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Amyloid goiter related with Crohn's disease: A rare association Amyloid goiter secondary to Crohn's disease

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ABSTRACT

INTRODUCTION: Amyloid goiter (AG) is characterized by enlargement of the thyroid gland as a result of extensive amyloid deposition in a bilateral and diffuse manner.
PRESENTATION OF CASE: A 58-year-old male patient was diagnosed of Crohn's Disease (CD). He was admitted to our clinic with complaint of respiratory distress and rapid growth swelling in the neck. Ultrasound examination revealed huge multinodular goiter on both sides of thyroid gland. We performed bilateral total thyroidectomy. Pathological evaluation revealed AG.
DISCUSSION: Amyloid leads to degeneration in tissues, thereby disrupts the function of the relevant organs. It is important to distinguish AG from other reasons of goiter, particularly thyroid medullary cancer that can cause amyloid deposition in thyroid gland. Secondary amyloidosis frequently involves thyroid gland at microscopic level, but rarely causes goiter. An analysis of current literature revealed that only few cases of AG occurred secondary to CD. Herein we presented a case of AG who has rapidly growing goiter that associated with CD.
CONCLUSION: AG must be kept in mind in case of rapidly growing goiter, especially in patients with chronic inflammatory bowel diseases.

1. Introduction

Amyloid is an amorphous and proteinaceous material that accumulates in tissue and causes amyloidosis.[1] It may effect a single organ or cause systemic involvement.[2,3] Amyloidosis is classified as primary or secondary. The latter accompany to neoplasms or chronic inflammatory diseases. "Serum Amyloid A" is an acute phase reactant that accumulates in various organs of patients with secondary amiloidosis.[1,4,5] In 1948, Olson et al. reported the first case of secondary amyloidosis caused by Crohn's disease (CD).[6] Amyloid accumulates in thyroid gland in case of amyloidosis or medullary thyroid cancer. Amyloid depots exist in thyroid gland at microscopic level up to 50–80% in patients with systemic amyloidosis.[7] These patients are mostly euthyroid and usually present with pressure symptoms due to rapidly growing thyroid gland.[8–10] Herein, we reported a very rare cause of AG, which developed during the course of CD.

2. Presentation of case

A 58-year-old male patient with intermittent diarrhea for about 20 years, endoscopic biopsy performed to confirm the CD. Symptoms were treated with budesonide and 5-aminosalicylic acid orally. After six months, therapy was discontinued because of drug intolerance. Afterwards, patient was admitted to hospital with rapidly growing swelling of neck, respiratory distress, dyspnea and fatigue. Physical examination revealed rather large thyroid gland as multi-nodular pattern on both lobes (Fig. 1). Thyroid function tests were all within the normal in range. Ultrasonography (USG) examination revealed that the size of the left lobe was 137 mm × 103 mm × 35 mm and right was 119 mm × 76 mm × 43 mm. Thyroid tissue was detected as diffuse heterogeneous and micro-nodular pattern. Multiple nodules were also detected bilaterally with the largest of them is measured 19 mm in size. Cervical and thoracic computed tomography examination showed an excessive growth of the thyroid gland and its lower boundaries extended to the mediastinum. Thyroid scintigraphy showed a heterogeneous distribution of radioactivity in both lobes. Fine-needle aspiration biopsy (FNAB) revealed a benign thyroid tissue. We performed total thyroidectomy. Thyroid tissue was extremely soft and fragile, macroscopically. And right lobe of the thyroid gland was 12 cm × 7.5 cm × 4.5 cm and left lobe was

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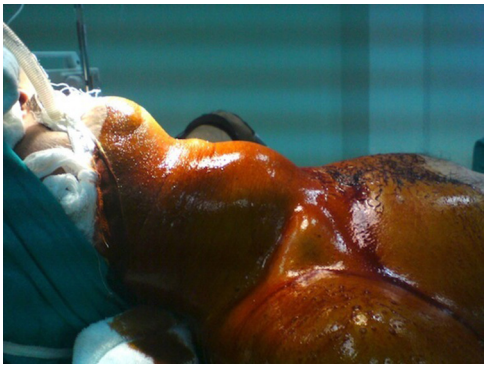

Fig. 1. The appearance of multinodular goiter in the neck.

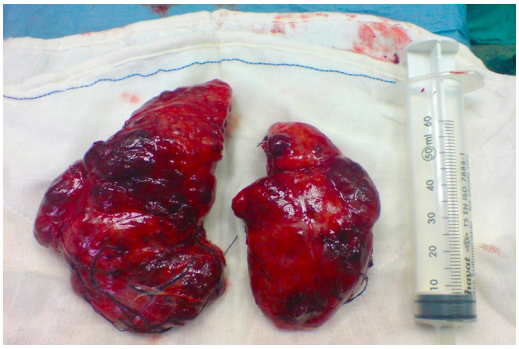

Fig. 2. Macroscopic aspect of thyroidectomy specimen.

14 cm × 10.5 cm × 3.5 cm in size (Fig. 2). There were nodular and diffuse areas in the cross-sectional samples, and the microscopic evaluation revealed extracellular amorphous proteinaceous material widely deposited in the stroma with Hematoxylin and Eosin (H&E) staining (Fig. 3). Crystal violet and Congo red also showed a positive staining (Figs. 4 and 5). There were no malignancies or inflammatory diseases detected, unless CD. Therefore we consider the patient as an amyloid goiter due to CD. Patient discharged second day after operation, without any complications. Amyloid deposition was not observed in biopsy specimen taken during colonoscopy. There were no recurrence or other organ involvement associated with amyloidosis after 24-month follow-up.

3. Discussion

Amyloid is a fibrillar protein that accumulates in various organs and disrupts the function of these organs.[11] It is determined microscopically as an intercellular and semi-transparent material.

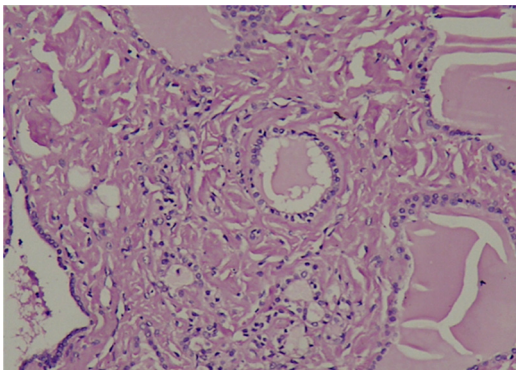

Fig. 3. Extracellular amorphous material is deposited widely in the stroma (H&E).

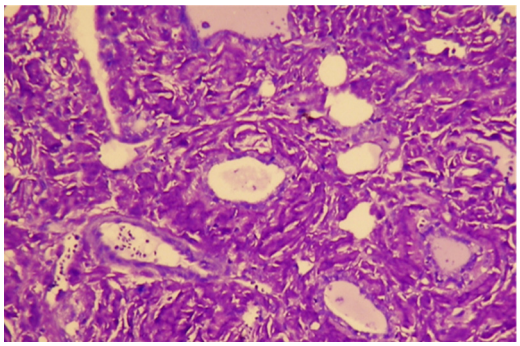

Fig. 4. Crystal violet showed a positive staining for amyloid.

Amyloid has many subtypes that two of them are the major. Despite the similarity in their staining patterns, amyloid is not a single chemical entity. Therefore, it will be more appropriate to describe amyloidosis as a heterogeneous group of diseases characterized by fibrillar protein deposits that have similar staining characteristic.[12,13]

Amyloidosis is classified as primary and secondary. In primary amyloidosis, amount of monoclonal plasma cells increase in bone marrow, similarly immunoglobulin light chains (AL type) increase in the affected tissue. Secondary amyloidosis accompanies malignancies or chronic inflammatory diseases such as tuberculosis, rheumatoid arthritis, ankylosing spondylitis, cystic fibrosis, bronchiectasis and inflammatory bowl disease.[6] Approximately, 75% of patients with secondary amyloidosis that accompanying to CD are male, just as our case.[14–16] It was reported in autopsy studies that amyloid deposition is found in the thyroid gland of 50% of primary amyloidosis and 80% of secondary amyloidosis.[7,8,17] AG is observed in about 0.04% of cases with primary systemic amyloidosis, and these patients have poorer prognosis.[9,18] Symptoms depend on the involved organ.

Amyloid deposition in thyroid gland was first published by Rokitansky in 1855. Beckmann reported a clinically visible thyroid enlargement due to amyloidosis in 1858.[19,20] The association of CD with amyloidosis is a rare entity that first time reported by Moschkowitz in 1936.[14,15,21] Afterwards, Olson et al. reported a case of secondary amyloidosis related to CD, in 1948.[6] Since then, some similar cases and reviews reported by various authors, between 1982 and 2009.[22,23] Greenstein et al. investigated and reported 3050 patients with inflammatory bowel disease in 1992. According to this report, 22 (1.29%) of 1708 patients with CD were complicated by secondary amyloidosis.[24,25] Nevertheless AG is very rare in this group of patients. So far, only few AG cases secondary to CD were reported in the literature.[6,26–29]

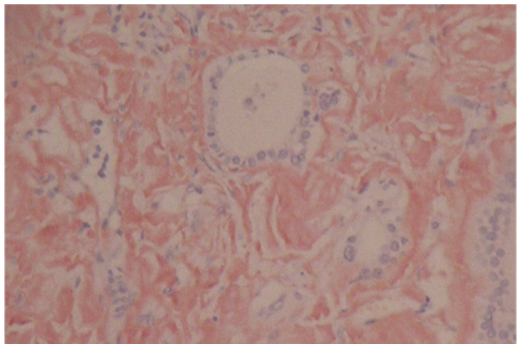

Fig. 5. Congo red showed a positive staining for amyloid.