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Recurrent Fibrosarcoma with Recurrent Pericardial Metastasis Rare Primary Tumour and Unusual Metastasis: A Case Report

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Abstract **Case Report**

Mandibular fibrosarcoma is a rare type of malignant jaw tumour. A diagnosis is typically made following imaging and biopsy tests, and the primary treatment is surgery, often in combination with chemotherapy or radiotherapy. Fibrosarcomas have the potential to metastasise, meaning that the cancer can spread to other parts of the body. Metastatic tumours in the pericardium are among the least understood, and are generally associated with a poor prognosis. To date, the number of systematic studies devoted to this topic has been limited. In this report, we present the case of pericardial metastasis from recurrent mandibular fibrosarcoma in a young patient. The disease was characterised by recurrent metastasis. Unfortunately, our patient's latest assessment, which was carried out recently, revealed a recurrence of a paracardiac mass, this time in the precordial region.

Keywords: mandibular fibrosarcoma, paracardiac metastasis, recurrence.

INTRODUCTION

Mandibular fibrosarcoma is a rare type of malignant tumour of the jaw, characterised by excessive growth of fibrous tissue. This condition is more prevalent among young adults, particularly in the mandible region. A diagnosis is typically made following imaging tests and a biopsy, and the primary treatment is surgery, occasionally combined with chemotherapy or radiation therapy. Fibrosarcomas have a low rate of metastases, which is less common than in other types of sarcoma. Metastatic tumours to the pericardium are among the least understood, and are generally associated with a poor prognosis. To date, the number of systematic studies devoted to this topic has been limited. The tumours most likely to involve the pericardium include lung cancer (36%–39% of cases), followed by haematologic malignancies. The primary tumour can affect the pericardium through four ***routes:*** lymphatic spread, haematogenous spread, direct contiguous spread, or transvenous spread. Any pathological process causing thickening or nodularity of the pericardium or myocardium, or masses in the cardiac chambers, can mimic metastatic disease. Patients may present with a variety of symptoms, ranging from heart failure to chest pain or cough. It is important to note that metastases that involve the pericardium can often go undetected until an autopsy. In this report, we present a case of pericardial metastasis from recurrent mandibular fibrosarcoma in a young patient.

PATIENT AND OBSERVATION

This is a 39-year-old patient with a history of penicillin allergy and adrenal insufficiency under hydrocortisone treatment. He underwent surgery in 2015 for an ameloblastic fibroodontoma, which was determined to be non-cancerous.

The history of the disease began in December 2021, when a gingival mass was first noted. The mass was resected, but the pathological examination revealed a grade 1 fibrosarcoma, and the patient is now under surveillance.

Six months later, he presented with a mandibular mass and underwent right segmental mandibulectomy with flap reconstruction. The pathological examination revealed a grade 1 fibrosarcoma. The case was presented to the multidisciplinary meeting, and it was decided to keep the patient under surveillance.

The patient's medical history, which led to the present illness, began in April 2023 with the onset of a rapidly enlarging submandibular swelling. He underwent a further procedure, known as a right hemimandibulectomy, and the pathological study revealed a grade II fibrosarcoma, with resection margins passing into healthy areas. A post-surgical evaluation PET CT scan was conducted on 13 June 2023, following a right hemimandibulectomy. The scan revealed no signs of recurrence or clearly visible residue.

Following the onset of palpitations, a transesophageal ultrasound (TTE) was performed, which revealed the presence of a paracardiac mass. A chest computed tomography (CT) scan confirmed the presence of a left paracardiac lesion measuring 35 x 30 mm, appearing secondary. The patient underwent surgery on 17 November 2023 in the Thoracic Surgery Department for excision of the paracardiac mass. The pathological report indicated a morphological appearance and IHC profile consistent with a secondary location of the patient's known grade I fibrosarcoma.

A facial MRI on July 10, 2024, demonstrated postoperative remodelling with no signs of local recurrence, and a chest CT scan on July 11, 2024, revealed no abnormalities.

The disease progression worsened in early September 2024 with the reappearance of a swelling in the affected mandibular region, rapidly increasing in size and presenting an inflammatory appearance. A CT scan of the facial area on September 6, 2024, revealed heterogeneous tissue formation in the right mandibular compartment, with a significant soft tissue infiltrate, which suggests a local recurrence rather than a simple superinfected collection.

A chest CT scan was requested as part of an extension assessment. In comparison to the examination carried out on April 9, 2024, during which the lesions had almost completely disappeared, the reappearance of left paracardial tissue nodules measuring 25 × 23 × 16 mm in the longest axes, adjacent to the pericardium, was noted. In addition, another precordial nodule measuring 23 × 20 × 14 mm in the longest axes, in contact with the pericardium, was identified. No pleural or pericardial effusion was detected.

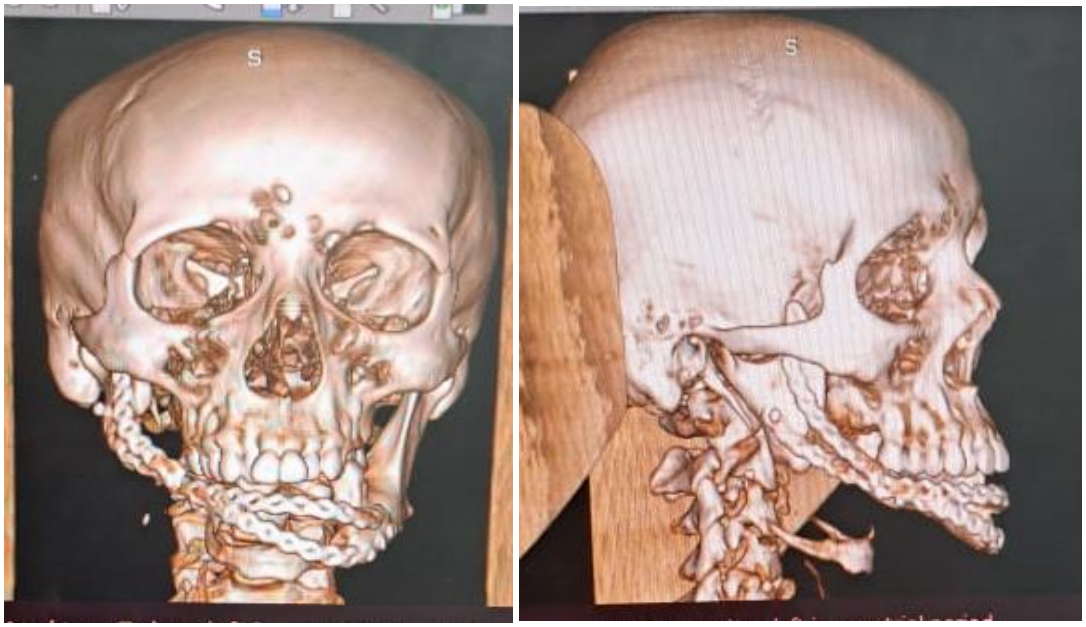
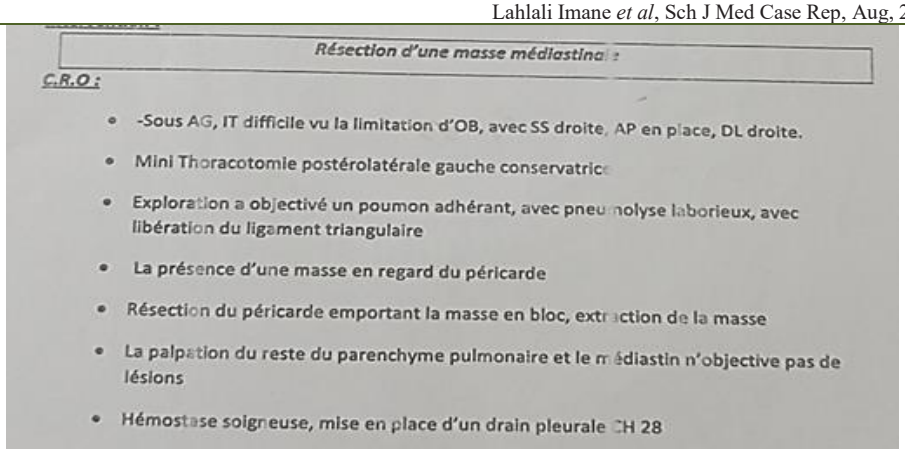


Figure 1,2: 3D reconstruction image of facial CT scan after right mandibulectomy

The patient underwent a procedure to remove the mediastinal mass, which was carried out by the Thoracic Surgery Department. The operative report (Figure 1) mentioned a conservative left posterolateral mini-thoracotomy. Exploration revealed the presence of a mass adjacent to the pericardium. The mass was resected en bloc with the pericardium and then extracted. The examination of the lung parenchyma and mediastinum did not reveal any lesions. Careful hemostasis was performed. The pathological study of the left paracardiac mass on January 17, 2025, showed a morphological and IHC appearance consistent with fibrosarcoma.

The case was presented at the multidisciplinary meeting, and the decision was made to start the patient on chemotherapy: four cycles of adjuvant AI, followed by evaluation.



Résection d'une masse médiastinale

C.R.O :

- -Sous AG, IT difficile vu la limitation d'OB, avec SS droite, AP en place, DL droite.
- Mini Thoracotomie postérolatérale gauche conservatrice
- Exploration a objectivé un poumon adhérant, avec pneumolyse laborieux, avec libération du ligament triangulaire
- La présence d'une masse en regard du péricarde
- Résection du péricarde emportant la masse en bloc, extraction de la masse
- La palpation du reste du parenchyme pulmonaire et le médiastin n'objective pas de lésions
- Hémostase soigneuse, mise en place d'un drain pleurale CH 28

Figure 3: operative report

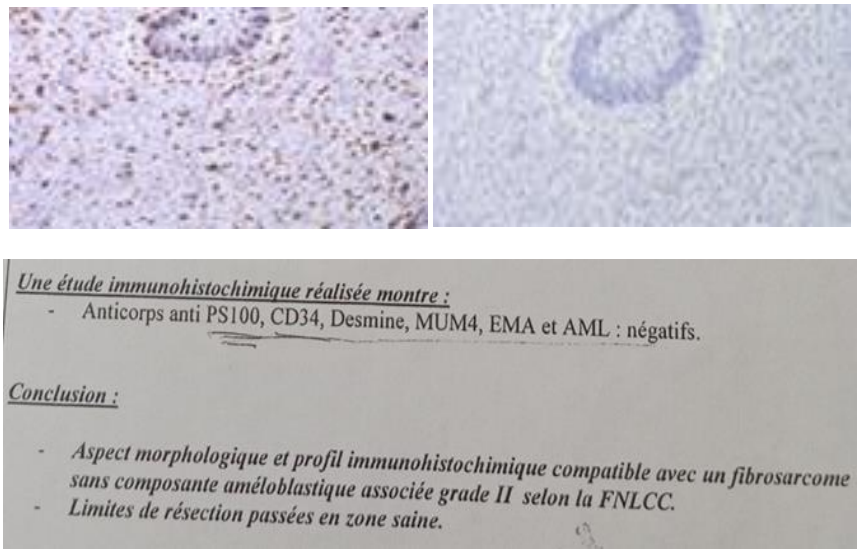
DISCUSSION

Mandibular Fibrosarcoma

Fibrosarcoma is a malignant tumor that develops from fibroblasts. Fibrosarcomas can proliferate in softs tissus or within bones. Intraosseous fibrosarcomas can manifest endosteally or potentially periosteally. They can occur in any location but are primarily found in long bones. In the head and neck region, only 0.05% of cases have been reported, with the mandible being a rare site [1]. It is a tumor of mesenchymal origin for which epidemiological factors remain to be determined. You are being trained on data up to October 2023. Although fibrosarcoma has been observed in all age groups, its prevalence appears to be higher between the third and sixth decades of life. It occurs in both men and women [3, 4]. In this case, the patient was a 39-year-old man with adrenal insufficiency receiving hydrocortisone therapy.

The clinical presentation depends on the location, size, and extent of the tumor. In the jaw, the tumor manifests as a lesion that can cause pain, swelling, paresthesia, and occasionally tooth loss and ulceration of the overlying mucosa [5,6,7]. Paresthesia is a late manifestation of the tumor, a sign of nerve involvement [3, 8]. In this case, the patient presented with swelling of the right mandible.

The exact cause of this tumor is unknown. A genetic alteration is thought to be one of the mechanisms. Causes include fibrous dysplasia, a bone cyst, osteomyelitis, malignant transformation of a giant cell tumor of bone, and radiation therapy [3, 9]. In our case, the primary tumor originated in the right mandible. The patient had a history of ameloblastic fibroodontoma, which was operated on in 2015 without evidence of malignancy.



Une étude immunohistochimique réalisée montre :

- Anticorps anti PS100, CD34, Desmine, MUM4, EMA et AML : négatifs.

Conclusion :

- *Aspect morphologique et profil immunohistochimique compatible avec un fibrosarcome sans composante améloblastique associée grade II selon la FNLCC.*
- *Limites de résection passées en zone saine.*

Fig 4,5,6: Report and image of anatomical pathology of the mandibular mass

Radiographically, the tumor resembled an osteolytic lesion with poorly defined contours, with eroded and permeative geographic patterns of bone destruction [10] comparable to those observed in chondrosarcomas and osteogenic osteolytic sarcomas [5, 7, 11].

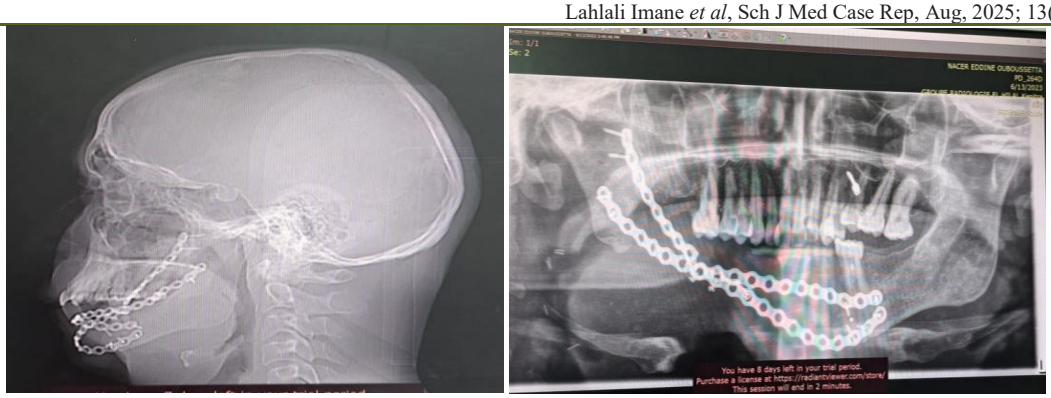


Fig 7,8 : Standard radiology images after right mandibulectomy

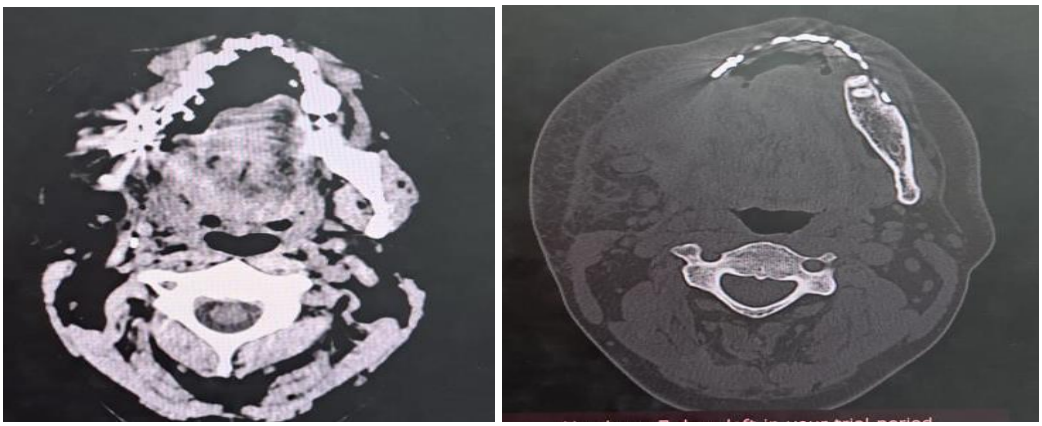


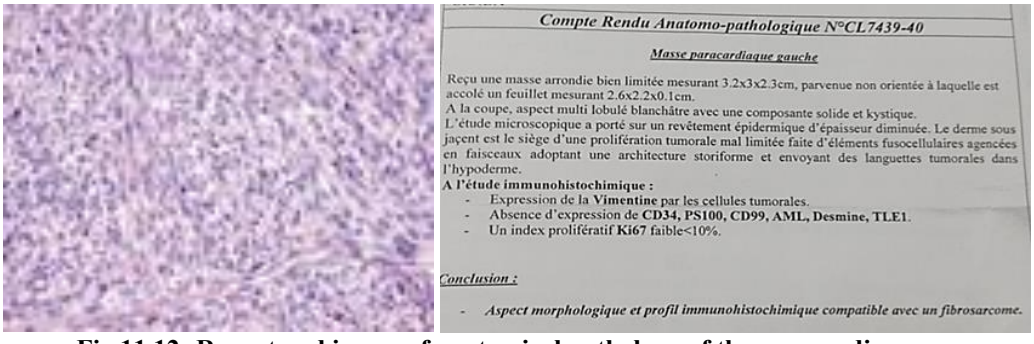
Fig 9,10: Images from facial CT scan after right mandibulectomy

Histologically, the level of differentiation can vary, and may be comparable to that of an anaplastic tumour. Low-grade fibrosarcoma is characterised by homogeneous spindle cells, organised in bundles, displaying a fishbone-like growth pattern with low to moderate cell density and a herringbone appearance. Mild nuclear pleomorphism and rare mitosis are noted, accompanied by a collagenous stroma [5, 12, 13].

High-grade lesions exhibit intense nuclear pleomorphism, increased cellularity, and atypical mitoses [14]. Nuclei may have a spindled, oval, or round shape [12]. High-grade fibrosarcoma may histologically resemble other tumour types, such as malignant fibrous histiocytoma, liposarcoma, or synovial sarcoma [5]. Our case was a low-grade fibrosarcoma.

The differential diagnosis includes fibromatosis, fibrous histiocytoma, liposarcoma, or synovial sarcoma of fibrosarcoma. In fibromatosis, mitosis is absent and the level of cellular atypia is extremely low. Odontogenic sarcomas show odontogenic tissues that are absent in fibrosarcoma [8,9].

Immunohistochemical (IHC) analysis was performed for the following markers: vimentin, pancytokeratin, desmin, actin, and S-100 protein. IHC revealed the presence of vimentin-positive cells, while other immunomarkers were not. By definition, a fibrosarcoma is primarily a diagnosis of exclusion and should not show any expression of actin, keratin, S-100, or desmin [15]. In this particular case, the tumor showed negativity to Desmin, PS-100, and CD-34.



Compte Rendu Anatomo-pathologique N°CL7439-40

Masse paracardiaque gauche

Reçu une masse arrondie bien limitée mesurant 3.2x3x2.3cm, parvenue non orientée à laquelle est accolé un feuillet mesurant 2.6x2.2x0.1cm.
A la coupe, aspect multi lobulé blanchâtre avec une composante solide et kystique.
L'étude microscopique a porté sur un revêtement épidermique d'épaisseur diminuée. Le derme sous jaçent est le siège d'une prolifération tumorale mal limitée faite d'éléments fusocellulaires agencées en faisceaux adoptant une architecture storiforme et envoyant des languettes tumorales dans l'hypoderme.

A l'étude immunohistochimique :

- Expression de la **Vimentine** par les cellules tumorales.
- Absence d'expression de **CD34, PS100, CD99, AML, Desmine, TLE1.**
- Un index prolifératif **Ki67** faible<10%.

Conclusion :

- ***Aspect morphologique et profil immunohistochimique compatible avec un fibrosarcome.***

Fig 11,12: Report and image of anatomical pathology of the paracardiac mass

The treatment of choice for fibrosarcomas is radical surgery [2,15]. Radiotherapy and chemotherapy can be used in inoperable cases or as palliative treatment [5], as their role in treatment is still unclear. They are administered for high-grade tumors, as these tumors may present with subclinical or microscopic metastases. [3,8,9] The five-year survival rate for this disease is low, ranging from 20 to 35% [15, 16].

Clinical stage, histological grade, and local recurrence are important prognostic factors. Prognosis depends on histological grade, tumor size, and adequate surgical treatment with clear margins [12]. In the present case, the patient underwent multiple surgeries and was then referred for adjuvant radiotherapy. However, due to the development of a paracardiac metastasis, the therapeutic strategy was modified and the patient was referred for chemotherapy.

CARDIAC METASTASES

Metastatic tumors to the heart are poorly understood and poorly documented in the literature. The frequency of secondary cardiac tumors has been reported to be between 0.7% and 3.5% in autopsies of the general population [19, 20] and between 10% and 15.4% in autopsies where a malignant neoplasm had been diagnosed [21].

All malignant tumors can give rise to cardiac metastases. Some cancers have a high potential for cardiac metastasis, but they are rare. Cardiac metastases are observed in approximately 30% to 10% of cases of lung, esophageal, renal, and hematologic malignancies, which represent 13% of secondary cardiac locations [22, 23]. The following table shows the different etiologies of cardiac metastases.

Tumeurs cardiaques primitives
Bénignes (75 % des cas)
- Myxomes (50 % des cas bénins)
- Fibro-élastomes papillaires
- Lipome
- Fibrome
- Rhabdomyome
- Hémangiome
- Tératome bénin
- Phéocromocytome
- (hypertrophie lipomateuse du septum interauriculaire)

Malignes
- Sarcomes (75 % des cas)
- Angiosarcome
- Mésothéliome
- Rhabdomyosarcome
- Fibrosarcome

Tumeurs cardiaques secondaires
- Poumon
- Lymphome
- Sein
- Estomac
- Mélanome
- Leucémie

Kystes cardiaques
- Echinoccocose
- Kyste sanguin
- Kyste péricardique

Thrombus-végétations
Corps étranger (exemple : cathéter de Swan Ganz)

Figure 13 : Etiologies of Cardiac Metastases

In osteosarcomas, for example, cardiac involvement secondary to osteosarcoma was identified in 20 of 480 patients at a single institution over the previous six years, but only three of these (0.6% of 480 patients) had direct cardiac involvement, while in the remaining cases, the involvement involved the great central cardiac vessels or tributaries of the primary tumor sites [20]. In an early study published in 1975 by Jeffrey et al., cardiac involvement was identified in 9 of 43 autopsies (21%). Another autopsy study by Siebert et al. [21] and published in 1982 reported cardiac involvement by osteosarcoma in 4 of 20 patients (20%).

Cardiac metastases are usually small, presenting as a nodule (24). Metastases to the heart generally first affect the pericardium, then the myocardium, and rarely the endocardium (25). Distant spread can occur directly by local extension in the case of bronchial or mediastinal tumors, indirectly via the bloodstream using the coronary arteries, or via the lymphatic route [26]. It can also occur via intravascular extension from the inferior vena cava or pulmonary veins (as in the case of hepatocellular and renal carcinomas) [27].

Clinically, manifestations can be highly variable: involvement of the right atrium can occasionally lead to its obstruction, resulting in a picture similar to pericardial constriction tamponade or restrictive cardiomyopathy. In addition, the tumor can ulcerate and release neoplastic microemboli into the pulmonary arterial circulation. [21]. Cardiac involvement can be completely asymptomatic or associated with certain signs, such as dyspnea, rhythm

disturbances, tamponade, or heart failure [25]. In our patient, the clinical signs were rather subtle and manifested as palpitations. The diagnosis was made incidentally during an ultrasound examination.

The subtlety of the clinical signs and the nonspecific nature of simple paraclinical examinations explain the value of other non-invasive imaging techniques. These can determine whether the tumor is a thrombus or a tumor. Once the tumor diagnosis is made, imaging can determine its origin (primary or secondary tumor) and establish a topographic and extension assessment to guide the appropriate therapeutic strategy, once histological confirmation has been obtained.

The diagnosis of cardiac metastases is generally made by echocardiography, CT scan, MRI, or, more recently, positron emission tomography (PET) scan. (PET scan). Echocardiography is the basic examination to detect these tumors and confirm the diagnosis [26]. MRI provides interesting information to characterize the tissues and vascularization. It is also useful for diagnosing several forms of cardiac pseudo-tumoral masses [28].

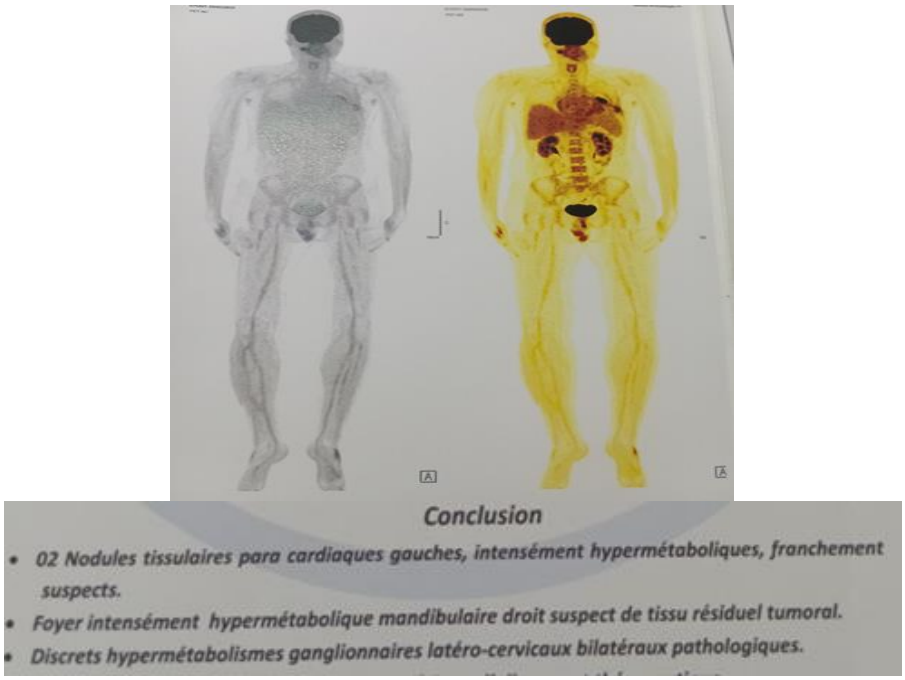


Figure 14, 15 : Image and report from the PET scan showing mandibular and paracardiac hypermetabolism

CT scans offer lower tissue contrast, but allow for better clarification of the anatomical relationships of a lesion (coronary, pericardium), thanks to their high spatial resolution.

No signs can formally differentiate malignant from benign cardiac tumors based on imaging. However, the presence of intratumoral necrosis, tumor size greater than 5 cm, tissue inhomogeneity, and pericardial invasion are elements that point toward a malignant origin, as is a history of neoplasia that may indicate a potential metastatic origin [29, 30]. In cases of disseminated disease, diagnostic biopsy of the cardiac lesion is not performed. The therapeutic decision is determined by the extent of the disease. When the heart is the only metastatic site, excisional surgery may be attempted [25, 31].

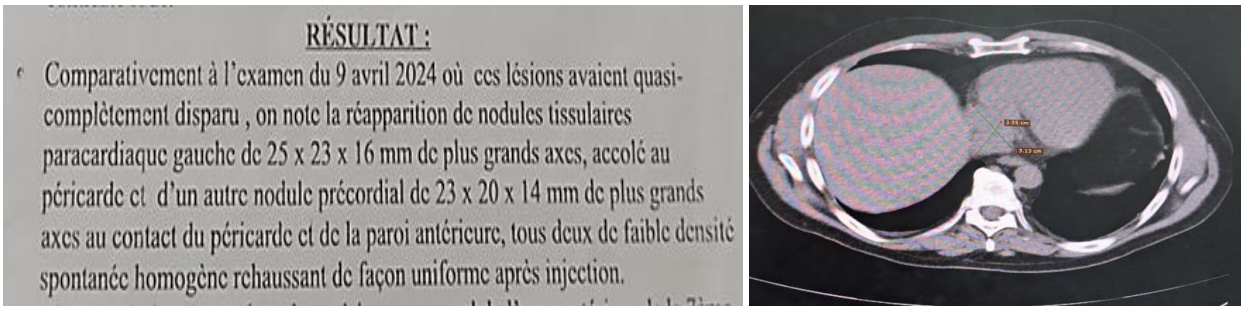
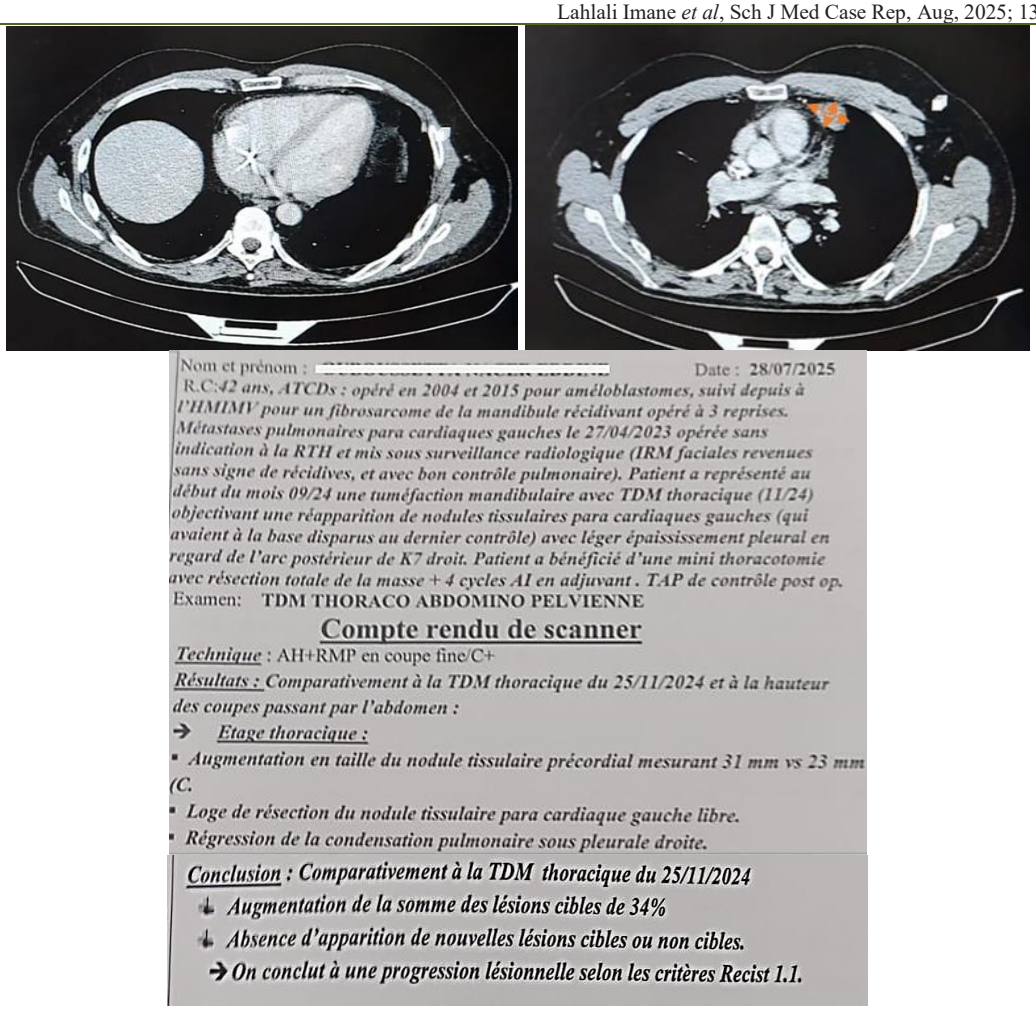


Figure 16, 17 : image and report of chest CT scan showing recurrence of the para-cardiac mass



Nom et prénom : Date : 28/07/2025

R.C:42 ans, ATCDs : opéré en 2004 et 2015 pour améloblastomes, suivi depuis à l'HMIMV pour un fibrosarcome de la mandibule récidivant opéré à 3 reprises. Métastases pulmonaires para cardiaques gauches le 27/04/2023 opérée sans indication à la RTH et mis sous surveillance radiologique (IRM faciales revenues sans signe de récidives, et avec bon contrôle pulmonaire). Patient a représenté au début du mois 09/24 une tuméfaction mandibulaire avec TDM thoracique (11/24) objectivant une réapparition de nodules tissulaires para cardiaques gauches (qui avaient à la base disparus au dernier contrôle) avec léger épaississement pleural en regard de l'arc postérieur de K7 droit. Patient a bénéficié d'une mini thoracotomie avec résection totale de la masse + 4 cycles AI en adjuvant . TAP de contrôle post op.

Examen: TDM THORACO ABDOMINO PELVIENNE

Compte rendu de scanner

Technique : AH+RMP en coupe fine/C+

Résultats : Comparativement à la TDM thoracique du 25/11/2024 et à la hauteur des coupes passant par l'abdomen :

→ *Etage thoracique :*

- Augmentation en taille du nodule tissulaire précordial mesurant 31 mm vs 23 mm (C.
- Loge de résection du nodule tissulaire para cardiaque gauche libre.
- Régression de la condensation pulmonaire sous pleurale droite.

Conclusion : Comparativement à la TDM thoracique du 25/11/2024

- Augmentation de la somme des lésions cibles de 34%
- Absence d'apparition de nouvelles lésions cibles ou non cibles.

→ On conclut à une progression lésionnelle selon les critères Recist 1.1.

Figure 19-22 : Images and report of chest CT scan showing good local control of the paracardiac mass and the appearance of the precordial mass

CONCLUSION

This disease was characterised by recurrent metastasis. Unfortunately, our patient's latest assessment, carried out recently, revealed a recurrence, a paracardiac mass, this time precordial.

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