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Urethrovesical foreign body: Report of two cases and literature review

Sefu Juma Uledi, George Lodewijk Pape, Fauzia Ayubu Masumai

ABSTRACT

Introduction: Urethrovesical foreign bodies have been fairly reported. However, hitherto their diagnosis and subsequent management still pose challenge to clinicians. Different types of urethrovesical foreign bodies have been described. Broadly, they can be categorized as inserted, iatrogenic and migratory foreign objects. Regardless of their diversity, nature and origin, they do often lead to similar presenting symptoms and beset by multitude of complications. Case Series: We hereby report two cases: first, being self inserted wire into the male urethra and urinary bladder. Second case is a foreign body iatrogenically introduced into urinary bladder of a 52-year-old male during open prostate surgery. Conclusion: Patients with urethrovesical foreign bodies are highly susceptible to infections and other life threatening complications. Therefore, the use of broad spectrum antibiotics after culture and sensitivity studies coupled with safe removal of foreign bodies remains the mainstay of treatment. The precise modus operandi of retrieval always depends upon factors such as the type, size, shape and location of foreign object. Never the less, minimally invasive retrieval modalities are encouraged whenever deemed appropriately. Urethrovesical foreign bodies are frequently encountered in our clinical practice. It is therefore very essential to have high index of suspicion when reviewing patients with acute or chronic lower urinary symptoms.

Keywords: Iatrogenic foreign body, Urethra, Urinary bladder, Foreign body, Self-insertion

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INTRODUCTION

Urinary bladder and urethra remain the main sites where foreign bodies often get stuck along the genitourinary system [1]. Urethrovesical foreign bodies are encountered in both male and female patients. However, they are more common in the latter group due to the presence of a short urethra.

Innumerable objects have been retrieved from the urethra and urinary bladder ranging from hairpin to a toothbrush [1–7]. Presence of a foreign body in the urinary bladder or urethra is a urologic emergency and should always be treated as such.

Late presentation may be fraught with undesired sequelae such as urinary bladder stone formation,
diverticulum, urethral stricture, and erectile dysfunction.

Two cases are presented herein, first being a 34-year-old male paraphiliac who inserted flex electrical wire into his own urethra and urinary bladder for auto-erotic stimulation purposes. Second case is a foreign body iatrogenically introduced into urinary bladder of a 52-year-old male during open prostate surgery. After few days the foreign body sluggishly migrated towards the meatal opening.

Our case series are primarily intended to document atypical presentation of urethrovesical foreign bodies and concurrently reminds clinicians to consider foreign body as a differential diagnosis when evaluating patients with lower urinary tract symptoms.

**CASE REPORT**

**Case 1**

A 34-year-old male who was referred to our hospital with one day history of dysuria, suprapubic pain and dribbling of blood tinged urine. Patient reported that these symptoms had followed self insertion of electrical wire into his own urethra for purpose of achieving sexual ecstasy.

Patient gave no history of associated fevers or chills. He also gave no history of having been admitted or treated for mental illness prior to this incidence. He does not smoke or use tobacco in any form; neither does he take alcohol or use any drugs for pleasure. Patient is single and peasant famer by occupation and gave no history of previous surgery.

Patient attempted to remove the wire several times with no success and in the process the inserted wire was pushed even deeper. (It is worth noting that this piece of information was obtained after repeated and protracted conversions with the patient who initially seemed not willing to readily divulge any information).

On examination, major findings were on local examination however on general examination we saw a young man, in good nutritional status, afebrile, not pale, well oriented but looked worried and rather anxious.

Local perineum and genitalia examination revealed few drops of blood stained urine per urethral meatus, but no visible foreign body noted. Also there was no any sign suggestive of genital trauma. However, unusual object could be palpated around the penoscrotal junction.

Per abdominal examination revealed no bladder distension but he had obvious mild suprapubic tenderness. The rest of systemic examination was essentially normal.

Patient had baseline investigations done which included urinalysis and full blood count. The former showed plenty of red blood cells but no pus cells, where as the latter was normal.

The most informative investigation was the pelvic radiograph anterior-posterior view which showed a long coiled up radio-opaque shadow extending along the entire urethra to the urinary bladder region (Figure 1). This confirmed the presence of urethrovesical foreign body.

Considering the pelvic radiograph findings patient was planned for suprapubic cystotomy to remove the foreign body. A long white flex electrical insulated wire with a complex knot was successfully extracted. The wire was 164 cm long when fully stretched (Figure 2). His postoperative period and serial follow up visits one year later was uneventful.

**Case 2**

A 52-year-old male, who presented to our centre with three hours history of protruding foreign body per urethral opening. Patient’s medical records and reports suggested that three weeks earlier he had undergone open prostatectomy.
He reported that he had fairly uneventful postoperative period and was discharged home ten days later. He had no catheter at time of discharge.

Nine days after discharge, he developed acute urine retention which necessitated suprapubic puncture and catheterization. There after he was referred to our hospital. However, before his arrival to our centre, he started experiencing migratory excruciating urethral pain which was associated with burning sensation and an intense urge to void.

Initially pain was more marked around the root of the penis and later kept on migrating towards the anterior urethra. Two days later, he was shocked to note a white plastic object protruding per his urethral opening (Figure 3).

On examination, we saw a middle aged man, anxious, well oriented, afebrile, not pale, and had no pedal oedema. Local examination of the genitalia revealed a conical shaped plastic object protruding per urethral meatus, otherwise normal genitalia.

Per abdomen, he had an indwelling suprapubic catheter, with midline sub umbilical incisional scar. Urinary bladder was not distended. The rest of abdominal examination was normal and the other systemic examination was unremarkable.

Laboratory work up revealed mild elevation of blood urea and creatinine. Urinalysis revealed many pus cells with insignificant bacterial growth.

Patient was prepared for emergency surgery whereby simple meatotomy was performed and the foreign body was easily removed out (Figure 4).

The foreign body turned out to be a cap of 60 mL irrigation syringe which was inadvertently forgotten inside the urinary bladder during open prostatectomy. The syringe cap gradually migrated towards the anterior urethra.

It was conical shaped with wider base diameter than the apex, a factor that may have prevented spontaneous expulsion and instead got stuck in the meatus (Figure 5). Patient had quick and uneventful recovery. Follow up visits, two years later revealed no urethral or meatal stricture.

**DISCUSSION**

Foreign bodies may get access to the urethra and urinary bladder either from deliberate act of self insertion, so as to attain sexual gratification or iatrogenically introduced into lower urinary tract during various therapeutic manoeuvres [1–5].

Seldom, urethrovesical foreign bodies have been reported to have migrated from contiguous structures [6]. Other factors that may prompt patients to inserts foreign bodies into their own genitourinary tracts include psychiatric disorders, senility, intoxication and occasionally curiosity in minors [4, 5].

A myriad of objects have been retrieved from the urethra and urinary bladder. Such foreign objects include safety pins, pocket battery, pencil and drinking straws just to mention a few [1–7].
In such scenarios management goals include, attaining correct diagnosis, preventing complications and safe extraction of a foreign body. Usually patient’s clinical presentation differ, however majority may present with hematuria, urethritis, cystitis and urinary retention [4].

Thorough history taking, physical examination coupled with appropriate investigations often leads to correct diagnosis. Occasionally obtaining a correct history from paraphilia, minors, drug abusers and mentally disturbed individuals may be difficult. Patience and high index of suspicion is therefore required.

Investigations selection more often hinges on patient’s presentation and clinician discretion. Investigations are usually specifically tailored and made appropriate to the case at hand. However, apart from hematological and biochemical work up more specific radiological investigations such as plain X-rays, ultrasonography, intravenous urogram, urethrogram and cystogram may be employed [3, 4].

While plain X-rays are useful to delineate radiopaque objects, the rest are useful in case of radiolucent foreign bodies. When available, cystoscopy can be used for both diagnostic and therapeutic purposes.

Definitive treatment goal is removal of the foreign body with no or minimal trauma. By far there is no widely acceptable treatment algorithm with regards to the presence of foreign bodies in the lower urinary tract.

Usually, decision regarding the best treatment depends on a number of factors such as patient’s condition, the foreign body’s shape, size, location and associated complications like encrustation and stone formation.

In most cases, transurethral cystoscopic removal is considered ideal, usually utilizing endoscopic forceps, snares, ballon-wires and stone-retrieving baskets. Large foreign bodies may be removed by suprapubic cystotomy in case endoscopic attempt is futile or no scopes available. Laparoscopic extraction is also becoming popular in some centers. Sometimes combined approaches may be required depending on circumstances [2–5].

In case of paraphilia, mentally unsound individuals and minors, a holistic approach is a prerequisite.

Thus, therefore a psychiatrist or medical psychologist should always be part of the multidisciplinary treatment team.

CONCLUSION

Urethrovesical foreign bodies are fairly common. Their diagnosis and subsequent management may be challenging due to multifaceted clinical presentation and diversity of objects that are incriminated. It is imperative therefore to always consider foreign body as a differential diagnosis when evaluating patients with acute or chronic lower urinary tract symptoms.

Author Contributions
Sefu Juma Uledi – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published
George Lodewijk Pape – Acquisition of data, Drafting the article, Final approval of the version to be published
Fauzia Ayubu Masumai – Conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflicts of Interest
Authors declare no conflict of interest.

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REFERENCES
Mitral valve endocarditis with high level aminoglycoside resistant *Enterococcus faecalis* in breast cancer patient

Ashish Bhargava, Vasavi Paidpally, Pragati Bhargava

**ABSTRACT**

Introduction: Enterococci represent the third most common cause of infective endocarditis, after *streptococcus* and *Staphylococcus aureus*, and are responsible for 5–20% of all cases of endocarditis. We present a rare case of native valve *Enterococcus faecalis* (E. faecalis) endocarditis of mitral valve in a breast cancer patient. Case Report: A 56-year-old breast cancer patient presented with complaints of fever and found to have *E. faecalis* bacteraemia. Echocardiogram showed mitral valve vegetations. This *E. faecalis* strain was also found to have high level aminoglycoside resistance (HLAR). She responded well to six weeks ampicillin and ceftriaxone combination therapy despite loss of aminoglycoside synergy. Conclusion: Most commonly observed risk factors for enterococcal endocarditis were rheumatic fever, valvular heart abnormalities, gastrointestinal neoplasia, surgery (dental surgery, cardiovascular surgery, and abdominal surgery), gastrointestinal procedures and diabetes. Left-sided enterococcal endocarditis in our patient responded well to combination regimen with ampicillin and ceftriaxone despite loss of aminoglycoside synergism.

**Keywords:** *Enterococcus faecalis*, Endocarditis, Mitral valve, Breast cancer

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**INTRODUCTION**

Enterococci are feared nosocomial pathogen since the emergence of resistant strains, which are challenging to treat. Despite the common occurrence of *E. faecium* bacteraemia, endocarditis caused by this organism is relatively rare, with frequency occurrence less than 10% among patients with enterococcal bacteraemia [1, 2]. Infective endocarditis is also unusual in cancer patient in absence of intravenous drug abuse. According to our medline literature search, there is no case reported about enterococcal endocarditis associated with breast carcinoma till now. We present a rare case of native valve *E. faecalis* endocarditis of mitral valve in a breast cancer patient. This challenging case was also complicated with loss of treatment synergism due to high level aminoglycoside resistance (HLAR).

**CASE REPORT**

A 56-year-old African-American female patient came to the infusion centre for her chemotherapy and found
to have high fever. She had stage IV estrogen/progestrone and her 2/neu receptor negative invasive ductal carcinoma of right breast status post palliative mastectomy which was diagnosed in 2010. For this locally advanced tumor she was started on chemotherapy with carboplatin and gemcitabine. Three months prior to her current admission, she received her last dose. During that visit, she was noted to have positive blood cultures with *E. faecalis*. This bacteremia last for four days and was noted secondary to an infected PICC line. She was given intravenous ampicillin and bactereemia resolved. Due to prolonged bacteremia, a transeosophageal echocardiogram was done which revealed no valvular vegetations. She was then treated with line removal and total two weeks of ampicillin treatment from PICC line removal and negative culture.

At this admission, patient was noted to have temperature of 38.4°C. She denied any other complaints such as chest pain, shortness of breath, palpitations, nausea, vomiting, diarrhea, dysuria, rash, chills or mediport site pain. Her blood pressures were stable with normal range pulse rate. On examination, mediport was noted on right side of the chest. There was no erythema, swelling or tenderness over the site. She was noted to have chronic lymphedema of her right arm since her mastectomy. Laboratory findings were normal except for anemia (see Table 1). Chest X-ray showed a left internal jugular port catheter tip over upper cavoatrial junction without any other significant findings. She was started on empiric therapy with vancomycin.

Blood cultures drawn upon admission showed *E. faecalis* which was ampicillin sensitive but resistant to aminoglycosides. So, she was switched to intravenous ampicillin 2 g every 4 h. She was noted to have persistent bacteremia for four days. Transthoracic echocardiogram showed 2 nodular, immobile echogenic densities measuring 0.8x0.6 cm attached to tip of the anterior mitral valve leaflet along atrial and ventricular aspects. Transesophageal echocardiogram revealed a large mobile bilobar echodensity measuring 1.7x0.8 cm attached to the atrial aspect of anterior mitral leaflet tip, consistent with vegetation. She was then started on ceftriaxone 2 g daily for synergy as *E. faecalis* had high level aminoglycoside resistance. Her blood cultures became negative after 48 h of combination therapy. She was discharged on intravenous ampicillin and ceftriaxone for six weeks. Repeat cultures done after discharge remained negative. During her three months follow up after completion of her intravenous therapy, patient has been doing well.

**DISCUSSION**

Enterococci are gram-positive, catalase-negative, facultative anaerobic bacteria, which usually inhabit the alimentary tract of humans in addition to being isolated from environmental and animal sources. Although initially enterococci were generally considered harmless commensals, studies have documented the pathogenic potential of these organisms and, in fact, shown to be the third most common cause of nosocomial bacteremia [3]. The other factor which draws attention is serious enterococcal infections are often refractory to treatment and with a higher mortality [4]. Enterococci represent the third most common cause of infective endocarditis, after *Streptococcus* and *Staphylococcus aureus*, and are responsible for 5% to 20% of all cases of endocarditis [5].

*E. faecalis* is responsible for the vast majority of cases of enterococcal endocarditis [6]. Only a minority of cases are caused by other species, such as *E. durans*, *E. hirae*, and *E. avium*. The presentation of enterococcal endocarditis is typically subacute and infrequently associated with peripheral stigmata of endocarditis. Most cases of enterococcal endocarditis are left-sided [7]. The most commonly observed risk factors were rheumatic fever, valvular heart abnormalities, gastrointestinal neoplasia, surgery (dental surgery, cardiovascular surgery, abdominal surgery), gastrointestinal procedures and diabetes [4, 8] (see Table 2). From International collaboration of endocarditis database, enterococcal endocarditis was most frequently seen in elderly men, frequently involved the aortic valve and tended to produce heart failure rather than embolic events [7].

The other significant factor about enterococci is its notorious nature of increasing antibiotic resistance which makes it feared pathogen that is challenging to treat. After the first report, in the late 1970s, of clinical isolation of *E. faecalis* with high level of aminoglycoside resistance, the number of infections caused by HLA strains are increasing. Enterococcus species do not possess cytochrome enzymes and thus cannot produce

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**Table 1: Biochemical profile of patient on admission.**

<table>
<thead>
<tr>
<th>Chemistry – Value (Range)</th>
<th>Complete Blood Cells</th>
<th>Microbiology (Blood Culture)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sodium – 135 mmol/L (135–145)</td>
<td>WBC – 8.3 K/CUMM (3.5–10.6)</td>
<td>Blood cultures (2 sets) –</td>
</tr>
<tr>
<td>Potassium – 4.6 mmol/L (3.5–5.3)</td>
<td>Hemoglobin – 8.7 g/dL (11–15)</td>
<td><em>E. faecalis</em></td>
</tr>
<tr>
<td>BUN – 24 mg/dL (7–20)</td>
<td>Hematocrit – 26.5% (34.4–44)</td>
<td></td>
</tr>
<tr>
<td>Creatinine – 1.4 mg/dL (0.4–1.1)</td>
<td>Platelets – 282 K/CUMM (150–450)</td>
<td></td>
</tr>
</tbody>
</table>
Table 2: Risk factors for enterococcal endocarditis and HLAR enterococcal infection.

<table>
<thead>
<tr>
<th>Risk factors for enterococcal endocarditis</th>
<th>Risk factors for HLAR enterococcal infection</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Gastrointestinal malignancy</td>
<td>Prior antibiotic therapy</td>
</tr>
<tr>
<td>2. Surgery</td>
<td>Patient received more</td>
</tr>
<tr>
<td>3. Rheumatic fever</td>
<td>than four antibiotics</td>
</tr>
<tr>
<td>4. Diabetes</td>
<td>Patient treated with cephalosporin</td>
</tr>
<tr>
<td>5. Valvular heart abnormalities</td>
<td>Urinary catheter</td>
</tr>
<tr>
<td>6. Gastrointestinal procedures</td>
<td></td>
</tr>
</tbody>
</table>

Ineffective initial treatment may require valve replacement for cure [13] or can be devastating with loss of patient life even with valve replacement [14].

**CONCLUSION**

In conclusion our patient with left-sided enterococcal endocarditis responded well to combination regimen despite loss of aminoglycoside synergism.

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**Author Contributions**

Ashish Bhargava – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Vasavi Paidpally – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Pragati Bhargava – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, 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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**REFERENCES**


An unusual case of pigmented villonodular synovitis after unicompartamental knee arthroplasty

Tomohiro Onodera, Hiroshi Tanji, Tokifumi Majima, Tamotsu Kamishima, Akio Minami

ABSTRACT

Introduction: Pigmented villonodular synovitis (PVNS) after replacement arthroplasty is a potential cause of postoperative pain and hemarthrosis. The disease is difficult to diagnose after arthroplasty, since magnetic resonance imaging (MRI) appearances are subject to metal artifact making them less than ideal. Case Report: A case of a 61-year-old woman with PVNS after unicompartamental knee arthroplasty (UKA). She was treated with an arthroscopic synovectomy with good result. Conclusion: MRI evaluation of the residual side of the knee joint after UKA is valuable for the differential diagnosis of PVNS.

Keywords: Pigmented villonodular synovitis (PVNS), Unicompartamental knee arthroplasty (UKA), Hemarthrosis, magnetic resonance imaging (MRI)

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INTRODUCTION

Pigmented villonodular synovitis (PVNS) is a relatively rare, proliferative disease affecting the synovial joints resulting in villous or nodular changes in the synovial tissue, large effusion and bony erosions. PVNS after replacement arthroplasty is rare and to our knowledge, a few cases have been reported in literature [1–5]. We report a case of diffuse PVNS after unicompartamental knee arthroplasty (UKA). This is the first report that uses magnetic resonance imaging (MRI) for differential diagnosis of PVNS after UKA. This disease is difficult to diagnose after arthroplasty, due to artifact typically observed on MRI. However, the appearance of the residual side of the joint on MRI can contribute to the diagnosis of PVNS in UKA patients who present with knee pain and recurrent effusion/hemarthrosis [6].

CASE REPORT

A 61-year-old woman presented with a 5-year history of progressive pain in her knee. Examination and X-ray findings were consistent with a diagnosis of osteoarthritis. She underwent cemented unilateral knee
arthroplasty (Zimmer Unicompartmental High Flex Knee System, Zimmer, Warsaw, Ind) without complication. At the time of surgery, no abnormal synovial membrane findings were detected, and synovectomy was not performed (Figure 1). Although postoperative radiographs demonstrated the oversized prosthesis, the patient progressed well in the first year after the UKA, with a range of motion from 0° to 130° and excellent ligamentous stability. However, three years after surgery, she had a spontaneous onset of right knee pain and swelling without history of trauma. She had mild medial-sided tenderness and a moderate knee effusion without erythema or warmth upon examination. Inflammatory markers at the time were normal. Plain radiographs were unremarkable (Figure 2). Arthrocentesis of the right knee showed bloody aspiration, and revealed no growth with cultures. MRI of the right knee was performed using a 0.35 Tesla. Axial T1-weighted spin-echo (TR/TE = 600/15 m-s) and gradient-echo (TR/TE = 800/30 m-s, 30° flip angle) MR images revealed villous shaped synovial proliferation with a large amount of joint effusion (Figure 3). Mild blooming artifact was seen in the synovium on the gradient-echo image. These imaging findings in addition to the bloody aspiration were compatible with PVNS. The patient was then considered to be a candidate for total arthroscopic synovectomy. The synovium was observed to be diffusely inflamed and hemosiderin colored at the time of surgery (Figure 4). The components were well positioned, tracked well, and no polyethylene wear was observed. There was no growth on operative cultures, and histologic analysis of synovial samples revealed PVNS (Figure 5). The patient had no pain with a mild effusion at the one-year follow-up visit after arthroscopic synovectomy.

DISCUSSION

Pigmented villonodular synovitis is a benign proliferative synovial disorder of unknown etiology that can affect bursae, tendon sheaths, and joints [7, 8]. The incidence of PVNS is 1.8 per one million in the general population and the knee is the most common joint to be affected [9].

It has been reported that PVNS is diagnosed an average of 4.4 years after presentation without MRI [10]. The decision to perform arthroscopic observation in a patient who previously underwent arthroplasty is difficult due to the artifact with the MRI and the risk of infection. Articular implants are considered as devices, which limit MRI visualization due to the induction of susceptibility artifacts. However, the MRI in the residual portion of the joint in patients with a history of UKA is visible [6]. In this case, MRI was safely applied and was valuable in making the decision to proceed with the arthroscopy. Our case shows the clinical value of MRI of patients with UKA.

The etiology of PVNS is unknown, although chronic recurrent micro-trauma and hemanarthrosis have been suggested [9]. There were two previous reports in
CONCLUSION

The evaluation of the residual side of knee joint after UKA by MRI is potentially valuable for the different diagnosis of PVNS.

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Author Contributions

Tomohiro Onodera – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Hiroshi Tanji – Substantial contributions to conception and design, acquisition of data, Drafting the article, Final approval of the version to be published

Tokifumi Majitma – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Tamotsu Kamishima – Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Akio Minami – Substantial contributions to conception and design, 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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REFERENCES


CASE REPORT

Cystadenoma of the appendix presenting with an intestinal obstruction of the ileum: A case report

Tadahiro Nozoe, Katsuo Sueishi, Mayuko Kohno, Tomohiro Iguchi, Takashi Maeda, Takahiro Ezaki

ABSTRACT

Introduction: Small bowel obstruction due to an appendiceal mucocele is comparatively rare. Case Report: A rare case of a small bowel obstruction due to a mucocele of the appendix, which had been histopathologically diagnosed as a mucinous cystadenoma. Partial resection of the ileum and appendectomy through traditional laparotomy relieved the patient from abdominal pain and congestion from the intestinal obstruction. Conclusion: Small bowel obstruction due to an appendiceal mucocele should be considered among the differential diagnosis of intestinal obstruction for patients who have no previous history of laparotomy.

Keywords: Appendix, Mucocele, Cystadenoma, Small bowel obstruction

INTRODUCTION

Intestinal tumors [1], mucosal edema of the intestine due to enteric anisakiasis [2], and malrotation of the bowel [3] are often the causal lesions in patients with small bowel obstruction and no previous history of laparotomy or intestinal tumors. Appendiceal mucocele is a comparatively a rare disease and intestinal obstruction other than intussusception of the bowel caused by appendiceal mucocele is quite atypical. A case of small bowel obstruction caused by compression by a mucinous cystadenoma of the appendix has been given here.

CASE REPORT

A 74-year-old male came to our institute with complains of abdominal pain and vomiting. He had no previous history of laparotomy.

A long tube was inserted to reduce the symptoms of abdominal fullness and a small bowel series demonstrated a complete obstruction of the ileum (Figure 1). Magnetic resonance imaging (MRI) suggested a cystic lesion that might be correlated with the small bowel obstruction (Figure 2).

Laparotomy revealed a cystic lesion located in the ileum, 80 cm from the terminal ileum causing a complete compression of the small bowel. Moreover, the entire cystic lesion was continuous with the appendix, a partial resection of small bowel and appendectomy was performed. The cystic lesion was found on the edge of the appendix (Figure 3A). Although the lumen of the ileum was completely occluded by the cystic lesion, the mucosa of the ileum was not involved with the tumor...
(Figure 3B). The cystic lesion contained the transparent mucin (Figure 3C).

The wall of the cystic lesion was lined with intestinal epithelium without atypia associated with chronic inflammation of the appendiceal mucosa and there was mucin secreting epithelium lining the appendix (Figure 4). The lesion demonstrated the pathological features of mucinous cystadenoma of the appendix.

**DISCUSSION**

Appendiceal mucoceles are defined as a group of lesions in which the appendiceal lumen becomes distended with mucus. The lesions are histopathologically divided into three subtypes: mucosal hyperplasia, mucinous cystadenoma, and mucinous cystadenocarcinoma [4].

Appendiceal mucocele is a comparatively rare disease and it often remains asymptomatic during the clinical course, and the disease itself has been accepted to be a neoplasm with low grade malignant potential, other than cystadenocarcinoma associated with pseudomyxoma peritonei.

Figure 2: Magnetic resonance imaging. A 5 cm cystic lesion caused the small bowel obstruction.

Figure 3: Macroscopic findings. (A) The cystic lesion was on the edge of the appendix, (B) No abnormal finding was observed in the mucosa of the ileum, (C) Cystic lesion containing transparent mucin.

Tumorous lesions including GIST, carcinoid tumor, lymphoma, primary cancer, and metastasized carcinoma usually metastasize from extra-intestinal melanoma; lung cancer and breast cancer have been reported as causal lesions for obstruction of the small intestine [5, 6].
Although there are a few reports of intussusception caused by appendiceal mucocele [7, 8], small bowel obstruction caused by a simple compression of appendiceal mucocele has been rarely presented [9].

Magnetic resonance imaging demonstrated a cystic lesion obstructing the ileum in the current patient, and a GI series showed the intestinal obstruction was far from the terminal ileum. These findings suggested that a cystic tumor in the small intestine or an appendiceal mucocele. While no definitive diagnosis could be made preoperatively and the latent possibility of cystadenocarcinoma could be also considered as a differential diagnosis.

An absolutely perfect dissection of the cystic lesion without any injury is required to avoid peritoneal dissemination [10], especially in case of appendiceal cystadenocarcinoma in association with an aggressive pseudomyxoma peritonei [11].

A conventional laparotomy was performed in the current case, and the lesion was eventually diagnosed as cystadenoma of the appendix. The patient recovered with no adverse events.

CONCLUSION

Appendiceal mucocele should be considered one of the causal lesions for small bowel obstruction in patient who does not have any history of laparotomy.

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Author Contributions

Tadahiro Nozoe – 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

Katsuo Suseishi – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

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Guarantor

The corresponding author is the guarantor of submission.

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Benign or malignant? Extensive pulmonary metastasis of an intracranial meningioma—unique radiographic and histopathologic features

Roman Leonid Kleynberg, Leonid Markus Kleynberg, Vera M Kleynberg

ABSTRACT

Introduction: Meningiomas are slow growing benign tumors of the central nervous system. Although local recurrence does occasionally occur, extracranial metastasis of meningiomas is exceedingly rare and rarely reported, occurring in less than 1 per 1000 cases. Most commonly, metastasis occurs to the lung, and most cases of pulmonary cases are detected incidentally, followed by liver, lymph nodes, and bones. Case Report: A 67-year-old Caucasian male with a past medical history of intracranial meningioma status-post resection and radiation treatment who developed multiple pulmonary metastases 29 years following complete cranial tumor resection. He presented to our hospital with respiratory failure and subsequent imaging revealed multiple pulmonary nodules in his chest. These lesions were biopsied: the final diagnosis was metastatic malignant meningioma. He developed mitral valve endocarditis and passed away from multisystem organ failure. Discussion: Extracranial metastasis of meningiomas is rare, occurring in approximately 0.1% of all meningioma cases. Several factors that are commonly associated with metastatic meningiomas include previous intracranial surgery for meningioma, dural sinus invasion, malignant histology, local recurrence, and a high Ki-67 labeling index. Conclusion: A doctor must be able to obtain a good history in the initial evaluation of a patient—the facts of which can help in narrowing down the list of differential diagnosis. Therefore, although distant metastases is a rare event, the possibility of pulmonary metastasis nevertheless is real and must not be ignored in patients with a past history of meningioma presenting with pulmonary symptoms.

Keywords: Pulmonary metastasis, Intracranial meningioma.

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INTRODUCTION

Meningiomas are slow growing benign tumors of the central nervous system. Although local recurrence does occasionally occur, extracranial metastasis of meningiomas is exceedingly rare and is rarely reported. A patient who developed multiple pulmonary metastases 29 years after a complete tumor resection is given here. He presented to our hospital with respiratory failure and subsequent imaging revealed multiple pulmonary nodules in his chest. These lesions
were biopsied which resulted in the diagnosis of metastatic malignant meningioma. Therefore, although distant metastases is a rare event, the possibility of pulmonary metastasis nevertheless is real and must not be ignored nor overlooked in those patients with a past history of meningioma presenting with pulmonary symptoms.

CASE REPORT

A patient (BZ) presented to our emergency department in the month of March 2011 with chest pain and dyspnea. He is a 67-year-old ex-smoker male with a past medical history significant for systemic hypertension, hyperlipidemia, coronary artery disease status post two vessel bypass surgery, history of congestive heart failure, mitral valve replacement in 1987, as well as a right-sided cerebral meningioma in 1979 status-post resection and radiation treatment. In 1982, BZ, then a 38-year-old, suffered a motor vehicle accident and underwent extensive brain surgery for treatment of complications from the accident. In late 1995, on a routine chest radiogram, multiple well-defined pulmonary nodules were discovered (no prior chest radiograms at the time were available for comparison). The nodules were subsequently biopsied and were found to be benign. A second biopsy one year later, in 1996 yielded the same results. The patients’ pulmonary nodules were followed over a course of several years and they remained unchanged.

The patient did well during follow-up until 1999, when he was admitted for fatigue, weight loss and shortness of breath and was found to have lymphadenopathy. Subsequent imaging, blood analysis and biopsy confirmed the diagnosis of stage IV non-Hodgkin’s lymphoma. He was then treated with CHOP systemic chemotherapy and went into remission. In March 2011, he presented to our emergency department with acute shortness of breath and a workup revealed acute mitral regurgitation as the underlying cause. During his stay at our facility, multiple pulmonary nodules were discovered on chest radiogram (Figure 1) and computed tomography (CT) scan showed multiple multilobar lung masses, measuring up to 8.4x5.3 cm. Magnetic resonance imaging (MRI) of the brain showed recurrent versus residual meningioma (Figure 2)—however this was not further biopsied. A left upper lobe nodule fine-needle aspiration revealed metastatic malignant meningioma via light microscopy (Figure 3) and immunohistochemistry staining (Figure 4). A surgical resection was indicated because of the doubling of one of the lung masses (RLL nodule from 3x3 cm to 3x6 cm in less than twelve months), in order to confirm the diagnosis. Again, metastatic malignant meningioma was confirmed by biopsy. PET scan showed multiple hypermetabolic lesions throughout the lung. Unfortunately, soon after, the patient deteriorated after developing mitral valve endocarditis and died from multi-system organ failure.

![Figure 1](image1.png)  
**Figure 1:** (A) Chest roentgenogram shows multiple pulmonary nodules, two in the right and two in the left lung lobes (marked off with measurements), (B) The chest computed tomography showing multilobar lung masses during a CT-guided fine needle aspiration of a right posteriomedial pleural mass.

![Figure 2](image2.png)  
**Figure 2:** Imaging of malignant meningioma. (A) Transverse magnetic resonance imaging T1-weighted postgadolinium image followed by, (B) Transverse T2-weighted postgadolinium image showing postsurgical changes status-post parietal-occipital craniotomy with extensive encephalomalacia (softening of cerebral tissue) and old hemorrhage in the right parietal region. Lobular tissue extending along the posterior falx presumably related to the patient’s history of meningioma, suggesting residual/recurrent tumor.

![Figure 3](image3.png)  
**Figure 3:** Light microscopy with hematoxlin and eosin staining of metastatic malignant meningioma from lung biopsy. (A) Whorled clusters of spindle cells, (B) Cells with a high mitotic index with a calcified psammoma body in center of image (arrow).

DISCUSSION

Meningiomas are slow-growing benign tumors confined to the intracranial space and are generally treated with surgical removal [1]. Most originate from
CONCLUSION

Surgical resection is currently the gold standard for treatment of pulmonary metastatic meningiomas [3]. In addition, local control of primary intracranial meningiomas and en bloc resection of metastatic tumors confined to the lung are other options for management of the disease [3]. In conclusion, this case brings to the forefront two important points. The first is that in patients with a history of intracranial meningioma, the possibility of extrapulmonary metastatic meningiomas must be considered in those that present with pulmonary nodules. The second is that although surgical excision remains as the standard of care for patients with pulmonary metastatic meningiomas, chemotherapy may be beneficial in patients that cannot tolerate surgery, and follow-up with CT, FDG-PET/CT or 111In-octreotide scintigraphy is necessary in staging patients with metastatic meningioma who are considered for further treatment. Because of the rarity of metastatic meningiomas, no controlled study has been performed to establish a efficacious chemotherapeutic regimen for metastatic meningiomas, and only a single case has shown any efficacy of chemotherapy for metastatic meningiomas [9]. Therefore, to improve disease outcomes, further chemotherapy regimens must be explored and researched.

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Author Contributions
Roman Leonid Kleynberg – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published
Leonid Markus Kleynberg – Conception and design, Critical revision of the article, Final approval of the version to be published
Vera M Kleynberg – Acquisition of data, Drafting the article, Final approval of the version to be published

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The corresponding author is the guarantor of submission.
Conflict of Interest
Authors declare no conflict of interest.

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Ameloblastomatous calcifying ghost cell odontogenic tumor: A case report

Monal Bhaurao Yuwanati, Jagdish Vishnu Tupkari, Shubhangi Mhaske, Avadhoot Avadhani, Pradnya Joshi

ABSTRACT

Introduction: Calcifying ghost cell odontogenic tumor (CGCOT) is an uncommon developmental odontogenic cyst first described by Gorlin et al. in 1962; represent a heterogeneous group of lesions that exhibit a variety of clinicopathologic and behavioral features. Case Report: A 63-year-old female reported with a painless swelling in mandible for three months. After detailed clinical and histopathological examination, it was diagnosed as ameloblastomatous CGCOT (Type III) and operated. Conclusion: CGCOT (Type III) is considered to be rare and accounts for only 1% of jaw cysts reported. CGCOT has been classified under two basic groups namely, cystic and neoplastic. Because of its diverse histopathology, there has always been confusion about its nature as a cyst, neoplasm or hamartoma and its behavior. A very few cases of ameloblastomatous CGCOT (Type III) have been reported in literature. The proper surgical procedure is to be followed to minimize chances of recurrence.

Keywords: Calcifying odontogenic cyst, Ameloblastomatous calcifying ghost cell odontogenic tumor (CGCOT), Calcifying ghost cell odontogenic tumor, Ghost cell


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INTRODUCTION

The odontogenic cysts are group of lesions frequently observed in the head and neck regions. The odontogenic epithelium is involved in the formation of cysts in majority of the cases. A calcifying odontogenic cyst (COC) occurs occasionally in the oral region [1]. It was first described by Gorlin et al. and now included in the group of odontogenic tumors in a World Health Organization (WHO) international classification proposed in 1992, in which there is odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation. It is well known that this lesion often occurs in association with odontogenic tumors e.g., odontoma ameloblastoma and adenomatoid odontogenic tumor and all recent
histopathological classifications of COCs advocate categorizing the variants associated with these tumors. Calcifying odontogenic cyst may clinically be diagnosed as calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor, unicystic ameloblastoma, ameloblastic fibro-odontoma, odontoma, dentigerous cyst or other odontogenic cysts. In a Recent WHO classification [2, 3], the term COC has been replaced by calcifying ghost cell odontogenic tumor (CGCOT). The association of ameloblastoma with this lesion is important as it will affect the clinical and treatment outcome. The CGCOT has been classified under two basic groups namely, cystic and neoplastic as per dualistic concept. According to Hong et al. [4], there is an ameloblastomatous variant of COC which is characterized by a unicystic structure. The epithelial lining shows unifocal or multifocal intraluminal proliferative activity that resembles ameloblastoma. Although it also contains isolated or clustered ghost cells and calcifications. Since it is associated with ameloblastomatous proliferation, there is a confusion regarding the nature, behavior as well as prognosis of this rare entity. The purpose of this paper is to describe a case of ameloblastomatous COC/CGCOC and to present a review of cases found in literature.

CASE REPORT

A 63-year-old female patient (Indian) was referred to the Department of Oral and Maxillofacial Pathology, Government Dental College and Hospital, Mumbai. Complaining of an asymptomatic swelling in the right anterior region of mandible that was present since approximately three months. The clinical examination revealed a painless, non-tender swelling over the right parasympysis region of mandible along with slight facial asymmetry. The mucosa overlying the lesion was normal with expansion of both the cortical plates and showed obliteration of the labial vestibule (Figure 1). The Panoramic radiograph revealed a well-defined multilocular radiolucency from the lower left side central incisor to the right first molar (Figure 2). Calcifications was not clearly evident on radiograph. On aspiration a blood tinged fluid was seen. The clinical diagnosis of an odontogenic cyst was made and an incisional biopsy was obtained from the right parasympophysial area to establish a definitive diagnosis. The differential clinical diagnosis included muticystic ameloblastoma and odontogenic keratocyst.

Microscopic examination revealed a cystic lesion lined by odontogenic epithelium with eosinophilic ghost cells (Figure 3) and calcification within the ghost cells. The stroma of the cyst also demonstrated ameloblastic islands, (Figure 4), sheets of eosinophilic ghost cells with areas of calcification (Figure 5). A histopathological diagnosis of ameloblastomatous COC/CGCOT (Type III) was made. The patient was surgically operated for the lesion and healing was uneventful. Ten months of follow up showed no recurrence.

DISCUSSION

Calcifying odontogenic cysts though rare, are not uncommon in the jaw bones, due to the embryonic odontogenic epithelium or its remnants. The odontogenic cysts are either of developmental or
inflammatory origin. The COC is an uncommon well circumscribed, either solid or cystic lesion derived from odontogenic epithelium that resembles follicular ameloblastoma but contains ghost cells and spherical calcification. Since the first description by Gorlin et al., sporadic cases of COCs have been reported in literature.

From the time when it was first described in literature, it has become clear that COC has a number of variants, including features of a benign odontogenic tumor. The histologic variation of COC has led to different terminologies such as epithelial odontogenic ghost cell tumor, calcifying ghost cell odontogenic tumor, dentinogenic ghost cell tumor and odontogenic ghost cell tumor.

The combined microscopic features of COC and ameloblastoma, merging from one to the other had been

reported in the past. Hong et al. [4] described cases of ameloblastoma occurring in neoplastic variant of COC and suggested the two variety of COC associated with ameloblastoma–ameloblastomatous cystic and neoplastic variant with ameloblastoma. According to Hong et al, the cysts can occur as four variants:

1. Non-proliferative COC: characterized by a simple unicystic structure
2. Proliferative COC: characterized by a cystic structure with multiple daughter cysts, extensive ghost cell formations, and marked tendency for calcification
3. Ameloblastomatous COC: characterized by ameloblastoma-like, cyst-lining epithelium with ghost cells and calcifications
4. COC associated with odontome

The calcifying ghost cell odontogenic cyst (CGCOC) is of central and peripheral variety. The central CGCOC (intraosseous) presents as an asymptomatic hard swelling of the jaw that produces expansion than erosion of bone and pain in case of secondary infection [5]. Intraosseus variety of ameloblastomatous COC has been reported [6]. The lesion usually presents as a painless, unilateral swelling in the premolar–molar region with a well circumscribed mixed radiographic appearance. However, the present case radiographically showed a well circumscribed radiolucency extending contralaterally up to the mesial of left lateral incisor.

Histopathological findings described in previous reported case of ameloblastomatous COC were evident in present case. It was characterized by a unicystic structure in which the lining epithelium showed intrameluminal proliferative activity that resembles ameloblastoma and also contains isolated or clustered ghost cells and calcification. It can be differentiated from ameloblastoma ex COC which shows Vickers and Gorlin criteria [7] and lacks ghost cells and calcifications.
The peculiar features of CGCOC is the presence of ghost cells. These ghost cells are mostly found within the epithelium but they can also be found in the connective tissue. These ghost cells are reported to be a type of aberrant keratinization. Takata et al. [8] reported that the ghost cells in calcifying odontogenic cysts, as opposed to ghost cells in dermal calcifying epitheliommas, contain enamel-related proteins in their cytoplasm accumulated during the process of pathological transformation and should not be considered as metaplastic. It was suggested that these cells undergo abnormal terminal differentiation as an apoptotic process [9]. The mechanism for this abnormal terminal differentiation is still not known. The similarity of the immunostaining patterns of cystic and solid calcifying odontogenic cysts supports the view that these lesions are two morphologic variants of the same entity.

Irregular calcified bodies of varying size and opacity may be seen in the radioluent area and in some cases the calcification may be substantial and occupy the greater part of the lesion but in this case calcification was not seen radiographically. Several authors have suggested that, if COC is associated with an ameloblastoma, its behavior and prognosis will be similar to an ameloblastoma, not that to COC. Yoshida et al. [10] based on their immunohistochemical study suggested that COCs with various histological features have neoplastic potential and may not be separate entities within the same histological spectrum. Unlike most of the earlier reported cases the present case did not show any evidence of recurrence after the conservative treatment and follow up of 10 months.

Till date, only 27 cases including the present case are reported (Table 1). The most common clinical symptom was painless swelling unless secondarily infected. The ameloblastomatous COC was more commonly showed equal distribution in jaws as well as in gender predilections. It was reported in age range 11–58 years. Premolar—first molar region was most common site of occurrence. There was no deviation in ameloblastomatous COC in age, gender, site distribution when it was compared with all reported cases of COC in literature irrespective of type of COC. No recurrence was noticed in all ameloblastomatous COC cases which was suggestive of non-neoplastic nature of COC, hence, they can be treated in conservative way. Unfortunately, the mural development of ameloblastoma or

Table 1: Ameloblastomatous COC/CGCOT (Type III) in Reported literature.

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>No</th>
<th>Age</th>
<th>Sex</th>
<th>Clinical symptom</th>
<th>Site/location</th>
<th>Region Area</th>
<th>Follow-up/ Recurrence</th>
<th>Side</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hong et al. [4]</td>
<td>1991</td>
<td>11</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Mitsuhide Yoshida et al. [9]</td>
<td>2001</td>
<td>7</td>
<td>11 to 38 yrs</td>
<td>3 Male, 4 Female</td>
<td>Swelling (5), No symptom (2)</td>
<td>6 Maxilla, 1 Mandible</td>
<td>All cases were in premolar-molar region</td>
<td>No recurrence in 6 case expect in one case after 6 years follow up</td>
<td>Right (4), Left (3)</td>
</tr>
<tr>
<td>Aithal D et al. [1]</td>
<td>2003</td>
<td>1</td>
<td>28</td>
<td>Female</td>
<td>Painless swelling</td>
<td>Mandible</td>
<td>Posterior-Premolar</td>
<td>2 years</td>
<td>No recurrence</td>
</tr>
<tr>
<td>S Iida et al. [2]</td>
<td>2004</td>
<td>1</td>
<td>17</td>
<td>Male</td>
<td>Painless swelling</td>
<td>Mandible</td>
<td>Posterior-Molar premolar</td>
<td>13 years</td>
<td>No recurrence</td>
</tr>
<tr>
<td>Kamboj M et al. [7]</td>
<td>2007</td>
<td>1</td>
<td>58</td>
<td>Female</td>
<td>Pain + Swelling</td>
<td>Mandible</td>
<td>Ant-Post-canine ramus</td>
<td>No recurrence</td>
<td>right</td>
</tr>
<tr>
<td>Mashhadi Abbas F [12]</td>
<td>2009</td>
<td>1</td>
<td>13</td>
<td>Male</td>
<td>Painless swelling</td>
<td>Mandible</td>
<td>Ramus</td>
<td>15 months</td>
<td>No recurrence</td>
</tr>
<tr>
<td>Kamran Nosrati [6]</td>
<td>2009</td>
<td>1</td>
<td>22</td>
<td>Male</td>
<td>Painless swelling</td>
<td>Mandible</td>
<td>Molar</td>
<td>14 months</td>
<td>No recurrence</td>
</tr>
<tr>
<td>Present case</td>
<td>2011</td>
<td>1</td>
<td>63</td>
<td>Female</td>
<td>Painless swelling</td>
<td>Mandible</td>
<td>Anterior Mandible</td>
<td>No recurrence</td>
<td>(Right /Left)</td>
</tr>
</tbody>
</table>

Abbreviations: NA- Not available
ameloblastomatous changes in COC is of unknown clinical significance at this time because of the limited number of cases and limited follow-up information.

CONCLUSION

Ameloblastomatous COC/CGCOT (Type III) shows varying degree of presentation which can be mistaken of any other odontogenic lesion. This review provides some insight to the findings from literature and improves the understanding of this rare entity. Moreover, the present case provides further evidence of a non-aggressive behavior of this lesion.

********

Authors' Contributions
Monal Bhaurao Yuwanati – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published
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Pradnya P Joshi – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

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Abdominal wall hernia following blunt trauma

Navaratnam R

ABSTRACT

Introduction: Despite the relatively high incidence of blunt trauma, traumatic abdominal wall hernia remains relatively rare. Many of the cases reported in literature involve children following handlebar type injuries. Advancing surgical techniques have stimulated much debate with regards to management of such hernias. Case Report: An unusual case of a 59-year-old patient presenting to our emergency department following a road traffic accident suffering from such a hernia is reported here. After appropriate investigation, abdominal exploration was undertaken and a primary mesh repair of the hernia was undertaken, with good result. A discussion of literature pertinent to the infrequent trauma scenario is presented. Conclusion: Traumatic abdominal wall hernias in adults are a rare entity and, in the absence of significant intra-abdominal injury, may be repaired electively.

Keywords: Hernia, Blunt trauma, Laparotomy

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INTRODUCTION

The incidence of trauma continues to rise worldwide while the incidence of traumatic hernia following a blunt injury remain relatively rare with only a handful of cases reported in literature. It is postulated, hernia following blunt abdominal injury accounts for 2% of all traumatic hernia. Traumatic hernia typically follow penetrating injuries, commonly with herniation of bowel. Blunt injuries often do not result in such hernia and this thought to be due to the elasticity of the abdominal wall resisting the shear forces generated by impacts. More typically seen in pediatric patients, they remain infrequent in the adult population. We report one such case and discuss the management of these traumatic hernia.

CASE REPORT

An unusual case of a 59-year-old female patient who presented to the emergency department having been involved in a high-speed vehicular collision has been reported here. She was hemodynamically stable on arrival, with a Glasgow Coma Score of 15/15. On physical examination, only bruising across the abdomen and chest consistent with seatbelt injuries was visible. Abdominal examination revealed a large, left sided swelling. It was tender on palpation and irreducible. Bloods tests and the chest radiography were unremarkable. Computed tomography scan revealed a large left sided abdominal hernia containing mesentery and small bowel. (Figure 1A–B). A laparotomy was performed with the patient’s consent. The bowel was found to be ischemic and a subsequent small bowel resection and end-to-end anastomosis was performed.
The abdominal wall defect was primarily closed and reinforced with a non-absorbable polypropylene mesh. Postoperative recovery was uneventful and the patient was discharged from hospital after 10 days. At most recent follow up her hernia repair was intact with no evidence of recurrence.

DISCUSSION

A traumatic abdominal wall hernia is defined as herniation of viscera through the muscles and fascia of the abdominal wall with the overlying skin remaining intact. First described in 1906 [1], it has a reported prevalence in trauma patients of approximately 1% [2]. Three forms of hernia have been described: (i) a small defect resulting from impact against a blunt object, i.e. handle bars, (ii) a larger defect typically seen following motor vehicle accidents, and (iii) intra-abdominal bowel herniation seen after a deceleration injury. Hernias are more commonly seen in patients involved in high velocity impacts, where a large force is applied to a relatively small area of the abdominal wall. They are typically found in the lower quadrants of the abdomen, possibly due to the absence of the posterior rectus sheath [3]. Though most herniations are evident at the time of presentation, reports exist in literature of delayed presentations, following weakening of the abdominal wall secondary to infection or hematoma formations [4]. As was in our case, in the hemodynamically stable patient, the advent of CT has allowed delineation of abdominal wall defects as well as associated intra-abdominal injuries prior to surgery [5]. However, the association with other intra-abdominal visceral injury is infrequent. We did not find any in our patient. Damschen et al. reported that 17 of 28 patients in their case series had no intra-abdominal injury. It has been argued that this is due to hollow organs being resistant to blunt injury combined with the fact that in reported cases the trauma was delivered to areas away from parenchymatous abdominal organs. Others have argued that given the forces needed to generate such a hernia, a high index of suspicion should be maintained for intra-abdominal injuries that though not initially clinically apparent, may have been missed on initial examination and CT scan. They have advocated a repeat CT scan in the following days [6]. Extra-abdominal injuries have been reported in literature including lumbar fractures and pelvic fractures. Humeral fractures have also been found in association with such herniation. These imply the high kinetic forces that have been transmitted to the body as a result of the trauma [7]. A review of literature drew three conclusions with regards to management of traumatic abdominal wall hernias. Firstly the mechanism of injury should be considered when planning any intervention. Secondly the clinically apparent hernias warrant laparotomy and, finally, that occult hernias may be managed expectantly [2]. However, this paradigm has changed. Traditionally, laparotomy was performed to avoid the complications of associated intra-abdominal injury such as bowel perforation. Now the management of the hemodynamically stable patient with a traumatic abdominal hernia has adopted a more conservative approach with reports suggesting that delayed repair occurring is both safe and feasible [8]. With the increasing use of minimally invasive surgery, reports of laparoscopic repairs of such hernias are growing. Indeed, this method has even been described for the immediate repair of traumatic hernias [9]. Although mesh repair is the procedure of choice for the repair of most abdominal wall hernias, there is consensus in favor of a primary sutured repair [10].

CONCLUSION

Traumatic abdominal wall hernias in adults are a rare entity and, in the absence of significant intra-abdominal injury, may be repaired electively.

**********

Author Contributions
Navaratnam R – Substantial contribution to conception and design, Acquisition of data, Drafting the article, revising it, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

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Postauricular sebaceous cell carcinoma

Suresh K Bhatia, Shivani Atri, Arshad Anjum, Minakshi Sardha, Syed Asmat Ali, Sufian Zaheer, Ruby Bhatia, Prem Singla

ABSTRACT

Introduction: Sebaceous cell carcinoma is an aggressive tumor arising from the sebaceous glands. It is relatively rare tumor and seen almost exclusively on the eyelids (75%). Despite the high concentration of sebaceous glands over head and neck region, true neoplasm, i.e. sebaceous cell adenoma and carcinomas are infrequent. It accounts for just 0.2–0.7% of all eyelid tumors in USA and very few cases that have originated in areas other than the eyelids have been reported. Case Report: A 70-year-old male presented with complaints of rapidly growing swelling (3x4 cm), on the right post auricular region, since one and half months. The patient developed discharging ulcer over the swelling associated with progressive tinnitus and hoarseness of voice, since last twenty days. Fine needle aspiration cytology (FNAC) was advised, which suggested the diagnosis of sebaceous cell carcinoma and excision biopsy was done. Histopathological examination of excised tissue confirmed the diagnosis. Conclusion: Extraorbital sebaceous cell carcinoma is an aggressive and invasive malignancy. It clinically mimics other diseases and is difficult to diagnose. Hence, an accurate and prompt diagnosis is crucial because of its fulminant course, serious associations with Muir–Torre syndrome and high potential for regional and distant metastasis.

Keywords: Sebaceous gland, Sebaceous cell carcinoma, Extraocular

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INTRODUCTION

Sebaceous cell carcinoma is an uncommon, cutaneous tumor first well-described by Allaire in 1891 [1]. Most sebaceous gland carcinomas have no obvious etiology and only a few are associated with Muir–Torre syndrome. This tumor is thought to arise from sebaceous glands in the skin and, thus, may crop up anywhere on the body, where these glands exist, including the genitalia [2]. Despite this relatively high concentration, true neoplasm, i.e., sebaceous cell adenomas and carcinomas are infrequent and very few
cases have been reported in scientific journals [3]. This tumor has a fulminant clinical course, with a considerable propensity for both local recurrence and distant metastasis. Diagnosis and therapy tend to be delayed because sebaceous carcinoma is frequently mistaken for more common benign entities, further complicating treatment of this aggressive malignancy [4]. In addition to its varied clinical appearance, a varied histologic appearance may occur, and delayed diagnosis or misdiagnosis following a biopsy is not uncommon [5]. Ocular region accounts for nearly 75% of all reported cases, as head and neck region of the body has the greatest density of sebaceous glands and its ectopias. Classically, it occurs in females and older population at 6-7th decade, and arises in ocular region from the meibomian gland of tarsal plate and upper eyelid [6]. The parotid gland is most common site outside the ocular region, accounting for about 20% of cases [7]. Overall, it is uncommon tumor with orbital sebaceous carcinoma accounting for only 0.2–0.7% of all eyelid tumors [8]. So far only few cases of extra orbital sebaceous carcinoma are reported in literature and none in the postauricular region [7].

**CASE REPORT**

A 70-year-old male, presented in surgery outpatient department with complaint of rapidly growing swelling, on the right postauricular region (Figure 1), since one and half months. Initially, a very small asymptomatic lesion was present for which no medical opinion was sought. The lesion progressively increased in size and for last 20 days patient had developed discharging ulcer over the swelling associated with progressive tinnitus and hoarseness of voice. His past history was unremarkable and there was no family history of similar lesion. On local examination, a moist greyish pink ulcerated growth of 3x4 cm was present behind his right pinna with features suggestive of facial nerve paresis. The lesion was non tender, fixed to overlying skin and underlying tissue. No regional lymphadenopathy was present. ENT examination and systemic examination was unremarkable. His routine investigation on blood and urine were within normal limits. Further his radiological investigations, i.e. X-ray skull, X-ray chest and USG whole abdomen too revealed normal study.

Fine needle aspiration (FNA) of swelling was advised which suggested the diagnosis as sebaceous cell carcinoma and excision biopsy was performed. Biopsy tissue on histo-pathological examination of cut surface revealed yellowish-grey tumor with area of necrosis and hemorrhages (Figure 2). Microscopic examination (Figures 3 and 4) exhibited lobules of tumor cells separated by fibrovascular stroma. The nuclei showed pleomorphism, hyperchromasia and cytoplasm of the cell was characteristically finely vacuolated to foamy. Some areas showed focal necrosis with comedo-like pattern (Figure 3-Inset), while other areas displayed several mitotic figures, globules and gland like structure formation. The distinctive microscopic findings confirmed the diagnosis of sebaceous cell carcinoma and as the extent of the lesion precluded further surgical treatment, radiotherapy was advised for palliation.
Sebaceous carcinoma is traditionally classified into two groups: tumor arising from the ocular adenaxa, particularly the meibomian glands and glands of Zeiss, and those arising in extra ocular sites. Extraocular sebaceous carcinoma most commonly involve the head and neck region, the parotid and submandibular glands, the external auditory canal, the trunk and upper extremity, sole, the dorsum of the great toe, and laryngeal or pharyngeal cavities. The sex distribution of extra-orbital sebaceous carcinoma appears to be about equal for male and female patients and the mean age of occurrence is 63 years [9].

The disease exhibits such a variety of clinical presentations and histological growth patterns that the diagnosis is often delayed for months to years. The clinical appearance of extraocular sebaceous carcinoma is not pathognomonic, but the lesion may be a pink to red-yellow nodule. Extraocular sebaceous carcinoma, in association with nevus sebaceous, in the postauricular region or in external auditory canal is very rarely described in literature [10]. Although extraocular sebaceous carcinoma, compared to orbital sebaceous carcinoma, is generally considered less aggressive, visceral metastasis has been reported [11]. Draining lymph nodes may be involved in few cases. The possibility of MTS must be considered in every case of sebaceous tumor. Criteria for diagnosis of Muir–Torre syndrome include the presence of at least one sebaceous adenoma, sebaceous epithelioma, or sebaceous carcinoma and at least one visceral cancer [12].

In our patient there was neither any regional lymphadenopathy nor any evidence of other internal malignancies as associated with Muir–Torre syndrome. However, immunohistochemistry, an important aid to diagnosis of Muir–Torre syndrome, could not be performed on the tumor to look for expression of mismatch repair genes because of local non-availability and financial constraints. Metastases have been reported to occur as late as five years after the initial diagnosis, lending support to the continual surveillance of patients with sebaceous carcinoma [9].

Histologically, sebaceous carcinomas are often poorly differentiated neoplasms mainly within the dermis. Multiple lobules of basaloid undifferentiated cells are present within the dermis. In the central portion of lobules, more mature cells are present. Marked nuclear atypia, pleomorphism and mitosis are common [13]. This neoplasm may be confused with tumors composed of basal cells, squamous cells (mucoepidermoid and spindle cell carcinoma), clear and balloon nevus cells as well as other sebaceous neoplasm [14]. Histochemically, the clear cells of sebaceous carcinomas are negative with periodic acid-Schiff and Alcian blue staining. Immunohistochemically, the tumor cells of sebaceous carcinomas show positive reactions for EMA and Ber-EP4, in contrast to squamous cell carcinoma which are negative for Ber-EP4. Human milk fat globules subclass 1 and 2 (HMFG1 and HMFG2) are positive in basal cell carcinoma which helps it to distinguish it from sebaceous cell carcinoma in which it is usually negative [15].

**DISCUSSION**

Sebaceous gland carcinoma is an aggressive, uncommon, cutaneous tumor first well described by Allaire in 1891 [1]. But this disease was not firmly accepted until 1956 when Straatsma thoroughly studied the histological and clinical presentation of this disease. This tumor is thought to arise from sebaceous gland in the skin and thus, may arise anywhere on the body where these glands exist [1]. It may appear on the top of pre-existing dermatosis, such as nevus sebaceous and actinic keratosis or may follow radiation therapy for other diseases. It may also occur in Muir–Torre syndrome, characterized by occurrence of sebaceous tumors in association with visceral malignancies.
CONCLUSION

We conclude that sebaceous carcinoma is a rare tumor and extraorbital sebaceous cell carcinoma is an aggressive and invasive malignancy. It clinically mimics other diseases and is difficult to diagnose. Further, correct preoperative diagnosis of sebaceous cell carcinoma is rarely made and its malignant potential is usually underestimated by the surgeon. Hence an accurate and prompt diagnosis is crucial because of its fulminating course, serious associations with Muir–Torre syndrome and high potential for regional and distant metastasis.

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Author Contributions
Suresh K Bhatia – Acquired the data, Revised it critically for important intellectual content, Final approval of the version to be published
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REFERENCES

Primary mucinous adenocarcinoma of the parotid gland: 
A case report

Ali Dehghani Nazhvani, Reza Tabrizi, Sara Amanpour

ABSTRACT

Introduction: Primary colloid carcinoma or mucinous adenocarcinoma (MAC) of the salivary glands, is an extremely rare neoplasm. Only six cases of major salivary glands origin have been occurred. Case Report: A case of MAC of parotid gland in a 55-year-old male and its diagnostic approach is given here. Conclusion: MAC is a high grade malignancy with a significant risk of recurrence and metastasis that need special treatment considerations.

Keywords: Mucinous adenocarcinoma, Colloid carcinoma, Parotid, Cytokeratin 7 (CK7)

Introduction

Primary colloid carcinoma or mucinous adenocarcinoma of the salivary glands, is an extremely rare neoplasm. The WHO defines mucinous adenocarcinoma as a malignant tumor composed of epithelial clusters with large pools of extracellular mucin [1]. Only 21 cases with major or minor salivary gland involvement have been reported to date [2, 3]. Only three cases in the parotid gland, two in the submandibular gland and one in the sublingual gland have been occurred [1, 4]. Mucinous adenocarcinoma of minor salivary glands belongs to a high-grade category with a significant risk of local recurrence, lymph node metastasis and fatal outcome.

We described the clinicopathologic findings of a MAC case arising in parotid gland. Our goal is to comprehend the profile of MAC in the salivary glands and the approaches leading to definite diagnosis.

CASE REPORT

A 55-year-old man was admitted to our clinic with complaint of a painless swelling in his left side of the face, parotid area, for 18 months. The patient's medical history was unremarkable. A complete physical examination revealed no obvious systemic problems.

The preliminary diagnosis was tumor of salivary glands. The mass was excised surgically beneath the parotid capsule and on gross examination, the specimen measured 5×3×1.5 cm and appeared to be multilobular with areas of gelatinous consistency. Samples of the tumor were embedded in paraffin. Sections were cut and stained with hematoxylin and eosin.

Microscopically, the tumor was composed of circumscribed large pools of extracellular mucin in direct contact with the stroma. The mucin lakes surround small cluster and nests of malignant epithelial cells with bland to pleomorphic and hyperchromic nuclei. The nests were solid or formed secondary lumens. A few cells contained small amounts of intracellular mucin, but signet-ring cells were not observed. Mitotic figures
were sparse (Figure 1). Colloid component was more than 50% and no cystic spaces was seen.

Periodic acid-Schiff (PAS) and mucicarmin staining confirmed its mucinous origin (Figure 2). Small sections of serous salivary gland tissue was observed in close relationship to tumor.

Complete clinical work-up and whole body scan was done which showed no trace of metastasis. Immunohistochemical staining for cytokeratin 7 (CK7) and 20 revealed only CK7 positivity which ruled out the metastasis of other malignancies and confirmed the salivary gland origin of the lesion (Figure 3A–B).

These findings identified the lesion as a primary mucinous adenocarcinoma of the parotid gland. Postoperative radiation administered to prevent future recurrence and regional metastasis.

**DISCUSSION**

The real frequency of MAC remains unknown; it comprises less than 0.1% of epithelial salivary gland tumors in Armed Forces Institute of Pathology (AFIP) files [5]. In chinese population, MAC represented 0.1% of all salivary gland tumors and 0.4% of carcinomas. Its incidence was also estimated to be 0.07% of malignant salivary gland tumors in West China [6]. Recent study of minor salivary gland tumors from the United States indicated that MAC accounted for 0.2% of all tumors and 0.4% of malignancies [5]. MAC is more common in the intraoral minor salivary glands, with an approximately 2:1 predilection for the minor over major glands. It occurred primarily in the palate, followed by cheek, floor of the mouth and base of the tongue. MAC can also arise in the stomach, lower gastrointestinal tract and lacrimal glands [4].

Patient ages have ranged from 42 to 86 years (mean 66.6 years), and the tumor was slightly more common in males, with a male to female ratio of 10:8. The most frequent presenting symptom was a painless mass of four weeks to 11 years duration. The histopathologic feature of this tumor is nearly identical to mucinous eccrine carcinoma of the skin, mucinous carcinoma of the breast and colloid carcinoma of the intestine [2, 5]. Alternatively, definitive diagnosis of MAC as a primary salivary gland tumor is achieved by exclusion of metastatic diseases [5, 7]. Distinction between primary MAC from metastatic tumor is impossible, based on histologic features, alone. The immunotyping of CK7/CK20 can aid to determine the tumor origin beside careful clinical history and evaluation. CK7(+) / CK20(-) phenotype may show a salivary primary tumor, whereas CK7(-) / CK20(+) profile may be a clue to an intestinal origin [8].

Most literature on mucinous adenocarcinoma of the salivary glands is in the form of single case reports [3]. The differential diagnostic consideration consist of mucinous cystadenocarcinoma (MAC), mucopidermoid carcinoma (MEC), mucin-rich salivary duct carcinoma (SDC), signet–ring cell adenocarcinoma, metastatic tumors and mucin extravasation phenomenon (MEP) [4]. MCAC has prominent cystic spaces with malignant epithelial lining. An MEC has typical areas of squamous, intermediate and mucin–secreting cells. An MEP shows inflammatory

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**Figure 1:** Colloid carcinoma composed of Nests of malignant epithelial cells floating in a mucin pool forming secondary lumens. The cells have pleomorph and hyperchrome nuclei (H&E staining, 200X).

**Figure 2:** Malignant epithelial nest with a large lumen. Mucicarmine staining confirmed mucinous origin of the pool’s material surrounding the nest (400X).

**Figure 3:** Immunophenotyping shows cytokeratin 7 cytoplasmic positivity in malignant epithelial cells (a, 200X) and cytokeratin 20 negativity (b, 400X) which suggests salivary origin of the tumor.
changes and fibrosis with no evidence of neoplastic epithelium. Quantities of mucinous component in others are not as extensive as in MAC [4].

As mucinous adenocarcinoma of the salivary glands is a rare entity, it is difficult to compare the behavior of them to a similar tumor in a different location. Tumors of the breast and stomach are associated with a better prognosis, whereas those of the colorectum are worse.
The prognosis of salivary gland tumors varies with location, histological type and grade. The clinical stage of the disease is considered the most prognostic factor [9].

Gnepp stated that primary colloid carcinoma of the salivary glands is an aggressive tumor with a significant risk of recurrence and metastasis therefore should be considered as a high-grade carcinoma which needs surgical excision with free margins, cervical lymph node dissection and adjuvant radiotherapy depending on tumor stage [4].

CONCLUSION

Mucinous adenocarcinoma of the salivary glands is a rare entity, it is a high grade malignancy with a significant risk of recurrence and metastasis that need special treatment considerations. So one should make distinction between primary and metastatic tumors by immunotyping.

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Author Contributions
Ali Dehghani Nazhvani – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Reza Tabrizi – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sara Amanpour – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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REFERENCES
A case of simultaneously developed sweet’s syndrome and systemic lupus erythematosus in a man

Hiroyuki Hounoki, Maiko Okumura, Koichiro Shinoda, Reina Ogawa, Hirofumi Taki, Kazuyuki Tobe, Kyoko Shimizu, Teruhiko Makino, Tadamichi Shimizu

ABSTRACT

Introduction: Sweet's syndrome is characterized by the development of tender, erythematous plaques, fever, arthralgia and leukocytosis. Sweet's syndrome is reported to be associated with underlying infections, malignant neoplasms, pregnancy, drugs, and autoimmune diseases. However, Sweet's syndrome associated with systemic lupus erythematosus has been rarely reported. Although both Sweet's syndrome and systemic lupus erythematosus predominantly occur in women, we are reporting a case of simultaneously developed Sweet's syndrome and systemic lupus erythematosus in a man. Case Report: A 48-year-old previously healthy man was admitted for intermittent fevers, arthralgias, and eruption. Laboratory investigations revealed elevated C-reactive protein, lymphocytopenia, and low complements with elevated immune complex. Antinuclear antibodies, anti-DNA antibodies were positive. Systemic lupus erythematosus was diagnosed based on the presence of arthritis, lymphopenia, anti-DNA antibodies and antinuclear antibodies. A biopsy specimen from an erythematous papule from the right sole showed a slight lichenification and an infiltration of neutrophils and lymphocytes in dermis without leukocytoclastic vasculitis. Thus, this case also met the modified criteria for Sweet's syndrome proposed by von den Driesch. Prednisolone was begun at a dose of 20 mg/day, resulting in diminishing skin eruption and resolution of arthralgia. Conclusion: Sweet's syndrome is categorized into classical, drug-induced, and malignancy-associated form. Only one case of concurrence of classical Sweet's syndrome and systemic lupus erythematosus has been reported in a man. This is the second case of coincidence of classical Sweet's syndrome and systemic lupus erythematosus in an adult man.

Keywords: Sweet's syndrome, Systemic lupus erythematosus, Erythematous plaques, Arthralgia

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INTRODUCTION

Sweet’s syndrome, or acute febrile neutrophilic dermatosis, was first described by Sweet RD in 1964 [1]. This syndrome is characterized by the development of tender, erythematous plaques, fever, arthralgia and leukocytosis [2, 3]. The syndrome is reported to be associated with underlying infections, malignant
neoplasms, pregnancy and drugs [2, 3]. In addition, Sweet’s syndrome is also associated with autoimmune diseases [2, 3]. However, Sweet’s syndrome associated with systemic lupus erythematosus (SLE) has been rarely reported [4–6]. Here, a case of simultaneously developed Sweet’s syndrome and SLE in a man is reported.

CASE REPORT

A 48-year-old previously healthy man was admitted for intermittent fevers, arthralgias, and eruptions. He had never taken any medications before occurrence of the symptoms. Laboratory investigations were C-reactive protein 2.31 mg/dL (normal range: <0.3 mg/dL), white blood cells 4300/mm³ with lymphocytes 830/mm³, CH50 18 U/mL (normal range: 30–46 U/mL), C3 37.0 mg/dL (normal range: 71–129 mg/dL), C4 5.7 mg/dL (normal range: 12–34 mg/dL) and immune complex 165.0 μg/mL (normal range: <3.0 μg/mL). Viral markers for hepatitis were negative. Antinuclear antibodies (ANA), anti-DNA antibodies were positive at 1:40 speckled pattern and 19 U/mL (normal range: <10 U/mL), respectively. Anti-SS-A antibodies, anti-SS-B antibodies, antineutrophil cytoplasmic antibodies against proteinase 3 and myeloperoxidase, anti-RNP antibodies, anti-Sm antibodies, anti-Jo-1 antibodies, anti-cardiolipin antibodies and lupus anticoagulant were all negative. Screening for malignancy by means of gallium scintigraphy, chest and abdominal computed tomography, and gastrointestinal fiberscope indicated no evidence of malignancy. Leukocytosis, which is a predominant feature of Sweet’s syndrome, was not always observed in this case [1]. Rather, lymphopenia in addition to positive ANA and anti-DNA antibodies with hypocomplementemia and high levels of immune complex in the serum were observed in this case. SLE was diagnosed based on the presence of arthritis, lymphopenia, ANA and anti-DNA antibodies according to the revised classification criteria of SLE.

A biopsy specimen from an erythematous papule from the right sole (Figure 1) showed a slight lichenification and an infiltration of neutrophils and lymphocytes in dermis without leukocytoclastic vasculitis (Figure 2A–B). Furthermore, immunofluorescence was positive for complement deposition at dermal-epidermal junction, indication a positive for lupus band test. This finding further helps to confirm the diagnosis of SLE. Oral administration of prednisolone was begun at a dose of 20 mg/day, resulting in diminishing skin eruptions and a resolution of arthralgia. Thus, this case met the modified criteria for Sweet’s syndrome proposed by von den Driesch [1].

DISCUSSION

Sweet’s syndrome is reported to be associated with underlying infections, malignant neoplasms, pregnancy, drugs, and autoimmune diseases [2, 3]. Thus, Sweet’s syndrome is categorized into classical, drug-induced, and malignancy-associated form. We diagnosed this case as a classical Sweet’s syndrome.

The characteristic histopathologic features of Sweet’s syndrome are diffuse infiltrate of predominantly neutrophils with leukocytoclasia, papillary dermal edema, swollen endothelial cells and an absence of leukocytoclastic vasculitis. The pathological findings of this patient were consistent with Sweet’s syndrome. In addition, immunofluorescence of immunoglobulin and complement deposition at the basement membrane of the skin was positive along with serum hypocomplementemia and high immune complex levels, suggesting a coexistence of SLE.

Concurrence of lupus including drug-induced lupus and subacute cutaneous lupus with Sweet’s syndrome had been reported previously in adults [7, 8]. However, only three cases of concurrence of classical Sweet’s syndrome and SLE have been reported, in whom two are women and one is a man [4–6]. Although both classical Sweet’s syndrome and SLE predominantly occur in women, this is the second case of coincidence of classical Sweet’s syndrome and SLE in an adult man.

CONCLUSION

Sweet’s syndrome associated with SLE has been rarely reported. This is a second case of concurrence of
classical Sweet’s syndrome and SLE observed in a male patient.

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Author Contributions
Hiroyuki Hounoki – Acquisition of data, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Maiko Okumura – Acquisition of data, Analysis and interpretation of data, Final approval of the version to be published
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REFERENCES
**Nocardia otitidiscaviarum** pneumonia in an immunocompetent host

Haroon Yousaf, Irfan Saddique

**ABSTRACT**

Introduction: Nocardiosis is typically regarded as an opportunistic infection, but approximately one-third of infected patients are immunocompetent. We herein report a case of immune competent female with pulmonary *N. otitidiscaviarum* infection in whom complete cure was provided with a seven months of antibiotic combination including trimethoprim sulfamethaxazole. Case Report: A rare case of *Nocardia otitidiscaviarum* pneumonia, in an otherwise healthy 44-year-old woman who complained of febrile illness associated with sore throat, dry cough, myalgia and diarrhea. She had never been a smoker. Her clinical symptoms showed a rapid deterioration in the two weeks before admission, despite a course of oral antibiotics. Gram-staining of her sputum taken at the time of his admission showed the presence of gram-positive branching filamentous bacilli. Sputum culture and PCR confirmed the diagnosis of *Nocardia otitidiscaviarum*. The patient was treated with a combination of antibiotics and showed excellent clinical and radiological response one month after treatment initiation. Conclusion: The majority of patients with nocardial infection are immunocompromised, most often with cell-mediated abnormalities. However, nocardiosis should always be considered in the differential diagnosis of indolent pulmonary disease even in immunocompetent patients.

Keywords: *Nocardia otitidiscaviarum*, Pneumonia, PCR

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**INTRODUCTION**

Nocardiosis is an uncommon gram-positive bacterial infection caused by aerobic actinomycetes in the genus *Nocardia*. *Nocardia* spp. have the ability to cause localized or systemic suppurative disease in humans and animals [1–5]. Nocardiosis is typically regarded as an opportunistic infection, but approximately one-third of infected patients are immunocompetent [6]. We herein report a case of immunocompetent female with pulmonary *N. otitidiscaviarum* infection in whom complete cure was provided with a 7 months of antibiotic combination including trimethoprim/sulfamethaxazole.

**CASE REPORT**

A 44-year-old Caucasian woman presented with a five-week history of a febrile illness associated with sore throat, dry cough, myalgia and diarrhea. Her clinical symptoms showed a rapid deterioration during the last two weeks before admission, despite a course of oral antibiotics (macrolide and b-lactam). She was a lifetime
nonsmoker and used to work in an environment with no known exposures to chemicals, fumes, dust and other environmental or occupational allergens.

Medical history of patient consisted of hypertension, for which she was taking hydrochlorothiazide, and several episodes of gout, for which she was taking allopurinol. There were no complaints of loss of weight or appetite. There was no past history of tuberculosis, diabetes mellitus or steroid therapy. Her physical examination revealed that she was obese, had a body temperature of 39.0°C, a regular pulse of 112 beats/minute and blood pressure of 145/90 mm Hg. Her respiratory rate was 20 breaths/minute with oxygen saturation of 94% on room air, which decreased to 89% on ambulation. There was no pallor, icterus, cyanosis, finger clubbing or pedal edema. Her neck was supple. The examination of the lungs revealed bilateral expiratory wheezes 65 and rare rhonchi Cardiac examination demonstrated normal first and second heart sounds with a regular rhythm and no murmurs. His abdomen was soft and nontender, extremities were warm and the skin was dry. Laboratory work up revealed; sodium 142 mmol/L, potassium 4.7 mmol/L, chloride 108 mmol/L, bicarbonate 25 mmol/L, BUN 10 mg/dL, Creatinine 0.9 mg/dL, Glucose 108 mg/dL, White blood cell 12.5×10^9/L, Neutrophils 70%, Hemoglobin 7.1 mmol/L, Hematocrit 33%, Platelets 423×10^9/L and LDH 128 U/L. Results of an HIV screening test were negative, and the patient's CD4+T lymphocyte count was 1080 cells/mm³. Chest X-ray and CT of the chest revealed left lower lobe consolidation and multiple nodular opacities. A direct gram-staining of his sputum taken at the time of his admission showed the presence of gram-positive branching filamentous bacilli (Figure 1). A standard culture and conventional identification of his sputum sample yielded 104 CFU/mL of Nocardia spp. after three days of incubation (Figure 2). Accurate identification at species level was achieved after 16SrDNA amplification and sequencing leading to the discovery of N. otitidiscaviarum. Using the disk diffusion method we found out that the isolate was resistant to rifampicin, erythromycin, gentamycin, doxycycline and vancomycin. Meanwhile, we found it susceptible to amoxicillin, trimethoprim/sulfamethoxazole, imipenem and ciprofloxacin (Figure 3). The patient improved clinically while receiving a three-week course of intravenous ciprofloxacin, amikacin, and trimethoprim sulfamethoxazole. One month later, his leukocyte count decreased to 6.4×10^9/L (50% PMNs) and the culture was negative. Patient was then started on a six-month course of oral trimethoprim and sulfamethoxazole and nebulized amikacin in order to eradicate the bacterium. During a follow-up examination after one year patient denied any fever, cough and his shortness of breath was fully resolved.

**DISCUSSION**

The majority of patients with nocardial infection are immunocompromised, most often with cell-mediated
abnormalities [3, 4]. However, nocardiosis should always be considered in the differential diagnosis of indolent pulmonary disease.

CONCLUSION

Our case illustrates the need for a high index of suspicion of pulmonary nocardiosis even in immunocompetent patients.

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Author Contributions
Haroon Yousaf – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Irfan Saddique – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

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Authors declare no conflict of interest.

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REFERENCES

Application of diode laser for excision of non-inflammatory vascular epulis fissuratum

Amit A Agrawal, Mahendra Mahajan, Aarti Mahajan, Swagat Devhare

ABSTRACT

Introduction: Epulis fissuratum is essentially one of overgrowth of fibrous tissue from vestibular mucosa, that most commonly develops when a full denture or partial denture flange begins to impinge/irritate on the tissues in this area. It must be surgically removed with scalpel, electrosurgery or lasers. As a component of the treatment, the denture must usually be re-made or substantially adjusted to prevent recurrence. Case Report: A fold of fibrous tissue in the anterior left segment of maxillary alveolar ridge with its base in vestibule, in a 65-year-old male patient, was excised using a diode laser. Follow-up was done after 15 days and 1 month and a new complete denture was fabricated for the patient. Conclusion: Based on the results obtained, it can be concluded that, diode laser is an excellent tool for surgical excision of fibrous overgrowths. It also helps in clean field with good vision, less postoperative bleeding or discomfort. Early recovery is an added advantage, since the patient can be delivered denture early and they can resume their regular activities in lesser time.

Keywords: Epulis fissuratum, Denture induced fibrous hyperplasia, Granuloma fissuratum,

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INTRODUCTION

Epulis fissuratum refers to the tissue growth into the oral cavity, located over the alveolar ridges but originating from the soft tissues of the vestibular sulcus. The term itself is old fashioned, but it is so ingrained in literature that we continue to apply it. The condition is essentially one of overgrowth of fibrous tissue that most commonly develops when a full denture or partial denture flange begins to impinge on the tissues in this area. The treatment involves elimination of the causing factors and surgical removal of the lesion, if required. In early stages, when fibrosis is minimal, nonsurgical treatment with a denture in combination with a soft liner is frequently sufficient for reduction or elimination of this tissue. If the causal factor persists, the tissue becomes more fibrous over time, and because this does not respond to nonsurgical treatment, excision is frequently required. The most common techniques are: surgical scalpel, electrical scalpel, carbon dioxide laser,
Erbium: YAG laser, Neodymium: YAG laser, and diode laser. Diode laser is one of the best lasers as an alternative to the surgical scalpel on oral soft tissues. Conventional surgical procedures, such as removal of epulis fissuratum with a scalpel, cause bleeding and postoperative pain, and require sutures and sometimes tissue grafts. In contrast, with diode laser, a dry treatment area is provided, there is minimal pain after surgery, and no sutures are needed.

This report presents a case of massive ‘epulis fissuratum’ lesion excised using diode laser. Follow-up was done after 15 days and one month and a new complete denture was fabricated for the patient. Unlike other reports in literature where CO₂ laser was frequently used, this report further discuss the comparison of diode laser as against CO₂ lasers and Nd:YAG lasers for management of similar lesions.

**CASE REPORT**

A 65-year-old edentulous male patient was referred with a complaint of enlarged mass covering the anterior left segment of maxillary alveolar ridge with its base in vestibule. The non-ulcerated mass in this region was split in center to form two folds. The superficial mass of the fold, towards the labial mucosa was smaller and was approximately 1x1.5x0.7 cm (Figure 1A). The deeper fold, towards the alveolar ridge, was larger and approximately 4x3x0.7 cm (Figure 1B). The color and surface texture of the mass was same as normal tissue. There was no history of any relevant systemic disease. Patient gave a history of using broken denture for last 6 months (Figure 2). Based on the clinical examination, examination of the broken denture and patient’s history a diagnosis of ‘epulis fissuratum’ was made. Then, the patient was planned for excision of the lesion by diode laser followed by a new complete denture.

The patient, assistant and the surgeon himself were protected with laser safety glasses and masks. Under aseptic condition adequate local anesthesia was achieved. The mass in left anterior segment was excised at its base in vestibule using diode laser (840 nm, 2W, pulsed mode). Excised mass was stored in formalin for histopathologic investigation. All tissue tags were removed so that postoperative surgical site is relatively even (Figure 3A). Patient was advised to apply vitamin E gel 2–3 times a day for two days, antibiotic and analgesics were prescribed for three days. Follow-up examination was planned after 15 days and after one month. Since satisfactory healing was achieved after 15 days (Figure 3B), clinical procedures for new complete denture were initiated at the same sitting. Follow up visit after six weeks showed excellent healing (Figure 3C) and patient was happily using the new denture (Figure 4).

**Histopathology**

Hematoxylin and eosin stained section showed underlying connective tissue composed of parallel bundles of collagen intermixed with plenty of dilated capillaries, some of which were large with intravasated RBCs filling the lumen of capillaries (Figure 5). In the deeper portions, aggregates of minor salivary glands

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**Figure 1:** (A) The smaller superficial fold of Epulis fissuratum in maxillary left anterior vestibule, (B) The larger fold of lesion lying towards the alveolar ridge. Note the base of the lesion in originating from vestibular mucosa.

**Figure 2:** Broken complete denture which the patient has been using since six months.

**Figure 3:** (A) Immediate post-operative view of the surgical site. Diode laser (2W, 810 nm, pulsed mode) was used to excise the lesion, (B) 15-day postoperative view of maxillary left surgical site shows satisfactory healing in terms of color and texture, and (C) 6-week postoperative view shows excellent healing of the operated site and the vestibular depth is also maintained adequately.
‘fibrous’ clinically, histologically it is more vascular than the typical fibroma. Hence the term fibrous hyperplasia cannot be applied to all lesions of so called epulis fissuratum. It has also been referred to as ‘granuloma fissuratum’ [3], however, this term was subsequently noted as misnomer, as the major histologic features included epidermal hyperplasia with fibrosis and chronic inflammation; with occasional notation of hyperkeratosis and parakeratosis. Since epulis fissuratum are frequently induced by irritation of broken or ill-fitting denture flange, they occur in vestibule, mostly in the anterior region of the upper or lower jaws and origin of the lesion is from vestibular mucosa. The most significant difference in histologic appearance of this lesion was the presence of multiple dilated blood vessels which was not reported in literature. Hence, instead of a histological basis for nomenclature, the clinical appearance and location of the lesion can be more generalized. In this view, term ‘vestibulum fissuratum’ would be more apt. ‘Vestibulum’ is the latin name for oral vestibule and ‘fissuratum’ means ‘fissure’, which by definition is a natural cleft in a substance of an organ or is a break or slit in tissue.

Advantages of diode laser over conventional surgery includes convenient mucosa removal, excellent hemostasis with a bloodless field, high precision in tissue destruction, no need for sutures, bactericidal properties that minimize the possibility of infection and minimal postoperative pain and edema. Majority of case reports in the literature have used CO₂ laser for excision of such lesions [4], but advantage of using diode over CO₂ laser is a matter of further debate. When compared with the CO₂ lasers, Goharkhay et al. [5] found one characteristic difference from the diode laser, namely that no trend of greater damage to lateral tissues with the constant wave mode at higher power levels can be observed. They also found no charring of bone underlying 0.8 mm thick soft tissue, with continuous wave mode, or with the pulsed mode at an average power of 4.5 W. On the other hand, several authors have reported that the use of CO₂ laser can result in possible damage to the underlying bone around teeth when cutting tissues with either pulsed or continuous wave CO₂ lasers [6]. Contrary to other investigations [7], deeper incisions could be achieved with the diode laser than were achieved by other authors with the CO₂ or Nd:YAG laser at the same power setting, even with fewer movements of the delivery system. Even the horizontal and vertical zones of thermal damage are in comparable range [5]. When compared with Nd:YAG lasers, the radiation of a diode laser shows a greater absorption and a smaller penetration depth than that of a Nd:YAG laser, especially in blood-rich tissue. The wavelength of the diode laser is considerably more absorbed due to hemoglobin than that of the Nd:YAG laser. This causes not only a better incision performance but also an excellent coagulation of tissues. The thickness of the charring layer and the coagulation layer, and incision depth, are similar for the diode laser and the Nd:YAG laser with the same laser setting [8].

which were mucous in nature and salivary ducts were also evident. Histopathologic features were suggestive of non-neoplastic, non-inflammatory vascular lesion.

**DISCUSSION**

Ill-fitting dentures are often used by the patients without any major complaints. Overtime some patients can even manage to use broken denture for years together. Only when some pathology like ‘epulis fissuratum’ arise then patients seeks attention of a dentist. Mere excision of the lesion without eliminating the causative factor would definitely result in recurrence. This fact should be made very clear to the patient before undertaking surgical excision.

The term epulis, first used by Virchow, that means over the gums, it is not appropriate to these lesions as the affected mucosa is oral mucosa of vestibular sulcus and not gingival mucosa [1]. In this view, some authors prefer to call these lesions denture-induced fibrous hyperplasia [2]. However, even if the appearance is
In the present case no hemorrhagic episodes or infection occurred during postoperative period. With lasers, a coagulum of denatured collagen on the surface is formed and with laser sterilization of wound, the acute inflammation reaction is delayed and minimal. Reduced pain can be attributed to the fact that the inflammatory reaction associated with laser application is reduced, since blood and lymphatic vessel sealing occurs, with prevention of the extravasation of fluids responsible for inflammation and pain. Moreover, laser irradiation cause sealing of the nerve endings in the surgical contact area and the denaturalized collagen layer formed on the surface of the surgical wound serves to isolate from the oral fluids.

Pogrel [9] has reported a decrease in vestibular depth with conventional epulis fissuratum surgery when the wound is closed with sutures. In addition, laser wounds have been reported to contain fewer myofibroblasts, which are responsible for lesser wound contraction [10]. This can explain the excellent postoperative vestibular depth in the present case.

CONCLUSION

Diode laser is an excellent tool for surgical excision of vestibulum fissuratum. It also helps in clean field with good vision, less postoperative bleeding or discomfort and lesser wound contraction. Early recovery is an added advantage with lasers; since the patient can be resume their regular activities with the new/ altered prosthesis in comparatively less time than conventional surgery.

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Author Contributions

Amit A Agrawal – 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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REFERENCES

Early cutaneous metastasis from colonic adenocarcinoma

James W F Kynaston, Victoria L Reid

CASE REPORT

A 74-year-old female presented acutely to the surgical department with two rapidly growing, painful skin lumps. Her medical history included an emergency right hemicolectomy for perforated transverse colon secondary to a Dukes’ C1 T4 N1 M0 poorly differentiated colonic adenocarcinoma one month previously. Physical examination revealed a tender 3 cm cervical nodule (Figure 1) and a 2 cm nodule in left iliac fossa at the previous drain site (Figure 2). Laboratory testing revealed a mild normocytic anaemia (hemoglobin 10.4 g/L) and acutely deranged liver function tests. Computed tomography scan of the neck, chest, abdomen and pelvis confirmed a 3x3 cm necrotic left supraclavicular lymph node, 2.8x1.9 cm abdominal wall nodule in the left iliac fossa, and mild ascites. Fine needle aspiration cytology established a diagnosis of poorly differentiated metastatic adenocarcinoma. This patient was not a candidate for chemotherapy and she was treated with palliative care.

DISCUSSION

Cutaneous metastasis from colonic cancer is rare with an incidence of around 4% [1] and only a few cases have been reported [2–4]. Cutaneous metastasis may present at a variety of sites such as the abdominal wall,

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Figure 1: A three cm cervical nodule.

Figure 2: A two cm nodule in left iliac fossa at the previous drain site.
particularly in scar sites, or less commonly as the first sign of an underlying unknown malignancy such as breast or lung cancer. Uniquely our case highlights a rapid occurrence of cutaneous metastasis with the involvement of multiple sites in a treated patient with colonic adenocarcinoma.

Only one lymph node was found to be involved in the resected specimen, and revealed an advanced tumor but a preoperative CT scan was not suggestive of distant metastasis. It is unusual for such advanced lymphatic spread and cutaneous metastasis without CT evidence of liver metastasis [3]. It has been stated that cutaneous metastasis from colorectal cancer usually takes years to develop [3]. In our case the patient had only undergone a right hemicolectomy one month earlier with no evidence of metastasis at that time. Subsequently she presented with not only a metastatic nodule at the previous drain site but also a large rapidly evolving nodule in her neck.

Cutaneous metastases can present as nodules, ulcers, or fibrous deposits and have varied histological classification depending on their type. In this case, the investigation of choice was fine needle aspiration cytology, which provided an accurate diagnosis and can prove vital in cases where the primary tumor is unknown. Cutaneous metastasis is associated with advanced disease and although the basis of treatment is treating the primary tumor the majority of patients are treated with palliative care. With local symptoms there is a role for radiotherapy or surgical resection/debridement. Median survival in those patients with cutaneous metastasis from colonic adenocarcinoma has been reported at an optimistic 18 months [5] but some reported figures are as low as 3.3 months [4].

CONCLUSION

This case of rapidly developing metastasis highlights the unpredictable nature and presentation of colonic adenocarcinoma. Advanced tumor staging should raise the suspicion of metastasis despite negative CT findings. In such cases care must be taken to make an accurate diagnosis and provide the most appropriate and sensitive treatment for the patient.

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Victoria L Reid – Conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Guarantor

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REFERENCES

MRI appearances of a destructive giant cell tumor and aneurysmal bone cysts of the lumbosacral spine

Muhammad Yunus Amran, Jumraini Tammasse

CASE REPORT

We report a case of a 35-year-old man who was hospitalized on January 23, 2008 who complained of experiencing weakness on both legs for six months prior to his hospital admission. The weakness started on his left leg after he played badminton and lasted for three months. Two weeks later, he felt weakness in his right leg. He also felt cramps and pain on his lower back, and it spread down to the lower leg. The pain was aggravated by coughing and sneezing. He could not normally urinate and defecate. There was no history of trauma, fever, chronic cough, diabetes and cardiac disease. Based on neurologic physical examination, movement on the lower limb had decreased. Additionally, pathological reflex at the lower limb, the muscle tone and the physiological reflex decreased as well. Sensory functions were anesthetized from acral until S1 dermatome and hypoesthesia from the S1 dermatome until L3 dermatome in both sides. Routine and blood chemistry analyses revealed the following results: white blood cell 8.43×10⁹/mm³, red blood cell 4.21×10¹²/mm³, hemoglobin 12.7 g/dL, hematocrit 38%; platelet 310×10⁹/mm³, blood sedimentation rate 1 hr – 11 mm and 2 hr – 30 mm (normal value for males below 10 mm/¹ hr glucose (random) 95 mg/dL, total cholesterol 194 mg/dL, HDL 46 mg/dL, LDL 70 mg/dL, triglyceride 146 mg/dL, uric acid 4.6 mg/dL, urea 29 mg/dL, creatinine 0.73 mg/dL, SGOT 15 mg/dL, SGPT 11 mg/dL, alphafetoprotein 1.59 mg/mL (Normal value <12.5 mg/mL), anti TB IgG - negative and prostate-specific antigen (PSA) 2.73 mg/mL (normal value <4 mg/mL). The perspiration test showed no color change from acral up to the knee (spinal cord dermatome L3). An anterior posterior (AP)/lateral view of lumbosacral X-ray showed that alignment of lumbosacral vertebrae was changed; destruction of posterior part of lumbar 4–5 vertebrae corpus and destruction of right lumbar 4 pedicles and bilateral lumbar 5 pedicles; narrowing in the vertebral discus of L4–5; bone mineralization in normal range and soft tissue was normal limit suggesting metastatic tumor to the vertebral corpus.

An magnetic resonance imaging (MRI) examination was performed with and without administration of intravenous Gadolinium (Gd-DOTA). The result revealed an extradural mass with mix-intensity (iso, hypo and hyperintense) in sagittal T1-weighted images of the lumbosacral region (Figure 1) and extradural mass with mix-intensity (iso and hyperintense) in sagittal T2-weighted images of the lumbosacral region (Figure 2). Sequential sagittal T2-weighted images FatSat with MR-myelography revealed a clear border, lobulated side with the solid component of the mass, cyst containing blood (level fluid appearance) in the large areas that were spreading to the anterior and posterior regions and also damaging the surrounding bones, and compressing thecal sac and nerve roots bilaterally, that cause stenosis canalis spinalis at that level (Figure 3). These findings suggested destructive giant-cell tumor and aneurysmal bone cysts in the lumbosacral spine of vertebrae corpus L₃–S₂ level. The patient was recommended neurosurgical operation. The tumor was biopsied and revealed the histopathology as carcinoma metastases at the vertebrae (it was very
Figure 1: Sagittal T1-weighted image of the lumbosacral region without (A) and with (B) intravenous Gd-DOTA contrast showing degenerative disc disease, lumbar and extradural mass which was mix-intensity (iso, lipo and hyperintense) without Gd-DOTA contrast and became hyperintense after injecting Gd-DOTA contrast intravenously.

Figure 2: Sagittal T2-weighted image of the lumbosacral region showing, degenerative disc disease, lumbar and extradural mass with mix-intensity (iso and hyperintense) signal.

Figure 3: Sequential sagittal T2-weighted image Fat-Sat with MR-Myelography. (A) Right parasagittal T2WI, (B) Right parasagittal T2WI, (C) Sagittal T2WI, all of them showing a mass at the level of vertebrae corpus L3-S2 with clear border, lobulated side with the solid component, cyst containing blood (level fluid appearance) in the large area that spread to anterior and posterior aspects and also destroying the surrounding bones and compressing the thecal sac and nerve roots bilateraly that cause stenosis canalis spinalis at that level.

difficult to determine the primer. Patient himself wanted to be discharged from the hospital after eleventh day of operation.

DISCUSSION

Spinal tumors which can affect the spinal column consist of primary and metastatic tumors. The primary vertebral tumors of the spine are about 11% of all primary tumors of musculoskeletal, and only 4.2% of all spinal tumors, including aneurysmal bone cysts (ABC) and giant cell tumors (GCT) [1, 2]. Aneurysmal bone cyst (ABC) was first reported by Jaffe and Lichtenstein in 1942 [3]. They described it as 'blood-filled sponges' of cavernous cysts with walls of woven bone. This tumor may be found in association with other tumors such as giant cell tumors or as a secondary feature in a variety of osseous lesions, including giant cell tumors [4, 5]. In our case report, we showed that the patient complaining weakness of both of his legs was diagnosed with a giant cell tumor and aneurysmal bone cysts of the lumbosacral spine along with primary tumor. The diagnosis was based not only from the MRI but also from all the examinations, including blood examination to identify primary tumor of giant cell tumors and aneurysmal bone cysts of the lumbosacral spine.

CONCLUSION

Giant cell tumors and aneurysmal bone cysts of the lumbosacral spine are rare tumors. Radiography can be
used to diagnose spinal tumors, but CT scan and MRI is more useful and needed to determine the extent of the tumor. Specifically, in the case of imaging diagnosis, MRI is the gold standard for diagnosing this tumor.

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Author Contributions
Muhammad Yunus Amran – 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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REFERENCES

CASE REPORT

A 61-year-old male with a history of peptic ulcer disease and prostatectomy presented to the emergency department with massive lower gastrointestinal bleeding, melena and diffuse abdominal pain. The patient had a history of lower gastrointestinal bleeding associated with diffuse abdominal pain and changes in bowel movements, alternating between diarrhea and constipation. Colonoscopy (Figure 1, 2) and computed tomography (CT) scan (Figure 3) revealed numerous submucosal lipomatous lesions in several bowel segments with the largest measuring approximately 7.2 cm, found in the transverse colon. Pre- and post-contrast CT scan showed lesions with radiodensity consistent with fatty tissue in the lumen of the transverse colon (arrow) (Figure 3). CT scan and colonoscopy showed no bowel obstruction or intussusception. We were unable to perform biopsy of the lesion due to risk of perforation. The patient was kept on watchful waiting and showed improvement in signs and symptoms during hospitalization.

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DISCUSSION

The lipomas are lesions formed by fat cells, surrounded by a fibrous capsule from which septa, penetrate the substance of the lipoma. It is believed that alcoholism is a risk factor, but there is insufficient
evidence to prove this [1]. According to the existing literature, the diagnostic methods of choice for colonic lipoma are colonoscopy and CT scan [5, 7]. As this is a rare and often asymptomatic condition, our case was unusual in that the patient presented with symptoms suggestive of neoplastic disease, which should be included in the differential diagnosis [1, 2, 4, 8]. There have been few reports of this condition in the literature [1–8]. Most patients are female and the small bowel is most often affected [3, 9, 10]. In our case there was no expulsion of lipoma which is quite common according to some authors [10]. We choose CT scan for diagnosis as it can easily differentiate for density from other tissues and other colorectal tumors. Timely diagnosis is essential in order to prevent the main complication of colonic lipoma i.e. intussusception, for which surgical intervention is the treatment of choice [1, 3, 6, 10].

In this case the patient was managed with medical treatment for the symptoms; keeping him hemodynamically stable. We advise the patient that surgery, hemicolectomy or segmentectomy would be necessary if there were signs of total obstruction or intussusception. As the patient refused surgery; we were successful in diagnosing the condition; patient’s symptoms improved on medical management and there were no associated complications; based on our clinical experience and literature references we successfully followed the patient clinically without the need for surgery.

CONCLUSION

Lipomas of colon are rare lesions which may present with signs and symptoms resembling those of neoplastic disorders. Colonoscopy and CT scan are diagnostic methods of choice. In intussusception is the main complication which may necessitate surgery.

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Josué A Victorino – Contributions to analysis and interpretation of data, drafting the case report, revising it critically for important intellectual content, final approval of the version to be published
Richard F Kühne – Substantial contributions to conception and design, acquisition of data, analysis and interpretation of data, drafting the case report, 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.
REFERENCES

Cecal bascule with a mesenteric band acting as a ‘point of basculation’

Jashodeep Datta, Joseph V Sakran

CASE REPORT

A 72-year-old female with a past history of cesarean section presented with a three day history of right-sided abdominal pain and distension. She had a low-grade fever, was normotensive, and without leukocytosis or metabolic acidosis. Her exam revealed moderate abdominal distension with rebound tenderness localized to the right lower quadrant. An abdominal X-ray showed significant cecal distension without small bowel dilatation (Figure 1). Given her worrisome clinical picture, the patient was emergently taken for an exploratory laparotomy. There was anteromedial folding of a massively distended cecum about a mesenteric band traversing the terminal ileum extending to the ascending colon (Figure 2; arrow). This created a closed-loop obstruction of the ascending colon. There was no axial torsion of the cecal mesentery. There was no colonic perforation or feculent spillage noted. After lysis of this band, inspection of the cecum demonstrated that it was not only severely dilated but also tethered in its abnormal anteromedial position by multiple adhesions. These adhesions were lysed and a right hemicolecction with primary anastomosis was performed. The patient had an uneventful postoperative course, with return of bowel function prior to discharge.

Figure 1: Abdominal X-ray showing massively distended and anteromedially positioned cecum without appreciable small bowel dilatation.

DISCUSSION

Although the cecal bascule was first described by Treves in 1899, Weinstein characterized it as a subtype of cecal volvulus in 1938 [1]. Cecal bascule—which in French means ‘sawed’—accounts for only 5–10% of all cecal volvulus cases and has been reported in elderly patients, in postpartum women, and in those with prior abdominal surgery [2, 3]. Unlike the classic volvulus, basculation does not occur via axial torsion of the mesentery. Rather, the cecum folds upon itself about an acquired inflexion point or as a result of congenital hypoxification. This results in a ‘flap-valve’ occlusion
of the ascending colon, impeding anterograde cecal emptying. Retrograde decompression is impossible because of a competent ileocecal valve, and gas production from bacterial metabolism compounds cecal distension [3]. Adhesions form between the anterior cecal wall and the ascending colon, reinforcing the malpositioned cecum anteromedially across the proximal colon. Progressive cecal distension precipitates venous outflow obstruction, bowel ischemia and subsequent perforation. Cecal bascule is under-recognized because it mimics other conditions such as volvulus, Ogilvie syndrome, and cecal obstruction [3]. Lack of awareness of this entity preoperatively may lead to confusion in correlating clinical, radiographic, and operative findings. More importantly, it may delay intervention and increase mortality [4].

AXR is the diagnostic modality of choice and typically demonstrates marked cecal distention. Often, the ‘comma’ or ‘coffee bean’ signs associated with the classic volvulus are not seen [1]. Non-surgical treatment options such as colonoscopic detorsion with minimal air insufflation have been advocated if basculation is detected early. Since only 5% of colonoscopic attempts are successful, the majority of patients will require urgent surgical exploration. While open detorsion with coceopsy or cecal decompression with tube ceccostomy have been described as surgical options, they are infrequently practiced owing to the high rates of recurrence and complications. Detorsion alone is associated with a 30% recurrence rate, while ceccostomy drainage results in tube leakage, colocutaneous fistulization, intra-abdominal infection, and recurrent basculation [2]. Consequently, most cases benefit from right hemicolectomy and primary anastomosis. If colonic perforation or feculent spillage is found at laparotomy, a diverting ileostomy with ileocolonic anastomosis or end ileostomy can be considered. At laparotomy, a constricting band—similar to that seen in our patient—is classically found traversing the ascending colon acting as a point of basculation [5]. These bands are hypothesized to be inflammatory, congenital, or related to prior surgery [3]. Our patient had a history of cesarean section, lending credence to our hypothesis that postoperative changes had contributed to the formation of this band.

CONCLUSION

Cecal bascule is a rare subtype of cecal volvulus. A ‘point of basculation,’ classically a constrictive mesenteric band, causes a distended cecum to fold upon itself resulting in closed loop obstruction of the ascending colon. Prompt diagnosis and urgent intervention can prevent the high rates of morbidity and mortality associated with this condition.

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Datta J, Sakran JV. Cecal Bascule with a mesenteric band acting as a ‘point of basculation.’ International Journal of Case Reports and Images;3(9):54–56.

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Author Contributions
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Esthiomene: An unusual presentation of elephantiasis

Anand Pai, Umadevi V, Narayanasamy S

To the Editors

Genital elephantiasis is an important medical problem in the tropics as it is associated with physical disability and extreme mental anguish especially in females. Genital elephantiasis is a very rare condition and due to prevalence of sexually transmitted infections (STIs) it is even rarer. Elephantiasis and chronic genital ulceration in women is called 'Esthiomene' [1].

A 40-year-old married female, mason by occupation, presented with swelling of the vulva and genital ulcer for the past five years. She complained of difficulty in micturition and difficulty in walking. She also had history of painful swellings in the inguinal region, on both sides, for the past seven years. There was no history of fever, cough with expectoration or weight loss. Her menstrual cycles were regular.

She had two healthy children. She gave a history of her husband’s extramarital contacts. On genital examination two large nodular swellings of size 15x7x5 cm were present involving both the labia majora (Figure 1). Multiple nodules were present on the mons pubis. Multiple superficial and deep ulcers of varying size were present on the inner aspect of both labia majora.

Multiple, firm, non-tender, matted lymph nodes involving both the horizontal and vertical groups of inguinal lymph nodes were found bilaterally. Groove sign was present. Per rectal examination was normal. Systemic examination was normal. Mantoux test was negative. X-ray chest and ultrasound abdomen were normal. Hemogram, LFT and renal parameters were normal. Blood VDRL was non-reactive. Serology for HIV antibodies and filariasis was found to be negative. Smear for malarial parasite and microfilaria was negative. Tissue smear for Donovan bodies was negative. Albumin/Globulin ratio was reversed (0.6). Tissue biopsy of the swelling was taken and histopathological study revealed lymphangiectasia (Figure 2). Culture of discharge in McCoy’s medium showed growth of lymphogranuloma venereum strains. Polymerase chain reaction study showed features specific for Chlamydia trachomatis L1-L3 serovars which confirmed the diagnosis. The patient was started on doxycycline 100 mg twice daily for 15 days and was

Figure 1: Vulval elephantiasis with chronic genital ulcerations
referred to plastic surgery. As no major structures were involved excision of the swelling was done. Postoperatively patient was followed up and had no further complaints.

Among sexually transmitted infections, lymphogranuloma venereum and donovanosis are the most common, others being syphilis and infection with non-LGV strains of Chlamydia trachomatis. Chlamydia trachomatis serovars L1-L3 are the causative agent of LGV and initiates the disease process primarily in the lymph channels leading to thrombolympangitis and perilymphangitis [2]. Extension of inflammatory process to draining lymph nodes causes perianoditis, matting of lymph nodes, formation of abscesses, fistulae and sinuses. Healing takes place by fibrosis. A combination of chronic oedema, sclerosing fibrosis and active lymphogranulomatous infiltration in the subcutaneous tissue results in the massive enlargement of genitalia. These late complications occur in the tertiary stage of LGV and are frequent in women. The diagnosis of LGV associated elephantiasis is supported by high titre serology and identification of the organism in the pus or bubo fluid by cytology or culture [3]. Chlamydia trachomatis is an obligatory intracellular organism and diagnosis is usually made by tissue culture on McCoy’s medium, direct immunofluorescence and polymerase chain reaction. Microimmunofluorescence test is the only serological means of distinguishing LGV strains of Chlamydia trachomatis from other serovars. Polymerase chain reaction amplification and sequence analysis of the omp1 gene is also useful in identification of L1-L3 serovars [4]. History of exposure to infection, a small transitory primary lesion, followed by chronic inguinal adenitis resulting in typical suppuration and fistulation is characteristic of LGV. With the exclusion of tuberculosis, filariasis, Hodgkin’s disease, gonorrhoea, syphilis, malignant disease and chancroid, the diagnosis of the condition becomes at once easy.

Treatment modalities of genital elephantiasis due to STIs require an interdisciplinary approach involving genitourinary medicine physicians, urologists, and dermatologists. The objectives of treatment are to reduce swelling, restore shape and normal sexual function, and prevent inflammatory episodes. Medical therapy should be the first line of treatment and any surgical intervention should be undertaken only under the cover of appropriate antibiotics. In case of genital elephantiasis caused by LGV, doxycycline 100 mg twice daily should be given for prolonged periods (improvement reported up to 13 month therapy). Surgery is the only effective option for a select group of patients in whom the disorder is disabling and persistent. Reduction procedures include labial reduction which is easily achieved by wide elliptical excision with a single suture line [5, 6]. Recurring swelling in a minority of patients will be benefited by another similar procedure. Larger labial defects may be covered with myocutaneous, floating island or fasciocutaneous flaps [7]. However, there are no studies available demonstrating the long-term efficacy of reduction procedures in STI-related genital elephantiasis. Due to the manifold aspects of the disease and of the possibility of the lesions remaining insignificant for a long time, every case of genital elephantiasis or rectal stricture needs to be subjected to a critical investigation in regard to the possibility of this specific infection. Hence we found it worthwhile to report this case.

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Author Contributions

Anand Pai – Conception and design, Acquisition of data, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published

Umadevi V – Conception and design, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published

Narayanasamy S – Analysis and interpretation of data, Critical revision of the article, 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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