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Title: Leiomyoma of the hand in an adolescent

Running Title: Hand Leiomyoma

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Abstract

Leiomyoma is a solitary benign tumor of smooth muscle and vascular tissue. It can occur anywhere in the body where smooth muscle is present though it is seen for less than 1% of all soft tissue tumors of the upper extremity. It is rarely reported in the hand or in children and is usually seen in the age range of 5-84 years and usually in males.

In this report, we present the case of a 16-year-old patient presenting leiomyoma of the hand.

Keywords

Smooth muscle, Tumor, Hand, Adolescent, Tendon

Introduction

Leiomyoma is a rare benign slow-growing tumor of smooth muscle and vascular tissue and mostly originated from uterine myometrium (1). The development mechanism of the leiomyoma is unclear but congenital origin, disturbances in blood flow, infection and estrogen exposure are suggested (2). It is even more rarer in the upper extremity where it is seen for less than 1% (3-5). There are several isolated primary hand leiomyoma in the literature of which only 108 cases are reported in the literature in English although many cases have been presented at various ages, with male dominance, between the ages of 5-84 (3). The tumor was located from web spaces, digits and volar dorsal surface of the hand (3). In the present report, we present a case of a 16-year-old patient presenting leiomyoma of the hand.

Case Report

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A 16-year-old boy was admitted to our out-patient clinic with loss of motion in the fourth finger and marked swelling on the palm. The patient had his complaints for two years and the mass had grown slowly.

Physical examination revealed a soft, immobile painless mass 3 x 3 cm in diameter. The vascular and sensorial examinations were normal.
Plain radiographs showed no bony involvement. Advanced evaluation with Magnetic Resonance Imaging (MRI) demonstrated a 36 x 22 x 17 mm mass inside the palmar side of the hand which showed an isointense signal on T_1 weighted images and hyperintense on T_2 weighted images. Following the intravenous administration of contrast agent mass showed mild absorption (Fig 1). Before the case presentation, the informed consent form was obtained from the patient's parents.

Removal of the tumor was planned, and routine pre-operative surgical procedures were completed. Under general anesthesia, the skin incision was made directly on palpable mass. Following the careful dissection, the outer surface of the mass was exposed. However, the flexor digitorum profundus tendon was surrounded by leiomyoma. The tumor was dissected carefully from the surrounding tissue along with its capsule. The flexor digitorum profundus tendon had to be sacrificed because of tumor invasion.

The immunohistochemical study showed positive results for smooth muscle indicators such as actin and vimentin but showed no staining for S-100 and CD34 (Fig. 2).
A rehabilitation program was started immediately following the third day post-operatively. The patient was followed_up closely to clarify any possible recurrence. At the one_year follow_up, an MRI showed no sign of recurrence.

Discussion

Leiomyomas are benign tumors of smooth muscle and vascular tissue and mostly originated from uterine myometrium (3). Extremity located leiomyomas are seen dominantly in the lower

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limb (6). In the upper extremity, leiomyomas are seen less than all other soft-tissue tumors (7). These tumors in the lower limb are mostly encountered in females between the ages of 20 and 50 years. However, in the upper extremity there is male dominance (3, 5). Oliver et al. studied 108 leiomyomas localized in the hand (3). Leiomyoma in the pediatric age group is even more uncommon the youngest case being a 5-year-old patient who presented with hand leiomyoma (8). In a recent study, Komforti et al. reported a 3*2 mm subungual leiomyoma in the left thumb of a 16-year-old female with no recurrence after 18 months postoperatively (9). In the present study, we presented a rare case of a leiomyoma with unusual localization in a 16-year-old male adolescent. Our case had a 36*22*17 mm leiomyoma inside the palmar side of the hand and after surgical removal, there was no recurrence at follow-up.

In the review published by Boutayeb in 2008, the pain was the chief complaint of patients and seen in 80% of patients with leiomyomas on the hand (10). Even though leiomyomas might be painless in the early stages, they mostly manifest themselves with pain due to the compression of nerves or intratumoral necrosis (11). In this case, the patient was admitted to the out-patient clinic because he had swelling without pain.

Differential diagnosis of the leiomyoma includes giant cell tumor of the tendon sheath, ganglion cyst, foreign body granuloma. myxoid cyst, inclusion cyst, glomus tumor, angiolipoma, schwannoma, hemangioma and fibromatosis (3, 12). Leiomyomas mostly have well-demarcated borders. In the MRI scan, these lesions are shown hyperintense in T_2, and isointense in T_1 images. However, there are many other soft tissue tumors observed hyperintense in T_2 and isointense T_1 images as well as leiomyomas. Therefore, the histopathologic examination is required to confirm the exact diagnosis (13). In our case, the MRI images were compatible with the literature, but definitive diagnosis could only be made by histopathological evaluation.

In addition, malignant transformation has been shown in these tumors. Malignant transformation has been observed in a patient in literature seven years after the initial surgery (9). In light of these reports, the clinician should always be suspicious about possible recurrence and malign transformation of the tumor. Even though low rates of malign transformation have been reported, the possibility of malign transformation should not be underestimated by the physician. In general, leiomyoma tends to be slow-growing and has a non-changing structure.

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Therefore, patients should be followed up closely for possible future complications. In our case, there was no recurrence after the one-year follow-up.

Conclusion

Following the surgical excision and histopathological examination, a close follow-up of the patient is recommended annually to avoid a late diagnosis of undesired complications such as recurrence and malign transformation.

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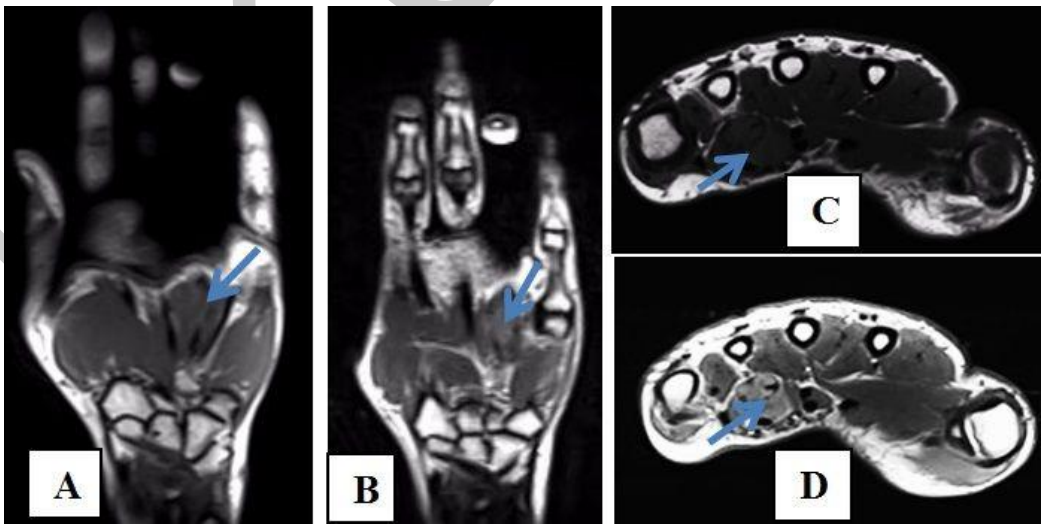
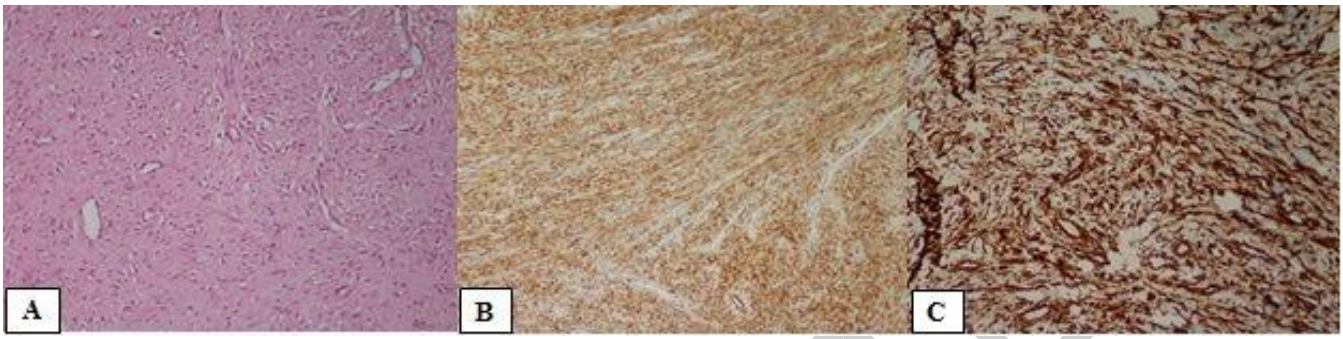
FIGURE LEGENDS

Figure 1: A) Coronal T_{1a} sequence: isointense lesion at the deep part of fourth flexor tendon (blue arrow); **B)** Coronal T_{2a}: mild hyperintense lesion at the deep part of fourth flexor tendon (blue arrow); **C)** Axial T_{1a}: isointense lesion at the deep part of fourth flexor tendon (blue arrow); **D)** Axial T_{1a} with contrast: mild contrast enhancement (blue arrow).

Figure 2: A) Spiculated shaped cytoplasmic smooth muscle cells forming fascicles and bundles (HE staining, x20); **B)** Vimentin staining positive filaments of smooth muscle cells; **C)** Desmin staining positive filaments of smooth muscle cells.





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