CASE REPORT

Thyroid abscess in case of Pre B acute lymphoblastic leukaemia: a rare presentation

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Accepted 11 October 2016

SUMMARY

Thyroid abscess is a very rare clinical condition. It usually occurs in immunocompromised individuals or those with underlying malignancy. We report a case of multiple thyroid abscesses in the patient with Pre B acute lymphoblastic leukaemia which developed secondary to hematogenous spread from pyomyositis of right calf muscle. The patient developed sepsis-associated disseminated intravascular coagulation, which got resolved after thyroidectomy. He became afebrile after surgical intervention. Unfortunately, all the cultures were negative. Since there are few case series and reports, there are no clear guidelines for management of thyroid abscess. We conclude that though rare, thyroid abscess may be the cause of persistent fever in immunocompromised patients.

BACKGROUND

Since thyroid contains iodine (which is an antiseptic), suppurative thyroid infections are rare. There are no clear guidelines on management of such condition as there are few case reports or small case series available. We share our experience and problems faced in balancing therapy for acute lymphoblastic leukaemia (ALL, which made the patient more immunocompromised) and control of infection. Though rare, thyroid gland infections should be kept in possibility when fever is not responding in neutropenic patients.

CASE PRESENTATION

A man aged 18 years was admitted with symptoms of fever on and off for 1 month. On examination, he had severe pallor, hepatomegaly of 2 cm (below right costal margin) and splenomegaly of 2 cm (below left costal margin along the axis of spleen). No lymphadenopathy was observed. Petechial rashes were present over both the lower limbs. Complete haemogram and peripheral blood examination showed pancytopenia with atypical mononuclear cells (20% cells). Bone marrow aspiration (BMA) was performed and was diagnosed as acute leukaemia with 95% blasts. Immunophenotyping confirmed it to be a case of CALLA positive Pre B ALL (HLA DR, CD 10, CD 19 positivity with coexpression of CD10/CD19 in 98% cells). The cerebrospinal fluid study was normal. Cytogenetic analysis was inconclusive as there were no dividing cells, and BCR-ABL was negative. So, complete diagnosis was CALLA positive Pre B ALL/CNS-1/intermediate risk group (according to BFM 2002 for age)/BCR-ABL negative/inconclusive cytogenetics (non-dividing cells).

Therapy was started according to the BFM 2002 protocol. The patient received pre-induction with tab prednisolone for 7 days, and the day 8 blast count in peripheral blood was 0/mm³ (prednisolone good responder). Induction therapy started thereafter. Day 15 bone marrow examination was M1 (morphological remission). On day 25 of induction therapy, he reported pain in right calf. There was no swelling, warmth or erythema of the region. The patient was afebrile initially; however, within 48 hours, his entire right lower limb got swollen and he became febrile. He was neutropenic at that time (total WCC count 290/mm³). Doppler of right lower limb did not show any evidence of deep vein thrombosis. Injection meropenem and teicoplanin were started after sending blood culture. Even though the swelling decreased in size, fever persisted. Over the period of 8 days, patient's condition worsened. There was the appearance of fluctuation in calf. Repeat ultrasonography (USG) of calf was reported as pyomyositis with the presence of air pockets. Incision and drainage of abscess was performed, and injection clindamycin was added. Despite drainage and broad spectrum antibiotics, fever persisted. He developed disseminated intravascular coagulation (platelets 18 000/mm³, prothrombin time 20 s, and fibrinogen 123 mg/dL).

About 15 days after starting antibiotics for calf abscess, he reported pain in the neck and on examination thyroid gland appeared swollen. USG of thyroid was performed, and it was reported to have multiple hypoechoic areas. Contrast-enhanced CT scan of neck was performed, which showed hypodense lesions in both the lobes of thyroid (figure 1). USG-guided aspiration was tried, but only scanty amount of pus was aspirated. As there were multiple abscesses involving both the lobes of thyroid, we had to go for a total thyroidectomy (with transfusion support), for complete removal of the focus of infection (figure 2). The specimen was sent for histopathology examination (figure 3). The patient became afebrile within 24 hours postthyroidectomy and all the coagulation parameters normalised within 3 days.

Chemotherapy was reinitiated after a delay of 4 weeks. Postinduction bone marrow was in remission (day 78 BMA). Currently the patient has completed consolidation therapy.

INVESTIGATIONS

Complete haemogram at presentation: haemoglobin: 5.6 g/dL, platelets: 26 000/mm³, total leucocyte count: 3400/mm³ with 20% atypical mononuclear cells.

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To cite: Kalantri SA, Datta S, Maitra D, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-216523

Figure 1 CT scan neck: enlarged heterogenous lobes of thyroid. Note made of hypodense lesion in both lobes of thyroid. The lesion is thick walled with contrast enhancement.

Figure 2 Intraoperative image: abscess cavity visible.

Figure 3 Gross specimen of excised thyroid gland.

Bone marrow aspirate: acute leukaemia with 95% blasts.
Immunophenotyping from bone marrow aspirate: CALLA-positive Pre B ALL (HLA DR, CD 10, CD 19 positivity with coexpression of CD10/CD19 in 98% cells).
Cytogenetics: non-dividing cells.
CSF analysis: CNS-1 (no blasts and total cell count <5).
Doppler right lower limb: normal venous and arterial flow.
USG calf: pyomyositis with air pockets.

Gram stain calf abscess: no organism detected.
Anaerobic culture calf pyomyositis: no growth.
Aerobic culture calf pyomyositis: no growth.
USG thyroid: multiple bilateral hypoechoic lesions both lobes of thyroid.

CT scan neck: enlarged heterogenous lobes of thyroid. Note made of hypodense lesion in both lobes of thyroid. The lesion is thick walled with contrast enhancement.

Thyroid profile: euthyroid (thyroid-stimulating hormone 2.5 mU/L, T4–8 µg/dL).

Examination of thyroid pus

- ▶ Gram stain: occasional Gram-positive cocci seen.
- ▶ Fungal stain: no fungal elements seen.
- ▶ Acid-fast stain: no acid-fast bacilli detected.
- ▶ Aerobic culture, anaerobic culture, fungal culture, mycobacterium tuberculosis culture: no growth.

All blood cultures sent prior to start of antibiotics and at time of change of antibiotics: no growth.

Serum calcium post thyroidectomy: 6 mg/dL.

Histopathology thyroid gland: extensive areas of haemorrhage, necrosis and fibrosis (figure 4) with the destruction of thyroid follicles by inflammatory infiltrates consistent with thyroid abscess (figure 5). Both viable and degenerated neutrophils were seen in abscess cavity.

DIFFERENTIAL DIAGNOSIS

Thyroid swelling:

1. suppurative thyroiditis,
2. de Quervain's thyroiditis.

Right lower limb swelling:

1. deep vein thrombosis,
2. pyomyositis.

TREATMENT

1. Broad spectrum antibiotics, including Gram-positive coverage.
2. Clindamycin: 4 weeks when air pockets were documented.
3. Incision and drainage of calf pyomyositis.
4. Thyroidectomy: since the patient had bilateral multiple abscess.
5. Calcium and thyroid hormone replacement.

Figure 4 Histopathology of thyroid gland: section shows thyroid tissue and wall of abscess cavity consisting of fibrous tissue, inflammatory infiltrates (neutrophils) and areas of necrosis (HE ×10).

Figure 5 Histopathology of thyroid gland: section shows necrosis and destruction of thyroid follicle by inflammatory cells, predominantly comprising of neutrophils. Degenerated and viable neutrophils can be seen in this section (HE $\times 40$).

OUTCOME AND FOLLOW-UP

The patient is currently afebrile, is in morphological remission and has completed consolidation therapy for acute lymphoblastic leukaemia.

DISCUSSION

Thyroid is relatively resistant to suppurative infections due to high iodine content, anatomical isolation from other neck structures (because of presence of capsule), rich blood and lymphatic supply.¹ Acute suppurative thyroiditis is a rare condition even in patients with impaired host defences.² To the best of our knowledge, there are only two case reports of thyroid abscess in patients of ALL. Da'as *et al*³ described a case of thyroid abscess where the organism isolated was candida. In another case of thyroid abscess in a patient with acute lymphoblastic leukaemia described by Bahat *et al*, no organism was isolated from pus culture; however, Gram-positive cocci were documented on Gram staining as it was in our case. The authors opined that cultures may have been negative as the patient was already on broad spectrum antibiotics.⁴ Similarly, in a case report by Imai *et al* where the authors described two cases of suppurative thyroiditis in children with acute myeloid leukaemia, no organism was isolated from pus culture. Bacterial infections were suspected in these cases because the patients were successfully treated with antibiotics.²

Most common organisms isolated in cases of suppurative thyroid infections are Gram-positive bacteria, especially staphylococci predominate as causative agent in adults and children.⁵ In immunocompromised patients, opportunistic pathogens have been isolated.³ In a case series of 15 children with acute suppurative thyroiditis, pathogenic organisms were isolated by pus culture in 8 (53.33%) cases only.⁶ Thyroid abscesses are generally limited to one lobe. They may be single or multiple. As against majority of cases where abscesses were localised to one lobe, in this case abscesses were present in both the lobes of thyroid making this case a unique one. In present

era with widespread use of antibiotics, suppurative thyroiditis rarely go to the stage of abscess formation.⁷ In a review article by Yu *et al*,⁸ the authors documented that most patients (83.1%) with bacterial infections were euthyroid, whereas those with fungal or mycobacterial infections tended to be hypothyroid (62.5%) and hyperthyroid (50%), respectively. In a series of children with thyroiditis, 93% had normal thyroid function tests, 4% were slightly hyperthyroid and 2% were slightly hypothyroid.⁹ Thyroid function tests were normal in our case, probably supporting bacterial aetiology.

We believe that as the patient was already on broad spectrum antibiotics, we could not isolate any organism. Broad spectrum antibiotics and surgical drainage are pillars of management. Current practice of management is less aggressive with incision and drainage or USG-guided aspiration. We had to go for total thyroidectomy as the patient had disseminated intravascular coagulation and was immunocompromised. Also, the abscesses were multiple and bilateral.

Learning points

- ▶ Though rare, thyroid gland infections should be kept in possibility when fever is not responding in neutropenic patients.
- ▶ Antibiotics alone are not enough for management of thyroid abscess and require prompt surgical intervention.
- ▶ Pyomyositis may require 4–6 weeks of antibiotic therapy in neutropenic patients and may require a prolonged duration of therapy prior to complete resolution of symptoms.

Acknowledgements The authors thank Dr Koel Mitra of Department of Anesthesia, MCH, Kolkata and Dr Kiran Nair, a post graduate trainee, Department of Surgery, for kind cooperation. They also thank Mr Dharmesh Vora for proof reading the manuscript.

Contributors SAK wrote this case report and participated in patient management. MB edited the case report. SD is responsible for HPE images and involved in patient management. DM operated this case.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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