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Inflammatory myofibroblastictumor of the maxillary alveolus: A rare entity presenting as a jaw swelling

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Abstract: *An inflammatory myofibroblastictumor (IMT) is an immunohistochemically diverse entity demonstrating neoplastic and nonneoplastic qualities.IMT occurs most commonly in the lungs. Head and neck constitutes14-18% of extrapulmonarysites.The common head and neck site of IMT is the larynx and the intraoral site for IMT is the buccal mucosa and it also occurs in other head and neck sites such as the tonsils, para-pharyngeal space, sino-nasal tract, and trachea. The exactetiology and pathogenesis of IMT are not well known. Initially it was thought to be of purely inflammatory in origin and later it was found to have potential for recurrence, infilterative local growth and even malignant transformation. Here, we are discussing a rare case of 65years old female having IMT in left maxillary alveolus region.*

Keywords- *Neoplastic, inflammatory, lungs, maxillary alveolus.*

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III. Introduction

An inflammatory myofibroblastictumor (IMT) is an immunohistochemically diverse entity demonstrating neoplastic and nonneoplastic qualities. It was first described by Brunn in 1939 [1, 2] as "inflammatory pseudotumor". Umiker and Iverson in 1954 first described it as a tumor.[1]IMT is a distinct borderline lesion composed of myofibroblastic cells with a variable admixture of inflammatory cells and collagen. IMT occursmost commonly in the lungs.Head and neck constitutes14-18% of extrapulmonarysites. The etiopathogenesis of IMT are not well known. Here, we describe a case of IMT in the maxillary alveolus which clinically and radiographically mimics odontogenictumor.

IV. Case Report

A 65yrs old female reported in the OPD with swelling of the left maxillary region since 4years. On palpation, well-defined non-tender round swelling of approximately 5-6cm in size was evident on left maxillary region extending mediolaterally from ala of nose to zygomatic prominence of maxilla and superoinferiorly from infraorbital margin to maxillary alveolus. It was associated with paresthesia. The overlying skin was normal in respect to surface texture, color, temperature (FIGURE-1).Intraorally, Well-defined round swelling was evident involving hard palate and left maxillary buccal vestibule. Also, there wasextrusion of left maxillary anteriors, distally displaced third molar and left maxillary premolars and molars were missing (FIGURE-2).

Orthopantomograph(OPG) showed well defined radiolucent osteolytic lesion with corticated margins in left maxillary region extending from nasal septum to zygomatic process of maxilla, displacing the left maxillary sinus laterally and superiorly. The left maxillaryposterior teeth were missing and distally displaced third molar.

Contrast enhanced computed tomograph(CECT) ofmaxillofacial region- Axial section revealed well-defined expansile lytic lesion of approximately 4.5*5*7cm, with non-enhancing soft tissue density contents and few air foci seen in left half maxilla, causing thinning, remodelling and cortical breach of maxilla. Superiorly, the lesion was causing upward displacement of inferior wall of maxilla and compression of maxillary sinus.The lesion extending to the left nasal cavity displacing nasal septum to right.Inferiorly, it was bulging in the oral cavity with thinning of hard palate(FIGURE-4).

Based on clinical and radiographic findings, the differential diagnosis was made as benign odontogenictumor (Ameloblastoma, keratocysticodontogenictumor).

Incisional biopsy was been done and histopathologic examination revealed fibrous lesional tissue composed of plump of spindled fibroblasts with vesicular nuclei. Numerous myofibroblasts with eosinophiliccytoplasm were also noted. The supporting stromal tissue was made up of bundled collagen fibres with areas of homogenisation. Moderate degree of chronic inflammatory infiltration comprising of plasma cells, lymphocytes and histiocytes was observed. Numerous thin walled vascular channels were seen distributed throughout the lesional tissue. Thefindings were suggestive of inflammatory fibroblastic tumor(FIGURE-5). To confirm the diagnosis and pathogenesis of the lesion, immunohistochemistry was been done.

Immunohistochemical(IHC) analysis revealed that the spindled cells were strongly positive for vimentin and focally (20% of cells) shown positivity for smooth muscle antibody (SMA).Stains for Ki67, cytokeratin, S-100 was negative(FIGURE-6). On the basis of above histopathologic and IHC findings, a diagnosis of Inflammatorymyofibroblastictumor was made.

Patient was planned for excision of the lesion and the resected lesional tissue was sent for histopathological examination and was found to be low grade inflammatory myofibroblastictumor. The patient kept under follow-up.

V. Discussion

Inflammatory myofibroblastictumors are classified by WHO classification as an intermediate soft-tissue tumor comprising spindle cells that exhibit myofibroblast differentiation and numerous inflammatory cells, plasma cells, and/or lymphocytes.[3]Because of variation in histologic appearance it has various synonyms as: inflammatory pseudotumors, plasma cell granuloma/pseudotumor, pseudosarcomatoustumors, fibrous xanthoma, xanthomatouspseudotumors, fibrous histiocytoma.[3]

Initially it was thought to be of purely inflammatory in origin and later it was found to have potential for recurrence, infilterative local growth and even malignant transformation.[4,5]Different hypothesis for origin of IMTs has been suggested like infection, auto immune, and trauma. The exact pathogenesis and etiology of IMT are not clear. The common head and neck site of IMT is the larynx and the intraoral site for IMT is the buccal mucosa.[6] However, in our case IMT occurred in the maxillary alveolus region. IMTs of upper aero-digestive tract are common in adults (median age of 59) and more often seen in the males[7], whereas in the present case the patient was 65years old female. Binmadi *et al*. have shown that lesion in the maxilla presented as a small sessile nodule.[8] However, in our case it presented as a large roundsmooth swelling on the left maxillary alveolus region extending mesially on hard palate. IMTs are characterized by ill-defined infiltrative bony erosion on CT scan,[2] and in the present case there was a well-defined homogenous, osteolytic enhancing lesion with destruction of the maxillary alveolus on the CT scan.

The histological differential diagnosis of IMT includes benign tumors like nodular fasciitis, solitary fibrous tumor, benign fibrous histiocytoma, follicular dendritic cell tumor, myofibroma and malignant tumors.[9] IHC for the present case was positive for the expression of SMA, which is a diagnostic feature on IHC for IMT. Histologically the present case was a low-grade lesion.Gallego *et al*. has also demonstrated that in IMT there is faint positivity of Ki67 on IHC study.[10] However, in the present case IHC for Ki67 and cytokeratin was negative for nuclear stain.

Non-surgical management of IMT, includes chemotherapy in the form of cyclosporine, methotrexate, azathioprine, and cyclophosphamide but it has little role and corticosteroids is not so effective in the treatment of IMTs of the head and neck region.[3] Moreover, in the present case conservative treatment with corticosteroids could not be started due to the lack of definitive diagnosis of the mass preoperatively, so complete resection of the lesion was been done. In the present case the lesion was clinically and radiologically mimicking benign odontogenictumor, histopathological examination followed by IHC confirmed the diagnosis. Follow-up for 10 years to detect local recurrence following simple excision of oral IMTs has been suggested.[10] In the present case, only surgical excision was done, and there was no local recurrence at the follow-up of 1year after treatment.

VI. Conclusion

IMT of the jaw are extremely rare, andmimics as odontogenictumors, clinically and radiographically. Histopathogical examination followed by IHC is mandatory for lesion to know the aggressive nature of such lesions. Complete surgical excision of alveolar IMT is the treatment of choice. The patients with oral IMT require periodic postsurgical follow-up to detect local recurrence.

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LEGENDS OF FIGURES

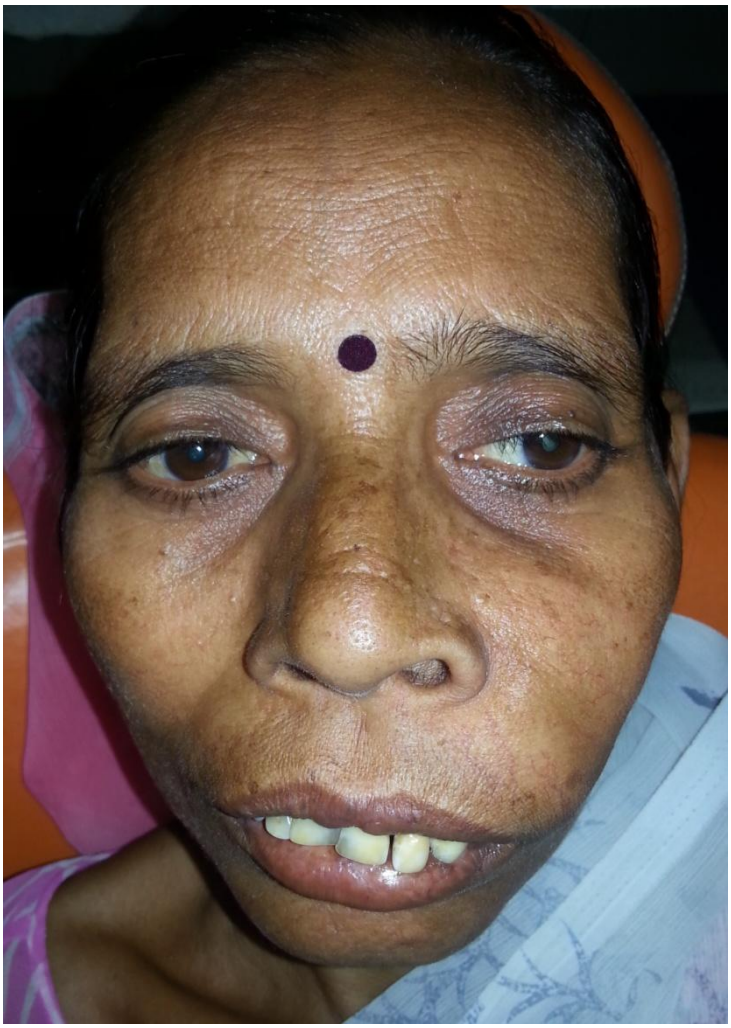


FIGURE-1 – Extraoral photograph showing diffure swelling in left maxillary region. Extrusion of left maxillary incisors is evident.

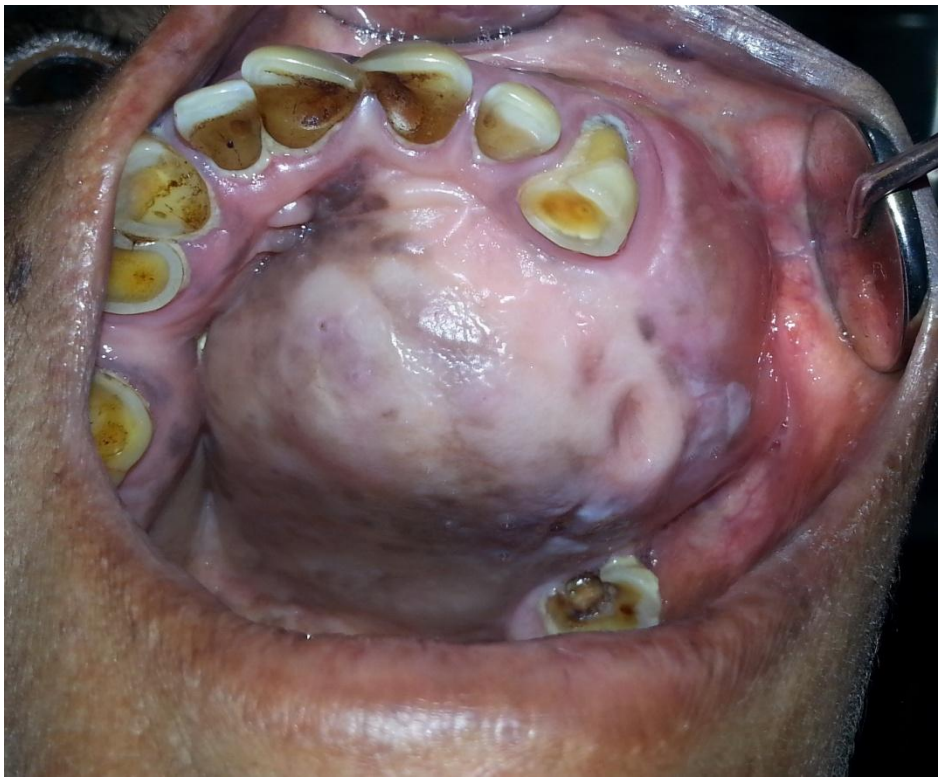


FIGURE-2 – Intraoral photograph showing well-defined round swelling in left maxillary alveolus region involving the whole hard palate and obliterating the buccal vestibule.

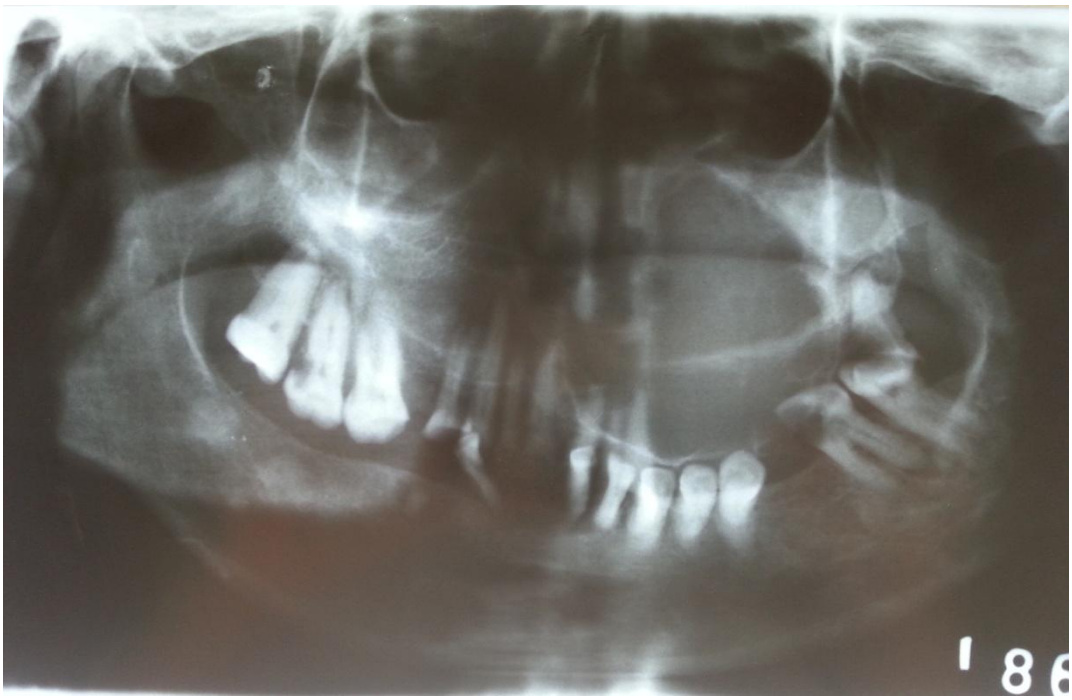


FIGURE-3 – OPG showing well defined radiolucency in left maxillay alveolus region displacing the left maxillary sinus laterally and superiorly.

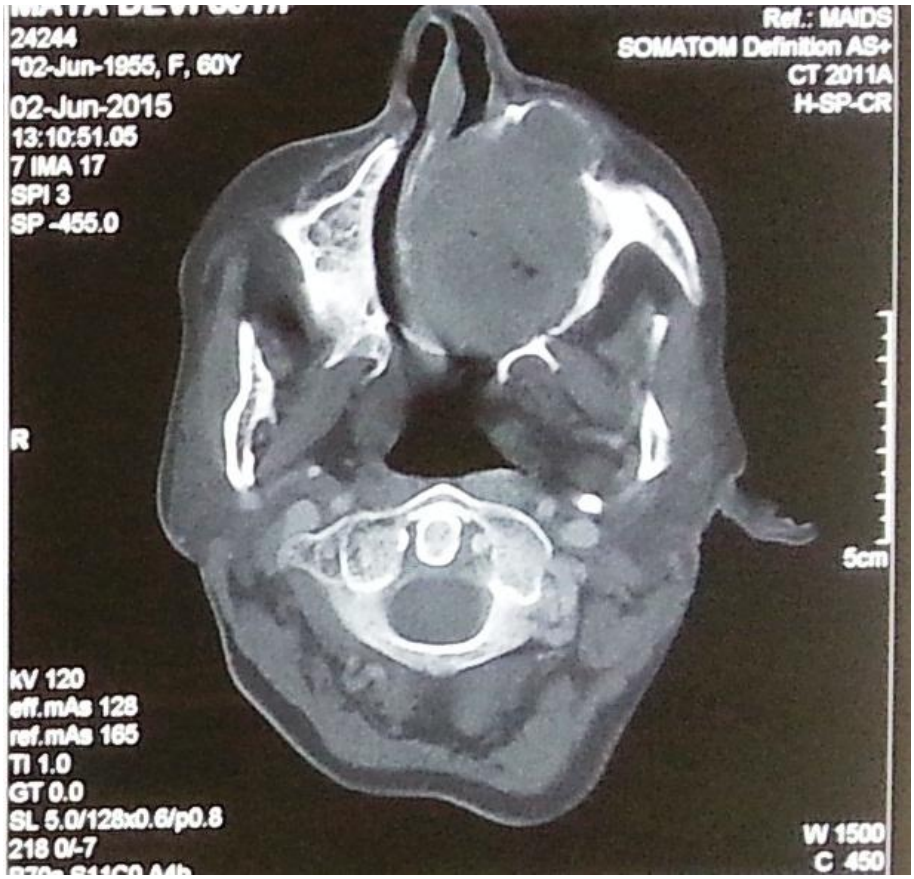
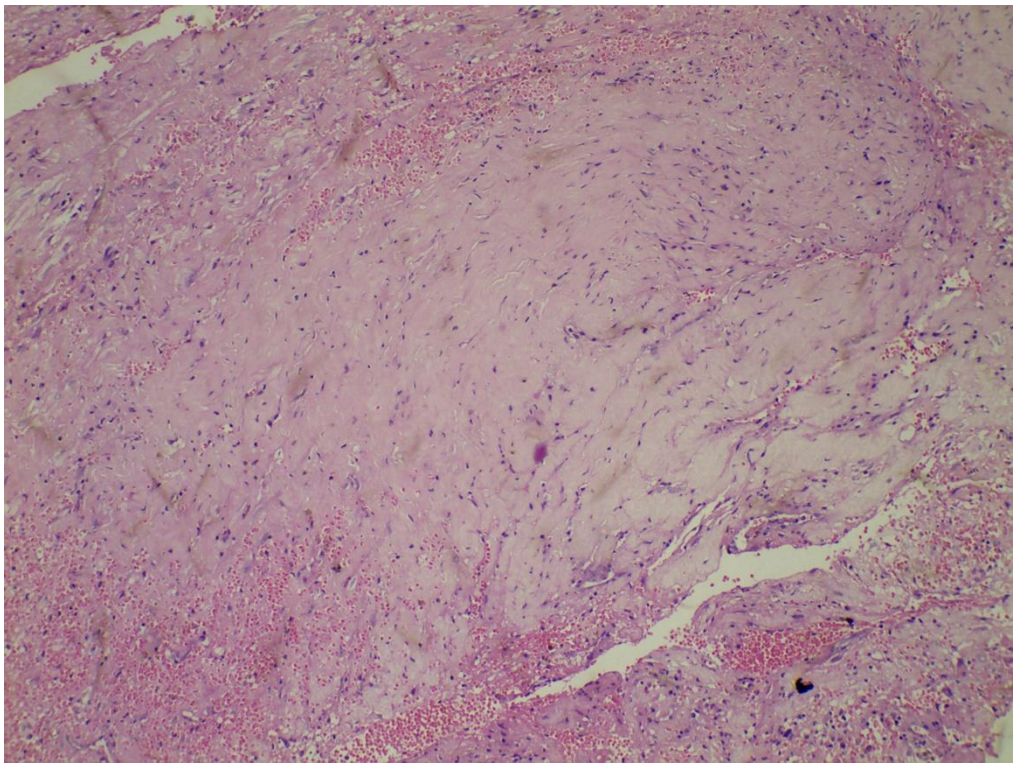
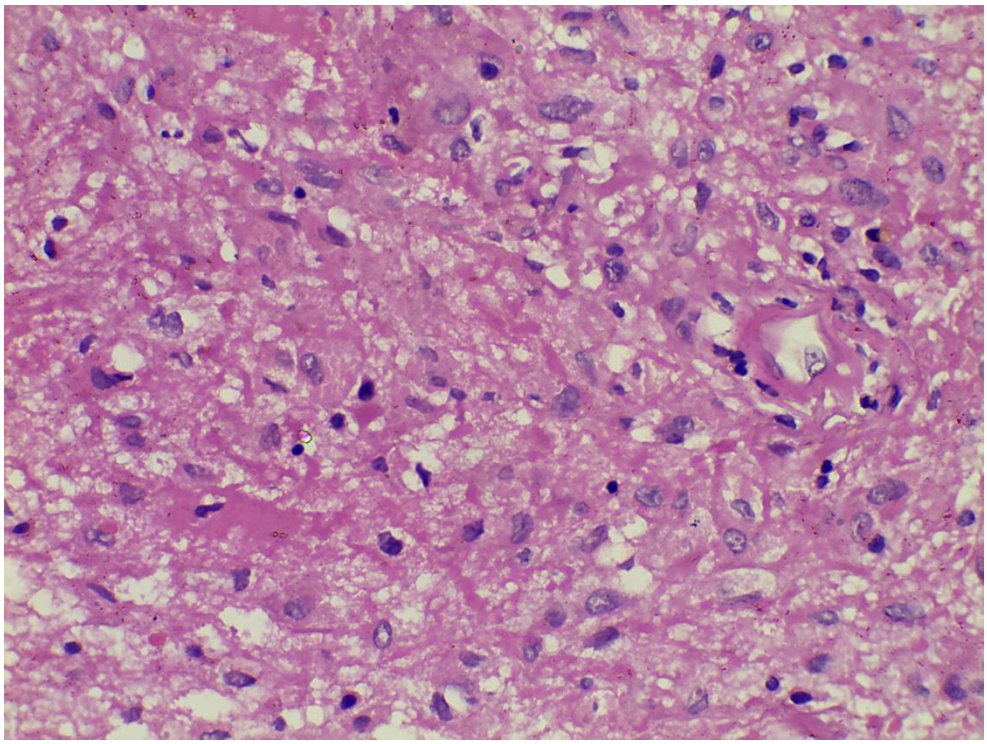


FIGURE-4 – Axial section of contrast-enhanced computed tomograph showingwell-defined expansile lytic lesion with non-enhancing soft tissue density contents and few air foci.

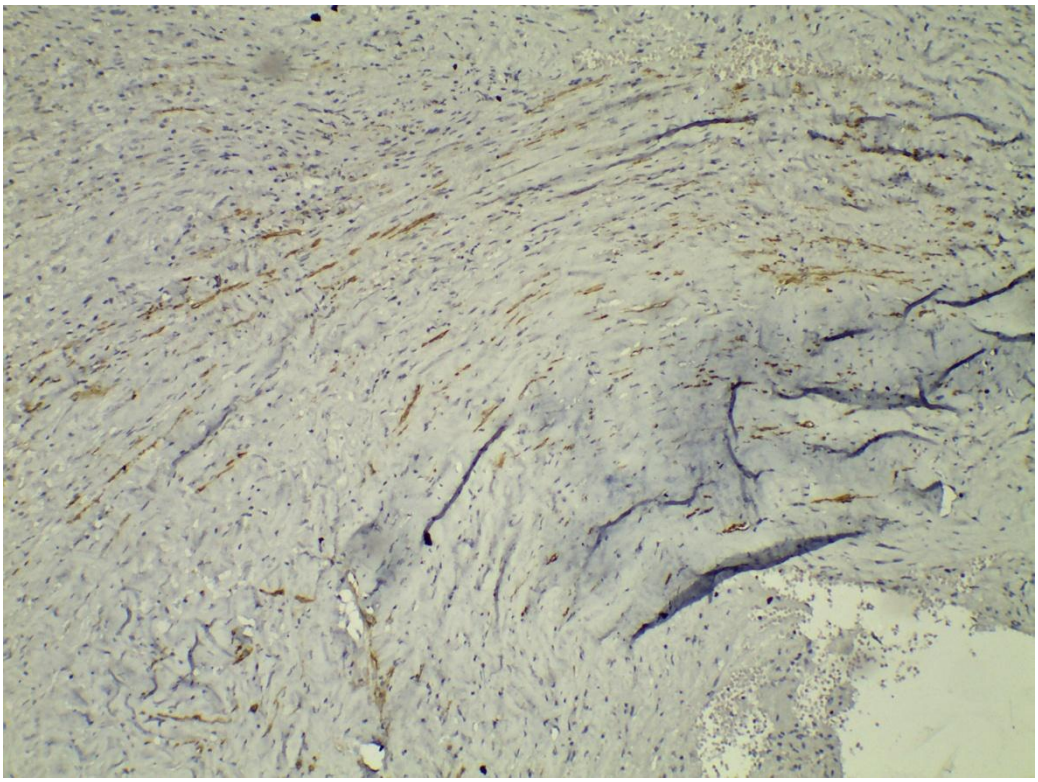


(a)

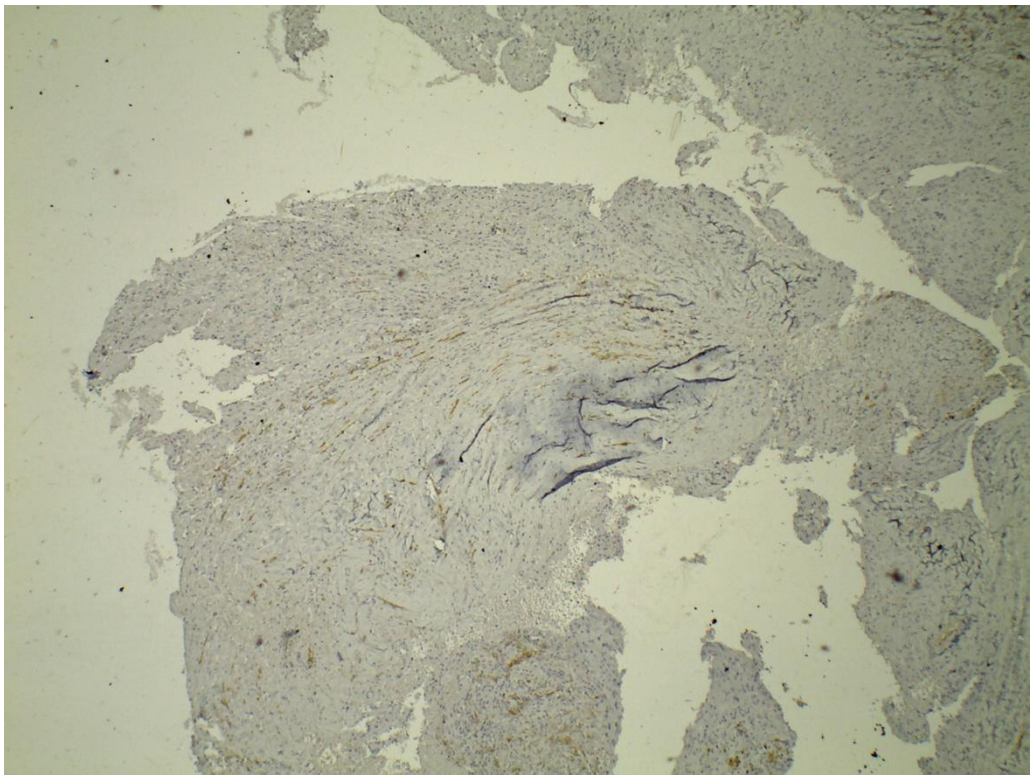


(b)

FIGURE-5 (a and b)-Histomicrophotograph ofhematoxylin and eosin staining showing fibrous tissues with plump of spindled fibroblasts, numerous myofibroblasts with eosinophilic cytoplasm, chronic inflammatory infilteration in collagenous stromal tissue.



(a)



(b)

FIGURE-6 –Histomirophotograph of immunohistochemical stains showing spindled cells positive for Vimentin and focally (20% of cells) positive for smooth muscle antibody(SMA).

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