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CYTODIAGNOSIS OF SUBMANDIBULAR SIALADENITIS WITH CRYSTALLOIDS MIMICKING METASTASIS

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ABSTRACT : *Sialomegaly of submandibular gland, whether inflammatory or neoplastic can be clinically mistaken for lymphoid metastasis especially when encountered in an elderly patient. Other than the neoplastic lesions and usual inflammatory lesions rarely amylase crystalloids can evoke a granulomatous response and can result in sialomegaly. We report an interesting case which presented as a submandibular swelling and was clinically diagnosed as metastasis where the diagnosis of granulomatous response to crystalloids was made at FNAC. The aim of this report is to bring an awareness of the existence of such lesions and to highlight the role of FNAC in diagnosis of such lesions in order to avoid unnecessary surgeries.*

Key Words: *Amylase crystalloids, sialadenitis, FNAC*

INTRODUCTION

Mass in the submandibular region would be due to enlargement of salivary glands or of regional lymph nodes. Both of these could be either inflammatory or neoplastic in nature. Metastatic process is the commonest cause of the cervical lymph node enlargement especially in elderly patients. Certain physiological substances can crystallize in the salivary glands causing sialadenitis which clinically mimics a metastatic process.

Fine needle aspiration cytology (FNAC) plays a vital role in the evaluation of mass in the cervical region and accurately distinguishes between salivary gland lesions and lymph node metastasis. It is a well established technique in the management of salivary gland disease where it helps in distinguishing inflammatory from neoplastic lesions and avoids unnecessary surgeries in about one third to one half of the cases.

We report an interesting case of an elderly male patient who presented with a submandibular swelling which was

clinically suspected to be metastasis, but on FNA was diagnosed to have sialadenitis with amylase crystalloids.

CASE REPORT

A 60-year-old male presented to the surgical out patient department of St Johns' Medical College Hospital with

Fig.I : Rectangular crystals in the background of necrosis with macrophages. (10x)

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history of swelling in the left submandibular region of three months duration. He had no other constitutional symptoms or history suggestive any other malignancies. His general physical and systemic examinations revealed no significant findings. Routine hematological investigations were normal.

Local examination revealed a swelling in the region of the submandibular gland measuring 2x2cms. The lesion was hard to feel, non-tender with mild restriction of mobility noted in both planes. No other enlarged lymph nodes were noted in the neck. No foci of infection or inflammation in the oral cavity or in the head and neck region were noted. He was referred to the department of cytology for evaluation by Fine Needle Aspiration Cytology for a primary diagnosis with a need to rule out a metastatic process in a lymph node. FNAC was performed with two passes and about 1ml of whitish opaque blood tinged material was obtained. Smears were made and fixed in 95% alcohol. The smears were stained with standard Papanicolaou and Hematoxylin and eosin stains.

Cytological findings:

The smears showed a high cell yield composed chiefly of acute and chronic inflammatory cells including many macrophages and epithelioid like cells with a few foreign body type of multinucleate giant cells. The background showed necrotic material with much karyorrhectic debris. Dispersed in the background were crystalline structures which were rectangular having blunt ends and varied in size from 40 to 70 microns. They stained red to orange with the Papanicolaou stain and were colorless and refractile with the Hematoxylin and eosin stains. A few crystalline fragments were seen within the giant cells. Scattered salivary acini were seen along with fragments of fibro connective tissue (Fig. 1). No malignant cells or lymphoid cells were seen. The periodic acid Schiff stain and Gomori's methamine silver stain were negative which ruled out fungal infection. The Ziel-Nielsen stain for acid fast bacilli was negative. With these features a cytological diagnosis of chronic sialadenitis with granulomatous response to crystalline material was made. The nature of the crystalloid substance could not be ascertained. However a diagnosis of malignancy was ruled out and close follow up was suggested.

On a literature search, it was found that these crystalline structures morphologically resembled amylase crystalloids which may be associated with sialadenitis and granulomatous response.

The patient received symptomatic treatment and the swelling gradually regressed and disappeared over a period of 15 months with no other complaints. Therefore an excision biopsy was not done in this case.

DISCUSSION

Fine needle aspiration cytology is an important technique in the evaluation of neoplastic and non-neoplastic lesions at any site. It plays an important role in confirming or refuting a diagnosis of malignancy. It is an excellent diagnostic modality in distinguishing sialomegaly and cervical lymphadenopathy. FNAC is the first line of investigation in the diagnosis of lymph node metastasis.

Submandibular salivary gland enlargement may be due to neoplastic or inflammatory processes. A FNA procedure is mandatory before any surgical intervention to distinguish the two. Of the rare causes for sialadenitis, crystalloids in the salivary gland are rarely encountered. A few reports exist in the literature as to the occurrence and incidence of the lesion. Of the cases reported, most of them occurred in the parotid gland, and rarely has been reported in the submandibular gland.^{1,2} Amylase crystalloids were first described by Takeda and Ishikawa in 1983 where they also confirmed the nature of the substance by scanning electron microscopy and electron probe x-ray microanalysis³. They are classically described on routine stains as rods squares, rectangles, needles which are non-birefringent. They do not stain positive with mucin stains like alcian blue or mucicarmine. They do not stain with Prussian blue and von Kossa which rules out any mineral content in these crystals².

Quantification of amylase activity of these crystals can be done by electrophoresis or mass spectrometry. MALDI mass spectrometry has confirmed that these crystalloids have a molecular weight of 57,768⁴. On transmission electron microscopy these crystals are polyhedral with no periodicity. Biochemical analysis can be done by automated analyzers. In the absence of non-availability of these techniques a cytopathologists should be aware of the appearances of these crystals on light microscopy to arrive at the correct diagnoses.

Amylase crystalloids may be found in normal and cystic lesions of salivary glands¹. It has been suggested that they may be responsible for the initiation of calculi formation or a granulomatous response^{5,6} resulting in a firm swelling in the region of salivary glands as seen in our case, where the other causes for granulomatous inflammation were

ruled out. These can clinically present as swellings with no associated symptoms which can be misdiagnosed as a neoplastic process. Most of the cases reported in the literature have been wrongly misdiagnosed as tumors, but FNAC has resolved the issue in every case^{1, 2}. These patients have been found to have no recurrence on a follow up. The present case also received symptomatic treatment and was found to be free of disease after a follow up of 15 months.

An awareness of such an occurrence is important for both cytopathologist and the clinician. FNAC plays a major role in the diagnosis of these lesions and avoids unnecessary surgeries.

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SOLITARY MALIGNANT SCHWANOMA OF PARA PHARYNGEAL SPACE -A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT : *Malignant Schwannoma is an aggressive soft tissue tumor that is more commonly found in association with Von Recklinghausens disease. The solitary tumors although rarer have a better prognosis when compared to those associated with Von Recklinghausens disease. Parapharyngeal tumors are rare and a majority of these are benign salivary or neurogenic tumors. A malignant Schwannoma at this site is very infrequent with only four cases reported so far. The case of a 16 year old girl with a parapharyngeal malignant schwannoma is presented for its rarity and our experience in dealing with it. A combined modality of treatment with surgery, radiotherapy and chemotherapy was used. Pertinent literature is reviewed.*

Key Words : *Malignant Schwannoma, Parapharyngeal Space Tumors*

INTRODUCTION

Parapharyngeal tumors constitute 0.5% of all head and neck tumors [Kerry Olsen] and although 20-40% of these are of neurogenous origin they are mostly benign schwannomas. About 2% of all neurogenous tumors turn out to be malignant. Malignant schwannomas (MS) arise in patients with and without Von Recklinghausens (VR)

Disease. The risk of developing malignant neurogenous tumors is 4600 times more in patients with Rr disease than in the general population [Ducatman et. Al].

In a retrospective study done at the Mayo Clinic, 120 cases of MS were detected over a period of 71 years. Of these 23 arose in the region of the head and neck (an

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