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Primary bladder amyloidosis: About a case of incidental discovery

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ABSTRACT

Primary bladder amyloidosis is rare. Hematuria is the most common form of it's revelation. There are no specific clinical and paraclinical signs to differentiate it from urothelial tumors. The diagnosis is histological.

We are reporting you a case of incidental discovery.

Introduction

Amyloidosis is defined as an accumulation in the tissues of an amyloid glycoprotein.[1] Amyloidosis can affect several organs: larynx, tongue, lung, skin, bladder, and the area around the eye.[2] Amyloidosis can be primary (primary), or secondary. It can be systemic, or localized. Primary bladder amyloidosis is rare[3] and was first described in 1897 by SOLOMIN[4] during an autopsy. Macroscopic hematuria is generally the mode of revelation. There is often a diagnostic problem, with a radiological presentation in the form of neoplastic lesions and a deceptive cystoscopy appearance. The diagnosis is histological.

We are reporting a case of primary bladder amyloidosis of incidental discovery.

Observation

This is Mr. M.L, a 55-year-old patient with a medical history of: type 2 diabetes treated with oral anti-diabetes drugs, active smoking with 10 packs/year weaned off in 1991, trauma of the left lower limb treated by external fixator in 1982.

He had been referred for urological consultation, for the management of bladder thickening found on thoraco abdominal pelvic computed tomography scan (TAP CT SCAN). The TAP CT SCAN was requested, during the etiological assessment of a pulmonary embolism, on deep venous thrombosis of the external iliac vein. He was treated by APIXABAN.

On examination there were no urological symptoms, the clinical examination was normal. The TAP CT SCAN requested as part of the etiological assessment of pulmonary embolism, revealed an irregular thickening of the bladder (Fig. 1).

A cytobacteriological testing of urine carried out had isolated an *Escherichia coli* which was treated.

A bladder fibroscopy had been found: a yellowish cardboard infiltration aspect under the mucous membrane very suspicious of the bottom of the dome, and of the lateral faces of the bladder (Fig. 2).

Biopsy specimens, and a urine sample for urine cytology, had been taken.

The result for urinary cytology was: acute inflammatory product in the presence of reactive changes.

And that of the biopsy was: subacute inflammatory changes in the presence of focal keratinized squamous metaplasia. Absence of obvious malignancy.

A transurethral resection of the bladder was programmed and performed, to complete the management.

The result of the histological examination, of the resection chips was as follows: histological appearance in favor of a parietal amyloidosis, which may correspond either to a localized amyloidosis of the urinary system, or to a generalized amyloidosis.

A typing of amyloid deposits was requested. New chips were then collected, frozen and sent in dry ice. The sample was studied in full after staining according to HES, Congo red, and in immunofluorescence (anti-Kappa, lambda, SSA, TTR and fibrinogen antibodies)

The result was as follows: bladder localization of amyloidosis, whose typing in immunofluorescence is in favor of AL amyloidosis, predominantly Lambda (Fig. 3).

The patient was finally referred to internal medicine for a full check-up of amyloidosis. There were no other organs affected by amyloidosis, nor any associated homeopathy.

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Abbreviations

TAP CT SCAN	thoraco abdomino pelvic computer tomography
HES	HEMATEIN EOSIN SAFFRON
SSA	Senile systemic amyloidosis
TTR	Transthyretin

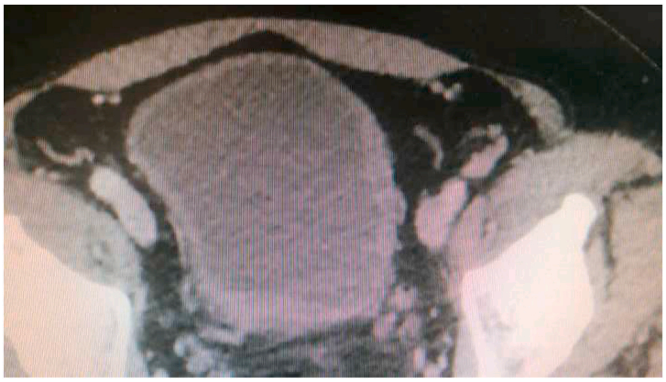

Fig. 1. TAP CT SCAN: irregular thickening of the bladder.

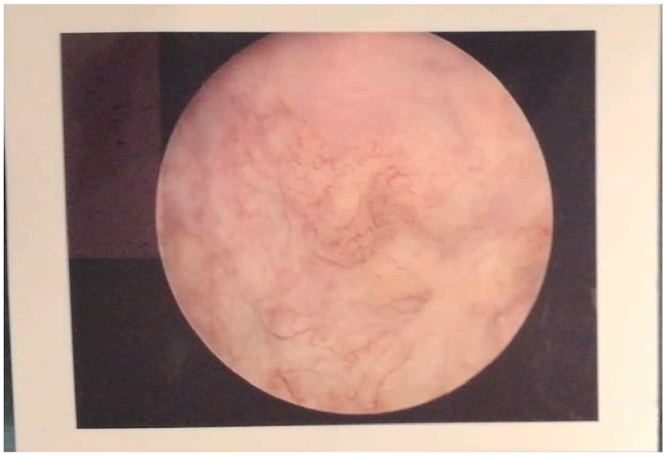

Fig. 2. Bladder fibroscopy: a yellowish cardboard infiltration aspect under the mucosa suspected.

Comments

Bladder amyloidosis is rare. It may be due to primary amyloidosis, or secondary amyloidosis.[1] Primary amyloidosis is a systemic disease related to dysglobulinemia, including multiple myeloma. Secondary amyloidosis is associated with an inflammatory disease, most commonly rheumatoid arthritis.[1,2] In our patient, it was difficult to predict before the paraclinical results, what type of amyloidosis it was, since he had no history of dysglobulinemia or inflammatory disease in his background. In the literature, there are two theories that may explain the occurrence of bladder amyloidosis: an abnormality in protein metabolism on the one hand and on the other hand, a chronic infection of the bladder mucosa[5]

The clinical signs of bladder amyloidosis are non-specific: Hematuria, Dysuria, Pollakiuria, Acute retention of urine.[1] These signs may suggest an urothelial tumor in the first line. Tirzmann et al.[4] found that 77% of their patients had hematuria, and that the average age of onset

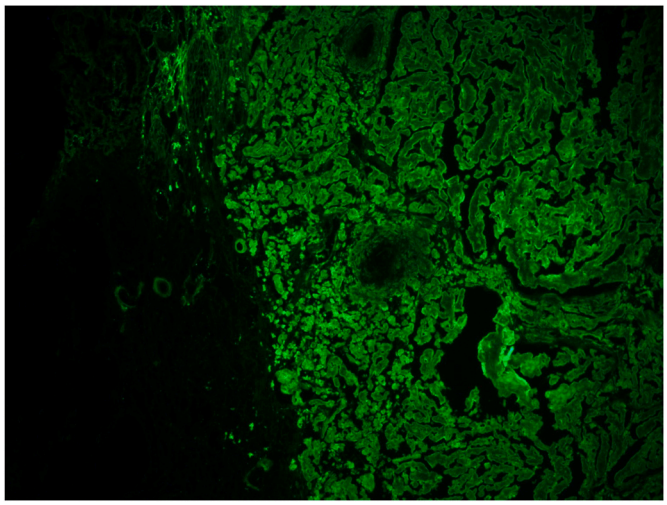

Fig. 3. Histology: bladder localization of amyloidosis, whose typing in immunofluorescence is in favor of AL amyloidosis, predominantly Lambda.

was 55 years. The peculiarity of our presentation is the absence of clinical signs, the discovery being fortuitous, during the etiological assessment of a pulmonary embolism.

A TAP CT SCAN requested had found: irregular thickening of the bladder. Liwen zhao,[5] found the same lesions in her patient. Adil,[2] for his part, found nodular lesions on the posterior surface of the bladder in his patient on the CT scan.

Cystoscopy also finds non-specific signs, which can make one think of a bladder tumor: erythematous, protuberant, polyploid lesions.[1,2,4] In our patient, we noted rather: an aspect of yellowish cardiac infiltration, under mucous membrane, very suspicious of the bottom of the dome and the lateral faces of the bladder.

Histology is therefore the only way that allows us to differentiate a bladder tumor from vesical amyloidosis.

The histological examination will show amyloid deposits in the submucosa which can reach the muscularis.[1] The identification is done with the Congo red coloring, which differentiates the amyloid substance from other hyaline substances, such as fibrin and collagen.

It is then necessary to do a typing of amyloidosis, using direct immunofluorescence techniques, with the use of specific antibodies: (Ac anti light kappa and lambda chains, Anti-SAA, anti lysozyme)

In our case, localized AL amyloidosis was found, predominantly lambda.

Conclusion

Bladder amyloidosis remains a rare pathology, with non-specific clinical and radiological signs. The diagnosis is histological.

Asymptomatic bladder amyloidosis is rare. No cases had been published so far.

Author contributions

Komi Hola SIKPA: Conceptualization, writing, Agathe BERNARD: Data curation, investigation, Denis SEGUIER: Methodology., project administration, Chamseddine CHAABANE: Methodology; Visualisations, Samuel MAKKE: supervision; visualization, Philippe DANJOU: Review.

Declaration of competing interest

None.

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