A Case Report -Bilateral Giant Achilles Tendon Xanthomas Resection with a Tendon transverse Technique

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ABSTRACT

Background : Xanthomatosis is a disease in which large tendon tumors can occur, especially in the Achilles tendon. This disease is a rare interesting orthopaedic condition.

Case Report:A case of a twenty eight year old girl patient with giant bilateral Achilles tendon xanthomas in which both tumors were resected. There was no ulceration on the both sides. The patient was treated by total resection of the lesion and reconstruction using tendon transfer of the Peroneus brevis and Flexor hallusis longus. Postoperative treatment consisted of six weeks lower leg cast immobilization followed by partial weight bearing. After 4 months the patient was able to walk pain free without any difficulties. It has been suggested that total resection with augmentation had associated with fewer complication of recurrent as compare with a subtotal resection. 10 months after surgery our patient had no signs of recurrence of Achilles tendon swelling.

Conclusion: Complete excision of the lesion is needed to reduce recurrence. Reconstruction of the defect is a challenge due to the large defect. Tendon transfer augmentation results in good functional outcome of the patient even in large defects.

Keywords: Xanthomatosis, tendon transfer, Achilles tendon.

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INTRODUCTION

Xanthomatosis is a disorder which is produced due to disturbance in lipid metabolism [1, 2 3, 4, 5, 6]. Xanthoma of the tendon may involve the tendon but can involve the ligament, fascia, or periostium [2]. There is a predilection for metacarpophalangeal joints but the Achilles tendon and patellar tendon may also be involved [2, 6, 7]. Less commonly the lesion may affect the triceps and extensor tendon of toes.

Xanthomatosis is usually slowly growing. The cause is an accumulation of fatty lipids in tendons, especially the Achilles tendons. No data have been published on the incidence, but the benign bilateral Achilles tendon fat accumulation is a rare disease [8,9]. It is mainly found in patients with heterozygous familial hypercholesterolemia [10]

A strong tendency to recurrence was observed in surgically removed Achilles tendon lesions [4,6]. .Large swelling of the tendocalcaneus results in weakness of plantar flexion and difficulty in walking hence surgery is carried out for these lesions We report a case of a patient with giant bilateral Achilles tendon xanthomas, not known with familial hypercholesterolemia, in which both tumors were resected with a tendon transfer technique.

CASE REPORT

A twenty eight year old woman presented at consultant clinic in Al-Kindy teaching hospital for a second opinion. She had complaints of pain and swelling in both Achilles tendons. The swellings begun several years ago and were progressive. Finally she had difficulties wearing footwear and increasing pain sensation especially during long distance walking , swelling was insidious in onset, gradually progressive not associated with fever and no history of trauma. There was no discharge from the ulcer. There was no history of familial

hypercholesterolemia. On examination the patient was moderately built. The swelling in both the heel were measuring about 18cms in length and 7 cm width. The non-tender swelling was firm in consistency with multiple nodules. The swelling were mobile, no ulcer was found in both ankles. There were 10 degrees of dorsiflexion and 20 degrees of plantar flexion in both right and left ankle.

Investigations: The blood count was normal. ESR was 12, blood glucose was 100 mg/dl, and uric acid was 3.2mg/dl, cholesterol of 160mg/dl and triglycerides 141 mg/dl. Ultrasound showed diffuse nodular thickening in the tendoachilles with hypoechoic areas suggestive of xanthomata. MRI showed diffuse speckled appearance on both axial images. This speckled appearance was more obvious on T1fat suppressed images than on T1water suppressed images which is suggestive of xanthomatosis

Treatment

Patient was operated under spinal anesthesia with 25cm midline incision. On exploration the tendon was replaced by yellowish hard nodular mass extending from the musculotendinous junction to the calcaneus. The lesion was measuring about 18cms in length and 7 cm in width (maximum at the base).Since there was total involvement of the tendon it was resected completely. Tendons transfer of Peroneus brevis and Flexor hallusis longus were done and it was sutured distally to the calcaneus by passing drill holes through bone and proximally to the musculotendinous junction using non-absorbable sutures, below knee plaster in plantar flexion was given postoperatively. Fig 1,2 ,3 and 4

Partial weight bearing was started after 8 weeks. Four months after the surgery patient was able to bear full weight. Surgery on the right ankle was done after 3 months .Clinical assessment was done after 9 months post-surgery on left ankle and 6 months on right side. There were 15 degrees of dorsiflexion and 35 degrees of plantar flexion on both ankles.

There was no calf muscle atrophy .Patient was able to walk without a calcaneus limp. Patient was able to stand on tiptoe without support. Physical activity at home and at work was normal. After 9 months of follow up there was no recurrence of the lesion clinically and confirmed by Ultrasound.

Histopathology: Microscopic examination showed foam cells, polyhedral cells with pale vesicular cytoplasm with round shaped nuclei. The stroma consists of stromal cells with granular cytoplasm and oval nuclei. Numerous vesicular lipoid filled spaces were found in the tissues. The tumor was surrounded by fibrous capsule .Giant cells were present in the areas of hemorrhage. There were numerous fibrous septae which divided the tumor into lobules. The microscopic diagnosis was xanthomata.

Fig 1: incision and Xanthoma before excise

Fig 2: Xanthoma after excise

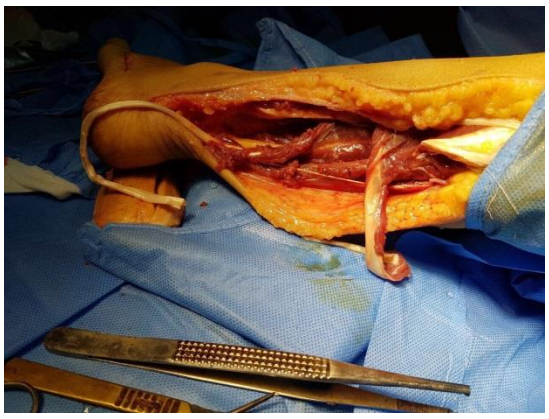

Fig 3: Tendons transfer before suturing

Fig 4: Tendons transfer after suturing

DISCUSSION

Achilles tendon xanthomatosis is a rare disease. It is strongly linked to familial hypercholesterolemia. Interestingly, our patient was not known with familial hypercholesterolemia. A raise of serum LDL cholesterol could not be found in multiple serum samples. However, this does not exclude a dyslipidemia because we did not perform a detailed lipid analysis [11].

Xanthomatosis of the ankle is a rare lesion and tends to recur after incomplete excision as stated by Tetsuya et al [3]. The tendon lesions are invariably multiple and may be associated with other lesions like tuber xanthoma, xanthelesma of the skin, arcus cornea and coronary artery disease [1]. Although xanthoma is a lesion that occurs due to disturbance of lipid metabolism S.K Roy [2] as in our patient, reported that the lesion can also occur in patients without hypercholesterolemia. The xanthomatous lesions of the tendon usually involve the extensor tendon of metacarpophalngeal joints of the fingers [2]. This is in contrast to a review done by Morris S. Friedman [6], in which it is stated that about 60 percent of the tumors in the wrist and fingers were found on the flexor surface and 40 percent on the

extensor surface. Larger tendon like patellar tendon, triceps tendon is involved less commonly. The lesion may affect the extensor tendons of the toe and plantar fascia. Involvement of Achilles tendon occurs in 50% of patients [6].

We decided to perform a tendon transfer resection of the xanthomas. However macroscopically both tumors were excised in total, its infiltration into the tendon made a subtotal resection inevitable. Carranza-Bencano et al. stated, in case of severe xanthomas infiltration, total resection is the best surgical technique [8] to avoid the risk of recurrence. As summarized by Huang et al. there are several procedures for Achilles tendon reconstruction. Autogeneous tendon grafts such as the peroneus brevis or flexor hallucis longus are commonly used. The authors described a reconstruction with a tibialis posterior auto-graft, which was necessarily because of a large tendon defect after extensive resection [12].

In our patient the size of the lesion was eighteen centimeters and the largest size reported by far is twenty-two centimeters described by Morris S. Friedman [6]. Treatment is primarily directed towards lowering the serum cholesterol level some patients require surgical removal of the lesion which is painful and disfiguring [1, 2, 5, 6]. Radiation treatment has proven not to be beneficial in treating these lesions [6]. Local excision or subtotal resection of the lesion and its association with recurrence has been reported by many authors [4, 6]. Recurrence rate of partial resection of the lesion ranges from 12% to 15% [6]. Because of the high rate of recurrence after partial resection of tendon total resection of the tendon between the musculotendinous junction and calcaneus is recommended especially when there is severe infiltration of the lesion [3].

Reconstruction of the Achilles tendon from musculotendinous junction to calcaneus remains challenging task. A number of procedures has been described for reconstruction of tendocalcaneus. The use of a local tendon like flexor hallucis longus and peroneus brevis has been suggested [13,14].Boopalan et al[15], suggested the use of free autologous tendon graft like gracilis, semitendinosis and fascialata. The use of free tendoAchilles tendon allograft has also been reported [16], other use Tensor fascia lata be sufficient for reconstruction Tetsuya tomita et al[3]. In our patient was carried using peroneus brevis and flexor hallucis longus tendons will be sufficient, we did not consider allograft because it is not available to us. We believe that autograft will integrate better than an allograft and it agreement with Huang et al [12].

The patient started weight bearing after 6 weeks and this parallel to Moroney et al [10], where others reported non-weight bearing periods for nine weeks [12].

CONCLUSION

Xanthomatosis of ankle is a rare interesting orthopaedic condition. Recurrence is more common in these lesions due to incomplete removal of the lesion. Reconstruction of the defect after complete excision remains challenging task. Though variable options are available reconstruction using peroneus brevis and flexor hallucis longus tendons remains a good option and the results are comparable to reconstruction done by tendocalcaneus allograft.

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