

Diffuse Nodular Gastritis Revealing Primary Sjogren's Syndrome in Young Woman

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Abstract : Primary Sjogren's syndrome (pSS) is the most common connective tissue disease in the world. Gastric involvement during pSS is estimated at 23% and is by far dominated by chronic atrophic gastritis.

Nodular gastritis (NG) is an exceptional variant of gastritis with annual incidence estimated at 0.18-0.94%. It is usually associated with *Helicobacter pylori* (HP) infection, predominates in the gastric antrum, and exposes to a significantly higher risk of pre-malignant lesions and gastric cancer.

We report the original observation of ND associated to pSS in a 26-year-old Tunisian woman.

Our observation is distinguished by its primary immunological origin apart from any HP infection or lymphomatous transformation, its diffuse character, and its chronology revealing pSS.

Keywords : Nodular gastritis, primary Sjogren's syndrome, gastritis, sicca syndrome.

1. INTRODUCTION

With a prevalence of 1:1000 to 1:100, primary Sjögren's syndrome (pSS), also known as sicca syndrome or Gougerot-Sjogren's syndrome, is the most common connective tissue disease in the world [1]. It is a non-organ-specific autoimmune disease, which is characterized by lymphocytic infiltration of the affected organs and the presence of antinuclear autoantibodies, particularly anti-SSA and/or anti-SSB type [1,2].

His clinic is dominated by glandular manifestations; in particular xerostomia and xerophthalmia, but 25-30% of patients may have extra-glandular involvement: pulmonary, renal, gastrointestinal, neurological, cutaneous, musculoskeletal, and hematological involvement [2,3].

The frequency of gastric involvement during pSS is estimated at 23% and is by far dominated by chronic atrophic gastritis [3].

Nodular gastritis (NG) is an exceptional variant of gastritis [4-6], usually associated with *Helicobacter pylori* (HP) infection [7,8], and predominates in the gastric antrum [6,9].

We report the original observation of a diffuse ND, not associated with HP infection, and revealing pSS in a young woman.

2. CASE REPORT

26-year-old Tunisian female, with no pathological medical history, was explored for chronic epigastralgia and dyspepsia not improved by symptomatic treatment. The gastroscopy showed diffuse nodular gastritis with an aspect of "goose flesh" affecting the antrum, the fundus, and the corpus of the stomach (Figs. 1, 2, and 3). The biopsy showed a diffuse lymphocytic infiltration with organization in lymphoid follicles, without signs of malignancy, nor of *Helicobacter pylori* infection.



Fig.1: Gastroscopy: nodular gastritis of the antrum of the stomach.



Fig.2: Gastroscopy: nodular gastritis of the corpus of the stomach.

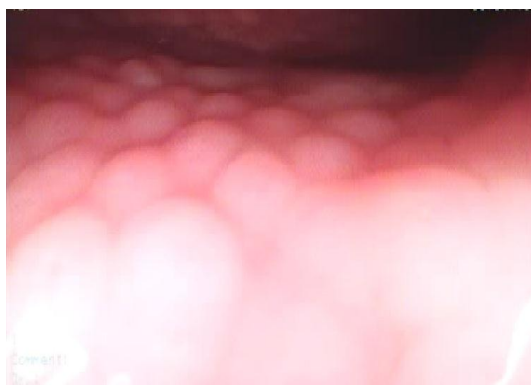


Fig.3: Gastroscopy: nodular gastritis of the fundus of the stomach.

The erythrocyte sedimentation rate was 68mm/H1. The other basic laboratory tests were within normal limits: hemoglobin, leukocytes, platelets, fasting blood sugar, calcemia, ionogram, creatinine, transaminases, muscle enzymes, lipid parameters, reactive C protein, and serum protein electrophoresis. The chest X-ray, abdominal ultrasound, and thoraco-abdomino-pelvic CT scan were without abnormalities.

Subsequent investigations concluded at pSS (abnormal Schirmer test, bilateral superficial punctate keratitis, Chisholm grade 3 lymphocytic sialadenitis, positive anti-nuclear antibodies at 1/320, and positive anti-SSA antibodies). Investigations for other extra-glandular visceral involvement were negative, as were tests for lymphomatous transformation.

Under systemic glucocorticoids (1mg/kg/day), the evolution was favorable with rapid disappearance of digestive functional complaints and normalization of the endoscopic aspect at one month.

3. DISCUSSION

NG is an exceptional endoscopic variant of acute gastritis. Its incidence is estimated at less than 1% in large series: 59 of 32,404 adults who

received endoscopic examination in Nakamura S et al series (0.18%) [4], 185 of 97,262 adult gastroscopies in Miyamoto M et al series (0.19%) [5], and 62 of 6623 patients who underwent endoscopy for abdominal symptoms in Kitamura S et al series (0.94%) [6].

It was initially described in children in association with HP infection, but is currently recognized to occur at any age with a female predominance [4,6]. This gastritis also appears to be associated with a significantly higher risk of pre-malignant lesions and gastric cancer, particularly for diffuse forms of NG [6-8].

Nodular gastritis is exceptional and unusual during pSS. It may later progress to atrophy, especially if associated with *Helicobacter pylori* infection [10].

Chronic atrophic gastritis is the most common and specific clinical presentation of pSS [3,11]. More rarely, other types of gastritis have been reported: hypertrophic gastritis [12], eosinophilic gastritis [13], hemorrhagic gastritis [14], and phlegmonous gastritis [15].

Our observation is distinguished by its nodular endoscopic presentation apart from any HP infection or lymphomatous transformation, its diffuse character, and its chronology revealing pSS. Indeed, gastritis, whatever its endoscopic aspect, remains an unusual presenting symptom of pSS [15].

4. CONCLUSION

As rare as it is, this unusual presentation of primary Sjogren's syndrome deserves to be known by health professionals given the high prevalence of this connective tissue disease.

Likewise, special clinical and endoscopic monitoring is necessary in patients followed for pSS and having NG given the significant risk of gastric cancer, particularly in diffuse forms.

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