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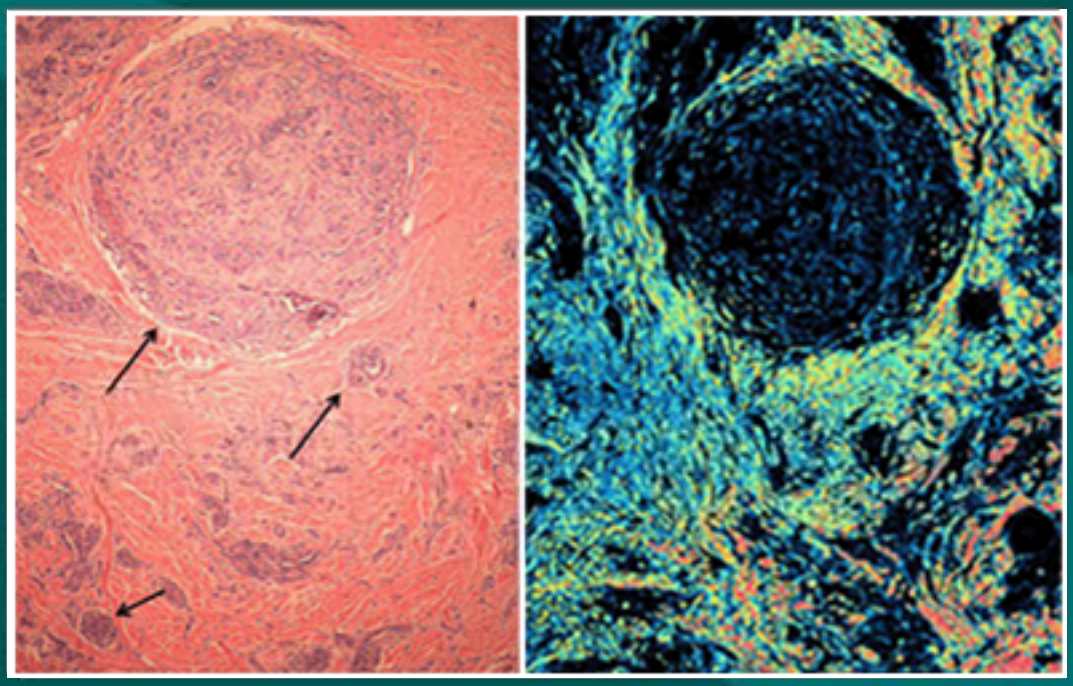

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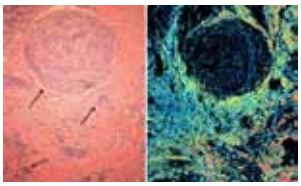

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CASE SERIES OPEN ACCESS

Pouch and tunnel technique: Minimally invasive periodontal plastic surgery for root coverage

Sangeeta Singh

ABSTRACT

Introduction: The indications for root coverage surgery are very well defined today eliminating unnecessary repeated surgeries due to failures resulting from improper case selection. The results of tunnel technique and its modifications combined with the use of connective tissue grafting have been quite predictable. Case Series: This case series comprises three cases of multiple adjacent recessions which were treated using pouch and tunnel technique with connective tissue grafting from palate using Liu's Class 1a incision. This technique resulted in excellent root coverage and complete healing of the donor site. The cases have been followed postoperatively varying from 9 to 18 months. Conclusion: The pouch and tunnel procedure is ideal for treatment of multiple adjacent recessions in a single surgical procedure demonstrating early healing and highly predictable root coverage results.

Keywords: Marginal tissue recession, Minimally invasive technique, Peridontal plastic surgery, Pouch and tunnel procedure, Root coverage surgery

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INTRODUCTION

Marginal tissue recession as a clinical entity has been documented quite elaborately in literature [1, 2]. The indications for surgical intervention are quite well defined and it is essential to carry out root coverage surgery whenever concerns such as aesthetics, sensitivity, susceptibility to root caries pulpal symptoms due to root exposure, food lodgment and plaque accumulation exists [3]. Surgical techniques for root coverage have been invented and modified over the last few decades with encouraging results and most of the procedures followed today result in 80–90% root coverage. Currently, accepted procedures for root coverage include coronally advanced flap, sub-epithelial connective tissue graft, guided tissue regeneration and acellular dermal matrix. The tunnel technique followed in this case report is the one described by Langer et al. where multiple adjacent recessions can be covered in a single surgery using connective tissue graft from the palate.

CASE SERIES

The three cases selected for root coverage using the pouch and tunnel technique had multiple adjacent recessions. The most important criterion for selecting this procedure was sufficient thickness of the marginal and papillary gingival at the recipient site so as to facilitate complete undermining for creating the pouch and tunnel without detaching the papillary tip.

Case 1: A 36-year-old female reported with the chief complaint of sensitivity in lower right posterior teeth. On examination there was Millers Class III recession in relation to 44 and 45 (Figure 1). The width of attached gingiva was found to be inadequate since the tension test was positive. After phase I therapy, a pouch and tunnel technique utilizing a connective tissue graft was planned for root coverage. A sulcular incision was made through each recession area and the tissues gradually undermined including the base of the interdental papilla without the tip and the undermining extended up to the mucogingival junction so as to relax the flap sufficiently to allow placement of the connective tissue graft. Thus gradually a pouch and tunnel was prepared connecting the recipient sites for placement of the graft (Figure 2). The connective tissue graft was harvested from the palate using Liu's Class 1a incision (Figure 3). This graft was then placed using a technique described by Zabaluigi et al. where two resorbable sutures of different colors were placed, one on either side of the graft. Using these sutures, the graft was gradually manipulated into the pouch and through the tunnel to cover the adjacent recipient sites. Once the graft was completely inside the tunnel, it was positioned coronal to the cemento-enamel junction. The ends were sutured with a simple square knot (Figure 4). A periodontal pack was placed both at the recipient site as well as the donor site using an acrylic stent for the palatal placement.

Case 2: A 31-year-old male presented with the chief complaint of sensitivity in the lower teeth and was clinically diagnosed as a case of Millers Class II recession in relation to 33 and 34 (Figure 5). The same surgical technique, as in Case 1, was used to place the connective tissue graft in the pouch created in relation to 33 and 34.

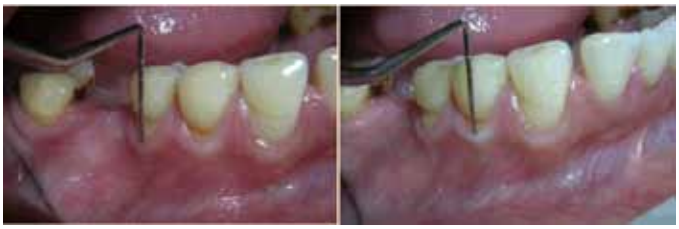

Figure 1: Millers Class III recession in relation to 44 and 45 (Case 1).

Figure 2: Pouch and tunnel preparation (Case 1).

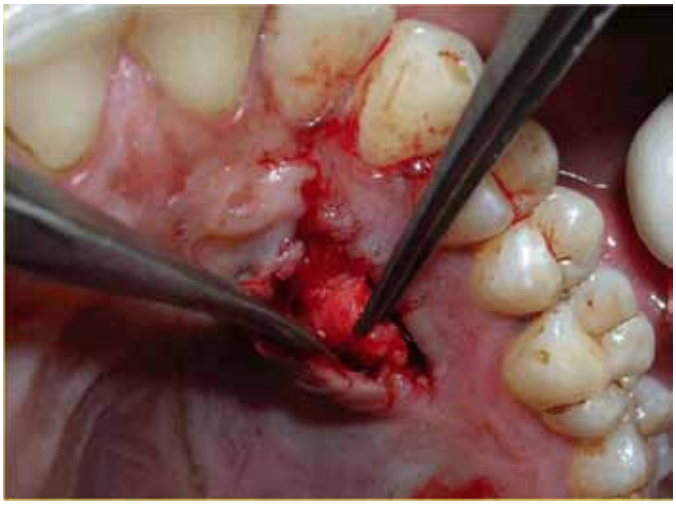

Figure 3: The connective tissue graft was harvested from the palate using Liu's Class 1a incision.

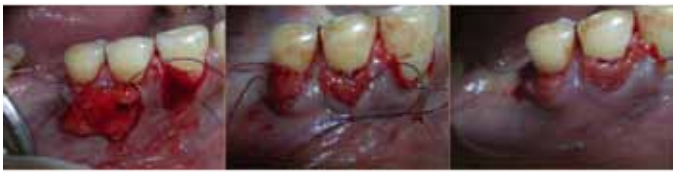

Figure 4: The graft placed into the tunnel and sutured.

Case 3: A 35-year-old female patient presented and her main concern was the gums 'going down' in relation to the lower teeth on the left side. She was diagnosed as a case of Millers Class II recession in relation to 34 and 35 (Figure 6). This case too was treated using the pouch and tunnel technique with placement of CTG procured from the palate. The donor site appeared normal in color and healthy after four weeks and the recipient site was healthy with excellent color match with adjacent tissues in all four cases. These results were stable and maintained at the time of review, 12 months in Case 1 (Figure 7), 18 months in Case 2 (Figure 8), six months in Case 3 (Figure 9). The mean root coverage achieved in these cases was an average 90% which is close to the mean root coverage of 91.6% in the Zabaluigi study.

DISCUSSION

The concerns of hypersensitivity, root caries, fear of tooth loss and an increasing interest in aesthetics have led to development and modifications of surgical procedures that not only enhance the width of attached gingival but also achieve maximum root coverage addressing these concerns. The subepithelial connective tissue graft technique gives the dual advantage of excellent healing of the donor site as well as excellent color match of tissues

[4]. The tunnel technique was developed as a modification of the envelope technique to manage multiple adjacent recessions using a single surgical procedure and this technique has resulted in consistently favorable results [5].

The pouch and tunnel technique of connective tissue grafting, if performed correctly, is the most predictable periodontal plastic surgery procedure. It is a very sensitive procedure and requires use of operating microscope, microsurgical instruments, delicate handling of the tissues and ample patience while undermining the interdental papilla. The common complications that can occur are detachment of the interdental papilla, thinning of the flap, too thick connective tissue graft harvested which can lead to ischemic necrosis of the inter dental papilla or too thin connective tissue graft harvested which can lead to insufficient coverage of the recession defect [6].

The use of tunnel procedure preserves the interdental papilla and this facilitates an early and accelerated initial wound healing. The tunneling also applies less traction and preserves the gingival height [7]. The elimination of vertical incision which is used in subepithelial connective tissue grafting, ensures complete coverage of the connective tissue graft by the flap thus aids in faster healing as well as excellent color matching. The pouch and tunnel procedure may be of advantage in multiple

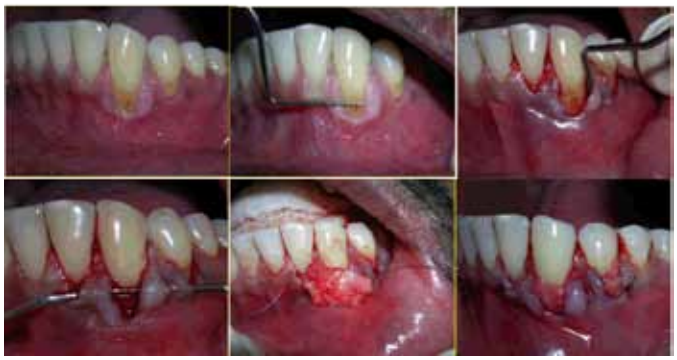

Figure 5: Millers Class II recession in relation to 33 and 34.

Figure 6: Millers Class II recession on 34 and 35.

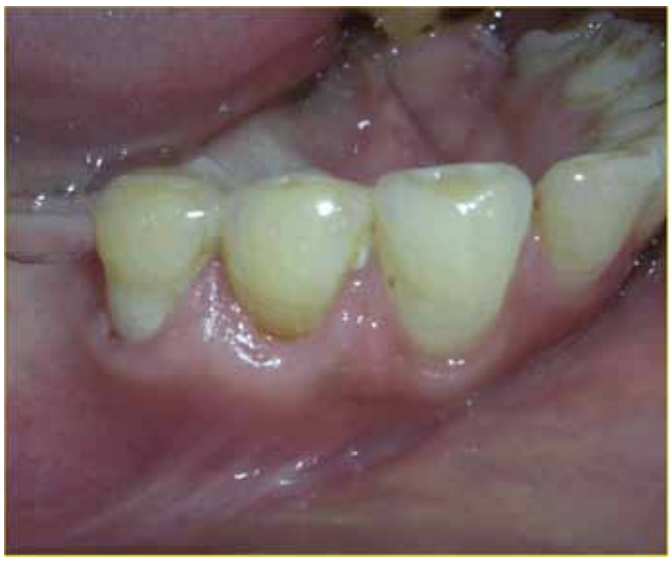

Figure 7: Postoperative image after 12 months (Case 1).

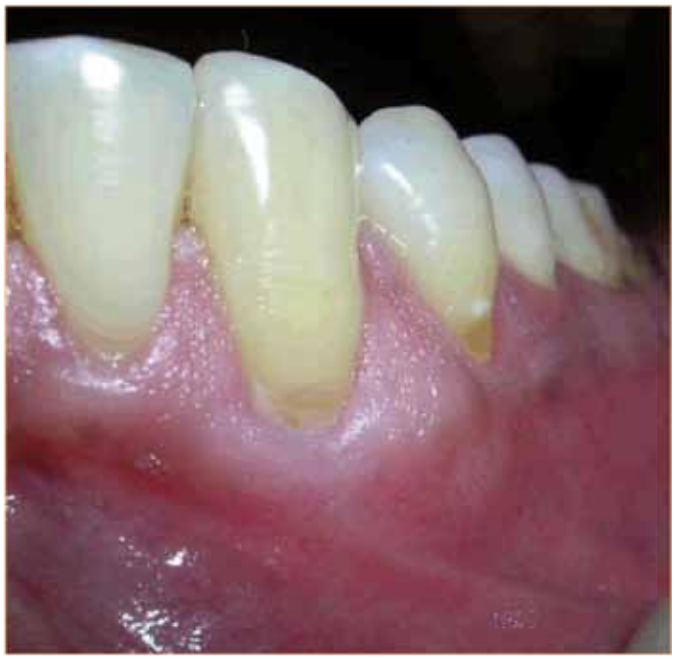

Figure 8: Postoperative image after 18 months (Case 2).

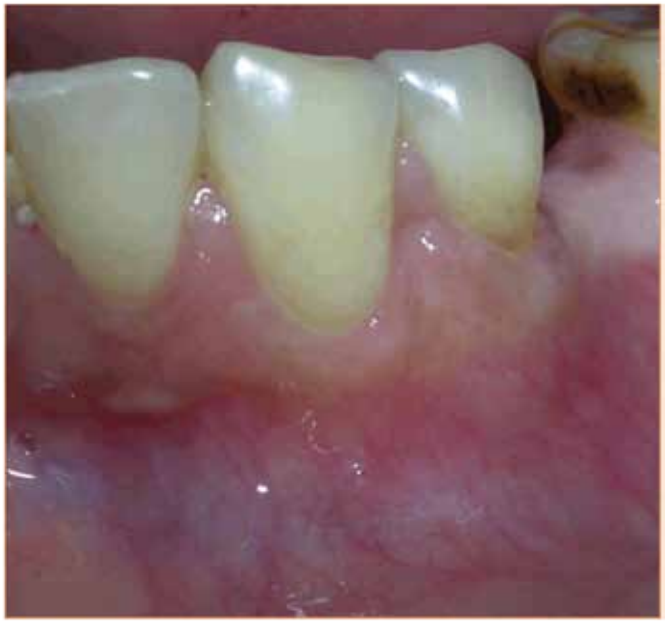

Figure 9: Postoperative image after six months (Case 3).

adjacent recessions as compared to coronally repositioned flap as there is minimum trauma to the recipient site and there is predictable root coverage [8].

CONCLUSION

The pouch and tunnel technique combines the advantages of subepithelial connective tissue grafting as well as the envelope technique thus making it an ideal choice for treatment of multiple adjacent recessions in a single surgical procedure demonstrating early healing and highly predictable root coverage results.

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Author Contributions

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

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Clamping of external carotid artery rather than embolization during surgical removal of a huge carotid body tumor

Abubakr Hashim Elrofaie Sayed Ali, Saif Eldin Mohammed Ali Ibrahim, Ashraf Mohamed Mokhtar Ali

ABSTRACT

Introduction: Carotid body tumors (CBT) are neoplasms that develop in the carotid body and are usually benign tumors. Malignant forms are less frequently. Carotid body tumors are widely known as paragangliomas. The carotid body is a gland located behind the carotid artery at the site of its bifurcation on either sides of the neck and originates from the neural crest and acts as a peripheral chemoreceptor. Case Report: A 42-year-old female presented with progressively enlarging neck swelling over five years which was associated with a recent difficulty in swallowing and hoarseness of voice. On examination there was anterolateral pulsatile neck swelling, ovoid in shape, about 6×6×7 cm in size, mobile from side to side only and firm in consistency. Computed tomography angiography (CTA) and magnetic resonance angiography (MRA) showed a mass arising at the bifurcation of common carotid artery (CCA), which has rich blood supply, splaying the external carotid artery (ECA). Blood test for vanillylmandelic acid (VMA) was negative. The patient underwent surgical excision of the tumor through an endarterectomy approach after clamping of the ECA. We report this case where clamping of ECA is being applied rather than endovascular embolization of the feeding vessels to minimize bleeding during removal of the tumor, to avoid embolization related complications and to lessen the cost of the procedure. Conclusion: Carotid body tumors are rare, however, early diagnosis and prompt treatment is essential. In a large tumor, preoperative endovascular embolization is widely used for devascularization. Clamping of the external carotid artery is a good alternative way to avoid embolization related complication with better control of bleeding, less operative time and improved cost-effectiveness.

Keywords: Carotid body tumor, Endarterectomy approach, Glomus gland, External carotid artery clamping, Paraganglioma

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INTRODUCTION

The carotid body (also known as glomus gland) is a gland located behind the carotid artery at the site of its bifurcation on either side of the neck, normally about

5 mm in size and covered with a fibrous capsule. The gland acts as peripheral chemoreceptor which releases neurotransmitter primarily in response to decrease in the arterial partial pressure of oxygen and by a lesser extent to increase in the partial pressure of CO2 and decrease in arterial pH. The gland receives afferent nerve fibers from the glossopharyngeal nerve which transmits impulses from the gland to the respiratory center in the medulla.

Carotid body tumors (CBT) are neoplasms that develop in the carotid body and are usually benign tumors. Malignant forms are less frequent. CBT's are widely known as paragangliomas. Carotid body tumors are common in 45-year females and in small portion are related to multiple primary tumor syndrome [1].

CASE REPORT

A 42-year-old female presented with a left neck swelling for five years. The swelling was small in size for the last four years. She was seen early by an ENT surgeon and advised just observation, during the last year the swelling started to increase in size with difficulty in swallowing and hoarseness of voice. There was not any other cardiovascular or neurological symptoms.

On examination she was unwell, not pale, jaundice or cyanosed. Vital signs were normal. There was anterolateral neck swelling, ovoid in shape, measuring about 6×6×7 cm in size, mobile in horizontal plane but fixed in the vertical one. It was firm but not tender with transmitted pulsation, no lymph nodes were palpable. Cranial nerves examinations were intact, and both carotid arteries were palpable, with no bruit.

Systemic examination revealed no signs of neurological or vascular deficit computed tomography angiography (CTA) and magnetic resonance angiography (MRA) of the carotid vessels showed a mass arising at the bifurcation of common carotid artery (CCA), which has rich blood supply, splaying the external carotid artery (ECA) (Figure 1). Blood test for vanillylmandelic acid (VMA) was negative.

The patient was planned for surgical removal of the tumor under general anesthesia (Figure 2). An endarterectomy approach was used, skin, subcutaneous tissues, superficial cervical fascia and platysma muscle were dissected respectively. The sternocleidomastoid muscle and jugular vein were identified and retracted laterally. The facial vein was identified, ligated and divided to reveal the bifurcation of the CCA. The carotid sheath was dissected until the tumor was identified splaying CCA, ECA, internal carotid artery (ICA), hypoglossal and glossopharyngeal nerves. Meticulous dissection and retraction of the nerves was done to maintain their integrity. Vessel loops were applied for the carotid vessels plus clamping of the origin of the ECA was carried out to facilitate dissection and minimize bleeding. Complete excision of the tumor was done in the sub adventitial plane. Following de-clamping, blood flow was restored with minimal bleeding. A drain was put in place and the wound was closed in layer (Figure 3).

The tumor specimen was sent for histopathology and results were consistent with paraganglioma.

The postoperative period passed uneventfully without neurological or vascular complication (Figure 4).

DISCUSSION

Paragangliomas are chromaffin negative neuroendocrine tumors. There are three different types

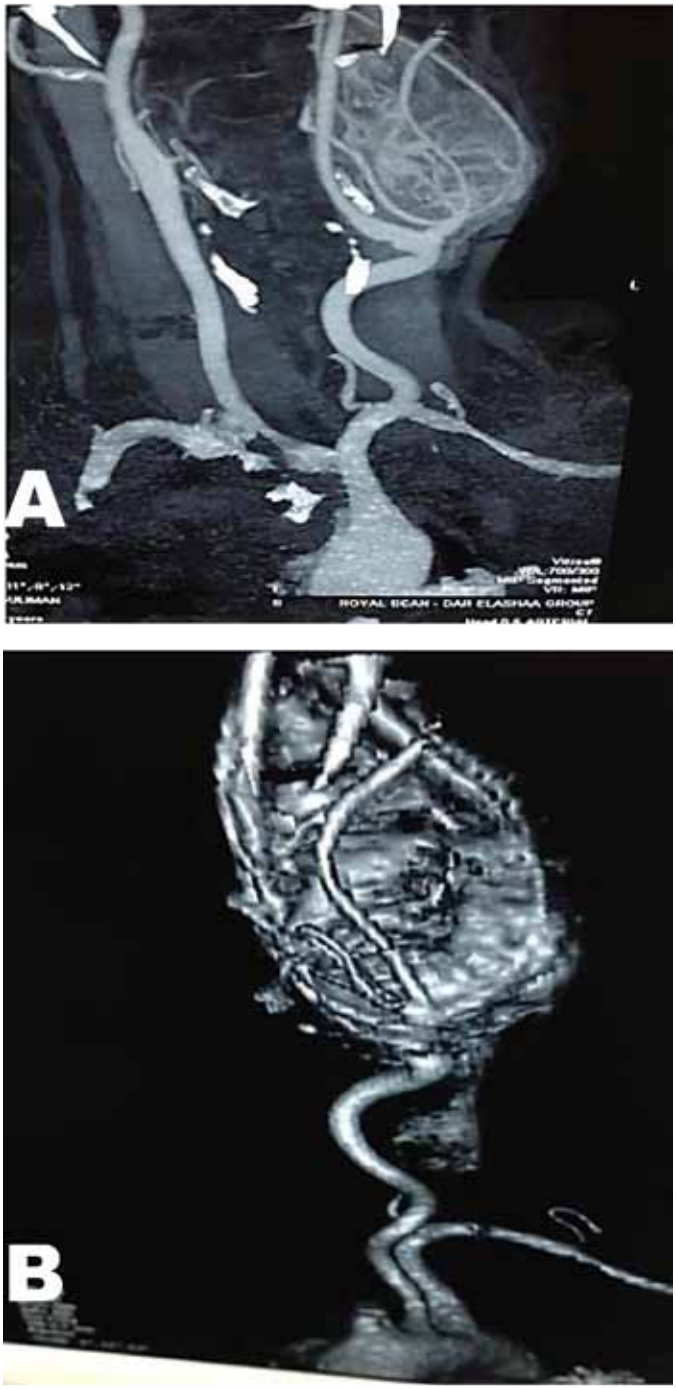

Figure 1: Imaging studies (A) MRA (B) Computed tomography angiography showing a carotid body tumors arising at the bifurcation of common carotid artery, which has rich blood supply, splaying the external carotid artery.

Figure 2 Preoperative carotid body tumors marking.

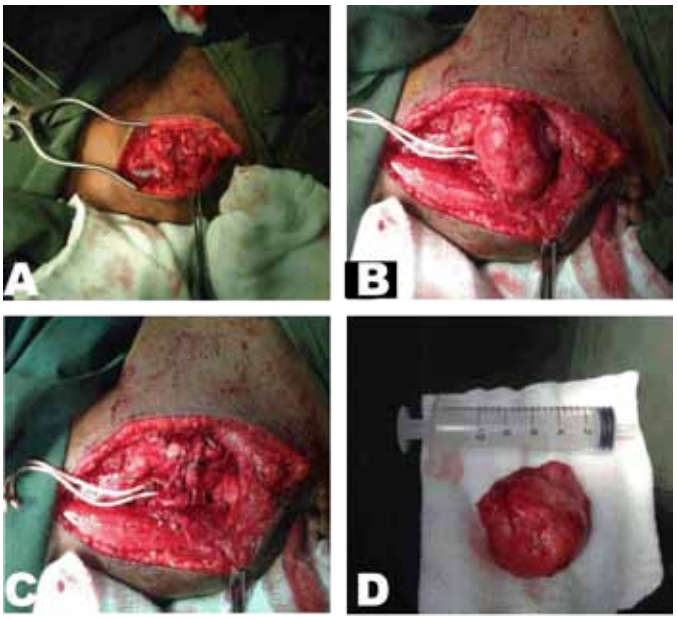

Figure 3: Operative pictures showing: (A) Incision, (B) During dissection, (C) Operative field after excision of carotid body tumors, (D) Huge excised tumor: 7 cm in length.

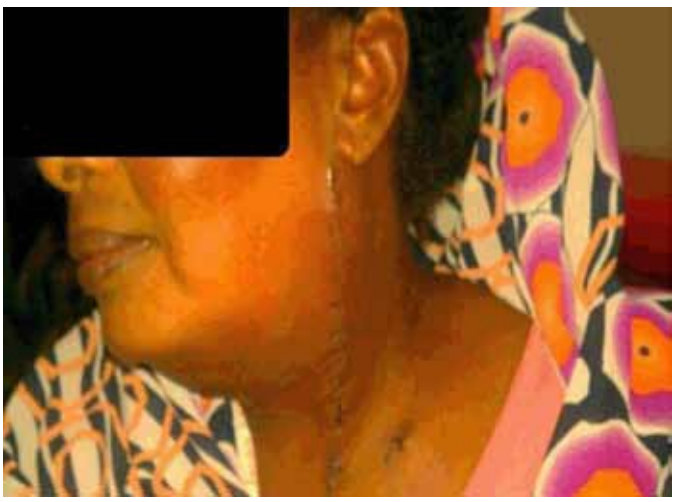

Figure 4: Completely healed scar.

of CBTs: and sporadic hyperplastic familial. The sporadic type is the most common type and represents 85% of CBTs. Hyperplastic form is very common in patients with chronic hypoxia or chronic obstructive pulmonary disease; the familial type may present in a younger age group and bilaterally.

Shamblin staging is commonly used to classify CBT into three types. Type I describes tumors which are small and easily resectable. Shamblin type II tumors are larger, slightly more adherent tumors and type III tumors are those which completely surround the carotid bifurcation.

The patient we are reporting was staged as a Shamblin type III CBT using MRA which had more than 270 degrees circumferential contact with ICA as described by Arya et al. and for smaller tumor Shamblin type I and type II were described as being less than 180 degrees, and between 180 and 270 degrees of circumferential contact with ICA, respectively [2]. The standard management is surgery and careful preoperative assessment of vascular status and the functional status of the CBT is required. Functionally, active paraganglioma necessitate the use of alpha adrenoreceptor blockade [3] but in the case of the patient we are reporting, VMA and cardiovascular examination were normal.

Much controversy lies with regards to the management of large (Shamblin grade II and III) CBT's. Some authors suggest that elderly patients with Shamblin III (large tumor more than 5 cm), which are associated with significant morbidities, should avoid surgery and receive radiotherapy instead [4]. Although surgery for CBT removal carries a 35% morbidity and a 1% mortality rate and a 19% risk of cranial nerve injury [5], surgical excision is the standard treatment as suggested by most of authors. The problem is surgical resection of large CBT is challenging, technically difficult and had an increasing risk of neurovascular injuries [6]. In order to avoid common postoperative neurovascular complication many suggested preoperative embolization of the feeding vessels could reduce blood loss and improve tumor excision [7, 8]. The ascending pharyngeal artery is the most common major feeding artery for CBTs. Tumor embolization with polyvinyl alcohol particles after super selection of feeding arteries is an option but numerous feeding arteries that could reduce the effect of embolization and the potential high risk of cerebral infarction by embolic particles is major limitation of embolization [9]. Embolization helped to some extent but it is expensive and as mentioned not suitable for all patients and is not quite safe and efficient as shown by study done by Ozay et al, of 14 patients who underwent CBT surgery and concluded that embolization before CBT surgery does not decrease blood loss or facilitate tumor removal [10].

Others suggest early ligation or deliberate resection of the ECA in Shamblin types II and III CBT resection which simple and significantly reduces the risk of stroke as opposed to percutaneous embolization which carries a significant risk of stroke [11, 12].

Spinelli et al. suggested a simpler technique for large

CBT which is clamping the origin and the distal part of the ECA with heparinization, which is much easier, and effective [13]. We used similar sufficient technique in this reported patient to create a relatively blood less field and to allow for visualization and carful resection of CBT with preservation of adjacent and closely related neurovascular structures by clamping of the origin of the ECA only and without heparinization.

External carotid artery clamping also used for other tumors resection as shown by a study conducted by Yadav et al., with regards to clamping of the ECA in excision of large meningiomas, they concluded that temporary clamping of ECA is a safe, simple and cost-effective alternative method to embolization [14].

Regarding the administration of heparin intraoperatively, any injury to the carotid vessels requiring clamping of the CCA, ICA or ECA needs heparinization to lower the risk of complications [15], however, we did not use heparin during clamping of the ECA because excision of the tumor was performed without arterial injury.

CONCLUSION

Carotid body tumors are rare, however, early diagnosis and prompt treatment with complete surgical excision is essential. In a large tumor preoperative endovascular embolization is widely been used for de-vascularization. Clamping of the origin of external carotid artery is a good alternative way to avoid embolization related complication with better control of bleeding, less operative time and improved cost-effectiveness.

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Author Contributions

Abubakr Hashim Elrofaie Sayed Ali – Acquisition of data, Drafting the article, Final approval of the version to be published

Saif-Eldin Mohamed Ali Ibrahim – Conception and design, Critical revision of the article, Final approval of the version to be published

Ashraf Mohamed Mokhtar Ali – Conception and design, Critical revision of the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Ameloblastic carcinoma of the mandible: A case report

Soumithran C. S., Sudha S., Ikram Bin Ismail P. T., Ambadas, Jibin Jose Tom, Seeja P.

ABSTRACT

Introduction: Ameloblastic carcinoma is an extremely rare, malignant epithelial tumor of the jaws with a poor prognosis. The most common site of occurrence is the posterior mandible. Clinically, it is very aggressive and has potential for extensive local destruction. Majority of the cases arise de novo (primary type), but a few cases arise from a pre-existing ameloblastoma (secondary type). Case Report: A 30-year-old female presented with a chief complaint of swelling on the left side of the face for the past one year. An incisional biopsy was performed and the histopathology was consistent of ameloblastic carcinoma. On the basis of the histopathology report, left hemimandibulectomy was done taking a safe margin of 2 cm and the defect was reconstructed using titanium reconstruction plate. Chemotherapy and radiotherapy were not advised. The patient is under regular follow-up. No recurrence nor metastases reported during the follow-up period. Conclusion: Early diagnosis and wide local excision on the primary site is the treatment of choice. The risk of malignant transformation should always be considered when a classic ameloblastoma is diagnosed and the prompt definitive management of an ameloblastoma is essential to eliminate this risk. Finally, meticulous, long-term follow-up is mandatory because recurrence and metastasis in the lung and regional lymph nodes have been reported.

Keywords: Ameloblastoma, Ameloblastic carcinoma, Carcinoma, Odontogenic tumor

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INTRODUCTION

Ameloblastic carcinoma is an extremely rare, aggressive, malignant neoplasm of the jaws with a poor prognosis. It belongs to the family of malignant epithelial odontogenic tumors [1]. It combines the histological features of ameloblastoma with features of cytological atypia regardless of whether it has metastasized [2, 3]. In contrast to ameloblastoma, ameloblastic carcinoma exhibits more aggressive clinical behavior, such as rapid growth, perforation of the cortex and painful swelling [3, 4]. It may metastasize to the regional lymph nodes or lung [3]. In rare cases, brain or multiple bone metastases have been reported [3]. Ameloblastic carcinoma occurs in a wide age range of 15–84 years, but the mean age is approximately 30 years [5, 6]. There is no apparent sex

predilection [5]. The most commonly involved area is the posterior portion of the mandible [5, 6]. Majority of the cases of ameloblastic carcinoma appear to arise de novo, and are termed primary ameloblastic carcinoma. Secondary ameloblastic carcinoma is defined as a tumor with malignant transformation within a pre-existing benign ameloblastoma, regardless of the presence of metastasis [4]. Secondary ameloblastic carcinoma is extremely rare. We report a case of secondary ameloblastic carcinoma of mandible in a 30-year-old Indian female.

CASE REPORT

A 30-year-old female presented to our department with a chief complaint of swelling on the left side of the face for the last one year (Figure 1). The swelling was associated with mild pain and the patient had difficulty in mastication and mouth opening. The patient gave a history of mild swelling, pain and mobility of teeth in left lower back region four years ago and she reported to the same centre. Her mandibular left 2nd and 3rd molars were extracted and an incisional biopsy was carried out. The biopsy report was suggestive of ameloblastoma and surgical excision of the lesion was advised. But the patient was not willing for the same and she did not report for the review. She noticed an increase in size of swelling and reported to our department after three years.

Clinical examination revealed a diffuse swelling over the left body-ramus region of the mandible, which extended antero-posteriorly from the left corner of mouth to the pre-auricular region. Superiorly, the extent was up to the infra-orbital region and inferiorly to the submandibular region. The overlying skin was normal in color and texture. On palpation, the swelling was mildly tender, smooth, and uniformly bony hard in consistency, with no local rise in temperature. The margins were ill defined. No paresthesia was associated with the swelling and no palpable regional lymph nodes noticed. Mouth opening was 32 mm and no deviation of mandible noted.

Intra-oral examination revealed a diffuse swelling of the left lower buccal vestibule extending from the canine region to the retromolar area. Posterior extension was not visualized clinically. A discontinuity of the overlying mucosa was noted with irregular rolled margins posteriorly (Figure 2). On intra-oral palpation, the swelling was firm to hard in consistency and mildly tender.

The orthopantomograph (OPG) showed a well-defined radiolucency involving the left body, angle and ramus of the mandible (Figure 3). The OPG revealed root resorption of left lower first molar tooth. Plain and contrast axial and coronal computed tomography (CT) scans showed a lobulated expansile lytic lesion involving body, ramus, and coronoid process of mandible measuring 6.3x8.3x10 cm in size, with thinning of the cortices (Figure 4). A chest radiograph ruled out the presence of any metastatic deposits. An incisional biopsy was done and

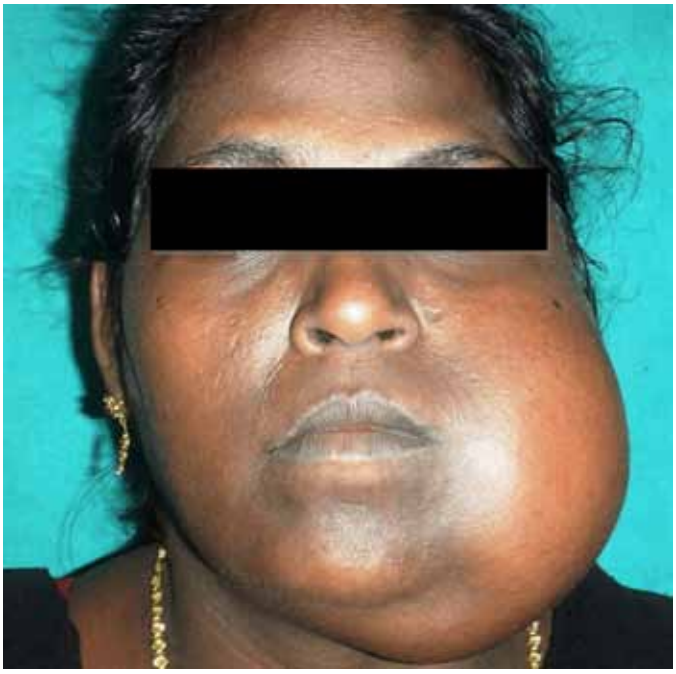

Figure 1: Preoperative extra oral view.

Figure 2: Preoperative intra oral view.

the tissue was sent for histopathologic examination. The histopathology was consistent of ameloblastic carcinoma (Figure 5). On the basis of the histopathology report, left hemimandibulectomy was done taking a safe margin of 2 cm and the defect was reconstructed using Titanium reconstruction plate. Chemotherapy and radiotherapy was not advised. The patient is under regular follow-up. No recurrence or metastases reported during the follow-up period.

DISCUSSION

Ameloblastic carcinoma is a rare odontogenic tumor that poses a real challenge to the clinician in diagnosis, treatment planning and prognosis [7]. It is a neoplasm demonstrating histological evidence of malignant

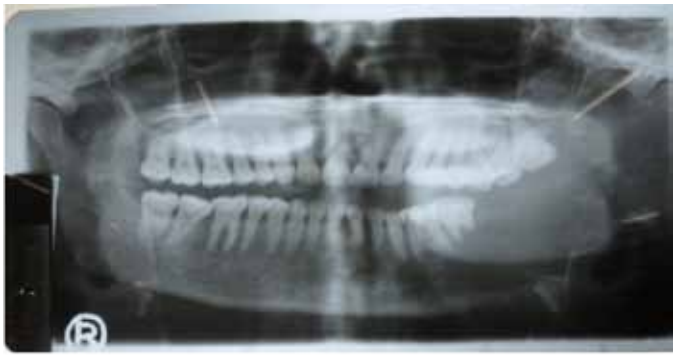

Figure 3: Orthopantomogram showing the lesion.

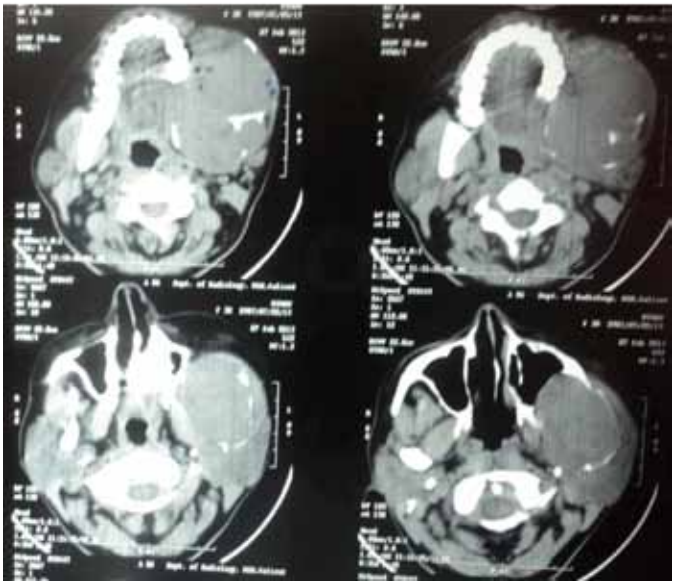

Figure 4: Computed tomography (CT) scan showing the lesion.

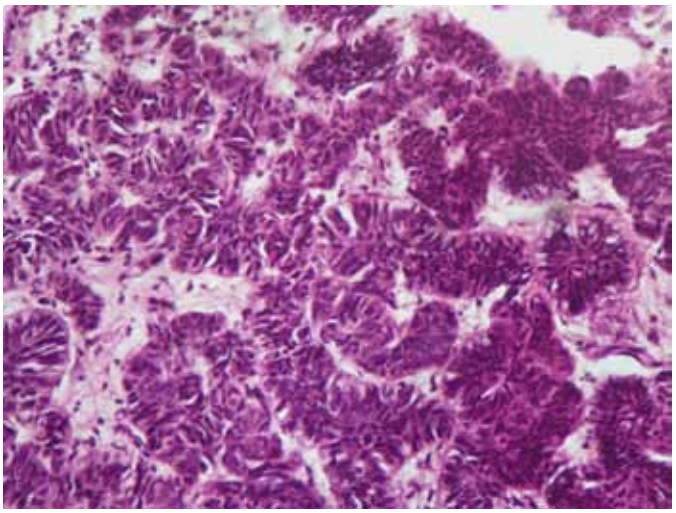

Figure 5: Histopathology of lesion.

transformation of the ameloblastoma-like epithelial component in the primary tumor whether or not it has metastasized [8]. WHO defined ameloblastic carcinoma as a rare odontogenic malignancy that combines the histological features of ameloblastoma with cytological atypia even in the absence of metastases [7]. In the updated histologic classification of the WHO in 2005 [7], ameloblastic carcinoma is classified as primary type and secondary type. Primary ameloblastic carcinomas are those arising de novo whereas the secondary type arise from a pre-existing ameloblastoma. Ameloblastic carcinoma, secondary type is extremely rare. The exact mechanism for the malignant transformation of ameloblastoma is currently unknown because of the limited number of cases [4].

Clinically, ameloblastic carcinoma is well known for its aggressiveness and extensive destruction of local structures [8]. The most common sign described has been swelling, although others include associated pain, rapid growth, trismus and dysphonia [5]. In most cases, radiographic findings show ill-defined radiolucency, however, focal radiopacity may be detected in radiolucent lesions [3]. Microscopically, it resembles the features of conventional ameloblastoma, except for the epithelium which shows various cytological features of malignancy [8]. Histologically, ameloblastic carcinomas have characters of both a benign ameloblastoma and carcinoma [9]. A palisade arrangement of epithelial cells with nuclei away from the basement membrane (reverse polarity) is a common feature of benign ameloblastomas [9]. The epithelial cells of ameloblastic carcinomas have features of hyperchromatism, a high mitotic rate, and a high nuclear-to-cytoplasmic ratio [9].

Recommended surgical treatment is jaw resection with 2–3 cm bony margin [5]. Ameloblastic carcinoma can recur locally 0.5–11 years after definitive therapy. Distant metastasis may occur as early as 4 months or as late as 12 years postoperatively and it is usually fatal [10]. Metastatic deposits of ameloblastic carcinoma are usually found in the lung, followed by bone, liver and brain [10]. Metastasis can occur even without any evidence of local recurrence [10]. The prognosis is much worse when distant metastasis occurs [9]. Cervical lymph node dissection should be considered when there is obvious lymphadenopathy [7]. The efficacy of adjuvant radiation or chemotherapy as a post-surgical treatment is not clear, because there is insufficient evidence that either provides a significant advantage [3]. However, radiotherapy and chemotherapy need to be considered in locally advanced cases and metastatic lesions not amenable to surgical resection [3]. The prognosis of ameloblastic carcinomas is poor [9]. The fatal factor includes the rapid growth of the primary tumor and distant metastasis [9]. Close periodic reassessment with a long-period of follow-up (at least 10 years) is mandatory [7]. Radical surgical resection, strict screening for and early detection of metastatic lesions, and periodic follow-up after surgery are needed to improve patient prognosis [3].

CONCLUSION

Ameloblastic carcinoma is a rare entity of jaw neoplasm that combines the histological features of an ameloblastoma with features of cytological atypia, with a

poor prognosis. This case report described a secondary type, ameloblastic carcinoma of mandible that originated from a pre-existing ameloblastoma which was left untreated.

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Soumithran C.S. – 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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Ikram Bin Ismail P.T. – 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
Ambadas – 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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Seeja P. – 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
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Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Rare sequelae of a testicular tumor: A 'burnt-out' tumor

Francesca Kum, Faisal Ghumman, Matin Sheriff

ABSTRACT

Introduction: Testicular tumors typically present as a palpable lump. Some tumors have been known to spontaneously regress, thus have been referred to as 'burnt-out' tumors or vanishing tumors. Case Report: A 28-year-old male was seen in urology clinic with clinical findings of a small atrophic left testis, with a prior history of a painful left testicular lump not responsive to antibiotics, which had spontaneously resolved. Ultrasound scan revealed an initial solid testicular lesion of 19x12x10 mm with internal vascularity, which upon repeat ultrasound scan two months later found a 12-mm coarse calcification and a hypoechoic region, but regression of the initial solid lesion. The diagnosis of a burnt-out tumor was made and he underwent urgent radical orchidectomy and prosthesis insertion. Computed tomoraphy (CT) staging scan showed significant paraortic lymphadenopathy, therefore the patient further underwent chemotherapy. Treatment has been successful and the patient is progressing well with no complications. Conclusion: The diagnosis of a burnt-out testicular tumor is an important differential to consider in patients with spontaneously regressing testicular lumps, abnormal findings on ultrasound scan and subsequent presentation of testicular atrophy of unknown cause. This diagnosis should also be considered in patients with secondary metastatic tumors of unknown primary.

Keywords: Burnt-out tumor, Cancer, Fast-track (2-week-wait) referral, Orchidectomy, Self-resolving tumor, Testicular cancer, Testicular tumor, Vanishing tumor

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INTRODUCTION

Testicular cancer primarily affects younger men typically aged 20–55 years. Presence of a testicular lump is an indication for referral to urology under the fast-track cancer pathway as an estimated 97% of testicular cancers present with a palpable mass [1]. Some tumors have exhibited the ability to spontaneously regress in size, thus presenting as an occult primary tumor. The diagnostic workup and management of these 'burnt-out' or vanishing tumors is identical to that of a typical testicular lump. This article presents a case of a burnt-out tumor, the important characteristic radiological findings and subsequent management.

CASE REPORT

A 28-year-old male presented via the fast-track cancer pathway to urology clinic with a preceding history of a

new onset left sided testicular pain and swelling. His general physician initially prescribed antibiotics for suspected epididymitis, but there was poor response over the ensuing weeks. An ultrasound scan was also requested and performed shortly after his first presentation (Figure 1). However, the report was not reviewed until the point when he was referred to urology two months later. This initial ultrasound scan reported a well-defined solid lesion in the lower pole of the left testis, measuring 19x12x10 mm with a calcific area and internal vascularity noted (Figure 1). The right testis was normal, as were both epididymis. A right sided varicocele was also noted.

At this point, a fast-track 2-week-wait referral to urology as *'swelling in body of testicle suspicious of cancer'* was made. When seen in urology outpatient clinic, the left testis was found to be notably smaller than his right testis with a small palpable solid component in the lower pole.

Subsequent testicular ultrasound (Figure 2) was performed some two months after the initial one and documented changes in the left testis from previous ultrasound scan with a few micro-calcification foci, coarse calcification of 12 mm length and an adjacent smaller low echoic area measuring 8x7 mm. The right testis was again normal. The report concluded a hypoechoic area in left testis adjacent to coarse calcification of which a burnt-out tumor should be excluded.

The patient was counseled and he decided not to take the risk of possible future tumor spread, therefore underwent left radical orchidectomy and insertion of prosthesis three days after being seen in urology clinic.

Macroscopically the left testis sample was 5x3x2.5 cm + epididymis + 5 cm cord attached. A poorly circumscribed greyish-white matter was noted within the testis, measuring 0.8 cm in diameter, which is a significantly smaller volume compared to the initial ultrasound scan, thus highly indicative of spontaneous reduction in tumor size without intervention. Upon microscopic examination the tumor type was a malignant teratoma with a cell composition of 60% embryonal carcinoma and 40% choriocarcinoma. The presence of background intratubular germ cell neoplasia, the precursor cell type to neoplasia, was noted in the surrounding testicular tissue. The tumor was confined to the testis, but vascular invasion was present. Histopathology of concluded a pT2 staging and β-hCG was elevated at 5172, thus correlating with that of a teratoma.

After orchidectomy, computed tomography scan of chest, abdomen, and pelvis with contrast was performed to assess for lymphatic involvement or metastatic spread (Figure 3). Computed tomography (CT) scan showed evidence of left para-aortic lymph node enlargement 22x21 mm with a necrotic centre. A few small volume lymph nodes were also noted adjacent to this enlarged necrotic node. There was no evidence of lung, liver or bony metastasis or lymphadenopathy and bony windows were unremarkable.

The patient was treated with adjuvant chemotherapy (BEP – bleomycin etoposide cisplatin) under the care of a tertiary Urology cancer centre. Shortly after his second cycle, he presented with a dry cough, coryzal symptoms, headache, tachycardia and pyrexia of 39.1°c. He was successfully treated with Tazocin and GCSF (granulocyte

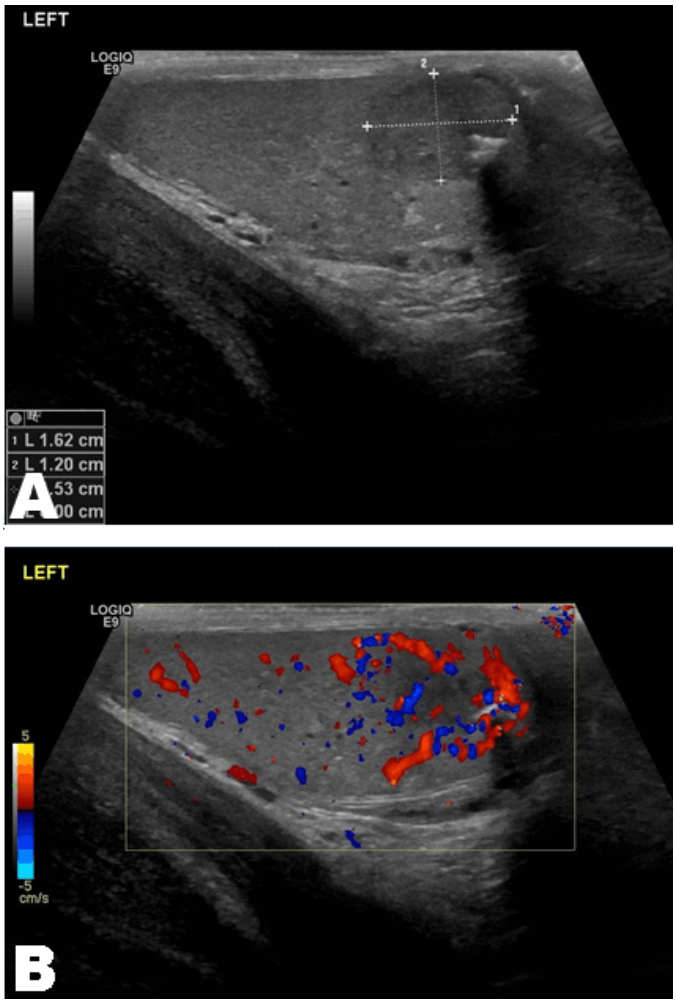

Figure 1: Ultrasound scan left testis, (A) Testicular well-defined solid lesion with calcific area, (B) Internal vascularity of lesion demonstrated.

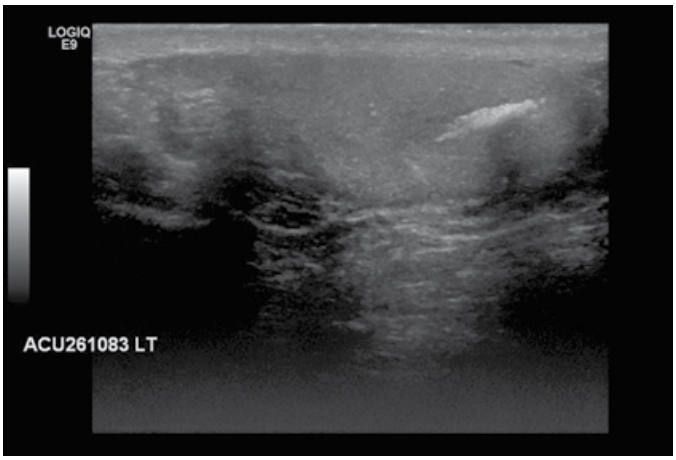

Figure 2: Ultrasound scan left testis, 12 mm coarse calcification and hypoechoic areas, (note absence of previous solid lesion).

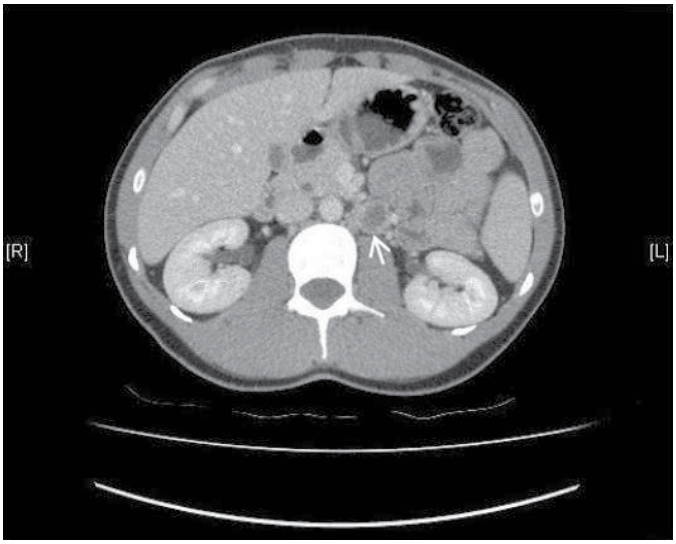

Figure 3: Computed tomography scan of chest, abdomen, pelvis with contrast showing an enlarged left para-aortic lymph node of 22x21 mm (arrow) with a necrotic centre and a few adjacent small volume lymph nodes.

colony stimulating factor) for neutropenic sepsis. The patient has subsequently completed chemotherapy and is progressing well.

DISCUSSION

This study presents a case of mixed cell type testicular teratoma with para-aortic spread, in which the primary neoplasm had spontaneously regressed within the two-month interval between ultrasound assessments. Testicular cancer usually presents with a palpable testicular lump in 97% of cases [1]. In contrast, clinical findings of burnt-out tumors on examination are often occult, such as an apparently normal sized testicle; or an atrophic testis, which develops insidiously; or an incidental finding of a retroperitoneal mass on imaging [2]. In such cases, it is essential that investigation and further imaging of the inguinoscrotal area should be performed to search for the tumor source.

Primary extragonadal germ cell tumors are rare and account for 5–10% of all germ cell tumors [3]. It is estimated that of the apparently primary extragonadal germ cell tumors, 10% are in fact due to underlying burnt-out testicular neoplasms [4]. In recent literature, these testicular tumors have also been described as 'vanishing' or 'self-resolving' tumors. However, although the initial testicular lesion may spontaneously regress, this does not negate the metastatic potential from the primary neoplasm to para-aortic and retroperitoneal lymph nodes and surrounding structures [2].

Existing literature reports specific ultrasound and histopathological findings which are indicative of previous presence of a testicular tumor [3, 5, 6]. Primary testicular tumors typically appear as a hypoechoic area on ultrasound. In the case of burnt-out tumors, some hypoechoic tumor tissue usually persists, in addition to histological findings of fibrous scar tissue with hemosiderin deposition, clustered macrocalcification or microcalcification, microlithiasis and increased hematoxyphilic bodies [5, 6]. Intratubular malignant germ cells and calcifications may also be present relating to Sertoli cells. Computed tomography scan remains the gold standard for assessing, staging lesions and metastases and guiding management plans after initial ultrasound reveals a testicular abnormality.

The underlying pathophysiology of tumor involution thus leading to the 'burning out' remains a topic of postulation [7–9]. It is thought that the initial aggressive nature of tumor growth eventually exceeds its vascular supply, thus resulting in subsequent death and spontaneous regression. Immunological theories are also considered such that cell-mediated immunity and recognition of tumor antigens may contribute to tumor destruction. Such autoimmune theories have previously been established in the response to in situ carcinoma of the testis.

The pathway of cancer referrals from primary to secondary specialist care is another current topic. It is widely known that delay in assessment adversely affects disease progression and patient outcome, therefore the National Health Services (NHS) two-week-wait referral system endeavors to ensure this [10]. The estimated incidence of testicular cancer in England is 7.3% with one-year survival rates of 98%. The criteria for activating this urgent referral pathway is the presence of a swelling or mass in the testicle. The National Institute for Health and Care Excellence (NICE) further recommends that the general physician should consider requesting an ultrasound scan if the lump cannot be distinguished from the testicular body itself. As with many other cases, imaging and further investigations are required, but these should not delay referral to specialist services [11]. Furthermore, this case illustrates the need for efficient review of investigation results by general physician and prompt of these suspected cancer cases to ensure better patient outcome.

CONCLUSION

In summary, this case report highlights the importance of early referral and subsequent management of a testicular cancer case. Early ultrasound scan is essential in the investigation of any testicular lump. Burnt-out testicular tumors are an important differential to consider in patients with spontaneously regressing testicular lumps, and subsequent presentation of testicular atrophy of unknown cause. This diagnosis should also be considered in patients with secondary metastatic tumors of unknown primary. Prompt surgical management of orchidectomy is usually required and subsequent computed tomography scan for disease

staging and grading.

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Author Contributions

Francesca Kum – 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
Faisal Ghumman – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Matin Sheriff – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

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CASE REPORT OPEN ACCESS

Strongyloidiasis after corticosteroid therapy: A case report

Jangala Mohan Sidhartha, Barabari Man Mohan, Maddinani Penchalaiah, Lomati Venkata Pavan Kumar Reddy

ABSTRACT

Introduction: Strongyloidiasis is a parasitic infection which can be divided into asymptomatic carriage, intestinal disease, hyperinfection with or without dissemination and it is also capable of a free living cycle. It is caused by *Strongyloides stercoralis* in humans. Generally in a healthy host, it is an asymptomatic infection but it causes hyper infection involving multiple organs in immunocompromised patients. Case Report: We report an intestinal strongyloidiasis hyper-infection in a chronic obstructive pulmonary disease (COPD) patient of 66 years old. The patient had a history of receiving corticosteroid therapy frequently for acute exacerbations of COPD symptoms, during one of such episode the patient presented to our center, after receiving corticosteroid therapy the patient recovered from the episode but developed diarrhea, investigations done for diarrhea evaluation revealed larvae of *Strongyloides* in stool sample. Anthelmintic therapy, albendazole for three days followed by ivermectin for five days were given, the frequency of stools decreased to a great extent even though not completely cured. Conclusion: Strongyloidiasis a helminthic infection common in southeast asia and other sub Saharan countries, the longevity of this infection is for decades due to autoinfection so, regular screening for this parasite in Immunocompromised patients should be done.

Keywords: Asymptomatic infection, Immunodeficiency, Nematodes, Strongyloidiasis

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INTRODUCTION

Strongyloidiasis is a parasitic infection caused by *Strongyloides stercoralis*. It is dominant in the temperate, tropical and subtropical areas [1]. *S. stercoralis* is distinguished by its ability—unique among helminths to replicate in the human host. This capacity permits ongoing cycles of autoinfection as infective larvae are internally produced. Strongyloidiasis can thus persist for decades without further exposure of the host to exogenous infective larvae. In immunocompromised hosts, large numbers of invasive *Strongyloides* larvae can disseminate widely and can be fatal [2]. Uncontrolled hyper infection manifestations are seen in many organs where the host-parasite relationships can be altered by certain predisposing factors such as corticosteroid therapy, malnutrition and anticancer drugs [3]. However, the intestinal involvement is very rare [4]. Many individuals infected with this parasite are asymptomatic. However,

in patients with defects in cell-mediated immunity as a result of malnutrition, immunosuppressive drugs, an unrecognized asymptomatic infection may convert into severe infection.

CASE REPORT

A 66-year-old male, resident of Kamalapuram, a small town in Kadapa region of the Andhra Pradesh (India), presented to General Hospital on 11 April 2014. His chief complaints were progressive breathlessness from last two weeks, pedal edema, fever, headache, cough with sputum, abdominal pain prior to admission. He was an ex-smoker and alcoholic.

On examining the patient, he was a lean built man with significant dehydration. His temperature was 100°F, pulse rate 90/min, blood pressure 150/90 mmHg. He maintained a saturation of 93 on ambient air and respiratory rate 24/min. There were few exacerbation in left lower zone. He had no icterus and there was no significant lymph adenopathy. Epigastrium tenderness was observed in the patient. The liver, spleen and kidneys were not palpable.

Patient's Medication History and Treatment

Patient's past history was notable for chronic obstructive pulmonary disease (COPD) and old pulmonary tuberculosis.

Investigations

Patient's reports showed HIV non-reactive and HBsAg was negative. Other laboratory findings were white blood cell count 11700 cells/mm^3 with a normal differential count and 3% eosinophils. Absolute eosinophil count 340 cells/mm^3 and erythrocyte sedimentation rate was 15 mm/h, blood urea 74 mg/dL and 1.9 mg/dL was serum creatinine. He had a grade one renal parenchymal changes and normal sized kidneys on ultrasound. Computed tomography (CT) scan of chest showed thick and thin walled cystic bronchiectatic changes in apical segments of right upper lobe (Figure 1A), small modular infiltrates were seen in left lower lobe and in lateral basal segment of right lower lobe (Figure 1B). Stool microscopy showed heavy loads of "strongyloidiasis" (Figure 2). Sputum culture grew *Pseudomonas* species and *Klebsiella oxytoca*, sensitive to gentamycin; however, it was thought to represent colonization. Sputum smear for acid-fast bacilli (AFB) was negative.

Treatment

A diagnosis of COPD with acute exacerbation was observed, for which he was given intravenous ceftriaxone 1 g twice daily, azithromycin 500 mg once daily, salbutamol and ipratropium bromide nebulizers. He was not responded and on 30 April 2014 patient received

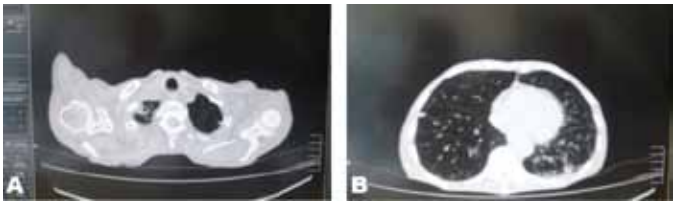

Figure 1: Computed tomography of chest showing (A) Thick and thin walled cystic bronchiectatic changes in apical segments of right upper lobe, (B) Small modular infiltrates are seen in left lower lobe and in lateral basal segment of right lower lobe.

Figure 2: Stool microscopy showing heavy loads of strongyloidiasis.

systemic corticosteroid (hydro cortisone 100 mg IV TID). After three days of initiation of corticosteroid therapy, the patient developed loose stools (10 times per day). After obtaining stool examination and conformed to "Strongyloidiasis" he was treated with albendazole 400 mg daily for three days, patient was not responded then ivermectin 12 mg daily and metronidazole 400 mg twice in a day for next five days. Decrease loose stool frequency four times per day and improved his vitals. However, he discharged against medical advice.

DISCUSSION

Strongyloidiasis is intestinal parasitic infections caused by two species in humans; they are *Strongyloides stercoralis (S. stercoralis)* and another *Strongyloides fülleborni* (*S. fülleborni)*. The symptoms related to strongyloidiasis depends on nematode's systemic passage, its local cutaneous involvement, or both. Most of the patients who develop hyperinfection syndrome are receiving corticosteroids often for COPD. Pulmonary strongyloidiasis may mimic COPD exacerbation [5]. Hyperinfection may develop as early as four days after the onset of corticosteroid therapy and as late as several years up to 20 years [6].

Detection of larvae in stool or sputum is the absolute finding of strongyloidiasis. Sometimes screening is inadequate in performed stool studies are alone. However, approximately 50% sensitivity was noted in a single stool exam for making the diagnosis [5]. Albendazole, thiabendazole, mebendazole and ivermectin are the effective drug therapy for *S. stercoralis* infection. In recent, more studies presenting ivermectin as the drug of choice in the treatment of strongyloidiasis. Monitoring response of the treatment is problematic in the stool specimen with recognition of *Strongyloides* larvae. Recent study by Edmilson Bastos de Moura et al. proved

that therapy with subcutaneous ivermectin was successful in immunocompromised disseminated strongyloidiasis patient [7].

The presentation in our patient was initially with severe pulmonary disease like in various aspects to that called by others. These clinical manifestations are non-specific and there is a no improvement after three weeks of therapy. Later he received a short course of hydrocortisone therapy (three days); he developed the hyperinfection syndrome. Similarly, a study showed that a patient diagnosed with *S. stercoralis* after receiving a course of corticosteroid therapy (13 days) [8]. *S. stercoralis* inactive in the host's intestine, it can stay nearly 30 years; it may apparent when the host immunity decreases either immunosuppressive drugs and/or disease [9]. Our case suggests that there is a need to be careful in such patients to allow for early diagnosis and institution of proper treatment in order to avoid such unfortunate effect. It may not be difficult to screen and serology before initiating steroid therapy to prevent the progress of Strongyloidiasis hyperinfection in immunocompromised patients from endemic areas. Screening may not be commended patients before starting short courses of corticosteroids for COPD. It may consider in severe cases of COPD using frequent steroids courses and they are coming from endemic areas.

Screening is necessary for preventing the strongyloidiasis hyperinfection in severe cases of COPD using frequent steroids courses and they are coming from endemic areas. To promote improving the living conditions of rural population as providing a safe drinking water supply and good sanitary measures, and instructing about the disease in high-risk populations. Early diagnosis of any strongyloides infection may helpful for outcomes of the therapy; otherwise increased mortality rate due to fatal complications.

The therapy with subcutaneous ivermectin was successful in immunocompromised disseminated strongyloidiasis patient.

CONCLUSION

Strongyloides hyperinfection requires prompt diagnosis and initiation of antihelmintic therapy. Even with appropriate therapy, the mortality in disseminated Strongyloidiasis is much higher. In light of this, appropriate screening of high risk individual should be carried out prior to the initiation of therapy that is associated with immunosuppression.

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CASE REPORT OPEN ACCESS

Intracystic papillary carcinoma of the breast with invasion accompanying lymph node metastasis

Takaaki Fujii, Reina Yajima, Hiroki Morita, Satoru Yamaguchi, Soichi Tsutsumi, Takayuki Asao, Hiroyuki Kuwano

ABSTRACT

Introduction: Intracystic papillary carcinoma (IPC) is a rare malignant tumor of the breast. Generally, IPC shows no invasive growth outside of the cyst, however, it is often associated with invasive carcinoma. Thus, an accurate preoperative diagnosis of invasion or spread plays a crucial role in the management of patients with IPC. Case Report: A 78-year-old Japanese female presented with a large right breast mass. Magnetic resonance imaging (MRI) scan showed an intracystic mass with a papillary lesion, indicating invasion. Breast conservative surgery with sentinel lymph node (SLN) biopsy was performed. Histological evaluation confirmed IPC with invasion to the outside of the cyst wall, as detected by preoperative MRI, and SLN metastasis was also detected. Conclusion: It should be noted that IPC is frequently accompanied by invasion. MRI findings may help in making a definitive diagnosis of tumor invasiveness. The SLN biopsy should be considered in cases of IPC with invasion.

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INTRODUCTION

Intracystic papillary carcinoma (IPC) is a rare malignant tumor of the breast [1–5]. Generally, IPC shows no invasive growth outside of the cyst [6–8]. There is no consensus on the management strategy of IPC with invasion, however, the prognosis for IPC is usually excellent if completely excised [1, 9, 10]. Thus, an accurate diagnosis of invasion or spread may play a crucial role in the management of patients with IPC. We report here on a rare case of IPC with invasion that was detected prior to surgery by magnetic resonance imaging (MRI) scan, accompanying axillary lymph node metastasis.

CASE REPORT

A 78-year-old Japanese postmenopausal woman presented with a large right breast mass four months prior to admission (Figure 1A). Physical examination revealed one 5.0x5.0 cm palpable cystic mass that was elastically firm in the lower external quadrant of the right breast. There was no abnormal nipple discharge. Mammography revealed a well-defined large dense mass (Figure 1B). Sonography revealed a well-demarcated intracystic tumor with papillary growth in the right breast (Figure 2A), and MRI scan showed an intracystic mass with a papillary lesion, indicating invasion (Figure 2B). There was no evidence of axillary

lymphadenopathy. A biopsy had been performed at the previous hospital and revealed an invasive ductal carcinoma. Breast conservative surgery with sentinel lymph node (SLN) biopsy was performed, and a white intracystic papillary tumor was removed (Figure 2C). During surgery, an axillary SLN was detected with blue dye and a hot (radioactive) tracer. Briefly, twelve hours prior to surgery, the radioisotope with 99mTc-phytate colloid was injected subcutaneously in the periareolar region. Intradermal injection of a blue dye, indigo carmine, in the periareolar region was also performed immediately prior to surgery. Any lymph nodes with blue dye and radioactivity were regarded as SLN [11–13]. The intraoperative diagnosis of SLN was positive for metastasis, and additional conventional axillary lymph node dissection (ALND) was therefore performed. At final histology, none of the non-SLN was metastatic. Histological evaluation revealed a lesion containing papillary structure in the cystic space and a solid tubular carcinoma with invasion to the outside of the cyst wall that had been detected by preoperative MRI. These findings are compatible with intracystic papillary carcinoma associated with invasive ductal carcinoma (Figure 3).

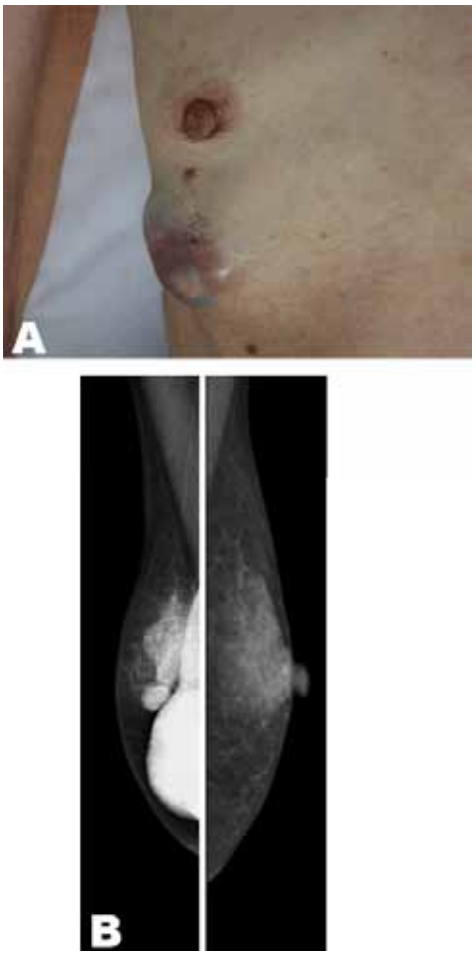

Figure 1: (A) A 5.0x5.0 cm palpable cystic mass was elastically firm in the lower external quadrant of the right breast, and (B) Mammography revealed a well-defined large dense mass.

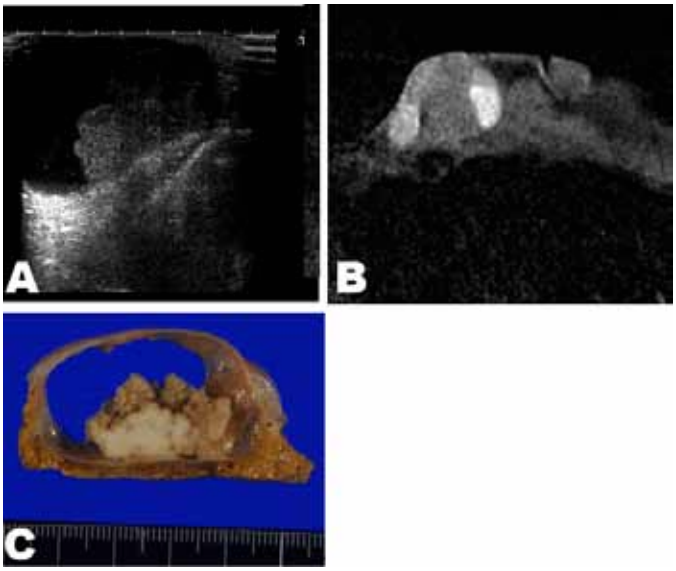

Figure 2: (A) Sonography revealed a well-demarcated intracystic tumor with papillary growth in the right breast, (B) Magnetic resonance imaging scan showed an intracystic mass with a papillary lesion, indicating invasion, and (C) Breast conservative surgery was performed. A Photograph of a gross specimen showed a papillary tumor within the cystic space.

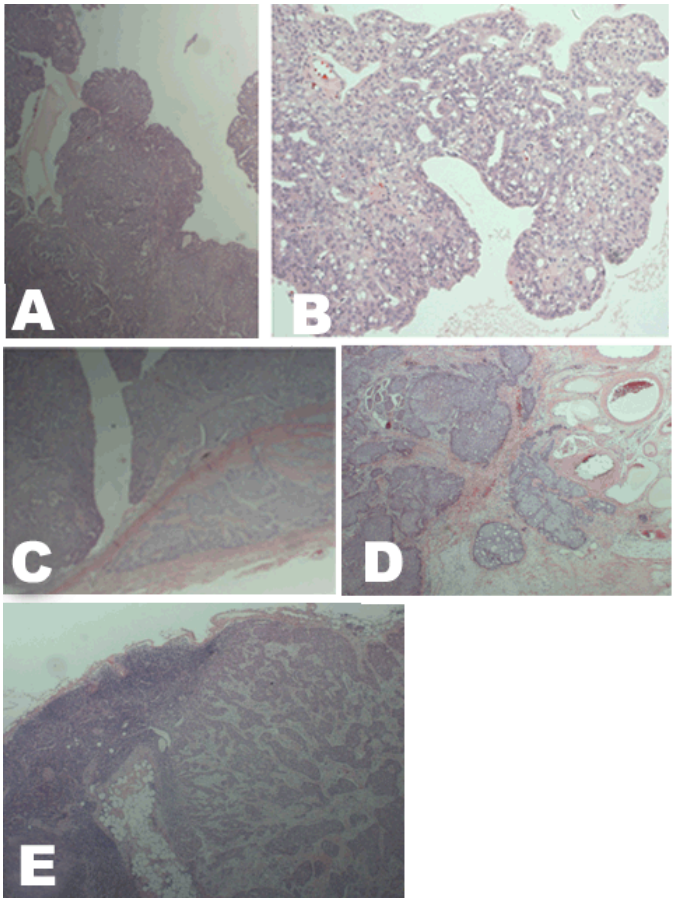

Figure 3: (A, B) Histological evaluation revealed a lesion containing papillary structure in the cystic space (H&E stain, x100, x200 respectively), (C, D) A solid-tubular carcinoma with invasion to the outside of the cyst wall (H&E stain, x100, x200 respectively). These findings are compatible with intracystic papillary carcinoma associated with invasive ductal carcinoma, and (E) Sentinel lymph node was positive for metastasis (H&E stain, x200).

Immunohistochemical staining showed estrogen and progesterone receptor were positive and the patient's HER2 score was 0. The patient has not received adjuvant therapy or radiotherapy because of severe dementia. The patient has remained alive for 14 months without locoregional or systemic recurrence of the tumor.

DISCUSSION

We report herein a rare case of IPC of the breast with invasion accompanying SLN metastasis. The IPC is an uncommon tumor that predominantly affects elderly women and accounts for only 1–2% of all breast cancers [1–4, 8–10]. This can be divided into a pure form (IPC alone), IPC with associated DCIS and IPC with associated invasive ductal carcinoma. The IPC is relatively frequently associated with invasive carcinoma or DCIS beyond the tumor [1, 2, 5, 10, 14]. The prognosis for this tumor is excellent regardless of whether it is noninvasive or invasive [3, 4]. Thus, an accurate preoperative diagnosis of invasion or spread may play a crucial role in the management of patients with IPC. Magnetic resonance imaging scan may be useful not only for reaching a differential diagnosis, but also for evaluating tumor invasiveness. In the present case, MRI was useful in diagnosing the invasiveness of the IPC and in decision-making for the surgical procedure. The SLN biopsy or ALND are often performed to assess the axillary lymph nodes [14]. The role of SLN biopsy has not yet been fully evaluated in IPC, but it may be an excellent alternative in patients with IPC with invasion. Among patients with IPC with invasive carcinoma, 30% of patients who underwent axillary dissection had histologically positive nodes [3, 10]. This case was also positive for SLN metastasis. Therefore, SLN biopsy is recommended for invasive cases.

CONCLUSION

In conclusion, we reported here on a rare case of intracystic papillary carcinoma (IPC) with invasion accompanying sentinel lymph node (SLN) metastasis. It should be noted that IPC is frequently accompanied by invasion. The SLN biopsy should be considered in cases of IPC with invasion. Magnetic resonance imaging scan may help in making a definitive diagnosis of tumor invasiveness.

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CASE REPORT OPEN ACCESS

Missed chronic impaction of dental crowns in the hypopharynx of a neurologically devastated child

Michael Zaki, Soroush Zaghi, Jonathan Ghiam, Alisha West

ABSTRACT

Introduction: Ingested foreign objects that impacted in the upper aero-gastrointestinal tract are fairly common and potentially serious problems. Dental objects are the most common ingested foreign bodies. Longstanding impacted foreign objects are complicated by failure to thrive or recurrent aspiration pneumonia and other serious complications such as viscus perforation, neck infections, hemorrhage, or esophago-aortic and tracheoesophageal fistulas. Case Report: An eight-year-old neurologically devastated boy with tongue biting and bruxism was brought into the emergency department for evaluation of two foreign bodies found incidentally on modified barium swallow study. Plain radiography of the neck soft tissue showed two radiopaque densities within the hypopharynx. The foreign objects were removed in the operating room under anesthesia using direct laryngoscopy and forceps and gross pathology was consistent with two golden dental crowns. The patient appeared to be at his baseline with respect to swallowing, breathing, and pain despite the fact that the crowns had been impacted for at least eight months based upon review of prior available radiological images. Conclusion: We hypothesize that our patient's continuous bruxism and ill-fitting crowns led to dislodgement and impaction of the crowns. His neurologic impairment and lack of gag reflex may have allowed the dental crown impaction to remain asymptomatic. Impaction of dental prosthetics in the upper aero-gastrointestinal tract may be masked and under-recognized in children, psychiatric patients, and individuals with neurologic impairment. Careful and regular evaluation of dental prosthetics in this population should be undertaken to prevent complications secondary to ingestion.

Keywords: Accidental ingestion, Aero-gastrointestinal tract, Dental prosthesis, Foreign body impaction, Hypopharynx, Neurological impairment, Psychiatric patient

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INTRODUCTION

Ingested foreign objects that are impacted in the upper aero-gastrointestinal tract are fairly common and

potentially serious problems in the pediatric population. In the US, 1500 deaths per year are attributed to foreign object ingestion [1]. Dental objects are among the most common ingested foreign bodies along with bones, disk batteries, and coins. The incidence of ingested and impacted dental prostheses is 0.7% [2]. Commonly, ingested foreign objects of dental origin can include: tooth picks; endodontic instruments such as files and burs, impression and denture lining materials, dental appliances such as inlays, onlays, crowns, and rubber dam clamps, fixed and removable prosthesis such as orthodontic retainers, bands, and wires. Risk factors for dental object ingestion include alcoholism, psychiatric disorders, incarceration, senility, dental trauma, developmentally delayes, individuals with neurological disorders including Parkinson's, seizure, dementia, and stroke, individuals with loose or ill-fitting dental appliances, individuals with single tooth cast, and prefabricated restorations, and age less than 15 years [3].

As much as 40% of foreign object ingestions in children are asymptomatic and resolve spontaneously. However, longstanding impacted foreign objects are complicated by failure to thrive or recurrent aspiration pneumonia. In less than one percent of foreign object ingestions, patients may suffer from serious complications such as viscus perforation, neck infections, esophageal obstruction with aspiration risk, hemorrhage, or esophago-aortic and tracheoesophageal fistulas, which in turn leads to the diagnosis when it is already too late [1].

CASE REPORT

An eight-year-old neurologically devastated boy residing in a specialized nursing home for medically frail children was brought into the emergency department for evaluation of two foreign bodies found incidentally on modified barium swallow study (MBSS). The patient has a history of posterior fossa arteriovenous malformation (AVM) rupture causing large left sided cerebellar hemorrhage that left the patient gastrostomy tube, tracheostomy, and ventilator dependent. Since the AVM rupture, the patient experienced continuous bruxism and tongue biting that required him to wear a mouth guard. The swallow study was performed for evaluation of swallowing prior to advancing his oral intake. Oddly, the patient had been completely asymptomatic with no changes in his ventilator requirements despite the impaction of these foreign bodies. He had been tolerating his gastrostomy tube feeds and did not seem like he was in pain. X-ray of the neck soft tissue lateral view (Figure 1) and anteroposterior view (Figure 2) showed two radiopaque densities ~5–10 mm each, which have the appearance of teeth within the hypopharynx/vallecula at the level of C3 and C4. Consultation to otorhinolaryngology team was initiated. Patient was scheduled for removal in the operating room under general endotracheal anesthesia. The foreign objects were removed under direct visualization with direct laryngoscopy and forceps. The remainder of the laryngoscopic examination was normal. Pathology was consistent with two irregularly shaped shiny golden metallic dental crowns measuring 1.0x0.6x0.6 cm and 1.1x0.8x0.6 cm. A photograph of the dental crowns was taken (Figure 3). It is unclear when our patient lost both his crowns and how long they were in his hypopharynx. However, on chart review, a lateral X-ray of the skull (Figure 4) performed eight months prior to discovery of the foreign bodies on the MBSS showed the two radiographic densities in the same place. This indicates that these dental crowns may have been impacted in the patient's hypopharynx for over eight months. The identification of these two radiographic densities was missed on the initial skull X-ray read. Additionally, a dental consult and examination was performed on the same day as the skull X-ray, which failed to detect the missing dental crowns but showed that his mouth-guard was loose and that teeth 7–10 and 23–26 were mobile/loose.

DISCUSSION

We hypothesize that our patient's continuous bruxism and ill-fitting crowns led to dislodgement and impaction of the crowns. Additionally, an ill-fitting crown can accumulate stress concentrations that may reduce the strength and long-term fixation of the restoration into the enamel [4]. Therefore, it is important to have the restoration evaluated periodically to ensure proper seal and fitment. Both complications associated with long-standing foreign object impaction (failure to thrive and recurrent aspiration pneumonia) were not possible to detect in our patient due to his gastrostomy tube and

Figure 1: An eight-year-old boy with two radiopaque densities incidentally found on modified barium swallow study. Findings: Two radiopaque objects (red arrow) at the level of the hypopharynx near the cervical esophagus. Technique: Lateral neck soft tissue plain radiograph.

tracheostomy dependence. Fortunately, our patient did not suffer from any of these complications or any of the other serious ones. However, his neurologic impairment and lack of gag reflex may have allowed the dental crown impaction to remain asymptomatic. Interestingly, our patient's dental crowns were impacted in his hypopharynx, which is a relatively uncommon site for impaction of dental prosthesis without causing any symptoms. According to a case series of dental prosthesis ingestion, most foreign bodies are lodged in the cervical

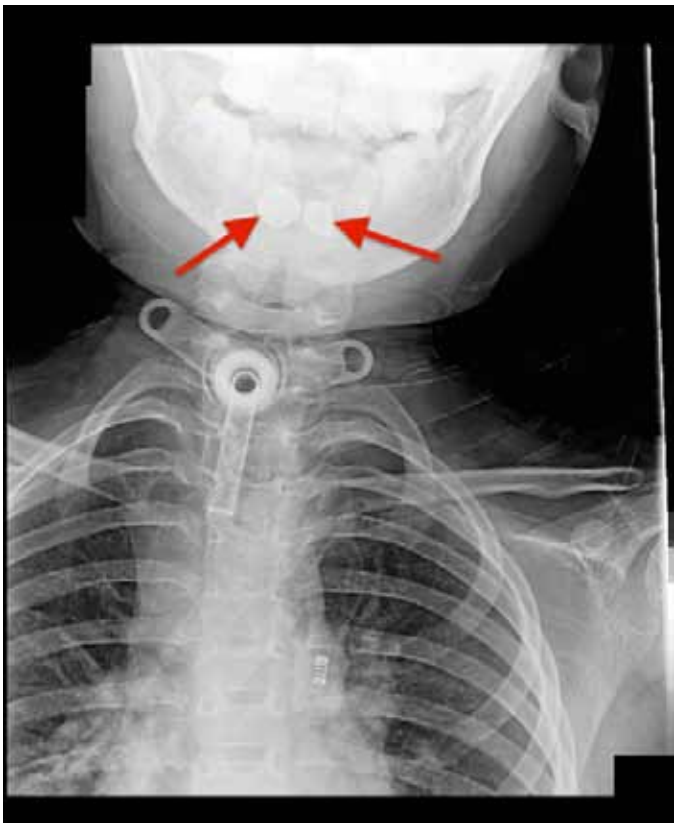

Figure 2: An eight-year-old boy with two radiopaque densities incidentally found on modified barium swallow study. Findings: Two radiopaque objects (red arrows) at the level of the hypopharynx near the cervical esophagus. Technique: Anteriorposterior neck soft tissue plain radiograph.

Figure 3: Two irregularly shaped shiny metallic crowns measuring 1.0x0.6x0.6 cm and 1.1x0.8x0.6 cm were removed from the hypopharynx.

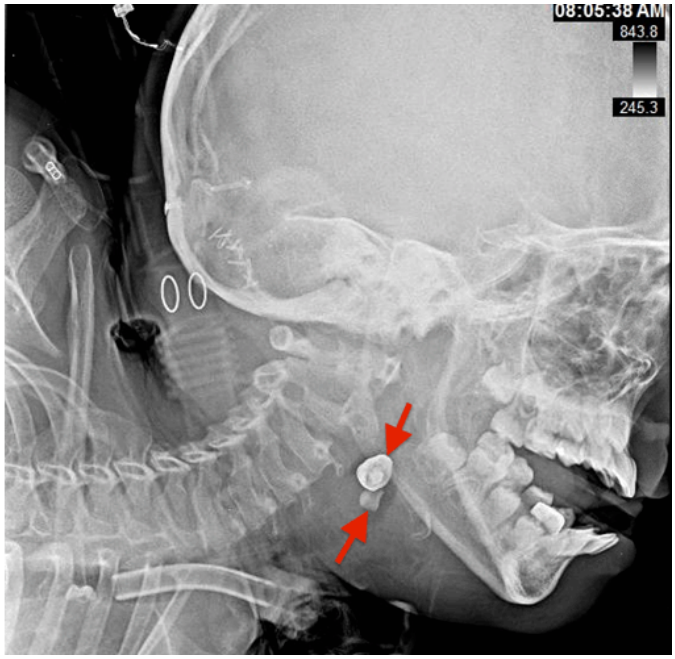

Figure 4: An eight-year-old boy with two radiopaque densities incidentally found on modified barium swallow study. Findings: Two radiopaque objects (red arrows) at the level of the hypopharynx near the cervical esophagus. Technique: Lateral skull plain radiograph performed eight months prior to discovery of foreign bodies on modified barium swallow study.

esophagus, mid and lower esophagus or rest of the gastrointestinal tract, with the hypopharynx being the least likely location [5].

If foreign body ingestion is suspected, plain radiography should be the initial method of investigation. Plain radiographs can confirm the size, location, and shape of ingested foreign objects and help rule out aspiration. They are also helpful in excluding some complications including pneumomediastinum (in case of viscus perforation) or crepitus (if there is an infection). Since, the skull X-ray was ordered to evaluate for a left-sided head lump, it is possible that most of the reading radiologist's attention was focused on ruling out a fracture. For this reason, radiologists should keep an open mind about the differential diagnosis and remain open to the possibility of unusual findings, as they are least likely to be biased by patient's clinical presentation. We also suggest that performing serial plain radiographic evaluation of neck, chest and abdomen when missing dental prosthetics is suspected in individuals at high risk for asymptomatic accidental ingestion (such as our patient). Initial plain radiographs will confirm if accidental ingestion of radiopaque objects has occurred. Subsequent serial plain radiographs will ensure timely passage of these foreign objects or dictate appropriate further management to prevent further complications associated with foreign object impaction in the aero-gastrointestinal tract.

Fortunately, the dental crowns were also made of a radiopaque metal that was easily detected on subsequent

neck soft tissue plain radiographs. It should be noted that other dental objects may be made out of radiolucent materials and may require computed tomography scan to localize them based on the soft tissue changes cause by the trauma from the foreign body [6].

The most relevant differential diagnoses for foreign body impacted in the aero-gastrointestinal tract are long objects, disk batteries, drug packets, and sharp pointed objects. The management of each of these is different and some require special considerations. It is important to categorize the type of object impacted radiographically or endoscopically (if radiography is inconclusive), to plan management accordingly. Disk batteries require urgent intervention because voltage burns and direct corrosive effects can occur as early as four hours after ingestion [1]. If the batteries are past the duodenum, serial radiographs should be performed every 3 to 4 days to ensure passage. For long and sharp or pointy objects, the risk of perforation is higher and early intervention with the use of overtubes to protect the airway and esophageal mucosa from lacerations is warranted. Since rupture and leakage of drug packet contents may be lethal, endoscopic removal should not be attempted and surgical intervention is preferred only if passage of packets fail or if there are signs of small bowel obstruction [7].

The first and most important priority in approaching a patient with foreign body ingestion is stabilization of the airway and breathing. If missing dental prosthesis is suspected, eliciting a specific history of dental work and thorough examination of teeth followed by serial plain radiographic evaluation of neck, chest, abdomen will confirm occurrence of accidental ingestion of radiopaque objects and will ensure timely passage of these foreign objects or dictate appropriate further management to prevent longstanding foreign body impaction in the aero-gastrointestinal tract and associated complications. Impacted objects can be managed conservatively if they are asymptomatic and evidence of progress through the gastrointestinal tract is present. Objects impacted in the esophagus are unlikely to resolve without intervention after 24 hours and may require endoscopy in 10–20% of the cases or surgery in 1% of cases [1]. According to the guidelines set by the ASGE for management of ingested foreign bodies, the mainstay of treatment for accidental ingestions is flexible endoscopy accompanied by retrieval devices. The guideline also recommends an otorhinolaryngology consultation for foreign bodies at or above the level of the cricopharyngeus [6].

CONCLUSION

Longstanding impaction of dental prosthesis in hypopharynx can present without a positive history in children with neurological impairment and should be included in the differential when reading c-spine/ neck X-rays. Regular thorough examination of the oral cavity and dentition in high risk population and careful attention to their history of dental work would detect missing teeth and dental appliances allowing for a more timely diagnosis of accidental dental prosthesis ingestion/impaction and prevention of further associated complications.

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Author Contributions

Michael Zaki – 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
Soroush Zaghi – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Jonathan Ghiam – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Alisha West – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Systemic to pulmonary arteriovenous fistula in a patient with tuberculosis sequels: A sign of inflammatory neoangiogenesis

Maria Fernanda Saavedra, Maria Juliana Valenzuela, Monica Ocampo, Carlos Garavito, José Federico Saaibi, Mauricio Orozco-Levi

ABSTRACT

Introduction: The study of the causes of hemoptysis entails multiple diagnostic choices and tests. However, in 30% of the cases, a cause is not clearly identified (cryptogenic hemoptysis). Even though pulmonary arteriovenous malformations are uncommon, they must be taken into account as possible causes of hemoptysis. Particularly, acquired systemic-to-pulmonary vascular fistulas are atypical and represent both diagnostic and therapeutic challenges. Case Report: We describe a case of systemic to pulmonary arteriovenous malformation between the internal mammary and right subclavian arteries to the upper lobe pulmonary veins, in a 47-year-old male with a past history of pulmonary tuberculosis who presented with hemoptysis. The patient had received anti-tuberculous treatment in another institution after he was diagnosed with pulmonary tuberculosis made conclusive by positive smear tests and chest radiologic examination. He was subsequently treated on a second occasion in spite of the negative smears following the first treatment because of the persistent hemoptysis. Despite recurrent hemoptysis, the patient was brought to our institution six years after the beginning of the hemoptysis. Pulmonary arteriography was done revealing a right internal mammary and subclavian arteriovenous malformation in communication with the vessels of the right upper pulmonary lobe, which was successfully treated with endovascular embolization. Conclusion: We believe systemic to pulmonary fistulas can represent a sign of inflammatory pulmonary and extrapulmonary neoangiogenic process.

Keywords: Acquired arteriovenous fistula, Hemoptysis, Inflammatory neoangiogenesis, Internal mammary artery, Tuberculosis

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INTRODUCTION

The expectoration of blood, or hemoptysis can vary from blood streaking of sputum to the presence of pure blood without associated sputum. It is not considered a disease but a sign of it. Otherwise massive hemoptysis is a life-threatening manifestation of a disease comprising the volume of 100–600 mL of blood in a 24-hour period [1]. This symptom remains as a daring field of multiple probable causal diseases. We report this case because of its interesting background in physiopathology that includes the neoangiogenic process to consider as a differential diagnosis.

CASE REPORT

A 47-year-old male presented to our institution with massive hemoptysis in which approximately 150 cm^3 of blood was expectorated at least twice a week in a period of 18 months. He had been treated in another institution for pulmonary tuberculosis six years ago. This was after the diagnosis with serial spontaneously sputum stains positive for acid-fast bacilli. He had to repeat this treatment in spite of the following negative smears because of the persistent hemoptysis, which had also increased in volume and frequency. Despite recurrent hemoptysis, the patient was brought to our institution four years after he finished treatment. On examination, he had low weight, and we found a continuous murmur at the level of the right superior thorax. We did not find vascular lesions in his mouth or on his skin. The rest of the physical examination was normal. The laboratory data including complete blood count and blood chemistries were also normal. Pulse oximetry at rest showed normal values. The patient denied any past history of liver disease. Computed tomography (CT) scan of chest revealed signs of volume loss and bronchiectasis involving the right superior lobe. We found an unusual image of vascular characteristic at the level of the right internal mammary artery. There were no caverns or suspicious images of pulmonary arteriovenous malformations (Figure 1). We decided to undertake an angiographic evaluation in which we observed a high flow systemic to pulmonary arteriovenous fistula from the internal mammary and right subclavian arteries, to the right upper lobar veins. We proceeded to perform embolization using two interlock coils and two amplatzer vascular plugs until the internal mammary artery was completely cut-off (Figure 2). The spirometric data showed a FEV1=2820 mL (82% ref), FVC=3050 mL (69% ref), and %FEV1/FVC=92. The patient was discharged after two days of hospital stay with resolution of the hemoptysis. The patient remains asymptomatic for dyspnea and hemoptysis.

DISCUSSION

Minor or major hemoptysis is a frequent sign of active pulmonary tuberculosis including its sequels such as bronchiectasis or caverns and its associated complications like aneurismal lesions. We describe the case of a patient with tuberculosis sequels and major hemoptysis in which we observe a potential causal relationship among tuberculosis disease and arteriovenous systemic to pulmonary fistula.

The pulmonary arteriovenous malformations (PAVM) are anomalous communications between pulmonary arteries and veins. Their etiology is not well known.

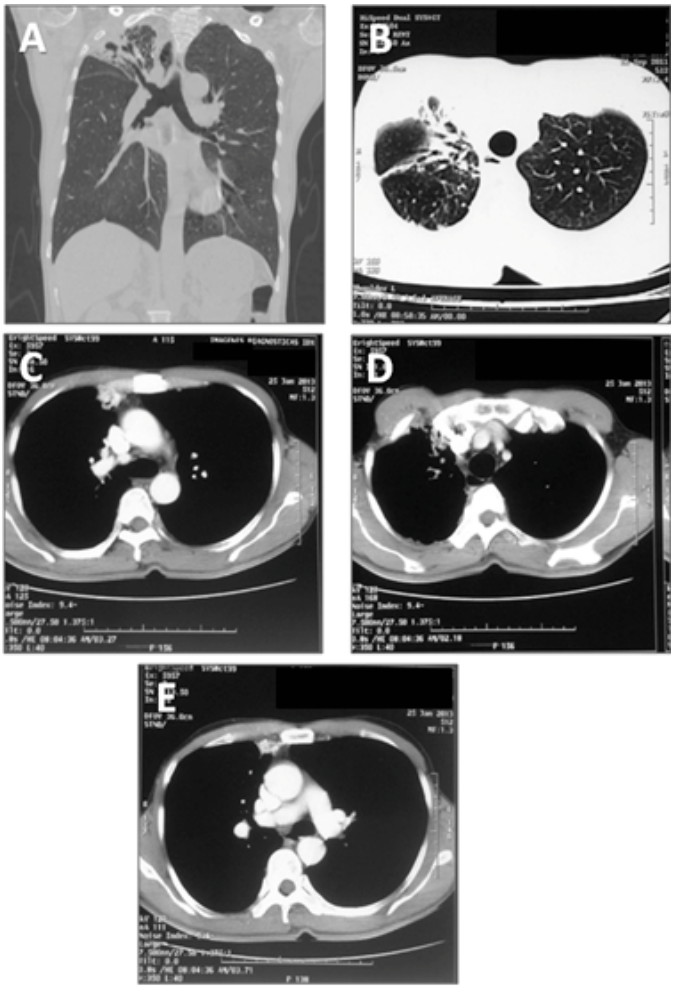

Figure 1: Computed tomography pulmonary angiogram using Omnipaque™ contrast medium and the smart prep protocol. (A) Coronal multiplaner reconstruction identifying fibrotic changes in the right upper lobe. (B) Lung window axial image showing bronchiectasis and volume loss in the right upper lobe, and some tree-in-bud lung opacities at the right inferior lobe spical degment. (C, D, E) Axial images showing multiple tortuous vascular structures in the right thoracic wall, adjacent to the mammary vessels. It also illustrates prominence of the right internal mammary vessel.

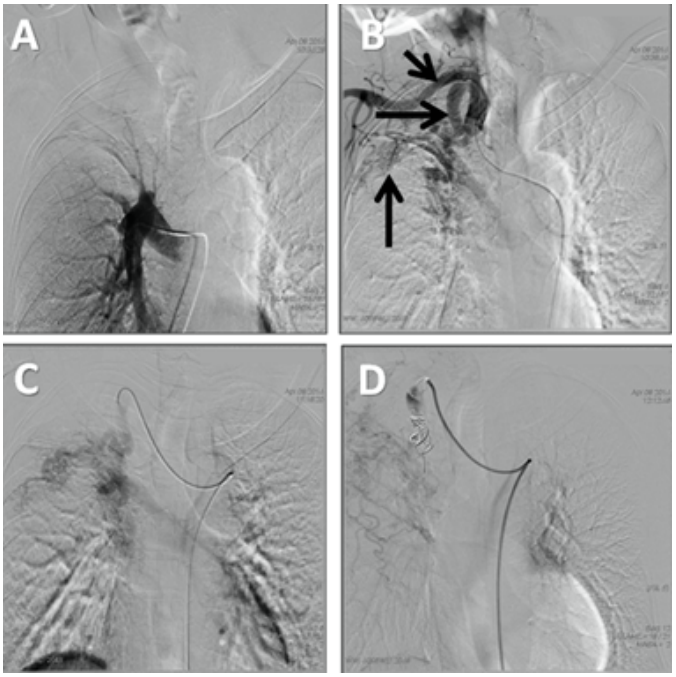

Figure 2: Pulmonary angiography using Omnipaque™ contrast media. (A) Selective angiography of the right pulmonary artery showing no intrapulmonary fistula (B) Selective angiography of the right subclavian artery that illustrates a normal subclavian artery (arrow 1), hypertrophic right mammary artery with multiple collaterals to the right upper lobe (arrow 2). It shows rapid filling of right superior pulmonary vein proving the diagnosis of systemic-pulmonary arteriovenous fistula (arrow 3) (C) The venous phase of selective arteriography of the right internal mammary artery shows a high flow fistula with large collaterals from the mammary artery to the pulmonary vein. (D) For selective embolization of the internal mammary artery, two coils (Interlock, Boston Scientific) and two vascular closure devices (Amplatzer Vascular Plug, AGA Medical Corp.) were used. Complete occlusion of the mammary artery is evident, although the filling of the pulmonary vein persists through two small branches from of the right subclavian artery.

Some authors have proposed theories explaining their origin from the fetal formation of the vessels. However, there are also many acquired medical conditions that can explain their formation. That is why PAVM can be congenital or acquired [2]. The congenital are related in 40% of the cases to the Osler-Weber-Rendu syndrome. The acquired PAVM are related to neoplasms, trauma, chronic inflammatory processes such as myocardial revascularization or sternotomy sequels, or post-infectious diseases (tuberculosis or fungal infections). The mechanism by which these medical conditions become correlated with PAVM is unknown. PAVM are supplied by pulmonary or systemic arteries, when PAVM are fed by systemic arteries, the presence of hereditary hemorrhagic telangiectasia or Osler Weber Rendu syndrome are ruled out since they are not commonly the cause. Hemodynamics in patients with PAVM represent a right to left shunt that may not be manifested in the clinical aspect, nevertheless PAVM is one of the multiple causes of hemoptysis and can be associated with other symptoms like dyspnea and signs as a continuous murmur. Patients with this disease may even have normal pulmonary artery pressures.

In our literature research, only four case reports mentioning the coexistence between pulmonary tuberculosis and systemic to pulmonary fistulas were found [3–6]. In most of the cases, the authors describe a fistula involving the internal mammary artery and the pulmonary veins. In these cases, endovascular treatment was performed. Some authors explain the possibility of a “recruitment” of extrapulmonary local arteries and their connection to pulmonary vessels. Thomas et al. hypothesized that the inflammatory process surrounding a tuberculosis foci can help recruit local and systemic arteries to supply the inflammatory mass [4]. Pierce et al. postulated that the destruction of the pulmonary parenchyma secondary to tuberculosis disease might contribute to the parasitic arrival of the systemic artery supply to this area [6]. In this case, the primary hemoptysis cause cannot be defined, the diagnosis of a vascular malformation such as systemic PAVM is clearly exposed by the angiographic findings. One limitation of this case is that there are not previous or serial angiographic studies to objectively define the natural history of the identified systemic-pulmonary arteriovenous fistula. This implies that a causal relationship between tuberculosis and the genesis of the fistula in this patient cannot be established. Nevertheless, several possibilities can be mentioned in order to explain the association. A first possibility is the existence of congenital vascular malformations in this patient. If this is the case, tuberculosis disease may have been an epiphenomenon casually affecting a vascular area with a single preexistent fistula, in such case an additional fistula would be expected, but in both the invasive angiographic study and the CT scan a vascular malformation in the right upper lobe of the patient was the only finding. Moreover, the patient did not show other common clinical features of an Osler-Weber-Rendu syndrome. A second possibility to explain the systemic to pulmonary fistula is potential neoangeogenic process secondary to the chronic inflammatory events of tuberculosis and its sequel. The acute, subacute, and chronic inflammatory events during and/or after the disease could be chronic angiogenic triggers for vascular proliferation, erosion, and parietal transgression resulting in the formation of fistulous tracts between the internal mammary and subclavian arteries to the upper lobe pulmonary veins. This second alternative deserves special attention in our case. These findings allow us to emphasize that the growth of new vessels must be considered nonexclusive of the bronchial vascular area, but also can affect areas out of the pulmonary parenchyma in those patients with sequels of pulmonary chronic inflammatory diseases.

CONCLUSION

In conclusion, neoangiogenic stimuli and vascular remodeling resulting in acquired pulmonary arteriovenous malformations must be taken in mind when facing a patient with recurrent massive hemoptysis and a previous history of pulmonary tuberculosis.

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Author Contributions
Maria Fernanda Saavedra – 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
Maria Juliana Valenzuela – 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
Monica Ocampo – Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Carlos Garavito – Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
José Federico Saaibi – 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.
Mauricio Orozco-Levi – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published.

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Intraduodenal hematoma presenting with small bowel obstruction, pancreatitis, and subsequent obstructive jaundice in a hemophiliac patient

Anas K. Gremida, Matthew Stotts, Bali Gill, Haripriya Maddur

ABSTRACT

Introduction: Intraduodenal hematoma (IDH) is a rare cause of small bowel obstruction. IDH usually results from blunt trauma to the abdomen. Case Report : We report a case of a 24-year-old male with a history of hemophilia A who presented with intraduodenal hematoma presenting with small bowel obstruction, pancreatitis, and subsequent obstructive jaundice. Conclusion: Spontaneous intraduodenal hematomas are rare clinical entity warrants high index of suspicion in patients with underlying risk factors such as hemophiliac patients.

Keywords: Spontaneous intraduodenal hematoma, Pancreatitis, Small bowel obstruction

How to cite this article

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INTRODUCTION

Intraduodenal hematoma (IDH) is a rare cause of proximal bowel obstruction. Traumatic injury is the most common identifiable cause [1]. Risk factors for spontaneous non-traumatic small bowel hematomas include pancreatitis, bleeding disorders, anticoagulant therapy, vasculitis and bleeding diathesis [2–4]. We report an unusual case of small bowel obstruction, obstructive jaundice, and pancreatitis caused by a spontaneous IDH in a hemophiliac patient.

CASE REPORT

A 24-year-old male with history of hemophilia a presented with abrupt onset of severe epigastric pain, nausea, and coffee ground emesis. He had been non-compliant with his factor VIII infusion and endorsed recent heavy weight lifting. There was no history of direct trauma. On examination, the patient's vitals were within normal limits. He had hypoactive bowel sounds and tenderness in his epigastric region with no rebound or guarding. The rest of his physical examination was unremarkable. His initial laboratory workup revealed hemoglobin 8.1 g/dL, AST 13 u/L, ALT 10 u/L, alkaline phosphatase of 45 u/L, total bilirubin 0.6 mg/dL, and lipase level was of 1322 mg/dL. A computed tomography (CT) scan of the abdomen revealed an intraduodenal mass measuring 8.1x14x14 cm (Figure 1) extending into the intraperitoneal and retroperitoneal spaces with decompression of the distal small bowel loops and abutting the head of the pancreas. The mass measured 40 HU on the Hounsfield scale which is consistent with a hematoma. A more radiopaque mass in the center with 60 HU reflects a central blood clot. The patient was admitted to the intensive care unit for supportive care. A nasogastric tube was placed for decompression and

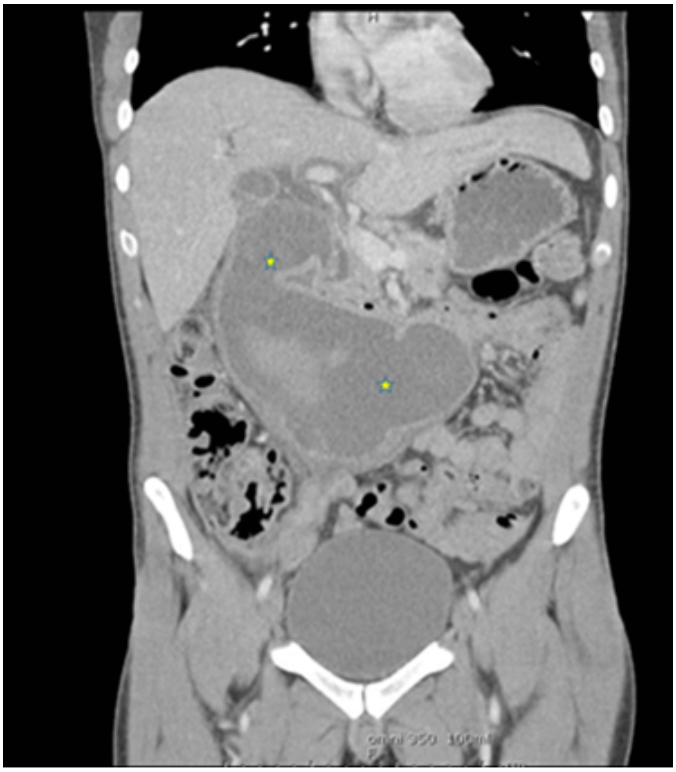

Figure 1: Non-contract computed tomography scan of the abdomen and pelvis showing a large intraduodenal hematoma (8x14 cm) extending into the retroperitoneal space (yellow asterisks).

packed red blood cells were transfused in addition to factor VIII (Humate P®) infusion.

On the third day of admission, after the patient was hemodynamically stable, an EGD revealed a Mallory Weiss tear near the cardia and a large hematoma in the lumen of the duodenum. The patient continued to be managed conservatively. On hospital day seven, the patient was noticed to have new onset jaundice, laboratory revealed a total bilirubin of 6.6 mg/dL (direct bilirubin was 5.0 mg/dL), AST 33 u/L and ALT 42 u/L. Computed tomography (CT) scan of the abdomen showed biliary and pancreatic duct dilation which was likely secondary to external compression from the duodenal hematoma. With further conservative management and factor VIII infusions, jaundice resolved with reduction of the hematoma on serial CT scans.

DISCUSSION

Intramural duodenal hematoma (IDH) is a rare cause of obstruction of the proximal gastrointestinal tract. Blunt abdominal trauma is the most common cause of IDH, a finding that has been attributed to the duodenum's relatively fixed location and its rich blood supply [1]. While anticoagulation with warfarin is the most common risk factor for spontaneous non-traumatic intramural bleeding, other causes of non-traumatic IDH include endoscopic biopsies, aneurysms, pancreatitis and bleeding diathesis [2–4].

IDH was first described in 1838 by Mclachan in a patient with pseudoaneurysm [5]. In 1908, Van Khautz reported the first case of spontaneous intramural hematoma in a hemophiliac patient [6]. Whereas traumatic small bowel hematomas commonly happen in the duodenum, most spontaneous hematomas tend to involve the jejunum [7, 8]. Our patient had no risk factors which could explain why the bleeding occurred in the duodenum rather than the jejunum. Even though pancreatitis can lead to an intraluminal bleeding, it is very unlikely that pancreatitis can cause such a large hematoma. The clinical presentation of IDH is mostly abdominal pain. Subsequent vomiting may indicate that the hematoma becomes obstructive. Presence of jaundice should raise the suspicion of external compression of the biliary tree, but could also result from hematoma breakdown. Diagnosis of IDH relies largely on clinical suspicion and radiographic studies. The CT scan with contrast is the most useful diagnostic tool in detection of intraluminal hematomas. Hounsfield units scoring system is a quantitative scale used to describe radiodensity and gives a clue about the nature of the structures on CT scans. Freely flowing blood has a density of 20–45 Hounsfield units (HU), while blood clots are more radiopaque and generally score between 45 and 70 HU [9]. Magnetic resonance imaging (MRI) scan is a good alternative when the enhanced CT scan is not possible, typically showing the characteristic three-layered sign, which is caused by the tendency of hemosiderin to deposit into layers [10]. In hemodynamically stable patients with IDH, the management is usually conservative, which includes bowel rest, bowel decompression, and correction of coagulopathy. Resolution of the hematoma is usually expected within a few weeks which depends on the size of the hematoma [8]. Surgical intervention through laparoscopy or laparotomy is reserved for hemodynamically unstable patients or in cases of perforation or ischemia.

CONCLUSION

Spontaneous intraduodenal hematomas are one of the rare causes of intestinal obstruction and pancreatitis in a non-traumatic patients. High index of suspicion is required in patients with bleeding tendency who present with symptoms suggestive of high intestinal obstruction or pancreatitis.

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Author Contributions

Anas K. Gremida – 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

Matthew Stotts – Analysis and interpretation of data, revising the article for important intellectual content. Final approval of the version to be published.
Bali Gill – Acquisition of data, drafting the article. Final approval of the version to be published.
Haripriya Maddur – Analysis and interpretation of data, Revising the article critically for important intellectual content, final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Severe cytomegalovirus infection in an immunocompetent host potentially related to flood water exposure

Carmelo Blanquicett, Josh Denny, Vincent Morelli

ABSTRACT

Introduction: In the immunocompetent host, cytomegalovirus (CMV) infection is typically asymptomatic. However, symptomatic disease can also occur and will manifest as flu-like symptoms or as mononucleosis. Case Report: We report a case of severe CMV infection in an immunocompetent, previously-healthy adolescent, male, possibly as a result of flood-water exposure in the South-Central region of the United States. Conclusion: This case highlights the need to consider CMV infection as a cause for the presenting symptoms described herein, in immunocompetent patients.

Keywords: Antiviral therapy, Cytomegalovirus (CMV), Immunocompetent

How to cite this article

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INTRODUCTION

In the US, approximately 60–80% of the population has been exposed to cytomegalovirus (CMV) [1, 2]. CMV infection in the immunocompetent host is usually asymptomatic. However, it can also present with flu-like symptoms or more commonly, it can present as a mononucleosis syndrome [3, 4]. The illness is generally self-limited, with complete recovery over a period of days to weeks. Antiviral therapy is usually not indicated. The CMV complications can result in significant morbidity, including meningitis, encephalitis and pneumonitis; and thus, accurate diagnosis is necessary.

To our knowledge, there are few cases reported in literature demonstrating severe, multi-system CMV infection in previously healthy, immunocompetent hosts. These few reports describe CMV colitis and central nervous system (CNS) infection with sequelae such as meningitis and encephalitis [5]. A more recent case report describing CMV-induced colitis is illustrative [6].

This case report describes clinical, hematological and ocular manifestations of CMV infection in an immunocompetent, previously healthy, 19-year old male who acquired a CMV infection, possibly-although debatable- after being exposed to muddy flood waters, as a consequence of significant flooding in spring of 2010.

CASE REPORT

A 19-year-old Caucasian male was admitted to our medical-surgical floor with complaints of fever for two weeks and a papular rash five days prior that started on the trunk and extended to the extremities. Associated symptoms included nausea, vomiting, headache, dizziness, left flank pain, and periorbital swelling of the right eye. The patient denied any hiking activity, yard work, camping or tick bites. He denied sick contacts or

new sexual partners. The patient affirmed that he had been working in muddy floodwaters, approximately one month prior to his developing symptoms and seeking treatment. Past medical history was significant for ADHD, for which the patient was taking dextroamphetamine. He had unremarkable social and family history.

Physical examination revealed a blood pressure of 122/66 mmHg, heart rate of 96 bpm, respiration rate 18/min; temperature 98.7°F. The patient was alert and oriented, but unwell. An erythematous morbilliform rash was present on the trunk and extremities. No lesions were observed on the palms or soles of the feet. An HEENT examination revealed a swollen, right, superior eyelid that was non-tender and non-purulent. No icterus, oral lesions, or pharyngitis was present. Cardiopulmonary examination was within normal limits. Abdominal examination revealed no hepatomegaly or splenomegaly. Costovertebral angle tenderness was noted on the left. No focal deficits were observed. Cranial nerves were grossly intact, and no meningismus was appreciated.

Laboratory findings demonstrated white blood cell count that was within a normal range (7.9/mm^3); thrombocytopenia was present, with a platelet count of 74,000 and a lymphocyte percentage of 58; hemoglobin and hematocrit were 17.7 g/dL and 41.8%, respectively. A complete biochemistry panel was notable for elevated liver enzymes with an AST and ALT of 489 and 454 U/L, respectively. Alkaline phosphate was 417 U/L. Total bilirubin was 1.8 mg/dL and C-reactive protein was 51.6 mg/L. A urinalysis that was positive for bilirubin and ketones, but negative for nitrites or leukocyte esterase; urine culture was negative. Laboratory results further demonstrated a negative Epstein–Barr virus (EBV) panel, based on serological testing, influenza negative, according to rapid influenza diagnostic testing, gonorrhea and chlamydia by nucleic acid amplification test, negative,

A chest X-ray proved to be normal, and abdominal computed tomography showed mild hepatomegaly which was not detected on physical examination. The initial diagnosis that was assigned was fever of unknown origin, possibly attributed to a tick borne illness. The patient was treated for nausea with ondansetron; morphine, as needed (PRN) and ibuprofen for headache and flank pain. Hepatitis—A, B, and C, HIV and *Borrelia Burgdorferi* serology were ordered and subsequently negative. An infectious disease consult was requested RPR, EBV and leptospira test were either non-reactive or negative. CMV serology yielded positive results with CMV IgM and IgG both being elevated by serological methods. HHV-6, a close relative of CMV, was not investigated, and in retrospect, would have provided additionally-useful information, as a rash (and periorbital swelling) is an unusual symptom for CMV. A rash is more commonly related to HHV-6 but unfortunately, samples were not available for analysis. Follow-up CMV titers were also not obtained and could have proven useful. The patient's symptoms improved with supportive therapy, and he remained well during follow-up.

DISCUSSION

At least 60% of the US population has been exposed to CMV [7], with a prevalence of more than 90% in high-risk populations [8]. Demmler et al. [1] maintain that in developed countries, such as the United States or United Kingdom, as many as 60–80% of the population will be infected with CMV by adulthood.

The CMV manifestations in immunocompromised hosts have been widely documented, however, reports of those manifestations present in immunocompetent hosts are few and merit attention.

In the majority of hosts, primary CMV infection is clinically silent. When symptomatic, CMV disease in immunocompetent individuals may present as a mononucleosis syndrome or as flu-like symptoms. Individuals who are at increased risk for CMV infection include those who attend or work at daycare centers, persons who have multiple sex partners, and recipients of CMV mismatched organ or bone marrow transplants. Sexual activity may be an important risk factor for acquiring CMV infection, and its transmission by sexual acts has been well-documented [9]. The virus can spread by horizontal transmission (direct contact, person to person with virus containing secretions such as saliva, urine, cervical secretions or semen) or vertically via transplacental passage. Adolescence is another period of rapid acquisition of CMV. [10]

When CMV produces a mononucleosis–like syndrome, the most common manifestations are fever, fatigue, pharyngitis, adenopathy, and hepatitis. Headache, abdominal pain with diarrhea, arthralgias, and rash may also occur. Laboratory abnormalities include lymphocytosis or lymphopenia with thrombocytopenia and elevated transaminases. However, the heterophile antibody titers or monospot tests will be negative. This presentation fits closely with the presentation of the patient described in this case report. However, our patient presented with ocular symptoms, which are not frequently observed in these cases.

The confirmation of an acquired CMV infection is best accomplished by documenting a CMV IgG seroconversion with the presence of CMV IgM antibody. In our particular example, the patient had positive IgM and IgG antibodies. CMV cultures of the urine, saliva and blood may also be positive during the acute phase of the infection, but according to Fauci et al., the most sensitive way of detecting CMV in blood or other fluids may be by PCR [11]. The demonstration of the presence of CMV genome in blood by PCR would have been more helpful in defining the timing of infection in this case. Stagno et al., maintain that a positive IGM can persist up to one year after infection [12]. Irrespective of the detection methods used, we were, unfortunately, unable to categorically determine that the patient's source of infection was attributed to flood waters and could only highlight an association, in a speculative manner.

Subclinical transaminitis is the most common finding

in immunocompetent patients; elevations of alkaline phosphatase and total bilirubin are less typical [4]. The patient in this report had both transaminitis and elevations in total bilirubin and alkaline phosphatase. The patient improved with supportive therapy. Our case highlights a source of potential CMV infection that is not widely reported; although, an alternative cause cannot be excluded. Further, this immunocompetent patient presented with atypical signs of systemic CMV infection, including periorbital swelling, thrombocytopenia and dark urine with hepatosplenomegaly on imaging.

CONCLUSION

In conclusion, our case presents a case of severe CMV infection possibly related to flood water exposure- although debatable- and highlights the need to consider CMV infection as a cause for the presenting symptoms described herein, in immunocompetent patients.

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Author Contributions

Carmelo Blanquicett – 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
Josh Denny – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Vincent Morelli – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Dabska tumor of the tongue: A clinicopathological study with confocal laser scanning microscopy

Gianfranco Favia, Luisa Limongelli, Angela Tempesta, Eugenio Maiorano

ABSTRACT

Introduction: Dabska tumor, or papillary intra-lymphatic angioendothelioma (PILA), first described in 1969 by Dabska, is a rare vascular low grade malignancy that usually affects the skin and subcutaneous tissues of infants. Case Report: We report a case of PILA of the right tongue in a 65 years old male patient presenting as a white-bluish multinodular lesion, measuring 4x3 cm. After wide excision, histological examination was carried out with confocal laser scanning microscopy (CLSM). The histological features seen during optical examination were: thin-walled vascular spaces resembling a cavernous lymphangioma, and the formation of prominent intraluminal papillary tufts with hyaline cores lined by hobnail endothelial cells. The CLSM analysis highlighted muscle infiltration, and high fluorescence of intraluminal papillary core due to the presence of young thin collagen fibers devoid of cross-links. Thus, a final diagnosis of PILA was made.
Conclusion: Confocal laser scanning microscopy analysis could surely facilitate the histological diagnosis, allowing the identification of some features of the tumor.

Keywords: Benign vascular tumor, Confocal laser scanning microscope (CLSM), Dabska tumor, Papillary intra-lymphatic angioendothelioma (PILA)

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INTRODUCTION

Vascular tumors are very common and most frequently occur in the skin and oral cavity and they could be classified, according to WHO classification, in benign tumors, low grade and high grade malignant tumors. A rare low grade vascular malignant tumor is papillary intralymphatic angioendothelioma (PILA), also known as Dabska tumor, a locally aggressive, rarely metastasizing vascular lesion characterized by lymphatic-like channels and papillary endothelial proliferation. These tumors appear to be closely related to retiform hemangioendothelioma [1]. We report a case of Dabska tumor of the tongue emphasizing the importance of confocal laser scanning analysis for diagnosis and differential diagnosis.

CASE REPORT

A 65-year-old male referred to the oral surgery unit of the University of Bari, presenting a multinodular painless exophytic mass on the right posterior margin of the tongue 4 cm in size. The patient's clinical history

highlighted the rapid growth of the tongue nodule within two months on a pre-existing red plane lesion. The lesion was characterized by white or bluish overlying mucosa, and variable consistency in different areas, ranging from soft to soft-elastic (Figure 1). The regional lymph nodes were not enlarged. Computed tomography scan showed a lesion with endophytic pattern of growth, intermingling cystic and solid areas with irregular border, diffuse enhancement with contrast media and a depth extension of 2 cm.

The clinical differential diagnosis had been made among benign soft tissues vascular tumor (such as hemangioma or lymphangioma), vascular low grade malignant tumor (Dabska tumor or endovascular papillary hemangioendothelioma), vascular high grade malignant tumor (angiosarcoma or Kaposi's sarcoma), vascular tumor-like lesions (intravascular papillary endothelial hyperplasia), cysts and cystic tumor of salivary gland (mucoepidermoid carcinoma), and cystic metastatic tumor. A definitive diagnosis could not be reached because of the clinical aspects of the lesion. Therefore, a fine-needle aspiration biopsy (FNAB) was performed. The specimen was formalin-fixed, paraffin-embedded, stained with hematoxylin-eosin, PAS, Van Gienson, and Picrosirius red and then sent to the Department of Pathological Anatomy of the University of Bari. The histological examination was carried out using a Nikon Eclipse E600 microscope (Nikon Corporation, Tokyo – Japan), equipped with Argon-ion and Helium-Neon lasers, emitting at 488 nm and 543 nm wavelengths, which allows both optical and confocal laser scanning analysis. The Nikon EZ C1 software (Nikon Corporation, ver. 2.10 Coord Automatisering) was used for bi-dimensional image processing. Under standard histological analysis, the tumor showed a submucosal location, with extension and involvement of superficial muscle layer of the tongue. It was composed by dilated, thin-walled vascular spaces resembling a cavernous lymphangioma and different size neoplastic nodules infiltrating the tongue intrinsic muscular tissues (Figures 2 and 3). The characteristic finding was the formation of prominent intraluminal papillary tufts with hyaline cores lined by hobnail endothelial cells. The endothelial cells had a pale or dark eosinophilic cytoplasm and round or ovoid nuclei. At high magnification it was possible to highlight, between atrophic remnants of intrinsic muscular tissues, ecstatic lymphatic vessels with endovascular papillary neoplastic proliferation associated with empty small and semilunar shaped vascular spaces, in more solid nodular worly shaped neoplastic cells. Absent or minimal cytological atypia and rare mitotic figures were seen. Peripherally, the tumor showed dilated anastomosing blood vessels. Confocal laser scanning microscopy (CLSM) analysis highlighted lack of fluorescence in muscle infiltration area, depending on the resumption of soft tissues collagen fibers. Moreover, CLSM showed the high fluorescence of intraluminal papillary core due to the presence of immature thin collagen fibers devoid of cross-links (Figures 4 and 5).

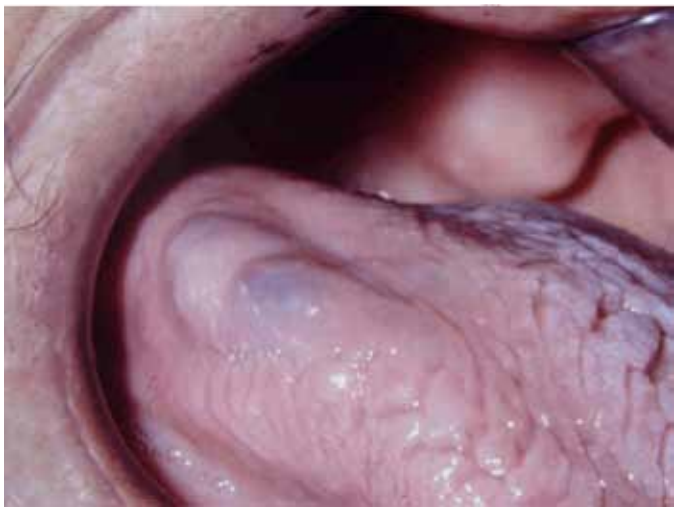

Figure 1: Multinodular white-bluish painless exophytic mass on the right posterior margin of the tongue, of 4 cm.

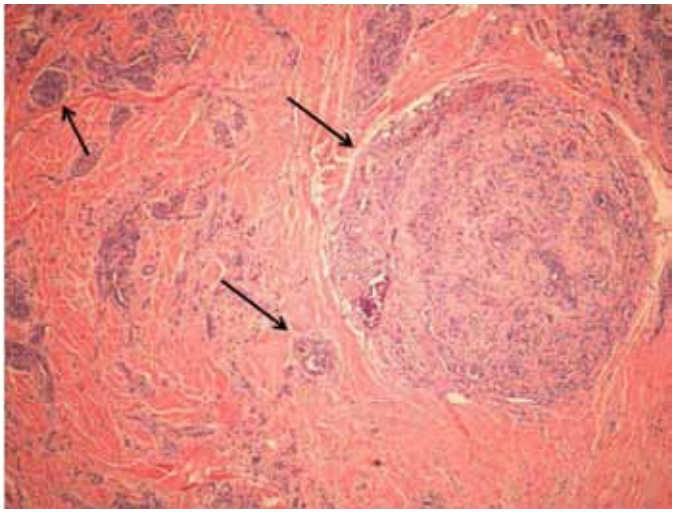

Figure 2: Hematoxylin-eosin staining (x100) showing different size neoplastic nodules infiltrating the tongue intrinsic muscular tissues.

Under general anesthesia, the surgeon performed radiofrequency wide local excision with intraoperative histological negative control of resection margins. No evidence of local recurrence or lymph nodes metastasis during the follow-up of three years.

DISCUSSION

Papillary intralymphatic angioendothelioma is a locally aggressive, rarely metastasizing vascular lesion characterized by lymphatic-like channels and papillary endothelial proliferation. This tumor is closely related to retiform hemangioendothelioma [1], also known as endovascular papillary angioendothelioma or Dabska tumor, it was described for the first time in 1969 by

Maria Dabska, reporting six cases as childhood tumors in the skin and subcutis [2]. PILA has predilection for infants and children, although 25% of the cases occur in adulthood [3]. Male to female ratio is 1:1. PILAs appear as plaques or nodules with asymptomatic growth, are ill defined and usually involve the dermis and subcutaneous tissues. In literature, these tumors are thought to arise from different anatomical locations such as the spleen, tongue, testicles and bone [4, 5]. Usually, they are diagnosed when they are 2–3 cm in size and they are more commonly diagnosed as hemangioma [6]. In this case report, Dabska tumor arise on the right posterior margin of the tongue presenting as multinodular painless exophytic mass 4 cm in size. The clinical diagnosis of this tumor is very difficult in view of its variable consistency and color, as well as its multinodular growth pattern. In this case here reported, the clinical differential diagnosis had been made among benign soft tissues vascular tumor, vascular low grade malignant tumor, vascular high grade malignant tumor, vascular tumor-like lesions, cysts and cystic tumor of salivary gland, and cystic metastatic tumor. Benign soft tissues vascular tumor, vascular tumor-like lesions and cysts of salivary gland are usually described as painless soft exophytic masses with bluish overlying mucosa, although generally in CT imaging they lack in endophytic pattern of growth, and solid areas with irregular border. Multinodular appearance, exophytic and endophytic patterns of growth, size, growth rate, depth extension and alternation of cystic and solid areas could prompt diagnoses of vascular low grade malignant tumor, vascular high grade malignant tumor, cystic tumor of salivary gland, or cystic metastasis, even though the lack of ulcers, pain, bleeding and lymph nodes involvement.

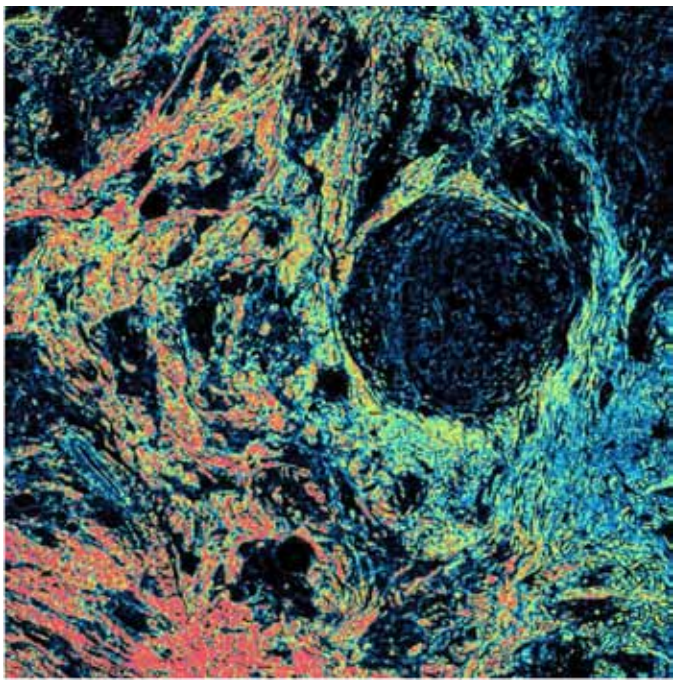

Figure 3: At confocal laser scanning microscopy examination showing poor fluorescence of tumoral tissues clashing with high fluorescence of non-neoplastic connective stroma.

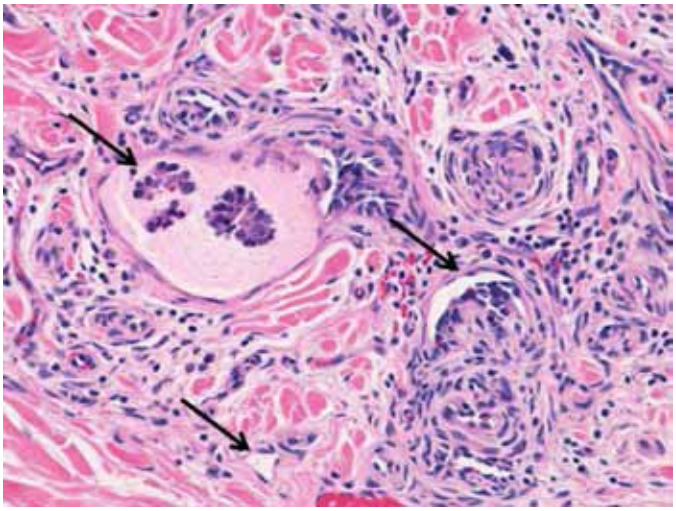

Figure 4: Hematoxylin-eosin staining (x250) between atrophic remnants of intrinsic muscular tissues are present evident ecstatic lymphatic vessels with endovascular papillary neoplastic proliferation associated with empty small and semilunar shaped vascular spaces, in more solid nodular worly shaped neoplastic cells.

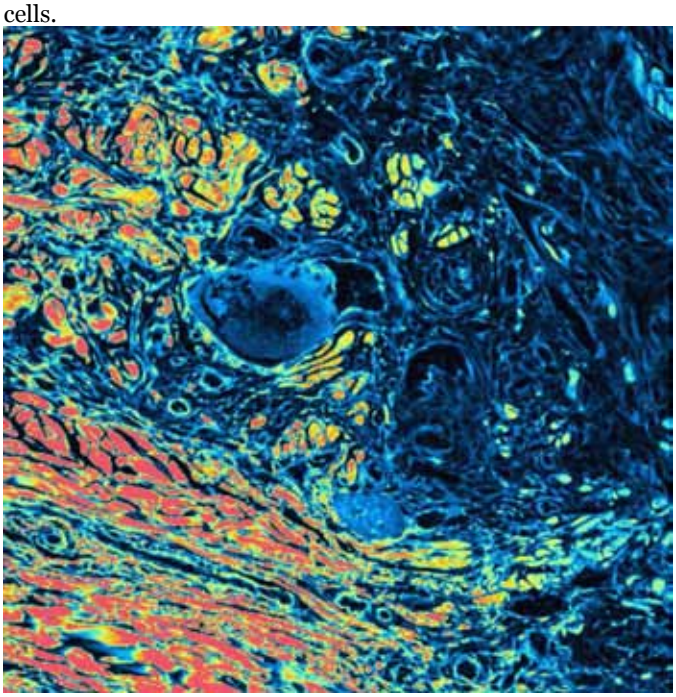

Figure 5: Confocal laser scanning microscopy highlighted lack of fluorescence in muscle infiltration area, due to the resorption of soft tissues collagen fibres.

Although PILAs may show different presentations depending on the analyzed area, FNAB or biopsy could be performed to achieve a certain diagnosis. Microscopically, PILAs are composed by endovascular papillary proliferations that project into dilated, thin-walled, lymphatic spaces [7]. At high-power view, they demonstrate a pathognomonic hobnail-like appearance [8] with scant pink cytoplasm and a prominent nucleus, with little or no cytological atypia [1]. Mitotic figures are rare [9]. Endothelial cells are

immunoreactive for CD34, CD31, and factor VIII-related antigen [10]. Histologically, PILA should be distinguished from other lesions presenting endothelial papillary overgrowth and vasoproliferation such as intravascular papillary endothelial hyperplasia (IPEH) [9], hemangioma, epithelioid hemangioendothelioma [10], hemangiopericytoma, and angiosarcoma. Confocal laser scanning microscopy examination allow to evaluate the high fluorescence of intraluminal papillary core due to the presence of immature thin collagen fibers devoid of cross-links and lack of fluorescence in muscle infiltration area, depending on the resumption of soft tissues collagen fibers.

Wide local excision is considered the treatment of choice for this low grade malignancy: the use of radiofrequency excision results in a reduction in bleeding and improved healing of tissues after surgery. Moreover, radiofrequency excision resets the postoperative edema, pain, and infection to zero. A tendency for local recurrence and lymph nodes metastasis was reported [1].

Nevertheless, a few cases of PILA have been reported thus far, and this suggests the need for a standardized clinicopathological approaches to achieve an accurate diagnosis and effective treatment, as clear-cut criteria for the differential diagnosis between PILA and PILA-like lesions are still lacking. CLSM analysis allows to achieve a certain histological diagnosis.

CONCLUSION

Confocal laser scanning microscopy analysis could surely facilitate the histological diagnosis, allowing the identification of some features of the tumor.

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Author Contributions

Gianfranco Favia – 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
Luisa Limongelli – Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Angela Tempesta – Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Eugenio Maiorano – Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS

Catamenial pneumothorax: A rare cause of recurrent pneumothorax

Waqas Jehangir, Jay Harman, Nneka Iroka, Abdalla Yousif

ABSTRACT

Introduction: Primary spontaneous pneumothorax is a common clinical occurrence. Although primary spontaneous pneumothorax is twice as common in men as in women. The recurrence rate is significantly higher in women. The two primary causes for recurrence in women are catamenial pneumothorax and endometriosis related pneumothorax. In the past, catamenial and/or endometriosis related pneumothorax were greatly underdiagnosed. The incidence has increased in the past decade because it is more easily recognized today. Spontaneous pneumothorax is a lung compression that occurs spontaneously due to air in the pleural space in a patient with no underlying lung disease. It can occur in men or women but occurs most often in men. It occurs through many different causes. Catamenial pneumothorax is a spontaneous pneumothorax that occurs at the time of menses in a woman that allows air to enter the thoracic space. Non-catamenial endometriosis related pneumothorax is a spontaneous pneumothorax that occurs when endometrial tissue ascends through diaphragmatic defects to the pleural space and allows air to enter. It can occur at any time and not just during the menses in a woman. Catamenial pneumothorax and noncatamenial endometriosis related pneumothorax are independent entities and are not synonyms. They may occur simultaneously but do not necessarily have to occur at the same time. Case Report: We present a case of recurrent pneumothorax diagnosed as catamenial pneumothorax in an otherwise healthy 34-year-old female. Conclusion: The percentages of catamenial pneumothorax are still unclear but it should be suspected and affectedly treated.

Keywords: Primary spontaneous pneumothorax, Catamenial pneumothorax, Endometriosis, Video assisted thoracoscopic surgery

How to cite this article

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INTRODUCTION

Primary spontaneous pneumothorax was first described in 1932 as a separate medical condition that occurs in patients without lung disease [1]. The incidence is thought to be increasing and the recurrence rate is between 20–60% [1]. The male to female ratio in primary spontaneous pneumothorax is 2:1 while the recurrence rate is significantly higher in women [1]. The term recurrent does not define the exact number of episodes although the mean is very high: five episodes with as many as 10 episodes in some cases [2]. At least two episodes are

required to be considered recurrent pneumothorax [3]. Therefore, the occurrence of recurrent pneumothorax is a significant problem especially in women. Catamenial pneumothorax and/or endometrial related pneumothorax are one of the main causes of recurrent pneumothorax in women. Although known since the 1950s, catamenial pneumothorax was considered an extremely rare condition and was greatly underdiagnosed [2]. Although catamenial and/or endometriosis related pneumothorax are recognized more now than in the past, their real frequency remains unclear [4]. The signs and symptoms of recurrent pneumothorax are the same as for other kinds of pneumothorax—chest pain, shortness of breath, and cough [2].

CASE REPORT

A 34-year-old Hispanic female non-smoker with no significant past medical history stated that she had been having pain in the scapula four days ago which was associated with shortness of breath. She described the pain as a sharp, constant, radiating to the front and back of the right side of the chest. The patient also stated that shortness of breath is associated with chest pain and dry cough. She was diagnosed with pneumonia by her primary medical doctor and she was prescribed antibiotics and pain medications which did not help her. She gave a history of spontaneous pneumothorax on her right side six months ago. She denied any fever, chills, nausea, and vomiting but stated that the pain gotten progressively worse. She did not have a history of tuberculosis or endometriosis. She had breast lifting seven years ago. Her last menstrual period was three days ago. She had three abortions which were induced and she has two babies. On physical examination, she was in mild respiratory distress and vital signs were temperature 98°F, blood pressure 127/58 mmHg, pulse 84/min, respirationrate 20/min, and PO2 96% on 2 litre/minute of oxygen per nasal canula. Lung examination revealed decreased air entry on the right side and decreased breath sounds on the right side. Rest of the physical examination was unremarkable. Laboratory data showed white blood cell count $10.4x10^3$/μL, hemoglobin 13 g/dL, hematocrit 41%, platelets $343x10^3$ K/uL, neutrophils 68%, lymphs 21%, BUN 8 mg/dL, creatinine 0.7 mg/dL, calcium 9.4 mg/dL, albumin 4.4 g/dL, total protein 6.7 g/dL, sodium 137 mmol/L, potassium 3.9 mmol/L, chloride 99 mmol/L, CO_2 26 mmol/L. Chest X-ray showed 90% of pneumothorax (Figure 1). She was admitted and emergency right closed thoracostomy drainage done and mechanical pleurodesis was performed. Alpha 1 antitrypsin later came to be 106.00 IU/mL (90–200 IU/mL). Patient was diagnosed with catamenial pneumothorax. She was discharged home and has remained free of recurrence six months after re-treatment.

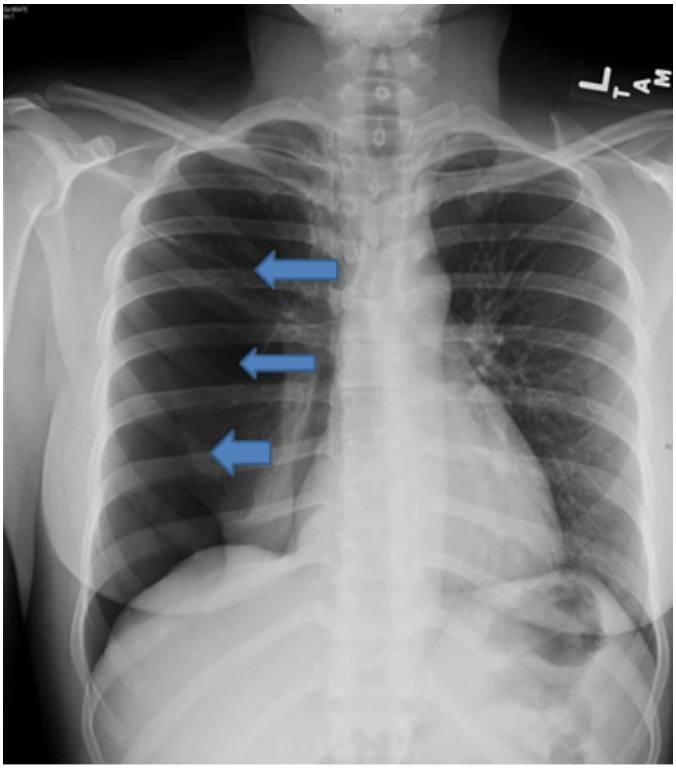

Figure 1: Chest X-ray showing 90% of pneumothorax on the right side.

DISCUSSION

While men are twice as likely to have primary spontaneous pneumothorax as women, whereas women are much more likely to have a recurrence. The current theory is that a woman's menstrual cycle and/or endometriosis play a role in this reoccurrence. The mean age for recurrent pneumothorax in a woman is 32 years [5].

Catamenial pneumothorax is a recurrent pneumothorax occurring 24 hours before and up to 72 hours after the onset of menses [4]. There are two hypotheses concerning the causes of catamenial pneumothorax. The first hypothesis is that the open connection between the atmosphere and the peritoneal cavity during menses allows air to enter the thoracic cavity through diaphragmatic fenestrations and porosities [5]. In the menstrual period the cervical mucus plug is absent, thus permitting communication between the peritoneal cavity and the outside through the uterine and the fallopian tubes. Air may be forced to enter the peritoneum by uterine contractions, physical exercise, or sexual intercourse. The air then reaches the pleural space through diaphragmatic defects because of negative intrathoracic pressure [2]. One argument supporting the theory of transdiaphragmatic passage of air is the observation that recurrent catamenial pneumothorax may be prevented by tubal ligation. The second hypothesis is that prostaglandin F2, a potent constrictor of bronchioles may destroy alveolar tissue causing alveolar rupture and

pneumothorax [5]. In the menstrual period, many women have increased levels of prostaglandin F2-a. Catamenial pneumothorax is unilateral and right sided in almost all instances [2].

The other hypotheses are that endometriosis plays a role in the reoccurrence of pneumothorax. The endometrial tissue reaches the thoracic cavity through auto transplantation to ectopic sites through lymphatic or vascular embolization or after retrograde menstruation [4]. The endometrial tissue can reach the thoracic cavity through diaphragmatic defects possibly caused by endometriosis [5]. This mechanism explains why there is right sided predominance in recurrent pneumothorax. The peritoneal fluids along with air and endometrial tissue exit from the pelvis along the right paracolic gutter up to the right subphrenic space [2, 3] and then through the diaphragmatic fenestrations and porosities. Endometriosis related pneumothorax is considered proven when endometrial glands and stroma are demonstrated by immunohistochemistry staining [4]. Thus endometriosis related pneumothorax can occur in the intermenstrual period [2] as well as the menstrual period, while catamenial pneumothorax only occurs in the menstrual period 24 hours before and up to 72 hours after menses.

The diagnosis of thoracic endometriosis has improved over the past two decades because of Video Assisted Thoracoscopic Surgery (VATS). The VATS is considered the gold standard for both definitive diagnosis and surgical treatment of catamenial and/or endometriosis related pneumothorax [5] and has been applied since 2000 [3]. Some clinicians influenced by the 50% recurrence rate of catamenial and/or endometriosis related pneumothorax advocate an aggressive approach with early surgical treatment [1]. Diaphragmatic involvement by either endometrial tissue or perforations is probably best treated by diaphragmatic resection. Talc pleurodesis is recommended instead of pleural abrasion because of higher recurrence rate with pleural abrasion [2]. Current studies have concluded that surgery has better results than hormone treatment in preventing recurrence. The best results have been obtained using surgery followed by either GnRH agonists or the antigonadotropic progestins cyproterone acetate for six months to induce amenorrhea [2]. If the stapling of the diaphragmatic lesions, the pleurodesis, or the hormone treatment does not prevent recurrence, then hysterectomy and bilateral salpingo-oophorectomy are the treatments of last resort [6].

In our case, the patient had all the classical symptoms of catamenial pneumothorax. She presented with SOB along with chest pain and a cough. Her last menstrual period was three days previously at the time she first had the pain. The lung exam and X-ray revealed a right sided pneumothorax. She had a history of spontaneous pneumothorax six months prior to this. For treatment the patient had a thoracostomy drainage done and mechanical pleurodesis. She has remained free of recurrent pneumothorax at six months.

CONCLUSION

The percentages of catamenial and/or endometriosis related recurrent pneumothorax are still unclear so additional research is needed to clarify this. But it should be suspected and affectedly treated. Operations for catamenial and/or endometrial related pneumothorax have practically zero mortality and no significant morbidity.

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Author Contributions

Waqas Jehangir – 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
Jay Harman – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Nneka Iroka – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Abdalla Yousif – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

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CLINICAL IMAGES OPEN ACCESS

The potential hazards of ear self-cleaning

Iain R. M. Bohler, Wickham M., McCaffer C., Schiszler T., Chin A.

CASE REPORT

A 77-year-old male was brought in by ambulance to the resus bay of the emergency department with major bleeding from his left ear. Attempting to remove remnant cotton wool from a failed 'cleaning' attempt with tweezers, the patient had slipped and fallen on a wet bathroom floor. Tweezers in situ in the external auditory canal, the patient fell to his left side sustaining direct trauma to the auditory meatus resulting in uncontrollable hemorrhage. There was no immediate loss of consciousness. The patient was not known to suffer any coagulopathic disorder, nor prescribed anticoagulant medication.

The patient suffered a syncopal episode in the ambulance, responsive to fast IV fluids, resulting on return of consciousness (GCS 15) on arrival.

With continued bleeding, he subsequently deteriorated into a peri-arrest state with further syncopal episodes and an accompanying hemoglobin drop of 4 g/L. Further crystalloid resuscitation and blood (PRC) transfusion of two units stabilized the patient whilst hemostasis was achieved with a Bismuth Iodoform Paraffin Paste (BIPP) dressing (Figure 1) by the ENT team. The injury was localised to the anterior wall of the external auditory canal raising suspicion of superficial temporal artery trauma. Secondary survey revealed no injury to the facial nerve, and the patient proceeded to radiological imaging.

Computed tomography (CT) scan demonstrated an intact tympanic membrane with absence of blood within

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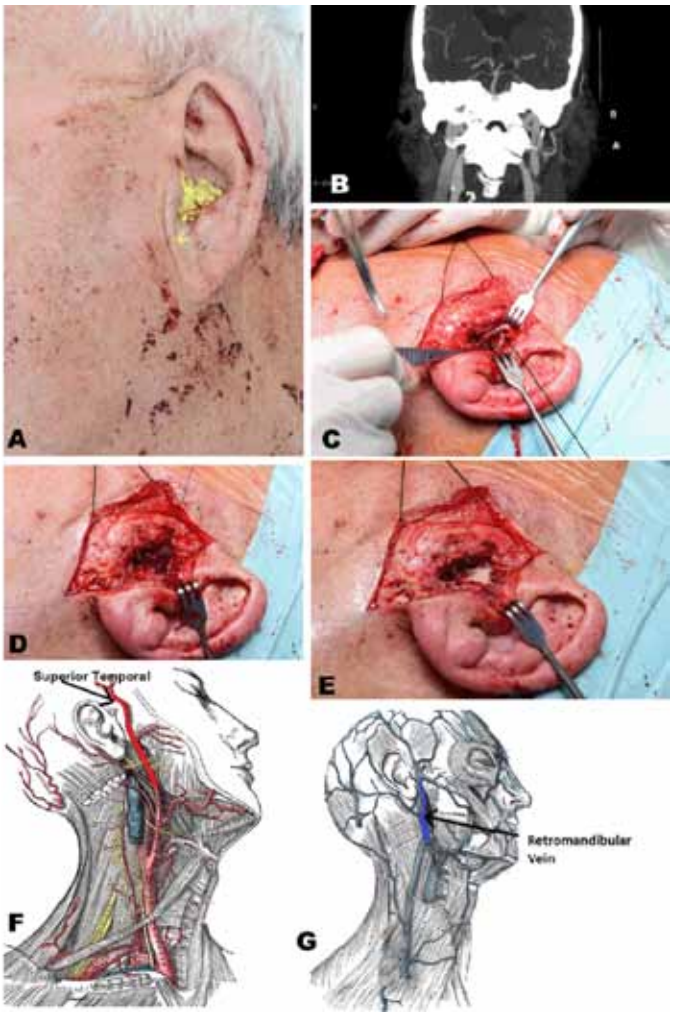

Figure 1: (A) Bismuth Iodoform Paraffin Paste dressing, (B) Coronal view of computed tomography scan showing loss of contrast between points A + B, where pseudoaneurysm has formed, (C) Partially tied off pseudoaneurysm of superficial temporal artery, (D) Tied off Pseudoaneurysm of superior temporal artery, (E) Excised pseudoaneurysm of superior temporal artery, (F) Superior temporal artery, and (G) Retromandibular vein.

the middle ear and mastoid cavity, excluding internal carotid artery and internal jugular vein injury. As suspected, the direction of trauma had sustained injury to the superior temporal artery with additional injury to the retromandibular vein to a lesser extent. The patient was observed on an ENT ward for five days with packing removed as an outpatient on day-7. Broad-spectrum antibiotic prophylaxis provided the only other active medical intervention throughout his initial admission, post resuscitation.

Four weeks post trauma, the patient was seen to have a pulsatile mass on examination of the ear. Computed tomography (CT) scan showed a 1.7-cm pseudoaneurysm arising from the left superior temporary artery. The patient underwent emergency surgery to tie off the pseudoaneurysm and following a brief stay for treatment of a postoperative wound infection, was discharged. The patient has since gone on to make a full recovery.

DISCUSSION

The elegantly contoured pinna of the outer ear acts to funnel air through the external auditory meatus towards the tympanic membrane. The auditory canal is approximately 3 cm long in adults with a slight s-shaping as it meanders towards the middle ear. Cartilage supports the ear at its opening with bone predominating medially. The keratinizing stratified squamous epithelium of the external canal is continuous with the eardrum and peppered with varying numbers of sebaceous and ceruminous glands [1].

The cerumen (wax) produced by the glands of the external ear facilitates the removal of dead epithelium. Aided by the fine hairs of the auditory canal, cerumen prevents airborne pathogens reaching the middle ear, where their accumulation or growth can lead to painful and profoundly debilitating disease [2]. Although this action represents normal bodily function, cerumen is commonly misconceived to be solely a waste product and is often actively removed from the ear. This active removal and the complications associated as such are the basis for a significant number of emergency ENT referrals (foreign body in ear, impaction of wax and associated disease).

This case is an infrequent consequence of a foreign body in the external auditory canal, and despite the severity of bleeding and post injury complications; the patient has gone on to make a full recovery to previous function. Computed tomography (CT) scan demonstrated an injury to the superior temporal artery. Arising from the external carotid artery, it passes anterior to the ear and is palpable superior to the zygomatic arch [3]. The retromandibular vein, also injured, is formed by the union of superficial temporal and maxillary veins. It descends through the parotid gland, deep to the facial nerve and superficial to the external carotid artery. Both vessels were damaged in a plane transverse to the external auditory meatus, as they peregrinate anterior to the ear [4].

Although this case represents a highly atypical presentation, its extremity highlights an avoidable workload on national health services (NHS). Enhanced education of patients through patient-doctor communication and health awareness campaigns, of normal bodily functions, could relieve the burden of patient numbers on emergency units.

CONCLUSION

Although this case report demonstrates an exceptionally unusual case whereby inner ear trauma can lead to massive hemorrhage and pseudoaneurysm formation, it serves well to provide a much broader general message. Ears are self-cleaning; there is no need to insert anything into the ear.

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

Chin A. – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CLINICAL IMAGES OPEN ACCESS

Pneumatosis intestinalis of the cecum mimicking emphysematous cystitis

Jun-Jie Ng, Kon-Voi Tay

CASE REPORT

A 73-year-old diabetic female was presented to the emergency department with lower abdominal pain and distension for five days. Physical examination revealed tenderness over her right iliac fossa and supra-pubic region. An abdominal X-ray (Figure 1) revealed a spherical structure in the pelvis in the expected position of the bladder with presence of intramural gas and a provisional diagnosis of emphysematous cystitis was made by the emergency physicians. Urinary catheterization performed yielded clear urine and urine dipstick did not reveal any evidence of urinary tract infection. The patient's abdominal pain and distension worsened and a surgical consult was made. Further history corroborated with the patient's family revealed that the patient has had associated absolute constipation for last five days. The abdominal X-ray was reviewed which also showed a loop of dilated transverse colon. There was suspicion of colonic intestinal obstruction. Computed tomography (CT) scan of her abdomen and pelvis (Figure 2) showed a massively dilated cecum extending into the pelvis with evidence of pneumatosis intestinalis secondary to a closed loop obstruction from a stenotic splenic flexure colonic tumor. The patient underwent an exploratory laparotomy which revealed a large circumferential and stenotic splenic flexure tumor causing resultant proximal large bowel obstruction. The cecum was massively dilated measuring

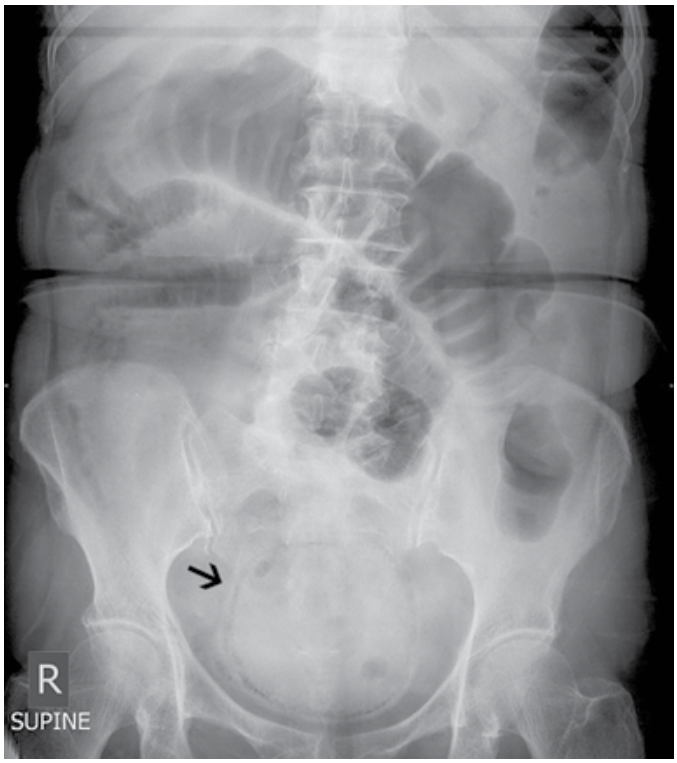

Figure 1: Plain supine abdominal X-ray of the patient showing a circular radiolucent rim (as indicated by the arrow) at the expected position of the bladder in the pelvis suggestive of emphysematous cystitis.

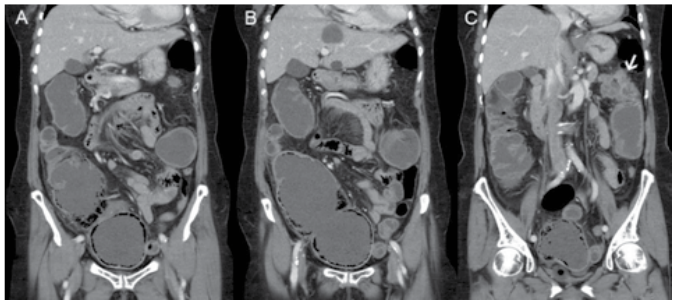

Figure 2: (A, B) Coronal views of a contrast-enhanced computed tomography scan of the patient's abdomen and pelvis showing a massive dilated cecum with pneumatosis intestinalis directed towards the pelvis causing an X-ray appearance that mimicked emphysematous cystitis, and (C) The massive cecal dilatation was due to a closed loop intestinal obstruction secondary to a stenotic splenic flexure colonic tumor (indicated by the arrow).

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up to 14 cm in diameter and the cecal wall appeared thinned out and dusky. The ascending and transverse colon were also dilated up to 5 cm in diameter but still appeared healthy. The patient underwent a subtotal colectomy and was discharged well on postoperative day-10. Histopathological examination of the resected colon revealed a moderately differentiated adenocarcinoma of the splenic flexure exhibiting transmural invasion with involvement of the serosa and omental fat. There was no malignancy identified in 28 harvested lymph nodes and resection margins were free of tumor. The cecum demonstrated mural attenuation with mucosal ulceration and a small focal perforation. The histopathological findings corresponded to a provisional tumor node metastasis (TNM) staging of T4N0.

DISCUSSION

Emphysematous cystitis is a rare but potentially life-threatening clinical entity where there is presence of gas within the bladder wall and lumen during a urinary tract infection. It usually occurs in elderly women with poorly controlled diabetes mellitus [1–3]. Common causative pathogens are gas forming organisms like *Escherichia coli* and *Klebsiella pneumoniae* [4]. Patients with emphysematous cystitis frequently present with symptoms of abdominal pain, gross hematuria and fever but a significant proportion of patients can be asymptomatic with diagnosis of emphysematous cystitis made incidentally on abdominal imaging [5]. Intra-mural gas can be seen on plain abdominal or pelvic X-rays as curvilinear or circular areas of increased radiolucency in the expected position of the bladder and intra-luminal gas can be seen as an air-fluid level. In our patient, the abdominal X-ray revealed a circular radiolucent rim centrally located within the pelvis which was highly suggestive of emphysematous cystitis. Although she presented with lower abdominal pain, she did not have any other symptoms of cystitis such has hematuria, dysuria, urinary frequency or urgency. Urine dipstick analysis of the urine also did not reveal any evidence of urinary tract infection such as the presence of leucocytes or nitrites. In view of the discordance between her presenting symptoms, urine dipstick analysis and X-ray findings, a CT scan was subsequently performed which revealed that the rim of gas in the pelvis was actually pneumatosis intestinalis of the cecum. The cecum was massively dilated and directed into the pelvis secondary to a closed loop obstruction from a stenotic transverse colon tumor and competent ileocecal valve. We hypothesize that apart from pneumatosis intestinalis of the cecum, pneumatosis intestinalis of the sigmoid colon and rectum could also possibly mimic the X-ray findings of emphysematous cystitis. The subsequent management and disposition of a patient with pneumatosis intestinalis of the colon or rectum will differ significantly with that of emphysematous cystitis.

CONCLUSION

Plain X-ray finding of a curvilinear or circular radiolucent rim at the expected position of the bladder in the pelvis usually represents a diagnosis of emphysematous cystitis. However, it is prudent to investigate with further imaging such as a computed tomography scan if clinical findings are not congruent with emphysematous cystitis as adjacent structures such as the cecum, sigmoid colon or even rectum with pneumatosis intestinalis can mimic the X-ray finding of emphysematous cystitis and if not picked up may lead to delayed diagnosis and disastrous complications.

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CLINICAL IMAGES OPEN ACCESS

Sump syndrome: Endoscopic management of biliary stent induced choledochoduodenal fistula

Anju Malieckal, Kinesh Changela, Zeyar Myint, Sury Anand

CASE REPORT

A 43-year-old female presented with severe right upper quadrant abdominal pain, nausea, vomiting and diarrhea for four days. Three years ago, she was admitted for cholecystitis, cholelithiasis and cholangitis requiring an endoscopic retrograde cholangiopancreatography (ERCP) with plastic stent placement for drainage. Unfortunately, the patient was lost to follow-up after initial plastic stent placement. On admission, she was afebrile and her liver function tests were normal. The ERCP was performed which revealed the distal end of the biliary stent, perforating proximal to the periampullary area causing a large choledochoduodenal fistula (Figure 1). The guidewire was successfully negotiated through the native papilla as well as the fistulous tract. No obstruction, mass or stone was noted. A sphincterotomy was done through the fistula (Figure 2) and through the actual sphincter at the ampulla (Figure 3). The sump was opened, swept clean with removal of abundant debris. Her abdominal pain completely resolved and patient clinically improved within 24 hours after the procedure.

DISCUSSION

As long as the sphincter of Oddi functions normally, bile and enteric contents will not accumulate in the distal common bile duct (CBD). Biliary sump syndrome occurs when accumulation of debris, stones and static bile acts as a nidus for bacterial proliferation, predisposing the patient to cholangitis. Biliary sump syndrome is a rare complication of biliary enteric anastomosis after cholecystectomy [1]. After side to side choledochoduodenostomy, the CBD between the anastomosis and the ampulla of Vater becomes a potential sump (a recess or reservoir serving a drain for liquids) (Figure 4). In this patient, a choledochoduodenal fistula formed due to the biliary stent migrating to the CBD, perforating proximal to the ampulla. The CBD segment between the fistula and ampulla became a sump. There have been cases reported of spontaneous choledochoduodenal fistulas occurring after biliary metallic stent placement [2]. In those cases the cause

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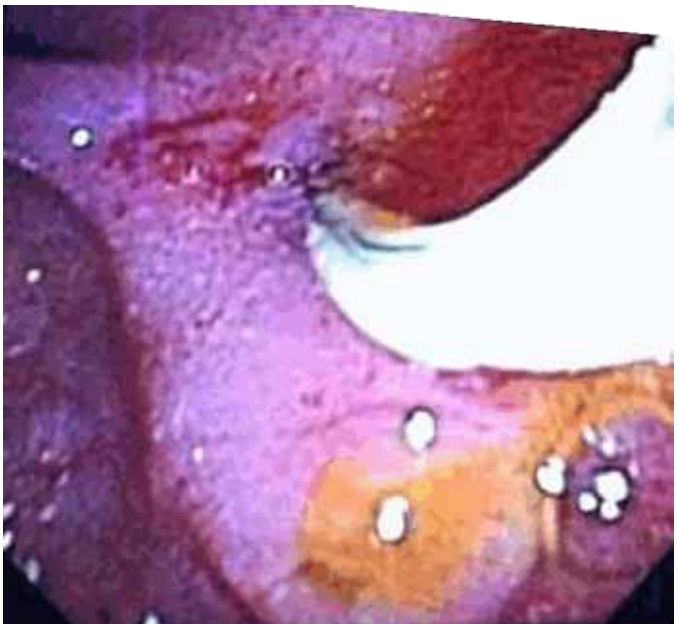

Figure 1: Endoscopy picture reveling distal end of the biliary stent, perforating proximal to the periampullary area causing a large choledochoduodenal fistula.

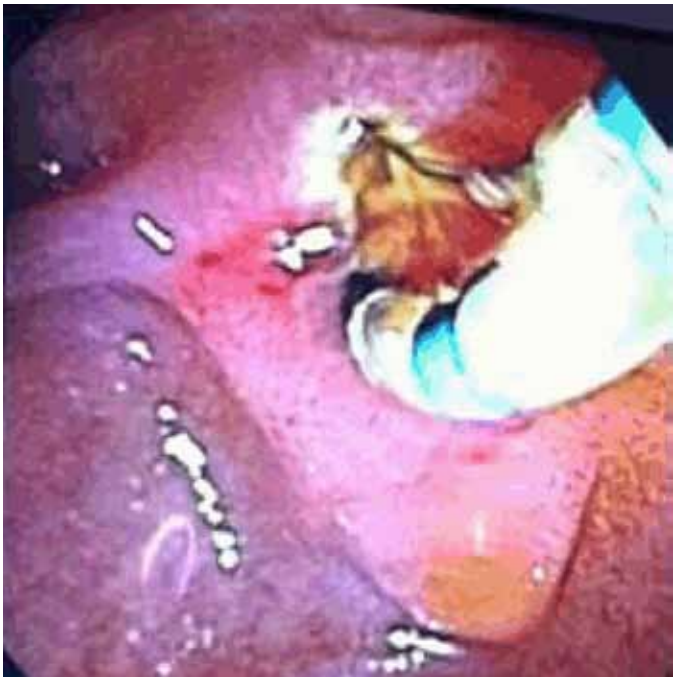

Figure 2: Passage of guidewire through the native papilla as well as the fistulous tract.

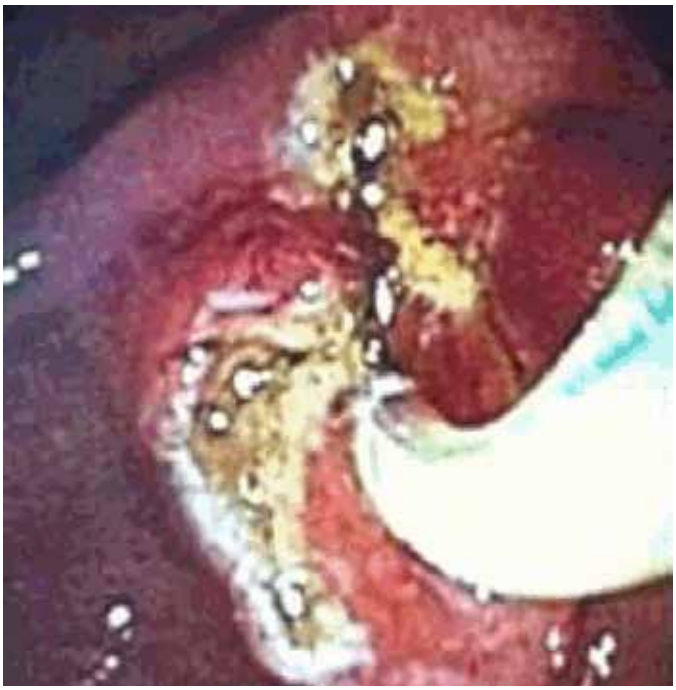

Figure 3: Sphincterotomy through the fistula and through the actual sphincter at the ampulla.

may have been inflammation caused by impacted calculi and food material. In this case, there is no stone or masse noted.

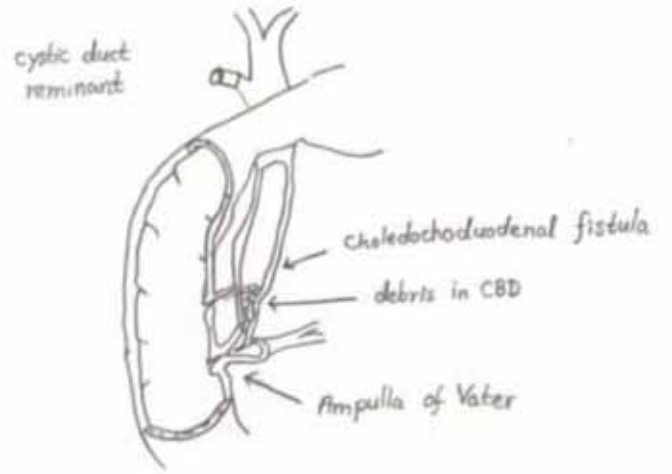

Figure 4: Diagrammatic explanation of SUMP syndrome.

CONCLUSION

The formation of a choledochoduodenal fistula resulting from plastic biliary stent induced perforation is rare. Several cases of biliary stent related perforations secondary to metallic stents have been reported. We report a case in which a spontaneous choledochoduodenal fistula occurred after plastic biliary stent placement, leading to sump syndrome that was treated successfully by sphincterotomy.

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Author Contributions

Anju Malieckal – 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

Kinesh Changela – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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