Case reports

Mucinous cystadenoma of the appendix: a case report

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Introduction

Tumours of the appendix are emerging as diseases of increasing concern due to a rising incidence[1]. We present a case of mucinous cystadenoma of the appendix in an elderly patient. To our knowledge, this is the first report of mucinous cystadenoma of the appendix from Nigeria.

Key Words: Appendiceal tumour, Appendicectomy.

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Case report:

A 74 year old man presented with a one-month history of recurrent episodic dizziness and occasional excessive sweating. The dizziness was precipitated by turning his neck to the right side. He presented to the hospital after the third episode of fainting spells where he was seen at the medical out-patients clinic. Examination revealed an elderly man who was afebrile, and was not pale. His blood pressure was 145/90mmhg; pulse rate was 78/minute, regular, with some hardening of his radial artery. Both lung fields were clear on auscultation His abdomen was flat and soft with a firm sausage-shaped non-tender and non-mobile mass in the right iliac fossa. The clinical diagnosis was caecal tumour; differential diagnosis of carcinoid syndrome. Abdominal ultrasonography revealed a 21cm x 6cm x 7.5cm septate, cystic mass in the right pelvis, and a normal liver architecture. ECG revealed supraventricular extrasystoles. Cervical X-rays revealed prominent osteophytes on the cervical vertebrae. He was subsequently prepared for an exploratory laparotomy. Intra-operative findings included scanty serous ascites, a sausage shaped, firm appendiceal tumour, which was not attached to the surrounding structures as shown in figure below.

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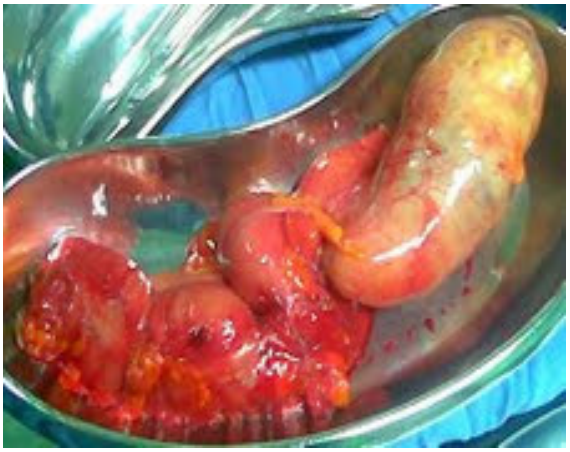
Figure showing Mucinous cystadenoma of the appendix



He had a right hemicolectomy. The tumour weighed 480g. The histology of the appendix was reported as 'showing extensive pools of mucin in a cystic cavity lined by tall columnar epithelium with apical mucin. Features are those of mucinous cystadenoma of the appendix'. His postoperative recovery was delayed due to prolonged ileus. He however resumed bowel function by the 9th postoperative day. His clinical status thereafter was unremarkable, and he was discharged home on the 13th postoperative day.

Discussion

Mucinous cystadenoma is a rare tumour of the appendix associated with cystic dilatation, to which the more general term of mucocele has been applied[2]. Mucocele of the appendix denotes an obstructive dilatation of the appendiceal lumen due to abnormal accumulation of mucus, which may be caused by a retention cyst, mucosal hyperplasia, cystadenoma and cystadenocarcinoma[3, 4]. It is a rare entity found in 0.3% of appendiceal specimens[5, 6],

with a slight female predominance and an average age at diagnosis of over 50 years[7].

Mucocelesare often asymptomatic and are discovered as incidental findings at appendicectomy[6], during laparotomy for another indication, or during histologic examination of an operative specimen[8]. However, it may be diagnosed clinically from features of acute appendicitis[8]. Association with concomitant colon cancer is recognized[9]. A link with irritable bowel disease (IBD) has been reported and IBD is a known risk factor for colorectal neoplasia[10]. It may also be an incidental finding during ultrasonography, computed tomography, and other radiological examinations of the gastrointestinal tract. Four pathological entities are described from the epithelial characteristics:

(1) Simple or retention mucoceles due to obstruction of the appendiceal outflow; usually by a faecolith, characterized by normal epithelium and mild luminal dilatation

(2) Mucoceles with hyperplastic epithelium with mild luminal dilatation. These constitute 5%-25% of mucoceles.

(3) Mucinous adenoma/cystadenoma is the most common form, accounting for 63%-84% of cases. These exhibit mostly epithelial villous adenomatous changes with some degree of epithelial atypia. There is marked distention of the lumen up to 6 cm.

(4) Malignant mucinous cystadenocarcinomas, represent 11%-20% of cases. These demonstrate glandular stromal invasion and/or presence of epithelial cells in the peritoneal implants.[11]

There is always the risk of rupture, either spontaneous or accidental, with consequent development of pseudomyxoma peritonei[2,11, 12], which may present with features of intestinal obstruction[13].

Treatment is usually an appendicectomy; right hemicolectomy is performed when the caecum is involved[8,11,12, 14, 15]. Our patient has been seen for follow-up thrice at the surgical out-patients clinic; initially at 2 weeks post-discharge, then 6 months and then after 1 year. At the time of this report he was in good health.

References

1. Robertson RG, Geiger WJ, Davis NB. Carcinoid tumours. *Am Fam Physician*. 2006; 74:429-434 PubMed .
2. Lo RH, Kan PS. Appendiceal mucinous cystadenoma presenting as "porcelain" appendix with myxoglobulosis- a rare cause of a right lower quadrant mass. *Singapore Med J*. 1998; 39:174-176.
3. Haritopoulos KN, Brown DC, Lewis P, Mansour F, Eltayar AR, Labruzzo C, Hakim NS. Appendiceal mucocoele: a case report and review of the literature. *Int Surg*. 2001; 86:259-262 PubMed .
4. Lee Y-T, Wu H-S, Hung M-C, Lin S-T, Hwang Y-S, Huang M-H. Ruptured appendiceal cystadenoma presenting as right inguinal hernia in a patient with left colon cancer: A case report and review of literature. *BMC Gastroenterol*. 2006; 6: 32.
5. Zagrodnik DF 2nd, Rose DM. Mucinous cystadenoma of the appendix: diagnosis, surgical management, and follow-up. Curr Surg. 2003; 60:341-343.
6. Zullo A, Botto G, Pastormerlo M. A case of appendiceal mucocele associated with cancer of the colon. *Minerva Chir*. 1995; 50:1095-1098.
7. Emmi S, Galasso MG, Ursino V, Scala R, Guastella T. Appendiceal mucoceles. A case report. Minerva Chir. 1998; 53:807-810.
8. Soreide K, Gudlaugsson E, Kjellevold KH. Appendiceal mucinous cystadenoma. Tidsskr Nor *Laegeforen*. 2005; 125:289-291.
9. Pitiakoudis M, Tsaroucha AK, Mimidis K, Polychronidis A, Minopoulos G, Simopoulos C. Mucocele of the appendix: a report of five cases. Tech *Coloproctol*. 2004; 8:109-112 PubMed .
10. Orta L, Trindade AJ, Luo J, Harpaz N. Appendiceal mucinous cystadenoma is a neoplastic complication of IBD: case control study of primary appendiceal neoplasms. *Inflamm Bowel Dis* 2009; 15; 415-421 PubMed .
11. Dixit A, Robertson JHP, Mudan SS, Akle C. Appendiceal mucoceles and pseudomyxoma peritonei. *World J Gastroenterol* 2007; 13; 2381-2384 PubMed
12.
13. Landen S, Bertrand C, Maddern GJ, Herman D, Pourbaix A, de Neve A, Schmitz A. Appendiceal mucoceles and pseudomyxoma peritonei. *Surg Gynecol Obstet*. 1992; 175:401-404.
14. Machado NO, Chopra P, Pande G. Appendiceal tumour-retrospective clinicopathological analysis. *Trop Gastroenterol*. 2004; 25:36-39.
15. Rampone B, Roviello F, Marrelli D, Pinto E. Giant appendiceal mucocele: report of a case and brief review. *World J Gastroenterol*. 2005; 11:4761-4673

Unusual cause of thyroid abscess

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Abstract

Thyroid abscess is a rare condition of the thyroid gland. The common causative organisms responsible for thyroid abscess are *Staphylococci* and *Streptococci* species. We described a case of thyroid abscess due to *Klebsiella pneumoniae* in an infant. The patient was successfully treated with open surgical drainage and appropriate antimicrobial agents.

Keywords: Thyroid, Abscess, *Klebsiella Pneumoniae,* Acute Suppurative Thyroiditis

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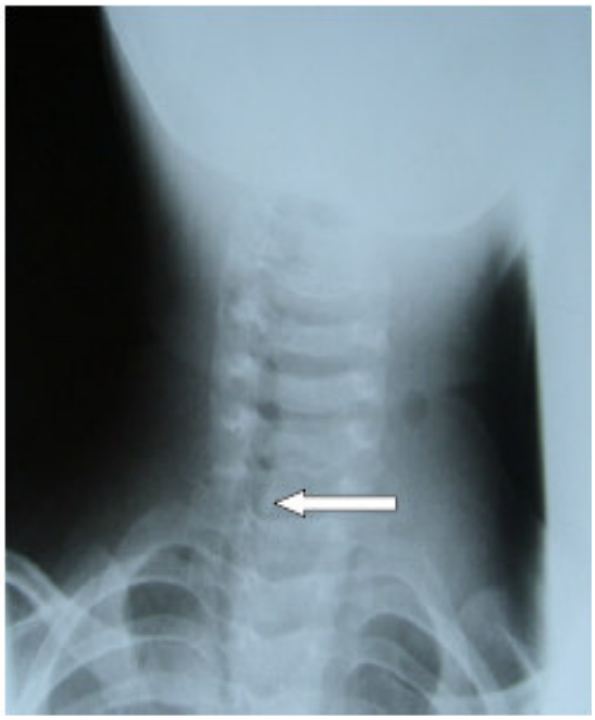
Case report

A 14 month old female child was admitted with a two week history of high grade fever which did not resolve with use of analgesics or anti-malaria drugs, and an enlarging, painful neck mass. Physical examination showed an acutely ill patient with axillary temperature of 38.4°C, tachycardia, a large tender, warm, and fluctuant anterior neck mass with no further extension.

The trachea was shifted to the right. The haemoglobin level on admission was 9.6 g/dl and plain radiographs of the neck and chest in frontal and lateral views showed a homogenous soft-tissue density anterior to the trachea, displacing it to the right. (Figure1).

Figure 1: Plain X-ray of the neck (AP view) showing the displacement of the trachea (arrow)



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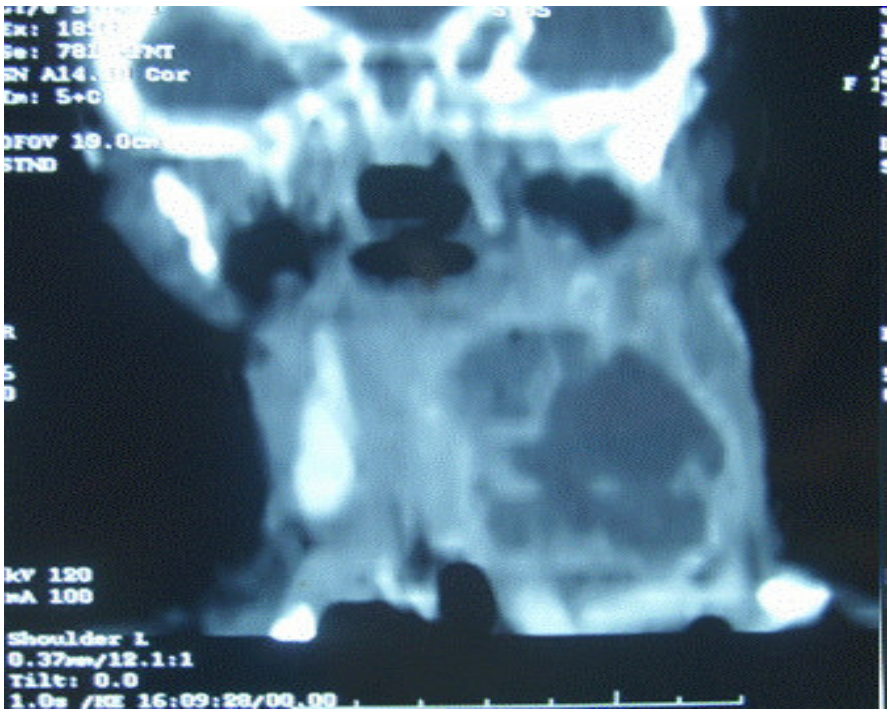
A cystic non homegenous collection in the left lobe of the thyroid gland was seen on computed tomography scan. This extended into the superior mediastinum without any distortion of the mediastinal structures (Figure 2).

Figure 2: CT scan showing a cystic collection in the left lobe of the thyroid gland (arrow)



Thyroid scintigraphy also showed a cold spot in the left thyroid gland and a retroviral screening for Human Immunodeficiency Virus was non reactive. A diagnosis of thyroid abscess was made and she underwent an incision and drainage procedure under general anaesthesia that yielded 150mls of greenish pus. She was treated with intravenous ceftriazone at a dose of 250 mg twice a day and metronidazole at a dose of 90 mg three times a day. Cultures of the drained pus yielded *Klebsiella pneumoniae*. After two week course of intravenous antibiotics; she improved symptomatically and was discharged home 14 days after admission. On review at the out-patient clinic she had sustained clinical improvement.

Discussion

Acute suppurative thyroiditis (AST) is a rare clinical event[1] and an uncommon form of thyroiditis[2]. The progression of the condition to thyroid abscess is equally unusual[3]. Both AST and thyroid abscess represent 0.1 to 0.7% of thyroid lesions managed surgically.[4] A tender thyroid lesion is the hallmark of AST but other causes of a tender thyroid includes de Quervain thyroiditis (the commonest cause of a painful thyroid), acute hemorrhage into a cyst or thyroid nodule, a rapidly enlarging thyroid carcinoma, or radiation thyroiditis [5,6].

The thyroid gland possesses some characteristics that help to make thyroid abscess an uncommon clinical event. These include: total encapsulation of the gland, its secluded anatomic position, an iodine-rich environment, extensive lymphatic drainage, and good blood flow from bilateral anatomising superior and inferior arteries. These provide protection by hindering the invasion of bacteria and its subsequent growth. Haematogenous spread from a distal site of infection is believed to be a common cause of thyroid infection; however the exact infectious source or pathway is frequently unknown. In our patient the exact cause of infection was not known.

Congenital thyroid gland pathology such as pyriform sinus fistula can also lead to acute suppurative thyroiditis[3] other causes includes trauma such as fine-needle aspiration[7] and foreign bodies. None of this was however demonstrated in the index case. AST has also been associated with immunosuppression, especially human immunodeficiency virus [6], however a retroviral screen in our patient was negative. Although

Staphylococci and *Streptococci* have been described as the most frequent causes of AST,[7] many other organisms such as *Aspergillus*, *Brucella*, *Klebsiella*, *Eikenella*, *Salmonella*, and *Acinetobacter* have been identified in infection of the thyroid gland and oftentimes the infection is polymicrobial.[6]

Thyroid abscess have been observed to be more usual in females than males[7] with a wide age range of 16 days to 79 years,[8] with the left side of the gland more commonly involved. Our patient is 14 months old and she developed the lesion on the left side,but the reason for the commoner involvement of the left lobe is not known. Thyroid abscesses usually start after upper respiratory tract, pharynx, or middle-ear infections.[9] Clinical signs include tenderness of the gland, dyspnea, pain, hoarseness, dysphagia, fever, and chills. The unexplained fever which did not resolve despite usage of analgesics or anti-malaria drugs might have suggested the onset of AST in our patient.

Although some patients may be asymptomatic laboratory results indicating infection such as leukocytosis, elevated erythrocyte sedimentation rate and thyroid scintigraphies showing hypo-functional areas with reduced uptake, may also be present.[10] Plain x-rays of the neck may show tracheal displacement, ultrasonography and computerized tomography may identify the underlying structure and extent of the abscess. A fine-needle aspiration to confirm the diagnosis of thyroid abscess and to determine the responsible organism and its antibiotic susceptibility can be performed.

The management of a thyroid abscess is surgical, consisting of incision and drainage, combined with culture and appropriate antibiotic therapy. Broad-spectrum antibiotic therapy covering aerobic, anaerobic, and oral flora should be started early after obtaining a specimen for microbiological studies and this can be changed when sensitivity profile is available. Complications such as destruction of the thyroid or parathyroid glands, internal jugular vein thrombophlebitis, local or hematological spread to other organs, sepsis, and even abscess rupture or fistula formation into the esophagus or trachea can follow thyroid abscess.[9]

The index case was caused by *Klebsiella pneumoniae*. *K. pneumoniae* is a common hospital-acquired pathogen though it may also be a community-acquired pathogen. Only four other cases of thyroid abscess due to K. pneumoniae have been reported in the English literature.[11] The antibiotic treatment should be based on the antibiotic susceptibility testing of individual isolates.

This case exemplifies a rare lesion of thyroid abscess due to an unusual causative agent: *Klebsiella pneumoniae*. It also illustrates the need for high index of suspicion in cases of fever of unknown origin in children and the relevance of excluding less common causes (such as Gram-negative bacilli like *Klebsiella* species) in the management of thyroid abscess.

References

1. Imai, C., T. Kakihara, A. Watanabe, Y. Ikarashi, H. Hotta, A. Tanaka, and M. Uchiyama. Acute suppurative thyroiditis as a rare complication of aggressive chemotherapy in children with acute myelogenous leukaemia. *Pediatr. Hematol. Oncol.* 2002; 19:247-253 PubMed .
2. Tien KJ, Chen TC, Hsieh MC, Hsu SC, Hsiao JY, Shin SJ, and Hsin SC Acute Suppurative Thyroiditis with Deep Neck Infection: A Case Report Thyroid 2007; 17 (5): 467-469
3. Rohondia, O. S., R. S. Koti, P. P. Majumdar. T. Vijaykumar and R. D. Bapat. 1995. Thyroid abscess. J. Postgrad. Med. 41:52-54.
4. Schneider, U., R. Birnbacher, S. Schick, W. Ponhold, and E. Schober. Recurrent suppurative thyroiditis due to pyriform sinus fistula. *Eur. J. Pediatr.* 1995; 154:640-642 PubMed .
5. Farwell AP, Braverman LE 1996 Inflammatory thyroid disorders. *Otolaryngol Clin North Am* 29:541–556.
6. McLaughlin SA, Smith SL, Meek SE 2006 Acute suppurative thyroiditis caused by Pasteurella multocida and associated withthyrotoxicosis. *Thyroid* 16:307–310
7. Nishihara E, Miyauchi A, Matsuzuka F, Sasaki I, Ohye H, Kubota S, Fukata S, Amino N, Kuma K 2005 Acute suppurative thyroiditis after fine-needle aspiration causing thyrotoxicosis. *Thyroid* 15:1183–1187.
8. Jacobs A, Gros DC, Gradon JD Thyroid Abscess Due to Acinetobacter calcoaceticus: Case Report and Review of the Causes of and Current Management Strategies for Thyroid Abscesses Southern Medical Association Volume 96(3), March 2003, pp 300-307
9. Schweitzer VG, Olson NR. Thyroid abscess. *Otolaryngol Head Neck Surg* 1981;89: 226–229.
10. Rohn RD, Rubio T. Neck pain due to acute suppurative thyroiditis and thyroglossal duct abscess. *J Adolesc Health Care* 1980; 1: 155–158.
11. Yu EH, Ko WC, Chuang YC, Wu TJ. Suppurative Acinetobacter baumanii thyroiditis with bacteremic pneumonia: Case report and review. *Clin Infect Dis* 1998; 27: 1286–1290 PubMed .