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Complicated Left-Sided Amyand’s Hernia in an 18-Month-Old Boy: A Case Report and Literature Review

Victor I. C. Nwagbara1*, Maurice E. Asuquo1, Ayi E. Archibong2, Emmanuel Etuk1, Ijeoma O. Uchejeru1

1Division of General Surgery, Department of Surgery, University of Calabar Teaching Hospital, Calabar, Nigeria
2Division of Paediatric Surgery, Department of Surgery, University of Calabar Teaching Hospital, Calabar, Nigeria
Email: aikay_en@yahoo.com

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Abstract
The rare finding of the vermiform appendix within an inguinal hernia sac is known as Amyand’s hernia. It was first described by Claudius Amyand in 1735, in a right inguinal hernia. A much rarer find is a left-sided Amyand’s hernia. This is a report of a case of complicated left-sided Amyand’s hernia in an eighteen month old male child. He presented as an emergency with an obstructed inguino-scrotal hernia and the diagnosis of Amyand’s hernia was made intra-operatively. He made uneventful recovery after surgery. Treatment options depend on findings during operation and clinical status of the patient.

Keywords
Obstructed Hernia, Vermiform Appendix, Left-Sided

1. Introduction
In the year 1735, a surgeon named Claudius Amyand performed a right inguinal hernia operation for an 11-year-old male patient named Havil Henderson and found the vermiform appendix in the hernia sac [1] [2]. He subsequently published his findings in 1736 in the Philosophic Transactions of the Royal Society [3] [4]. Subsequently, the finding of the vermiform appendix in a hernia sac has been ascribed the eponym Amyand’s hernia. This makes no distinction of whether the appendix is normal, inflamed or ruptured1 [3] [5] [6]. The presence of

*Corresponding author.

the vermiform appendix in a femoral hernia sac is however known as Garengeot’s hernia [7].

The incidence of Amyand’s hernia is variously reported as ranging from 0.5% to 1% of cases of Appendicectomy while an inflamed appendix is seen in 0.08% to 0.1% of cases. [2] [5] [8]. Amyand’s hernia is less well known than the much rarer Littre’s hernia (Meckel’s diverticulum in the sac) although Meckel’s diverticulum is rated as being present in 2% of the general population. The age and sex distribution is not known but from reported cases there is a male preponderance. The reported age ranges from a 15-day-old premature neonate to a 92-year-old man [8] [9]. Our search in English literature found a few recorded left-sided cases of Amyand’s hernia and none from the West African sub-region. This submission is the first report of a case of left-sided Amyand’s Hernia from our practice in Calabar-Nigeria and perhaps the first reported case from West Africa. This case had many peculiarities including a tear in the caecal wall besides being complicated.

2. Case Report

An 18-month-old boy, weighing 8 kg was referred from a private clinic with a 3-day history of sudden onset of abdominal pain maximal over a pre-existing reducible left inguinal swelling. Patient was irritable, vomited once and refused feeds for 2 days. He also developed fever, constipation and abdominal swelling a day before referral.

Clinical evaluation showed an irritable, pale, febrile and dehydrated infant with tachypnoea and tachycardia. The abdomen was moderately distended. Umbilical hernia was present and bowel sounds were hyperactive. He had a hyperaemic, tender, left inguino-scrotal mass but both testes were separately palpable. A working diagnosis of Strangulated Left Inguino-scrotal hernia was made. The patient was rapidly resuscitated and scheduled for an emergency operation. Suddenly, the crying and irritability increased in tempo before the beginning of the operation. It was observed that the scrotum was rapidly increasing in size and tensed with the attendant fear of bursting (Figure 1).

Immediate intervention was done through a left inguinal incision and intra-operative findings were:
1) An obstructed, sliding indirect left inguinal hernia sac with faeculent smell.
2) The contents of the sac shown in Figure 2.
   ➢ A gush of gas with faeculent odour upon opening the hernia sac.
   ➢ An inflamed (hyperaemic and turgid) vermiform appendix.
   ➢ An enclosed part of the circumference of the caecal wall with a 2 cm linear tear (grasped by Babcock’s forceps in Figure 2).
   ➢ A scanty amount of purulent exudates.
   ➢ Viable terminal ileum and caecum.
3) Normal left testis and vas deferens

The constricting fascial band at the neck was released. Appendicectomy was done, caecal tear closed extra-abdominally with a single layer of Polyglactin 910 sero-muscular suture. Local toileting was done with saline soaked sponge and contents reduced. The hernia sac was closed by purse-string and redundant part excised. Inguinal anatomy was reconstituted over a drain removed after 48 hours. Antibiotic prophylaxis commenced pre-operatively was continued post-operatively in therapeutic doses.

Figure 1. Pre-operative picture showing tense, shiny, scrotal swelling.
Figure 2. Intraoperative picture showing vermiform appendix held with Allis forceps and the tear in the caecal wall held by Babcock forceps.

The postoperative course was uneventful. Patient commenced oral feeding on second post-operative day. He was discharged home on the 5th postoperative day. Abdominal Ultrasound scan and Chest Radiograph were requested. However, the patient was lost to follow-up and never did the requested investigations.

3. Discussion

Amyand’s hernia implies that the vermiform appendix is contained within the inguinal hernia sac [1]-[9]. This usually suggests a right-sided inguinal hernia as the vermiform appendix and caecum are normally found in the right lower quadrant of the abdomen. The incidence of Amyand’s hernia ranges from 0.5% to 1% of hernias and almost always an incidental operative finding. Most cases of Amyand’s hernia contain a normal appendix while inflamed appendix is noted in 0.08% to 0.1% of cases [2] [5] [8]-[11], thus making it a rare condition.

We note at this point that Amyand’s hernia is less often mentioned in the medical literature than the rarer eponymous Littre’s hernia. Meckel’s diverticulum is said to occur in about 2% of the general population (post-mortem) but this not confirmed from operative findings. The incidence of finding a ruptured appendix in a hernia sac whether Amyand’s or not, is not known though expected to be rarer than the inflamed variety. The tear in the caecal wall as noted in this case was spontaneous. This a peculiar isolated finding and perhaps precisely occurred at the time of sudden increase in the tempo of crying and rapid expansion of scrotal size. This tear, in the absence of any gangrenous segment of gut or rupture of the appendix, may be due to a rise in intraacaecal pressure above its bursting point especially in the presence of a competent ileo-caecal valve.

Left-sided Amyand’s hernia is less reported in literature than the right-sided because the appendix is normally located in the right lower quadrant of the abdomen. Our search in English medical literature produced about 20 reported cases of left-sided Amyand’s hernia from all continents and more recently from Africa as well [11]. This is the first report of a case of Left-sided Amyand’s hernia from the West African region, and a peculiarly complicated one.

An inguinal hernia sac may be empty or contain omentum, small bowel or both. Atypical cases may contain part of the wall of the caecum or urinary bladder (sliding hernia), vermiform appendix (Amyand’s hernia), Meckel’s diverticulum (Littre’s hernia) or part of the circumference of the bowel wall (Richter’s hernia) or even an “arrested testis”. We may someday witness an appendix tumour within the hernia sac. It has been suggested that pre-operative investigations such as ultrasonography, hernioscopy, laparoscopy or computed tomography [11] [12] may be helpful for diagnosis. These modalities are usually not employed since diagnosis is made intra-operatively, and will amount to waste of resources and lead to delay in treatment. Amyand’s hernia has no typical clinical features. It shares common clinical features with other hernias with the possibility of being complicated by obstruction or strangulation. However, the presence of an inflamed vermiform appendix within a hernia sac will result in accentuated presentation and risk of rupturing or bursting as in our index patient.

Hernia is a surgical disease that requires surgical intervention. In emergency situations like our index case, surgical therapy must be expedited. The proposed management classification by Losanoff and Basson [13] [14] (Table 1) may be more useful for right-sided Amyand’s hernia diagnosed before operation than for the left-
Table 1. Losanoff and Basson management classification.

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<th>Description</th>
<th>Management</th>
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<td>Type 1</td>
<td>Normal appendix within hernia sac</td>
<td>Hernioplasty (Mesh repair) without appendicectomy</td>
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<td>Type 2</td>
<td>Acute appendicitis within the Hernia sac, no abdominal sepsis</td>
<td>Appendicectomy and repair of hernia without mesh</td>
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<td>Type 3</td>
<td>Acute appendicitis within the hernia sac with abdominal sepsis</td>
<td>Laparotomy and proceed to appendicectomy and primary repair of hernia without mesh</td>
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<td>Type 4</td>
<td>Acute appendicitis within the hernia sac, related or unrelated abdominal pathology</td>
<td>Manage as types above, investigate treat secondary pathology as appropriate</td>
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sided cases. From our experience herein, we advocate appendicectomy in all left-sided cases irrespective of the clinico-pathological status of the appendix. This is justified in that the appendix in the left may lead to misdiagnosis if left in situ. Diagnosis of hernia is a normal clinical work up not requiring the various radiographic procedures with their attendant hazards, expense and treatment delay. However, in cases like ours, during the post-operative period, various imaging techniques may be employed to resolve other possible diagnostic dilemma such as situs inversus, malrotation or wandering caecum. The selected modality must be justifiable hence we think that the use of computed tomography will add no advantage.

4. Conclusion

Left-sided Amyand’s hernia is an uncommon surgical entity. It is usually a chance finding at operation. It is more likely to harbour an inflamed appendix than the right side. Immediate appendicectomy should be done routinely in left-sided Amyand’s hernia if only to prevent future diagnostic confusion. The management is incomplete until situs inversus, malrotation or mobile caecum is ruled out.

References


Left Atrial Myxoma—A Case Report

Ramachandran Muthiah
Thoothukudi Medical College Hospital, Thoothukudi, India
Email: cardioramachandran@yahoo.co.uk

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Abstract
Atrial myxomas are the most common primary cardiac tumors. More than 90% are solitary. A large myxoma occupying in the left atrium producing mitral stenosis and regurgitation was demonstrated by 2D echocardiographic images in this case. It remained asymptomatic for a long period with a survival up to the age of 75 years in an elderly female.

Keywords
Myxoma, Left Atrium, Mitral Valve Obstruction

1. Introduction
The most frequent benign tumor of the heart is myxomas and accounting in approximately 30% of all primary cardiac tumors [1]. 75% are located in the left atrium [2]. Myxoma is a benign polypoid neoplasm, usually originates from endocardial cells in the region of fossa ovalis and is attached to the interatrial septum. Myxomas are pedunculated, friable and appear as a soft, gelatinous, mucoid, usually gray-white mass often with areas of haemorrhage or thrombi. They are slowly growing and usually do not produce symptoms or signs until they enlarge. Their size, shape and texture can be quite varied. Myxomas may be smooth surfaced but are more often irregularly shaped or have the appearance of a “cluster of grapes”. They are typically nonhomogeneous in texture with lucent centers or areas of calcification. Myxomas can be quite large, occupying most of the left atrium and resulting obstruction to left ventricular filling. The diagnosis can be established by the demonstration of a characteristic echo producing mass in the left atrium by two-dimensional (2D) echocardiography [3] [4]. Although asymptomatic patients with myxomas have been reported, most present with one or more effects of a triad of constitutional, embolic and obstructive manifestations [5]. Typically, these large pedunculated tumors advance through and obstruct the atrioventricular valves during diastole and are expelled retrogradely into the atrium during systole. The most common clinical presentation mimics that of mitral valve disease [6]—either stenosis due to tumor prolapse into mitral orifice or regurgitation due to tumor induced valve trauma.

An asymptomatic large myxoma in the left atrium in an elderly is rare and so this case has been reported.
2. Case Report

A 75-year-old woman was admitted with exertional dyspnea and palpitations for the past 6 months. The patient developed the symptoms especially in sitting posture when raising from the bed in the early morning hours and get relieved when lying down. The symptoms were intermittent for a short period recently Constitutional symptoms such as arthralgia, myalgia, and weight loss were also present. Blood chemistry showed elevated erythrocyte sedimentation rate and platelets were within normal range. Pulse was irregular (112 bpm) and blood pressure was 140/90 mmHg. Symptoms and physical findings were often “positional”. Physical examination revealed mild clubbing and auscultatory findings were consistent with mitral valve stenosis and insufficiency. A loud first heart sound, low pitched “tumor plop”, a holosystolic murmur that was loudest at the apex and resembled mitral regurgitation and a mid diastolic murmur well noticed in the sitting position were the characteristic findings masquerading as Rheumatic mitral stenosis and regurgitation. ECG revealed atrial fibrillation. X-ray chest revealed cardiomegaly. Echocardiography revealed a large myxoma occupying in the left atrium, producing mitral valve obstruction as shown in the Images 1-4.
She was given symptomatic treatment with small doses of diuretics and beta blockers such as propranolol, symptom free thereafter and advised to avoid exertion. Since the age was advanced and well maintaining basic life style, surgical intervention was not preferred in this patient.

She was followed up to two years and no further adverse events noticed in her life style. Echocardiographic screening of family members revealed no abnormality.

3. Discussion

The most common primary cardiac tumor presenting in the left atrium is the benign myxoma, which in the large majority of cases is solitary [7]. In a large single-Institution series of primary cardiac tumors, 42% were cardiac myxomas [8]. The proportion of myxomas in comparison with other tumors was increased to 77%. The mean age of presentation with sporadic myxoma is 56 years and 70% are females in the age group of 3 to 83 years. The cause of syndrome myxoma is unknown [9]. It has been proposed to result from a widespread abnormality resulting in excessive proliferation of certain mesenchymal cells and excessive glycosaminoglycans production by them. Most tumors are histologically benign and potentially lethal due to intracavity or valvular obstruction,
peripheral embolism and conduction disturbances. The association of constitutional symptoms is likely due to synthesis and secretion of interleukin-6.

Left atrial myxomas prolapse to various degrees into mitral valve orifice, resulting in obstruction to AV (atrioventricular) blood flow and frequently, mitral regurgitation. The resultant signs and symptoms often mimic those of mitral valve disease [10], especially mitral stenosis. Symptoms of sudden in onset, intermittent and related to the patient’s body position [11] should raise the suspicion of left atrial myxoma. When atrial myxomas obstruct the AV valves, the patient may experience dyspnea, dizziness or syncope when sitting or standing with alleviation of symptoms on lying down. Sudden death may also occur. The loud first heart sound that occurs in patients with left atrial myxoma may be due to the late onset of mitral valve closure resulting from prolapse of the tumor through the mitral valve and frequently split, with the second component corresponding to the expulsion of the tumor from the mitral orifice. In many cases, an early diastolic sound, termed a “tumor plop” can be identified and it is thought to be produced as the tumor strikes the endothelial wall or its excursion is abruptly halted. If the obstruction is incomplete, the tumor plop may be followed by a diastolic rumble. When the obstruction becomes more severe, cardiac output may fall precipitously.

Given a typical presentation, Echocardiography is virtually diagnostic of myxoma. The ability to detect atrial myxoma of the heart by means of echocardiography was first reported more than two decades ago by Effert and Domanig. 2D echocardiography is the non invasive procedure of choice for the diagnosis of left atrial myxoma. The virtual pathognomonic finding of an atrial myxoma is that of a large pedunculated tumor mass traversing through the AV valves in a to-and-fro motion. Large atrial myxomas have been classified by echo appearance as follows [12].

Class I-small and prolapse through the mitral valve.
Class II-small and non prolapsing.
Class III-large and prolapsing.
Class IV-large and non prolapsing.

Myxomas are mottled in appearance and in some atrial myxomas, areas of echolucency corresponding to areas of haemorrhage within the tumor which are not seen in thrombi or infective lesions. In this case, 2D echocardiographic Image 1 and Image 3 illustrate the myxoma in the left atrium (size 54.8 × 45.6 mm) and Colour Doppler Image 2 and Image 4 illustrate its obstructive features masquerading as Rheumatic mitral regurgitation and stenosis. Echocardiographically, the mitral valve appears as normal without any evidence suggestive of Rheumatic process such as commissural fusion, valve thickening and calcification. The resultant mitral stenosis and regurgitation are due to the movement of the myxoma across the mitral valve and so they are functional, masquerading as Rheumatic mitral stenosis and regurgitation in this case.

Surgical excision is the treatment of choice for most benign cardiac tumors and in many cases results in a complete cure. Recurrence is 1% to 5% after resection. Large size of myxoma together with location on posterior left atrial wall necessitates complete removal of the heart which was followed by “auto transplantation” i.e., reimplantation of the patient’s excised heart.

4. Conclusion

A large myxoma occupying in the left atrium remains asymptomatic up to the age of 75 years in a female is a rarest presentation in tropical countries such as India at Thoothukudi coastal region in Tamil Nadu State.

References


A Rare Cause of Chest Pain: Disseminated Elemental Mercury Microthromboembolism

Brandon Allen, Bobby Desai, Lars K. Beattie
Department of Emergency Medicine, Gainesville, FL, USA
Email: brandonallen@ufl.edu, bdesai@ufl.edu, lars.beattie@ufl.edu

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Abstract

Background: The clinical manifestations of mercury poisoning vary based on chemical form, dose, and route of administration. In the medical field, many of the cases of mercury exposure have been in mining agriculture, or chemical industrial workers via inhalation. Aim: To discuss the diagnosis of parenteral mercury poisoning with radiographic identification and potential sequelae. Case Presentation: We present the case of a young male with a chief complaint of chest pain and intravenous mercury administration found to be intentional as a suicide attempt. The chest radiographs lead to a wide differential diagnosis in a hemodynamically stable patient. Conclusion: While ingestion rarely causes toxicity due to the poor absorption of mercury through the gastrointestinal tract, intravenous elemental mercury can range from producing minimal symptoms to being severely toxic.

Keywords

Mercury Toxicity, Chest Pain, Microthromboembolism

1. Introduction

Mercury has been used widely for many centuries in various treatment modalities and medical instruments. Chronic exposure to vapor of mercury has been reported to result in neurologic symptoms like weakness, ataxia, and tremors. Acute exposure is more dramatic and can be fatal [1] [2].

Toxicity with metallic mercury injection, acquired both accidentally, and following attempted suicide is extremely rare. There have been a few published cases of intravenous metallic mercury injection over the past 30 years most commonly found in patients who are younger males.
2. Case Report

2.1. Emergency Department Evaluation

A 31-year-old male with the past medical history of spontaneous pneumothorax and hepatitis C presented to the Emergency Department with a complaint of severe, stabbing left-sided chest pain which began abruptly 4 hours prior to arrival and was worse with deep inspiration throughout the chest. He also reported that this pain was different from his previous pneumothorax chest pain. He acknowledged a non-productive cough, but denied fevers, chills or rigors. His only medication was ibuprofen 800 mg every six hours as needed.

The pertinent social history included that the patient was an active full pack per day smoker with frequent use of marijuana. He denied IV drug use and worked as a part-time mechanic whose duties consisted of welding and grinding metal. He reported no protective equipment during these activities.

At presentation to the emergency department his initial vital signs and physical exam showed: temperature 37.9 degrees Celsius, pulse rate 96 beats per minute, blood pressure 124/83 mmHg, respiration rate 24 breaths per minute, and pulse oximetry 99% on room air. He was in apparent distress, writhing in pain, tachypneic, and he appeared disheveled and unkempt. His chest wall was non-tender to palpation, bilateral normal percussion, vocal fremitus, symmetrically clear breath sounds without crepitation or audible wheezing. His heart sounds were normal without murmur, rub or gallop. Besides multiple excoriations and a few pustules in accessible areas of his body, the rest of the patient’s physical exam was normal and unrevealing.

Initial laboratory data revealed white blood cell count of 12.1 thousand/cu mm with 62.7% neutrophils and no bandemia. The cardiac biomarkers, basic metabolic panel, coagulation studies and urinalysis were all normal. No inclusion criteria were met based on the Pulmonary Embolism Rule-Out Criteria (PERC) to warrant a d-dimer in this patient [3].

The radiograph from his most recent emergency department discharge (Figure 1 and Figure 2) showed no acute cardiopulmonary disease and a two-view chest radiograph was performed (Figure 3 and Figure 4) which showed scattered radiopaque material between both lungs consistent with inhalation of radiopaque material. He was then admitted to the hospital.

Figure 1. Posterior/anterior chest radiograph. Interpretation: no acute cardiopulmonary disease.
Figure 2. Lateral chest radiograph. Interpretation: no acute cardiopulmonary disease.

Figure 3. Posterior/anterior chest radiograph. Preliminary radiology interpretation: scattered radiopaque material between both lungs consistent with inhalation of radiopaque material.
2.2. Hospital Course

During his hospital stay patient continued to have severe pleuritic chest pain with moderate relief from parenteral narcotics. The patient underwent a CT scan (Figures 5-9) to evaluate for thromboembolism and innumerable metallic foreign bodies were found scattered throughout each hemithorax, right ventricle and the liver which were not present in a prior study from one week before his hospital admission.

Considering his presentation and wide spread metallic microthromboemboli outside the lung tissues, inhalation of metallic objects due to unprotected welding was deemed unlikely. An intravenous metallic injection was suspected, and the Poison Control Center was contacted. A heavy metal urine toxicology collection was ordered. The following morning after urine collection, the patient’s chest pain had improved and he requested hospital discharge. He was advised to wait for urine toxicology results in order to guide therapy, but he consequently signed out against medical advice. The results returned three days later revealing urine mercury 54 µg/L (Reference range 0 - 10), urine mercury ug/day 104 (Reference range 0 - 15 µg/day) and urine mercury ug/g creatinine was 79.4 (Reference range <= 35) [4].

Once the toxicology report had resulted, the patient was appropriate for chelation therapy to remove the mercury as the primary therapeutic intervention. He was unable to be contacted despite numerous attempts and a law enforcement visit to the patient’s last known address. Based upon historical chart review, the patient has returned to the ED with the complaint of chest pain. However, he has not shown evidence or sequelae of toxicity from his intravenous mercury exposure.

2.3. Diagnosis

Disseminated elemental mercury microthromboembolism.
Figure 5. CT scan of the chest. Interpretation: multiple metallic foreign bodies found scattered throughout each hemithorax and right ventricle.

Figure 6. CT scan of the chest. Interpretation: multiple metallic foreign bodies found scattered throughout each hemithorax and liver.
Figure 7. CT scan of the chest. Interpretation: multiple metallic foreign bodies found scattered throughout each hemithorax and right ventricle.

Figure 8. CT scan of the chest. Interpretation: multiple metallic foreign bodies found scattered throughout each hemithorax and bilateral lung fields.
3. Discussion

While some patients with disseminated elemental mercury microthromboembolism have reportedly presented with fever, cough, GI symptoms, gingivitis, intention tremor, and/or bizarre behavior, this patient was unique as he only had symptoms of severe chest pain [5].

Because elemental mercury is radiopaque, it can be easily detected radiographically [6]. The chest radiograph usually shows tiny metallic opacities throughout the lung fields. A CT scan of the chest, abdomen and pelvis shows the full extent of mercury deposits throughout various organs [7]. The degree of mercury toxicity can further be evaluated by obtaining blood and urine mercury levels. Blood concentrations of >2 microgram/dL and urinary concentrations of >20 microgram/dL indicate toxicity [4]. Our patient’s mercury level was 54 ug/L.

Management of metallic mercury toxicity depends on severity of the illness, which in turn depends on the quantity injected, and the extent of tissue distribution. Embolization to the lung is seen in most cases. In rare cases, the mercury passes throughout the pulmonary vascular bed and may enter into systemic circulation resulting in liver disturbances, acute renal failure, anemia, various GI and neurologic abnormalities. Based on the mercury level, DMSA (Dimercaptosuccinic acid) or British Anti-Lewisite (BAL) may be given as a chelating agent to remove the mercury. Mercury may remain in the body for long periods in some instances. However, it is believed that the emboli are converted slowly to metallic oxides and organic compounds of mercury and slowly eliminated primarily through the renal system [6] [8]. Those with large mercury deposits in accessible areas may benefit from surgical excision [7]. Those who develop renal failure often require hemodialysis [6].

4. Conclusion

Disseminated elemental mercury thromboembolism is a rare diagnosis to make with the emergency department complaint of chest pain. The chest radiograph will provide the provider with radiograph evidence of a radiopaque intravascular exposure that will require confirmation via blood and urine mercury levels. The patient’s need for chelation therapy for removal of the mercury is based on clinical symptomatology and/or the mercury level on blood or urine toxicology screens. The amount of mercury detected does not have a direct relationship to clinical symptoms or end-organ damage. This patient was fortunate to have no long-term sequelae from his intentional parenteral mercury administration [4] [5].
References


Alveolar Ridge Preservation of an Extraction Socket of Fractured Maxillary Lateral Incisor

Hasan Ayberk Altug*, Abdullah Tugrul Coskun, Aydin Ozkan, Tamer Zerener, Metin Sencimen
Department of Oral and Maxillofacial Surgery, Gulhane Military Medical Academy, Ankara, Turkey
Email: aybork@yahoo.com

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Abstract

Background: Alveolar ridge resorption still continues to be a problem in oral surgery. Cause of bone resorption is including tooth extraction, periodontal disease and inflammatory periapical pathologies. Various methods and materials have been suggested to minimize this resorption. Aim: Goal of this case report is to present alveolar ridge preservation (ARP) following horizontally fractured maxillary lateral incisor with allograft in the aesthetic zone. Case presentation: 30-year-old female patient with fractured tooth was treated by grafting and insertion dental implant. Fractured tooth extraction was performed and extraction socket augmentation was performed by allograft and covered with collagen membrane. Augmented area was treated with bone-level implant. Definitive prosthesis single-tooth porcelain-fused-to-zirconia restorations were fabricated. Conclusions: Before implant insertion, extraction and grafting socket procedure is appropriate treatment for of fractured teeth with granulation tissue.

Keywords
Dental Trauma, Maxilla, Incisor, Allograft, Dental Implant

1. Introduction

Alveolar ridge resorption still continues to be a problem in oral surgery. Cause of bone resorption includes tooth extraction, periodontal disease and inflammatory periapical pathologies. Age, gender, systemic diseases (diabetes, etc.) and trauma are the predisposing factors that affect alveolar bone resorption [1]-[3]. Root fractures caused by a trauma constitute approximately 7% of dental injuries and may give rise to complicated tooth extraction. This situation may cause the placement of dental implants to be difficult. Alveolar ridge resorption is accelerated after tooth extraction in the following six months [4]-[6].

*Corresponding author.
Various methods and materials have been suggested in order to minimize this resorption [3] [7]-[11]. Although guided bone regeneration increases alveolar ridge height and width, because of its various difficulties, socket preservation has become a more popular method lately [9].

Aim of this case report is to present alveolar ridge preservation (ARP) following horizontally fractured maxillary lateral incisor with allograft in the aesthetic zone. Allograft was performed immediately in an extraction socket for implant region development.

2. Description of the Case

A 25-year-old female patient has referred to our clinic with infection complaints of the left maxillary lateral incisor. As the patient told us, her anterior maxillary teeth were injured as a result of being hit by a stone four years ago. Root canal treatment was applied to right maxillary central incisor and left maxillary lateral incisor (had horizontal fracture in the middle third) by patient's local dentist (Figure 1(a) and Figure 1(b)).

In radiographic examination by Cone Beam CT (CBCT), the left maxillary lateral incisor fractured horizontally had a root-filling and a granulation tissue in middle thirds (Figure 2). The luxation of this tooth was determined clinically (Figure 3).

Figure 1. (a) A periapical radiograph taken four years ago; (b) A periapical radiograph taken two years ago. Granulation tissue was seen.

Figure 2. A CB-CT of the left maxillary lateral incisor horizontally fractured.
Patient informed about potential risks of operation. Her consent form was taken. Local infiltration was administered using articaine hydrochloride 4% with epinephrine 1:200,000 (Ultracaine DS, Hoechst Marion Roussel). Envelope flap design was applied to expose bone margin of the tooth to be extracted. #14 extraction was performed and socket was curetted to remove any granulation tissue (Figure 4(a) and Figure 4(b)). Extraction socket augmentation was performed by mineralized solvent-dehydrated bone allograft (Puros, 250 - 1000 micron particle size) and covered with collagen membrane (BioMend Extend, 15 * 20 mm). The wound was closed with 4 - 0 nylon suture. After surgery, patient used amoxicillin (500 mg, four times a day for five days), chlorhexidine oral rinse (0.12%, six times a day for 10 days). Sutures were removed 10 days after the surgery.

The augmented area (Figure 5) was treated with bone-level implant (Institute Straumann AG, Waldenburg, Switzerland) after six months. Implant length 12 with 3.3-mm diameter (Figure 6). Prosthetic procedure was performed after four months (Figure 7).

Allograft was performed immediately in the extraction socket for implant region development. The primary stability of inserted implant was 53 ISQ and secondary stability was 71 ISQ. Postoperative healing follow-up was uneventful. Definitive prosthesis single-tooth porcelain-fused-to-zirconia restorations were fabricated for all four anterior incisors. There were no symptoms like infection, bleeding or pocket around the implant.

3. Discussion

Maxillofacial trauma can disrupt the integrity of the tooth, therefore its extraction might be inevitable. Major etiological factor of alveolar bone resorption is the extraction of a tooth [12]-[14]. The presence of sufficient buccal alveolar bone is essential for alveolar ridge preservation. Bone graft as autogenic, allogenic, and heterogenic with or without barrier membrane, synthetic materials, platelet rich fibrin gives a good result for ARP [3] [6] [7] [9] [11] [15].

In contrast to our case, the horizontal fractures are described in the central maxillary incisors in the literature. These conditions lead to aesthetic, functional, and healing problems [16]. According to Andreasen et al. [4], recovery of root fractures is divided into four different types: a) recovery with hard tissue, b) recovery with conjunctive tissue interposition, c) recovery with bone and conjunctive interposition between the fragments, and d) recovery with granulation tissue between the fragments. Recovery was with granulation tissue all over the middle thirds of root in our case at the end of four years. Therefore, we extracted the left maxillary lateral incisor, persevered and augmented the socket, and inserted bone-level implant. Although Gu et al. [17] asserted tissue-level implant uses in anterior maxilla, many authors [18] [19] published articles which state that bone-level implant is a more concise surgical procedure and it leads to less bone resorption.

Covani et al. [20] reported that immediate implant placement is a valid treatment which induces predictable results. In this current case, we have extracted fractured tooth, augmented the socket and inserted dental implant step by step because of the granulation tissue around the root.

Barone et al. [3] indicated that use of corticocancellous porcine substitute and resorbable membrane succeed in reducing alveolar contour from remodeling when compared to non-preserved extraction sockets.
Figure 4. (a) A view of extraction area; (b) Extracted left maxillary lateral incisor.

Figure 5. A CB-CT of the grafted area.

Figure 6. A periapical radiograph of dental implant.
Kim et al. [15] reported that autogenous tooth bone graft material can be a preferable bone substitute for extraction socket graft because of its good bone remodeling and osteoconductivity in the series of their cases. We have reached reasonable aesthetic and functional results by using allograft.

Cheah et al. [9] demonstrated that both calcium sulfate-platelet rich plasma and calcium sulfate were effective in socket preservation application and the amount of the mineralized bone component was significantly higher in the calcium sulfate-platelet rich plasma group in comparison to the calcium sulfate group.

According to the aforementioned literature, if the volume of the alveolar ridge can be maintained after extraction of tooth, dental implant insertion would be simplified. Although insertion of dental implants right after tooth extraction is a valid treatment method, extraction and grafting socket procedure is more pertinent for treatment of fractured teeth with granulation tissue.

4. Conclusion

Although dental implants insertion right after tooth extraction are a valid treatment application, extraction and grafting socket procedure is more suitable for treatment by implant of fractured teeth with granulation tissue.

References


Cutaneous Metastatic Disease: Case Series in a Tropical Setting

Maurice E. Asuquo1*, Aniefon N. Umana2, Victor I. C. Nwagbara1, Martin Nnoli3, Theophilus Ugbem3

1Department of Surgery, University of Calabar, Calabar, Nigeria
2Department of Otorhinolaryngology, University of Calabar, Calabar, Nigeria
3Department of Pathology, University of Calabar, Calabar, Nigeria

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Abstract

Background: Cutaneous metastasis is valuable though with infrequent occurrence in clinical practice. It is of esteem value in diagnosis as well as treatment of cancer due to the ease of accessibility for clinical examination and biopsy. Case Series: This is a presentation of 5 consecutive patients with histologic diagnosis of cutaneous metastatic malignancies at the University of Calabar Teaching Hospital, (UCTH), Calabar, Nigeria from 2010 to 2013. They were studied in an attempt to evaluate the clinical significance of cutaneous metastatic nodules/disease in a tropical setting. This was compared with total cutaneous malignancies and total malignancies seen over the same period. Conclusion: The spectrum in the reported cases ranged from localised and barely noticeable nodules to generalised nodules. Clinicians are urged to show renewed interest in cutaneous nodules in view of the estimable value by subjecting such for histological evaluation.

Keywords

Cutaneous Malignancy, Metastatic

1. Introduction

Cutaneous metastases arising from carcinomas though rare are important findings in clinical practice [1] [2]. It is reported to occur in 0.7% - 5% of cancer patients [3], some have reported up to 9% [4], and others 10.4% [5]. In Calabar, the authors setting metastatic skin cancer comprised 6.3% of total histologically diagnosed skin cancer
It is relatively uncommon but often a reflection of the presence and course of an internal disease [2] [7]. Cutaneous metastasis is very rare in children and varies according to sex. Metastasis from the colon including lungs is common in males while that from the breast commoner in females [1] [2]. Clinical findings may be barely noticeable and require a high index of suspicion [2]. Skin metastasis presents commonly as skin nodules (single or multiple), usually painless and may mimic dermatofibroma, neurofibroma, lipoma and granuloma [1] [2] [8]. Some malignancies have a predilection for areas of metastasis, which can be useful in directing search for the primary site [2]. Presentation is classified into the following settings: a) develop after initial diagnosis of primary malignancy, b) carcinoma of unknown primary site (CUPS), c) detected as the same time of diagnosis [1] [2]. Diagnosis is based on histology and the pattern provides clue to the location of the primary malignancy [1]. In addition, immunohistochemistry provides a useful adjunct in directing search of the primary tumour [2] [9]. Olfactory neuroblastoma is a rare slow growing malignant tumour that arises from neuroepithelial cells of the olfactory membrane. Scalp metastasis from olfactory neuroblastoma has been described [10]. Treatment is palliative and simple excision is the treatment of choice [2] [3]. We present this study to enkindle the significance of cutaneous metastasis in clinicians and urge for renewed interest.

2. Case Series

Case 1
H. I, a 65-year-old male presented with recurrent abdominal pain and swelling of 9 months duration. He noticed recurrent episodes of right sided abdominal pain and admitted to episodes of alternating history of constipation and diarrhea. However, there was no history of passage blood in stool but admitted to history of weight loss and gets tired easily. His attention was drawn to the presence of an umbilical swelling as part of his clinical evaluation 6 months after the onset of his ailment.

Examination revealed a chronic ill-looking patient, anicteric, pale, afebrile with no peripheral lymphadenopathy. Pulse was 76 beats per minute and Blood pressure 140/70 mm/Hg. Chest examination was normal. Abdominal examination revealed no organomegally except for a mild tenderness on palpation of the right flank in addition to an umbilical nodule that was non tender, firm, measuring 2 × 1.5 cm. Rectal examination was not remarkable. A preoperative diagnosis of colonic cancer was made.

Haemogram showed a packed cell volume (PCV) of 29%, white blood count (WBC) 5.3 × 10^9/l, (lymphocytes 27%, neutrophils 70%, monocytes 2%, and eosinophils 1%), Platelets 257 × 10^9/l, ESR 80 mm/hr. Faecal occult blood was positive. Carcino embryonic antigen (CEA) was 7.0 ng/ml. Chest X-ray (CXR) was normal. Abdominal computerized tomogram (CT) was reported as revealing features suggestive of malignant mass in the ascending colon. Biopsy of the umbilical nodule revealed metastatic adenocarcinoma.

Patient was commenced on haematinics counseled for surgery which he declined and was lost to follow-up after a month.

Case 2
A. E, was a 75-year-old female, she presented with a mass located at the upper part of the back of 4 months duration. She however did not present with any other symptoms.

Examination revealed a patient in apparent good health, anicteric, not pale, afebrile with no peripheral lymphadenopathy. Pulse and Blood pressure 140/70 mm/Hg. Chest examination was normal. Abdominal examination was essentially normal but for 3 discrete nodules distributed along the laparotomy scar. They were non tender, hard with measurements that ranged from 1.5 × 2 cm.

Haemogram revealed a PCV 34%, WBC 5.8 × 10^9/l, (Neutrophils 70%, lymphocytes 28%, monocytes 1%, eosinophils 1%), platelets 276 × 10^9/l. Biopsy of the mass revealed metastatic adenocarcinoma suggestive of colonic site.

Patient however, declined further evaluation on account of cost and was lost to follow-up after one month.

Case 3
J. T, was a 56-year-old female who presented with multiple swellings on the scar of a previous operation of 3 months duration. She was offered a right hemicolectomy 3 months ago for a diagnosis of carcinoma of the caecum that presented as an emergency with malignant intestinal obstruction.

Examination revealed a patient in relative good health, anicteric, afebrile, not pale with no peripheral lymphadenopathy. Pulse and Blood pressure 140/70 mm/Hg. Chest examination was normal. Abdominal examination was essentially normal but for 3 discrete nodules distributed along the laparotomy scar. They were non tender, hard with measurements that ranged from 1.5 × 1.0 to 1.5 × 2 cm. Rectal examination was normal.
Haemogram showed a PCV of 33%, WBC $5.0 \times 10^9$/l (Neutrophils 70%, lymphocytes 26%, monocytes 2%, and eosinophils 2%), platelets $272 \times 10^9$/l. CXRay and abdominal ultrasound scan (USS) were reported as normal. CEA was 5.3 ng/ml. Nodules were excised and histology revealed metastatic adenocarcinoma. She was commenced on adjuvant chemotherapy with 5FU but was however lost to follow-up after 2 months.

Case 4

I. S. E, a 37-year-old male barber presented with multiple skin masses located on the forehead, left shoulder, anterior abdominal wall, back, gluteal region and left foot; (Figure 1(a) and Figure 1(b)) of 2 months duration as a referral from the ear, nose and throat (ENT) clinic.

Nine months prior to this he presented to the ENT clinic with a 2 month history of left eye protrusion and left nasal obstruction of 6 months duration. Clinical examination then revealed a chronically ill-looking man with left cervical lymphadenopathy ($3 \times 4$ cm) with supraorbital proptosis of the left eye with a firm mass on the medial canthus. Visions in both eyes were intact. CT of the paranasal sinuses showed left orbital mass involving the nasal cavity, ethmoidal, maxillary and sphenoidal sinuses. Histology and immunohistochemistry of the mass revealed olfactory neuroblastoma.

He was offered 3 courses of 5FU, cisplatin and zofran fortnightly and 2 weeks of radiation which initially resulted in near resolution of the mass and nasal obstruction. However, 6 weeks after commencement of chemoradiation, multiple skin masses appeared.

Excision of the masses at the shoulder and anterior abdominal wall revealed metastatic olfactory neuroblastoma (Figure 2(a) and Figure 2(b)). He succumbed to the metastatic disease within 11 months of onset of the illness and 2 months of the development of cutaneous metastases.

Figure 1. (a) Clinical Photograph-Showing metastatic nodule forehead, (b) clinical photograph showing metastatic nodules on the left side of the back and buttocks.

Figure 2. (a) Photomicrograph H & E $\times 40$ metastatic olfactory neuroblastoma, (b) photomicrograph H & E $\times 100$ metastatic olfactory neuroblastoma.
Case 5

H. E, a 60-year-old retired female civil servant who presented to the surgical outpatient department (SOPD) as a referral from the General Hospital in Calabar with a 5 month history of abdominal pain and progressive abdominal distension of 4 months duration following umbilical herniorrhaphy. She reported to have had a painless reducible umbilical swelling since childhood that became painful and irreducible and was offered herniorrhaphy, Figure 3. The post operative period was reported as uneventful; however, 4 weeks after surgery, she developed above presenting complaints. Pain initially located in the epigastrium was burning and radiated to both flanks. It later became colicky slightly relieved by lying on her sides. There was associated non bilious vomiting, intermittent constipation and passage of pelletized faeces with periods of dark tarry stool.

She was not a known peptic ulcer disease patient, known hypertensive and non diabetic but admitted to loss of weight. She was commenced on anti Kock’s therapy to no avail prior to referral. The onset of increasing dyspnoea, abdominal swelling and pedal oedema necessitated her referral.

Examination revealed acute-on-chronic ill looking patient in respiratory distress, anicteric, pale, afebrile with bilateral pitting oedema and no peripheral lymphadenopathy. Respiratory rate was 30 cycles/min. Pulse rate was 92 beats/min, BP 160/90 mm/Hg. Abdominal examination revealed a grossly distended abdomen that moved with respiration. There was a transverse scar across the umbilicus with 2 Sister Mary Joseph’s nodules, Figure 3. Organs were not ballotable due to gross ascites. Vaginal and digital rectal examinations were unremarkable. A diagnosis of metastatic colonic cancer was made.

Full blood count showed, PCV 32%, WBC 4.5 × 10^9/l (lymphocytes 24%, neutrophils 72%, monocytes 4%), platelets 252 × 10^9/l, ESR 90 mm/hr. CXRay revealed features of hypertensive heart disease. Abdominal CT showed features in keeping with advanced transverse colonic tumour. Histology of umbilical nodule was reported as metastatic poorly differentiated adenocarcinoma, (Figure 4(a) and Figure 4(b)).

She was commenced on antihypertensive drugs, iron dextran and had 2 courses of 5FU. Dyspnoea persisted despite 2 episodes of abdominal paracentesis an succumbed to metastatic disease 5 months post appearance of the umbilical nodule.

3. Discussion

Cutaneous metastasis from primary internal malignancies is uncommon. The total number of cutaneous malignancy that presented in the authors setting (2010-2013) was 60 [55(92%) primary, while 5(8%) were metastatic], and total cutaneous malignancy accounted 10% of total malignancies during the same study period, Table 1. The 5(8%) consecutive patients presented accounted for 0.8% of total malignancies. Aksoy S et al. reported a frequency of cutaneous metastasis that ranged between 0.7% and 10.4% of all patients with malignancies [11]. This compares favourably with this report (0.8%) and our earlier report in which cutaneous metastasis accounted for 6.5% of total cutaneous cancer and 0.65% of total malignancy [1]. Virtually every cancer can present with cutaneous metastasis, however some do so more often than others do. The most frequent non-cutaneous tumours associated with skin metastasis are breast, lung, colorectal [11]. This study revealed the colon as the commonest
Figure 4. (a) Photomicrograph H & E × 40 metastatic adenocarcinoma, (b) photomicrograph H & E × 100 metastatic adenocarcinoma.

Table 1. Summary of clinicopathological features (2010-2013).

<table>
<thead>
<tr>
<th>S/N</th>
<th>Age</th>
<th>Sex</th>
<th>Clinical presentation (Diagnosis)</th>
<th>Site of cutaneous nodule</th>
<th>Histology</th>
<th>Remarks</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>68</td>
<td>M</td>
<td>Recurrent large bowel adenocarcinoma (ascending colon)</td>
<td>Umbilicus</td>
<td>Metastatic adenocarcinoma</td>
<td>Refused surgery. Lost to follow up</td>
</tr>
<tr>
<td>2.</td>
<td>75</td>
<td>F</td>
<td>Irregular hard cutaneous mass</td>
<td>Left scapula region</td>
<td>Metastatic adenocarcinoma</td>
<td>Lost to follow up</td>
</tr>
<tr>
<td>3.</td>
<td>56</td>
<td>F</td>
<td>Multiple nodules scar of previous laparotomy</td>
<td>Anterior abdominal wall (midline scar)</td>
<td>Metastatic adenocarcinoma</td>
<td>Adjuvant chemotherapy 5FU. Lost to follow up</td>
</tr>
<tr>
<td>4.</td>
<td>37</td>
<td>M</td>
<td>Multiple cutaneous masses Figure 1</td>
<td>Fore head, abdomen back gluteal, left foot</td>
<td>Metastatic olfactory neuroblastoma Figure 2(a) and Figure 2(b)</td>
<td>Mortality. 2 months (post cutaneous nodules)</td>
</tr>
<tr>
<td>5.</td>
<td>60</td>
<td>F</td>
<td>Abdominal distension umbilical nodules Figure 3</td>
<td>Umbilicus</td>
<td>Metastatic adenocarcinoma Figure 4(a) and Figure 4(b)</td>
<td>Hospital mortality 5 months (post cutaneous nodules)</td>
</tr>
</tbody>
</table>

Total cutaneous malignancy 60 (55 (92%)—primary, 5 (8%)—Metastatic). Total cutaneous malignant 10% of total malignancy. Total cutaneous metastatic malignant—0.8% of total malignancy.

primary site for cutaneous metastasis in these series in our setting. Cutaneous metastases from abdominal cancers are uncommon occurring in less than 5% of patients [12].

Cutaneous metastases are detached extensions of primary tumours to the skin. Four metastatic pathways namely regional spread (through body cavities), transplantation (via surgery or other invasive procedures), lymphatic (common for carcinomas), and haematogenous (common for sarcomas) have been documented [2]. In keeping with these pathways, metastatic sites are usually predictable though unusual sites and wide spread metastases may occur in keeping with our patients with colonic primary sites spreading to the anterior abdominal wall/back and metastatic olfactory neuroblastoma respectively.

Majority of cases present with painless nodules after diagnosis [2]. This was the experience in all our patients. Most cutaneous metastases occur near to the primary as depicted in this study, 2 (40%) patients presented with Sister Mary Joseph’s nodules with primary malignancies in the colon, while another developed nodules on the
laparotomy scar following right hemicolectomy for carcinoma of the caecum. However, in rare circumstances, (Case 2), cutaneous metastasis may be the only sign of the disease post operative or present as carcinoma of unknown primary site (CUPS), [11].

Cutaneous metastasis may occur at any time in the course of the malignancy and spread may follow, direct extension, lymphogenous, intravascular dissemination or surgical implantation as noted in a patient [4] [11]. Development of extensive cutaneous metastatic disease as demonstrated in the patient with metastatic olfactory neuroblastoma while under treatment with chemotherapy and radiotherapy represents failure of current treatment and possibly an indicator of poor prognosis. A similar phenomenon has been described with gastric carcinoma with cutaneous metastases on the head, neck, axilla, chest and upper limb [13]. Late presentation with attending complications is a common event in our setting. Two (40%) patients presented with malignant intestinal obstruction with metastatic nodules already manifest.

Diagnosis is by histology and comprehensive evaluation includes assay for markers, imaging, and immunohistochemistry especially when in search for the primary site as in CUPS [2]. Cutaneous lesions may be primary or metastatic, certain features are characteristic of metastatic lesions as depicted in this study. Most of the tumour is found in the deep reticular dermis and subcutaneous Figure 4(b) and neoplastic cells in between collagen bundles. The epidermis is usually not involved, Figure 4(a), except in later stages with direct spread [1] [2] [4]. Metastasis is classified into adenocarcinoma, Figure 4(a) and Figure 4(b), squamous cell carcinoma or undifferentiated (anaplastic). The later require immunohistochemistry to establish the tissue of origin [1].

Treatment is palliative aimed at improving the quality of life especially when indicated by disfigurement or pain. Wide local excision of the cutaneous lesion is recommended. Other modalities include cryotherapy, photodynamic therapy, radiation, intralesional, or topical chemotherapy [1] [2] [10].

The outcomes were generally poor, 2 (40%) patients were recorded mortalities (5 months and 2 months after diagnosis cutaneous metastases), while others were lost to follow up with advance primary disease. Mortality rate is high; however, some improvement has been recorded with chemotherapy. Overall survival has been reported between 3 - 9 months in keeping with this report [2]. Others have reported median survival of 6.5 months after cutaneous metastasis and 50% of patients die with 6 months with cutaneous metastasis [14].

Following the increasing incidence of malignancies, dermatologists including dermatopathologists and generally clinicians are likely to encounter cutaneous metastases hitherto regarded as insignificant and are of esteemed value in directing search for the associated primary malignancy [15]. Cutaneous metastatic disease detected in patients in with malignant intestinal obstruction may be an indicator of poor prognosis, [16]. Clinicians are encouraged to search for this and subject skin nodules to pathologic evaluation in view of the treasured value in diagnosis and search for primary malignancies.

4. Conclusion

Cutaneous metastasis though not a frequent event shows a pattern of clinical significance hence the request for renewed interest by clinicians. Recognition may be a pointer to an undiagnosed disease and when detected warrant a thorough metastatic work up that may result in accurate staging of the disease. Meticulous surgical techniques can prevent skin metastasis from implantation while wide spread metastases may be an indicator of poor prognosis.

References


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