Case Report

Cytological Diagnosis of Fibromatosis Coli: A Rare Presentation of Infantile Neck Swelling

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ABSTRACT

Fibromatosis coli is a rare benign mesenchymal lesion, usually presenting with neck mass in infant. Although etiology is not known, about half of the patients have history of birth trauma. It is self limiting fibroblastic lesion. Fine needle aspiration cytology is used as first line of investigation. If the diagnosis is made early, it shows an excellent prognosis. Most cases are managed conservatively. Here we present two interesting cases of fibromatosis coli which were diagnosed cytologically & showed complete regression after conservative management.

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Introduction

Fibromatosis coli also known as sternocleidomastoid tumor of infancy, is a benign self limiting fibroblastic lesion. Its occurrence is rare and around 0.4% of live birth.[1] The tumor is seen characteristically between 2-8 weeks of life as a firm mass of around 2-3 cm in diameter in the mid or lower part of sternocleidomastoid muscle.[2] Male infants are affected more commonly than females.[2] In about 50% of the cases there is history of complicated delivery.[2] One fourth of them may present with mild torticollis. Various congenital anomalies may be associated with fibromatosis coli. These tumours show spontaneous regression & eventually disappear completely with conservative management. Cytological diagnosis is non invasive, non expensive & invaluable to differentiate it from other inflammatory or neoplastic conditions of infancy.

Case Report(S)

We present two cases of fibromatosis coli one in a 3 weeks old female and other in a 17 days old male infant where both the cases were diagnosed on FNAC.

Case 1: A 3 weeks old female infant presented with neck swelling in the middle part of sternocleidomastoid muscle, noticed by her mother for one week. The swelling was firm; measuring 1x1 cm. There was history of preterm labour with breech presentation. On clinical examination, there was restricted neck movement on the affected side. There were no other congenital anomalies. FNAC was advised. Cytology showed bland appearing fibroblasts with oval to spindle nuclei and muscle giant cells (Fig 1, 2) in a clear background. Mother was reassured about the management. The baby was treated conservatively in the form of physiotherapy, passive stretching of muscles. The swelling started reducing after 2 weeks of physiotherapy & neck movements returned to normal. After 3 months of treatment the mass disappeared completely.

Case 2: A 17 days old male infant presented with torticollis, noticed by his parents for 2 weeks. On clinical examination there was a firm, fusiform swelling, 2x1.5 cm in diameter in the lower part of sternocleidomastoid muscle. There was no history of prolonged labor or birth trauma. Contact history of tuberculosis was positive. Clinically diagnosis of both tuberculous lymphadenitis & sternocleidomastoid tumor of infancy were suggested. Ultrasonography of neck was advised, which was suggestive of cervical lymphadenopathy. For confirmation FNAC was done. Cytosmear showed benign looking oval to spindle fibroblasts, degenerated atrophic muscle & muscle giant cells (Fig 3, 4, 5). Diagnosis of fibromatosis coli was given. Conservative management including gentle passive stretching was advised. Parents were educated about the techniques. After one week of physiotherapy, the swelling started to reduce with returning to near normal neck movements.

Discussion

Sternocleidomastoid tumour of infancy is an infrequent lesion presenting as neck mass. Fibromatosis coli is seen around 0.4% of all newborn. It usually occurs in the middle or lower part of the sternocleidomastoid muscle.[3] Etiology is not clearly understood, although 50% of cases are associated with a history of birth trauma.[4] Most infants present to OPD with congenital torticollis.[5] It usually appears at 2-4 weeks of age with a male predominance.[6] One of our patients is a female. Most of cases are diagnosed before 6 months of age. Both our patients were in the same age group. Causative factors are not known, however various theories have been proposed, which include fetal malposition, birth trauma, ischemic necrosis following vascular compression during birth, infections & presence of other endogenous factors.[7] Among these birth trauma is more commonly associated with fibromatosis coli, which was present in our first case. Another study by Kurtyez et al also showed the similar findings.[8] These tumors are sometimes associated with other congenital anomalies like club foot, congenital dislocation of hip etc,[1] however no such congenital anomalies were seen in our cases. Differential diagnosis of fibromatosis coli include congenital lesions like branchial cyst, thyroglossal cyst, inflammatory lesion like tuberculous lymphadenitis, benign neoplastic lesions like hemangioma, cystic hygroma or malignant neoplasms like neuroblastoma, rhabdomyosarcoma & lymphoma.[1] FNAC is the first line of investigation & helps to exclude

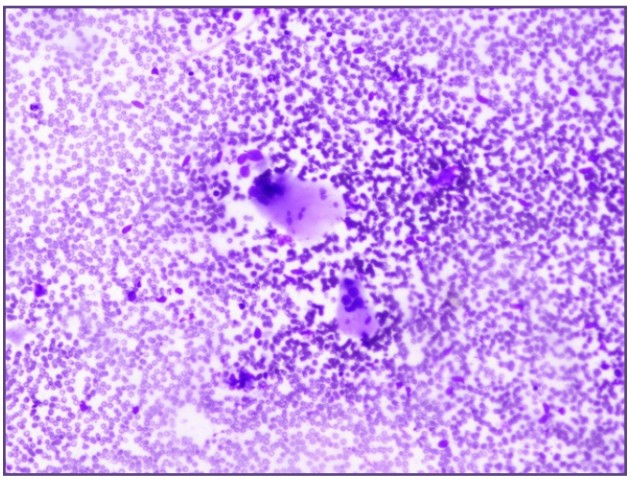

Fig. 1: Cytosmear shows bland appearing fibroblasts and muscle giant cells (Giemsa, 100X).

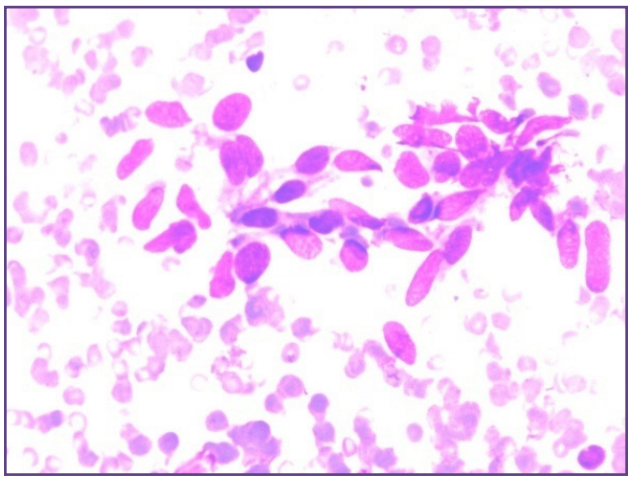

Fig. 2: Cytosmear shows bland appearing fibroblasts with oval to spindle nuclei (Giemsa, 400X).

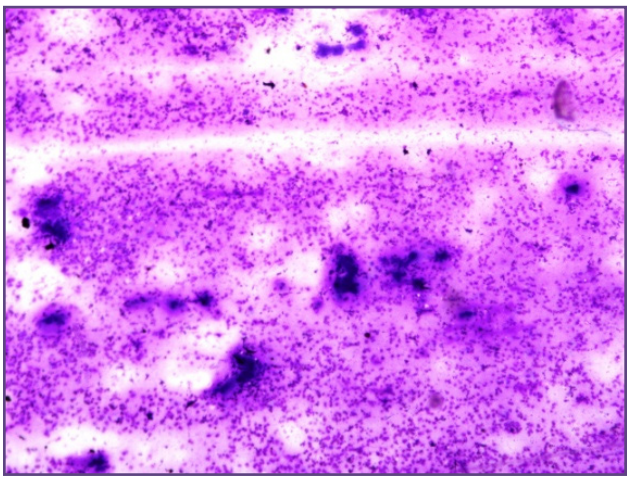

Fig. 3: Cytosmear showed benign looking oval to spindle fibroblasts, degenerated atrophic muscle & muscle giant cells (Giemsa, 40X).

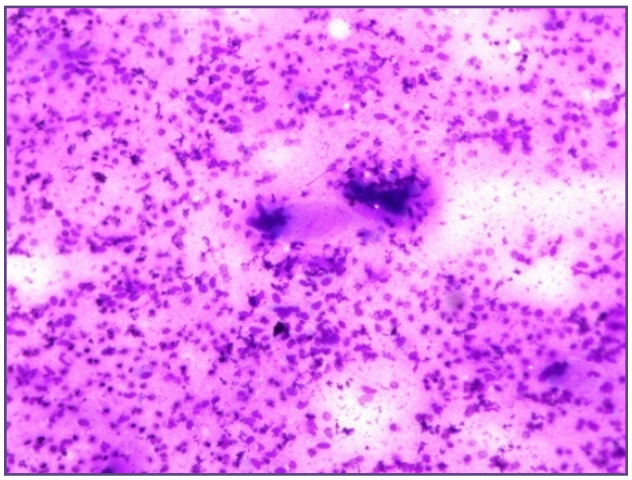

Fig. 4: Cytosmear shows benign looking oval to spindle fibroblasts, degenerated atrophic muscle & muscle giant cells (Giemsa, 100X).

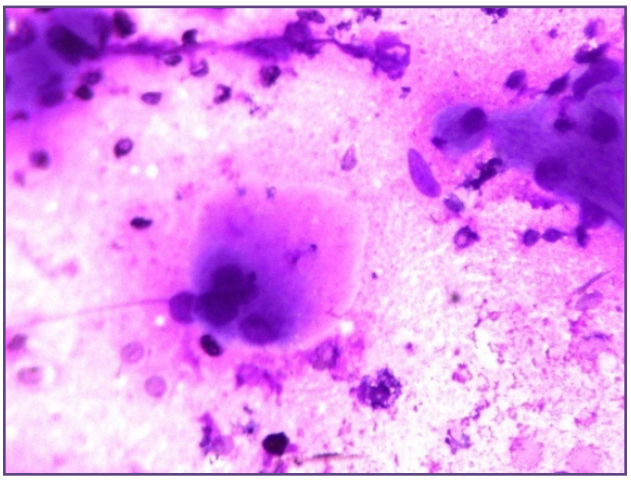

Fig. 5: Cytosmear showing degenerated atrophic muscle & muscle giant cells (Giemsa, 400X).

other differentials, as both our cases were diagnosed on FNAC. FNAC is cheap, non invasive, reliable diagnostic tool that helps to avoid nore invasive & costly procedures. Imagings like ultrasonography, computed tomography may be of help in diagnosis of fibromatosis coli, however high cost & less availability limit their roles.[9] There are few studies available in the literature that described cytological features of fibromatosis coli.[1,10,11] The characteristic cytological features described are bland appearing fibroblasts, degenerative atrophic skeletal muscles & muscle giant cells in a clear background. Similar findings are also seen in our cases. These rare infantile lesions are mostly managed by conservative treatment. Managements include passive stretching of the muscle & physiotherapy in early diagnosed cases, while for children greater than one year or refractory cases, surgical interventions may be required.[12] Late diagnosed cases show worst prognosis.[1] Both of our patients were diagnosed confidently at an early stage & following conservative treatments both are doing well and are disease free.

Conclusion

Fibromatosis coli is a self limiting fibroblastic lesion of infancy & it has a characteristic clinical presentation & history. These cases highlight vital roles of FNAC in diagnosis & reassurance of anxious parents. Cytology avoids unnecessary surgical intervention & guides for conservative managements. Hence fibromatosis coli should always be considered in the differentials of neck swelling in infants.

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Competing Interests
None Declared

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