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Case Report

A rare case report of fibrous hamartoma of infancy: Cyto-histopathological correlation

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ABSTRACT

Fibrous hamartoma of infancy is a rare, benign soft tissue tumor that typically occurs within the first two years of life. The histogenesis is unclear. It is most commonly found in the axilla, shoulder, inguinal region, and chest wall and is usually a solitary malformation located in the subcutaneous tissue or reticular dermis. Local recurrence is uncommon and treatment is largely successful by local excision. The clinical course is typically benign and prognosis excellent. We describe a 7-month-old male with a mass on his left arm that progressively increased in size. The management of a subcutaneous mass in the pediatric patient presents a clinical challenge. The differential diagnosis includes numerous benign and malignant soft tissue tumors. The physical appearance and characteristics of a subcutaneous mass on a child may suggest a malignant process; however, Fibrous Hamartoma of Infancy should be included within the differential diagnosis.

1. Introduction

Subcutaneous soft tissue masses in infants and children encompass a diverse clinical and pathologic spectrum of entities including choristomatous or hamartomatous lesion, malformation, benign, intermediate, and malignant neoplasms. The benign nature, but the rarity and diversity of infantile soft tissue tumors present a diagnostic and therapeutic challenge. The most common category is vascular, followed by fibroblastic-myofibroblastic, fibrohistiocytic, neurogenic and adipose.[1]

"Fibrous hamartoma of infancy is a distinctive, fibrous growth first described by Reye in 1956 as a subdermal fibromatous tumor of infancy."[2] This rare benign lesion with a characteristic histologic appearance usually occurs as a small, rapidly growing, superficial soft tissue swelling developing in the first two years of life. There are few descriptions of the fine needle aspiration biopsy (FNAB) features of this lesion

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2. Case History

7 month old male baby was brought with complaints of swelling in the posterior aspect of the left forearm, noticed for 1 month. Past medical history and family history were noncontributory. The mass had been present since birth and was slowly progressing in size. On examination, swelling in the posterior aspect of left forearm measuring 3X2cms, soft to firm in consistency, non-mobile, not attached with the overlying skin or underlying soft tissues.

1. FNAC- Fine-needle aspiration (FNA) was performed using a 23-gauge needle from multiple sites. Smears were stained with hematoxylin-eosin (H&E) and May-Graunwald-Giemsa (MGG) stain.

2.1. Cytology features

Cellular smears showed clusters of benign spindle cells having elongated nuclei with scant cytoplasm, embedded in fibroblastic stroma

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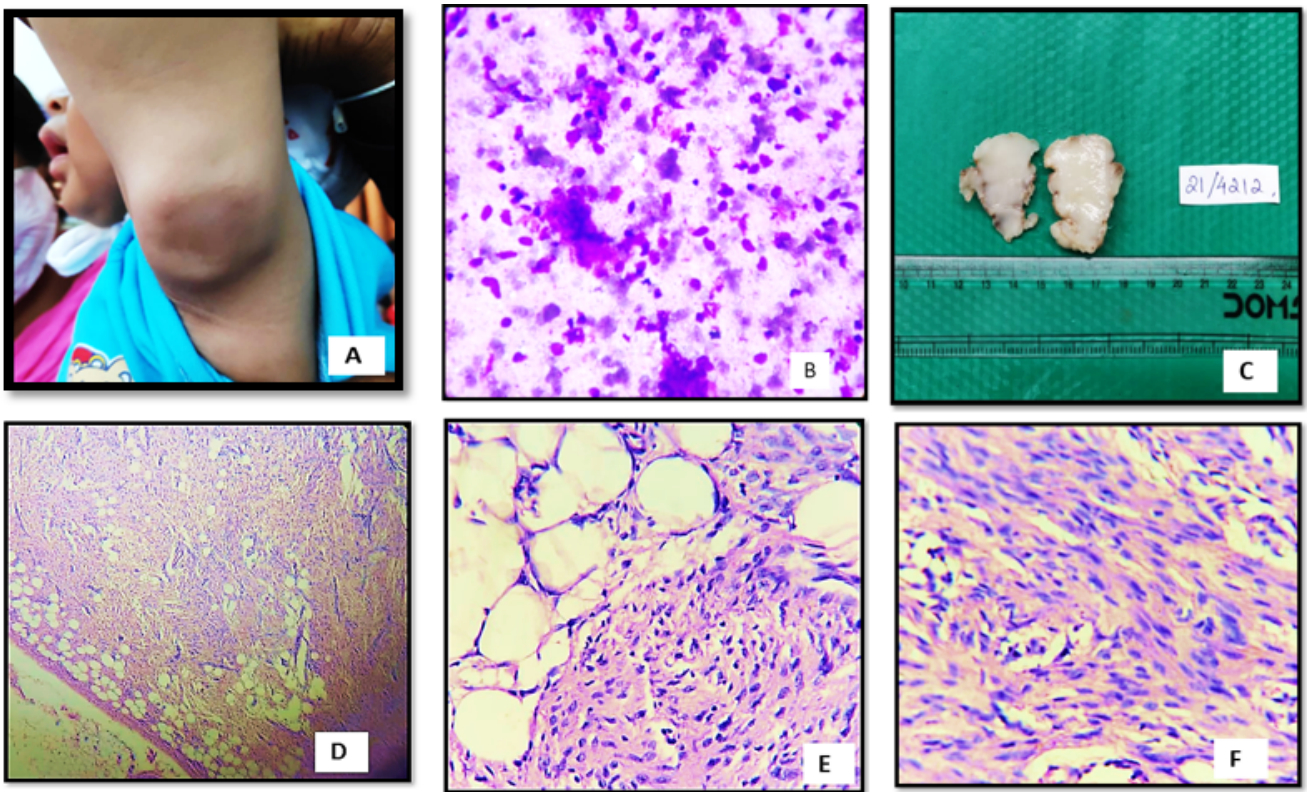

Fig. 1: A: Soft tissue mass in posterior aspect of left forearm measuring 3X2cms; **B:** Cellular smears showed clusters of benign spindle cells having elongated nuclei with scant cytoplasm, embedded in fibroblastic stroma; **C:** lobulated, grey brown. Cut surface- grey white, homogenous; **D,E,F:** poorly circumscribed benign tumor composed of three distinct structures in an organized pattern(H&E)

1. Gross findings- Received a soft tissue mass measuring 3.5x3.5x1.5cms. External surface- lobulated, grey brown. Cut surface- grey white, homogenous.
2. Microscopic features: Section studied shows a poorly circumscribed benign tumor composed of three distinct structures in an organized pattern. There are intersecting trabeculae of fibroblastic and myofibroblastic spindle-shaped cells with bland wavy nuclei. These cells are interspersed by loosely textured islands of small stellate mesenchymal cells with scant cytoplasm embedded in a myxoid matrix. Periphery of the lesion shows mature adipocytes. No mitotic figures were seen.

3. Discussion

Fibrous Hamartoma of infancy is an uncommon non-malignant lesion with a characteristic histologic appearance; it usually occurs as a small, rapidly growing, superficial soft tissue swelling, developing in the first 2 years of life. The majority of our patients were infants, as described in the literature. The affected children are generally healthy and association with other neoplasms or congenital malformations has not been reported.[3] Although the shoulder girdle area and upper arm are the favored and commonest locations, FH may occur anywhere in the body.[4,5] FHs are histologically characterized by an organoid mixture of 3 components: well-defined intersecting trabeculae of fibro-collagenous tissue; loosely textured areas of immature-appearing, small, rounded, primitive mesenchymal cells; and mature fat. In some cases, a pronounced and evident sclerosing process replaces the majority of the lesion.[6] Most FHs were less than 5 cm in diameter as described in the literature, and can reach up to larger than 10 cm. They are usually firm, and may be un-separable and affixed to the underlying tissue, thus causing the concern for malignancy.[7] Three distinct tissues came together to form an organoid structure in the histologic characteristics. The dense fibro-collagenous tissue's well-defined trabeculae were made up of fibroblastic spindle cells with bland, straight, or wavy nuclei. The fibrous trabeculae, which were made up of immature-appearing tiny, round or stellate, primitive mesenchymal cells with minimal/scants cytoplasm, were arranged between the loosely textured islands. These two elements were separated by the mature adipose tissue. The proportions of these 3 components varied greatly from region to region, and in other cases, fat was only visible at the edge. The fibroblastic and primitive cell regions lacked mitotic figures, and none of the

sections displayed nuclear atypia, irregular nuclear contours or hyperchromasia. Clinical differential diagnosis includes a number of entities such as epidermal inclusion cyst, inclusion body fibromatosis, juvenile hyaline fibromatosis, palmoplantar fibromatosis, benign fibrous histiocytoma, dermatofibroma, leiomyosarcoma and fibrosarcoma.[8] Local and conservative excision is usually conducted notably in symptomatic patients and the surgical procedures are often curative.[9] FHI exhibits its special pathological and clinical characteristics. The differential diagnosis of FHI from other soft tissue tumors before operation remains a challenge, which is what we should pay attention to. Sometimes, we have to choose aggressive therapeutic approaches for the treatment of FHI when the tumor is very big and deep to get a satisfactory life quality.[10]

4. Conclusion

FHI lesions have common histological features, but misdiagnosis is possible because clinical knowledge of FHI is limited and its incidence is rare. The clinical course is typically benign and prognosis is excellent. Simple FNAC and biopsy would help in the diagnosis which will prevent aggressive treatment.

5. Source of Funding

None.

6. Conflicts of Interest

There is no conflict of interest.

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