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CASE REPORT – OPEN ACCESS

Leiomyoma of the Hand in an Adolescent

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ABSTRACT

Leiomyoma is a solitary benign tumor of the smooth muscle and vascular tissue. It can occur anywhere in the body where smooth muscles are present, although it is seen in <1% of all soft tissue tumors of the upper extremities. It is rarely reported in the hand or in children and is usually seen in patients aged 5–84 years and in males. In this report, we describe the case of a 16-year-old patient presenting with leiomyoma of the hand.

Keywords: Smooth muscle, tumor, hand, adolescent, tendon

INTRODUCTION

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

CASE REPORT

A 16-year-old male was admitted to our outpatient clinic with the loss of motion in the fourth finger and marked swelling on the palm. The patient had the complaints for 2 years, and the mass had grown slowly.

A physical examination revealed a soft, immobile painless mass, 3×3 cm in diameter. The vascular and sensorial examinations were normal.

Plain radiographs showed no bony involvement. An advanced evaluation with magnetic resonance imaging (MRI) demonstrated a 36×22×17 mm mass inside the palmar side of the hand, which showed an isointense signal on T1-weighted images and hyperintense on T2-weighted images. Following the intravenous administration of contrast agent mass showed mild absorption (Fig. 1). Before the case presentation, an 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 the palpable mass. Following a 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 it showed no staining for S-100 and CD34 (Fig. 2).

A rehabilitation program was started immediately following the 3rd day postoperatively. The patient was followed up closely to clarify any possible recurrence. At the 1-year follow-up, an MRI showed no sign of recurrence.

DISCUSSION

Leiomyomas are benign tumors of the smooth muscle and vascular tissue, originating mostly from the uterine myometrium (3). Extremity-located leiomyomas are seen dominantly in the lower limbs (6). In the upper extrem-

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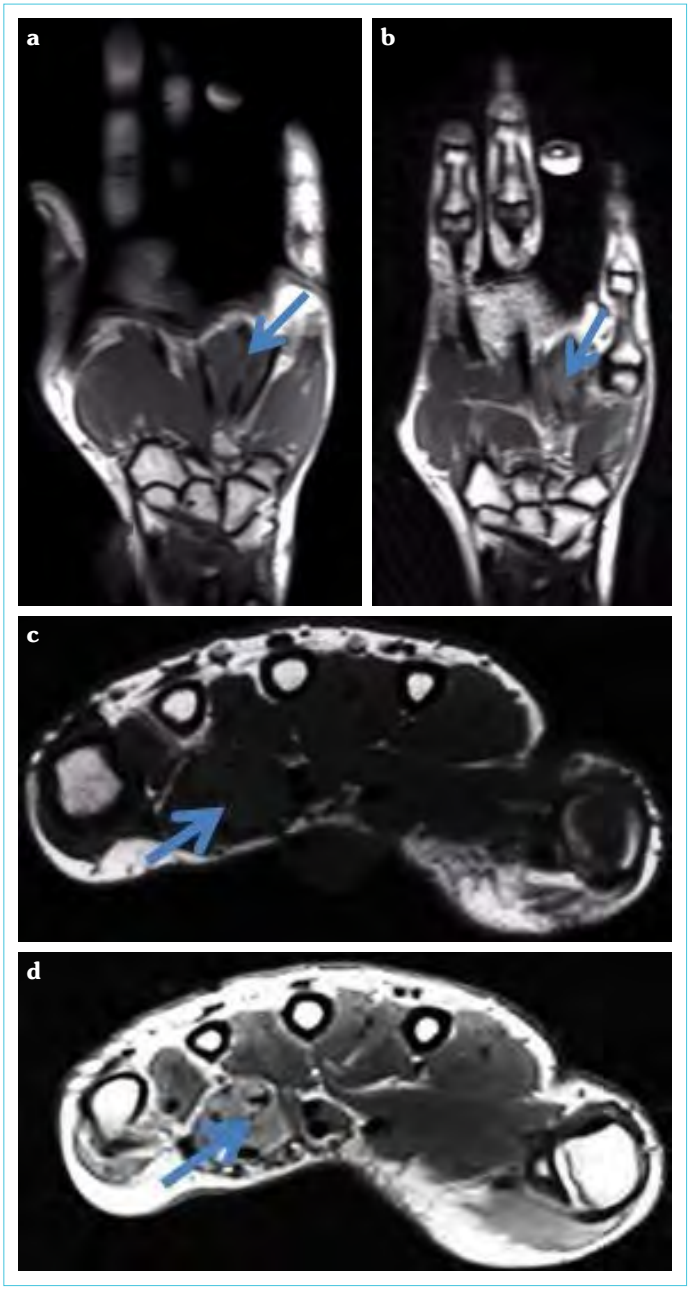

Figure 1. a–d. (a) Coronal T_{1a} sequence: an isointense lesion at the deep part of the fourth flexor tendon (blue arrow); (b) Coronal T_{2a}: a mild hyperintense lesion at the deep part of the fourth flexor tendon (blue arrow); (c) Axial T_{1a}: an isointense lesion at the deep part of the fourth flexor tendon (blue arrow); (d) Axial T_{1a} with contrast: a mild contrast enhancement (blue arrow)

ities, leiomyomas are seen less frequently than all other soft tissue tumors (7). These tumors in the lower limbs are mostly found in females aged 20–50 years. However, in the upper extremities, male cases are predominant (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

Figure 2. a–c. (a) Spiculated cytoplasmic smooth muscle cells forming fascicles and bundles (HE staining, ×20); (b) Vimentin-staining positive filaments of smooth muscle cells; (c) Desmin-staining positive filaments of smooth muscle cells

months postoperatively (9). In the present study, we investigated 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, pain was the chief complaint and seen in 80% of patients with hand leiomyomas (10). Even though leiomyomas might be painless in 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 outpatient clinic because he had swelling without pain.

A differential diagnosis of the leiomyoma includes a giant cells 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. On the MRI scan, these lesions are shown as hyperintense in T2 and isointense in T1 images. However, there are many other soft tissue tumors observed as hyperintense in T2 and isointense in T1 images, as well as leiomyomas. Therefore, a histopathologic examination is required to confirm the exact diagnosis (13). In our case, the MRI images were compatible with the literature, but a definitive diagnosis could only be made by histopathological evaluation.

In addition, malignant transformation has been observed in these tumors. Malignant transformation was found in a patient in the literature 7 years after the initial surgery (9). In light of these reports, the clinician should always be suspicious regarding 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. Therefore, patients should be followed up closely for possible future complications. In our case, there was no recurrence after the 1-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.

Informed Consent: Written informed consent form was obtained from the patient's parents.

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