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RESEARCH ARTICLE

INFLAMMATORY MYOFIBROBLASTIC TUMORS IN THE PECULIAR CONTEXT OF THE POSTPARTUM CASE REPORT

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Abstract

The debate persists about the inflammatory or tumoral, benign or malignant nature of the Inflammatory myofibroblastic tumors (TMI). Radioclinically, they can mimic a malignant neoplasm, although they are classified as "intermediate" by the World Health Organization (WHO). The diagnosis is almost always made by pathological examination. The treatment is poorly codified but the management is usually surgical. The removal of these tumors is a challenge when the mass is developed at the expense of or in the vicinity of noble organs. Several therapies have been tested to overcome the limitations of surgery, the results are variable. These tumors rarely affect the digestive tract. We propose the study of a case of myofibroblastic tumor developed at the expense of the omentum, five months after a cesarean section, in order to try to determine if it is a rare complication of the postpartum or a fortuitous combination.

Introduction:-

Inflammatory myofibroblastic tumor (IMT) is a relatively rare soft tissue tumor. The reactive versus neoplastic pathogenesis of this tumor is unresolved. Ubiquitous, they were initially described in the lungs. IMT are neoplastic proliferations that show a predilection for the visceral soft tissues of children and adolescents and have a tendency for local recurrence, but only a small risk of distant metastasis.Various pathogenetic backgrounds have been proposed as initiating factors such as infections, autoimmune and neoplastic processes, but the etiology of most remains unknown. Radiologically and clinically, they can mimic a malignant neoplasm, while they are classified in the “intermediate” category by the World Health Organization. The diagnosis is almost always made on the pathological examination. Histologically we find a variable amount of fibroblasts, myofibroblasts,lymphocytes, histiocytes, plasma cells, mast cells and foamy macrophages. We count varying modalities of treatment but the management is usually surgical. The tumor’s resection is a real challenge when the mass has developed at the expense of or in the vicinity of noble organs. Several therapies have been tested to overcome the limitations of surgical treatment, with variable results.

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Observation :-

A 39year-old patientpresented 5 months after the cesarean section a case of peritonitis. Physical examination on admission found a patient with fever and tachycardia. Abdominal examination revealed a distended abdomen with a painful mass in the left flank.Biological assessment found : Hemoglobin at 11.2 g/dl, leukocytes high at 19,500 U / mm^3, C-reactive protein high at 150 ;the rest of the examination is within normal limits. An ultrasound was performed revealing the presence of an intraperitoneal collection on the left flank measuring 9x4.8cm, this collection communicates with anotherparietal collection measuring 6x3 cm. An emergency laparotomy reveledamass located on the omentum measuring 12 x 6 cm. The tumor was completely resected. With uneventful postoperative course, the patient was discharged after 3 days. The pathological report found an omentum formation weighing 128g and measuring 12x5.5cm. Macroscopically,the lesion was whitish, measuring 8x4 cm. The histological examination of the various samples taken showed a spindle cell tumor proliferation including fatty lobules. Spindle-shaped cells have a fairly monomorphic elongated spindle-shaped nucleus, achieving a storiform appearance. Presence of loose edematous areas and dense lymphocyte clusters (Figure 1).

With the immunohistochemistry positivity of anti caldesmone antibodies in a focal manner, anti AML antibodies diffusedly positive on the fusiform component (Figure 2) and anti AE1 AE3 antibodies heterogeneously positive on the fusiform component as well; the anti CD34, anti PS 100, anti CD 117, anti DOG1 and anti ALK antibodies were all negative (Figure 3). The immunohistochemical and morphological study makes it possible to orient the diagnosis towards an inflammatory myofibroblastic tumor.

Magnetic resonance imaging(MRI) was performed one month after surgerywith the reappearance of pelvic pain revealing a parietal infiltration measuring 75x38x85mm with underlying peritoneal extension which may be related to a myofibroblastic tumor. The patient was presented at a multidisciplinary consultation meeting, where the decision was to prescribe anti-inflammatory COX-2 inhibitors for 3 months then to perform a follow-up computerised tomography (CT) after 6 months. The patient responded well to medical treatment; radiologically, an abdominal CT was performed after 6 months showing mass regression measuring 2 x 2 x 1.5 cm. After a one year follow-up, the patient remained asymptomatic no longer showing any discomfort or pain, no longer showing any discomfort or pain without treatment and a disappearance of the lesion on the control MRI finding only postoperative alterations of the abdominal wall.

Discussion:-

IMT constitute a heterogeneous group of lesions initially reported in the lung, which can affect multiple organs [1]. In the abdomen, the mesentery and the omentum are the most common locations[2]. It is a rare tumor that can be seen at any age, especially in young people, with more than 50% in the 30 years-old or younger group [3]. Itcan bepost-traumatic, it can follow an infectious episode, chemotherapy, an irradiation, or surgery like inour patient case, whose diagnosis was made 5 months postpartum of a cesarean [4]. Functional signs are usually chronical, but several studies report an acute evolution, with a complication revealing the tumor, such as fever, severe abdominal pain, digestive hemorrhages[5]… On ultrasound, the lesion appears rather well limited, heterogeneous hypoechoic, with the presence of posterior reinforcement. Its scannographic appearance is variable, the lesion appears hypodense or isodense spontaneously with an inconstant, homogeneous or heterogeneous and often late enhancement. An area of central necrosis and calcifications are sometimes associated. On the MRI, IMT is hypointense in T1 sequence, hyper-intense in T2 sequence with heterogeneous enhancement after gadolinium injection, MRI therefore proves to be a good monitoring tool, mainly for inoperable tumors [6]. Depending on the series, the size varied from 0.2 to 20 cm, in our case the size was 8 cm. Macroscopically, the mass appears whitish, firm when cut, with the presence of hemorrhagic, necrotic or even calcifications changes. In microscopy, there is a component of spindle-shaped cells that are not atypical of the myofibroblastic type, more or less fasciculated, in a hyaline stroma, associated with numerous inflammatory lympho-plasmacyt cells with little mitotic activity. The tumor is positive for AML and cytokeratin to a varying degree. The expression of the ALK gene, although not specific to TMI, is observed in about 50% cases. The ALK gene is involved in the pathogenesis of this lesion. The presence of clonal rearrangements of this gene in IMTs of children and young adults is in favor of its tumoral nature rather than a reactive one. The immunohistochemical study reveals the great biological plasticity of myofibroblasts which can express vimentin, actin or desmin [7]. In our case, the tumor was positive for anti AML antibodies (Figures 1 and 2), as well as for anti caldesmone antibodies and for anti AE1 AE3 antibodies, and negative for the rest of the antibodies tested, in particular ALK (Figure 3). The etiopathogenesis of IMT remains controversial, several causes are mentioned, in particular post-traumatic, autoimmune, infectiousor tumoral. The inflammatory nature is suspected due to the history of infection or inflammation, particularly postoperative, as is the case for our patient and the regression under

antiobiotherapy and anti-inflammatory drugs. Regarding the reaction hypothesis, several studies have reported an association with viral infections; thus the systemic signs observed would be related to the secretion of pro-inflammatory cytokines. Postoperative spindle cell nodules of the urogenital mucous membranes and inflammatory ganglionic pseudotumors secondary to mycobacterial infection fall within this scope. The HHV8 virus has recently been implicated. Its genome would code for proteins analogous to cytokines, oncoproteins and inhibitors of apoptosis. An association with EBV [8], detected by immunohistochemistry or by in situ hybridization, has been described in hepatic, splenic and lymph node locations. Tumor nature is suspected in the face of recurrence, infiltration potential, multifocalite or vascular invasion. From 1999, Dr Griffin and Hawkins's team have demonstrated non-random abnormalities of the 2p23 chromosomal band with translocations and rearrangements involving the ALK gene have been reported and are responsible for a constitutive overexpression of an ALK protein (tyrosine-type receptor–Kinase), detected by immunohistochemistry or in situ hybridization methods. However, only a few IMTs exhibit these clonal abnormalities. In our case, our tumor was also ALK negative [7,9]. Complications of IMT are uncommon, mainly represented by local recurrences and more rarely by metastases or malignant transformation, hemorrhagic complications are exceptional. Factors in favor of aggressive forms include large size, presence of necrosis, aneuploidy and p53 expression [10]. Regarding the treatment, the gold standard remains complete surgical resection. In this particular case, we suspect the participation of hormonal factors,since the tumor occurs in a postpartum context despite the absence of hormonal receptors in immunohistochemistry. Indeed, the literature review found, to our knowledge, only four cases of extra uterine IMT in a postpartum context: a mesovarian site IMT on day 1 postpartum, a liver IMT revealed on day 4 postpartum, a case of pulmonary IMT discovered 5 months postpartum, and a case of breast IMT also in a postpartum context. Three other cases have been described in pregnant women [11], and 28 cases of placental associated IMT with apparently benign course [12].With the largest case study on the matter so far (9 cases), Makhdoum and al raise an interesting question about placental and uterine IMT compelling they may be a subtype within the group of uterine IMT with specific molecular characteristics which may be related to the hormonal milieu[12].These studies have corroborated our hypothesis stating that hormonal changes during and after pregnancy may be one of the underlying causes of IMT in the per and postpartumperiod.

Figures

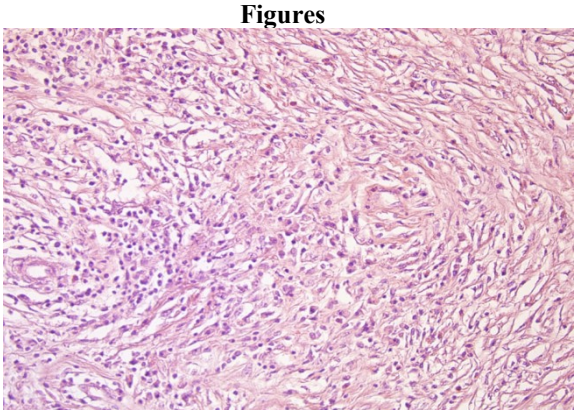

Figure 1:- Fusocellular tumor proliferation related to an inflammatory myofibroblastic tumor.

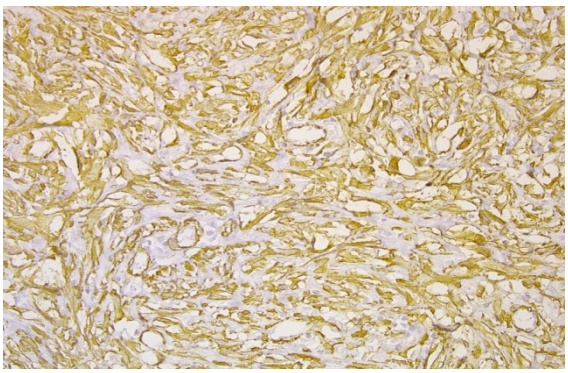

Figure 2:- Immunohistochemical expression of anti-AML antibody cytoplasmic and membrane staining in the inflammatory myofibroblastic tumor.

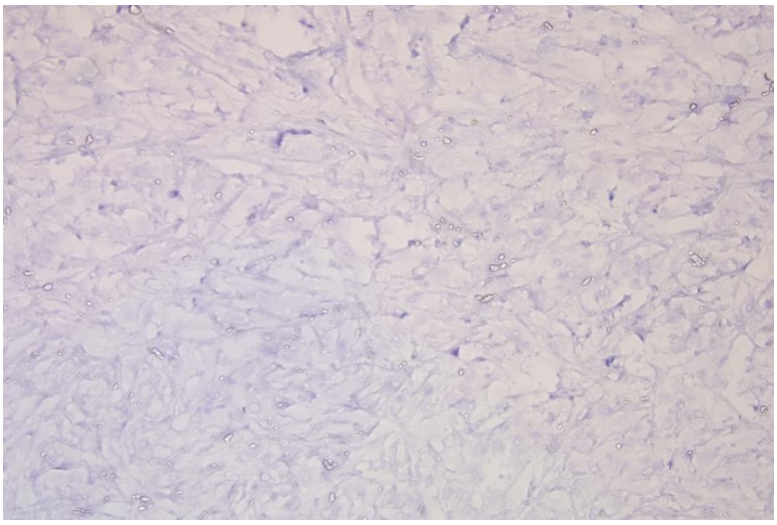

Figure 3:- Negative immunohistochemical expression of anti ALK antibodies in the inflammatory myofibroblastic tumor.

Conclusion:-

IMT is a rare ubiquitous tumor with a still unknown etiopathogenesis. Although the cases of IMT developed in a postpartum context and reported in the literature are exceptional, this rarity seems relative and raises the hypothesis of the influence of hormonal factors. This role nevertheless remains to be verified on larger series.

Declaration of interest:
The authors declare that they have no conflict of interest.

Author contributions :
All authors have participated in the patient's diagnosis and therapeutical management, and have partaken in drafting and reviewing the article.

The anatomopathological study was performed by Dr Asmaa El Kebir, and Professeur Mehdi Karkouri, head of department of Pathology.

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