**Case Report**

**Disseminated zygomycosis presenting as thyroid abscess in a renal allograft recipient**

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**Introduction**

In the current medical environment, thyroid abscess is a clinical entity that is seldom encountered; however, it was a common condition in the era before antibiotics [1,2]. Review of the published causes of thyroid abscess since 1980 demonstrated that although Gram-positive bacteria (*Staphylococcus* and *Streptococcus* species) remain the most common causes, there have been a number of cases in the literature, caused by *mycobacteria*, *Salmonella* species and anaerobes [2]. Fungal thyroid infections are very rare and described in immunosuppressed patients. Immunocompromised patients, such as those with leukaemia, lymphoma, autoimmune diseases and organ-transplant patients on pharmacological immunosuppression, are particularly at risk. Aspergillus is by far the most common cause of fungal thyroiditis [3]. However, zygomycotic thyroiditis is rarely reported in the literature. To the best of our knowledge, there are only four reports in the literature, of disseminated zygomycosis in which thyroid was also involved [4–7]. Possibly, ours is a fifth case in which a live unrelated renal allograft recipient (LURRAT) presented with thyroid abscess antemortem, which was diagnosed at autopsy with disseminated zygomycosis.

**Case**

A 53-year-old male received a renal allograft from his wife in September 2003 for end-stage renal disease (ESRD). He received triple drug immunosuppression along with IL-2 (interleukin) receptor antagonists. The patient had uneventful post-transplant period for 2 months with stable graft function (serum creatinine 1.5–1.7 mg%). Subsequently, he presented with painful swelling, on the right anterior aspect of neck, of 4 days duration, which was associated with low-grade fever without chills or rigors. There was a history of odynophagia along with anorexia for the last 10 days. The urine output was adequate. On local examination there was an 8 × 6 cm tender swelling present in the anterior part of the neck predominantly on the right side. The overlying skin was erythematous and indurated. The swelling was fluctuant and moved with deglutition and protrusion of tongue thereby indicating a thyroid swelling. In addition, a single 1 × 0.5 cm lymph node in the right axilla was also palpable, which was non-tender. Systemic examination did not reveal any significant abnormality. Routine investigations revealed raised blood urea (130 mg/dl, normal range = 20 to 30 mg/dl). Serum creatinine rose up to 3.1 mg/dl from an initial value of 2.4 mg/dl in 2 days duration. Haemogram was normal but for a low total leucocyte count (2300/cumm) a day before the demise. Liver functions and serum proteins were within normal range, however the random blood sugar was 196 gm%. The blood culture sent antemortem was reported sterile.

Radiological examination of the chest showed nodular opacities in left lower zone and right upper zone. These opacities increased in size and multiple new nodular shadows appeared in both lung fields during the hospital stay. The computed tomography (CT) showed multiple cavitary lesions involving both the lungs.

Ultrasound examination of the neck revealed enlargement of right lobe of the thyroid gland, measuring 24 × 27 × 26 mm in size, with a hypoechoic mass lesion having irregular borders suggestive of an abscess. Contrast-enhanced computerized tomography (CECT) of the thyroid revealed an intraglandular...
lesion in the right lobe of the thyroid with surrounding oedema, and the CT density was suggestive of necrotic material. An ultrasound-guided aspiration of the swelling was attempted, which revealed scanty bloody material, hence was not adequate for reporting or culture.

The patient had further deteriorated and terminally had one episode of malena, developed breathlessness (respiratory rate >30/min) and hypotension, for which central line was placed and fluids were given for low central venous pressure. Intravenous antibiotics (vancomycin) and amphotericin were started with clinical suspicion of staphylococcal/fungal pneumonia. However, the patient’s condition progressively deteriorated and the developed a cardio-respiratory arrest from which he could not be resuscitated. A partial necropsy was performed including the removal of the thyroid gland.

**Autopsy findings**

Externally, the diseased was thinly built and emaciated. On opening, the left pleural cavity contained 250 ml of straw coloured fluid.

On gross examination, the thyroid was enlarged, soft and weighed 50 g. Both the lobes and isthmus were swollen and oedematous. On serial slicing, discoloured, haemorrhagic and necrotic areas were seen more on the right lobe. Few preserved areas of thyroid parenchyma were noted in the left lobe. The histological examination revealed large areas of coagulative necrosis without any inflammatory reaction, with focal preservation of thyroid follicles and remnants of colloid. Angioinvasive broad aseptate fungal hyphae with right angle branching were seen within the necrotic and preserved areas, consistent with the morphology of zycomycosis. Many fungal thrombi were also noted. Periodic acid-Schiff and Grocott’s silver stains confirmed the same (Figures 1 and 2).

The transplant kidney, lungs, liver, spleen and small intestine also revealed numerous fungal profiles with fungal vasculitis, angioinvasion and fungal thrombi. Colonies of aspergillus-forming aspergilloma were also noted within the right lung. In addition, Cytomegalovirus inclusions were noticed in right lung, within glomeruli and tubules of transplanted kidney and bilateral adrenal glands.

**Discussion**

The ability of the thyroid gland to resist infection is well-known and infection in the thyroid gland is rare, particularly so with the advent of widespread usage of antibiotics. The remarkable resistance of the thyroid gland to infection is attributed to many factors. A prosperous lymphatic and vascular supply, well-developed capsule, and high iodine content of the gland are various mechanisms suggested to account for this relative resistance to infection [8,9].

Fungal infection of the thyroid is extremely uncommon. Most reported fungal infections of the thyroid have occurred concurrently with systemic infections in immunocompromised hosts, some of which had a pre-existing thyroid disease [10]. The index case had no pre-existing thyroid disease; however it was a post-LURRAT recipient with increased susceptibility to various kinds of infections. A recent review by Goldani et al. [3] described Aspergillus as the most common fungal infection of the thyroid. However, there are few case reports of candida, cryptococcal, coecidioidal and histoplasma thyroiditis as a part of disseminated disease [11–14]. Thyroid involvement by zygomycosis is rarely reported. There have been four case studies in the literature, of disseminated zygomycosis involving the thyroid (4–7, Table 1). All patients succumbed due to disseminated infection and were diagnosed only at autopsy. Of these, one case of Saksenaea vasiformis was immunocompetent and the other three patients were immunosuppressed. These included two patients...
of acute myelogenous leukaemia, myelodysplastic syndrome with desferrioxamine therapy, and one case of HIV infection as underlying predisposing factors.

In renal transplant patients, infections with zygomycetes are an uncommon cause of infection [15]. Common clinical presentations of zygomycosis include the rhinocerebral, pulmonary and gastrointestinal forms. It has also been reported to involve the liver, the genitourinary system and the skin [16]. The Thyroid has not been described as a site of involvement in renal transplant patients. Our case possibly represents the first case of disseminated zygomycosis with thyroid involvement in a post renal transplant patient. Most cases of mucormycosis infection in renal transplant recipients have occurred within a month of treatment for a rejection episode or within 2 months after transplantation [16,17]. The index case also presented within 2 months after renal transplant.

Clinically local signs and symptoms of fungal thyroiditis are indistinguishable from other infectious thyroiditis which included fever, anterior cervical pain, thyroid enlargement sometimes associated with dysphagia and dysphonia. Laboratory features of transient hyperthyroidism due to the release of thyroid hormone from follicular cell damage followed by residual hypothyroidism is described [3]. The clinical presentation in this patient was similar to that of the other reported cases of fungal thyroiditis. However, thyroid function tests were not done in this patient.

Antemortem diagnosis of fungal thyroiditis is made by direct microscopy and culture of a fine-needle aspirate, and/or biopsy in most cases [3]. Unfortunately in the index patient, attempted guided aspiration revealed only blood, therefore cytological diagnosis and mycological culture could not be made antemortem.

At autopsy, histopathologically, fungal hyphae were broad aseptate and showed right angle branching, conforming to the morphology of zygomycosis. However, postmortem culture was not sent in this case, hence proper classification of the fungus was not possible. Since most patients with fungal thyroiditis have disseminated fungal infection with delay in diagnosis and treatment, the overall mortality is high [3]. Fungal thyroiditis is diagnosed at autopsy as part of disseminated infection in a substantial number of patients without clinical manifestations and laboratory evidence of thyroid dysfunction. Treatment usually involves aggressive surgical debridement, in addition to systemic antifungal therapy, and reduction of immunosuppression may prove helpful [18].

In conclusion, to the best of our knowledge the index case described here represents the first reported case of disseminated zygomycosis presenting as acute thyroiditis in a renal-allograft recipient. This observation suggests that the thyroid should be added the list of tissues that can be the site of infection in immunocompromised patients with disseminated Zygomyosis. Since thyroid is an accessible and easy site for aspiration, a cytological examination may help in early diagnosis and treatment.

Conflict of interest statement. None declared.

References


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