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Cardiac scintigraphy allows both accurate localization and graduation of the intensity of radio-tracer retention: high uptake on apical and antero-apical walls (black arrow) and moderate on antero-medial-septal wall (white arrow).

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Therapy for small-cell lung cancer: When the past meets the future

Giovanni Leuzzi

Despite the incidence of small-cell lung cancer (SCLC) has decreased over the years to approximately 10–14% of all lung cancers [1], this tumor still represents a challenge for most physicians due to its rapidly-progressive behavior. As a rule, the outcome is relatively poor in those cases receiving oncological therapy with a median survival of 16–17 months and a 5-year overall survival (OS) of 10% [2], even worse in SCLC patients not receiving any kind of therapy (5-year OS of 5%) [3].

Contrary to that reported for non-small-cell Lung cancer (NSCLC), the quest for the optimal strategy in the setting of limited-stage SCLC has had a rapid rise from the 60s until the 80s and then had a stalemate until today. In fact, in the past some authors reported acceptable outcomes in selected cases undergoing surgery with radical intent with a 5-year OS of 25% [4]. Given these results, although the decision on whether administering chemo (CT) or surgery depended on the surgeons’ or oncologists’ experience, most physicians have recommended the use of surgery in those patients without metastases or bulky mediastinal disease. Subsequently, the “gold rush” was interrupted in the 90s when the Lung Cancer Study Group (LCSG) [5] reported the results of a randomized controlled trial comparing surgery with non-surgical treatment in early-stage SCLC. This study comprised of 146 patients who were randomized for surgery or for no further treatment after the administration of CT and chest radiotherapy (RT). The final results revealed comparable results with a 2-year OS of 20% in both study arms. The main limitations to this study were several, mainly the heterogeneous cohort (composed by patients either with or without nodal involvement) and the chemotherapeutic regimen (different from the current protocol with platinum and etoposide). Despite these biases, this study ratified the “banishment” of surgery for many years and the National Comprehensive Cancer Network (NCCN) and the American College of Chest Physicians (ACCP) have “prohibited” the use of surgery until some years ago, even in case of resectable and node-negative disease.

Although this “prohibition” was widely accepted, in literature some authors have simultaneously explored the role of surgery even in case of locally-advanced SCLC. A recent analysis on 2476 performed on the National Cancer Database (NCDB) [3] revealed acceptable 5-year OS rates with 49%, 32% and 27%, respectively, for p-stage I, II and III. In particular, this study evidenced a significant outcome benefit in surgically-treated N2 SCLC compared to non-surgical IIIA stage (5-year OS 18% vs 12%, respectively). Similarly, Takei et al. analyzed 243 patients reporting a 5-year OS of 52.6% for all resected SCLC [6]. In addition, the definition of the seventh TNM staging system has been based on the analysis of 349 SCLC patients undergoing surgical resection and suggested the utility of surgery in node-positive cases as well, given the 1-year and 5-year survival rates of resected N1 and N2 disease were 74% and 33%, and 54% and 6%, respectively [7].

Considering these interesting outcomes, early-stage SCLC patients undergoing surgery have been associated with improved outcome compared to non-surgical ones. As a result, the NCCN and ACCP have recently changed direction and would seem to be returned back to the starting point. In fact, the more recent guidelines recommend surgical resection (over non-surgical treatment) followed by platinum-based adjuvant CT only for clinical stage I SCLC [8]. However, when nodal involvement is evidenced at preoperative workup, surgery should be avoided.

Regarding the nodal involvement, two issues are actually debated. Firstly, the prognostic impact of
nodal involvement in resectable SCLC, that has been rarely explored so far. In this setting, Inoue et al. [9] reported better survival rates in N1 patients compared to N2 tumors, while, according to Miyamoto et al., no survival difference among node-positive patients was demonstrated regardless of pathological N status [10]. Likewise, Badzio et al. showed no prognostic difference when comparing N1 and N2 disease [11]. Based on these data, the kind of nodal involvement (N1 or N2) would not seem to be useful for stratifying the outcome of such patients. Thus, further studies are needed to assess this point.

The second issue that has not properly elucidated up to now is the optimal procedure to evaluate the nodal status. In fact, although the use of both conventional and nuclear imaging may improve clinical staging, some authors reported that N2 metastases are reported in more than 50% of clinical N1 patients [12, 13], reflecting a high underestimated metastatic potential of SCLC. As for NSCLC, mediastinoscopy represented the “gold standard” to assess mediastinum for many years. Concerning SCLC staging, Inoue et al. [13] reported that mediastinoscopy biopsy correlated with final pathology in about 80% of surgically-treated SCLC patients. Recently, a retrospective study on EBUS-TBNA has demonstrated a more accurate nodal staging with 96.4%, 100%, and 97.2% of sensitivity, specificity and accuracy, respectively [14]. Thus, the assessment of the optimal strategy to stage SCLC patients is an ongoing debate that needs to be further evaluated in order to obtain specific guidelines.

In conclusion, contrary to that advocated until few years ago, surgery for SCLC may be recommended as a part of multimodality therapies. Nowadays, it is difficult to apply the biased results of past trials [5] in the setting of SCLC patients who are potentially eligible for surgery, due to the recent improvements in terms of staging (PET, EBUS, etc.) and oncological treatments. Probably, it is time to perform a new randomized trial in Europe exploring the role of surgery in No patients as well as in those with nodal involvement and locally-advanced disease.

**Keywords:** Radiotherapy, Small-cell lung cancer, Therapy, Tumor

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Enhancing medical journalism from Internet Medical Society

Ricardo Correa, Eloy Cardenas-Estrada, Manuel Menendez

INTRODUCTION

Medicine in the 21st century has evolved into Evidence Based Medicine. By this, we mean that we have left behind the paradigm of unipotential knowledge given to us by our esteemed professors and have now entered the era of research where our hypotheses must be tested through scientific method.

Research and publishing is the foundation of our career. Through the application of research and subsequently the publishing of our work we can contribute to the scientific expansion of our chosen fields. We must be capable of producing and spreading knowledge in order for medicine to continuously develop. Each year scientific methodologies and the amount of medical researchers increase, urging the need for a platform to depict the medical community’s results.

Continue Medical Education is a challenging stage which is not only clinical but also scientific. Due to a lack of time and inherent deficit in the knowledge of scientific journalism much of the work done at this stage goes unpublished.

Open access system

In the 19th and 20th centuries, medical societies started publishing the first medical journals. These journals published articles with case reports, and ideas on new therapeutic options. Some of them also published the results of biological experiments. All these early journals were printed journals that were sold to subscribers [1]. That is, one needed to be subscribed to a journal to get access to the contents.

But the internet era arrived and the means of publishing changed in all fields of science. First, existing publishers adapted their journals, making them mixed -both printed and online-, and online only in most cases later on. New publishers opted for online only journals from birth.

The new way of publishing changed business for good: scientific publishing has never been more profitable [2, 3]. The fee-for-access model that made perfect sense for the printed journals was no longer consistent with the mission of sharing knowledge because it limited the reach of the scientific literature [1]. For this reason, many publishers adopted a new model: the open access model. In this model, instead of charging readers for accessing journals, publishers charge authors -or their institutions- a fee to cover the costs of editorial works.

The open access model is now an established model, but it also brought some challenges. Much debate has been done about open access itself and about the fees charged by publishers: are these fees suitable or excessive? Who should pay the fees, authors and their institutions or funding agencies? What about authors who do not have funds for addressing these payments? Authors from developing countries have many difficulties in getting the funds needed to cover the costs, even when some publishers offer discounts for them. This is generating a dangerous bias in the access to publishing.

The scientific world needs a fresh infusion of idealism where the obligation of paying for publishing open access can be removed. We know this may seem difficult at first glance since publishing, even online, has some important costs to face [4].

Internet Medical Society

Internet Medical Society is a worldwide, independent organization. IMS is aimed at promoting best practices in the use of internet in medical practice and medical education including publication. We also aim to promote an exchange of information and ideas on the science and
practice of medicine, both within the health professions and with responsible and informed public opinion [5].

We strongly encourage physicians from all around the world, particularly young physicians, to join IMS and to use this new source to publish their articles and create important discussion in the medical field [5].

**Keywords:** Medical Journalism, Medical societies, Open access, Open journal system

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**Author Contributions**

Ricardo Correa – 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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**REFERENCES**

Cancer survivorship care: Where are we going?
Shekhar Gogna, Priya Goyal, Prateek Thakur, Animesh Raj

INTRODUCTION

A cancer survivor is an individual who has been diagnosed with cancer, regardless of when that diagnosis was received and is still living [1]. Considering the diagnosis, response to treatment, morbidity and mortality of disease there are three distinct phases associated with cancer survival:

- The time from diagnosis to treatment
- Transition from treatment to immediate survival
- Long-term survival [2].

Over the past 25 years, the number of cancer survivors has increased from three million to nearly twelve million due to progress in the treatment of cancer. However, advanced treatment has often failed to provide survivors with care that address the side effects of both cancer and its treatment, which have a significant effect on their quality of life. American society of clinical oncology (ASCO) has identified biggest hurdle in care of cancer survivors, which is unassisted transition from an orderly system of hospital based care to a “non-system” in which there are no guidelines to assist them through the next stage of their life or help them overcome the medical and psychosocial problems [3]. This observation is true worldwide.

EVOLUTION OF CANCER SURVIVOR CARE

In 1986, National Coalition for Cancer Survivorship (NCCS) was formed with a goal to change the perception of cancer victim to survivor. This society stressed the need to include family, friends, and caregivers. National Cancer Institute established the Office of Cancer Survivorship in 1996 with the main objective to support research in cancer care. This office aimed to understand the physical, psychological, economic issues and attempted to increase the quality of life of survivors. It also intended to provide education to healthcare professionals and survivors about health related issues [4]. The Institutes of Medicine (IOM) published two subsequent reports on care of cancer survivors in 2006. Both reports recommended that survivors should get a detailed care plan to assess and treat the potential long-term effects due to cancer and its treatment. These reports also identified primary care providers as partners in the care of cancer survivors [5]. Table 1 shows the brief summary of 10 recommendations passed in the IOM meeting. National Cancer Survivorship...
Resource Center was formed in 2010, with collaboration between the American Cancer Society and the George Washington University Cancer Institute, and is funded through a cooperative agreement with the Centers for Disease Control and Prevention. This center identified 10 cancer sites based on survivor prevalence and the severity of long-term or late effects. These 10 cancer sites were colorectal, prostate, lung, breast, cervix, ovarian, endometrial, uterine, melanoma and head and neck cancers. This office has also recommended to strengthen and follow 2006 IOM guidelines as outlined in Table 1.

**Where are the evidence-based guidelines for healthcare workers**

National comprehensive cancer network (NCCN) is a pioneer society in establishing and publishing evidence-based guidelines for treatment site and stage specific cancer. The NCCN publishes and updates survivorship guidelines based on expert panel recommendations it incorporates steps that are related to late effects of cancer which are mostly neurocognitive such as anxiety, depression, cognitive function, fatigue, sleep disorders, chronic pain, and sexual function. The complaint specific guidelines are also available such as pain, emesis, infections and regular surveillance. American society of clinical oncology (ASCO) also publishes and regularly updates guidelines that focus on providing guidance for the management of long-term and late effects such as chemotherapy related neuropathy, deep vein thrombosis, mental symptoms such as stress depression, fatigue. ASCO has previously published and now updated follow-up guidelines for patients with prostate, breast colorectal cancer.

**CONCLUSION**

The Institutes of Medicine (IOM) report identified four essential components of survivorship care: prevention, surveillance, intervention, and coordination. Cancer care is continuously evolving process. Oncologist and primary care physicians both hold the equally important positions in the life of cancer survivor. The goal of cancer survivorship is to empower survivors and their families. Cancer survivors should be given detailed follow up plan after primary treatment for better improvement in their quality of life.

**Keywords:** Cancer care, Cancer survivorship, Guidelines

Table 1: Institutes of Medicine (IOM) recommendations

| Raise awareness of the needs of cancer survivors. |
| After completing primary treatment patients should be provided with a care summary and follow-up plan. |
| Health care providers should use evidence-based guidelines, to recognize and treat late physical and psychological effects of cancer. |
| Effective public/private healthcare sector partnerships should be formed to achieve health care measures. |
| Various designated cancer research societies will support and expand survivorship research. New research initiatives on cancer patient follow-up are urgently needed to guide the effective care. |
| Policy makers should act to ensure that all cancer survivors have access to adequate and affordable health insurance. Insurance companies should recognize survivorship care as an essential part of cancer care, and these companies should design easy reimbursement on plans for cost of cancer care. |
| Concerned authorities will ensure and eliminate workplace discrimination and minimize adverse effects of cancer on employment. |
| Provide educational opportunities to health care providers to be able to address the health care and quality of life issues of cancer survivors. |
| Congress should support the Centers for Disease Control and Prevention and other collaborating institutions, and the states in developing comprehensive cancer control plans. |
| Centers for Medicare and Medicaid Services, National Cancer Institute, Agency for Healthcare Research and Quality, Department of Veterans Affairs, and other qualified organizations should support the demonstration programs to test models of coordinated, interdisciplinary survivorship care across the society. |

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REFERENCE


Carbon monoxide poisoning: Same time, same place but different outcomes

Phee-Kheng Cheah, Fatin Salwani Zaharuddin, Nik Hisamuddin Rahman, Muhamad Yaakub Arifin, Mohd Hakimi Abdullah

ABSTRACT

Introduction: There have been numerous reports documenting the differences in outcomes after carbon monoxide poisoning between men and women. There are also several reports mentioning the differences in presentation and outcome of poisoning in adults and children. Case Series: We report two construction workers who had carbon monoxide poisoning while watching television. They used a petrol generator to power their television during a power outage, and the generator was placed in the same confined room. All windows were closed due to heavy rain and strong winds outside. Both patients were found motionless the next morning in front of the television. The two patients aged 16 and 22 years old presented to emergency department with almost similar complaints but had a very differing hospital course. The 16-year-old had status epilepticus needing intubation and ICU admission while the other was well throughout his stay. Both patients underwent two courses of hyperbaric oxygen therapy using the Royal Malaysian Navy treatment table 18-60-30. The 16-year-old was placed in the multi-place hyperbaric chamber while still intubated. Both patients were discharged without any neurological deficits. Conclusion: The clinical course of patients with carbon monoxide poisoning may differ due to factors other than gas concentration and duration of exposure.

Keywords: Carbon Monoxide Poisoning, Hyperbaric Oxygen Therapy, Status Epilepticus

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INTRODUCTION

Carbon monoxide poisoning has been dubbed as a silent killer as its presence goes undetected by human senses. It is a colorless, odorless, tasteless and a non-irritant gas. It produces variable extremes of symptoms and complications. Patients may be totally asymptomatic or present with only mild symptoms such as dizziness, headache and flu-like symptoms to extremes of severe cardiopulmonary failure, nervous system dysfunction and death [1].

Severity of carbon monoxide poisoning is related to the amount of carbon monoxide inspired [2]. However, patients exposed to the same amount of carbon monoxide may have different clinical manifestations. Few factors
have been studied as to what determines the severity of poisoning and the parameters contributing to these differences. The relation of these factors to the severity of poisoning is yet to be clearly proven although some explanations had been offered. We report two young healthy adult males, accidentally exposed to carbon monoxide at the same location with equal amounts of exposure. Surprisingly, they developed neurological complications of differing severity and had a very different clinical course in hospital. Both patients were young and well with no preexisting medical conditions that could have explained the differences.

Principles in managing carbon monoxide poisoning involves dissociating the carbon monoxide bond from hemoglobin which is 230 times stronger to split compared to the oxygen-hemoglobin bond. Hyperbaric oxygen therapy (HBOT) has been long recognized as the best modality to deliver super-saturated oxygen to hypoxic tissues and disable the carbon monoxide-hemoglobin bond [3].

CASE SERIES

Two construction workers, aged 16 years and 22 years, were found unconscious by their relatives. Both had been sleeping in a confined room with a petrol generator placed inside the room. The generator was running from 10.00 pm until the next morning due to a power failure. They presumably fell asleep or were rendered unconscious by the carbon monoxide while watching television, which was powered by the generator. All windows were closed, as it was raining heavily with strong winds. Both patients were previously well with no medical illness and neither had exhibited any features of depression, suicidal or parasuicidal tendencies. Neither was known to have abused illicit drugs. Both patients were brought immediately to the emergency department (ED) by their relatives who found them.

Patient A is a 16-year-old boy had Glasgow Coma Scale (GCS) of 10/15 (E3V2M5) on arrival to ED. His blood pressure was 120/71 mmHg and pulse rate 126 bpm. Initial oxygen saturation was 90%, rising to 100% after oxygen was given via a non-rebreather mask. His blood pressure was 115/62 mmHg and pulse rate 105 beats/min. Cardiovascular and respiratory examination were normal. His neurological assessment also indicated an upper motor neuron lesion. No evidence of meningism was found. Blood investigations showed mild leukocytosis with normal hemoglobin, platelet, glucose, electrolytes and creatinine levels. Similar to patient A, there was an elevated level of creatinine kinase with a reading of 2162 mmol/L. Serum lactate was 3 mmol/L. Initial blood gas shows, pH 7.30, pO2 105 mmHg, pCO2 27 mmHg and bicarbonate 16.1 mmol/L. Serial measurements of electrocardiogram did not show any ischemic changes and chest radiograph was normal. Computed tomography scan of brain was also normal. This patient developed generalized tonic-clonic seizure in casualty but aborted spontaneously after 2 minutes. After the seizure, he continued breathing spontaneously without needing any ventilatory support as compared to patient A. The patient underwent two sessions of HBOT using the Royal Malaysian Navy treatment table 18-60-30 in the multi-place hyperbaric chamber while still intubated. No complications were observed during the session. Subsequently, he was monitored in the Intensive Care Unit. On day-3, intropic support could be weaned off. He was extubated and regained full GCS with normal neurological assessment on day-5. He was discharged on day-8.

Patient B is a 22-year-old male also arrived with GCS of 10/15 (E3V2M5). Oxygen saturation was 86–88% on room air, which improved to 100% with oxygen via a non-rebreather mask. His blood pressure was 115/62 mmHg and pulse rate 105 beats/min. Cardiovascular and respiratory examination were normal. His neurological assessment also indicated an upper motor neuron lesion. No evidence of meningism was found. Blood investigations showed mild leukocytosis with otherwise normal hemoglobin, platelet, glucose, electrolytes and creatinine levels. This patient developed status epilepticus requiring emergency intubation. The patient needed inotropic support post intubation. The post intubation chest X-ray showed haziness in the right lower zone, hence patient was also treated for aspiration pneumonia. The patient underwent two sessions of hyperbaric oxygen therapy (HBOT) using the Royal Malaysian Navy treatment table 18-60-30 in the multi-place hyperbaric chamber while still intubated. No complications were observed during the session. Subsequently, he was monitored in the Intensive Care Unit. On day-3, intotropic support could be weaned off. He was extubated and regained full GCS with normal neurological assessment on day-5. He was discharged on day-8.

DIscUssION

Methods of defining severity of carbon monoxide poisoning are still being debated. It is generally agreed that it depends on the inspired concentration of carbon monoxide, the length of exposure towards the poisonous gas, and the general health status of the individual being exposed [2]. From the history, we can safely conclude that both patients were presumably exposed to carbon monoxide for an equal amount of time as they were in the same confined room. Therefore, we can also postulate that both had inspired an equal amount of carbon...
monoxide during that duration. Both are young, fit, male construction workers with no known medical illnesses.

Despite those similarities and the presumed same carbon monoxide exposure, they developed neurological complications with different extremes of severity. Age is one factor that influences outcome. It has been reported that children are more susceptible to high level of toxicity as they have a higher metabolic rate as compared to adults [4]. Although one of our patients were in the pediatric age group, they had an age difference of only 6 years. They showed a marked difference in their disease process with patient A needing intubation and ICU care. However, their final neurological outcome was similar. Did the difference in age become the deciding factor in the divergent outcome in these two cases?

Impaired consciousness level has been related to severity of carbon monoxide poisoning. Grieb et al. concluded in their study that there was an inverse correlation between initial GCS and severity of carbon monoxide toxicity [4]. Both patients presented with initial GCS of 10/15 but their clinical courses differ in severity. Elevated leucocyte counts and C-reactive proteins were found to be associated with severe carbon monoxide toxicity [5]. Both patients had elevation of white cell counts in their blood investigations, reflecting only severity of their carbon monoxide level but not the difference in clinical outcome. Lactate elevation results from anaerobic glycolysis due to inadequate oxygen supply. Reports concluded that blood lactate level might also be a useful indicator in predicting severity of carbon monoxide poisoning [6]. Both patients had a raised level of lactate with metabolic acidosis.

Measuring carboxyhemoglobin level is not yet available in our current setting. Many reports however have concluded that levels of carboxyhemoglobin have little or variable correlation with symptoms or severity of carbon monoxide poisoning [5]. A recent study only recorded 61.6% positive toxicology screening results in intubated carbon monoxide poisoned patients [7]. In a setting like ours which carboxyhemoglobin levels may not be readily available, we suggest a high index of suspicion for this common form of poisoning. Clinical history and physical examination is of utmost importance in making this crucial diagnosis. Certain laboratory parameters such as respiratory alkalosis are suggestive and should be able to increase suspicion of carbon monoxide poisoning [8].

Hyperbaric oxygen therapy remains the gold standard for the treatment of carbon monoxide poisoning. In the military hospital in Malaysia the treatment table used is the Royal Malaysian Navy Treatment Table 18-60-30 whereby the patient is pressurized to an atmospheric pressure equivalent to 18 m depth in a hyperbaric chamber within a few minutes. Once the depth of 18 m is reached, a cycle of 25 minutes of 100% oxygen and 5 minutes of air-break will be delivered and repeated once before the patient is surfaced from 18 m to 0 m while breathing 100% oxygen. The air-break in between the oxygen delivery is to prevent the effect of oxygen toxicity.

Although many case reports has documented the differences in carbon monoxide toxicity in mass group poisoning, the factors identified could not explain the difference in the clinical course of our patients. From our patients, we agree that impaired consciousness, elevated leucocyte and high lactate levels reflect the severity of carbon monoxide toxicity and may predict the final neurological outcome. However, questions still remain as to why the clinical courses are different and how these factors correlate with each other to produce a different outcome. Perhaps further understanding of this differing clinical course will shed more light on measuring the severity in carbon monoxide poisoning.

CONCLUSION

The clinical course of patients with carbon monoxide poisoning may differ due to factors other than gas concentration and duration of exposure. Management of these patients especially when there are multiple victims involving pediatric age group should be tailored to individual presentation and clinical course.

*******

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REFERENCES

A case of hardening of the skin and scleroderma like changes on biopsy: Is it scleroderma or not? A review of pseudoscleroderma and differential diagnosis

Travis C. Sizemore, Gulzar Merchant, Katharine Whitfield

ABSTRACT

Introduction: Scleroderma is an autoimmune connective tissue disease characterized by pervasive systemic multiple organ fibrosis. Definitive diagnosis can be difficult as this disease has a highly variable disease spectrum and course. Further complicating matters, there exist several less commonly known imitators, termed pseudoscleroderma, which force a more open differential than might be presumed initially. Case Report: We present the case of a 46-year-old female diagnosed with pseudoscleroderma. Conclusion: This case emphasizes the importance of skin biopsy while not on steroids and also the need to maintain a broad differential even in the setting of biopsy results.

Keywords: Pseudoscleroderma, Scleromyxedema, Scleroderma

INtrODUctION

Systemic sclerosis, known commonly as scleroderma, is an autoimmune connective tissue disease characterized by systemic multiple organ involvement with a highly variable disease spectrum and course. Exact etiology has yet to be elucidated, however, it is known that pathogenesis involves fibroblast dysfunction, autoimmune response with abnormal production of antibodies, and tissue hypoxia secondary to vascular abnormality [1]. Genetic involvement is also known to predispose patients to scleroderma, with genome-wide association studies illustrating the variable genetic components that contribute to the clinical sub-phenotypes of systemic sclerosis [2, 3]. The classification criteria for diagnosis was revised in 2013 by a collaborative initiative of the American College of Rheumatology/European League against Rheumatism (ACR-EULAR) and was shown to have improved sensitivity and specificity over the 1980 criteria set out by the ACR [4]. It includes a point system with weighted variables determined by multi-criteria decision analysis. There are two exclusionary criteria, one sufficient criterion, and seven criteria of varying weight in achieving a summative threshold score for classification of systemic sclerosis [4].

One of the exclusion criteria of the new initiative is that the scoring is not applicable to those with "a scleroderma-like disorder that better explains their manifestations." Narrowing the differential diagnosis is often difficult because of the existence of these less well known imitators of scleroderma. The term "pseudoscleroderma" is an umbrella term that has been used to describe skin lesions that imitate or resemble systemic sclerosis. We herein report a case of pseudoscleroderma. The patient, and a patient with diabetes with paraproteinemia initially presumed to be

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scleroderma based on biopsy but not consistent with current diagnostic criteria of scleroderma.

CASE REPORT

A 46-year-old Caucasian female presented as a new patient at an Internal medicine resident rheumatology clinic with a nine-month history of swelling in her arms and legs. She stated that she had noticed a thickening/hardening of the affected skin and pain in her joints that had been progressively worsening over the last six months. She had recently been seen by an oncologist after a previous lab work revealed an elevated serum IgG level, and by their assessment, she had a very small elevation in a monoclonal protein which was not deemed to be clinically significant. However, this physician was concerned with the “plaque-like lesions” all over her body. She was then referred to dermatology for biopsy, which also occurred prior to being seen at the Rheumatology Clinic.

This patient had a past medical history of two DVT’s in 1997 and 2002 involving her left leg for which she is on Coumadin. She had recently been diagnosed with type 2 diabetes mellitus with a HgbA1c of 7.5%. She has hypertension, hyperlipidemia, and depression. She stated that throughout these months, she had not experienced any fever, dysphagia, or Raynaud’s-like symptoms. The patient was morbidly obese, but did report an involuntary weight loss of 50 lbs since her symptoms began.

On examination, she was afebrile with elevated blood pressure 197/113 mmHg, heart rate 83bpm, temperature 97.1°C, weight 163.48 kg, and BMI 56.44. Skin exam showed thick indurated skin across her thighs, calves, abdomen, upper arms, and lower arms that stopped proximal to the phalanges. All extremities were involved equally. There were also changes in pigmentation with both hypo- and hyperpigmentation scattered throughout affected areas. Scabbed-over ulcers were present mainly on bilateral thighs that were not erythematous or warm to palpation (Figures 1–6). Ophthalmic, cardiac, and pulmonary examinations were within normal limits.

Laboratory investigations reveal IgG was elevated at 1679 mg/dl, with normal IgE (<4 mg/dl), CRP 51.4 mg/dl, ANA negative, ASO<50 IU/ml, scleroderma and centromere antibodies negative, anti-smith and anti-RNP antibodies negative, kappa-lambda ratio normal at 0.52. Computed tomography (CT) scan of the thorax exhibited hepatomegaly and mediastinal, retroperitoneal, hepatic, and pelvic lipomatosis. PFTs were performed, showing mild airway obstruction with FEV1/FVC 68.

Two punch biopsies were obtained by the consulting dermatologist. Right proximal forearm biopsy findings were deemed insignificant, but histopathological examination of the sample from the right anterior proximal thigh (Figures 7 and 8) revealed extensive dermal fibroplasia with hyalinization and sparse perivascular lymphoplasmacytic inflammation suggestive of scleroderma. It did not show the degree of cellularity or mucin deposition typical of nephrogenic systemic fibrosis or scleromyxedema. An atypical lymphoid infiltrate was not seen. Basketweave orthokeratosis was observed. The vital epidermis was normal in thickness and architecture. Throughout the full thickness of the reticular dermis there was prominent fibrosis. There were sparse superficial perivascular inflammatory infiltrate of lymphocytes, mononuclear cells, and plasma cells.

These initial punch biopsies were taken while the patient was on prednisone. Upon visit at the clinic, the patient was set up for a deeper biopsy to the fascia of the thigh that was to be done with the patient off of all steroids. This biopsy revealed collagen thickening with decreased intra-collagenous clefts and a focal decrease in pericellular fat. There were sparse patchy perivascular lymphoplasmacytic infiltrate present.

The limited fascial component is free of inflammatory elements and increased eosinophils are not seen. The overall features are morphologically often seen in scleroderma (Figures 9 and 10).

From the history, clinical examination, and histopathological findings, a diagnosis of nonspecific pseudoscleroderma was made and further evaluation and treatment of paraproteinemia was suggested.

DISCUSSION

Scleroderma was originally described by Neapolitan physician Carlo Curzi in a monograph dated 1752 [5]. The term is derived from the Greek words “sklerosis,” meaning hardness, and “derma,” meaning skin. Systemic sclerosis
prevalence is estimated between 3 and 24 per 100,000 persons and appears to be higher in North America and Australia as compared to Europe and Japan [6].

Two distinct clinical subsets exist in scleroderma, determined by the degree of skin involvement: limited cutaneous systemic sclerosis with skin findings usually limited to the hands and forearm, and diffuse cutaneous systemic sclerosis that can involve abdomen, chest, upper arms, and shoulders.

Specific histological findings of scleroderma include an increased amount of new collagen synthesis in the reticular dermis as well as an increased number of myofibroblasts, activated fibroblasts that express the smooth muscle marker (smooth muscle actin), presence of myofibroblasts, with intima proliferation and
parakeratosis [7, 8]. Serum antibodies, systemic sclerosis specific autoantibody-anticentromere antibodies, anti-topoisomerase antibodies and anti-RNA polymerase III antibodies are found in over 50% of patients and can be useful predictors of disease prognosis and organ involvement [9].

Prognosis in systemic sclerosis is poor and often fatal, with 10 year survival ranging from 54–66% [10]. However, many of the causes of pseudoscleroderma syndromes have much favorable prognosis and can be reversed by treatment of the underlying etiology or removal of offending agent.

The term “pseudoscleroderma” is an umbrella term that has been used to describe skin lesions that imitate or resemble systemic sclerosis. These disorders typically occur as either distinct pathological entities or a complication of malignancy. A smaller number are induced by medication or environmental factors [11, 12]. To list a few: eosinophilic fasciitis, sclerodermiform genodermatoses, scleredema adulterum of Buschke, acrodermatitis chronica atrophicans, porphyria cutanea
tarda, Graft-versus-host disease, nephrogenic fibrosing dermopathy (NFD), scleredema diabeticorum, and scleromyxedema. Pseudoscleroderma syndromes mimic scleroderma in general by causing thickening/hardening of the skin. Additional common factors of pseudoscleroderma include: Pathogenesis is thought to be secondary to activation of eosinophils and upregulation of fibroblast and collagen synthesis producing an overall increase in cytokines, specifically interleukin-4 and interleukin-13, as well as transforming growth factor beta [13].

It is often very difficult to differentiate from scleroderma and can result in delayed diagnosis and treatment. To be distinguished from scleroderma, the ACR/EULAR classification criteria have been adapted.

**CONCLUSION**

Herein, we report a case of nonspecific pseudoscleroderma, initially thought to be true scleroderma based on biopsy. However, patient did not meet diagnostic criteria for scleroderma. Although morphological features observed on patient’s histopathology can be seen in scleroderma, scleroderma is not a diagnosis based solely on pathology. In fact, ACR/EULAR updated classification criteria does not include pathology. The diagnosis of nonspecific pseudoscleroderma was made because clinically, patient did not meet criteria for scleroderma, having scored 0/34 points (Table 1). In addition, patient has concurrent paraproteinemia with elevated IgE that can be seen in different and distinct types of pseudoscleroderma. The patient’s clinical profile did not correspond completely with any one subset of pseudoscleroderma, thus, she was diagnosed with a nonspecific form pending further evaluation with bone marrow biopsy and flow cytometry and additional dermatologic evaluation. This case report is an important reminder that many rheumatological diseases cannot be made simply by pathology on biopsy but need to be taken within clinical context and case by case.

**Author Contributions**

Travis C. Sizemore – 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

Gulzar Merchant – 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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**Table 1: ACR/EULAR criteria for diagnosis and classification of systemic sclerosis adapted from American College of Rheumatology/European League against Rheumatism collaborative initiative [4].**

<table>
<thead>
<tr>
<th>Item</th>
<th>Sub-item</th>
<th>Weight/score</th>
<th>Patient</th>
</tr>
</thead>
<tbody>
<tr>
<td>Skin thickening of fingers of both hands proximal to metacarpophalangeal joint</td>
<td>Puffy fingers</td>
<td>9</td>
<td>NONE</td>
</tr>
<tr>
<td>Skin thickening of the fingers</td>
<td>Sclerodactyly</td>
<td>2</td>
<td>NONE</td>
</tr>
<tr>
<td>Fingertip lesions</td>
<td>Digital tip ulcers</td>
<td>2</td>
<td>NONE</td>
</tr>
<tr>
<td></td>
<td>Fingertip pitting scars</td>
<td>3</td>
<td>NONE</td>
</tr>
<tr>
<td>Telangiectasia</td>
<td>2</td>
<td>NONE</td>
<td></td>
</tr>
<tr>
<td>Abnormal nailfold capillaries</td>
<td>2</td>
<td>NONE</td>
<td></td>
</tr>
<tr>
<td>Pulmonary arterial HTN and/or ILD</td>
<td>Pulmonary arterial HTN</td>
<td>2</td>
<td>NONE</td>
</tr>
<tr>
<td></td>
<td>Interstitial Lung disease</td>
<td>2</td>
<td>NONE</td>
</tr>
<tr>
<td>Raynaud’s phenomenon</td>
<td>3</td>
<td>NONE</td>
<td></td>
</tr>
<tr>
<td>Systemic sclerosis related autoantibodies (anticentromere, anti-Scl-70, anti-RNA polymerase III)</td>
<td>3</td>
<td>NONE</td>
<td></td>
</tr>
</tbody>
</table>

Final score: 0
Katharine Whitfield – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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REFERENCES

Asymptomatic idiopathic right atrial rupture: An unusual presentation

Parminder S. Otaal, Rajesh Vijayvergia

ABSTRACT

Introduction: Atrial rupture is a very rare and fatal condition and has been only scantily reported in the past. Patients with this condition present with significant clinical symptoms requiring an urgent medical management. Case Report: A case of spontaneous idiopathic right atrial rupture detected on routine physical examination in a completely asymptomatic active young male. A defect of 18 mm in the right atrial wall just above the level of tricuspid valve was revealed in the transthoracic echocardiogram. Patient denied the option of surgical repair and remains asymptomatic during a two-year follow-up. Conclusion: Very rarely, patients with spontaneous idiopathic right atrial rupture may remain asymptomatic for a long period and can live a normal life without any active surgical intervention. However, the patient may develop severe right heart failure followed by death if the medical condition is left untreated. In view of the long-term complications, it is advisable to treat this condition with immediate surgical intervention.

Keywords: Right atrial rupture, Idiopathic, Spontaneous, Cardiology

INTRODUCTION

Atrial rupture of the heart is a rare condition which carries a very high mortality and requires urgent surgical repair [1, 2]. Almost all cases of right atrial rupture reported in literature had significant symptoms and either underwent urgent surgery or had limited survival [2–4]. Survival beyond short-term has not been reported in literature [1]. Here we report a patient with spontaneous right atrial rupture diagnosed two years back and followed-up since then. The case here is discussed in view of the existing literature.

CASE REPORT

A 25-year-old male was referred to us for cardiac evaluation in view of cardiomegaly found on chest X-ray during pre-employment medical examination. Patient was asymptomatic, active young man with no significant past medical history. Patient denied any history of malaise, fever, weight loss or fatigue. Patient also denied any history of chest discomfort, trauma, any hospitalization or intervention in the past. His clinical examination was normal with no evidence of systemic venous congestion. Electrocardiography showed no abnormality.
X-ray of chest revealed cardiomegaly with right atrial enlargement. Transthoracic echocardiogram revealed an 18 mm defect in the right atrial wall just above the level of tricuspid valve. This was supported by transesophageal echocardiogram (Figure 1). Color Doppler using transesophageal echocardiography showed flow across the defect into the pericardial cavity (Figure 2). Further, contrast echocardiogram using transesophageal echocardiogram revealed opacification of right atrium and right ventricle simultaneously with opacification of pericardial sac while the left side chambers showed no evidence of bubble contrast (Figure 3). Echocardiography also revealed compression of the right ventricle and its outflow tract suggestive of right sided tamponade. There was minimal effusion on either side of left ventricle or left atrium.

The patient was advised further evaluation and the option of possible surgical repair but patient denied the same. Patient is on follow-up from last two years and is completely asymptomatic.

**DISCUSSION**

Right atrial rupture cases can be classified into different types based on the causes cited in various case reports.

1. **Iatrogenic**

Live three-dimensional echocardiography helped diagnose a 54 year old woman of silent right atrial rupture following cardiac catheterization for atrial septal defect [4]. The patient had undergone cardiac catheterization and coronary angiography two months earlier and was complaining of tolerable exertional dyspnea.

2. **Post-traumatic**

Blunt traumatic cardiac rupture is associated with a very high rate of mortality. Rapid pre-hospital transportation and prompt surgical intervention contribute to survival in these patients [2, 5, 6]. Brathwaite et al. observed an overall mortality of 81.3% in their review of 32 patients comprising right atrial rupture (40.6%), left atrial rupture (25%), right ventricular rupture (31.3%), left ventricular rupture (12.5%) and rupture of two cardiac chambers (9.4%). The only survivors were those patients (6 of 12 patients) who presented with vital signs [2].

3. **Malignancy**

Primary heart tumors are extremely rare, with an incidence of 0.0017% as reported in the autopsy studies by American Medical Association [7]. However, cardiac metastases are more frequent than the primary heart tumors. Angiosarcomas have a mesenchymal origin and account for 25–30% of the malignant cardiac tumors [1]. Angiosarcoma of the heart almost exclusively involves the right atrium and appears between the 3rd and 5th
decades of life. Spontaneous rupture of an angiosarcoma is extremely rare with only few case reports till now [1, 8].

4. Infarction

Wessler et al. studied 20 hearts on autopsy and found that none of the ruptures was localized to the right side of heart and all cases had myocardial infarction in the territory of rupture [9]. Rupture of the right ventricle in acute myocardial infarction has been infrequently reported in literature. Atrial rupture in acute myocardial infarction is rare but has been reported [10].

5. Idiopathic

The present case is a unique case of right atrial rupture since this young man harbors cardiomegaly accompanied by right atrial rupture yet he is asymptomatic and clinically sound.

Most common symptoms of atrial rupture are recurrent or persistent chest pain, asthenia, dyspnea, syncope and distension of jugular vein. The diagnosis of atrial rupture is generally made based on physical examination and changes in the vital signs that can be confirmed through hematological tests followed by radiological investigations such as chest X-ray, transthoracic echocardiography (TEE), color Doppler, magnetic resonance imaging (MRI) scan etc. The differential diagnosis of right atrial rupture should include epicardial hematoma. Surgical correction of the rupture is the treatment of choice and patients can survive if the rupture is recognized and corrected in acute setting.

CONCLUSION

Very rarely, patients with spontaneous idiopathic right atrial rupture may remain asymptomatic and can live a normal life without any active surgical intervention. However, the patient may develop severe right heart failure followed by death if the medical condition is left untreated. In view of the long-term complications, it is advisable to treat this condition with immediate surgical intervention.

**********

Author Contributions

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

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REFERENCES

Portal vein thrombosis following laparoscopic sleeve gastrectomy: A rare case report

Nidal Abu jkeim, Ahmad Al Hazmi, Awad Alawad, Rashid Ibrahim, Ahmad Abudamis, Samir Tawfik, Mohammed Mansour

ABSTRACT

Introduction: Portal vein thrombosis (PVT) is a relatively uncommon complication after sleeve gastrectomy. Case Report: A 33-year-old female underwent an uncomplicated laparoscopic sleeve gastrectomy for the treatment of morbid obesity, and presented on postoperative day 14 with epigastric pain. Computed tomography scan revealed left portal vein thrombosis. She promptly improved after initiation of low-molecular-weight heparin and was discharged on hospital day-5 with oral warfarin. Conclusion: Although uncommon, PVT should be included in the differential diagnosis for unexplained abdominal symptoms after laparoscopic sleeve gastrectomy.

Keywords: Laparoscopic sleeve gastrectomy, Morbid obesity, Portal vein thrombosis

INTRODUCTION

Portal venous thrombosis (PVT) is a potentially lethal condition with multiple causes, both systemic and local. Postoperative portal venous thrombosis is a known complication following surgical operations that involve manipulation of splanchnic veins, such as splenectomy [1]. However, with the broad use of laparoscopic surgery over the last 30 years, case studies have emerged of PVT, in the absence of direct manipulation of the portal or mesenteric vessels. Among the laparoscopic operations with reported postoperative PVT are appendicectomy, cholecystectomy, and colectomy, as well as Nissen fundoplication, and sleeve gastrectomy for morbid obesity [2–6]. Although the cause of PVT following laparoscopic surgery is obscure, it is well known that increased intra-abdominal pressure induced by pneumoperitoneum (using carbon dioxide) results in reduction of blood flow in the portal vein, which may enhance thrombosis.

Laparoscopic sleeve gastrectomy is emerging as a popular operation for the treatment of morbid obesity, with acceptable morbidity and long-term weight reduction. Here we report an even more unusual case of PVT, 14 days after an uncomplicated laparoscopic sleeve gastrectomy.

CASE REPORT

A 33-year-old woman presented initially to our department with morbid obesity. Her past medical history was remarkable for diabetes mellitus and bronchial asthma. She had no remarkable family history of hypercoagulable state or thrombotic events. She was not taking oral contraceptive pills. After an extensive multidisciplinary workup, she was planned for a laparoscopic sleeve gastrectomy. Her preoperative liver function tests showed normal hepatic enzymes (alanine...
aminotransferase 27 U/L, aspartate aminotransferase 10 U/L), alkaline phosphatase (105 U/L), and total bilirubin (10.4 umol/L). Preoperative coagulation parameters were all normal. She underwent an uneventful laparoscopic sleeve gastrectomy. Water was administered orally on postoperative day-2 after an upper gastrointestinal contrast study revealed no leak from the staple line. Throughout her hospital stay, she was treated with subcutaneous injection of enoxaparin 40 mg once per day. She was discharged home on day-5 with subcutaneous enoxaparin.

At postoperative day 14, the patient re-presented to our outpatient clinic complaining of epigastric pain and nausea for one day. On physical examination, her blood pressure was 130/60 mmHg, pulse rate 94 beats/minute, body temperature 37.1°C. Abdominal examination demonstrated minimal epigastric tenderness. Her laboratory results were within normal limits. Ultrasonography of the abdomen showed thrombosed left portal vein (Figure 1), which was confirmed with a computed tomography (CT) scan of the abdomen (Figure 2).

The patient was admitted to our department. A hypercoagulability workup demonstrated normal findings. She was started on therapeutic low-molecular-weight heparin (LMWH, enoxaparin). She responded to the treatment with resolved abdominal symptoms. An abdominal ultrasonography revealed scanty color filling at the periphery suggesting partial recanalization of left portal vein (Figure 3). She was discharged in good condition on hospital day-5 with oral warfarin.

DISCUSSION

Portal vein thrombosis (PVT) is a recognized complication after laparoscopic sleeve gastrectomy. There are few cases reported in literature of PVT after laparoscopic sleeve gastrectomy [3, 7–8]. Patients in these case reports underwent what was described as uncomplicated laparoscopic sleeve gastrectomy.

Increased intra-abdominal pressure induced by pneumoperitoneum during laparoscopy results in disturbed clotting parameters, as manifested by changes in prothrombin time, international normalized ratio, fibrinogen, and fibrin degradation products. It affects portal and splanchic venous blood stream [2]. One study concluded that the radius of the portal vein and the mean portal blood stream were significantly diminished with pneumoperitoneum of more than 10 mmHg [8]. Positioning of the patient during laparoscopic surgery might likewise influence the portal blood flow leading to more stasis. Direct injury to the portal vein and its branches may also be a contributing factor but is unlikely in our case.

The diagnosis of PVT can be confirmed with either abdominal ultrasonography or computed tomography. Both imaging studies are usually used in the workup of unexplained abdominal pain after laparoscopic sleeve gastrectomy [2]. Our patient had both studies to confirm the diagnosis of PVT. The use of invasive portal venography is typically not necessary.

The treatment of PVT varies depending on the degree of the thrombosis and the presence or absence of intestinal ischemia. It is unknown whether prophylactic anticoagulants affect the incidence of PVT or not. Most patients who were diagnosed with PVT after laparoscopic procedures can be managed properly with supportive treatment and anticoagulation. One patient in literature required transhepatic portal vein thrombectomy for significant thrombosis [9]. Our patient promptly improved after initiation of LMWH and was discharged on day-5 with oral warfarin. The optimal duration of anticoagulation for these patients is unidentified. If the patient has primary clotting disorders, lifelong anticoagulation is needed. For those with no recognized risk factors, some recommend it is reasonable to re-evaluate the patient after approximately 6 months of anticoagulation and stop if declaration of the thrombosis is proved. Although still a subject of debate, some studies suggest that the threat of venous thrombosis following laparoscopic bariatric surgery extends long after discharge from the hospital, and prophylaxis should therefore be sustained for several weeks into the postoperative period [10–11].
of PVT, is not considered standard. In our case, LMWH was continued for 12 days after surgery.

CONCLUSION

In this report, we describe a female patient who presented with portal vein thrombosis (PVT) after uncomplicated laparoscopic sleeve gastrectomy. Although rare, PVT should be one of the differential diagnoses for unexplained abdominal pain after laparoscopic sleeve gastrectomy. Anticipation, careful clinical monitoring and early anticoagulation are crucial to reduce morbidity.

*********

Author Contributions

Nidal Abu jkeim – 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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Mohammed Mansour – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

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REFERENCES

A case of ganglioneuroma of the colon during routine colonoscopy

Michael Herman, Jean Abed, Wenjing Shi, Arzu Buyuk, Pavan Kumar Mankal, Donald Kotler, Gabriel Ionescu

ABSTRACT

Introduction: Colonic ganglioneuromas are classified as hamartomatous polyps that are composed of ganglion cells, nerve fibers, and enteric nervous system cells. The GNs of the gastrointestinal tract can be classified into three groups based on the size and the number of polyps: polypoid GN, ganglioneuromatous polyposis, and diffuse ganglioneuromatosis. Polypoid GNs, seen in patients with Cowden’s syndrome, are small, sessile or pedunculated polyps that have a similar appearance to hyperplastic and adenomatous polyps. Ganglioneuromatous polyposis, seen most commonly in patients with MEN IIb, NF1, Cowden’s syndrome, usually manifests as more than 20 sessile or pedunculated polyps. Lastly, diffuse ganglioneuromatosis, seen in MEN IIb7 and NF1, involves proliferation of neuronal cells in the entire colon, but does not extend into the ileum. Case Report: A 57-year-old African-American male with a history of untreated chronic hepatitis C cirrhosis with viral load of over 4 million copies, seizure disorder, mild mental retardation, hypothyroidism, hypertension, diabetes mellitus type 2, presented to the gastroenterology clinic for scheduling of a screening colonoscopy. At that time, he was completely asymptomatic. On colonoscopy, one 4 mm sessile polyp was resected in the sigmoid colon and was histologically diagnosed as a ganglioneuroma (GN). Conclusion: The finding of an asymptomatic, solitary GN in our patient does not warrant more frequent colon cancer screening given its benign nature.

Keywords: Cancer, Colonoscopy, Cowden’s syndrome, Ganglioneuroma, Neurocutaneous syndromes

INtroDuction

Ganglioneuromas (GNs) are a subset of neuroblastomas. They are rare, slow growing, well-differentiated large tumors that arise from sympathetic ganglion cells. They are often benign and have excellent prognoses even when the tumor is not completely resected, regardless of where they are located in the body. Epidemiologically, GNs are seen more frequently
in females, with 60% occurring before the age of 20 years [1]. The most common locations for GNs to appear are the mediastinum, retroperitoneum, and adrenal glands, and less commonly the colon. Patients with intestinal GNs usually have (1) neurocutaneous syndromes, such as neurofibromatosis type 1 (NF1) and tuberous sclerosis, (2) genetic polyposis syndromes, such as juvenile polyposis, polyposis coli, or Cowden’s disease, or (3) multiple endocrine neoplasia type IIB (MEN IIB). However, solitary lesions do not embody the same association. Colonic GNs are relatively asymptomatic and patients with the above syndromes have the same risk of having gastrointestinal manifestations of GNs as the general population [1].

CASE REPORT

A 57-year-old African-American male with a history of untreated chronic hepatitis C cirrhosis with viral load of over 4 million copies, seizure disorder, mild mental retardation, hypothyroidism, hypertension, diabetes mellitus type 2, presented to the gastroenterology clinic for scheduling of a screening colonoscopy. At that time, the patient was asymptomatic and did not report any fever, chills, nausea, vomiting, abdominal pain, diarrhea or constipation. He never had any alarming symptoms such as weight loss, melena and hematochezia. In addition to denying toxic habits (i.e., tobacco, alcohol, drugs), he also denied any personal or family history of colon or small bowel cancer. His physical examination was unremarkable, as he did not have scleral icterus, murmurs, wheezing, abdominal distention, organomegaly, café au lait spots or fibromas on his skin. At home, the patient was taking metformin, levothyroxin, ferrous sulfate, tamsulosin, lactulose, omeprazole, divalproex sodium, propranolol, aripiprazole, vitamin C, multivitamin, and saline nasal spray. Initially, patient underwent a screening colonoscopy, one 8 mm pedunculated polyp in the ascending colon was removed with a cold biopsy forceps and was histologically identified as a tubular adenoma. Additionally, a pink, tan sessile polyp-like lesion was identified, measuring 4 mm in diameter, and resected with a hot snare from the sigmoid colon (Figure 1). After the procedure, the patient had no complications and was discharged home. The histology of the biopsied 4 mm lesion revealed a solitary small sessile colonic polyp in the lamina propria (Figures 3 and 4).

They range from nearly normal ganglion cells with large round nuclei, prominent nucleoli and abundant cytoplasm to abnormal ganglion cells with hyperchromatic nuclei with irregular nuclear membrane, invisible nucleoli, and scanty cytoplasm.

Scattered spindle cells are also seen. Both of them are highlighted by immunostain S100 (Figure 5), but negative for AE1/AE3 and EMA (markers for 69 epithelium) or CD117 (the marker for GIST). No mitosis or cell necrosis is seen. Diagnosis of ganglioneuroma was subsequently made by the pathologist.

DISCUSSION

Colonic GNs are classified as hamartomatous polyps that are composed of ganglion cells, nerve fibers, and enteric nervous system cells. The GNs of the gastrointestinal tract can be classified into three groups based on the size and the number of polyps: polypoid GN, ganglioneuromatous polyposis, and diffuse ganglioneuromatosis [1].

All three types of GNs are usually incidental findings found on colonoscopy and typically do not present with symptoms. Polypoid GNs, which can be sessile or
pedunculated, are small and appear to be very similar to hyperplastic and adenomatous polyps endoscopically [1]. This subset is most commonly seen in patient with Cowden’s syndrome, characterized by ganglioneuromas found in the breast, thyroid, genitourinary tract, and mucocutaneous areas [2].

Ganglioneuromatous polyposis is seen most commonly in patients with MEN IIB, NF1, Cowden’s syndrome, often accompanied by more than 20 sessile or pedunculated polyps [3]. Lastly, diffuse ganglioneuromatosis involves proliferation of neuronal cells in the entire colon, but does not extend into the ileum. Polyps can be as large as 17 cm in diameter with a variable (intramural or transmural) penetration into the colonic wall. They are seen as a component of MEN IIB7 and NF1 [4]. Histologically, the polyp is confirmed to be a GN by immunohistochemical staining with S100 protein confirming the presence of ganglion cells. The histological features of isolated polyoid GNS show disturbed crypt architecture and expanded lamina propria at low magnification. Higher magnification demonstrates the spindle cells in the fibrillary matrix and irregular groups of ganglion cells within the expanded lamina propria. The isolated GNS may also present submucosal extension and a plexiform-like arrangement involving submucosal nerve plexus. This pattern is suggestive of neurofibromas, but the presence of ganglion cells distinguishes them from neurofibromas. The GNS in ganglioneuromatous polyposis have overlapping features with isolated GNS. However, they are more variable and consist of more numerous ganglion cells. Diffuse ganglioneuromatosis may exhibit fusiform expansion of the myenteric plexus or confluent transmural ganglioneuromatous proliferations involving nerve fibers, ganglion cells, and supporting cells of enteric nervous system. The management of GNS depends on the patient’s clinical history and presentation. For the polyoid subgroup, polypectomy is the cure, however, colectomy may be required for ganglioneuromatous polyposis and diffuse ganglioneuromatosis, particularly if the patient is symptomatic [5]. Rarely, large GNS may cause symptoms of abdominal pain, constipation, obstruction, or bleeding secondary to the size and location within the colon. In general, solitary polyoid GNS are asymptomatic [6, 7].

**CONCLUSION**

To our knowledge, no guideline is available for repeat colonoscopy, although there are reports of association
with tubular adenomas. The finding of an asymptomatic, solitary ganglioneuroma in our patient, does not warrant more frequent colon cancer screening given its benign nature.

*********

Author Contributions
Michael Herman – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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REFERENCES
Osmotic demyelination affecting extrapontine areas of brain

Uduman Ali Mohamed Yousuf, Heng Siang Ting, BM Yashodhara, Shashikiran Umakanth

ABSTRACT

Introduction: Inappropriate fluid management in sick patients has a rare potential consequence called osmotic demyelination syndrome (ODS); either central pontine myelinolysis (CPM) or extrapontine myelinolysis (EPM) or combination of both. As reported in the studies, the incidence of these varies from 0.05–5.7% as per the different autopsy studies. The clinical presentation is variable from “Locked in syndrome” to seizures, behavioral and personality changes. Case Report: A 58-year-old ADL (activities of daily living) independent, apparently healthy, social and friendly female presented with aggressive behavior, excessive talking and disorientation after being treated for vomiting and diarrhea in a hospital. All these symptoms and events occurred after initial treatment for pneumonia. Her CT scans of brain were normal and had normal blood reports on thorough assessment. Conclusion: Prompt evaluation with EEG and MRI scan clinched the diagnosis of osmotic demyelination syndrome (ODS). We report one such presentation in the patient who had complete recovery at one year of follow-up.

Keywords: Behavioral disturbance, Extrapontine area of brain, Osmotic demyelination, Sodium

INTRODUCTION

Osmotic demyelination syndrome (ODS), presenting either as central pontine myelinolysis (CPM) or extrapontine myelinolysis (EPM) or combination of both, is a rare non-inflammatory demyelination disorder involving pons and other areas of brain and occurs as a consequence of rapid correction of hyponatremia. It was renamed later when the pathologic findings of central pontine myelinolysis (symmetric area of myelin disruption) was also found in extrapontine area [1]. In a study done in Japan, out of 1000 consecutive autopsies done, of which 626 brains were examined, 37 cases were found, giving rise to the incident rate of 5.7% [2]. In a different larger scale retrospective study that was conducted, the incidence rate was found to be 0.05% (15 cases in over 3000 autopsies done). Clinically, asymptomatic CPM found at autopsy has always been at least as frequent as cases diagnosed premortem and serves as a reasonable indicator for the incidence of the disease [3].
CASE REPORT

A 58-year-old female, apparently healthy retired kindergarten teacher, was referred to the hospital for aggressive behavior, non-stop and irrelevant talking and reduced sleep for 3–4 weeks. There were no known medical illness except for thyroidectomy 20 years back and hypertension. She was ADL independent, not ill-tempered social and friendly before the onset of the illness and has many friends, as revealed by her daughter. Prior to this incident, the patient had fever, cough with yellowish sputum production for a few days, for which she was treated in a private hospital with i.v. antibiotics. Later, while she was still in the private hospital, she had profuse vomiting and diarrhea for one week. She received i.v. antibiotics, i.v. fluids, i.v. anti-emetics and i.v. pantoprazole during her stay hospital. A week after her stay in the hospital, she had an episode of tonic movements, developed blank stare and CT brain was done and it was reported as normal except for mild cerebral atrophy (Figure 1). She was moved from general ward to intensive care unit, in that hospital, in view of these developments while diarrhea and vomiting resolved. She was observed for one more week in general ward before she was discharged. She required sedation to sleep during her stay in that hospital as noted by the daughter. At discharge from that hospital, the patient would talk incoherently. During her stay at home for two weeks, the patient was able to feed and dress herself, and she could, recognize family members in the beginning. Two weeks after discharge patient developed symptoms of sleeping difficulties, abusive behavior and physical aggression. The day before admission to our hospital the patient needed to be physically restrained at home. There was no history of rash, neck pain, headache, UTI symptoms and memory disturbances.

Past history: Apparently healthy after thyroidectomy 20 years back. She has been on treatment for hypertension, had no history of psychiatric, neurological disorder or any drug allergy in the past. There was no history of recent travel to other places or jungle trekking.

The patient was married and had four children and all were healthy.

On examination the patient was restless, talks incoherently, answers only a few questions, no eye contact, not oriented to time, place and person, Glasgow coma scale was: E4, V4 M5. There was no neck stiffness and Kernig’s sign was negative. There were no rashes on skin. Cardiovascular, respiratory and abdominal examinations were normal. Central nervous system reflexes were increased bilaterally and planters were down going. Power: Normal in all limbs. There were no cranial nerves palsies, no cerebellar signs. Repeat CT scan of brain done in the emergency department was normal. Neurologist was consulted and a possibility of Herpes simplex/encephalitis was considered. All the listed differentials, as mentioned in Table 1, were systematically ruled out.

Table 1: List of differential diagnoses

- Acute delirium
- Meningoencephalitis
- Herpes simplex encephalitis
- Seizures with post epileptic confusion
- Metabolic encephalopathy
- Intracranial Space occupying lesions
- Dementia with delirium
- Stroke

On reviewing the reports from previous private hospital it was found that she had received IV augmentin 1.2 g BD for 5 days, IV azithromycin 500 mg OD for 3 days for pneumonia. Workup for Legionella and Mycoplasma were negative. We also noted from the previous hospital records that she had severe hyponatremia (sodium 107 mEq/L), following vomiting and diarrhea. During admission to that hospital her serum sodium was found be rapidly normalized to 128 mEq/L in 24 hours by i.v. 3% saline (▲ 21 mEq/L) and also, she received i.v. ceftriaxone 1 g BD for 5 days for diarrhea and for altered sensorium. We did not find serum osmolality and urine osmolality reports from previous hospital; however, these were normal in the present admission at our hospital. In view of these findings, an EEG and MRI brain scan were done. EEG showed bilateral cerebral dysfunction with
excessive bilateral frontotemporal beta wave activity and MRI scans were suggestive of osmotic demyelination syndrome; T1 weighted MRI scans showed bright signals over basal ganglia and hippocampus on both sides. Also, there were hyperintense areas over right occipital and right parietal lobes (Figure 2). T2-weighted scans showed hyperintense signals over basal ganglia and swollen hippocampus bilaterally (Figure 3). Her serum sodium reports were normal after discharge from previous private hospital. The cause of severe hyponatremia was probably due to severe vomiting and diarrhea. She was not on hydrochlorothiazide, did not have hypothyroidism, primary polydipsia, cortisol deficiency, renal disease or SIADH as revealed by investigations.

Investigations

Full blood count, routine urine examination, LFT, RFT, CRP, ESR, serum calcium, serum magnesium, and ABG were normal. Serum sodium, urine and serum osmolality were normal. CT scan of brain was normal (Figure 1). Blood glucose levels were normal. Hepatitis B, C and HIV tests were negative. EEG showed bilateral cerebral dysfunction with excessive bilateral frontotemporal beta wave activity. The MRI scan of brain (Figures 2 and 3): showed features consistent with osmotic demyelination in extrapontine locations in frontotemporal areas. Investigations for the cause of hyponatremia were also done. Her Thyroid function tests and morning serum cortisol were normal.

Treatment

She received neurobion, amlodipine and respiridone tablets, while she was in hospital. The family was counseled and subsequently the patient was followed in neurology outpatients department.

Outcome and follow-up

- She was under neurologist’s follow-up in the outpatients department. Three months after discharge on the said medications (vide supra) sleep was normal, patient was alert, walks without support, not aggressive in behavior, but talks irrelevantly.
- Six months after the discharge, the patient was able to sleep well, ADL independent, cheerful. The daughter was happy with the improvements.
- Ten months after discharge patient had fully recovered.
- One year after the discharge, the patient is completely normal.

DISCUSSION

Osmotic demyelination syndrome most frequently occurs in adults, [4] but multiple cases have also been reported in children [5]. There are many associations between ODS and other medical conditions like alcoholic, [1] liver transplant, [4] end stage renal diseases, [6] and burn patients (Table 2) [7]. However, the striking similarity found between all is a rapid change in serum osmolality, mainly due to an over rapid correction of hyponatremia, [7, 8] typically followed an elevation in serum sodium > 20 mEq/L/24 hours, [9] although rarely it had been reported to be associated with hypokalemia, [10, 11] ODS is a consequence of a hyperosmotically induced demyelination[1]. Its exact pathophysiology is poorly understood, but an experiment mainly involving hyponatremia in rats led to the hypothesis that osmotic injury caused by over rapid correction of hyponatremia is the main cause ODS. Most of the change of brain osmolality in chronic hyponatremia can be accounted by
Table 2: Causes of hyponatremia

<table>
<thead>
<tr>
<th>Hypovolemic causes:</th>
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<tbody>
<tr>
<td>• Dehydration</td>
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<tr>
<td>• Vomiting</td>
<td></td>
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<tr>
<td>• Diarrhoea</td>
<td></td>
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<tr>
<td>• Diuretics</td>
<td></td>
</tr>
<tr>
<td>Hypervolemic causes:</td>
<td></td>
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<tr>
<td>• Congestive heart failure</td>
<td></td>
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<tr>
<td>• Cirrhosis of liver</td>
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<tr>
<td>• Nephrotic syndrome</td>
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<tr>
<td>Euvolemic causes:</td>
<td></td>
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<tr>
<td>• SIADH</td>
<td></td>
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<tr>
<td>Other causes:</td>
<td></td>
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<tr>
<td>• Beer drinking/Chronic alcoholism</td>
<td></td>
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<tr>
<td>• Sertraline</td>
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<td>• Lithium</td>
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The changes in organic osmolytes and brain electrolytes; and rapid correction of hyponatremia is associated with an overshoot of brain sodium and chloride levels along with a low organic osmolyte level. The high cerebral ion concentrations in the absence of adequate concentrations of organic osmolytes may be relevant to the development of central pontine myelinolysis [12, 13]. The clinical features of ODS typically range in severity from mild and transient confusion to severe spastic quadriaparesis, pseudobulbar palsy, and impairment in the level of consciousness [9]. Rarely, in the cases where central pons are involved, patients can present as ‘locked in syndrome’. Extrapontine involvement however can present with psychiatric symptoms like catatonia, hallucinations, behavioral or personality change can easily lead to a misdiagnosis [1]. Moreover, it can be difficult to diagnose because the onset of clinical features can be delayed for 2–6 days after rapid correction of hyponatremia, [9, 14] as in our patient. High index of suspicion is needed to make a diagnosis especially if a patient with normal electrolytes presents with neurological complaints long after correction and previous history of rapid correction of hyponatremia may not be clearly evident, exemplified in our case study. Diagnosis is made with imaging studies mainly MRI scan, where myelinolysis lesions can be found at pontine or extrapontine areas [15]. The lesions appear hyperintense on T2-weighted and FLAIR MRI images. These lesions do not enhance with GDTPA. In our case, the demyelinations were found in extrapontine areas. EEG may show non-specific slow wave activity consistent with metabolic disorder as in our case. CT scan can be used occasionally to diagnose ODS with its typical hypodense lesions in pons, [16] however has been largely replaced by MRI scan due to the higher sensitivity [17]. It may take as long as 4 weeks for MRI scan to become positive after disease onset [15] hence making it worthwhile to repeat neuroimaging studies 10–14 days to confirm diagnosis. Latest technique of diffusion-weighted imaging has an advantage of earlier diagnosis whereby patient could be diagnosed within 24 hours of onset of symptoms [18]. Since there is no established treatments for ODS other than supportive management, [1] prevention becomes the mainstay of treatment. Rate of correction of hyponatremia needs to be closely regulated, especially in case of chronic hyponatremia and severe hyponatremia at time of presentation. The proposed rate of correction of hyponatremia is <8 mmol/L per 24 hours [19] and <12 mmol/L in 48 hours [20]. In terms of more careful and forethought approach as reported by the recently published “expert panel recommendations” would be to restrict the elevation of serum sodium to 4–6 mmol/L in 24 hours [20]. In case if there is any inadvertent elevation of serum sodium more than above recommended values does occur, re-induction of hyponatremia could be done by using intravenous 5% dextrose or desmopressin or combination of both. Other pharmacological approaches to prevent the myelin damage following over-correction of hyponatremia could be usage of any one the agents; corticosteroids, intravenous immunoglobulin, plasmapheresis, minocycline, urea and myoinositol. Besides, these measures physical therapy and symptomatic treatment could be used [20]. In our case, re-induction of hyponatremia was not done, as there was a gap of 3–4 weeks after rapid hyponatremia correction had happened in previous hospital. Only correct diagnosis, symptomatic treatment, family counseling and physical therapy were offered to the patient in our hospital. The prognosis is not that grave as in olden times of first recognition of this condition in mid-1970s. Mortality as low as 6% and recovery as high as 40% was reported in 44 German patients with ODS [20].

**CONCLUSION**

The formulae that are used to calculate and correct the serum sodium in hyponatremia are not accurate in replenishing dynamically changing serum sodium in a patient. The prevention of osmotic demyelination syndrome (ODS), central pontine myelinolysis (CPM) or extrapontine myelinolysis (EPM) can be done by frequent monitoring of serum sodium and its gradual correction. Close and frequent monitoring of serum sodium is vital in the prevention of ODS.

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**Author Contributions**

Uduman Ali Mohamed Yousuf – 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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Shashikiran Umakanth – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

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ABSTRACT

Introduction: Abdominal incisional herniae are well known complications after abdominal surgery and many methods for repair of the hernial defect have been tried and developed. These include open repair with or without mesh and laparoscopic repair. Case Report: We present an extremely unusual case where the hernia sac had dissected a plane between anterior sheath and the rectus muscles. Surgical development of this plane is associated with great risk of damage to nerves and vessels therefore not commonly used in hernia repairs. Conclusion: In this highly unusual case where traditional mesh placement was of little value, we chose a unique and as yet undescribed method of retro-anterior sheath mesh placement with good results at third month.

Keywords: Abdominal wall, Hernia, Hernia repair, Mesh

INTRODUCTION

An abdominal incisional hernia is a defect in the abdominal wall at the site of previous surgery and occurs with an incidence of 15% [1]. This may result in the abnormal protrusion of any part of an abdominal viscus and can lead to incarceration and strangulation. In addition to technical failure, there are several factors that affect a patient's risk of developing an incisional hernia which include an elevated body mass index and advanced age [2]. Prior to the development of polymerized meshes, incisional hernia were repaired by primary closure. Sutured repairs had the disadvantage of a high recurrence rate [3]. Comparative studies have found a 10-fold increase in recurrence following sutured repair when compared to mesh reinforcement [4].

Techniques of ventral wall mesh reinforcement utilize the different anatomical layers of the ventral wall. The ‘onlay’ technique involves fixing the mesh to the anterior aspect of the rectus sheath, directly beneath Scarpa’s fascia. It has a relatively high recurrence rate [5]. The ‘inlay’ technique, in which the mesh is secured to fascial edges between the two recti is best avoided due to the high rate of failure and need for re-intervention with quoted recurrence rates of up to 44% [6]. The ‘sublay’ technique of reinforcement is traditionally felt to be the technique with the most durable results. Documented recurrence rates are in the region of 3.6% [5]. In this
repair, the mesh is placed between posterior sheath and rectus muscles. With the evolution of the laparoscopic era the ‘intra-peritoneal onlay mesh’ (‘IPOM’) has more recently documented durable repairs [1]; at present long-term recurrence and patient satisfaction data are awaited.

In this case report, we describe the acute presentation and subsequent management of an elderly lady with a complex ventral intraparietal incisional hernia. Conventionally, retro-anterior-sheath placement of a synthetic mesh in ventral wall hernia repair is not undertaken. The risk of damage to vascular and nerve perforators in this plane, between rectus muscles and anterior sheath, is generally felt to preclude a safe repair. However, in this unusual case, the herniation of the small bowel had almost completely dissected the anterior sheath away from the anterior rectus muscle. The result was the development of a plane that is normally inaccessible which can be easily accommodated synthetic mesh (Figures 1 and 2).

**CASE REPORT**

A 78-year-old female was admitted with a large irreducible right iliac fossa incisional hernia with associated features of bowel obstruction. The incisional hernia was a complication of a previous Pfannenstiel incision for an extended abdominal hysterectomy for an endometrial malignancy. At the time of admission she was awaiting an elective laparoscopic hernia repair.

Following initial presentation, assessment and resuscitation, a computed tomography (CT) scan was performed. The CT scan confirmed the clinical suspicion of an acutely incarcerated ventral incisional wall hernia with threatening bowel strangulation. She subsequently underwent an emergency laparoscopy. This was converted to a full midline laparotomy early in the procedure due to the presence of small bowel within the hernia sac too adherent to be safely mobilized laparoscopically. At a specific point over the incision the anterior sheath had a 5x6 cm hernia defect which contained chronically obstructed but viable small bowel. The hernial defect was found to pass between the widely separated recti muscles and had dissected the anterior sheath away from the recti. Ventral wall reinforcement was achieved by suturing the rectus abdominis muscles together and by securing a DynaMesh anterior to the anterior rectus abdominis muscle but behind the anterior rectus sheath. After excision of the hernial sac, the anterior sheath was primarily closed with polydioxanone (PDS), with two Redivac ® drains (B. Braun Medical Ltd, Sheffield, U.K.) placed either side of the sheath in an attempt to reduce seroma formation, before finally closing the skin with clips.

Postoperatively, the patient recovered well, with no immediate complications and was discharged a few days after surgery. Review in outpatient department after one month revealed an excellent result with no evidence of recurrence. A focal point of overgranulation and mild serous discharge was treated in clinic with a silver nitrate cautery stick. Further review at third months showed complete resolution of the discharge, a sound repair of the hernia with no evidence of recurrence. Additionally, the level of satisfaction was high and the patient was subsequently discharged.

In this case, we strayed from traditional techniques for open hernia repair with mesh reinforcement. We placed the mesh in a retro-anterior plane which, to the best of our knowledge, has not previously been done.

**DISCUSSION**

Incisional herniae are a well-recognized complication of abdominal surgery. Predisposing risk factors include technical failure, patient age, sex, elevated body mass index and diabetes mellitus [2], approaches to hernia repair and ventral wall reinforcement include both conventional open techniques and more recently, as skills
and mesh design have evolved, laparoscopic methods of hernia reduction and intra-peritoneal mesh placement have become more commonplace. A Cochrane review found no difference in incidence of recurrence when comparing laparoscopic to open repairs but did elucidate a reduction in wound infection rates with laparoscopic repairs [1]. However, some studies have reported an increased risk of bowel injury during laparoscopy [7].

The development of synthetic meshes has revolutionized hernia surgery and significantly improved recurrence rates and patient satisfaction [1, 3, 8]. In open repairs, ventral wall reinforcement with onlay, inlay and sublay meshes have varied results. A Cochrane review from 2008 found no significant difference in recurrence rate when comparing sublay and onlay mesh position. However, found the mean operative time to be shorter in the sublay position [9]. In contrast to this a meta-analysis comparing sublay to onlay mesh repair from 2014 found sublay to be slightly superior both with respect to recurrence rate and surgical site infections. 1,948 patients were included in the meta-analysis [8]. In a randomized controlled study from 2011, comparing sublay to inlay hernia repair, with 100 patients in each group, sublay was also found to be preferable when comparing recurrence rates and complications between the two groups [10].

In this case, plane obliteration precluded securement of mesh in the traditional planes. Sublay placement was not possible as the hernial defect was lower than the posterior sheath. Similarly, inlay was not considered an option due to the extent of hernial dissection of the fascial layers off the recti muscles. Onlay placement of a synthetic mesh was considered feasible. However, as the retro-anterior sheath plane needed little development compared to the “onlay plane” between subcuticular fat and the anterior sheath it was felt that the alternative placement of the mesh would be advantages in this instance. After searching literature, we have been unable to find any previous description of a mesh placement in this plane.

CONCLUSION

In this extremely unusual case, we found the more conventional option of hernia repair with mesh (inlay, onlay and sublay) to be of limited value and instead chose a unique and as yet undescribed method of retro-anterior sheath mesh placement with good and safe documented results at the third month.

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Author Contributions

Susanne Irene Scott – 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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REFERENCES


Heterotopic pregnancy: Cause of a rare acute abdominal emergency

Birge Ozer, Ozgur Nazan, Erkan Mustafa Melih

ABSTRACT

Introduction: Heterotopic pregnancy is defined as the coexistence of intrauterine and extrauterine gestation. The incidence rates are higher in induced ovulation patients compared to spontaneous conception. Intrauterine gestational sacs can be easily overlooked during a routine ultrasonography examination if the physician is not paying enough attention for the accompanying ectopic pregnancy possibility. Case Report: Our case is a female previously operated with dilatation and curettage (D&C) on eighth week of gestation in another hospital. She came to our clinic with an acute abdominal emergency and was taken to surgery immediately after seeing an ectopic gestational sac during the examination. Herein, we would like to present this well-managed and treated case admitted to our hospital for acute abdominal emergency following termination of a spontaneously developed anembryonic pregnancy. Conclusion: Intrauterine gestational sacs can be easily overlooked during a routine ultrasonography examination if the physician is not paying enough attention for the accompanying ectopic pregnancy possibility. The clinical presentation of the patient plays a key role in choosing a treatment method.

Keywords: Acute abdomen, Anembryonic pregnancy, Emergencies, Heterotopic pregnancy

INTRODUCTION

Heterotopic pregnancy is defined as the simultaneous development of an intrauterine and an extrauterine gestational sac. Spontaneous heterotopic pregnancy was considered to be very rare with an incidence of 1 in 30,000 pregnancies [1]. The incidence rates are higher in patients with induced ovulation in comparison with spontaneous conception. Intrauterine sacs can be easily overlooked if the physician is not paying enough attention for the accompanying ectopic pregnancy possibility. Using serial hCG measurements for follow-up is redundant since the intrauterine pregnancy keeps hCG levels increased. The treatment for ectopic pregnancy is surgical intervention. After surgically removing ectopic sac, intrauterine pregnancy proceeds normally in most of the patients. There are also other techniques such as transvaginal or laparoscopic KCl injections. The clinical situation of the patient during initial admission defines the chosen therapeutic method. Early diagnosis is especially important in those type of pregnancies since it
directly affects morbidity, mortality and future fertility of the patients.

**CASE REPORT**

A 21-year-old female admitted into emergency room of Ağrı Obstetrics and Gynecology Hospital with syncope, nausea, emesis and vaginal hemorrhaging in staining character. She was not having her period for 2 months and was diagnosed with anembryonic pregnancy in another private medical clinic. Eight hours before admission, the patient was treated with dilatation and curettage (D&C) under elective conditions in the same medical clinic she was diagnosed without any complications and the patient was discharged after the operation. Postoperatively 8 hours, the patient gradually started to feel unwell and finally was brought to the emergency room of our hospital. The medical history stated that this was her first pregnancy and she had no history of a coexisting illness or medication. Following admission, no active hemorrhage was diagnosed during speculum inspection but routine hemorrhage in staining character was common following such procedures. Abdominal examination showed a widely distributed tenderness in all quadrants during palpation and rebound finding was prominent in lower quadrants. Dyspnea and palpitations were seen in supine position. Ultrasonography (USG) examination showed a normal uterus, endometrium thickness (10 mm) and normal bilateral ovary structures. Disseminated dense fluid retention in a hemorrhagic fashion was detected in Douglas and paraovarian areas in addition to intestinal segments. The image was consistent with an embryo in yolk sac with a gestational age of 6 weeks and six days in ampullar portion of right fallopian tube. (Figures 1 and 2). The patient vital signs were blood pressure 80/40 mmHg, pulse 98/minute and hemoglobin was 7.8 g/dl, HTC 22, Platelet count 182000 and WBC 14000. Patient was closely followed and the USG images taken before D&C procedure were obtained from the patient. The images revealed a gestational sac consistent with an anembryonic pregnancy measuring 33 mm located in uterine fundus and the procedure was actually a D&C for abortion (Figures 3 and 4). Patient’s relatives were notified of the medical situation of the patient and our initial diagnoses were either a uterine rupture or a coexisting pregnancy of a heterotopic type. Laparotomy was planned after assessment of the clinical situation of the patient and risks and details of the procedure was shared with the patient. After obtaining 3 units of erythrocyte suspension, the abdominal access was done using a Pfannenstiel incision. Dense disseminated fluid was detected and about 2 liters. of this hemorrhagic fluid was aspirated. After aspiration, no signs of perforation was found on the front or rear surface of the uterus. In tubal structures inspection, left tube and ovary was normal but right tube had an actively bleeding gestational sac about 2 cm in diameter on ampullary portion and a full thickness perforated tubular structure confirmed our diagnosis of heterotopic pregnancy (Figure 5). Salpingostomy was chosen after considering patient’s age and gravidity. The tube was repaired using 3-0 Prolene sutures after removal of all trophoblastic structures. Operation was finished after checking for hemorrhage and an abdominal drainage tube was inserted for follow-up. Three units erythrocyte suspension was used during the operation. Within 48 hours in postoperative period, 50 cm³ serohemorrhagic fluid was collected in the drain. The abdominal drainage tube was removed on the third day of operation and patient was discharged from the hospital with full recovery. The patient was advised to go to regular follow-up in gynecology and obstetrics clinic. β-hCG values dropped below 5 and no complications were seen in incisional scar area. She was also informed about the possible risks that might occur in future pregnancies.

![Image 1: Ectopic pregnancy discerned in right tubal structures.](image1.png)

![Image 2: Expansive fluid seen in abdomen of the case admitted with acute abdominal findings.](image2.png)
Heterotopic pregnancy is the development of intrauterine and ectopic gestations at the same time, but in different places [2]. First incidence rate of this condition is reported as 1/30000 in Devoe and Pratt’s study done in 1948. This incidence rate was revised to 1/3889 after new analysis methods and this went up to 1/100 with assisted reproductive techniques [2, 3]. Our main objective in this study is to present the diagnosis and treatment stages of such a rare case of tubal and intrauterine spontaneous heterotopic pregnancy with literature review. Although assisted reproductive techniques increased incidence rates dramatically, heterotopic pregnancy is still a very rare condition after spontaneous pregnancy. Predisposing conditions for this condition are the same for ectopic pregnancy; which are pelvic adhesion, previous tubal damage, previous tubal surgery and STD history. Early diagnosis of this condition is crucial since it plays an important role in morbidity and mortality of the patient as well as future pregnancies. Maternal mortality is around 1% and fetal intrauterine mortality varies between 45–65%. Each case deserves a different approach. Beta-hCG levels, progesterone levels and USG are all important for both diagnosis and follow-up. Serial progesterone level tracking can be used to determine a bad pregnancy prognosis. However, serial β-hCG level tracking is useless in those cases because of the ongoing intrauterine pregnancy. Diagnosis of a uterine pregnancy is relatively easy using ultrasonography, however gestational sac in adnexial region and fetal heartbeat visualization is rather difficult and rare. In addition, visualization of an intrauterine pregnancy might cause the obstetrician to overlook an ectopic pregnancy. Therefore, it is important to check carefully for an extratubal pregnancy using vaginal ultrasonography scans during the first trimester in patients that became pregnant with assisted reproductive procedures. Nonetheless, less than 50% of the patients were diagnosed during USG. Most of the patients were diagnosed during emergency laparotomy procedures after they become symptomatic. Likewise, our patient was also diagnosed after clinical symptoms were seen. It is very important to keep the possibility of a heterotopic pregnancy in mind during first 14 weeks in patients with abdominal pain and hemorrhage, especially in pregnancies occurred after IVF and intrafallopian transfer of gamets. Diagnosis rate before surgery is only 0% [4]. Patients usually present to emergency rooms with acute abdominal emergency findings due to delays in diagnosis of the condition. Intrauterine pregnancy should be differentiated using TV-USG before surgery for ectopic pregnancies in patients who underwent IVF and ovulation induction. Even after no intrauterine pregnancy was visualized, no curettage should be performed on these patients due to potential intrauterine pregnancy. Acute cases indicate laparotomy surgery to maintain existing intrauterine pregnancy. The operation should be completed with minimal anesthesia and minimal trauma to uterus. If there is a missed abortus situation, laparoscopy should be done for evaluation of tubes, especially in patients in which pelvic pain is the main symptom and complaint. Failure in diagnosis might cause higher mortality rates, major blood loss and a hindrance to conservative tubal surgery. Although TV-USG increases the odds for
correct diagnosis, laparoscopy is still one of the most credible methods. Today, laparoscopy almost ruled out laparotomy in heterotopic pregnancy cases. However, laparotomy was performed in this patient because of a rapid progression of acute abdominal symptoms and a rapid deterioration in hemodynamic parameters. There is a possibility that an existing ectopic pregnancy might ruin the intrauterine pregnancy by forming a hematoma. Our case was a patient that was operated previously in another hospital and no patient file existed before her initial admission to the clinic in our hospital. Ultrasonography images before curettage showed a normal structured but anembryonic gestational sac consistent with 8–9 weeks of age. If there is a missed abortus case, laparotomy should be considered in those patients, especially with pelvic pain complaints after curettage, for evaluation of fallopian tubes and ectopic pregnancy using TV-USG and β-hCG results. Diagnosis delay brings out an increased mortality risk, major blood loss and poses a hindrance to normal tubal surgery. In heterotopic pregnancies, those treated with laparotomy have 9% abortus or stillbirth, 16% preterm delivery and 75% term delivery [5]. During operation course, it is essential to maintain ovarian blood supply [6]. Reecce et al. stated continuing and healthy delivered pregnancy rates to be 75% whereas Molley et al. reported the same rate as 60% [5, 7]. Even though surgical interventions such as laparotomy and laparoscopy are the most used treatment modalities, transvaginal embryo aspiration can only be performed after local methotrexate and potassium chloride injections to ectopic sac with the aid of USG during the process [8, 9]. However, the detrimental effects of methotrexate, RU486 or prostaglandins on intrauterine pregnancy states that those techniques should not be used [10]. In acute cases requiring laparotomy, operation should be completed with minimal trauma to uterus and minimal anesthesia. Our case underwent a curettage operation before admittance and came to our hospital’s emergency room with suddenly increasing emergency acute abdominal symptoms. Today, laparoscopy ruled out laparotomy in the diagnosis and treatment of symptomatic heterotopic pregnancies [11, 12].

CONCLUSION

In all pregnancies, uterus and bilateral ovaries should be thoroughly evaluated as soon as pregnancy is revealed and heterotopic pregnancy possibility should always be kept in mind in patients with hemorrhage and abdominal pain, especially when other acute abdominal symptoms are present.

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REFERENCES


Cardiac electrical and biochemical abnormalities in acute stroke: A case report

M. Umair Bakhsh, Hassan Alkhawam, Feras Zaiem, Mohammed El-hunjul, Jasprit Takher, Anirudh Pareek, Robert Sogomonian

ABSTRACT

Introduction: Cardiac troponin T (cTnT) is a highly sensitive and specific marker of myocardial necrosis that aid in diagnosis of myocardial infarction. The cTnT elevations along with ECG changes noticed in patients with acute Stroke without major cardiac events. Case Report: We report a case of 87-year-old female presented with cough, lethargy, decreased oral intake, shortness of breath, chest pain and altered mental status. Head computed tomography (CT) scan did not reveal any acute intracranial pathology. Laboratory work showed cardiac troponin T elevation with ECG changes were concerning for cardiac ischemia. Careful examination was significant for right facial droop and right sided weakness. A repeat head CT scan showed acute stroke. Conclusion: Patient with ischemic stroke may have shown to have an association with elevated cTnT. They are at an increased risk of mortality from renal and/or cardiac failure. Though ECG changes and cTnT levels can be attributed to stroke, given the common risk factors shared by coronary artery disease and stroke, a cardiac work up is warranted to rule out any acute myocardial event or look up for the source of stroke which most likely to be cardiac.

Keywords: Acute stroke, Cardiac, Coronary artery disease, Myocardial infarction, Stroke, Troponin

INTRODUCTION

Cardiac troponin T (cTnT) is a highly sensitive and specific marker of myocardial necrosis that aid in diagnosis of myocardial infarction. The cTnT elevations along with ECG changes have been reported previously in patients with acute Stroke (ischemic or hemorrhagic) [1]. Literature search demonstrated that Electrocardiographic (ECG) changes might be seen with acute stroke. Various mechanisms have been set forth to validate the relation between acute stroke and troponin elevation with ECG changes which are implied to the increase in catecholamine, which in turn, lead to myocyte injury [2–4]. Elevated troponin is seen after stroke and is associated with increased mortality [5]. We reported a case and then reviews different mechanisms involved in troponin elevation in the setting of acute stroke.
CASE REPORT

We report a case of a 87-year-old female with past medical history significant for hypertension, lymphoma status post neck radiation, aortic stenosis, dementia and glaucoma. This patient presented with cough for 3 days, lethargy and decreased oral intake. The patient reports no fever, chills, nausea, vomiting, diarrhea or palpitations. About four hours prior to admission, patient experienced shortness of breath and increased cough and chest pain for which she was taken to the emergency department where she was found to have altered mental status. Laboratory work showed cardiac troponin T elevation, cTnT was found to be 1.6 ng/mL (normal range 0.0 – 0.2 ng/mL). ECG changes were concerning for cardiac ischemia - T wave inversions in lead V1-V3, with no prior ECG for comparison (Figure 1). Head CT scan did not reveal any acute intracranial pathology. Given her negative head CT scan and ECG changes she was given heparin bolus, clopidogrel 600 mg, aspirin 325 mg and cardiologist consulted for possible catheterization.

Upon arrival patient was alert, afebrile with stable vital signs. Initial physical examination was significant for altered mental status but later on, patient was noticed has right facial droop and right sided weakness. A repeat head computed tomography (CT) scan showed right cerebellar hypodensity consistent with acute stroke. Thereafter, she became lethargic and the arterial blood gas showed severe acidosis with hypercarbia due to which she had to be intubated. Neurosurgery was consulted and she was started on hypertonic saline and dexamethasone for concern of cerebral edema and risk of possible midline shift of the brain.

Her cardiac troponin T (cTnT) on admission was 1.37 ng/mL, which peaked to 1.5 ng/ml and then finally trended down to 0.2 ng/ml. Patient’s creatine kinase (CK) MB isoenzyme level on admission was 6.3 ng/ml which trended down to 2.4 ng/ml (CK-MB normal range 0–3 ng/mL ). Her initial 12 lead ECG on admission showed T wave inversion in leads V1-V2, with mild elevations in V4-V6, however, serial ECGs showed resolution of T wave inversion and ST elevations (Figure 1).

Echocardiography (echo) showed normal left ventricular size with hyperdynamic left ventricular systolic function and moderate to severe aortic stenosis. Carotid Doppler showed small atherosclerotic plaques at both carotid bifurcations however there was no evidence of obstruction to flow.

The serial head CT scan of the patient showed acute right cerebellar infarction with interval increase from 2.5 to 3.3 cm. Magnetic resonance imaging (MRI) scan showed multifocal infarcts with largest involvement within the right superior cerebellum, also additional infarcts were seen in the left cerebellum and left parietal lobe and areas of petechial hemorrhages in the right superior cerebellar infarct. Serial ECGs were stable with down trending troponin, therefore cardiology team at our center considered not to perform any cardiology intervention.

DISCUSSION

Stroke is the fourth leading cause of death [6] and the leading cause of long-term disability in United States. Stroke is defined as an acute neurological condition that occurs either due to brain ischemia (thrombosis, embolism, systemic hypoperfusion) or brain hemorrhage (intracerebral hemorrhage or sub arachnoid hemorrhage). 80% of the stroke is ischemic in nature while 20% is hemorrhagic. The cTnT is a highly sensitive and specific marker for myocardial necrosis that is used in risk stratification of patients with acute coronary syndrome and is used in the diagnosis of acute myocardial infarction. There is a complex overlap between cardiovascular and cerebrovascular diseases. Interestingly, elevations of cardiac troponin levels and ischemic ECG changes have been described previously in the setting of acute ischemic stroke [1]. Patients with elevated troponin levels were more likely to have myocardial ischemia, as seen on the ECG [7].

Elevated cTnT have been reported in 5–34% of the patients with acute ischemic stroke. This was shown by another study, which enrolled 222 subjects, which showed that 20% of the patient had troponin > 0.2 µg/L. It was seen that these patient also had higher catecholamine
levels when measured and ECG changes were suspicious of acute myocardial infarction [8]. Patients with elevated cTnT had evidence of myocardial damage in 10–34% of patients with acute ischemic stroke[9–13].

The mechanism associated with acute ischemic stroke causing cTnT elevation and ECG changes is not fully understood. A few possible explanation have been set forth describing this association. One possible explanation is associated with loss of the inhibitory effect of sympathetic stimulation that is exerted by the insular cortex. Right insular cortex maintains its inhibitory effect by suppressing the sympathetic system. Once this area is affected there is loss of sympathetic inhibition leading to catecholamine surge in the body which can lead to myocyteolysis [14]. This subsequent catecholamine surge may also induce LV dysfunction with LV apical ballooning [2–4]. Imaging showed that patient with raised troponin have stroke that more commonly involved the right posterior, superior, and medial insula and the right inferior parietal lobe exerting loss of inhibition of sympathetic pathway by insular cortex [13].

Another possible explanation of the elevated cTnT in stroke patient can be attributed to the common risk factors that are shared by stroke and myocardial infarction and increased prevalence of coronary artery disease in stroke patient [15]. Also, any stressful condition such as stroke would trigger the body’s natural response to release glucocorticoids and catecholamine that might be an additive factor contributing to elevated cTnT in the setting of stroke.

Patients who were admitted with an acute ischemic stroke, serum cTnT level at the time of admission was a powerful predictor of mortality [5]. Furthermore, patient with elevated cTnT had a higher frequency of heart and/or renal failure as well as increased risk of mortality within the following two years [16].

CONCLUSION

Patient with ischemic stroke have shown to have an association with elevated cardiac troponin T (cTnT). They are at an increased risk of mortality from renal and/or cardiac failure. Multiple explanations have been set forth to define the association. Though ECG changes and cTnT levels can be attributed to stroke, given the common risk factors shared by coronary artery disease and stroke, a cardiac workup is warranted to rule out any acute myocardial event.

REFERENCES


Author Contributions

M. Umair Bakhsh – 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.
Is 20s too young to have triple coronary vessel disease?

M. Umair Bakhsh, Hassan Alkhawam, Feras Zaiem, Jasprit Takher, Anirudh Pareek, Mohammed El-hunjul, Robert Sogomonian, Neil Vyas

ABSTRACT

Introduction: Coronary artery disease (CAD) remains the leading cause of death in the US in both men and women. This is less frequent in younger population, however, when present CAD can have devastating consequences on the patient and the family. Case Report: We reported a case of a 28-year-old smoker male with a family history of CAD who presented with dyspnea on exertion. Laboratory examinations were normal for cardiac enzymes and negative for D-Dimer. On admission ECG showed sinus rhythm at 71 bpm, RSR' in V1, S wave in I, Q wave in III, T wave inversion in III. Echocardiogram showed right ventricular systolic function reduced mid portion, preserved at apex and base (McConnell's sign) consistent with acute pulmonary embolism. Patient was started on anticoagulation. Computed tomography angiography (CTA) was also done which was negative for any acute pulmonary embolism, and anticoagulation was discontinued in the light of negative CTA. The patient was discharged from hospital with follow-up in outpatient department. However, the patient came back to emergency department with worsening dyspnea. Lab work was significant for elevated troponin I and ECG on admission showed normal sinus rhythm and S1Q3T3 pattern. Patient had coronary angiography which showed three vessel disease. Conclusion: Smoking is conventionally recognized as the most common risk factor for heart disease. The CAD in young adults has a poor long-term prognosis. Smoking cessation remains a significant challenge in younger population. More awareness and cessation measures are required to address this emerging situation. Given lack of warning signs in these patients, physicians need to be more aggressive in managing risk factors as CAD in younger patient can have adverse outcomes.

Keywords: Acute coronary syndrome, Coronary artery disease, Vessel disease, Young population

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INTRODUCTION

Coronary artery disease (CAD) is the leading cause of death in both men and women. In United States about 600,000 people die of heart disease every year [1]. Coronary artery disease is the most common type of heart disease, killing close to 380,000 people annually. Every year about 720,000 Americans have heart attack, out of these, 515,000 experience their first while 205,000 already had a heart attack [2]. The burden of coronary artery disease is significant; it costs $108.9 billion dollars annually [3]. African American men are 30% more likely...
to die from heart disease than non-Hispanic white men [4]. In men, the risk for CAD increases after age 45 while in women, the risk for CAD increases after age 55. The CAD is less frequent in younger population, however, when present this can have devastating consequences on the patient and the family. Younger men with myocardial infarction, less than 40 years old were found to be heavy smokers with a high incidence of angiographically normal coronaries. As opposed to traditional risk factors for CAD, younger patient have a lower prevalence of hypertension, diabetes mellitus and hyperlipidemia. Smoking is recognized as the most common risk factor for CAD in younger men and is associated with myocardial infarction. A positive family history of premature CAD was significantly more prevalent in younger men. The readmission rate is high and it is associated with smoking and decreased ejection fraction.

**CASE REPORT**

A 28-year-old male, South Asian descent, active smoker with no prior medical history presented to the emergency department with dyspnea on exertion. Family history was significant for premature coronary arterial disease and dyslipidemia in her mother. The patient denies any recreational drug and was not on any medications.

Three months ago the patient arrived to USA and since then he had been having dyspnea on exertion, progressively getting worse. Three months ago he used to run two mile without any complaints, however soon after returning back from his country he started to get short of breath.

In the emergency department, the patient was afebrile with normotensive and stable vitals. ECG on admission showed sinus rhythm at 71 bpm, RSR’ in V1, S wave in I, Q wave in III, T wave inversion in III (Figure 1)

Examination unremarkable for any murmur or lower extremity swelling. Laboratory studies revealed normal troponin I and normal CK–MB levels. His D-dimer was normal <0.150 ug/mL (0–0.223). Echocardiogram showed normal left ventricular systolic function – EF 60%, RV dilatation, right ventricular systolic function reduced mid portion, preserved at apex and base (McConnell’s sign) consistent with acute pulmonary embolism. Patient was started on anticoagulation—Lovenox 1 mg/kg BID. Computed tomography angiography (CTA) was negative for any acute pulmonary embolism and anticoagulation was discontinued in the light of negative CTA. The patient was discharged with outpatient follow up with primary care clinic.

Three weeks after being discharged patient presented to the emergency department with worsening dyspnea on minimal activity, exercise tolerance limited to less than one block and one flight of stairs. Denied any chest pain, palpitations, lower extremity swelling. Patient was afebrile, normotensive and vital signs stable.

The ECG on admission showed normal sinus rhythm. S1Q3T3 pattern (Figure 2). He was given 81 mg x 4 aspirin and admitted to medicine service for further management. His lab work was significant for elevated troponin I to 0.217 ng/mL (0.00–0.10 ng/mL), however negative CK–MB. Cardiologist was consulted and patient was upgraded to CCU. His troponins I peaked to 1.884 ng/mL. Patient was started on heparin drip and eptifibatide drip for non-STEMI. Lipid panel showed cholesterol 342 mg/dL (0–200 mg/dL), triglyceride 460 mg/dL (0–150 mg/dL), HDL 25 mg/dL (23–92 mg/dL), LDL 190 mg/dL (0–100). HbA1C was 5.2%.

Given ECG findings with worsening dyspnea and elevated troponins patient had D-Dimer done which was negative (<0.150, normal 0–0.223 ug/mL) concern for pulmonary embolism. Patient had echo done which showed normal LV function, mild MR, RV dilatation and McConnell sign seen on prior Echo was no longer seen.

Given elevated troponin with worsening dyspnea cardiology team performed coronary angiography which showed three vessel disease, minimal LV dysfunction, no MR or AS. Successful PCI of LAD/D1 and OM1. Cardiology team recommended Aspirin, Plavix with staged PCI of AV continuation.

The patient was admitted to cardiology in November 14 for staged PCI of AV continuation. The patient was discharged home with successful PCI of AV continuation, angiography further showed patent stent in LAD and OM1.

![Figure 1: ECG on admission showed sinus rhythm at 71 bpm, RSR’ in V1, S wave in I, Q wave in III, T wave inversion in III.](image1)

![Figure 2: ECG on admission showed normal sinus rhythm. S1Q3T3 pattern.](image2)
He was discharged home on aspirin, plavix and statin. His symptoms improved after PCI and continues to follow-up with cardiology as outpatient.

**DISCUSSION**

Coronary artery disease remains the leading cause of death in the US in both men and women [1]. This is less frequent in younger population however when present CAD can have devastating consequences on the patient and the family [5]. Younger men with myocardial infarction, less than 40 years old were found to be heavy smokers with a high incidence of angiographically normal coronaries [6]. As opposed to traditional risk factors for CAD, younger patient have a lower prevalence of hypertension, diabetes mellitus and hyperlipidemia. Smoking is recognized as the most common risk factor for CAD in younger men and is associated with myocardial infarction [7, 8]. A positive family history of premature CAD was significantly more prevalent in younger men [9, 10, 11]. The re-admission rate is high and it is associated with smoking and decreased ejection fraction [12].

Echo done also showed McConnell sign. McConnell sign has been reported to have high sensitivity and specificity for diagnosing acute pulmonary embolism, however, recent reports have shown a poor predictive value for diagnosis of acute pulmonary embolism [13]. ECG showed S1Q3T3 which is not neither sensitive nor specific for pulmonary embolism with the most common finding being sinus tachycardia.

Stable angina and multivessel disease is uncommon in the young [14]. Younger patients denied any history of chest pain prior to MI [15]. Older patient frequently have triple vessel disease [10, 16–20]. The long-term prognosis of the patient depends upon the number of vessels involved and the degree of LV dysfunction [11, 21, 22].

**CONCLUSION**

Smoking and family history of premature coronary artery disease (CAD) is seen more commonly in younger patients. The CAD in young adults has a poor long-term prognosis. Smoking cessation remains a significant challenge in younger population. More awareness and cessation measures are required to address this emerging situation. Given lack of warning signs in these patients, physicians need to be more aggressive in managing risk factors as CAD in younger patient can have adverse outcomes.

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**Author Contributions**

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**Guarantor**

The corresponding author is the guarantor of submission.

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**REFERENCES**

Nonsteroidal anti-inflammatory drugs: An unusual cause of multiple ileal perforations

Mohamed Ali Sheredi, Zakaur Rab Siddiqui

ABSTRACT

Introduction: In this case report, we describe a rare case of pneumoperitoneum with peritonitis due to multiple ileal perforations with a history of high doses of diclofenac sodium usage. Case Report: A young adult male with recent history of high doses of Nonsteroidal anti-inflammatory drugs (NSAIDs) presented in emergency department with acute onset pain in right iliac fossa with high grade fever and tachycardia for 12 hours. Abdomen was tender in the right iliac fossa with guarding. X-ray of chest showed free gas under the diaphragm. Exploratory laparotomy was done and showed multiple ileal perforations. Resection and anastomosis of the affected segment was done, both ends brought as double barrel ileostomy which was reversed later on without any complication. Conclusion: High doses of Nonsteroidal anti-inflammatory drugs can cause multiple ileal erosions and perforation and should be considered in the differential diagnosis, if other possibilities are excluded.

Keywords: Diclofenac sodium, Ileum, Intestinal perforation, Nonsteroidal anti-inflammatory drugs (NSAIDs)

INTRODUCTION

Nonsteroidal anti-inflammatory drugs (NSAIDs) are known to have adverse effects on the upper gastrointestinal tract, however, its effect on the small intestine are not well described [1]. It is proven that NSAIDs can be harmful to the small intestine and can be associated with multiple complications, such as, small intestinal strictures, ulcerations, perforations, diarrhea, and villous atrophy. The NSAIDs can have adverse effects in any part of the gastrointestinal tract including, oesophagus, stomach, duodenum, small intestine, or colon [2, 3]. Risk is increased with age, in first three months of treatment, smoking, associated cardiovascular or respiratory disease, high dose and multiple NSAIDs, and concomitant use of anticoagulant or steroids [1, 3].

Herein, we report a case of NSAID-induced multiple small bowel erosions and perforations that involved the proximal as well as distal ileum. Furthermore, similar cases of NSAID-induced enteropathy described in literature are also reviewed, in which other causes were ruled out by obtaining biopsy specimens for histologic analysis.

CASE REPORT

A 31-years-old male, with no known co-morbidity, presented in emergency department with a history of severe right lower quadrant pain of more than 12 hours
that was not responding to analgesics. There is associated intermittent high grade fever (up to 39.5°C), severe sore throat, and weight loss of 6 kg over 2 weeks, for which he received augmentin for a week and cefuroxime for another week with no improvement.

On physical examination in the emergency room the patient was tachycardiac, tachypneic, febrile and showed presence of severe follicular tonsillitis with oral ulcers. On further interrogation, the patient revealed that 16–20 tablets of diclofenac potassium per day for his pain for two weeks prior to the onset of his abdominal symptoms.

Systemic examination showed bilaterally equal air entry in the chest with regular heart sounds and no audible murmur. There was diffuse abdominal distention along with tenderness more in the lower abdomen. Although white cell count was high, however, all other laboratory tests including the Widal test were normal.

Urgent ultrasound of abdomen done which revealed free fluid in the abdomen. A chest X-ray showed free air under the diaphragm on the right side and an emergency laparotomy was performed for suspected peritonitis associated with intestinal perforation. Peroperatively, there were multiple small bowel perforation in the terminal ileum each approximately 5 mm in size along with a few impending perforations (Figure 1).

Approximately 15 cm of the ileum was resected due to very close and tinny perforations in which suturing was impossible, ileostomy was done with planned scheduled relook laparotomy through the laparostomy (mesh and VAC used).

Three relook laparotomies were performed and showed new perforations with impending perforations which were fixed by either primary repair or burying it inside.

Workup was done for the etiology including stool analysis for *Campylobacter jejuni*, *Yersinia enterocolitica*, *Salmonella*, *Shigella* and *Clostridium difficile* and found to be negative. Serum markers for vasculitis were also negative.

Histopathology report showed nonspecific inflammation and excluded inflammatory bowel disease, vasculitis, and vascular thrombi. Only positive history found was ingestion of high dose nonsteroidal anti-inflammatory analgesics.

Patient recovered slowly but well and discharged after two weeks of hospital stay. After six weeks ileostomy reversal was done without any complications and remained healthy till now (six months postoperatively)

**DISCUSSION**

The differential diagnosis of nonspecific erosions, ulcerations and perforation of the small intestine includes Crohn’s disease, Typhoid fever trauma (from ingested toothpicks or bones), infection (cytomegalovirus, tuberculosis, or *Yersinia*), Vasculitides (such as lupus, rheumatoid, or periarteritis nodosa) postradiation therapy, ischemia, and various medications (like enteric-coated potassium). In certain case reports, rheumatoid vasculitis and other collagen vascular diseases are supposed to be responsible for described intestinal strictures and inflammation, therefore, patients suffering from such diseases should be thoroughly examined, as NSAIDs are usually mainstay to treat these disorders [3, 4].

In contrast to the previously accepted belief, recently, it is documented that NSAID-induced gastrointestinal injury occurs more frequently in the small bowel than in the stomach [4, 5]. (NSAID) induced enteropathy may manifest acutely as well as after chronic use, being five patients described in one study after use of medication for less than 4 weeks [6, 7]. Likewise, the development of enteropathy does not seem to be related to the route of administration of NSAIDs, because, use of Osmosin, a now obsolete indomethacin rectal suppository, was also associated with ileal perforations [8]. However, the effect of intravenous route has yet to be established. Furthermore, in another study, majority of patients with NSAID-induced enteropathy were taking either indomethacin or piroxicam preparations for more than a month. Among these patients, 40% had intestinal obstruction, 40% had ulceration and anemia, and 12% had intestinal perforation, suggesting the NSAIDs as a possible cause. In a separate autopsy series, undiagnosed
small bowel perforations was found to be the cause of death in three patients who had received long-term Aspirin therapy, that is, for six months or more [9]. In the study described previously, most of the patient underwent intestinal resection while in about one-fifth of them, the only treatment was discontinuation of use of NSAIDs [9]. Considering that, if the disease is suspected and treated early, it may preclude the need of surgery, thus, considerably reducing the associated morbidity and mortality.

In the case described here, the patient high doses of diclofenac sodium orally for two weeks. The subsequent laboratory and histopathologic findings were unable to identify any etiology for his ileal perforations other than NSAID-induced enteropathy, including inflammatory bowel disease, trauma, typhoid, tuberculosis and amoebiasis. Therefore, a diagnosis of diclofenac sodium-induced multiple ileal perforation was reached.

In another retrospective study, it is noted that those patients who were taking NSAIDs were twice as likely to have small and large bowel perforations or hemorrhage in comparison with control patients [10], further supporting our presumption in this case.

**CONCLUSION**

High doses of nonsteroidal anti-inflammatory drugs can cause multiple ileal erosions and perforation and should be considered in the differential diagnosis, if other possibilities are excluded. Early suspicion and timely intervention may reduce the associated morbidity and mortality.

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**REFERENCES**

ABSTRACT

Introduction: Though thrombosis of a giant aneurysm is a common entity, the phenomenon of embolic phenomenon from an angiographically negative thrombosed aneurysm causing ischemic infarction is a very rare entity. Case Report: Herein we report a case of an ischemic episode from a thrombosed giant aneurysm which was angiographically occult. We then discuss the management and outcome in a 32-year-old female. Conclusion: Embolic phenomenon from an angiographically negative giant aneurysm can impose dilemma in the management plan. This differential should always be placed in mind while dealing with lesions in the vascular territory and correct algorithm should be taken in planning the correct therapeutic approach for the same.

Keywords: Angionegative thrombosed, Giant aneurysm, Emboli, Middle cerebral artery aneurysm, Management

INTRODUCTION

Though spontaneous thrombosis within a giant aneurysm is a fairly common entity [1], the embolic phenomenon through the same and that too in an angiographically occult aneurysm is a rare epiphenomenon. Such a clinical entity would surely spin the head of anyone concerned with the management of the same. Here we report such case and review the management done for the same in a 32-year-old female.

CASE REPORT

A 32-year-old female with no significant past medical or surgical illnesses, presented to our emergency department with sudden onset of weakness on the left half of the body since 1 day duration. There was no history of trauma, transient ischemic attacks, joint pain, photosensitive rashes, shortness of breath, hematuria or chronic medication usage. GCS of the patient was 15/15 and there was left sided hemiparesis of 2/5. Systemic examination was normal. NIHSS score was 4. Contrast computed tomography (CT) scan performed showed oval shaped lesion on the right distal Sylvian region with peripheral rim enhancement and central flow void suggestive of a thrombosed aneurysm. Contrast CT scan of brain showed oval shaped lesion with peripheral enhancement and
central flow void suggestive of thrombosed aneurysm (Figure 1). There was also hypodensities in the right striatal territory indicative of infarction. Magnetic resonance imaging (MRI) scan of brain showed evidence of hypodensities on the right striatal and frontal opercular region (Figure 2). Diagnostic angiography was negative for any obvious aneurysm on the middle cerebral artery (MCA) territory. Diagnostic angiography was negative for suspected right MCA aneurysm (Figure 3). Antinuclear antibody (ANA) and erythrocyte sedimentation rate (ESR) were within normal limit. EKG and ECHO to rule out cardiac cause for embolus was negative. After explaining the disease condition, the risk of further embolic episodes as well as that of rupture, the patient was taken up for surgery. We also detailed the option of flow diverters and thrombolytic therapy had it not been due to giant aneurysm. We harvested the superficial temporal artery (STA) for probable ST-MCA bypass for flow augmentation. Intraoperatively, a giant thrombosed MCA bifurcation aneurysm was seen with multiple perforators encircling its neck. After dissecting the branches from the neck the sac was opened (Figure 4), clot evacuated and clip reconstruction securing the MCA bifurcation branches was done. Clipping of the neck securing the MCA bifurcation (Figure 5). Patency of parent vessels was confirmed with intraoperative indocyanine green (ICG) study (Figure 6). Encephalo-myo-arterio-synangiosis was also done. Postoperative computed tomography scan of brain showed no evidence of vasospasm (Figure 7). Postoperative period was uneventful and the hemiparesis improved to 4/5.

DISCUSSION

Intra aneurysmal thrombosis has been reported to occur in around 50% of the cases with giant aneurysm [2, 3]. This was first described by Lyell [4] during an autopsy study. The most important factor contributing to the development of the thrombosis is the critical ratio between the aneurismal volume and its neck size [5]. While some argue that the lattice-like armor of the thrombus provides the protection against the rupture [6, 7], others argue that due to vortex flow pattern, there are areas of endothelial damage alternating with the fibro-calcified areas that may be the nidus for rupture [8, 9].

The incidence of thromboembolic phenomenon from a thrombosed aneurysm has been reported to be in the range of 5–59% [10–13]. Another risk can be parent vessel occlusion.

The hallmark of the entity is the disparity in the size of the lesions in the CT scan and angiography [14–16]. Others features are target sign, Peripheral rim enhancement and calcification of the wall [17, 18].

The management of the condition varies from observation, clipping, thrombectomy, clip reconstruction and trapping after bypass. Lawton et al. [19] have described a new classification scheme with type specific
treatment strategy. They proposed that the best result was after clipping. If the neck is not clippable, then the best approach would be bypass and aneurismal occlusion. Postoperative period can also be complicated by the Coandă effect [20] and also the vasospasm.

CONCLUSION

To conclude these subsets of aneurysm are extremely difficult to treat and remains an enigma. Detailed preoperative planning is the key to success. This case was unique in the sense that the angiography was normal and also there was embolic phenomenon observed. The effect could be due to mass effect of a giant aneurysm or of a emboli from the clot within the sac.
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Figure 7: Postoperative computed tomography scan of brain showing no evidence of vasospasm.


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Usefulness of $^{99m}$Tc-HDP scintigraphy in the diagnosis of suspected cardiac amyloidosis revealed by heart failure: A case report of an amyloidogenic transthyretin mutation

Geraldine Celine Bera, Lavinia Vija, Pierre Jean Fouret, Nathanaëlle Yeni

CASE REPORT

A 54-years-old Guadeloupean patient known to have hypertrophic cardiomyopathy with severe left ventricular systolic dysfunction was hospitalized for cardiogenic shock. Ventricular arrhythmia lead to an implantable cardioverter-defibrillator (ICD). While most investigations proved to be unsuccessful, cardiological assessment showed: a micro-voltage on electrocardiogram (ECG); an increased level of N-terminal form of B-type natriuretic peptide (nT-pro-BNP) to 12000 ng/L; a restrictive infiltrative cardiomyopathy with concentric and homogeneous left ventricular hypertrophy and a severe biventricular dysfunction on echocardiography (Figure 1). A cardiac single photon emission computed tomography (SPECT), performed three hours after the intravenous injection of 600 MBq of $^{99m}$-technetium oxidonrate ($^{99m}$Tc-HDP) revealed abnormal myocardial uptake (Figure 2A–B) that was prominent on apical and anteroapical walls and moderate on antero-medial-septal wall (Figure 2C). In this context, the scintigraphy was highly suggestive of cardiac amyloidosis. This diagnostic was confirmed thereafter by myocardial biopsy that showed extracellular amyloid deposits (Figure 3). Antibody anti-transthyretin revealed by immunohistochemical analysis of amyloidosis was in favor of a transthyretin form. Genetic sampling showed homoygous missense mutation valine to isoleucine substitution at position 122 (V122I) of TTR gene.

Figure 1: Short-axis echocardiography showing homogenous left ventricular hypertrophy.
Amyloidosis is characterized by extracellular tissue deposition in vital organs of misfolded proteins [1, 2]. Among systemic amyloidosis, immunoglobulin light-chain amyloid (AL-form) and transthyretin amyloid (ATTR-form) are known to infiltrate the heart leading to congestive heart failure, atrial fibrillation and conduction abnormalities [2, 3]. The ATTR-form of cardiac amyloidosis, an underdiagnosed cause of heart failure, is slowly progressive and clinically well tolerated while AL-form often lead to rapidly progressive cardiac dysfunction. An early recognition and accurate classification of both entities, potentially fatal, is necessary because treatment depends on amyloidosis type. The ATTR-form may be either familial (variant TTR) caused by a mutation, or sporadic (aged-related) due to misaggregation of a wild-type transthyretin [2]. TTR is a transport protein primarily expressed by the liver and circulates as homotetramer; under genetic mutation or aging, tetramers dissociate to monomers leading to amyloid fibrils [2]. Aggregate autopsy data show presence of wild-type TTR in 25–30% of hearts, leading to cardiac dysfunction in a smaller but significant elderly population [2, 4]. As for variant TTR more than 80 mutations have been described [1]. V122I TTR variant is one of the most widespread with a population prevalence of 4% in Afro-Caribbeans [2, 4]. Its diagnosis has to be confirmed by the immunohistochemical demonstration of amyloidogenic TTR-form on a biopsy specimen and by the presence of an ATTR mutation on protein/DNA analysis [2–4]. However myocardial biopsy is not often performed in practice because it is invasive, results are late with the possibility of false-negatives. Non-invasive diagnostic approaches such as ECG and echocardiography have low specificity and sensitivity, respectively [2]. Cardiac magnetic resonance (CMR) is hampered by limited availability and inability to image patients with pacemaker or ICD. This modality is able to identify amyloid infiltration by late gadolinium enhancement imaging (with a sensitivity and specificity of 90%), keeping in mind the potential risk of nephrogenic systemic fibrosis in these patients with frequent chronic kidney disease [2]. To note, radionuclide imaging shows a potential interest for identification cardiac amyloid deposits. With SPECT, a myocardial uptake of 99m-technetium diphosphono-propanodicarboxylic acid (99mTc-DPD) is able to differentiate mutation carriers from the AL-form [1], likely due to higher concentrations of calcium-containing products in TTR amyloid [5]. Quantification of amyloid burden by scintigraphy could predict cardiac events and thus have an impact on determining therapeutic strategies. 99mTc-DPD cardiac retention can be scored as follow:

- **Grade 0** for no visible myocardial uptake in both the delayed planar or cardiac SPECT;
- **Grade 1** for cardiac uptake on SPECT only or cardiac uptake of less intensity than the accompanying normal bone distribution;
- **Grade 2** for moderate cardiac uptake with some attenuation of bone signal
- **Grade 3** for strong cardiac uptake with little or no bone uptake [6]. The possibility of a prognostic assessment by quantification allows to follow-up the course of the disease while new specific therapies for TTR amyloidosis are emerging (such as TTR stabilizers and small RNA silencing molecules). 18fluorine-florbetapir positron emission tomography (18F-Florbetapir PET) approved for beta amyloid plaque in the brain, was investigated in cardiac amyloidosis and showed high myocardial radiotracer uptake and late retention of 18F-Florbetapir in amyloid subjects suggesting a correlation with amyloid protein. However, its value in the diagnosis of cardiac amyloidosis remains to be confirmed [7]. Another interesting SPECT tracer is 123iodine-metaiodobenzylguanidine (123I-MIBG) for imaging of myocardial innervation. The localization of MIBG relates to the presence of sympathetic nerves. Myocardial defects in MIBG activity seems to correlate
with impaired cardiac sympathetic nerve endings due to amyloid deposits and can be identified very early in cardiac amyloidosis. Furthermore, the clinical severity of disease correlates with a decrease in MIBG uptake [1].

CONCLUSION

In conclusion 99mTc-HDP or 99mTc-DPD scintigraphy appears to be a major examination to establish the aetiology of cardiac amyloidosis with a high specificity, especially as age-related ATTR could become the most common form of this disease in the future. This could facilitate the ATTR specific care (genetic counselling, preventing measures and interference with medications). Furthermore with a simple and non-invasive method, whole-body tracer retention and specifically myocardial tracer retention correlated with disease severity could be used for prognostic assessment.

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Apocrine hidrocystoma of eye lid mimicking as sebaceous cell carcinoma

Undrakonda Vivekanand, Heng Siang Ting, Shankar Nag Vattipulusu, BM Yashodhara

CASE REPORT

We report a case of a 58-years-old male who presented with a painless right upper eyelid swelling with mucoid discharge since two years. Ocular examination revealed phthisis bulbii in both eyes with nil perception of light. A nodular, reddish swelling of 1.5x2 cm was present in right eyelid involving middle one-third of upper lid margin (Figure 1). No such tumors were found in any other parts of the body of the patient. Wide excisional biopsy with Cutler–Beard lid reconstruction surgery was performed under local anesthesia (Figures 2 and 3). Histopathology (Figure 4A–B) revealed epidermis with underlying adnexa and basal incontinence of pigment, dermis showed multilocular cystic spaces lined by double layer secretory cells. Inner most cells were cuboidal with eosinophilic cytoplasm, outer layer cells were columnar suggestive of apocrine hidrocystoma of eyelid.

DISCUSSION

The eyelids are specialized structures of the ocular adnexa made up of various cells and tissues which can give to a variety of tumors of benign to malignant nature and inflammatory lesions. The management of each of these is variable. Papilloma, seborrheic...
keratosis, keratoacanthoma, inverted follicular keratosis, neurofibroma, pseudocarcinomatous hyperplasia and epidermal inclusion cyst are the common benign tumors. Basal cell carcinoma, squamous cell carcinoma, melanoma, sebaceous gland carcinoma and Merkel cell carcinoma are the malignant lesions of the eyelid. Tumors could also arise from sweat glands and some of them are: syringoma, eccrine hidrocystoma, apocrine hidrocystoma, syringocystadenoma papilliferum, pleomorphic adenoma and malignant sweat gland tumors. Likewise there are many tumors which could originate from vessels, hair follicles or sebaceous glands [1]. Apocrine hidrocystoma is a tumor originating from glands of Moll at the lid margin, usually solitary (sometimes multiple) cystic to nodular or papular lesions with bluish or black hue to iron deposit/lipofuscin/Tyndall effect in the cyst wall [2, 3]. Solitary tumors are treated by surgical excision. Recurrence of tumor following surgery is rare. Multiple lesions are treated with carbon dioxide laser and trichloroacetic acid or botulinum toxin. The differentials for this tumor are nevi, basal cell carcinoma, melanoma, milia and syringomyelia. The patient had a pinkish, fleshy lesion resembling sebaceous gland carcinoma which is a malignant lesion for which he underwent excision of tumor followed by Cutler-Beard reconstruction [4].
CONCLUSION

Apocrine hidrocystoma of eyelid may sometimes mimic as sebaceous cell carcinoma. Histopathology plays a crucial role in establishing a diagnosis.

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REFERENCES

Spontaneous subcutaneous orbital emphysema following nose blowing

Ozgur Tatli, Faruk Ozsahin, Selim Yurtsever, Gurkan Altuntas

CASE REPORT

A 29-year-old male presented to the emergency department with sudden swelling and pain in the left eye. Pain accompanied by swelling had developed immediately after blowing his nose two days previously. On examination there was left periorbital swelling with crackling sound (crepitus) on palpation suggestive of subcutaneous orbital emphysema. The eye was painful at palpitation. Swelling was not accompanied by redness or elevated temperature.

Bilateral ocular movements, pupillary reactions, funduscopic examination and visual acuity were normal. There was no diplopia or ptosis. The patient had no history of recent trauma to the head and penetrating the orbital area. Orbital computerized tomography revealed accumulations of air above the left lateral rectus muscle in the orbital ceiling and in the anterior palpebral area (Figures 1A–B). There was no bone fracture. The sinuses were normal. Retro-orbital fat planes had a normal appearance. The patient was started on prophylactic antibiotic therapy and kept under observation about 24 hours. As a prophylactic regimen cephalexin was started at a dosage of 500 mg every 12 hours for 5 days. The visual acuity in left eye was not dropped. Follow-up examinations showed no ophthalmological complications because of these normal findings, drainage and decompression were not performed. Swelling regressed entirely and clinical findings were normal by the 10th day of monitoring.

DISCUSSION

Orbital emphysema is a rare complication generally arising after trauma. Most cases involve an orbital bone...
fracture causing air ingress. It is a benign and temporary condition that usually develops in the first 24 h after paranasal sinus fractures, and spontaneous absorption generally takes place within two weeks [1]. Orbital emphysema is evaluated by means of palpation and radiological examination, particularly in patients with a history of trauma. Orbital fractures most frequently occur in the thinnest parts of the medial and inferior walls of the orbital bone, the ethmoid, maxillary and frontal sinuses, respectively [2]. The orbital bones are more flexible in children, and thus fracture less [3]. With paranasal sinus mucosa destruction, the fracture line has a valve-like effect and air enters the orbita [4]. Clinical symptoms such as swelling in the eye, closing of the eyelids, extraorbital subcutaneous emphysema, subconjunctival hemorrhage, sensitivity and pain may appear [5]. Rarely, occlusion of the central retina may develop with orbital compartment syndrome resulting from orbital emphysema. Air entering the orbital space may lead to occlusion and sight loss by creating a mass effect on the central retinal artery. Proptosis and diplopia may also occur. There are various sub-classifications within orbital emphysema. In genuine orbital emphysema, the air is localized behind the orbital septum whose integrity is not compromised immediately after orbital fracture [6]. Air can enter the orbital soft tissue during coughing, sneezing or nose blowing. There is no internationally agreed treatment modality for orbital emphysema. However, Hunts et al. published a study on the treatment of the condition in 1994 [7]. Patients should be advised not to blow their noses. Nasal decongestants, antibiotics and steroids may also be given [8]. If findings such as restricted ocular movement, disk edema or loss of sight occur in orbital emphysema, surgical procedures such as drainage and/or direct decompression may be performed.

In diagnosis, the most important radiological imaging technique in determining fracture and the location of the air pockets is orbital tomography [9].

CONCLUSION

Diagnosis of orbital emphysema is generally made with accurate anamnesis, physical examination and orbital tomography. Orbital emphysema is a complication that may frequently arise following orbital traumas, though it should not be forgotten that it can also have non-traumatic causes. Orbital emphysema developing after nose blowing is one such rare condition.

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