The patient was a chronic chain smoker and occasional social drinker. There was unremarkable past medical, family history and physical examination findings. Initial barium swallow demonstrated a stricture in the lower third of the esophagus with irregular outline (Figure 1). Esophagogastroduodenoscopy showed fungating lesion 32 cm from the upper incisor teeth, and 1 cm from the Z-line (Figure 2). Biopsy confirmed poorly differentiated squamous cell carcinoma of the lower third of the esophagus (Histologic Grade 3). At the aforementioned thoracic esophageal level, contrast-enhanced circumferential lesion was found (Figure 3). There was no apparent lymph node or distant organ metastasis. Minimally invasive esophagectomy was the recommended treatment of choice, and an informed consent was obtained from the patient.

The thoracoscopic and laparoscopic esophagectomy approach was adopted. Prior to operation, investigations were performed which deemed the patient fit for minimally invasive esophagectomy. Complete blood picture, clotting and electrolyte profile were normal. Liver, renal function tests and electrocardiogram were normal. Chest X-ray was unremarkable. Anesthesia was given, and ventilation was provided at tidal volume of 5 ml/kg and oxygen flow rate of 3 L/min in preparation for possible longer duration of one-lung ventilation during the thoracoscopic stage. Thoracoscopic mobilization was first performed with the patient in semi-lateral position. Five trocars were placed in respective locations to allow dissection (Figure 4).

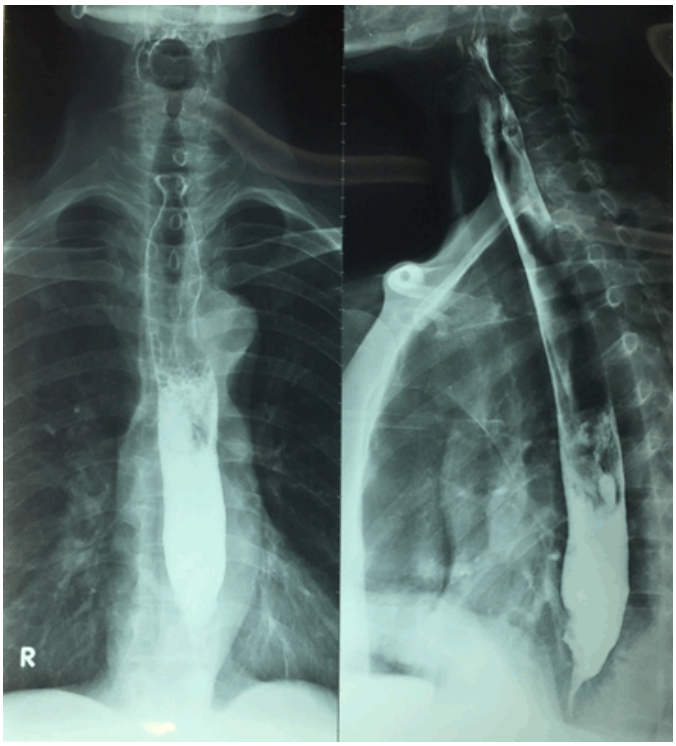

Figure 1: Barium swallow showing a stricture in the lower third of the esophagus with irregular outline.

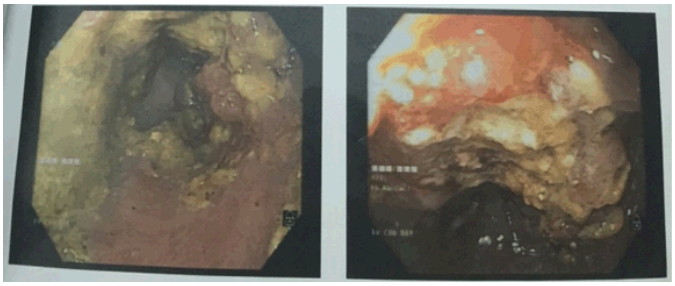

Figure 2: Esophagogastroduodenoscopy showing fungating lesion in lower esophagus.

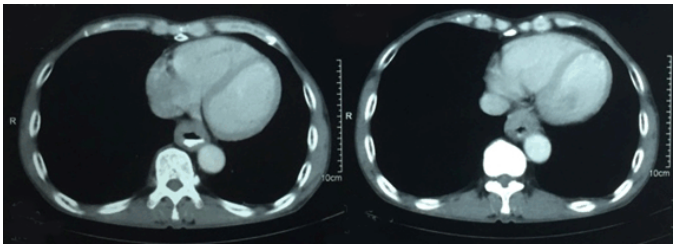

Figure 3: Contrast computed tomography showing circumferential lesion in lower esophagus.

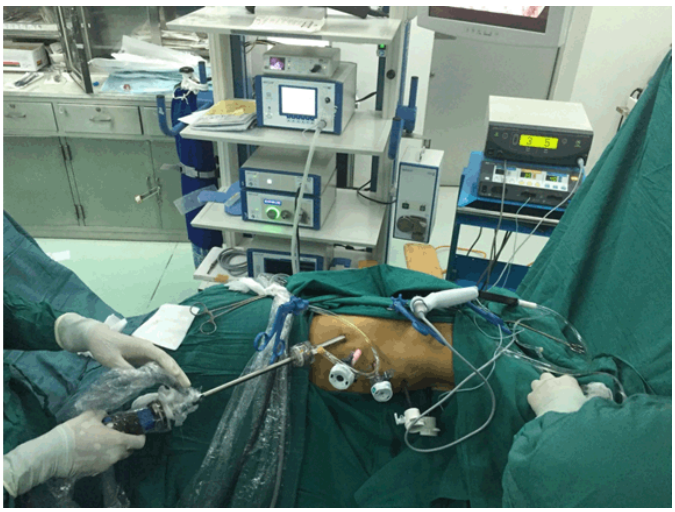

Figure 4: Thoracoscopic stage of minimally invasive esophagectomy.

The surgery had been uneventful up to this stage. The operation continued with localization of the central diaphragmatic tendon. However, the oxygen saturation of the patient dropped suddenly from above 95% to around 65%. The double-lumen tube for one-lung ventilation was immediately checked for misplacement, but this was not the case. The oxygen flow rate was increased from 3 L/min to 5 L/min, yet the oxygen saturation did not improve. Re-expansion of the nonventilated lung was performed to correct the situation. The oxygen saturation returned to above 95% after re-expansion, and the operation was attempted again with one-lung ventilation after the condition stabilized. However, the oxygen saturation fell again after brief dissection. Introducing a positive end-expiratory pressure of 5 cmH2O and tilting the operating table around 15 degrees to the ventilated lung was the

solution to maintain the oxygen saturation at above 95%. The operative was subsequently carried out with esophageal mobilization through dissection of the lateral pleura.

In the laparoscopic stage, the patient was placed in a supine position. The gastro-hepatic ligament and gastroesophageal attachments were dissected to achieve gastric mobilization. The less curvature portion of the stomach was stapled in preparation to form a gastric conduit 40 cm in length and 3 cm in width. The resected esophagus and stomach was removed and examined (Figure 5).

The cervical esophagus was then exposed through a horizontal neck incision above the clavicle. The stomach was guided through the posterior mediastinum and brought to cervical esophagus for end-to-side stapled anastomosis. Suture of all port sites completed the minimally invasive esophagectomy. Postoperative care was provided for the patient, and there was no complication.

DISCUSSION

In Western countries, such as the United States, esophageal cancer more commonly presented as adenocarcinoma of the lower thoracic esophagus, with Barrett's esophagus as a major risk factor. Whereas in other parts of the world specially in Southeast Asia squamous cell carcinoma was the predominant form [4].

In this case, computed tomography and bronchoscopy revealed a lack of adventitial involvement, as well as the absence of lymph node and distant metastasis. Surgical resection of the esophagus would be the treatment of choice. Traditionally, open transthoracic esophagectomy or transhiatal esophagectomy would be adopted. However, a minimally invasive approach was performed, as recent studies had indicated the use of thoracoscopic and laparoscopic approach in squamous cell esophageal carcinoma having similar morbidity and mortality rate.

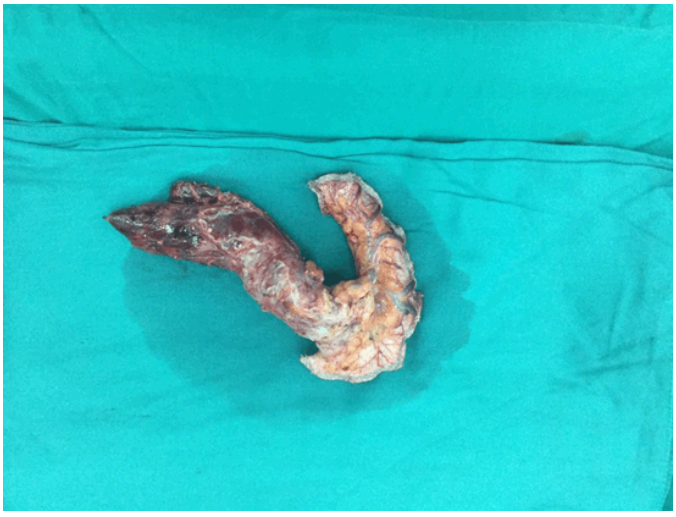

Figure 5: Resected esophagus and partially resected stomach.

At the same time, this technique provided faster recovery and shorter hospital stay when compared to open surgery [5].

One issue with minimally invasive esophagectomy was the risk of hypoxemia, as demonstrated in this case. The underlying cause could be attributed to one-lung ventilation. In addition, smoking which was a major risk factor in squamous cell esophageal cancer, may also played a role in hypoxemia during operation. Despite a normal chest X-ray and computed tomography scan, reduced lung function would arise from chronic smoking, and hypoxemia might be exacerbated especially when stressed during surgery [6]. It is suggested that pulmonary function tests, such as lung volume, capacity, peak flow rate and perfusion scans, could be performed for high risk patients to better assess the risk of hypoxemia during surgery.

In incidence of hypoxemia during the thoracoscopic stage of esophagectomy, mechanical causes like double-lumen tube misplacement should be checked immediately. This was shown to be a common and easily correctable cause of hypoxemia in one-lung ventilation [7].

When mechanical causes were excluded, the oxygen flow rate could be increased in moderation. However, hyperbaric oxygen posed threats to the central nervous system and the respiratory system, especially through reperfusion injury and the production of reactive oxygen species. Failure of maintaining oxygen saturation with increased oxygen flow rate would indicate re-expansion of the non-ventilated lung for stabilization of the patient [8]. Yet, there was a risk of barotrauma, and it would be impractical to perform esophagectomy with an expanded lung.

In this case, the maintenance of oxygen saturation was achieved by providing a positive end-expiratory pressure of 5 cmH2O, and tilting the operating table to the ventilated lung. Studies showed providing a positive end-expiratory pressure of 5–10 cmH_2O reduced atelectasis, especially in patients with impaired lung function, and thus correcting hypoxemia [9]. Thus, a positive end-expiratory pressure of 5 cmH_2O may be provided during minimally invasive esophagectomy to minimize the risk of hypoxemia.

In addition, the position of patient was also important. Rotating the operating table by 10–15 degrees to the non-dependent lung (ventilated lung) provided better oxygenation, because the effect of gravity allowed for a better perfusion of the lower-positioned lung [10]. It is, therefore, suggested that the thoracoscopic stage of minimally invasive esophagectomy may be approached in a lateral decubitus position with the operating table tilted to the ventilated lung whenever possible.

CONCLUSION

Hypoxemia in minimally invasive esophagectomy may not be a common occurrence, but poses substantial

threat to a successful operation. As demonstrated in this case report, one possible solution is to provide a 5 cmH2O positive end-expiratory pressure, and approaching the thoracoscopic stage in a lateral decubitus position with the operating table tilted to the ventilated lung.

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Author Contributions

Pak Lun Lam – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Trung, Lam Viet – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Giant granuloma pyogenicum of scalp: A rare presentation

Bhavinder Arora, Sunita Singh

ABSTRACT

Introduction: Granuloma pyogenicum or pyogenic granuloma is common benign vascular lesion of oral cavity mucous membrane and skin particularly in children and young adult fingers. Most of granuloma pyogenicum achieve a size of about 2 cm but giant variant can achieve a size of more than 4–5 cm. Giant granuloma pyogenicum of scalp is a occasionally reported. Giant and multiple granuloma pyogenicum are reported in immunocompromised patient. Case Report: A giant granuloma pyogenicum of scalp is reported here in a healthy child. This giant variant of granuloma of scalp was treated by excision with a small margin of normal skin and primary closure with sutures. Conclusion: Giant variant of granuloma pyogenicum on scalp is uncommon.

Keywords: Giant granuloma pyogenicum, Giant pyogenic granuloma, Granuloma telangiectaticum, Lobular capillary hemangioma, Vascular tumors

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INTRODUCTION

The granuloma pyogenicum is a variety of benign vascular tumor called lobular capillary hemangioma. It is also known by other names like pyogenic granuloma, and granuloma telangiectaticum. The most commonly used terminology pyogenic granuloma is a misnomer as neither it is not granuloma nor it contains pus. In 1897, Poncet and Dor gave the hypothesis that granuloma pyogenicum is a reaction to bacteria giving rise to florid granulation tissue, however, no bacterium could be isolated. In 1980 based on histology of this lesion the term granuloma pyogenicum was replaced by lobular capillary hemangioma, a vascular proliferative disorder [1]. Exact cause is not known but predisposing factors are trauma, chronic irritation, chronic inflammation, pregnancy, vascular malformation, drugs e.g., retinoid, mitozantrone, indinavir and erythropoietin [2].

Granuloma pyogenicum may present as solitary red glistening benign lesion that bleed spontaneously or with minor trauma. Granuloma pyogenicum is commonly a solitary lesion but multiple lesions are known in immunocompromised patients [3]. The most common site is oral cavity (marginal gingiva, palate, buccal mucosa, tongue and lips) in females due its relation

with pregnancy. External sites commonly involve the skin of face, eyes, neck, upper and lower extremities in children and young adults [4]. Granuloma pyogenicum usually presents as a solitary, red rapidly growing papule or a nodule. Most of these lesions are less than 2 cm in size. The giant variety is a rare variant of granuloma pyogenicum. The giant variety can acquire a huge size of more than 4–5 cm [5]. The giant granuloma pyogenicum has been reported from different sites like fingers, arms and thighs. Giant granuloma pyogenicum is a rare presentation on scalp. Only some cases of giant pyogenic granuloma of scalp have been described in literature [6]. We are presenting this rare giant variant of granuloma pyogenicum on scalp in a child.

CASE REPORT

An 11-year-old boy presented with a globular swelling which was red color in the vertex of scalp (Figure 1). There was no history of injury. It started as painless small swelling and grew to a large exuberant growth in about 20 days. It started bleeding spontaneously many times a day for last one day. On examination, there was a 4.5x4.0 cm spherical, sessile, firm, painless and raspberry like red color swelling in vertex of scalp. This ulcerated swelling started bleeding in the outpatient department (Figure 2). The hemoglobin value, bleeding time, clotting time, total and differential leucocytes counts were normal. The HIV test was negative. The surgical excision of this giant granuloma pyogenicum was done taking a margin 0.5 cm of healthy skin. The electrocautery was used for excision and hemostasis. The primary closure of the resultant elliptical defect was done using 2-0 polypropylene interrupted sutures. This excised swelling from the scalp was histopathology. Histopathology of this giant granuloma pyogenicum in H&E stain revealed stratified squamous epithelial lining with ulceration. Subepithelium revealed lobular accentuation comprising of chronic inflammatory granulation tissue with embedded dilated vascular channels separated by fibrous septae (Figure 3). Immunohistochemistry CD 31 revealed positive staining for vascular channels lined by endothelial cells (Figure 4) and SMA was positive for smooth muscle cells (Figure 5). This histopathological report confirmed the diagnosis of a giant granuloma pyogenicum. In the postoperative period, wound healed primarily without any infection or disruption. The sutures were removed on ninth postoperative day.

DISCUSSION

Granuloma pyogenicum can present clinically as a smooth or lobulated exophytic lesion with a sessile or pedunculated base. Mucosal lesions in oral cavity and cutaneous lesions in upper trunk and extremities are the common sites. McClintock et al. described two types of

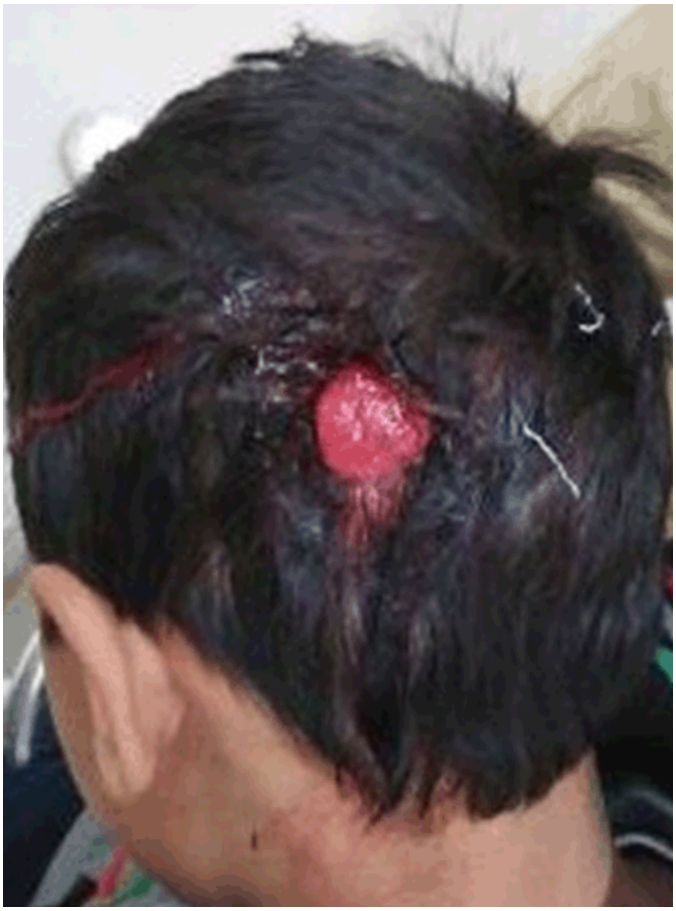

Figure 1: Giant granuloma pyogenicum of scalp.

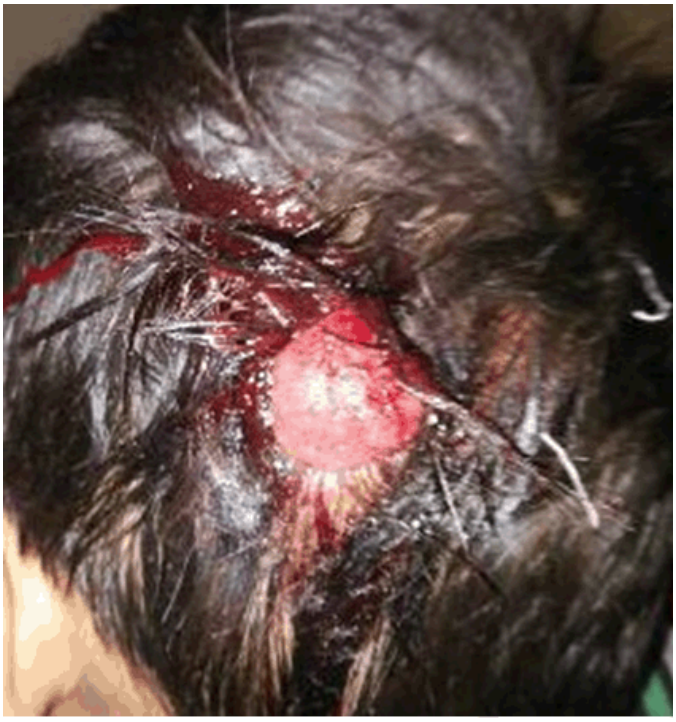

Figure 2: Hemorrhage in granuloma pyogenicum.

vascularized lesions as pyogenic and pseudopyogenic granulomas considering them partially neoplastic and inflammatory with little clinical differences between these two types. However, pyogenic granuloma is based on extremities while pseudopyogenic granuloma occurs on scalp and ears. There is no difference in treatment of these two histological types [7]. There are number of predisposing causes which can act as initiating cause. Trivial trauma particularly on fingers is the most common cause. Infection, drugs, pregnancy and oral contraceptive are other causes [8]. There was no apparent initiating cause in this child, but poor hygiene and hair care was thought to cause the granuloma pyogenicum in this child.

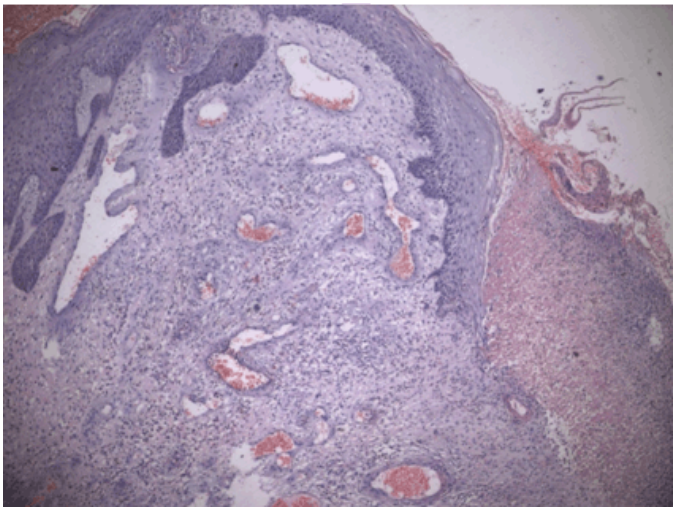

Figure 3: Lobular capillary angioma (H&E stain, x100).

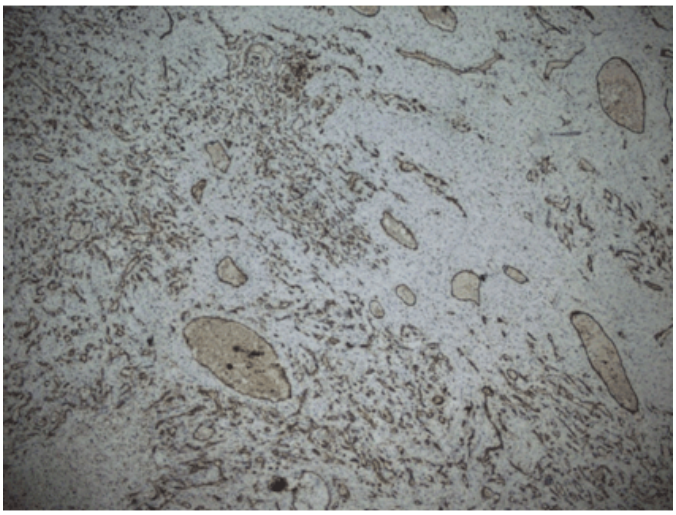

Figure 4: Immunohistochemistry CD 31 positive (magnification: x100).

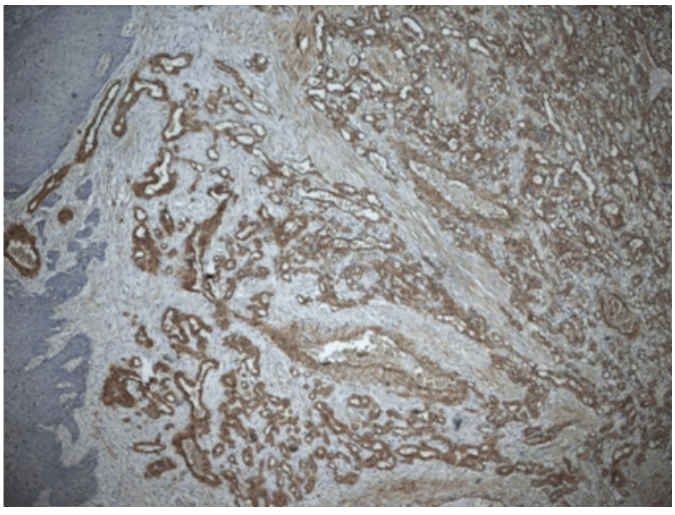

Figure 5: Immunohistochemistry SMA positive (magnification: x200).

Only some cases of giant granuloma pyogenicum of scalp have been reported in literature [9]. In this case, the size of granuloma pyogenicum was 4.5x4.0 cm which exceeds the standard size of less than 2 cm, hence labeled as giant granuloma pyogenicum. This size of giant granuloma pyogenicum is comparable to other case reports of giant pyogenic granuloma of scalp [10]. Giant granuloma pyogenicum are painless as nervous tissue proliferation is not there in the proliferating vascular tissue. So, even the giant sized granuloma pyogenicum is painless. Differential diagnosis of granuloma pyogenicum should include conventional granulation tissue, foreign body granuloma, hemangioma, angioendothelioma and angiosarcoma. Differentiation can be done on clinical basis and if suggestive biopsy should be done to rule out malignancy.

Giant granuloma pyogenicum of scalp can be easily treated with proper diagnosis. Millsop et al. have described current therapeutic modalities which include cryotherapy, electrodessication, curettage, excision, laser therapy, sclerotherapy, topical imiquimod and microembolization [11]. They reported a recalcitrant case pyogenic granuloma unresponsive to conventional surgical and nonsurgical treatment; and a complete resolution was achieved in this case by aggressive laser therapy, intralesional triamcinolone injection and topical application of timolol [11–13]. Lee et al. in an extensive retrospective review of treatment options and recurrence rates for pyogenic granuloma in 1162 patients observed surgical excision with lowest recurrence rate of 2.94% and cryotherapy in medical management with the lowest recurrence rate of 1.62%. They concluded that surgical excision and primary closure should be treatment of choice for small cutaneous pyogenic granulomas in non-cosmetic areas [14]. Patrice et al. in a study of 178 patients with pyogenic granuloma; 149 patients were treated by full thickness skin excision and linear closure with no recurrence, and 23 patients were treated shave excision or intradermal excision using cautery with a high recurrence rate [15]. Dissemond and Grabbe reported a case of giant pyogenic granuloma and its treatment. They observed that nonsurgical treatment that laser therapy or cryosurgery is associated with 50% recurrence rate so complete surgical excision as the first choice therapy [16]. The feeding vessels extend into surrounding skin in a cone like manner, so excision with surrounding margin of skin and primary closure is the best way to prevent recurrence of granuloma pyogenicum. Rapid healing can be observed

within a few days of treatment and as blood vessels are sealed, there is improvement, hemostasis and coagulation compared with surgical excision. Postoperative pain, discomfort, edema and bleeding are notably reduced. Complete surgical excision with a margin of normal skin is best with minimum recurrence, however if recurrence does occur re-excision may be required.

CONCLUSION

It is concluded that scalp is an uncommon site for giant granuloma pyogenicum which are generally small size. Huge or giant granuloma pyogenicum of scalp is a rare variant of pyogenic granuloma. This has tendency to bleed profusely. Giant cell granuloma pyogenicum can be diagnosed clinically. Proper treatment can be done by marginal excision using electrocautery. This leads to complete healing with no recurrence.

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Author Contributions

Bhavinder Arora – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sunita Singh – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE IN IMAGES PEER REVIEWED | OPEN ACCESS

Xanthogranulomatous cholecystitis: The great gallbladder carcinoma masquerader

Massimo Arcerito, John Moon, Kevin Tri Nguyen

ABSTRACT

Introduction: Xanthogranulomatous cholecystitis represents an uncommon chronic cholecystitis characterized by gallbladder wall thickening infiltrating to the adjacent liver parenchyma. This feature creates a challenge for the hepatobiliary surgeon in treating this rare disease, which mimics gallbladder carcinoma leading to unnecessary enlarged hepatic resection. Case Report: A 62-year-old male presenting at emergency department complaining of eight weeks history of right upper quadrant pain, nausea, with intermittent vomiting and severe weight loss. Surgical consultation was obtained. All imaging workup, including conventional abdominal ultrasound, computed tomography scan of the abdomen and pelvis, magnetic resonance imaging scan of the abdomen and positron emission tomography scan suggested gallbladder carcinoma. He underwent extended right hepatectomy, pancreaticoduodenectomy, with primary intrahepatic Roux-Y hepaticojejunostomy. The hospital course of patient was unremarkable being discharged six days after surgery. Final pathology was compatible with xanthogranulomatous cholecystitis. Conclusion: This rare entity can lead the hepatobiliary surgeon to face a challenge and sometime useless surgical treatment which might provoke morbidity and mortality to the patient. More define clinical and radiographic criteria are needed in diagnosing and treating xanthogranulomatous cholecystitis, which must be labeled as the great gallbladder carcinoma masquerader.

Keywords: Contrast enhancing ultrasound, Foamy cells, Pancreaticoduodenectomy, Xanthogranulomatous cholecystitis

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INTRODUCTION

Xanthogranulomatous cholecystitis is an inflammatory gallbladder disease characterized by destructive inflammatory process and infiltration of foamy cells of the gallbladder wall with associated proliferative fibrosis and thickening. This rare entity is almost impossible to be distinguished from the lethal gallbladder carcinoma based on clinical, radiographic and preoperative pathologic features. Furthermore, a very aggressive surgical treatment option is almost always performed provoking potential morbidity and possible mortality to our patients.

CASE REPORT

A 62-year-old male with a two-month history of right upper quadrant pain associated with nausea and vomiting causing 40 pounds weight loss. Empiric treatment with proton pump inhibitors did not achieve control of his symptomatology. The patient did not seek medical attention for all this time and he presented at the emergency department. Abdominal ultrasound showed features of acute cholecystitis characterized by pericholecystic fluid collection and thickness of the gallbladder wall. Contrast-enhanced abdominal computed tomography demonstrated extensive abnormal gallbladder wall thickening with direct invasion of the adjacent hepatic parenchyma and pylorus, with resulting gastric outlet obstruction (Figure 1). Subsequently, abdominal MRI scan confirmed the CT scan findings, and showed abnormal signal intensity and enhancement within the adjacent hepatic parenchyma, measuring up to 2.2 cm in thickness, invading the right hepatic duct as well compressing and encasing the pylorus (Figure 2).

Positron emission tomography scan showed hypermetabolic activity around the gallbladder suggesting a neoplasm (Figures 3A–B). The patient underwent diagnostic laparoscopy to rule out distant metastasis. After conversion to open technique, intraoperative ultrasound, extended right hepatectomy with cholecystectomy, resection of extrahepatic bile duct and pancreaticoduodenectomy with Roux-Y intrahepatic biliary anastomosis were performed (Figure 4). Figure 5 shows a detailed aspect of the gallbladder wall. Pathology showed xanthogranulomatous cholecystitis forming a large mass, extending into liver, duodenum, and distal stomach without involvement of pancreas and ten unremarkable lymph nodes (Figure 6). Figure 7 shows the xanthogranulomatous cholecystitis characterized by mixed inflammatory cells with many foamy macrophages in different magnification. A pathologic picture of gallbladder carcinoma is shown in Figure 8 for comparison to the benign xanthogranulomatous cholecystitis.

DISCUSSION

Christensen et al. first described this rare entity as “fibro-xanthogranulomatous inflammation” of the gallbladder in the seventies [1]. It is characterized by acute on chronic cholecystitis with dense fibrosis and aggressive accumulation of xanthomas in areas of destructive inflammatory response. This rare disease, based on its aggressive inflammatory behavior, mimics a gallbladder malignancy with involvement of surrounding anatomical structures, and this characteristic leads to perform an unnecessary extensive surgical resection of the surrounding organs. Jain et al. recently reported a retrospective study comparing the clinical and radiographic findings between the xanthogranulomatous and gallbladder cancer. Despite the absence of difference in clinical symptomatology between the two groups, contrast enhancement CT scan of the abdomen and pelvis helped to identify some radiographic properties

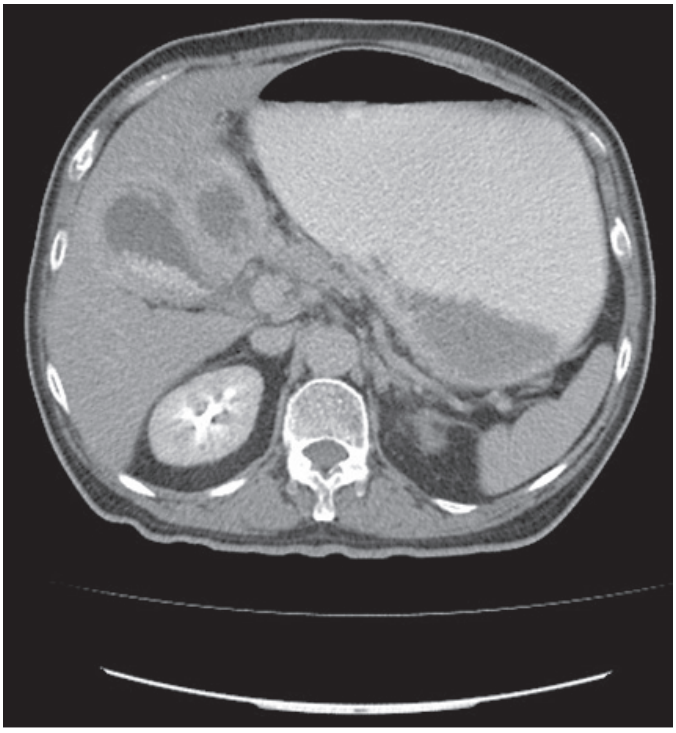

Figure 1: Triple phase computed tomography scan of abdomen and pelvis with gallbladder wall thickening suggesting gallbladder carcinoma.

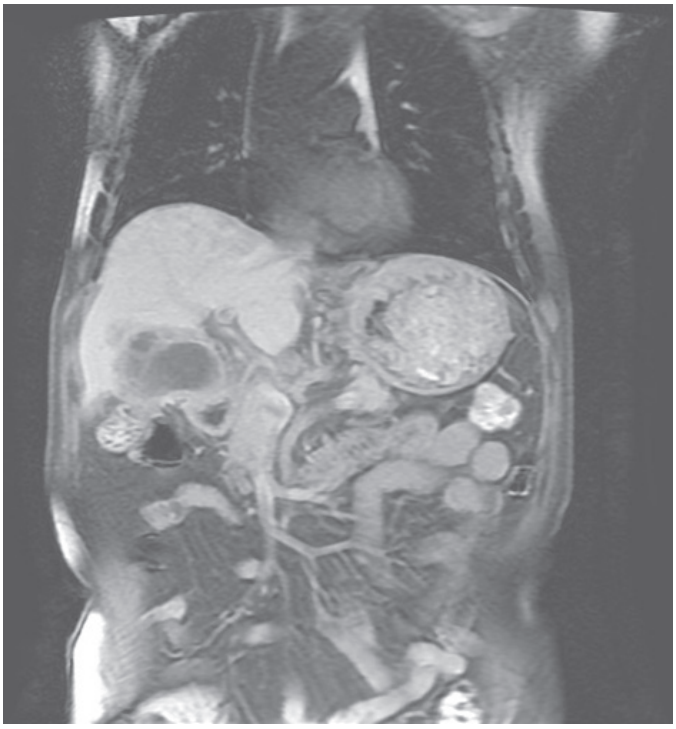

Figure 2: Magnetic resonance imaging scan showing markedly thickened gallbladder wall with perforation of the posterior superior gallbladder wall. Abnormal signal intensity and enhancement within the adjacent hepatic parenchyma measure up to 2.2 cm in thickness.

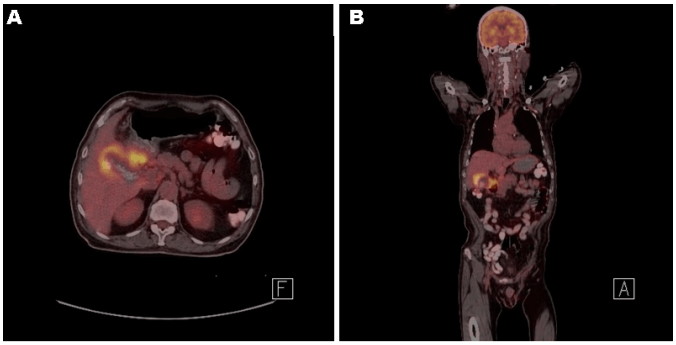

Figure 3: (A, B) PET-CT scan showing hyper metabolic activity around the gallbladder suggesting neoplasm.

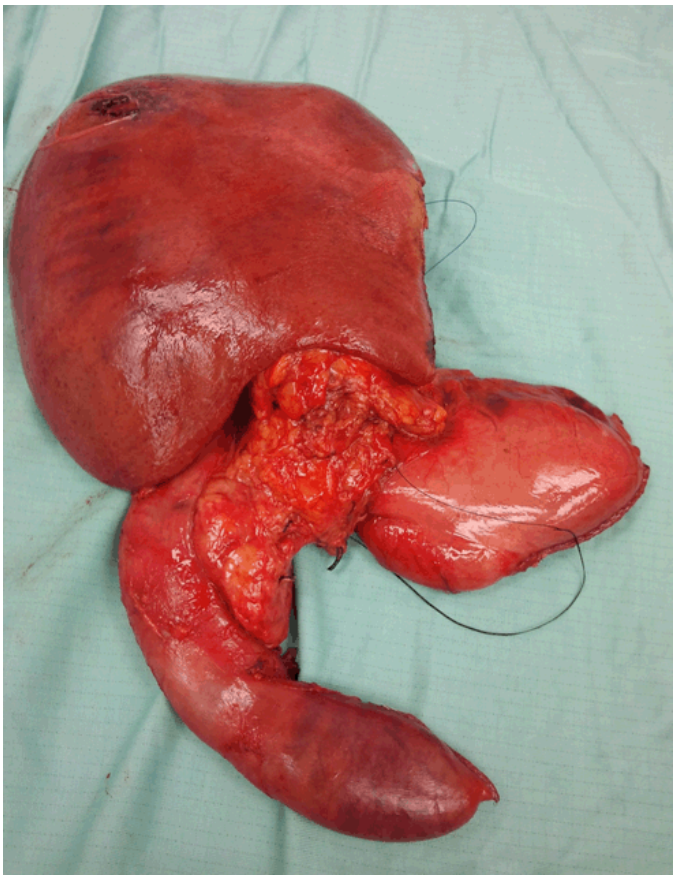

Figure 4: Pancreaticoduodenectomy with extended right hepatectomy.

commonly founded in xanthogranulomatous cholecystitis compared to gallbladder carcinoma [2]. Gallbladder wall thickness was similar in both groups, but the presence of intramural hypo-attenuating nodules in about 60% of xanthogranulomatous cholecystitis group in the absence of disrupted mucosal lining and obstructive features of the bile duct system, implied the inflammatory component. In support of these radiographic findings, Yuan et al. reviewed the clinical role of contrast-enhanced ultrasound technique in differential diagnosis of xanthogranulomatous cholecystitis. A total of 60 patients (17 xanthogranulomatous cholecystitis and 43 gallbladder carcinoma) were enrolled in the study. An

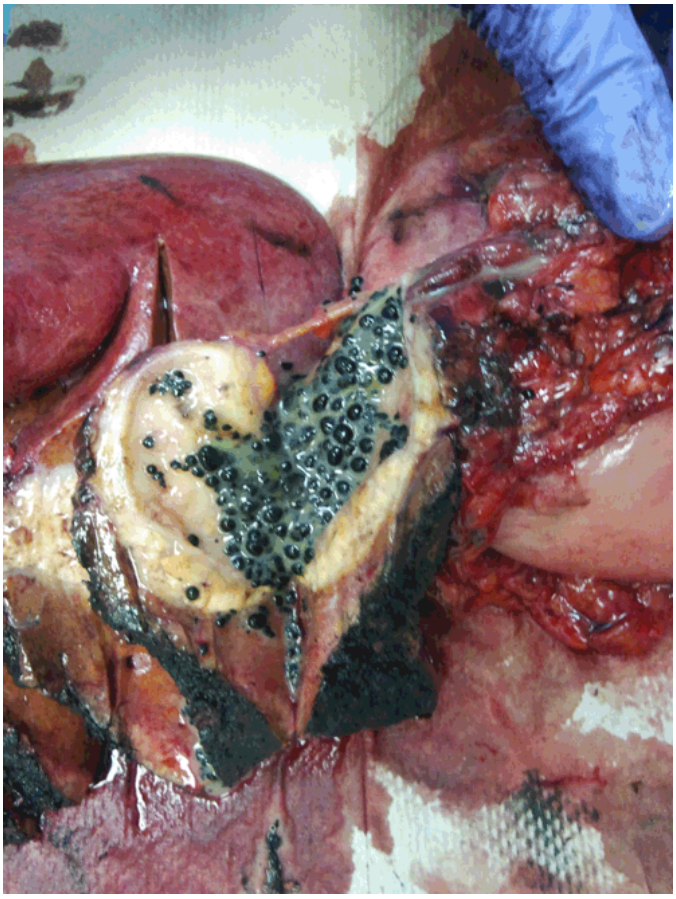

Figure 5: Xanthogranulomatous cholecystitis.

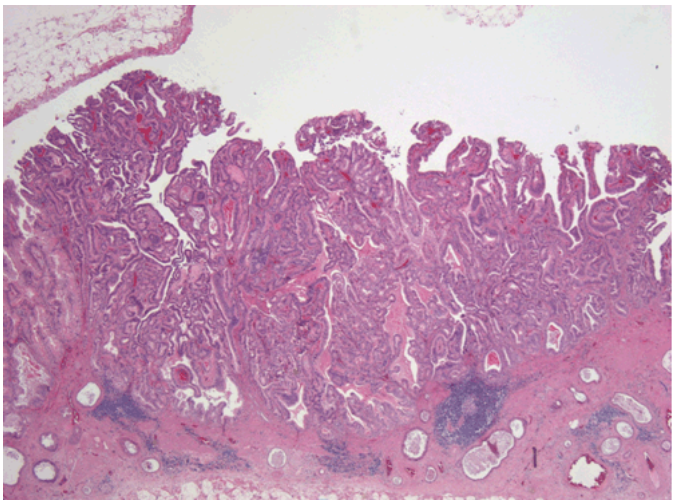

Figure 6: Pathologic view of gallbladder wall in xanthogranulomatous cholecystitis.

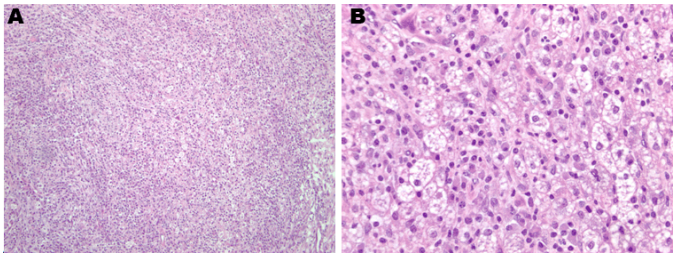

Figure 7: (A, B) Mixed inflammatory cells with many foamed macrophages.

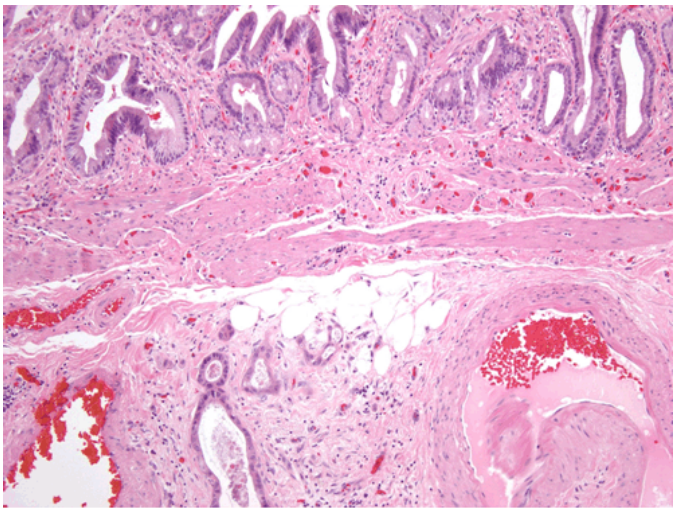

Figure 8: Gallbladder carcinoma; invasion of adipose tissue next to the gallbladder.

initial conventional ultrasound was performed in both groups and subsequently their data were compared with contrast-enhanced technique in searching for wall thickening characteristics, gallbladder stones and hypoechoic nodule frequencies. Contrast-enhanced ultrasound showed superior technique in analyzing the gallbladder wall thickening, gallbladder stones and hypoechoic nodules compared with conventional ultrasound.

Particularly, a hypoenhancement time greater than 80 seconds, a diffuse thickening and hypoechoic nodules were highly suggesting features of xanthogranulomatous cholecystitis [3]. An ultrasound-guided fine needle aspiration (UGFNA) of gallbladder lesions should be always be part of our armamentarium in differential diagnosis of xanthogranulomatous cholecystitis. We did not perform a UGFNA in our patient. Rana et al. proved in their largest experience the paramount safeness, reliability and cost-effective of this technique in diagnosis gallbladder diseases. Their conclusions prompted them to suggest for all the providers in hepatobiliary specialty to perform a UGFNA in all cases with gallbladder mass upon presentation [4]. The premalignant condition of xanthogranulomatous cholecystitis is unknown. Ghosh et al. addressed the premalignant property of xanthogranulomatous cholecystitis [5]. Proliferating cell nuclear antigen (PCNA), beta-catenin and p53 expression were studied comparing xanthogranulomatous cholecystitis, gallbladder cancer, chronic cholecystitis and cholelithiasis as control group. p53 mutation and PCNA were present in 52% and 60% of gallbladder carcinoma and only in 3% and 11% of xanthogranulomatous cholecystitis. No evidence of this mutation was found in chronic cholecystitis or cholelithiasis. Beta-catenin expression was positive in all four analyzed groups. We supported the inflammatory component of xanthogranulomatous cholecystitis without evidence of premalignant condition.

A new clinical scoring system has been proposed in differentiating xanthogranulomatous cholecystitis and gallbladder carcinoma [6]. The authors retrospectively reviewed their experience in treating almost 500 patients with gallbladder diseases focusing on clinical and imaging parameters. Patients with a long history of abdominal pain and gallbladder wall thickening, cholelithiasis and gallbladder submucosal hypoattenuated nodules compared with anorexia, weight loss are more prone to be diagnosed as xanthogranulomatous cholecystitis more than gallbladder malignancy. A scoring system closed to 11 to 13 helps to narrow the diagnosis towards xanthogranulomatous cholecystitis.

CONCLUSION

Xanthogranulomatous cholecystitis is a rare entity leading to potentially unnecessary extensive surgical resection with possible associated increase morbidity and mortality. A defined role of scoring systems should be implemented in differentiating this rare disease from the more lethal gallbladder carcinoma. Identifying radiographic features and performing preoperative ultrasound-guided fine needle aspiration (UGFNA) of gallbladder masses and possible intraoperative biopsy through the liver parenchyma may assist the hepatopancreaticobiliary surgeon in intraoperative planning and decision-making.

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Author Contributions

Massimo Arcerito – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
John Moon – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
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Guarantor

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Conflict of Interest

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CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Enterocutaneous fistula 30 years after prosthetic mesh repair of ventral hernia

Arron J. Gravina, Joshua A. Cuoco, Edward H. Cussatti

CASE REPORT

A 64-year-old female, with a one-week history of a mid-abdominal mass, presented to the emergency department with focal tenderness and cramp-like abdominal discomfort. She denied fever, chills, recent trauma, or changes in bowel habits. Her past medical history included diabetes, obesity, hypertension and asthma. Past surgical history was notable for a laparoscopic cholecystectomy and four ventral hernia repairs ultimately requiring mesh placement approximately thirty years ago. Computed tomography scan demonstrated an abscess in the right rectus sheath measuring 8.2x3.8x7.2 cm with phlegmon extension to the level of the umbilicus. Moreover, locules of gas and calcification were notable on the posterior aspect of the abscess. The patient was placed on broad-spectrum antibiotics. Interventional radiology was consulted for percutaneous drainage. A 5 French needle was subsequently used to drain 30 cc of purulent material. Pathology confirmed the presence of *Escherichia coli* within the abscess.

Six days after percutaneous drainage, surgical exploration demonstrated an infected polypropylene mesh intimately adherent to the abdominal wall and small bowel forming a fistula. The majority of the infected mesh was then removed, the abscess was further drained, and vacuum-assisted closure dressing was placed. A follow-up small bowel series with barium performed two days later demonstrated an ostomy within the abdominal wall and a corresponding enterocutaneous fistula (Figure 1). Conservative measures were initiated for the following weeks with little resolution of the fistula due to its high output (>1 L per day on an oral diet with somatostatin and 400–600 mL per day on parenteral nutrition). An exploratory laparotomy was consequently performed during which a portion of small bowel was resected with primary anastomosis and further removal of the infected mesh. The abdominal wound began to granulate after correction of the fistula and placement of the vacuum-assisted closure dressing.

DISCUSSION

Large or recurrent ventral hernias are best managed with placement of a prosthetic material to ensure support of the abdominal defect. Polypropylene mesh is a commonly used prosthetic material utilized in open and laparoscopic methods for management of ventral hernias. A prosthetic mesh can be placed in three ways including onlay, subfascial, and intraperitoneal. Intraperitoneal repair is the most popular method due to ease of technique

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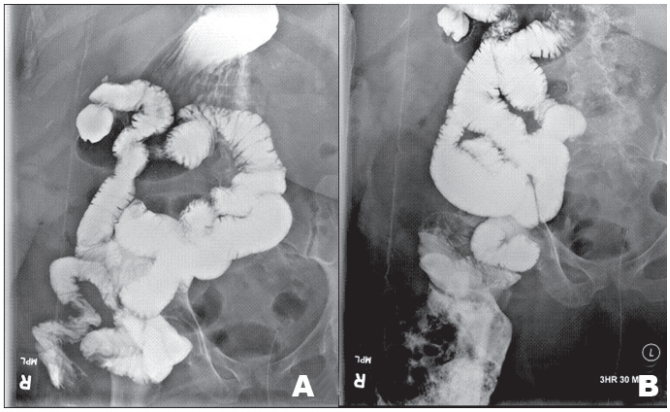

Figure 1: Small bowel series with barium illustrates (A) an ostomy within the abdominal wall and, (B) corresponding enterocutaneous fistula.

and lower recurrence rate [1]. Placement of uncoated polypropylene mesh is performed when the omentum is sufficient to cover the bowel, thereby protecting the intestines from interaction with the mesh [1]. When omentum is insufficient, double layered polypropylene and vicryl or newer anti-adhesive meshes may be utilized [1].

Although polypropylene mesh is well tolerated by most patients, the use of such apparatuses is not without risk. Known complications in using polypropylene mesh include bacterial infection, hematoma and/or seroma development, adhesion formation, sinus formation, bowel obstruction, mesh extrusion, mesh migration, and enterocutaneous fistula formation [2]. Although rare, the most serious complication associated with mesh placement is enterocutaneous fistulation, which is usually a consequence of intestinal erosion by the mesh [2].

Enterocutaneous fistulation consequent to erosion of the bowel after mesh placement for a ventral repair was first described in literature in 1981 [3]. Since then, studies have reported the incidence of enterocutaneous fistulation following mesh placement to be between 3.0–3.5% [4, 5]. Nevertheless, the 30 years delayed presentation of enterocutaneous fistulation after mesh placement in our patient is unique such that it indicates risk of fistulation does not completely dissipate with time. Rather, polypropylene mesh provides patients with a life-long risk for the development of enterocutaneous fistulation.

CONCLUSION

Here, we present a patient that exhibited enterocutaneous fistulation 30 years after mesh placement for repair of a ventral hernia. As such, this case demonstrates that mesh placement may predispose patients to a life-long risk for enterocutaneous fistulation.

Keywords: Complications, Enterocutaneous fistula, Surgical mesh, Ventral hernia

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Author Contributions

Arron J. Gravina – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Edward H. Cussatti – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Conflict of Interest

Authors declare no conflict of interest.

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CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Bilateral superficial angiomyxomas of lower limbs: Unique presentation of a benign cutaneous tumor

P. Gaspar da Costa, L. Lêdo, S. Braz, A. Teixeira,
L. Soares-de-Almeida, J. Meneses Santos

CASE REPORT

A 93-year-old female presented with painless grouped cutaneous nodules and tumors, ranging from 0.5–3 cm in diameter (Figure 1), which had slowly developed over the last five years. Examination revealed multiple firm, non-tender lobulated nodular lesions, varying from flesh-colored to erythematous, symmetrically distributed over the dorsal face of all toes and the dorsal face of both feet. A 6-mm skin punch biopsy was performed and showed poorly circumscribed myxoid dermal nodules containing a few of bland spindle-cells (Figure 2A–B), mucin deposits (Figure 2C) and small blood vessels (Figure 2D), without inflammatory cell infiltrate. These findings were consistent with the diagnosis of superficial angiomyxoma. Tumor resection was not proposed because of lesions extension and no other treatment was performed.

DISCUSSION

Superficial angiomyxoma is a rare benign cutaneous tumor that affects patients of all ages, with a peak incidence in 4th and 5th decades, and has a slight predilection for males [1, 2]. First reported by Allen et al. in 1988 it is usually reported as a solitary skin nodule, papule or polypoid lesion under 5 cm in diameter, most commonly presenting on the trunk, followed by the lower limbs and, finally, the head, neck and the upper limbs.

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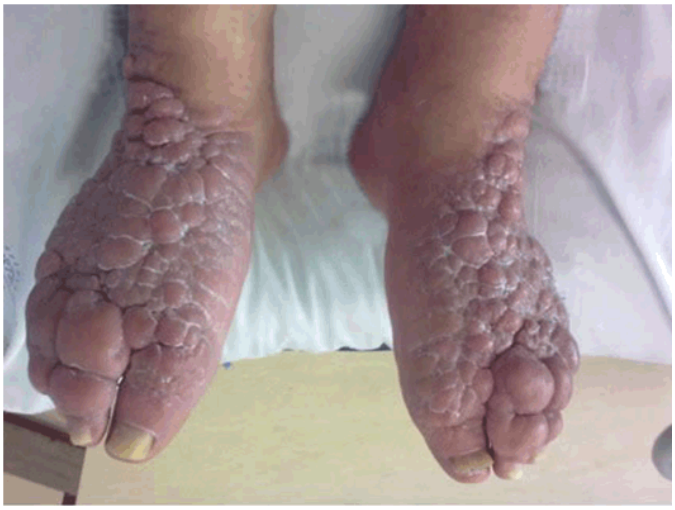

Figure 1: Multiple grouped skin tumors on the feet.

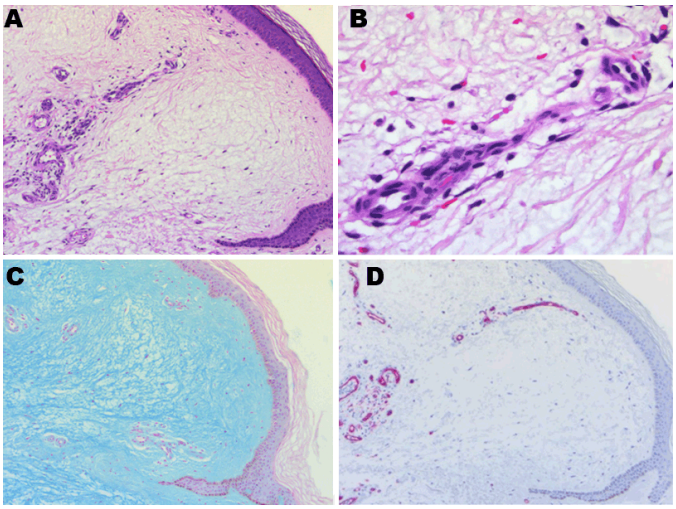

Figure 2: (A, B) Histological aspects of dermal mucin with scanty fibroblasts and blood vessels (H&E stain, A: x100 and B: x400), (C) Alcian blue stain highlights the mucin deposit (magnification: x100), (D) Immunohistochemical study: The blood vessels are in red with CD31 (magnification: x100).

Superficial angiomyxoma with unusual location such as the oral cavity mucosa and external genitalia and subungual neoplasms have also been reported [2, 3].

Histologically, lesions are described as well circumscribed dermal tumors with an abundant mucinous stroma, spindle-shaped and/or stellate cells and a prominent vascular pattern with multiple small to medium-sized thin-walled vessels. A sparse to moderate mixed inflammatory infiltrate is usually present within the myxoid matrix.

The myxomas described in the Carney complex are very similar to superficial angiomyxomas [4]. Their recognition is important because it can be the first manifestation of the syndrome. The patient had neither lentigines on the face, nor signs of endocrine overactivity.

Treatment consists of surgical excision. We did not find in literature any alternative therapy to surgical resection. These tumors are not known to metastasize. However, they have a 30–40% local recurrence rate mainly after incomplete excision [1, 2].

CONCLUSION

Multiple superficial angiomyxoma is a very rare occurrence. To the best of our knowledge, there is no similar case, as the one reported here, described in the English literature. The dimension of the tumors and their symmetrical distribution on the lower limbs make this case a unique superficial angiomyxoma.

Keywords: Angiomyxoma, Cutaneous tumor, Superficial angiomyxoma

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Conflict of Interest

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