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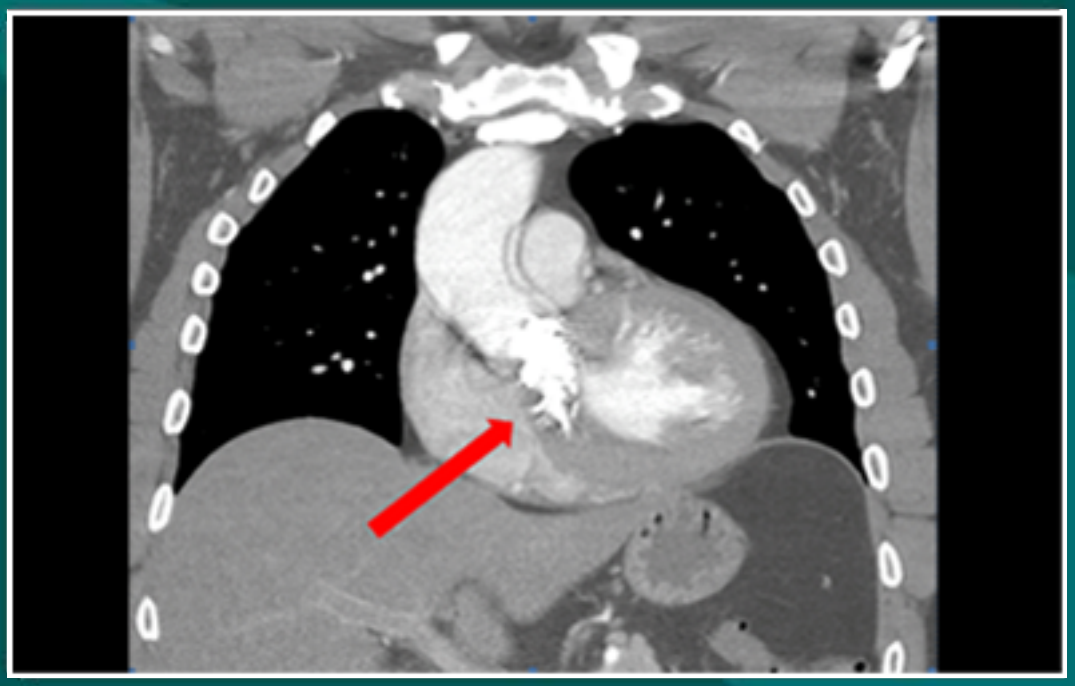

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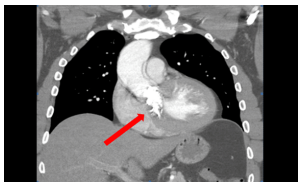

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EDITORIAL OPEN ACCESS

The clinical and scientific mind

Slobodan V. Marinković

The human mind, due to its intelligence, curiosity and creativity, has achieved a tremendous and fascinating progress in medicine, technology and science in general. Such progress, however, is complicated by certain setbacks in both the clinical work and the scientific research.

The most important of these is that the previous unitary disciplines have been divided into several subfields and even smaller subunits, each of them requiring a narrow specialization, specific equipment, methods and procedures, but with a consequent loss of certain connections among them. This usually enables, on one hand, good results and a progress in each subfield but, on the other hand, it made difficult to "see the forest for the trees," that is, to realize the entirety and myriad of interrelationships within a certain discipline. Such loose interconnections in a multidisciplinary subject can sometimes lead to the less reliable results, misdirection, or even a dead end. In addition, new data in each subfield arrive every day which must be incorporated into and processed within the preceding knowledge and skills. This is, obviously, a necessity but, at the same time, it may complicate an already complicated situation.

Some fields of medicine and science are complex per se. For example, a clinical cardiologist must have excellent knowledge of the heart anatomy, embryology, histology, biochemistry, physiology and electophysiology, as well as in hemodynamics, pathology, pathophysiology, and pharmacology, but also in microbiology, immunology, cardiosurgery, vascular diseases, pulmology, nephrology, oncology, radiology (especially in echocardiography, coronarography, CT scan and MRI scan), and in some other disciplines. In addition, cardiologists have immense responsibility for their patients, and are often engaged in research projects. Due to that, some cardiologists, especially the interventional ones who are under a permanent stress, are themselves at a higher risk of suffering myocardial ischemia.

Similarly, a neuroscientist, for example, must be very familiar with the macroscopic and microscopic anatomy of the brain, its biological evolution and embryological development, the structure, ultrastructure and immunohistochemistry of the glia cells, neurons and their synapses, biochemistry and pharmacology of certain neurotransmiters, neuromodulators, cellular receptors and ion channels, electrophysiology, functional principles of the nerve cells and certain neuronal networks, neuropathology, neurology, neuroradiology, endocrinology, psychology, psychiatry, philosophy, etc. In addition, knowledge of electronics, informatics, and statistics is also necessary for successful research.

There are only two solutions for the mentioned problem: an individual's enthusiastic engagement, or perfect team work.

Enthusiasts are completely devoted to their subject of interest. They will try his best to be experts in the entire discipline, regardless of a temporary engagement in this or another subfield, as well as to accept, process and take into consideration a huge number of new information. This often implies a neglect of many other activities and relationships, including his private lives occasionally. On the other hand, they receive a great intellectual reward, sometimes to the level of ecstasy when making a significant scientific discovery, or if managing to cure a large number of certain patients by way of a new medication or therapeutic procedure.

On the other hand, each member of a successful team, albeit engaged in a specific subfield, provides data in an interaction with other experts of the team from various fields and takes them into consideration in his own subject. This permanent exchange of the information,

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knowledge and skills obviously leads to more coherent and valuable results, and to original ideas and methods for future research. Similarly, a medical team consisting of various experts must be formed occasionally to solve a complicated health problem in certain patients.

Nevertheless, whatever choosing to solve problems of multidisciplinarity, a geometrical progress in medicine and science will be continued to an unimaginable level in the future.

Keywords: Creativity, Curiosity, Human mind, Intelligence

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Author Contribution

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

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

A possible new risk factor causing pneumomediastinum in a middle-aged patient: Anatomical weakness in the bronchial wall

Hirofumi Namiki, Kazuhiko Matsuno, Tadashi Kobayashi

ABSTRACT

Introduction: Pneumomediastinum is a rare condition that occurs after physical trauma or other situations that lead to air leakage into the mediastinum. The present report describes a middle-aged patient with pneumomediastinum caused by breath-holding at work. Case Report: A 59-year-old Japanese male presented sudden coughing immediately after breath-holding at work. A chest X-ray was normal, and a computed tomography scan showed pneumomediastinum. The patient had no other predisposing factors for pneumomediastinum, such as trauma, chronic obstructive pulmonary disease or airway infection. Bronchoscopy revealed an anatomical weakness in the bronchial wall. He recovered with two months of conservative treatment. Conclusion: Pneumomediastinum should be considered in middle-aged patients who develop sudden coughing immediately after breath-holding. Anatomical weakness in the bronchial wall should be considered as a possible new risk factor.

Hirofumi Namiki[1], Kazuhiko Matsuno[2], Tadashi Kobayashi[3]

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INTRODUCTION

Previous reports have shown that pneumomediastinum rarely occurs in young male especially children [1]. However, many authors understand that the occurrence of pneumomediastinum may be more frequent than initially believed because many patients refrain from medical visits [2]. In addition, almost patients without any risk factors are diagnosed to spontaneous pneumomediastinum [3, 4]. This report describes a middle-aged patient who developed pneumomediastinum after breath-holding at work. Bronchoscopy revealed a new possible risk factor for pneumomediastinum, an anatomical weakness in the bronchial wall.

CASE REPORT

A 59-year-old male developed sudden-onset coughing at working the evening before his presentation at a clinic. His symptoms appeared immediately after a few seconds when breath-holding for heavy lifting started, and had persisted for approximately 12 hours. He had no other

symptom except for dry cough. His medical history included only hypertension. He was undergoing regular medication (Indapamide 1 mg/day) for hypertension. There was no family history of respiratory disease. He was a not-smoker. Physical examination revealed normal vital signs, and no tenderness or crackles on his neck and chest. All other findings were normal. Blood test and X-ray findings were also normal (Figure 1). For differential diagnosis of pneumothorax, a computed tomography scan of his chest at the clinic was obtained, and pneumomediastinum was revealed (Figure 2). Thus, the patient was diagnosed with pneumomediastinum and received bronchoscopy at a general hospital in another island, which found an anatomical weakness in the bronchial wall (Figure 3). The patient had a conservative treatment with cough medicine (codeine phosphate, 2 g every 8 hours) and daily follow-up. After two months of treatment, his symptoms gradually disappeared.

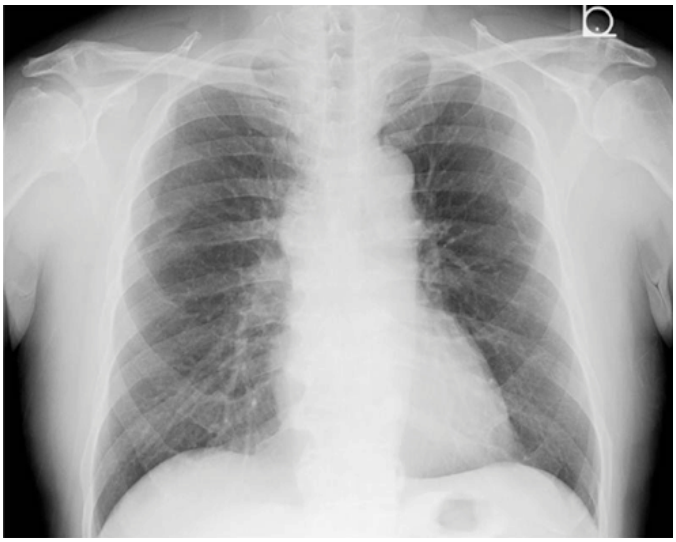

Figure 1: Chest X-ray showing normal findings.

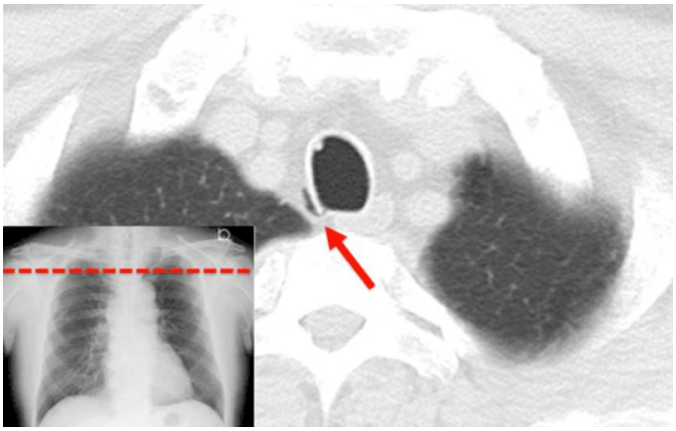

Figure 2: Computed tomography scan of the chest showing pneumomediastinum. Chest computed tomography scan revealed mediastinal emphysema (a red arrow) on the level of red dotted line on chest X-ray.

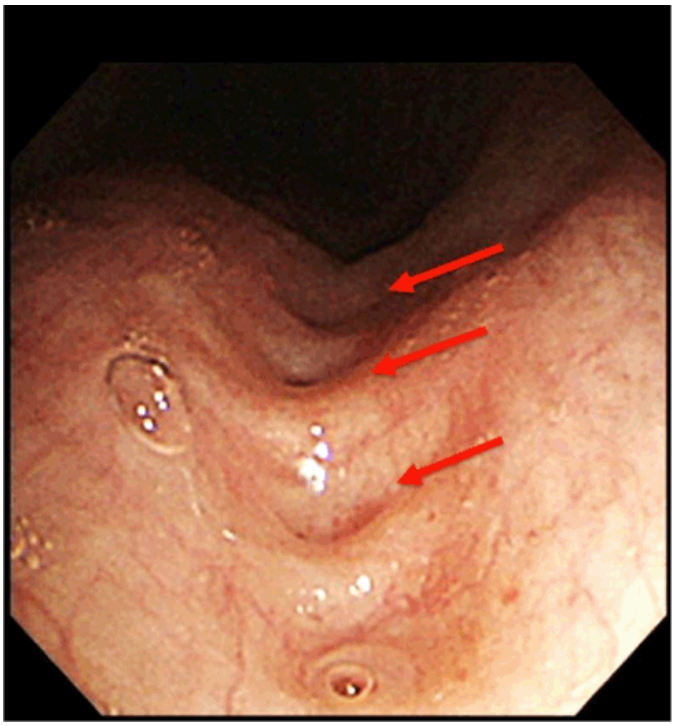

Figure 3: Bronchoscopy showing weakness (red arrows) in the bronchial wall.

DISCUSSION

Investigation of the epidemiology of pneumomediastinum has indicated that pneumomediastinum is a rare event in middle-aged and older patients, but that it occasionally occurs in natural births and patients aged 5–34 years [1]. Most affected patients (76%) are male [5]. In addition, several reports of pneumomediastinum in middle-aged patients reported that affected patients had some risk factors [6]. The development of spontaneous pneumomediastinum in a middle-aged man is rare. We considered that increased pressure on the bronchus secondary to a bronchial anomaly might have caused the pneumomediastinum in our patient. The mediastinal tissues in older patients are fibrosed, making air movement more difficult [7]. However, the increased pressure in the bronchus upon the breath-hold could have caused the air to rupture the bronchial wall that had a cratered surface [8, 9]. Our patient had no other predisposing factors for pneumomediastinum, such as chronic obstructive pulmonary disease or airway infection, post- tuberculosis. Some reports of spontaneous pneumomediastinum have revealed that no inducing factor may be found [3, 4]; however, bronchoscopy was not always performed in these studies. Therefore, in our patient, the potential causes of the pneumomediastinum were incidentally revealed by bronchoscopy and may have been related to the cratered surface of the bronchus.

With respect to therapy, conservative management was chosen in our patient and provided a good

outcome. However, the optimal management of pneumomediastinum depends on its severity and cause. Some patients might require hospitalization for over 24 hours of observation, and others might require drugs for pain, anxiety, or infection. Conservative treatment might be sufficient for patients with pneumomediastinum that causes only mild discomfort. In the present case, we strongly discouraged physical activity and instructed the patient to remain on bed rest and treatment with cough medicine. Spontaneous pneumomediastinum is generally considered to be a benign disease with a good prognosis. Both recurrence of spontaneous pneumomediastinum and prolonged cases (over two months) have been reported [4]. In general, however, few cases of recurrent pneumomediastinum have been reported, highlighting its benign nature [10]. Additional diagnostic evaluation should be conducted in patients with recurrence to detect underlying pathologies such as pulmonary or esophageal pathology. As our patient in middle-aged who had no other predisposing factors underwent bronchoscopy, we had detected anatomical weakness in the bronchus. To the best of our knowledge, no cases of pneumomediastinum associated with bronchial anomalies in middle-aged patients have been reported. Anatomical weakness in the bronchial wall should be considered as possible risk factor for pneumomediastinum in a middle-aged patient, if patient had no common predisposing factors.

CONCLUSION

Pneumomediastinum should be considered in middle-aged patients who develop sudden coughing immediately after breath-holding. Anatomical weakness in the bronchial wall should be considered as a possible new risk factor.

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Author Contributions

Hirofumi Namiki – 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

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

Tadashi Kobayashi – 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

Guarantor

The corresponding author is the guarantor of submission.

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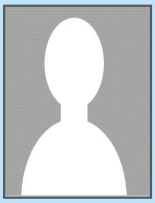

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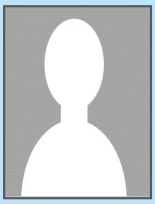

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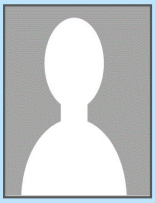

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Condylar osteoma unusual cause of facial asymmetry: A short surgical practice

Saman W. Boskani, Shakhawan M. Ali, Nazar A. Amin, Ali H. Neamat, Payman Kh. Mahmud

ABSTRACT

Introduction: Osteoma is a benign neoplasm resulting from the continuous formation of cortical or cancellous bone. Osteomas of the maxillofacial region mostly occur in the mandible. However, rarely osteomas seen in the mandibular condyle. Case Report: We present a case of a 33-year-old male patient reported with chief complaints of malocclusion, facial asymmetry, difficulty in chewing and deviation of jaw since two years also during childhood he had a history of trauma. Radiographic images and computed tomography suggested benign osteogenic neoplastic lesion involving right condyle which on histopathological examination confirmed it is osteoma. Conclusion: Osteoma should be considered as one of the differential diagnosis in a patient with malocclusion, facial asymmetry, deviation of jaw and difficulty in chewing, especially in a patient with the previous history of trauma.

Keywords: Facial asymmetry, Malocclusion, Mandibular condyle, Osteoma

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INTRODUCTION

Osteoma is benign osteogenic lesion composed of both cortical and cancellous bone that increases in size by continuous formation of bone [1]. The pathogenesis of osteoma is unknown. Some authors consider it a hamartoma, while others consider it a true neoplasm [2]. The first reported case of osteoma of condyle was described by Ivy in 1927 [3]. The reactional mechanism, trauma or infection is also suggested as possible causes [4]. Usually, osteoma located in muscle insertions, suggesting that the muscle pulls acts on the development of the lesion. It is possible that minor traumas, which are not even remembered by the patients, may have caused a sub-periosteal hematoma which, associated with the muscle pull, starts the lesion [4]. Really, it is slow growing, asymptomatic, usually solitary lesion, however, osteomas involving the mandibular condyle may result in morphologic and functional disturbances. There are two types of osteomas:

- Central osteoma, arising from an increase in cancellous bone.
- Peripheral osteoma arising from an increase in cortical bone [5].

Histologically, osteomas have two distinct variants. One is made up of relatively dense compact bone with scarce medullary tissue, while the other has lamellar or cancellous bone trabeculae with abundant medullary spaces of fibrous-adipose tissue. Osteoblastic activity is usually prominent [6]. The traditional radiographic image is usually enough to diagnose an osteoma. It is presented as a radiopaque mass with a density similar to that of a normal bone. The panoramic X-ray, Waters view or CT scan usually shows the location and the benign nature of the lesion [7]. Osteoma treatment is based on complete surgical removal, on the base, where the bone cortical is located. There are no reports of osteoma malignant transformation [8]. Osteomas recurrence is rare also it believed to be relatively uncommon [2].

CASE REPORT

A 33-year-old male patient arrived at the oral and maxillofacial surgery department, with the main complaints of malocclusion and facial asymmetry, he had noted a gradual change in the deviation of the jaw and inability to chew for two years duration. The patient was unable to occlude teeth with painless non-tender swelling on right preauricular region. He had a history of trauma (fall from the bike) during childhood. At the initial examination, extra-orally, asymmetrical facial morphology, the mandible was noted to be deviated to the left, and the median plane of the face was distinctly shifted to the left. On palpation, there was no pain on a temporomandibular joint region and movements of the joint were normal. Intra-oral examination revealed the mandibular central incisors deviated 7–9 mm to the left with respect to the maxillary central incisors (midline was shifted) (Figure 1) but there was no pain associated with mouth opening, and the mandible deviated to the right during the wide opening, the interincisal mouth opening was 35 mm. Based on clinical examination provisional diagnosis given as condylar hyperplasia. Radiographic investigations included OPG, computed tomography 3D and axial view showed a radiopaque osseous mass around the head of the condyle, well-defined pedunculated bony growth was seen on anteromedial aspect of the right condyle. Superiorly extending into left temporomandibular joint space and abutting articular tubercle, which causes dislocation of the condylar head to anterolateral (Figure 2).

Tomography Clinical and radiological findings were suggestive of benign tumor of the condyle: osteochondroma, condylar osteoma, chondroblastoma and osteoid osteoma considered for differential diagnosis.

While the patient under general anesthesia, the tumor was excised. The upper and lower compartments of the temporomandibular joint were accessed by standard preauricular approach with tragal modification for aesthetic purpose, skin prepared and infiltrated with local anesthesia to make bloodless field, high condylar incision done with preservation of meniscus (Figure 3), after wide opening of the mouth space created to approach the mass which was anteromedial to the condylar head, also mass was dissected and separation from the surrounding tissue mass extracted with a small piece of condylar head (Figure 4) and the specimen obtained was sent for histopathological examination. Based on the histopathological examination, diagnosis as osteoma. Postoperative follow-up the patient occlusion and deviation of jaw return normally with good ability to chewing no any facial palsy (Figure 5).

Figure 1: Clinical photography showing preoperative occlusion and facial asymmetry.

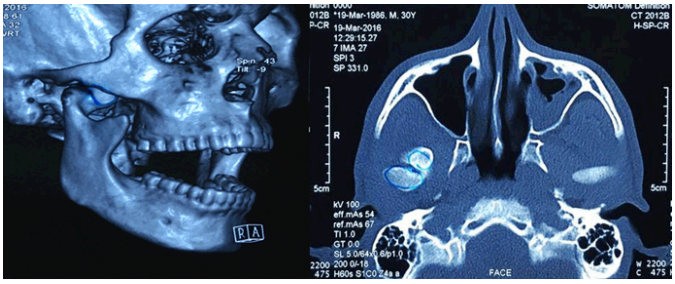

Figure 2: Computed tomography scan (3D and axial section) showing a well-defined pedunculated bony growth was seen on anteromedial aspect of the right condyle.

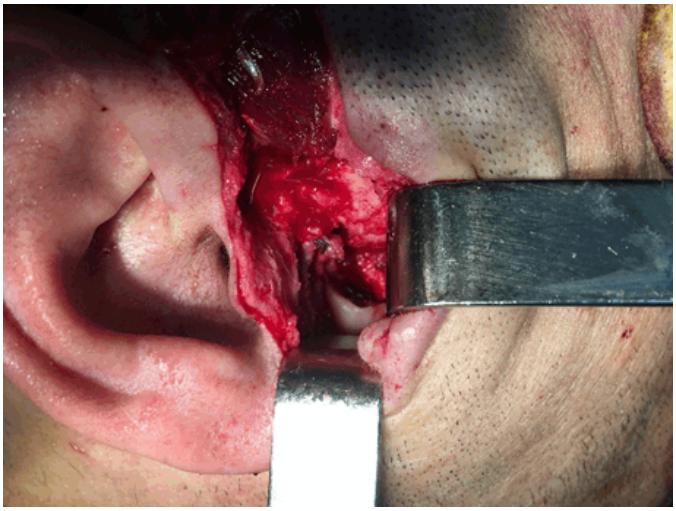

Figure 3: High condylar incision with preservation of meniscus.

Figure 4: Specimen obtained was sent for histopathological examination.

DISCUSSION

Osteoma of the condyle may cause a slow, progressive shift in the patient's occlusion with a deviation of the midline of the chin towards the unaffected side. This results in facial asymmetry and temporomandibular joint dysfunction. The most common clinical manifestations

Figure 5: (A) Postoperative directly (B, C) Occlusion and facial symmetry after three months.

involving the condyle are malocclusion and facial asymmetry [9]. The mandible is the common area for osteoma in the maxillofacial region. There are no sex predilection differences in the incidence of occurrence. Osteomas of the condyle are uncommon. Osteoma was first described by Monsarrat in 1913. The first reported case was described by Ivy in 1927 about condylar osteoma [3]. The etiology of osteomas is unclear. It may be developmental, neoplastic or, most likely, reactive in nature [6]. A combination of trauma and muscle traction, which may initiate an osteogenic reaction, has been suggested as the underlying pathogenesis of osteoma [6]. However, may be the possible precipitating cause in this case.

Radiographic images show osteomas as circumscribed masses similar in density to normal bone. They are a smooth surface with a thin sclerotic rim at the centers, these masses may exhibit a mixed radiolucent-radiopaque appearance depending on the amount of marrow tissues present. Osteomas can be confused with complex odontomas. Smaller endosteal osteomas are difficult to differentiate from foci of condensing osteitis or focal chronic sclerosing osteomyelitis or idiopathic osteosclerosis [10].

According to the pattern of proliferation, condylar process osteomas can be classified into two types:

1. Osteomas that form a pedunculated mass on the condyle [1]
2. Those that proliferate and cause replacement of the condyle by the osteoma.

In the current case, the osteoma is presented a bilobed structure, one lobe presenting as a pedunculated mass and the other lobe is seen as a replacement of the condyle.

Histologically, an osteoma consists of either normal appearing dense mass of lamellar bone with minimal marrow tissue (compact osteoma), or of trabeculae of mature lamellar bone with intervening fatty or fibrous marrow (cancellous osteoma) [11]. A large osteoma resulting in pain, facial asymmetry and malocclusion may require surgical excision (condylectomy) as in the present case, whereas for small, asymptomatic lesions periodic observation is necessary. Recurrence after excision is extremely rare [6]. Up to date, there is only one reported case of recurrence of a periosteal osteoma of the mandible following excision [12]. Furthermore, there are no reports of malignant transformation of osteomas [12].

CONCLUSION

Osteoma of the mandibular condyle is a benign, rare, bony growth that may cause a painless interference in mouth opening and facial asymmetry. Osteoma should be considered as one of the possible etiologies in a patient with facial asymmetry, malocclusion, deviation of jaw & difficulty in chewing, especially in a patient with the previous history of trauma. Surgery with complete lesion removal is an adequate treatment, with low recurrence rates.

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Author Contributions
Saman W. Boskani – Substantial contributions to conception and design, Analysis and interpretation of data, Final approval of the version to be published
Shakhawan M. Ali – 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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Guarantor
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Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Upper airway injury caused by gum elastic bougie

Ayça Tuba Dumanlı Özcan, Cemile Altın Balcı, Şemsi Mustafa Aksoy, Gökçer Uğur, Orhan Kanbak, Togay Müderrris

ABSTRACT

Introduction: Difficulties and complications associated with intubation are among the leading causes of surgery-related mortality in patients with obesity and obstructive sleep apnea. It is known that during perioperative intubations, the progression of the bougie may lead to serious injury and even rupturing in the trachea. Case Report: A 46-year-old ASA II patient was assessed preoperatively for uvuloplasty. His body mass index was 34.7. Preparation was completed for the difficult intubation. The patient could not directly intubated with laryngoscopy but was intubated with bougie in the second trial. After the surgery 200 mg bridion was administered and the patient was extubated. He was then followed-up in PACU. Due to the stridor, it was thought that there was edema in his upper airway. At 45 minutes of PACU follow-up, it was noticed that upper airway edema regressed but there was subcutaneous emphysema giving a sense of rattle during palpation in the periphery of the right eye. It spread rapidly over the face. Afterwards he was intubated again through video laryngoscopy due to the risk of upper airway obstruction. Fiber optic examination and thorax tomography revealed that the fistula line was on the left lateral wall following cricoid cartilage. Mucosal damage of the patient healed spontaneously and weaning was conducted three days later in the ICU. Conclusion: It was reported that tracheal injury and rupture occurs due to 'blind' advancement of the bougie during intubation. Due to the identification of subcutaneous emphysema at 45 minutes during follow-up, it was thought that the bougie caused injury in the patient. The close long-term postoperative follow-up is important in cases where difficult intubation is conducted with bougie and intubation is achieved through multiple trials.

Keywords: Difficult intubation, Gum elastic bougie, Obstructive sleep apnea syndrome, Upper airway injury

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INTRODUCTION

Elastic gum bougies are commonly employed during the intubation of the trachea, particularly in cases

where the glottic opening is difficult to visualize due to obstruction. Previous studies have shown a success rate of more than 94%, when elastic gum bougies are applied using direct laryngoscopy with the aid of a Cormack–Lehane 3 laryngeal view. Intubation can be further facilitated adjusting the shape of the bougie prior to the procedure. It was indicated that use of bougie along with lubricant, advancing it gently and withdrawing it a few centimeters or asking for help to stabilize it while placing tracheal tube could reduce airway injury associated with bougie. However the use of intubation may cause airway trauma in case of difficult airway. Herein, we present a male case of difficult intubation and challenging airway management [1–3].

CASE REPORT

A 46-year-old male with the American Society of Anesthesiologists (ASA) Class II patient was admitted for uvuloplasty and evaluated before surgery. His medical history revealed diabetes mellitus with a smoking history of seven pack-years. His Mallampati score was II, neck movements were intact, and mouth opening was 5 cm. The patient was scheduled for uvuloplasty procedure due to obstructive sleep apnea syndrome (OSAS) by the ear, nose, and throat (ENT) specialist. At baseline, his blood pressure was 150/95 mmHg, pulse rate was 85 bpm, and oxygen saturation was 93%. The body mass index was 34.7 kg/m^2 and his ideal body weight was 84 kg.

A written informed consent was obtained from the patient. Midazolam 2 mg was used in premedication, and anesthesia was induced with thiopental 500 mg, rocuronium bromide 60 mg, and remifentanil 60 μg. The patient was intubated at the second attempt using direct laryngoscope and a bougie. Respiratory sounds were equal bilaterally after intubation and harsh in the left upper zone, particularly. The anesthesia was maintained with sevoflurane 2% and remifentanil infusion with a starting dose of 0.125 μg/kg/min. Tidal volume was set to 475 mL and respiratory rate was set to 16/min on mechanical ventilator. Blood pressure ranged from 110/80 to 130/90, pulse rate was 70–75 bpm, end-tidal CO_2 was 33–34 mmHg, and oxygen saturation was 96–98%. Peak pressures had an elevated course after intubation. There were also harsh bilateral respiratory sounds on auscultation during inspiration and expiration in the right hemithorax, particularly. Harsh sounds were considered to be caused by bronchoconstriction for which the patient was administered as an intravenous bolus dose of methylprednisolone 250 mg and H_2 receptor blocker, and aminophylline 240 mg for an half an hour infusion. Respiratory sounds during control examination at 45 min improved, and the procedure continued for three hours. The patient was administered sugammadex sodium 200 mg to reverse the effects of muscle relaxant, and the patient was extubated without any complication once he re-gained his muscle strength and consciousness. The patient was transferred to the post-anesthesia care unit and he was administered anti-edema therapy including cold vapor and subcutaneous adrenalin due to hoarseness and mild stridor which were considered to be due to mild edema in the upper airway. Edema resolved at 45 min of follow-up and swelling occurred in the right eye which spread to the whole face within 10 min. There was crepitation on palpation and the patient was re-intubated without any difficulty using a video-assisted laryngoscope to investigate the cause of subcutaneous emphysema. Fiberoptic examination performed by an ENT specialist revealed a 0.5-cm rupture in the cricothyroid membrane and mechanical ventilator support was considered to be appropriate due to risk of upper airway obstruction. On the day of admission to intensive care unit, computed tomography scan revealed free air under the skin and between muscle planes of the submandibular and supraclavicular areas and also around the larynx and trachea in the mediastinum and intraluminal air of the left lateral wall of trachea following the cricoid cartilage extending linearly to the emphysema site at the left side of the neck (Figures 1–3).

The fistula line was thought to be the left lateral wall following the cricoid cartilage and the two bronchi were found to be normal. Mediastinum was enlarged on chest X-ray and there was an increase in radiolucency due to emphysema in the subcutaneous and soft tissue (Figure 4).

The patient was hospitalized for spontaneous recovery and he remained intubated for soft tissue repair and wound healing for two days. On day-3, the endotracheal tube was removed and no difficulty or complication was seen during follow-up. The patient was discharged with full recovery in the postoperative first week.

DISCUSSION

Difficulties during airway management and intubation can be encountered in obese patients. The

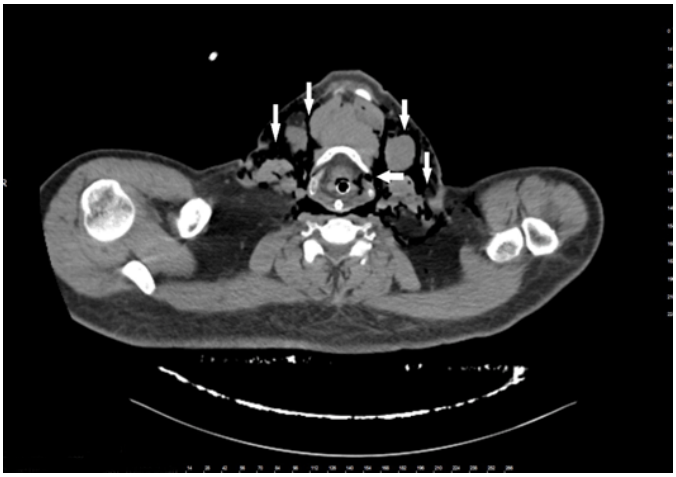

Figure 1: Free air under the skin and between muscle planes of the submandibular and supraclavicular areas, and also around the larynx and trachea in the mediastinum.

risk of intubation difficulty is two-fold higher in the obese patients [4]. Fat deposition around the neck and large tongue in obese patients complicate laryngoscopic view and intubation [5]. It is three times more difficult to perform mask ventilation in patients with a BMI of >26 kg/m^2 [6].

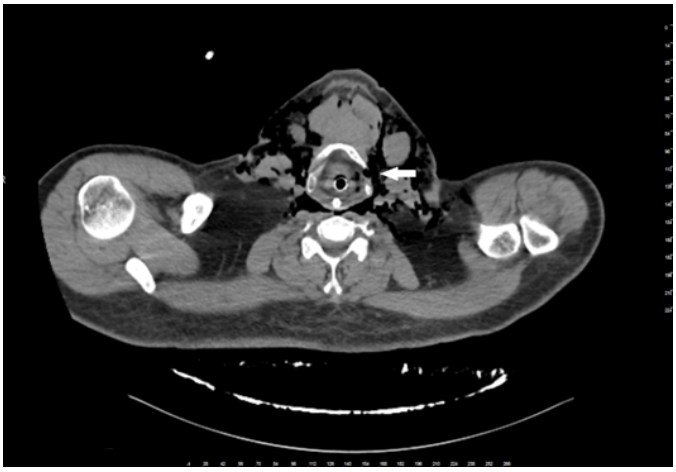

Figure 2: The fistula line was thought to be the left lateral wall following the cricoid cartilage.

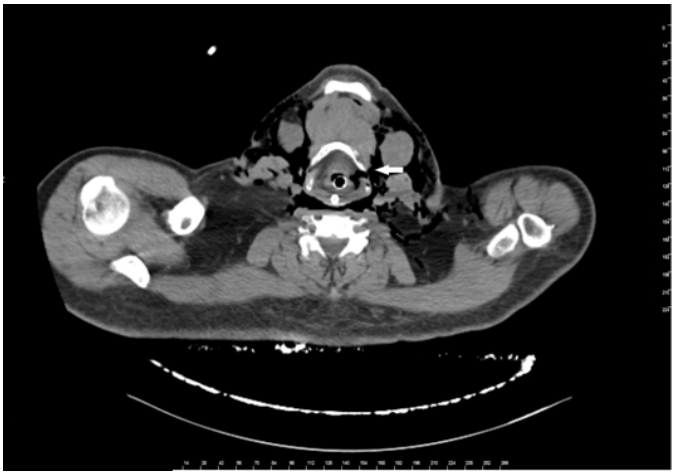

Figure 3: İntraluminal air of the left lateral wall of trachea following the cricoid cartilage extending linearly to the emphysema site at the left side of the neck.

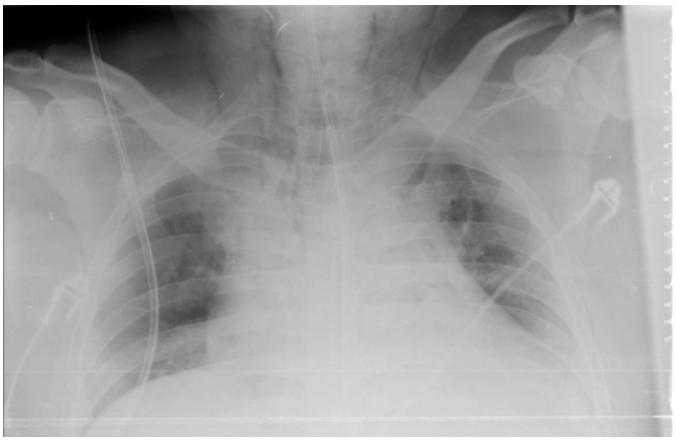

Figure 4: Radiolucency due to emphysema in the subcutaneous and soft tissue on chest X-ray.

Preoxygenation is less effective in these patients than normal-weight patients due to reduced expiratory reserve volume (ERV), and the fact that ERV is the primary back-up oxygen source during apnea [7]. In such cases, 25° head-up position and continuous positive airway pressure can be used for an effective preoxygenation [8]. To facilitate intubation with a laryngoscope, head can be placed in the ramp position by placing a pad under the patient's head [9]. Difficult airway equipment including a laryngeal mask airway and fiberoptic bronchoscope must be kept available due to risk of intubation difficulty [10].

The use of stylet/guidewire or bougie intubation in Grade 2–3 views of larynx provides 90% success rate in the management of a difficult airway using direct laryngoscopy [11]. These instruments are chosen due to their low cost and complication rate and ease of use. The tube is blindly inserted into the trachea or using the Seldinger technique using the tracheal click or distal hold-up signs [12]. The endotracheal tube is, then, slided over the bougie, which is removed after accurate positioning of the tube [13].

Advancing the bougie into the bronchi produces hold-up signs, which bring the risk of possible perforation or trauma [14]. Trauma has been mostly reported with disposable bougies [15–17]. Even 0.8 Newton power has been reported to be sufficient to induce a trauma [16]. While the bougie is placed in the trachea, intubation without withdrawing the laryngoscope increases the chance of intubation [18]. Bougies or stylets need to be pre-shaped under the guidance of a video-assisted laryngoscope [19]. The tube must be advanced to the oropharynx under direct vision to reduce the chance of trauma [20]. As blind advancement of the bougie causes trauma, it is not recommended in patients with Grade 3b and 4b views of larynx, in particular [21].

Trauma is rare with bougies and is always associated with intubation difficulty [22]. Most bougie-related complications, particularly perforations, occur during aggressive placement or pushing the tube against resistance, while sliding the tube over the bougie [23]. In addition, such complications can be related to proximal airways at a lesser extent. There are also reports of pharyngeal perforation [24], bleeding in the right main bronchus [25], laceration in the posterior tracheal mucosa below glottis [26], and tracheal abrasion diagnosed with hemopneumothorax on chest X-ray [27]. In the present case, there was a fistula tract extending from the cricoid cartilage to the left lateral wall of the trachea.

Furthermore, disposable bougies were manufactured in 1997 due to risk of infection [28]. Reusable bougies are coated with polyester-based resin, while disposable bougies are coated with plastic. Therefore, sliding the tube over the bougie may cause resistance, if not lubricated. Zwall and Gupta [29] reported a failed attempt of advancing endotracheal tube over the disposable bougie without using lubricant. Manufacturers also recommend the use of lubricants [30].

It has been previously described that applying lubrication on the bougie moving it forward cautiously and drawing the bougie slightly by several centimeters or stabilizing it with the assistance of other clinical staff during tracheal intubation may help prevent bougies from causing airway injuries [16].

In the present case, we were alert for a difficult intubation, as he was obese, however, intubation with a bougie was attempted due to Grade 2–3 view of larynx under direct laryngoscopy. The use of lubricant facilitated sliding the tube over bougie; however, injury occurred due to excessive force on the bougie. In addition, emphysema developed at 45 min of follow-up and, therefore, we suspected that the patient had suffered an airway injury from the bougie.

CONCLUSION

In conclusion, given the possibility of such bougie-related injuries, long-term postoperative follow-up is of utmost importance for patients in whom intubation is difficult with bougies with several attempts.

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Author Contributions

Ayça Tuba Dumanlı Özcan – 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
Cemile Altın Balcı – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Şemsi Mustafa Aksoy – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
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Togay Müderrris – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Sigmoid colon fistula with tubo-ovarian pelvic abscess: A case report

Pyong Wha Choi

ABSTRACT

Introduction: A tubo-ovarian abscess reflects an inflammatory adhesion of pelvic organs including the fallopian tube and ovary forming a palpable complex, which represents the ultimate process of pelvic inflammatory disease (PID). Fistula formation between the sigmoid colon and other pelvic organs such as the bladder, uterus, and ovary is mainly caused by colorectal cancer or diverticulitis. However, cases in which a tubo-ovarian abscess leads to sigmoidal fistula are extremely rare. Case Report: Here we present a case of sigmoidal fistula secondary to a tubo-ovarian abscess. A 46-year-old premenopausal woman presented with a two-week history of lower abdominal pain. The patient had received antibiotic therapy with the impression of PID at a local clinic prior to presenting to the emergency room, but her symptoms did not resolve. A pelvic examination showed severe tenderness in both adnexal regions but digital rectal examination findings were negative. Computed tomography (CT) scan showed PID with bilateral tubo-ovarian abscesses and an air-containing abscess in the rectovaginal pouch in which a suspicious fistula within the sigmoid colon was noted. Colonoscopy showed continuous excretion of a pus-like substance at the rectosigmoid colon. During the operation, the tubo-ovarian inflammatory complexes and the abscess cavity in the rectovaginal pouch abutting the sigmoid colon were revealed. Total abdominal hysterectomy, bilateral salpingo-oophorectomy, and resection of the sigmoid colon including the affected segment were performed with primary anastomosis. Conclusion: This case represents an unusual type of sigmoidal fistula caused by a tubo-ovarian abscess and describes its surgical treatment.

Keywords: Colon, Fistula, Pelvic inflammatory disease, Tubo-ovarian abscess

How to cite this article

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INTRODUCTION

Tubo-ovarian abscess is a severe inflammatory condition in the pelvis caused by the aggravation of pelvic inflammatory disease (PID) or local extension of an infection such as appendicitis [1]. Medical treatment using combination antibiotics is the mainstay of treatment. Failure to obtain medical treatment may lead to rupture into the abdominal cavity, which requires emergency surgery [2]. However, fistula formation with nearby organs in the pelvis is an extremely rare complication of

tubo-ovarian abscess. The bladder, rectum, and sigmoid colon have been reported as fistula-forming organs with tubo-ovarian abscess [3–6]. Here we present a case of sigmoidal fistula secondary to a tubo-ovarian abscess.

CASE REPORT

A 46-year-old premenopausal woman gravida 2 para 2 presented to the emergency department with lower abdominal pain. She had no specific past medical history including surgery and reported that the pain had started two weeks prior. The patient previously visited a local clinic. With the presumptive impression of PID, she received conservative management including analgesics for pain control and antibiotics for one week. The pain was transiently relieved. No improvement was seen during her local hospital stay, so she was referred to the department of obstetrics and gynecology. Her vital signs at admission were as follows: temperature 37.5°C, blood pressure 120/85 mmHg, pulse 92 beats/minute, and respiratory rate 18 breaths/minute. The lower abdomen was soft and tender without signs of peritoneal irritation. A pelvic examination showed severe tenderness in both adnexal regions, but a digital rectal examination was negative. Laboratory results were within the normal range except for the white blood cell count of 19,500/μL. There were no specific findings on chest or abdominal X-ray. Abdominopelvic computed tomography (CT) scan showed PID with a tubo-ovarian abscess and an air-containing abscess in the rectovaginal pouch with suspicious fistula in the sigmoid colon (Figure 1). Colonoscopy indicated an elevated hard mucosal change 7–20 cm from the anal verge. Although no definite fistula opening was detected, a pus-like substance was continuously draining from the rectosigmoid colon (Figure 2). With the diagnosis of sigmoid fistula and a tubo-ovarian pelvic abscess, elective surgery was performed. During the operation, the tubo-ovarian inflammatory complex and abscess cavity in the rectovaginal pouch abutting the sigmoid colon were revealed, while omental and small bowel adhesions were noted in the abscess cavity. After the colon, omentum, and small bowel were dissected from the abscess cavity and the pus was drained, total abdominal hysterectomy, bilateral salpingo-oophorectomy, and resection of the sigmoid colon including the affected segment with the primary anastomosis were performed. After the surgery, the fistula opening was identified in the surgical specimen that was not preoperatively detectable (Figure 3). The patient's postoperative course was uneventful and she was discharged on the 10th postoperative day.

DISCUSSION

Pelvic inflammatory disease is common, but the differential diagnosis of a surgical abdomen such as appendicitis is crucial because the optimal treatment can vary among disease conditions. Pelvic inflammatory disease is caused by an infection that ascends from the lower genital tract into the fallopian tube and peritoneal cavity [1]. Its treatment of choice is medical, but resistance to medical treatment or the spread of a local inflammatory condition like appendicitis, adnexal surgery, and cesarean section may lead to tubo-ovarian abscess [1, 2]. Although

Figure 1: Abdominopelvic computed tomography image showing pelvic inflammatory disease with bilateral tubo-ovarian abscesses and an air-containing abscess in the rectovaginal pouch with a suspicious fistula with the sigmoid colon (white arrow).
(A) Axial view, (B) Coronal view, and (C) Sagittal view

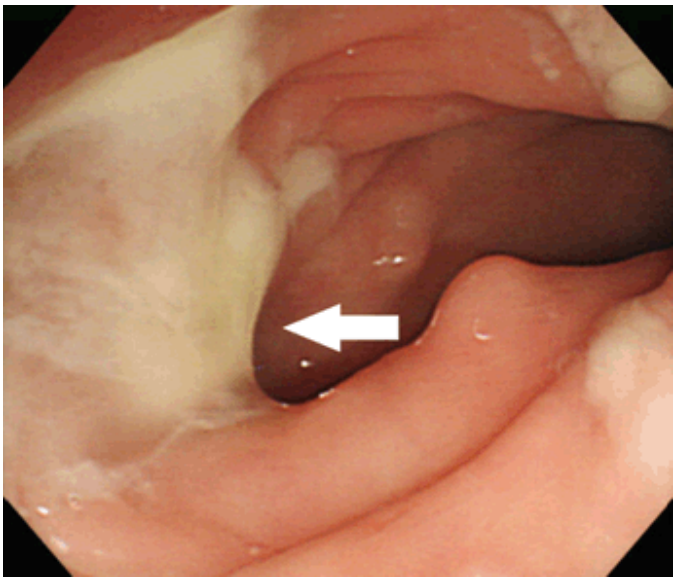

Figure 2: Colonoscopy image showing purulent discharge coming out of the fistula opening at the rectosigmoid colon.

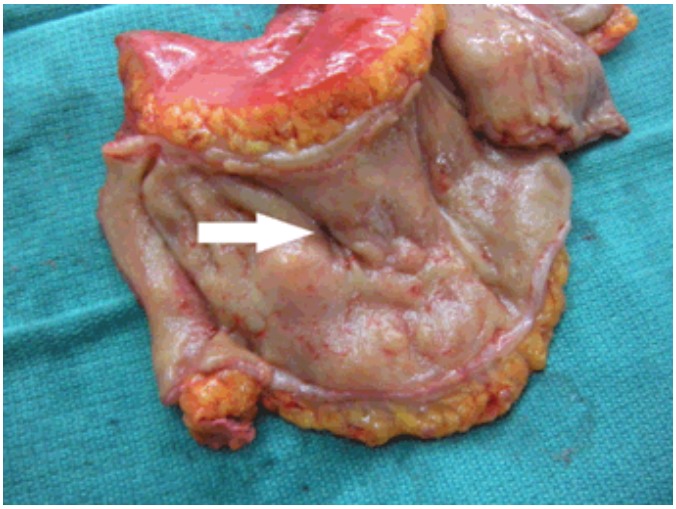

Figure 3: Gross findings, the white arrow indicates the fistula opening of the resected sigmoid colon.

antibiotics for gram-negative organisms combined with clindamycin or metronidazole are recommended in the treatment of tubo-ovarian abscess, medical treatment failure may result in rupture of the abscess into the peritoneal cavity, one of the most serious complications leading to sepsis and mortality. Emergency surgery is required in such cases [7]. Fistula formation around the organs is another rare complication of tubo-ovarian abscess, but it develops chronically and does not require emergency surgery.

The mechanism of fistula formation has not been well established, but the defense mechanism of the spread of inflammation in the abdominal cavity may be a clue to its pathogenesis. Since redundant organs such as the small bowel, sigmoid colon, and omentum play an important role in preventing the spread of inflammation into the abdominal cavity by adhesion to the pelvis, local tubo-ovarian and pelvic abscess may be confined in the pelvis without rupturing into the abdominal cavity. Thus, in such cases, when antibiotic therapy is not effective and an abscess does not rupture into the peritoneal cavity, it may invade other spaces such as the preperitoneal space and form an abscess in the abdominal wall or a nearby organ such as the bladder, rectum, or sigmoid colon and form a fistula as in the present case [3–6, 8]. Therefore, sealing off the inflamed pelvis using the small bowel, sigmoid colon, and omentum is a sort of defense mechanism but also may be a triggering factor of fistula formation in the pelvic organs.

Abdominal pain, the main symptom of fistula secondary to tubo-ovarian abscess, may be relieved by pus drainage into the sigmoid colon or bladder. Depending on the fistula-forming organ, pyuria or purulent diarrhea may also be present, and these symptoms may be pathognomonic in patients with tubo-ovarian abscess and fistula formation [3–6].

The diagnosis of tubo-ovarian abscess is made based on imaging studies. Transvaginal ultrasonography is the first-line imaging study because it provides high-resolution images and avoids radiation, but its interpretation may vary among practitioners [1, 9]. If the US result is equivocal or there is a suspicious lesion such as malignancy, CT scan can be a preferred modality [10]. However, the detection of a fistula tract or opening on a CT image may be limited. Thus, to detect a fistula tract or opening, a contrast study, magnetic resonance imaging (MRI), and colonoscopy can be useful [4, 6]. In the present case, ultrasound was performed in the local clinic, but the report was not available at admission. Although the CT image indicated a suspicious lesion of a fistula tract with the sigmoid colon, colonoscopy findings made it possible to consider fistula formation with the sigmoid colon. A tubo-ovarian abscess secondary to sigmoid colonic diverticulitis could be considered in the differential diagnosis because diverticulitis may be one of the etiologies for tubo-ovarian abscess [1]. However, there was no evidence of diverticulitis on the CT image; finally, the operative findings and gross findings of the surgical specimen led us to the confirmatory diagnosis.

Surgical treatment for tubo-ovarian abscess varies from drainage and unilateral salpingo-oophorectomy to total abdominal hysterectomy and bilateral salpingo-oophorectomy, but in cases of fistula formation, surgical options have not been well established [1]. Fistula opening sealing with a primary suture but no colon resection may be one option, while involved colon resection with or without diversion may be another [5, 6]. Surgery accompanied by colon resection requires longer operative time than that with fistula opening sealing only, but when the fistula opening sealing is not available due to severe inflammation, colon resection may be a suitable option as in the present case, and if the patient's condition is unstable, Hartmann's operation may be performed. However, secondary operations to reverse the colostomy might lead to general anesthesia–associated risks and postoperative morbidity, particularly in older patients. Thus, the optimal surgical option for fistula tract should be individualized to a patient's condition.

CONCLUSION

Sigmoidal fistula secondary to tubo-ovarian abscess is an extremely rare complication for which the optimal surgical treatment modality has not been established. Prompt preoperative diagnosis and individualized surgical treatment should be provided to avoid morbidity and mortality.

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Author Contribution

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

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Alcohol septal ablation through an anomalous dominant septal artery in hypertrophic cardiomyopathy patient: With separate ostium from right coronary sinus

Neelima Katukuri, Tjuan Overly, Raj Baljepally

ABSTRACT

Introduction: Hypertrophic cardiomyopathy (HOCM) is a genetic disease characterized by left ventricular hypertrophy that has variable morphologic and hemodynamic manifestations. Alcohol septal ablation (ASA) has emerged as a widely accepted alternative to surgical myomectomy for the management of HOCM. In this percutaneous, procedure, pure ethanol is injected into the septal perforator Septal perforator of left anterior descending that supplies the hypertrophied myocardium, leading to infarction and thinning of the myocardium thinning of the hypertrophied. Case Report: A 62-year-old female with known history of HOCM presented with progressive dyspnea, dizziness and intermittent chest pain, despite high doses of beta-blocker and calcium channel blocker calcium channel blockers. Echocardiography revealed asymmetric septal hypertrophy with septal thickness of 16–19 mm, systolic anterior motion of the anterior mitral leaflet and a resting LVOT gradient of 35–40 mmHg, which increased to 124 mmHg with Valsalva. Non-selective angiogram revealed an anomalous septal artery arising from a separate ostium in the right coronary sinus (Bonapace artery). Bonapace artery fed the hypertrophic basal septum and was a suitable target for alcohol septal ablation. Conclusion: In this report, we describe a rare case of clinically important Bonapace artery in the absence of coronary artery disease. Additionally, this artery was utilized to perform successful ASA to achieve gradient reduction and relief of symptoms in HOCM. The presence and clinical importance of an anomalous septal artery should be sought in HOCM patients with lack of dominant septal artery from the left coronary system.

Keywords: Aberrant septal artery, Alcohol septal ablation, Hypertrophic cardiomyopathy

How to cite this article

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INTRODUCTION

Hypertrophic cardiomyopathy (HOCM) is a genetic disease characterized by left ventricular hypertrophy that has variable morphologic and hemodynamic manifestations. Alcohol septal ablation (ASA) has emerged as a widely accepted alternative to surgical myomectomy for the management of HCM. The septum is usually supplied by septal arteries arising from the proximal left anterior descending (LAD) and distal right coronary artery (RCA).

Septal coronary branches arising from the proximal right coronary artery (RCA) or the right coronary sinus, known as descending septal artery (DSA) or Bonapace's branch, have rarely been described. However, the DSA plays an important role in certain situations like patients with hypertrophic cardiomyopathy, highlighting the need for its proper identification and evaluation [1, 2].

CASE REPORT

A 62-year-old female with known history of hypertrophic cardiomyopathy, hypertension and hypothyroidism referred to clinic with worsening dyspnea despite high doses of metoprolol and cardizem. Echocardiography revealed asymmetric septal hypertrophy with septal thickness of 16–19 mm, systolic anterior motion of the anterior mitral leaflet and a resting LVOT gradient of 35–40 mmHg, which increased to 124 mmHg with Valsalva (Figure 1A). Subsequently, she underwent cardiac catheterization as her symptoms were worsening and she had elevated cardiac biomarkers which showed origin of anomalous septal artery from separate ostium of right coronary sinus (Bonapace's artery) and no significant coronary artery disease (Figure 2, Video 1). Computed tomography scan of her chest for pulmonary embolus showed incidental right subclavian artery originating from descending thoracic aorta and no pulmonary embolus. Elevated cardiac biomarkers were assumed to be secondary to hypertensive urgency and her antihypertensive medications were adjusted. After six months, she returned to office with worsening dyspnea despite adequately controlled blood pressure. Electrocardiogram showed normal sinus rhythm with no interventricular conduction delays. Alcohol septal ablation of septal artery was planned as she refused to undergo surgery. Bonapace's artery fed the hypertrophic basal septum and was a suitable target for alcohol septal ablation. A temporary trans venous pacemaker was placed through the right internal jugular vein and AR modified guide was utilized to engage the septal artery. A extra sport wire was used to cross the septal artery. Echocardiogram was obtained to confirm localization of contrast in the basal septum (Video 2). Then 0.2 cc increments of alcohol was injected to a total of 1 ml over a period of 10 minutes.

Subsequently, there was reduction in gradient from peak of 40 mmHg to 15 mmHg. The temporary pacemaker wire was removed after 48 hours without any evidence of heart block. Two months post ablation, her gradient on resting echo remained low with LVOT gradients both at rest (10.5 mmHg) and with Valsalva (13 mmHg) (Figure 1B). This corresponded with symptomatic improvement, increase exercise tolerance and quality of life improvement for the patient.

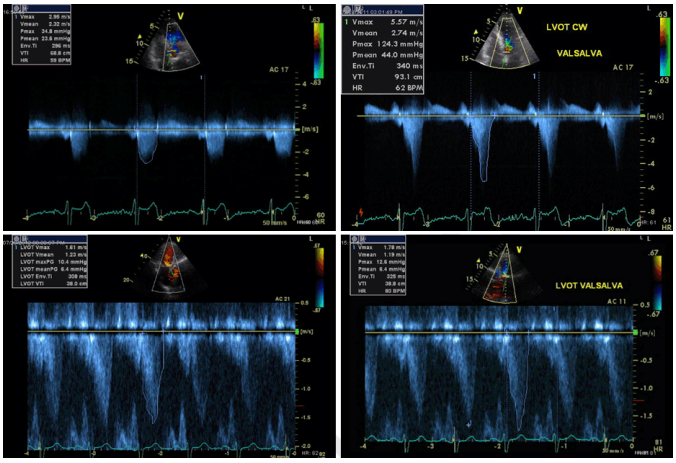

Figure 1: (A) Preablation echocardiographic gradients: Resting (B) Postablation gradients.

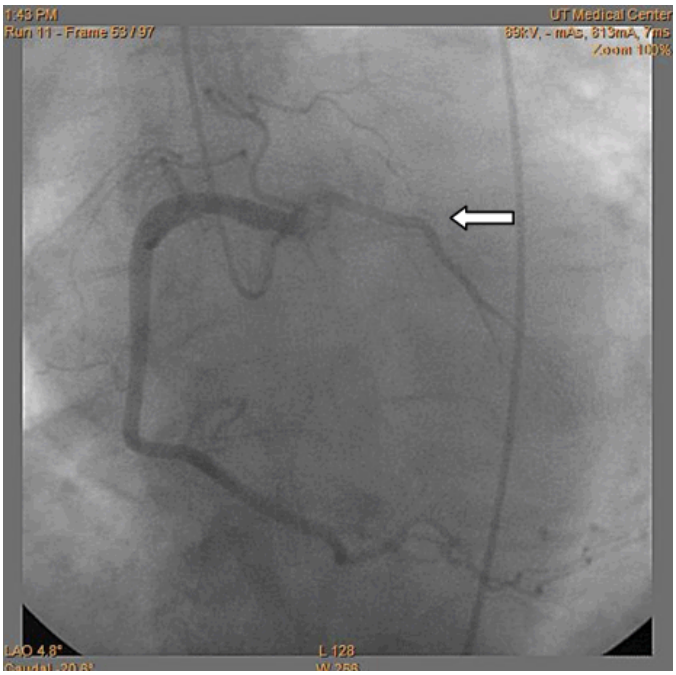

Figure 2: Coronary angiogram showing (double arrow) separate origin of septal artery from right coronary sinus.

VIDEOS

Video 1: Coronary angiogram showing separate origin of septal artery from right coronary sinus. The supplemental motion image also shows right coronary artery.

Video 2: Still and supplemental motion image of coronary angiogram showing sport wire crossing anomalous septal artery.

Video URL: www.ijcasereportsandimages.com/archive/2017/007-2017-ijcri/CR-10804-07-2017-neelima/ijcri-1080407201704-neelima-full-text.php

DISCUSSION

On review of literature there was case of dominant septal artery originating from right coronary artery

first described in 1995 [2]. This anomaly when present can be safely and successfully utilized for alcohol septal ablation [3]. Kurita et al. [4] performed a percutaneous septal alcohol ablation through a DSA, preceded by the demonstration of its contribution to the perfusion of the basal interventricular septum.

It is also important to understand the technical aspects concerning the evaluation of the DSA which can sometimes be missed. Deep cannulation of the RCA may prevent its identification and in some cases a DSA arising from an independent ostium was accidentally identified while attempting to find the RCA as in our case. Also, contrast back-flow during injections in the RCA might reveal its presence [1, 5].

CONCLUSION

In this report, we describe a rare case of clinically important Bonapace's artery, which has a separate ostium on right coronary sinus. Additionally, this artery was utilized to perform successful alcohol septal ablation to achieve gradient reduction and relief of symptoms in hypertrophic cardiomyopathy. The presence and clinical importance of an anomalous septal artery which should be sought in HOCM patients with lack of dominant septal artery from the left coronary system.

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Author Contributions

Neelima Katukuri – 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
Tjuan Overly – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Raj Baljepally – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Unusual gastric submucosal perforation following multiple magnetic bead ingestion in an infant

Ghassan Nakib, Valeria Calcaterra, Balaji Krishnamurthy, Gloria Pelizzo

ABSTRACT

Introduction: Magnetic beads are hazardous, having potentially lethal consequences if ingested. Case Report: A two-year-old girl, presented to the pediatric emergency department with an history of worsening abdominal pain, nausea and vomiting. The patient appeared in moderate distress. Physical examination revealed abdominal tenderness with guarding and rigidity. On abdominal X-ray examination, nine opaque foreign bodies (magnetic beads) were seen in the left hypochondrium. Explorative gastroscopy was performed and two magnets appeared at the lessure curvature, but not the reaming seven. These were pulled out and another gastroscopic attempt showed appearance of another two of the remaining ones. Removing these, the remaining five were all attached as seen on the X-ray. The magnets must have caused pressure necrosis on a gastric mucosa fold with subsequent limited submucosal perforation making a pouch was the spheres were logged. Conclusion: The ingestion of multiple magnets may require urgent surgical intervention and early removal because of very high risk of gastrointestinal perforation.

Keywords: Children, Ingestion, Magnetic beads, Perforation

How to cite this article

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INTRODUCTION

Accidental ingestion of foreign bodies is a common problem in children [1–2]. The majority of ingested foreign bodies pass spontaneously without treatment. Magnetic beads are hazardous, having potentially lethal consequences if ingested [3–6].

CASE REPORT

A two-year-old girl, weight 19 kg presented to the pediatric emergency department with an eight hour history of worsening abdominal pain, nausea and alimentary vomiting. The patient appeared pale with dry mucous membranes and in moderate distress. Vitals signs were as follows: temperature 36.7°C, heart rate 156 bpm, SPO2 100%, respiratory rate 26 rpm, blood

pressure 90/72 mmHg. Physical examination revealed abdominal tenderness with guarding and rigidity. The intravenous infusion of crystalloid fluids was started in pediatric emergency department.

On abdominal X-ray examination, there was no free air or dilated bowel loops; however, nine opaque foreign bodies (magnetic beads) were seen in the left hypochondrium (Figure 1A). Explorative gastroscopy was performed and two hypochondrium (Figure 1B), but not the remaining seven. These were pulled out and another gastroscopic attempt showed appearance of another two of the remaining ones. Removing these, the remaining five were all attached as seen on the X-ray (Figure 1C). Gastric perforation was suspected. Laparoscopic assisted enterolysis from the Treitz to terminal ileum was performed and no perforation was seen. Stomach anterior wall was free of any perforations. The magnets must have caused pressure necrosis on a gastric mucosa fold with subsequent limited submucosal perforation making a pouch were the spheres were logged (Figure 1D). The postoperative period was uneventful and the patient was discharged on the second postoperative day.

DISCUSSION

Complications from foreign body ingestion in children are uncommon but the ingestion of multiple magnets may require urgent surgical intervention and early removal because of very high risk of gastrointestinal perforation [1–3].

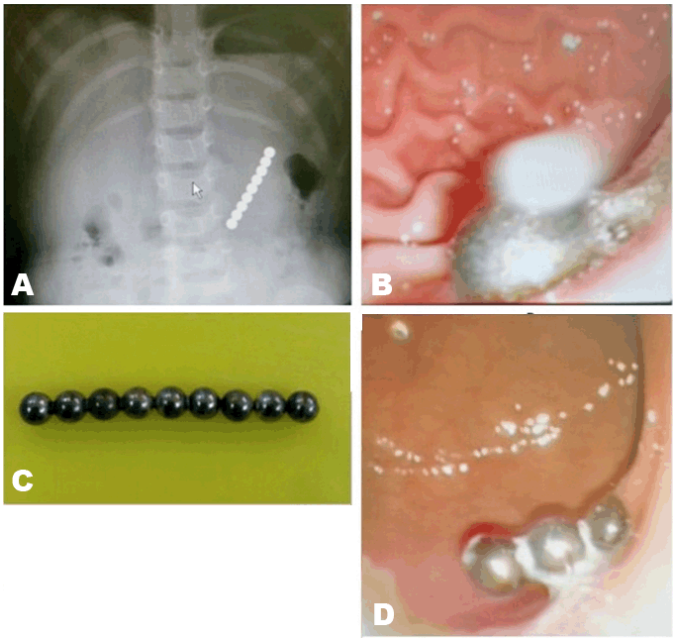

Figure 1: Gastric submucosal perforation following multiple magnet ingestion. Panel (A) Abdominal X-ray examination showing nine opaque foreign bodies with a round shape in the gastric area, (B) Two magnets appeared at the lessure curvature, (C) Ingested magnets, and (D) Submucosal gastric pouch in which the magnetic bead were lodged.

CONCLUSION

Prevention campaigns from health authorities should start about the hazards of magnetic toys to prevent the appearance of new similar cases.

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Author Contributions

Ghassan Nakib – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important content, Final approval of the version to be published

Valeria Calcaterra – Substantial contributions to conception and design of data, Drafting the article, Revising it critically for important content, Final approval of the version to be published

Balaji Krishnamurthy – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important content, Final approval of the version to be published

Gloria Pelizzo – Substantial contributions to conception and design of data, Drafting the article, Revising it critically for important content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

A case of pulmonary artery aneurysm

Aniket S. Rali, Tyler Buechler, Steven Whitfield

ABSTRACT

Introduction: The natural history of pulmonary artery aneurysm (PAA) is poorly understood due to the limited number of cases diagnosed ante-mortem. Aneurysms of the proximal pulmonary artery are found in approximately 1 in 14000 postmortem examinations. The pulmonary artery trunk is considered aneurysmal when the diameter exceeds 4 cm. Case Report: A 62-year-old woman with rheumatoid arthritis, moderate COPD (requiring supplemental oxygen), untreated obstructive sleep apnea, moderate pulmonary hypertension (mean pressure of 40 mmHg), 90 pack years smoking history and HFpEF presented to the hospital with acute on chronic hypoxemic respiratory failure. The patient was noted to have a pulmonary artery aneurysm measuring 6 cm in diameter on a computed tomography scan obtained to rule out pulmonary embolism. We believe the mechanism of PAA in our patient to be secondary to the structural changes in elastin and collagen due to increased pulmonary artery pressure leading pulmonary artery dilation and subsequent aneurysm. Due to poor pulmonary reserve, patient is not considered a good candidate for surgical intervention and conservative management was opted in her case. Conclusion: Pulmonary artery aneurysm continues to remain a poorly understood disease entity. Treatment options are often limited by late/acute presentations and multiple co-morbidities. However, advances in imaging and higher degree clinical suspicion allow for earlier identification of PAAs and allow for appropriate intervention. We hope that our case report will allow medical providers to be on the look-out for PAAs in patients with above mentioned risk factors.

Keywords: Aneurysms, Pulmonary artery, Pulmonary hypertension

How to cite this article

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INTRODUCTION

The natural history of pulmonary artery aneurysm (PAA) is poorly understood due to the limited number of cases diagnosed ante-mortem [1]. Aneurysms of the proximal pulmonary artery are found in approximately 1 in 14000 post-mortem examinations [2]. The pulmonary artery trunk is considered aneurysmal when the diameter exceeds 4 cm [3]. They have been associated with structural heart disease, with 50% of the postmortem cases associated with congenital heart defects, as well

as, structural vascular defects, vasculitides, pulmonary hypertension, infections, trauma, and a few cases of idiopathic pulmonary artery aneurysms have been reported [3]. Advances in diagnostic imaging have made earlier identification of PAAs easier, and allows for appropriate intervention prior to aneurysmal rupture. Here, we report a case of a patient with multiple co-morbidities and incidental pulmonary artery aneurysm.

CASE REPORT

A 62-year-old female with rheumatoid arthritis, moderate COPD (requiring supplemental oxygen), untreated obstructive sleep apnea, moderate pulmonary hypertension (mean pressure of 40 mmHg), 90 pack years smoking history and heart failure with preserved ejection fraction (HFpEF) presented to the hospital with acute on chronic hypoxemic respiratory failure. Chest X-ray was unremarkable except for prominent pulmonary vasculature (Figure 1A). A computed tomography (CT) scan showed no acute pulmonary embolism, pulmonary fibrosis or rheumatoid pleurisy. However, a 6.0-cm dilation of the main pulmonary artery was noted (Figure 1B–D). Right heart catheterization performed two days after the CT scan demonstrated mean right atrial pressure of 10 mmHg, right ventricle pressure of 57/12 mmHg, pulmonary artery pressure of 59/31 with mean pressure of 40 mmHg. The mechanism of patient's pulmonary artery aneurysm was believed to be multi-factorial including pulmonary hypertension, chronic emphysematous changes, untreated obstructive sleep apnea and heart failure.

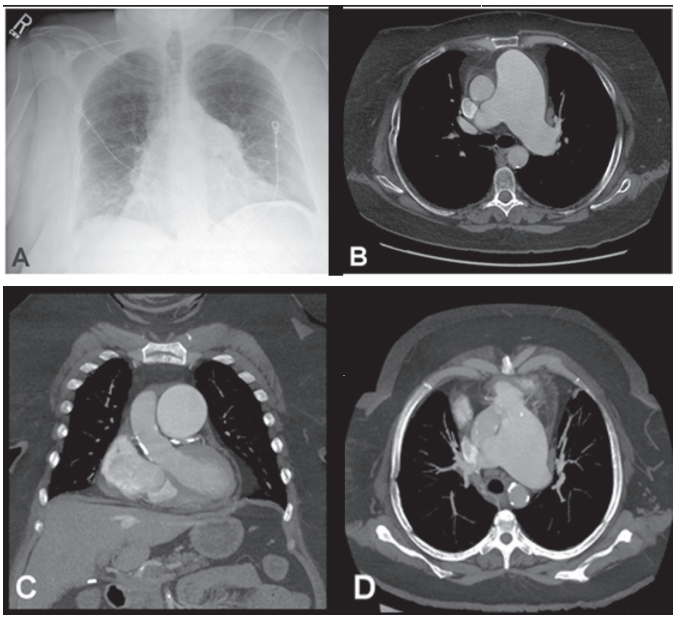

Figure 1: A: Chest X-ray showing the prominent pulmonary vasculature, and (B–D): Computed tomography scan showing various views of the pulmonary artery dilatation of 6 cm.

Due to poor pulmonary reserve, the patient is not considered a good candidate for surgical intervention and conservative management was opted in her case.

DISCUSSION

Pulmonary artery aneurysms are infrequently diagnosed ante-mortem and are a rare entity in general with less than 200 cases reported [4]. Congenital heart diseases are most commonly associated with PAA. Two studies, looking at proximal PAAs at autopsy found the incidence of congenital heart disease to be around 56% [3]. The most common congenital defects include patent ductus arteriosus, ventricular septal defects and atrial septal defects [5]. These entities increase the sheer stress on the pulmonary artery due to left to right shunt, causing weakness in the vascular wall.

The remaining causes of PAA include infectious, traumatic, iatrogenic and idiopathic causes. Of the acquired causes, pulmonary hypertension was noted in 66% of patients [3]. It has been proposed that structural changes in elastin and collagen secondary to increased pulmonary artery pressure leads to pulmonary artery dilatation and subsequent aneurysm [5]. We believe this to be the mechanism of PAA in our patient as well.

Infectious etiologies have classically been associated with syphilis and tuberculosis [6]. When pulmonary tuberculosis is the cause, patients develop a Rasmussen aneurysm, which is an inflammatory pseudo-aneurysmal dilatation of a pulmonary artery branch adjacent to a tubercular cavity [6]. Iatrogenic causes include Swan-Ganz catheter insertion, chest tube insertion, surgical resection, and catheter based pulmonary angiography [5].

While the natural history of PAA is poorly understood, PAAs represent a life-threatening disease if they progress to rupture or dissection. However, not all aneurysms progress to the rupture stage [5]. The risk of rupture is proportional to the stress placed on the aneurysm which is affected by pressure, wall thickness, and radius of the vessel [3]. In one systemic review, out of 66 case reports on pulmonary artery dissection and rupture, 62% of cases were associated with high pressure [4]. Furthermore, the duration of elevated pulmonary artery pressure is also an important consideration. Aneurysms seen in conjunction with congenital heart defects show the most unexpected deaths and most pulmonary artery dissections [4]. In the absence of elevated pulmonary pressures from pulmonary valve defects, pulmonary hypertensions, or left to right shunt, the risk of aneurysmal rupture appears to be very small [3]. A pulmonary artery diameter greater than 5.5 cm in conjunction with high pulmonary artery pressure was associated with more sudden unexpected death [4]. The systemic review by Duijnhouwer et al., classify high risk aneurysms for rupture or dissection as aneurysms with: diameter >7.5 cm, pulmonary artery pressure of 50 mmHg, and

aneurysmal growth rate >2 mm/year [4]. Although our patient did not have any of these high risk features, her ongoing contributors of increased stress on the aneurysm, including untreated sleep apnea, COPD and moderate pulmonary hypertension, placed her at an increased risk of PAA rupture or dissection.

Optimal treatment for PAAs remains unclear and treatment guidelines have not been established because of low disease incidence. Treatment can be either surgical intervention, management of underlying pulmonary hypertension or conservative management [3]. One study recommended surgical repair if aneurysms were larger than 6.0 cm [3]. Surgical interventions include aneurysmorrhaphy or arterioplasty, pericardial patch reconstruction, and interposition grafting [3]. Improvement in endovascular techniques have allowed for less invasive approaches that produce less damage to the lung parenchyma; recently, steel coil embolization and balloon embolization have been reported [5]. Patients with pulmonary hypertension should be treated appropriately directed at the underlying cause to lower pulmonary artery pressures and subsequently the sheer stress on the pulmonary arterial wall. Given her co-morbidities and poor lung reserve, our patient was not deemed to be a surgical candidate and hence after discussion with the patient and her family, we opted for conservative management.

CONCLUSION

Pulmonary artery aneurysm (PAA) continues to remain a poorly understood disease entity. Treatment options are often limited by late/acute presentations and multiple co-morbidities. However, advances in imaging and higher degree clinical suspicion allow for earlier identification of PAAs and allow for appropriate intervention. We hope that our case report will allow medical providers to be on the look-out for PAAs in patients with above mentioned risk factors.

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Author Contributions

Aniket S. Rali – 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
Tyler Buechler – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Steven Whitfield – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Calcitriol mediated hypercalcemia due to necrotizing sarcoid granuloma of the liver

Priyadarshini Balasubramanian, Deepak Kana Kadayakkara, Gregory Soloway, William Laskin, Sachin Majumdar

ABSTRACT

Introduction: Necrotizing sarcoid granulomatosis (NSG) is a rare variant of sarcoidosis with pathologic features of necrosis and vasculitis that overlap with rheumatologic and infectious diseases. Hypercalcemia is occasionally the presenting feature of classical sarcoidosis occurring in 10–20% of patients, most commonly in association with pulmonary involvement. However hypercalcemia in NSG is less common and has not been previously described when NSG primarily affects the liver. Case Report: We report a 65-year-old Caucasian female who presented with hypercalcemia and NSG of the liver without pulmonary involvement. Conclusion: Hypercalcemia was mediated by 1, 25-vitamin D which was most likely produced by hepatic granulomas and it resolved rapidly with prednisone. Further, we describe the time-course for the resolution of hypercalcemia and other biochemical abnormalities with treatment, and suggest potential roles for monitoring 1, 25-dihydroxyvitamin D and angiotensin converting enzyme in guiding therapy.

Keywords: 1,25-Dihydroxyvitamin D, Hepatic granuloma, Hypercalcemia, Necrotizing sarcoid granuloma

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INTRODUCTION

Sarcoidosis is a multisystem granulomatous disorder classically observed in young African-American females of 25–40 years of age. However, it can occur in all races and age groups, with about 30% of cases in elderly patients [1]. In over 90% of cases sarcoidosis involves the lung, and hypercalcemia, occurring in 10–20% of patients, is rare in the absence of pulmonary involvement [2]. Necrotizing sarcoid granulomatosis (NSG) is an uncommon variant of sarcoidosis characterized by granulomas with necrosis and vasculitic changes [3]. Necrotizing sarcoid granulomatosis can mimic rheumatologic and infectious diseases, yet like classical sarcoid it primarily affects the lungs, though rarely it can present with extra-pulmonary manifestations. When extra-pulmonary involvement occurs the most commonly affected sites are the eyes and the central nervous system. Unlike classic sarcoidosis which is more common in African-American females,

NSG is thought to be more prevalent in Caucasians. We present a case of calcitriol mediated hypercalcemia due to NSG of the liver in the absence of pulmonary involvement in an elderly Caucasian female.

CASE REPORT

A 65-year-old female presented to the hospital with a six-week history of headache, vomiting, weight loss, confusion and constipation. She denied fever, loss of appetite, cough, chest pain, visual disturbances or weakness.

Past medical history of the patient was remarkable for hypertension, depression and intra-ductal atypia of left breast s/p lumpectomy a year ago. Her medications at home included losartan, atorvastatin and quetiapine. She denied taking over the counter nutritional supplements. Physical examination of the patient was unremarkable.

Laboratory workup revealed a calcium level of 15.8 mg/dl (8.5–10.2 mg/dl), 25-hydroxyvitamin D 28 ng/ml (20–100 ng/ml), 1, 25-dihydroxyvitamin D (calcitriol) 119 pg/ml (20–62 pg/ml) and parathyroid hormone (PTH) of 7.3 pg/ml (7.5–53 pg/mL). Creatinine was 1.2 mg/dl, AST 126 U/L (14–36 U/L), ALT 156 U/L (9–52), and ALP 342 U/L (38–126 U/L). Chest X-ray was normal. The results indicated calcitriol-dependent hypercalcemia, and the diagnostic possibilities considered were malignancies (hematological cancers and solid tumors) and granulomatous disorders including tuberculosis and sarcoidosis. Computed tomography scan of the chest, abdomen and pelvis did not show evidence of malignancy or lymphadenopathy. Ultrasound of the liver showed normal echotexture with no focal lesions. A skeletal metastatic series was negative as were serum and urine protein electrophoresis. Upper gastrointestinal endoscopy with biopsies of stomach, duodenum, and distal esophagus were negative for malignancy, parasites and celiac disease. An angiotensin converting enzyme (ACE) level was 260 u/L (8–53 u/L).

With elevated liver enzymes, high ACE, and a negative malignancy workup, a liver biopsy was performed which showed necrotizing granulomas present in portal areas and in parenchyma. No GMS fungal elements or AFB-positive organisms were identified. Reticulin stain showed normal liver plates and iron stains were negative. The PAS with diastase failed to identify organisms but highlighted lipofuscin within Kupffer cell granulomas associated with necrotic hepatocytes consistent with the necrotizing variant of sarcoidosis, of NSG, as shown in (Figure 1).

The patient was started on 20 mg/d of prednisone and improved symptomatically along with significant reductions in calcium, calcitriol, ALP, liver enzymes, and ACE levels as depicted in (Figure 2). Serum calcium and calcitriol normalized within one week of starting steroids, ALP took at least one month to normalize, and liver enzymes and ACE declined significantly yet remained elevated. Calcitriol correlated with hypercalcemia and responded rapidly to steroid therapy with suppression maintained at low doses, which permitted rapid tapering while minimizing side effects. Angiotensin converting enzyme levels declined yet their persistent elevation appeared to reflect a more sensitive measure of disease activity with less clinical impact.

Prednisone was tapered and stopped in eight months. One month after discontinuation of prednisone the calcium was 9.2 mg/dl, 1, 25 vitamin D 39 pg/ml, ACE 89 U/L, AST 52 U/L, ALT 36 U/L and ALP 102 U/L.

DISCUSSION

Sarcoidosis is a multisystem disease of unknown etiology with the pathological hallmark being the presence of non-caseating granulomas. The NSG is a rare and poorly understood entity characterized by granuloma with caseous necrosis making it difficult to distinguish from infectious conditions like tuberculosis. It was first described in 1973 by Liebow as sarcoid-like granuloma with vasculitis and necrosis, not caused by rheumatological or infectious diseases [3]. There is a controversy as to whether the entity is sarcoidosis with necrosis of the vessels, or a separate vasculitic disorder. Therefore, it is important to exclude other conditions to avoid unnecessary delay in treatment. Necrotizing

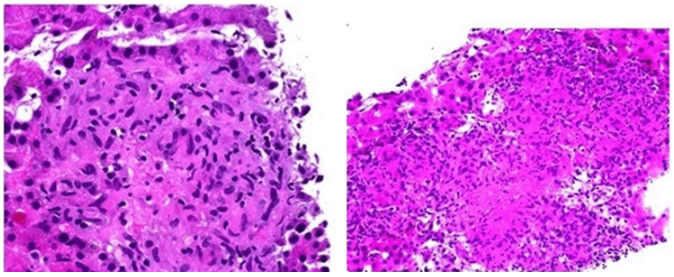

Figure 1: Biopsy of the liver. (A) Liver parenchyma showing granulomatous inflammation and necrosis (x200), and (B) Discrete sarcoid granuloma (x400). Acid-Fast Bacilli (AFB) and fungal stains were negative.

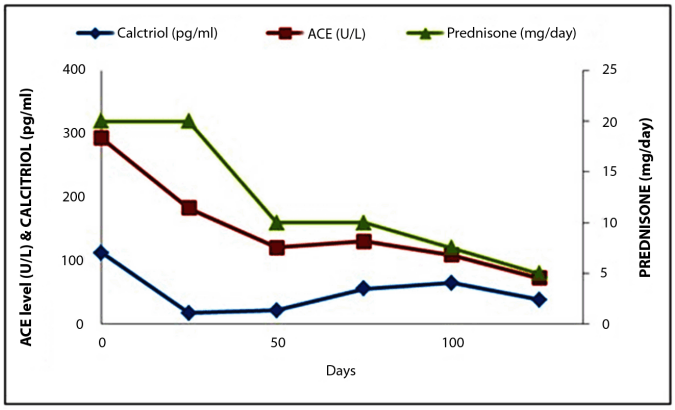

Figure 2: Graph showing angiotensin converting enzyme and calcitriol in response to prednisone.

sarcoid granulomatosis is thought to be more common in the lung with extra-pulmonary involvement being very rare, though when present the most commonly affected sites are the eyes and central nervous system [4]. While sarcoidosis is typically more common in African-American females, all of the 14 cases of NSG in a review published by Quaden et al. were Caucasians, of which 10 patients were females and 4 were males [5]. Therefore, one could postulate that NSG is more common in Caucasian females, and our case supports this hypothesis.

Necrotizing sarcoid granulomatosis of the liver with calcitriol mediated hypercalcemia has not been previously described. There are only two other cases of NSG of the liver described in literature. The clinical presentation was not described in one patient and the second patient did not have hypercalcemia associated with NSG of the liver [6]. Hypercalcemia in sarcoidosis and other granulomatous conditions appears to be mediated by elevated levels of circulating calcitriol resulting in increased intestinal calcium absorption [7]. Excessive calcitriol synthesis in sarcoidosis has been shown to originate from increased 1α-hydroxylase activity in alveolar macrophages [8, 9]. Hence hypercalcemia is rare in the absence of pulmonary involvement. Our patient had no evidence of pulmonary involvement suggesting calcitriol production in extra-pulmonary granulomatous tissue such as the liver or GI tract, yet we cannot exclude the possibility of occult pulmonary disease.

Angiotensin converting enzyme has historically been used to monitor disease activity in sarcoidosis, but calcitriol has not. However, when hypercalcemia is the major clinical manifestation, calcitriol may be useful in guiding therapy, particularly as a more direct marker of disease activity whose responsiveness to treatment could foster rapid reductions in steroid dose, while providing reassurance of therapeutic effectiveness. In our case, when the prednisone dose was lowered, calcitriol levels remained stable, and this was helpful in deciding to proceed with tapering steroid doses and ultimately discontinuing treatment while providing reassurance that treatment remained effective and the disease was stable. Monitoring calcitriol also provided the potential to prevent symptomatic hypercalcemia when steroids were discontinued, particularly in the setting of persisting ACE elevations, because if calcitriol levels were to rise, one could consider resuming steroid treatment before significant hypercalcemia could occur. Angiotensin converting enzyme levels were useful indicators of disease activity, but were a step removed from the pathogenesis of hypercalcemia in comparison to calcitriol, yet changes could be informative in regards to disease activity, with rises in ACE potentially prompting closer monitoring or treatment consideration. Hence, we propose that monitoring calcitriol can help guide steroid therapy in the acute setting when sarcoidosis is associated with hypercalcemia, while ACE levels may be useful for chronic disease monitoring rather than treatment guidance.

Corticosteroids are the mainstay of treatment in sarcoidosis. Recommendations suggest 1 mg/kg/day of prednisone which can be tapered over several weeks to months, however required to lower calcium level may be much lower. Drugs like methotrexate, hydroxychloroquine or azathioprine may be used in steroid resistant cases or as steroid sparing agents. Prognosis is variable and depends on gender, race, age, organ involvement, the signs and symptoms at presentation. In the majority of patients, sarcoidosis stabilizes in the first two years of illness with treatment. There are no gold standard tests available to monitor the response to therapy.

CONCLUSION

We describe the first case of calcitriol-mediated hypercalcemia associated with necrotizing sarcoid granulomatosis (NSG) of the liver in the absence of pulmonary involvement. The NSG appears to be more common in Caucasian females and sarcoidosis can present with hypercalcemia without pulmonary involvement. Elevated calcitriol and angiotensin converting enzyme levels can be useful markers of disease activity and also potentially guide treatment. Calcitriol monitoring can help guide therapy by matching steroid dose with its relative effectiveness on suppression of hypercalcemia, while monitoring angiotensin converting enzyme can provide information on disease activity potentially altering longer term care needs of the patient. In this context, both measures have the potential to add precision to medical decision making in efforts to minimize side effects of therapies, gauge their effectiveness, and provide more specific information about disease activity to the clinician and patient.

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Author Contributions

Priyadarshini Balasubramanian – 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
Deepak Kana Kadayakkara – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Gregory Soloway – 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
William Laskin – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published
Sachin Majumdar – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Temporary exacerbation of primary epiploic appendagitis: A case report

Masahide Hara, Yumiko Ando, Kazuhide Tohara

ABSTRACT

Introduction: Primary epiploic appendagitis (PEA) is a disease with a good prognosis, and numerous reports have indicated that it can be cured without surgical treatment. However, there have been few reports on the necessity of anti-microbial therapy. There is also a paucity of information regarding what kind of management should be provided if the symptoms do not improve. Case Report: A 69-year-old male presented to the hospital with left lower abdominal pain lasting 24 hours. Tenderness and rebound were found during the abdominal examination. The serum C-reactive protein level was mildly increased to 1.43 mg/dl, and the leukocyte count was 8,800/mm³. While abdominal radiography revealed no abnormalities, computed tomography (CT) scan revealed intraperitoneal panniculitis near the sigmoid colon. He was diagnosed with PEA. He received fluid therapy and levofloxacin intravenously. On the third day of hospitalization, his abdominal pain worsened. Subsequent CT scan reconfirmed this diagnosis. The administration of oral non-steroidal anti-inflammatory drugs (NSAIDs) led to improvement of the patient's symptoms, laboratory abnormalities, and imaging findings. He was discharged from the hospital on the eighth day after initial onset. Conclusion: Primary epiploic appendagitis is a self-limiting disease, and NSAIDs (rather than antibiotics) are effective for treating aseptic inflammation. It is important to differentiate PEA from acute abdomen requiring surgical treatment. Computed tomography scan is useful for diagnosing this disease and determining appropriate treatment.

Keywords: Antibiotics, Non-steroidal anti-inflammatory drugs, Primary epiploic appendagitis, Self-limiting disease

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INTRODUCTION

It is important to distinguish primary epiploic appendagitis (PEA) from other diseases in the clinical evaluation of patients with acute abdomen. The reported treatment modalities for PEA have primarily focused on surgical techniques [1]. In recent years, many reports have stated that if the diagnosis is correct, most patients have a good prognosis without surgical treatment [2, 3]. However, there are few reports on how to manage PEA in patients with deteriorated symptoms. In addition, there is limited information regarding whether antibiotics are

useful as conservative treatment. We describe a patient with PEA of the sigmoid colon, and we discuss available literature on the topic.

CASE REPORT

A 69-year-old male with myocardial infarction had been treated conservatively. He reported smoking 30 cigarettes per day for 52 years, and drinking one bottle of beer and a glass of *shochu* with hot water per day. He had no significant family medical history. He was receiving outpatient treatment for high blood pressure and ventricular extrasystole.

The presented to the hospital with left lower abdominal pain beginning the previous evening. He was admitted to the hospital on the same day with a suspected diagnosis of colonic diverticulitis. He was 164.8 cm tall, weighed 96.0 kg, had a body mass index (BMI) of 35.3 kg/m^2, and a temperature of 36.0°C. His blood pressure was 112/87 mmHg, and his pulse was regular at 86 beats per minute. On physical examination, his abdomen was bulging, and bowel sounds were normal. Tenderness and rebound were present in the left lower quadrant. There were no palpable masses in the abdomen.

The venous blood examination showed a C-reactive protein level of 1.43 mg/dL (normal range 0–0.14 mg/dL) and white blood cell count of 8,800/mm^3 (normal range 3,300–8,600/mm^3). The leukocytes comprised 56% neutrophils (normal range 38–72%), 29% lymphocytes (normal range 19–50%), 5% monocytes (normal range 4–11%), 8.4% eosinophils (normal range 0.5–9.6%), and 0.9% basophils (normal range 0.2–3.6%). No other laboratory abnormalities were present. The abdominal radiograph showed no abnormality. The abdominal computed tomography (CT) scan showed no intraperitoneal free air or signs of ileus. A hyper-attenuated nodular soft tissue mass surrounded by a fatty rim and localized peritoneal thickening was identified. The mass was located anterior to the sigmoid-descending colon junction. It was not continuous with the sigmoid colon, and he was diagnosed as having PEA (Figure 1A). Since secondary epiploic appendagitis could not be completely ruled out, he began fasting. He received fluid replacement therapy and 500 mg of levofloxacin per day intravenously.

On the third day of hospitalization, his abdominal pain exacerbated and abdominal tenderness and rebound were observed. Venous blood analysis revealed a WBC count of 7,300/mm^3, and the C-reactive protein level was 1.60 mg/dl. To reconfirm the diagnosis, CT scan was repeated. Computed tomography scan showed exacerbation of the density of the fat rim and peritoneal thickening (Figure 1B). There were no inflammatory findings on the intestinal wall, and abscess formation was not observed. Therefore, conservative treatment was continued accompanied by the oral administration of 180 mg daily of loxoprofen sodium.

On the seventh day of hospitalization, the patient's abdominal pain and tenderness gradually improved. Laboratory test results revealed a white blood cell count of 5,600/mm^3 and a C-reactive protein level of 0.53 mg/dL. The CT scan showed alleviation of the hyper-attenuated adipose tissue and peritoneal hyperplasia. Levofloxacin and loxoprofen sodium administration were completed, and he was discharged on the eighth day of hospitalization.

On outpatient follow-up on the eighth day after discharge, he no longer reported abdominal pain, and the white blood cell count and C-reactive protein level were normalized at 8,300/mm^3 and 0.11 mg/dL, respectively. Abdominal CT scan revealed further improvement (Figure 1C).

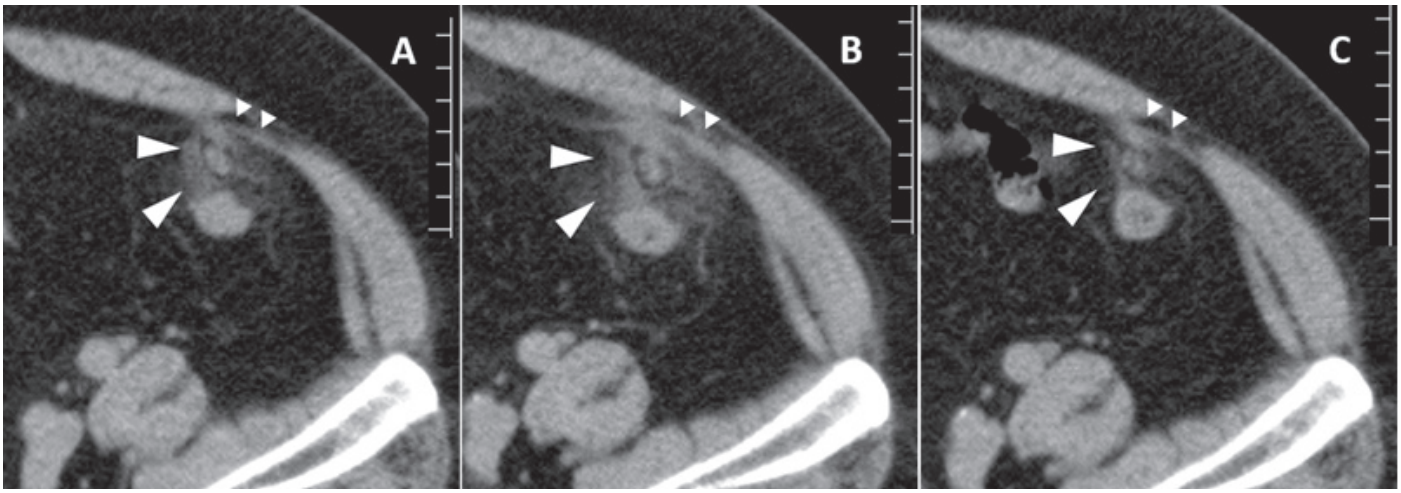

Figure 1: The panels showing non-contrast computed tomographic scans of the abdomen in a 69-year-old male with left lower quadrant pain. (A) A small lobular-shaped soft tissue density lesion surrounded by hyper-attenuated fat tissue (large arrowheads) is located in front of the sigmoid-descending colon junction (small arrowheads). Localized peritoneal thickening is also seen (small arrowheads). The lesion is pericolic; however, it is not adjacent to the colon. (B) Expansion of the hyper-attenuated adipose tissue (large arrowheads) and exacerbation of the peritoneal thickening (small arrowheads). (C) Reduction in the lesion density (large arrowheads) and improvement of peritoneal thickening (small arrowheads). One scale on the ruler represents 10 mm.

DISCUSSION

Epiploic appendages are pedunculated, leaf-like fat structures wrapped by serosa. The colon has approximately 50 to 100 and 2–5 cm long epiploic appendages. They protrude from the anterior and posterior portions of the large intestine along the tenia libera. Although their role is not precisely understood, they are believed to play a protective role as cushions, localized prevention against infection, and as energy reservoirs during starvation [4].

Epiploic appendages are nourished by small arteries and veins. Since they are pedunculated and rich in mobility, they are prone to ischemic infarction due to torsion. This is the reported pathomechanism of epiploic appendagitis [5]. However, secondary epiploic appendagitis is caused by the inflammation of adjacent organs, such as the colon diverticulum, gallbladder, and appendix [6]. Epiploic appendages exist along the entire length of the colon. Therefore, it is necessary to differentiate epiploic appendagitis from many other diseases. Diverticulitis, appendicitis, and omental infarction are often difficult to differentiate from epiploic appendagitis. Other potential diagnoses include tuberculous peritonitis, neoplasm, urachal cyst, mesenteric panniculitis, and trauma [7].

Symptoms of epiploic appendagitis include localized abdominal pain and rebound tenderness, sometimes with mild fever. In many cases, the white blood cell count remains normal or mildly increased. Choi et al. [8] examined 31 patients with epiploic appendagitis. They reported that abdominal tenderness was found in all cases, and rebound was found in 8 cases. The average body mass index was 25.9 kg/m^2. The diagnosis before the imaging examination was diverticulitis in 13 patients. On diagnostic CT scan, the epiploic appendage with inflammation is observed as an ovoid mass close to the colon. Intraperitoneal adipose tissue with hyperdensity due to inflammation surrounds the mass [6, 9, 10]. Symptoms are often relieved within a week with analgesic therapy alone [6]. There have been many reports that conservative treatment can be continued if typical CT findings are recognized [11, 12].

Schnedl et al. reported that 1,000 mg of ciprofloxacin daily was often used for epiploic appendagitis in their hospitals [13]. The Infectious Diseases Society of America guidelines for intra-abdominal infection provided recommendations for antibiotic therapy in 2010 [14]. According to the guidelines, adult patients with mild-to-moderate severity community-acquired infection should be treated with ticarcillin-clavulanate, cefoxitin, ertapenem, moxifloxacin, or tigecycline as single use. Cefazolin, cefuroxime, ceftriaxone, cefotaxime, levofloxacin, or ciprofloxacin are also preferable in combination with metronidazole. However, for adults with high-risk community-acquired infection, doripenem, meropenem, imipenem-cilastatin, piperacillin-tazobactam, ciprofloxacin, or levofloxacin accompanied by metronidazole should be chosen. In our case, a single intravenous infusion of levofloxacin was administered because secondary epiploic appendagitis due to diverticulitis could not be ruled out. Metronidazole was not available. Abadir et al. investigated 15 patients with omental infarction or epiploic appendagitis and reported that nine patients received antibiotics [15]. Six of 15 patients had peritoneal signs, and 12 patients were treated conservatively. Thus, there is no consensus on the role of antibiotics in the treatment of epiploic appendagitis.

Many studies have reported that CT scan is useful to rule out the need for surgical treatment [8, 16]. In our patient, antibacterial therapy and fasting were initiated on the first day of hospitalization, but his symptoms worsened on the third day of hospitalization. Secondary epiploic appendagitis and abscess formation were ruled out through subsequent CT scan. Mild diverticulitis was considered difficult to distinguish from PEA. Computed tomography scan showed that the lesion was a solid, lobular-like soft-tissue density mass, and it was not adjacent to the colon wall. On the basis of these characteristic findings, we diagnosed the lesion as PEA, not diverticulitis. In the case of diverticulitis, the interior of the lesion is not solid, and it has a three-layer structure. That is, there is a hyper-attenuated mucosal layer from the inside, thickened submucosa with a low density, and a large amount of serosa. Oral administration of non-steroidal anti-inflammatory drugs (NSAIDs) was initiated, and his symptoms subsequently improved. This indicates that NSAIDs may be more effective for the treatment of PEA than antibiotics. One study reported that diagnostic laparoscopy is required for patients without clinical improvement of diverticulitis [17]. However, we re-examined the CT scan instead of laparoscopy, and we confirmed that the diagnosis was PEA. Therefore, we decided to continue conservative management.

CONCLUSION

Primary epiploic appendagitis (PEA) is a self-limiting disease. To our knowledge, there are no other reports of patients with PEA with symptoms worsening after the administration of antibiotics alone. We consider this case valuable in terms of determining appropriate treatment for PEA. To alleviate the symptoms conservatively, a confirmed diagnosis by computed tomography scan and the administration of non-steroidal anti-inflammatory drugs from an early stage are necessary.

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Author Contributions

Masahide Hara – 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

Yumiko Ando – Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Kazuhide Tohara – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Extracorporeal membrane oxygenator venovenous in treatment of a fulminant varicella pneumonia in an adult

Rita Rei Neto, Sara Ferraz, Petra Monteiro, Margarida Correia, Carla Nogueira, Paula Castelões

ABSTRACT

Introduction: Varicella pneumonia is the most frequent and severe complication of varicella infection in adults, associated with a mortality of 30%. Case Report: A 46-year-old female with ulcerative colitis that starts fever, increasing dyspnea and a generalized rash with vesicles two days after admission. Laboratory tests showed renal impairment, acute hepatitis, a highly inflammatory state and chest radiography shows bilateral shadows with a rapid clinical deteriorating despite the anticipated treatment with antibiotic. 24-hours after the first symptoms, she develops an acute respiratory distress syndrome and was admitted to the intensive unit care and initiated protective lung ventilation. Despite acyclovir treatment and ventilator strategy, she maintained severe hypoxemia and need to started extracorporeal membrane oxygenator venovenous (ECMO-vv). The lavage fluid polymerase-chain reaction and vesicular liquid returned highly positive for varicella zoster virus (VZV) and completed treatment for seven days. The patient maintained ECMO-vv during nine days, was extubated after 13 days of mechanical ventilation, with all dysfunctions resolved. Conclusion: We can conclude that systemic varicella zoster virus infection, although rare, can cause serious and fatal complications. Early detection and proper treatment is mandatory for minimizing mortality and controlling infection, but often difficult because typical dermatological findings and the elevation of specific antibodies can be absent in immunosuppressed patients.

Keywords: Acute respiratory distress syndrome, Acyclovir, Pneumonia, Varicella

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INTRODUCTION

Varicella pneumonia is the most frequent and severe complication of varicella infection in adults [1–4]. Benign but highly contagious childhood disease, in adults is uncommon but potentially life-threatening [5], associated with an mortality of 30% even higher (50%) in those with mechanical ventilation [2, 3]. Transmission

occurs via respiratory droplets or direct contact with lesions [2]. Respiratory symptoms usually start 3–5 days after the onset of rash but in 10% precede cutaneous manifestations [3].

CASE REPORT

A 46-year-old female, non-smoking, admitted with a suspect of acute intestinal ischemia, confirmed by exploratory laparotomy without intestinal resection. Her medical history was significant for ulcerative colitis receiving 5 mg/day prednisolone at time of admission and had no history with contact with children recently. In second day after admission, patient starts fever, increasing dyspnea and a generalized rash with some vesicles. Laboratory tests showed renal impairment, acute hepatitis, a highly inflammatory state and chest radiography shows bilateral shadows. She starts antibiotic and intravenous acyclovir because of immunosuppression and rapid clinical deteriorating. Despite the anticipated treatment, she remains with hypoxemic insufficiency, was intubated and was admitted to the intensive unit care (APACHE II 20, SAPS II 38 and SOFA 11 at first day). Physical examination of the patient revealed systemic vesicular rash with lesions at various stage of progression and few crusts (Figures 1 and 2). 24-hours after the first symptoms, she remains hypoxemic, with bilateral infiltrates and under the diagnosis of acute respiratory distress syndrome (ARDS), she required treatment with protective lung ventilation. Despite treatment and ventilator strategy, she maintained severe hypoxemia and need to started extracorporeal membrane oxygenator venovenous (ECMO-vv). The polymerase-chain reaction in lavage fluid and vesicular liquid returned highly positive for varicella zoster virus (VZV). A hydrocortisone stress scheme was started because of chronic steroid treatment. Acyclovir was administered for seven days and the vesicles gradually formed clusters that were improved after 10 days. The patient maintained ECMO-vv during nine days, was extubated after 13 days of mechanical ventilation, with all dysfunctions resolved. She was transferred 16 days later and was discharged home at 21st day. Although his serum VZV-specific IgM levels were not elevated, the IgG levels were found to be elevated for three weeks.

DISCUSSION

Incidence of varicella pneumonia is one in 400 cases of varicella [1] and well recognized risk factors have been identified such as smoking, male gender, pregnancy, immunosuppression, pulmonary obstructive lung disease and the severity of rash (> 100 spots) [1, 2, 4]. Systemic VZV infection should be considered when an immunocompromised patient develops pulmonary symptoms, even in the absence of cutaneous lesions [3]. Varicella pneumonia can progress to hypoxemic acute insufficiency or ARDS [1, 2]. The pulmonary lesions caused by VZV consist in focal hemorrhagic necrosis, mononuclear infiltration of alveolar walls and fibrinous exudates [1, 4], causing an interstitial pneumonitis with alveolar hemorragae [1] and some bronchial vesicles can be found. Acyclovir has become standard therapy for patients with complications of Varicella infection and should be used early because reduces mortality [1].

The effect of corticosteroids in varicella pneumonia is unknown. In adult patients who progress to pneumonia or ARDS, corticosteroid therapy combined with acyclovir demonstrated a significant clinical response and had been shown to reduce the duration of hospital and ICU stay [2, 4]. It is possible that action of corticosteroids in varicella pneumonia is similar to that in pneumocystis pneumonia and military tuberculosis, both infections triggering T cell mediated responses [4]. The use of extracorporeal membrane oxygenation (ECMO) has been shown to be beneficial when patients have severe life-threatening refractory hypoxemia and they didn't respond

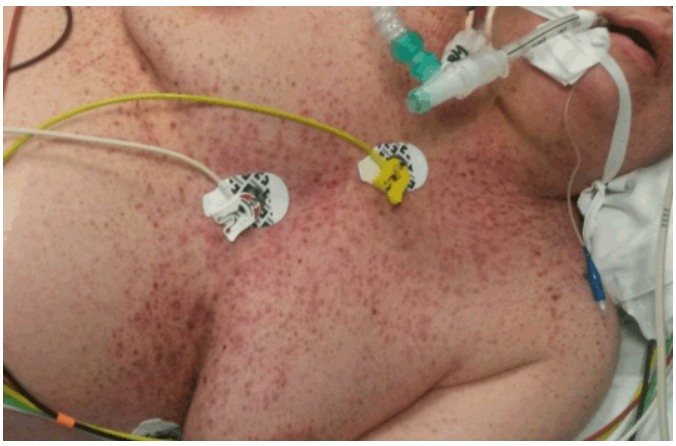

Figure 1: Systemic vesicular rash with lesions at various stage of progression and few crusts.

Figure 2: Bilateral infiltrates in thoracic X-ray.

to conventional rescue therapies, such as recruitment maneuvers, inhaled nitric oxide, prone positioning, high frequency oscillatory ventilation or extra-corporeal membrane oxygenation (ECMO) [1, 6].

Since the H1N1 influenza A pandemic in 2009 we saw a worldwide expansion in the use of ECMO. Several studies have reported that ECMO may improve survival in severe ARDS [6] but there have been few studies to review long-term survival and quality of life of this patients. In this case, our patient was a survival that not have profound muscle weakness and wasting because she starts early rehabilitation during the ICU period and she was discharge from ECMO as soon as possible.

A long-term morbidity shows that varicella pneumonia had effect on respiratory function, with a temporary reduction in forced vital capacity and forced expiratory volume in one second and may be associated with a restrictive lung disease pattern in the acute and recovery phase [1]. The prevention with immunoglobulin has proved to be effective and its use is recommended for all adults who have no evidence of immunity (history of chickenpox, being born before 1980 or having positive titers) and should be emphasized for those who come in contact with patients at high risk of severe disease and in individuals with a high risk of personal exposure [7] and to susceptible immunosuppressed patients and pregnant women who have been in contact with a proven source of Varicella infection [1].

CONCLUSION

Systemic varicella zoster virus (VZV) infection, although rare, can cause serious and fatal complications. Early detection and proper treatment is mandatory for minimizing mortality and controlling infection, but often difficult because typical dermatological findings and the elevation of specific antibodies can be absent in immunosuppressed patients. Varicella zoster virus prophylaxis may be required in some of immunosuppressed ICU patients to prevent fatal VZV infection.

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Author Contributions

Rita Rei Neto – Substantial contributions to conception and design, Acquisiton of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sara Ferraz – 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
Petra Monteiro – 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
Margarida Correia – 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
Carla Nogueira – 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
Paula Castelões – 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

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Penile strangulation due to a metal ring: A case report

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ABSTRACT

Introduction: Penile strangulation is a condition in which a foreign material compresses the penis, resulting in penile enlargement due to a circulatory disturbance. Case Report: A 68-year-old male patient was referred to our hospital in order to undergo the removal of a metal ring that had strangled his penis. The ring was successfully removed using an airtime cutter. His penis showed no ischemic lesions at one week after removal. Conclusion: We herein report a rare case of penile strangulation.

Keywords: Strangulation, Incarceration, Penile

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INTRODUCTION

Penile strangulation is a condition in which a foreign material compresses the penis, resulting in penile enlargement due to a circulatory disturbance [1]. We herein describe a case of penile strangulation due to a metal ring that the patient had worn to the reduce pain of varicocele.

CASE REPORT

A 68-year-old male was referred to our hospital with an enlargement of his penis due to a metal ring. He had been inserting his penis into a metal ring to reduce the pain associated with varicocele for ten years. Three hours before his initial visit, he could not remove this ring and experienced difficulty in urination (Figure 1). To treat his condition, the patient was referred to our hospital at six

Figure 1: Penile strangulation by a metal ring.

hours after penile strangulation. We initially attempted to remove the ring using a ring cutter and wire cutter; however, this failed to cut the ring. We then asked a dental surgeon to cut the ring using an airtime cutter. The ring was successfully cut and removed from his penis (Figures 2 and 3).

Figure 2: The metal ring after removal.

Figure 3: One week after removal.

DISCUSSION

Penile strangulation is caused when an object compresses the penis. In most cases, the removal of the foreign body is difficult because of priapism in the external penis. Penile strangulation represents an emergency disease, as the compression of the penis for an extended period of time interrupts the blood flow, resulting in penile ischemia [1].

In previous reports, the strangled penile body was divided by soft and hard materials. Rubber and vinyl have been reported as soft materials, while metal rings, metallic pipes, and the tops of PET bottles have been reported as hard materials. Soft materials compress the skin deeply, with ulceration occurring in severe cases. Hard materials first lead to edema rather than ulceration. The duration of strangulation in the previously reported cases has ranged from one hour to ten years [1].

In the present case, the patient had strangled his penis in order to reduce the pain associated with varicocele. Previous reports showed that the reasons for penile strangulation were divided into non-sexual and sexual purposes. The sexual purposes have included (but are not limited to) improving energy, extending erection time, and enlarging the penis [2]. The duration of strangulation varied according to the reasons. The patients in whom penile strangulation occurred due to non-sexual reasons presented to hospital earlier than the patients in whom the purpose was sexual [3]. In this case, the patient performed penile strangulation for a therapeutic reason. The patient presented to a hospital in the early stage of penile strangulation, after experiencing difficult urination. We have to remind that inappropriate usage of foreign bodies resulting in penile strangulation.

The removal of the object strangling the penis is the most important aspect of treatment in cases of penile strangulation. A previous report suggested that the object can be removed at up to 12 hours without any complications [1]. In our case, the rapid removal might have contributed to the lack of any complications. Instruments that have been used in removal have included pincers, a plaster cutter, a hammer, and a grinder [4–6]. However, air turbines for dental use are most frequently used to remove such objects [5]. The top of the air turbine is coated with diamonds and spins at a rate of 400,000 times per minute, which gives it a high cutting ability. Water can be poured to reduce the frictional heat and improve the safety of the procedure—as we did in our case [7]. When standard procedures including the use of a ring cutter fail to cut the object, the use of an air turbine with the assistance of a dental surgeon is sometimes necessary [8]. Followed-up is needed to confirm no ischemia after removal of metallic ring.

CONCLUSION

We herein describe a case of penile strangulation due to a metal ring that the patient had worn to the reduce pain of varicocele. The removal of the object strangling the penis is the most important aspect of treatment in cases of penile strangulation.

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Author Contributions

Kentaro Matsumiya – 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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The corresponding author is the guarantor of submission.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Incarcerated gravid uterus in a rectal prolapse: A case report

El-Nafaty Aliyu Usman, Obiano Sunday Kelvin, Mamman Tijjani Hinna, Rabiu Amina Baba, Ningi Adamu Bala, Farouk Halima Usman

ABSTRACT

Uterine retroversion is relatively common in 15% of women and rarely calls for concern. Such women will normally conceive and remain symptomless, as the pregnancy advances the uterus rotates spontaneously between 14–16 weeks to an anteverted position, thus allowing the growing uterus to expand into the abdomen. However, if the uterus become entrapped in the hallow of the sacrum it is then incarcerated, such uterus fails to return to anterior position. This usually presents as an emergency and requires immediate intervention to reposition the uterus. The risk factors include, deep sacral concavity, pelvic adhesions, uterine malformations, and pelvic tumors. Rectal prolapse is commonly seen in the children and the elderly but can occur at all ages. The risk factors include chronic constipation, severe or chronic cough, and pelvic floor dysfunction and pregnancy a contributory factor. The combination of prolapse of an incarcerated uterus in a rectal prolapse is rare and only one case reported in literature. This is a report of the second case of prolapse of an incarcerated gravid uterus in a rectal prolapse occurred in a 25-year-old multiparous. The prolapse incarcerated uterus with the rectum was reduced through laparotomy and an encirclement suture applied at the anal mucocutaneous margin. The pregnancy was carried to term and delivered via spontaneous vaginal delivery of alive female weighing 2.8 kg. There was also no recurrence of the rectal prolapse.

Keywords: Incarcerated uterus, Live birth, Rectal prolapse, Thiersch suture

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INTRODUCTION

Uterine retroversion is common phenomenon and considered as normal variation in women with 15% incidence rate. Such women will normally conceive and remain symptomless, as the pregnancy advances the uterus rotates spontaneously between 14–16 weeks to an anteverted position thus allowing the growing uterus to expand into the abdomen. However, in event the uterus fails to antevert and rise into abdominal cavity, it gets entrapped in the hollow of the sacrum and become incarcerated. This condition occurs in about 1:3,000–10,000 pregnancies [1]. The risk factors include, deep

sacral concavity, pelvic adhesions, uterine malformations, and pelvic tumors [1–4]. Patients in this situation often presents as an emergency with acute urinary retention, pelvic pain, back pain and constipation. The treatment requires urgent intervention to dislodge and reposition the uterus [2, 3].

Rectal prolapse is the protrusion of the rectum through the anus, a prolapse can be partial or complete, in the former the mucosal lining of the rectum bulges partly from the anus while a complete prolapse the entire rectum bulges through the anus. It is usually associated with weak pelvic floor musculature, increased intra-abdominal pressure, chronic constipation, lax muscles of the anal sphincter and pregnancy a contributory factor. It is commonly seen in the elderly and children but can occur in all ages. The presentation is usually protrusion after a bowel motion which retracts spontaneously, later protrudes more often, especially with straining, sneezing, etc. finally the rectum prolapses with daily activities such as walking which may progress to continual prolapse and patients may have to replace it manually [5, 6]. Rectal prolapse has been reported to occur in pregnancy [7, 8].

The occurrence of combined incarcerated gravid uterus with rectal prolapse is a rarity and we report a second case of incarcerated gravid uterus inside a massive rectal prolapse in a young lady.

CASE REPORT

A 25-year-old gravida 5 and para 4 with 2 live children (G5P4+0, 2 alive), presented to the gynecology emergency unit of Federal Teaching Hospital Gombe, with complaints of four months amenorrhea and sudden protrusion per rectum. The patient was pounding grains in a mortar when suddenly she felt like defecating, on straining to empty her bowels in the lavatory she suddenly felt the gush of protrusion per anus. She tried to push it back but with no success. She was then rush to a secondary health center where the protrusion was dressed with gauze and subsequently referred to our department.

The past history revealed she had been experiencing the protrusion per rectum since early childhood each time she goes to defecate it used to come out and retreat spontaneously, but since past four months she had to manually reduce the protrusion each time she goes to empty her bowels. There was no associated urinary symptoms or chronic cough but had recurrent episodes of constipation. She had never been to the hospital because of this protrusion.

Obstetrics history, she had four full term pregnancies all delivered spontaneous at home, the pregnancies where not supervised, the fourth delivery was a stillbirth, while the second child died at four years of age due to febrile ailment.

On examination she was in painful distress, afebrile and mild pallor. The vital signs were normal blood pressure 120/80 mmHg, pulse 96 beats/min.

The abdomen was scaphoid soft and non-tender, the bladder not distended, the uterus was not palpable. The other abdominal organs were un-remarkable. On pelvic examination, there was a massive reddish mass, edematous about 20 cm in diameter, in the perineum protruding through the Anus (Figure 1). With oozing of blood from the mucous surface. On digital vaginal examination revealed the cervix was pointing upwards about 4 cm long with OS closed. The fundus was displaced posteriorly deep into the prolapsed rectal swelling.

An ultrasound scan examination with the transducer on the swelling, confirmed a gravid uterus containing a live single active fetus with good cardiac activity and a calculated gestation age of 15 weeks.

Management an intravenous fluids 5% dextrose in water (D/S) 500 ml was set up, urgent pack cell volume (PCV) was 32%. Blood grouping and typing of 2 pints was done. Electrolytes were normal. Analgesia was given and the patient was counsel on the need for immediate intervention and consent for surgery obtained. The risk to the possible loss of the pregnancy was explained.

Procedure under general anesthesia, examination under anesthesia (EUA) was done and the findings confirmed as earlier noted. A manual reduction was attempted but did not succeed. Therefore, a double approach was used with the general surgeons assisting at the perineum. The abdomen was entered through a Pfannenstiel incision, and manually the fundus of the uterus was grasped and with the aid of pressure from the perineum it was dislodged and delivered into the abdomen (Figure 2). It was inspected and was found to be normal, with the tubes and ovaries. The abdomen was closed normally.

A Thiersch procedure was used to apply an encirclement suture round the anus with Nylon 2 to prevent prolapse of the rectum (Figure 3). The postoperative recovery was uneventful, she had analgesics, antibiotics, and Tocolytics and stool softeners. An ultrasound scan on the fifth day postoperative revealed a live fetus with active cardiac activity. She was subsequently counsel and booked for antenatal care. The pregnancy was carried to term uneventfully and she had a spontaneous vaginal delivery of alive female weighing 2.8 kg.

On examination of the perineum six weeks postpartum it was found to be intact the Nylon suture was in place and there was no recurrence of the rectal prolapse and she has been defecating normally (Figure 4), the uterus was in the anteverted position.

DISCUSSION

Uterine retroflexion and retroversion are common occurrence in women and rarely associated with any symptom. Majority will have seamless pregnancy with spontaneous version to the anterior position as the pregnancy advances. However, when spontaneous rotation fails to occur and the uterus gets trapped in the

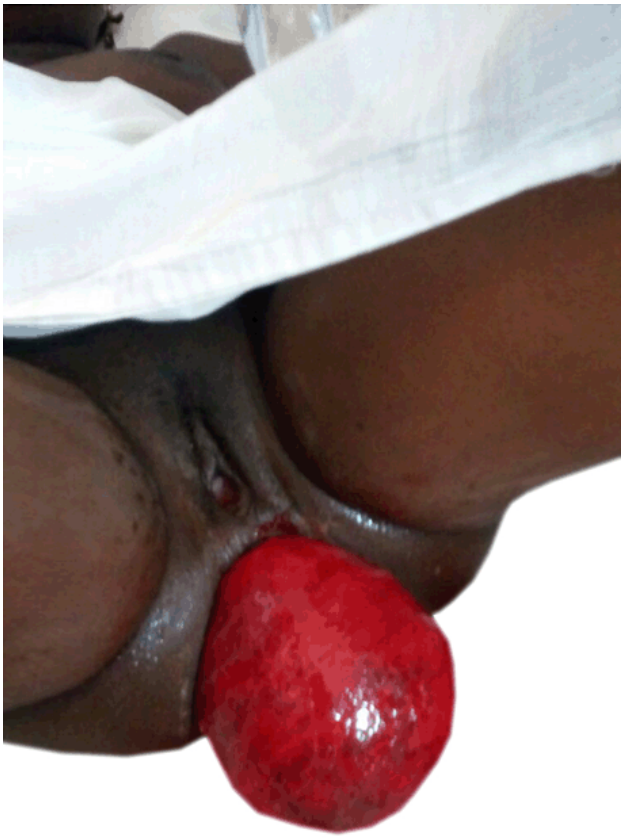

Figure 1: Rectal prolapse with Incarcerated uterus.

Figure 2: The dilated Anus after reduction of the prolapse.

Figure 3: After application of the Thiesch suture.

Figure 4: Six weeks postpartum.

hallow of sacrum incarceration occurs. Such patients present with acute urinary retention and severe pelvic pains. Immediate intervention is usually required to relief pain by repositioning the uterus [9, 10]. In this case the patient presented because of the prolapse containing the incarceration uterus.

The failure of the patient to seek medical attention for the rectal prolapse since childhood was a clear case of ignorance, even when symptoms worsen during the index pregnancy, that she had to manually reduce it on some occasions still did not make her report to the hospital. This contributed to the occurrence of this complication.

The combination of incarceration and rectal prolapse in pregnancy is rare and only one case reported in literature [11]. The presentation is usually as emergency because of the failure of the prolapse rectum and the uterus to reduce spontaneous or manually. A weak pelvic floor with a lax anterior rectal wall and anal sphincters provides as enabling room for the retroverted uterus to expand and push into the rectum which provokes undue desire to defecate and may carry the uterus along with straining to empty the bowels.

Failure to intervene will result in more complications with risk of losing the fetus, strangulation of the uterus and the rectum. In the first case, reported, manual reduction was done under anesthesia but the patient aborted few days later, undue handling could have contributed in the loss of that pregnancy. Therefore, we opted to do laparotomy and gently reduce the incarcerated prolapse uterus and rectum. This was done with aim to save the pregnancy, which actually allowed the pregnancy to be carried to term. The rectal prolapse was managed with Thiersch procedure which by applying an encirclement of Nylon suture at the anal margin prevented further rectal prolapse during the pregnancy and after. This procedure is simple to perform and associated with limited complications. Other more invasive procedures may not be suitable in a pregnancy state e.g., laparoscopic rectopexy [12], anticipated complications include recurrence of prolapse, constipation and mucosal ulceration, none of these occurred in this case. Recurrence of incarceration has been reported [13].

CONCLUSION

In this case, with a rectal prolapse predating the pregnancy means there was an existing factor that's favors the prolapse of the uterus with rectum. Therefore, obstetricians should be aware of the risk of having uterine prolapse in patients who present with symptoms of rectal prolapse they should be examined to determine if there is retroverted uterus, the risk of combine incarceration and prolapse should be born in mind. The conservative treatment with Thiersch suture has been found useful in this case and vaginal delivery uneventful.

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Author Contributions

El-Nafaty Aliyu Usman – 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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Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

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CLINICAL IMAGE PEER REVIEWED | OPEN ACCESS

Purpura fulminans: A rare presentation of antiphospholipid syndrome

Ahmed S. Mahmood, Noor Q. Omar, Sudheer Chauhan, Jose Cervantes

CASE REPORT

A 72-year-old male with no significant past medical history who was admitted with worsening bilateral lower extremity discoloration and swelling for three days. He complained of intermittent chest pain and generalized fatigue, but denied dyspnea, fever, weight loss, trauma or any episodes of bleeding. Physical examination was remarkable for non-blanching and non-tender erythematous rash with branched configuration extending up to the knees associated with cold and cyanotic toes but palpable distal pulses bilaterally. There was 1+ bilateral pitting edema extending up to the knees. Investigations (Table 1) were significant for neutrophilic leukocytosis, normocytic anemia and thrombocytopenia. Coagulation profile was unremarkable. D-dimer and fibrinogen levels were noted to be elevated. Renal and hepatic functions were within normal limits and there were no electrolytes or metabolic derangements. Deep vein thrombosis (DVT) studies and computed tomography (CT) pulmonary angiography failed to show large vessel thromboembolic phenomena. Immunology testing revealed positive RF and mildly reduced C4. Human immunodeficiency virus (HIV), mycoplasma, anti-nuclear antibody (ANA), C3, and anti-neutrophil cytoplasmic antibody (ANCA) serologies were negative. Ankle brachial index and peripheral vascular resistance were normal. Patient was started on prednisone 1 mg/kg and broad spectrum antibiotics on the first day of hospitalization to cover for septic versus immunologic phenomena. Blood cultures were negative and antibiotics were discontinued accordingly. Patient's condition was deteriorating evident by worsening skin necrosis and formation of widespread hemorrhagic blisters and ecchymosis (Figures 1 and 2). Platelet count further dropped. Patient remained hemodynamically stable during the course; however, he was transferred to medical ICU for close monitoring. Intravenous immunoglobulins (IVIG) and heparin infusion were initiated, and he was switched to high dose methylprednisolone intravenously. Serology testing revealed elevated phosphatidylserine IgA, IgM and IgG, and anticardiolipin (aCL) IgM levels. Serology testing for cryoglobulinemia and systemic lupus erythematosus (SLE) was negative. The diagnosis of idiopathic antiphospholipid syndrome was made based on the clinical picture and the positive serology heparin infusion was started. During the hospital course, patient's symptoms significantly improved. Platelet count trended up and skin necrotic changes started to resolve. Intravenous immunoglobulins and intravenous steroids were discontinued. The patient was started on warfarin and discharged to nursing home after a total duration of two weeks in hospital.

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DISCUSSION

Antiphospholipid syndrome is an autoimmune multisystem disorder characterized by arterial, venous, or small vessel thromboembolic events and/or pregnancy complications in the presence of persistent antiphospholipid antibodies (aPLs) [1]. Presenting symptoms typically include blood clots, stroke, peripheral arterial thrombosis, or repeat miscarriages [2]. The development of life-threatening acute retiform and widespread purpuric lesions (purpura fulminans) at the time of presentation has been rarely reported [3, 4]. The mainstay of treatment for antiphospholipid syndrome

Table 1: Lab results

Test	Result	Reference Range
White blood cell count	13.1x10³/µL	4.8–10.8x10³/µL
Hemoglobin	12.6 g/dL	14.0–18.0 g/dL
HCT	38.3 %	42–52 %
MCV	85.7 fL	80–94 fL
MPV	10.9 fL	7.2–10.4 fL
Platelet	65 K/µL	130–400x10³/µL
Neutrophils Auto.	87 %	44–80%
Lymphocytes Auto.	3.4 %	13–43 %
Monocytes Auto.	9.3 %	2.0–15 %
Eosinophils Auto.	0.1 %	0.0–3.0 %
Basophils Auto.	0.2 %	0.0–3.0 %
PT	26 secs	10.1–13.0 secs
PTT	26 secs	26.6–34.2 secs
INR	1.2	0.9–1.1
D–dimer	6500 ng/mL	45–500 ng/mL
Fibrinogen	930 mg/dL	230–458 mg/dL
BUN	30 mg/dL	9–20 mg/dL
Creatinine	0.7 mg/dL	0.7–1.3 mg/dL
Sodium	136 mEq/L	137–145 mEq/L
Potassium	3.8 mEq/L	3.5–5.1 mEq/L
CO2	26 mEq/L	22–30 mEq/L
Troponin	< 0.012 ng/mL	< 0.034 ng/mL
Creatine Kinase (CPK)	23 U/L	55–170 U/L
Rheumatoid Factor	80 IU/mL	< 15 IU/mL
C4	13.5 mg/dL	16–48 mg/dL
Anticardiolipin IgM	15 MPL	< 12 MPL
Phosphatidylserine IgM	49 U/mL	< 25 U/mL
Phosphatidylserine IgG	24 U/mL	< 10 U/mL
Phosphatidylserine IgA	45 U/mL	< 20 U/mL

Figure 1: (A) Necrotic, hemorrhagic and tense bullae noted on the dorsum of the right foot, and (B) Necrotic, hemorrhagic and tense bullae noted on the dorsum of the right foot.

Figure 2: Ecchymotic changes noted on the planter surface of right foot.

includes the following antithrombotic medication: heparin, warfarin and aspirin [5–8].

CONCLUSION

Antiphospholipid syndrome is a rare but potentially life threatening disease (especially catastrophic antiphospholipid syndrome). It should be suspected in patients with unexplained skin necrosis and thrombocytopenia, and those presenting with purpura fulminans, when no apparent etiology can be found. Prompt treatment with steroids, intravenous immunoglobulins and anticoagulation can be lifesaving.

Keywords: Anticoagulation, Antiphospholipid syndrome, Purpura fulminans, Thrombocytopenia

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Ahmed S. Mahmood – 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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Intramyocardial calcification in a 37-year-old patient with severe aortic stenosis

Aoife M. Granahan, Katie E. O' Sullivan, Sarah A. Early

CASE REPORT

A 37-year-old male with symptoms of progressive dyspnea and intermittent chest pain over a one-year period with a longstanding history of aortic stenosis was referred for aortic valve replacement. His personal cardiovascular history was unremarkable, most notably for tuberculosis, rheumatic heart disease, pericarditis or myocardial ischemia. Family history was non-contributory. Examination revealed an ejection systolic murmur consistent with aortic stenosis. Vital signs revealed an early warning score of zero, laboratory investigations were found within normal parameters and electrocardiogram (ECG) was in sinus rhythm. Echocardiography (ECHO) demonstrated an ejection fraction of 55% and confirmed severe aortic stenosis and moderate aortic incompetence with peak and mean gradients of 90 and 50 mmHg respectively. Extensive myocardial calcification on ECHO (Figure 1) and coronary angiography (Figure 2) prompted further investigation with computed tomography scan. Computed tomography scan was notable for a thick band of intramyocardial calcification extending from beneath the left coronary cusp to half way down the interventricular septum (Figure 3). Calcification was not noted in any other organ system and there was no evidence of malignancy. Serum calcium levels were within normal range. Cardiac catheterization revealed no evidence of coronary artery disease.

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Aortic valve replacement was performed. Intraoperative findings revealed a heavily calcified aortic valve. The native valve was meticulously excised and replaced using a 25-mm carbomedics mechanical prosthesis with interrupted sutures. Postoperative echocardiogram was satisfactory and postoperative recovery was uneventful.

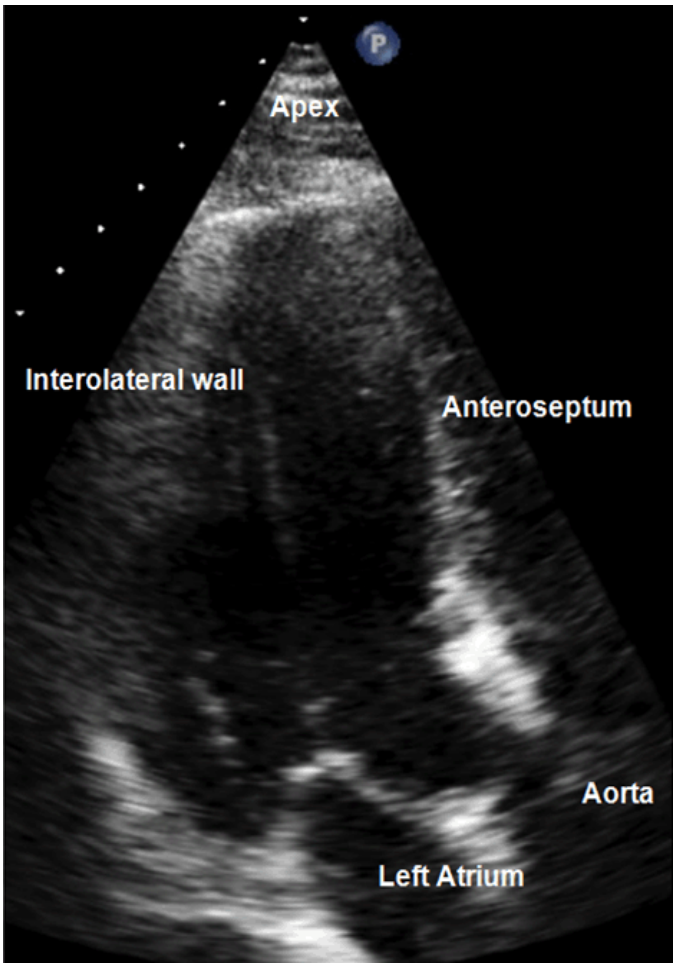

Figure 1: Preoperative transthoracic echocardiogram demonstrating abnormal calcification in the interventricular septum.

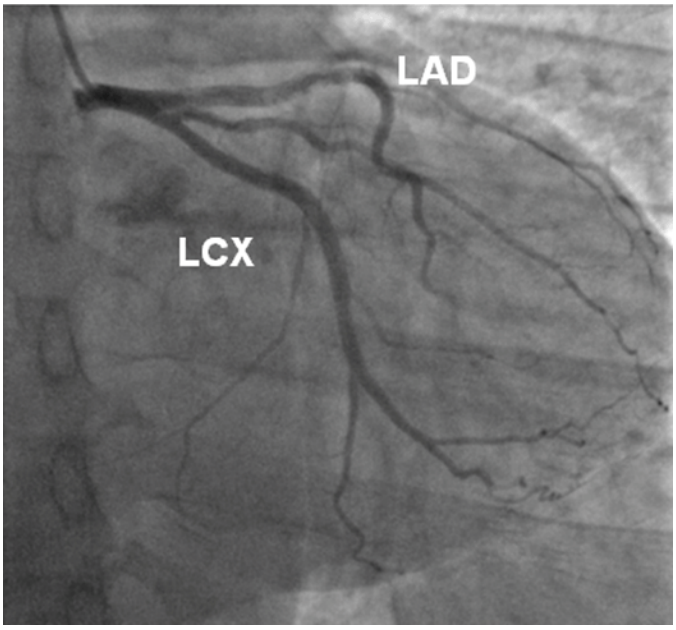

Figure 2: Coronary angiogram demonstrating intramyocardial calcification relative to the left anterior descending (LAD) and left circumflex (LCX) arteries.

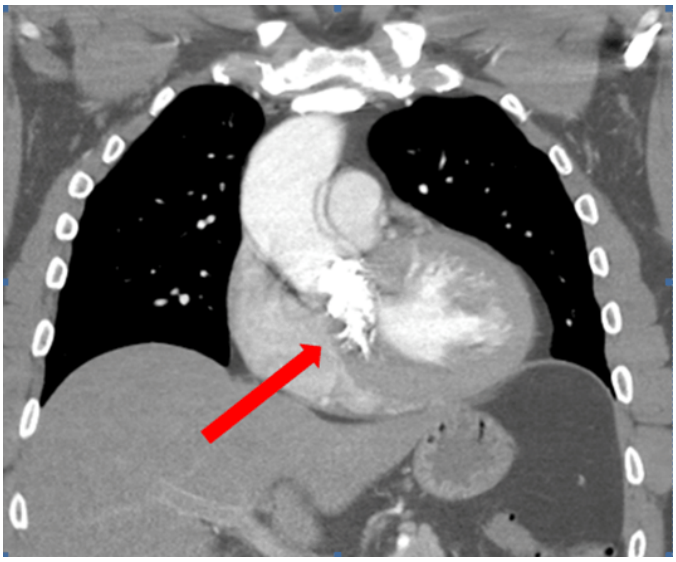

Figure 3: Computed tomographic thoracic angiogram (coronal section) demonstrating aberrant calcification extending from the aortic valve to the interventricular septum.

DISCUSSION

Epidemiological studies reveal that aortic stenosis increases with age, 25% of people aged over 65 years have aortic sclerosis, and 3% over 75 years have severe stenosis. Approximately, 16% of patients with sclerosis progress to stenosis in seven years [1]. The overwhelming cause of aortic stenosis is calcific degeneration, 2% of the population account for congenital bicuspid valves, rheumatic disease is now extremely rare [2]. Cases of myocardial calcification which are not associated with myocardial infarction or metastatic deposition are extremely rare [3, 4]. The calcification of myocardium can occur secondary to two mechanisms. Firstly, dystrophic calcification occurs in dead or degenerative tissue in the presence of normal calcium/phosphate balance such as previous myocardial infarction. Other potential causes of dystrophic calcification include myocarditis, ventricular aneurysms, tuberculosis, sarcoidosis and hemorrhage [5–9]. Secondly, metastatic calcification, which occurs when a derangement of calcium phosphate metabolism results in calcium deposition in normal tissue, such as chronic renal failure or hyperparathyroidism [3]. Our patient had no known history or aberrant biochemical analyses to suggest that any of the differentials mentioned were the causative pathology. Extensive calcification and porcelain heart due to endomyocardial fibrosis has also been reported in literature [10]. Endomyocardial fibrosis is characterized by the presence of fibrous tissue in the endocardium eventually extending into the myocardium, a finding not identified in our patient. Mitral annular and leaflet calcification occurs frequently with degenerative aortic stenosis and is known to reduce leaflet opening and result in significant mitral stenosis in 25% of patients with calcific aortic stenosis [11]. In our patient, however, no involvement of the mitral valve was noted.

CONCLUSION

This is an unusual case as significant calcification of the interventricular septum developed in the absence of any known etiologic factor or evidence of pre-existing myocardial injury. Valvular heart disease, is an underappreciated yet serious and growing public health problem.

Keywords: Aortic stenosis, Aortic valve replacement, Cardiothoracic surgery, Intramyocardial calcification, Symptomatic valvular disease

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Author Contributions

Aoife M. Granahan – 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
Katie E. O' Sullivan – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Sarah A. Early – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CLINICAL IMAGE PEER REVIEWED | OPEN ACCESS

Lower limb cellulitis associated with ski boot compression trauma

Claire Marie Murphy, Mark Conal Murphy

CASE REPORT

A 77-year-old male was admitted from the emergency department with left lower leg cellulitis failing to respond to oral penicillin V and flucloxacillin. There was no history of previous cellulitis, diabetes, venous disease or surgery to the leg. On physical examination the legs were red, warm to touch, edematous and the skin was intact and very dry. Likely sources of entry were either the dry flaking skin or a small ooze from a problematic in-growing toenail but which on routine culture was negative for pathogens. The C-reactive protein was raised at 165 mg/L as was his white blood cell count $11.7x10^9$/L, (72% neutrophils) but he had no fever. There was no observable lymphadenopathy or tracking lymphangitis. The C-reactive protein settled to 39 mg/L with four days of intravenous flucloxacillin and benzyl penicillin and he was discharged home on oral flucloxacillin after five days.

The patient required readmission within 48 hours for worsening symptoms of cellulitis although the C-reactive protein was 29 mg/L, (Figures 1 and 2). Doppler ultrasound showed no venous thrombosis. Venous duplex scan showed incompetence at the saphenofemoral junction and the great saphenous vein bilaterally and at the saphenopopliteal junction and the small saphenous vein on the right with normal arterial flow and patent deep veins. Antibiotics were changed to clindamycin and vancomycin but clinical improvement was very slow (Figure 3), and although a carrier site screen was negative for methicillin resistant *Staphylococcus aureus* the antibiotics were escalated to linezolid. He was not

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Figure 1: Lower leg cellulitis, day-7, medial view.

Figure 2: Lower leg cellulitis, day-7, lateral view.

Figure 3: Lower leg cellulitis, day-14, lateral view.

known to have diabetes but he had a body mass index of 40 kg/m^2, weighed 111 kg and had a high fasting sugar of 8–9 mmol/L with a HbA1c of 46 mmol/mol in keeping with a pre-diabetic state. Magnetic resonance imaging scan revealed edema over the lateral lower leg but no drainable collection. He was discharged 23 days after the initial admission with a C-reactive protein of 17 mg/L and WBC of 8.5x10^3/L.

At follow-up one year later his legs appeared normal on inspection, were not inflamed but on palpation there was a marked temperature difference in a lateral and distal 7x5 cm approximate area just above the ankle joint which was markedly cooler that the rest of the lower legs. Also present was definite edema demonstrated by finger pressure but also restricted to the same cooler lateral area of the calf without any more general or foot edema. Maximum calf diameter was 17 cm.

Unusual in the history was a prolonged compression injury to his calves from over tightened ski boots. He said he felt his lower legs were never normal since an event 20 years previously when he had skied all day in pain, without a break, from boots overtightened by an external screw control mechanism. The boot design was contemporary to 20 years ago and semi rear entry with wiring which could be tightened by an external screw control mechanism. The patient described deliberately overtightening the boots in the hope of achieving better ski control and he had persisted skiing despite considerable pain throughout the day.

DISCUSSION

As there was no clinical lymphoedema evident on presentation, other than a general lower leg swelling, lymphoscintigraphy was not performed although it has been shown that lymphatic abnormalities are commonly present in lower leg cellulitis without clinical signs of edema [1]. Conditions that reduce the circulation of blood in the veins or that reduce circulation of the lymphatic fluid (such as venous insufficiency, obesity, pregnancy, or surgeries) may also increase the risk of developing cellulitis, although different studies have given conflicting risk factors [2, 3]. Possible risk factors in this patient were obesity, pre-diabetes, superficial venous insufficiency, dry skin, and a slight ooze from an ingrowing toenail. Halpern et al. in a single centre case control study of 150 cases identified local risk factors to be of prime importance rather than systemic risk factors and also identified blunt injury as a significant risk factor. This patient reported a specific and very clearly remembered painful history of compression trauma with consequent residual abnormal signs in this lateral aspect of his lower leg which have never been described.

Most ski boots are designed to fit a 13–14 inch calf and this patient had a 17 inch calf which may have predisposed him to tight fitting boots [4]. The importance of good fitting ski boots is better recognized today and the skier with wider calves can now be accommodated with boots with lower cuffs and insoles with a heel lift. Also custom liners can be professionally made for a more personal fit and boots can have upper cuff adjustments to fit a wide calf.

Boot top fractures have also been recognized as a risk from overtightened boots and safety release designs but are less common today due to upward as well as rotational binding release design [5, 6]. It is likely this sustained compression trauma permanently damaged the superficial venous/lymphatic system leading to the tendency towards recurrent cellulitis. This presentation may have been more serious on this occasion by a pre-diabetic state which may have further compromised recovery.

CONCLUSION

We believe this case of lower leg cellulitis is the first described to identify a new risk factor for cellulitis with a chronic persistent abnormality attributable to ski boot sustained compression trauma 20 years previously.

Keywords: Cellulitis, Lower limb, Ski boot compression trauma, Tight fitting boots

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Author Contributions

Claire Marie Murphy – 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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Guarantor

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