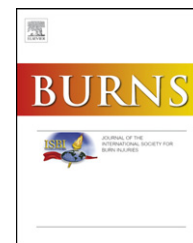


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Case report

Pacinian hypertrophy in a type 2A hand burn contracture and Pacinian hypertrophy and hyperplasia in a Dupuytren's contracture

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1. Introduction

Several distinctive encapsulated sensory receptors exist in mammalian skin. Specific examples are: Meissner corpuscles, Krause endings, Ruffini endings and Pacinian corpuscles. Filippo Pacini was an Italian anatomist (1812–1883), and described the corpuscle bearing his name. Pacini's corpuscle is a lamellated nerve ending, and its shape resembles that of a small onion. This laminar capsule acts as a filter, allowing only transient disturbances at high frequencies to activate the nerve endings concerned with the perception of pressure and known as corpuscula lamellosa. Pacinian corpuscles are also found in the mesentery, joints, ligaments and deep connective tissue, where they act as mechanoreceptors and innervation is by fast-conducting myelinated fibres.

Pacinian corpuscles make up 10–15% of the mechanosensory innervation in the hand, adapting more rapidly than Meissner corpuscles and with a lower response threshold. The properties of Pacinian afferents make them well suited to detect vibrations transmitted through objects that contact

the hand or are being grasped in the hand—no doubt important for using a wrench, cutting bread with a knife, writing, etc. [11,33–35].

Studies of Pacinian corpuscles are not only recent [1]. Although many of the relevant articles include case reports of one or two patients, there are also studies performed on a wide range of animal species from mice to elephants [2–6] and experimental studies concerned with Pacinian corpuscle analysis. In one of these, Pacinian corpuscles were transplanted from the cruris into the brain in rats; no reinnervation was observed on the corpuscles following transplantation [7]. In another study in rats, the sciatic nerve was dissected and, over a period from 1 to 20 weeks after surgery, a total of 15 corpuscles were observed for atrophy using a surgical microscope. As in a similar study in cats, the capsular layers were found to remain within the normal range following denervation [8,9].

In this paper, we report two cases of Pacinian corpuscle hyperplasia and hypertrophy, one associated with bilateral Dupuytren's contractures and another with a type 2A palmar

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burn contracture. To our knowledge, this is the first published description of such an association.

2. Cases reports

2.1. Case 1

A 72-year-old man presented complaining of inability to fully extend his hands and of cord-like tumours on his palms, but no pain; this indicated a Dupuytren's contracture. Further questioning revealed that he had not been receiving any drugs or alcohol except drugs for hypertension. Two teams performed bilateral palmar fasciectomy in one session, using a pneumatic tourniquet and general anaesthesia. A Penrose drain was inserted under the flaps after the operation. During surgery we observed some small white-coloured particles in the fatty tissue outside the palmar fascia. We decided to remove these with the palmar fascia while protecting digital neurovascular structures. Pathological examination revealed that the particles were hypertrophic Pacinian corpuscles.

2.2. Case 2

A 55-year-old woman presented complaining of pain and inability to extend her right second, third and fourth fingers. The initial history was a hand burn from touching a hot stove 2 years previously. Further questioning revealed that she had grand mal-type epilepsy and had been using phenobarbital. The wound had healed in 2 months using dressings only, but this resulted in contractures of the three fingers. After 2 years she sought medical assistance, and the condition was diagnosed as a type 2A burn contracture. Although simple Z-plasty would have corrected the burn contracture, we aimed to restore full extension of the fingers after releasing them with Z-plasties, and thus found a skin defect. During operation we did not remove any scar tissue. As the fingers were released, we observed in the second finger near the digital nerve one hypertrophic Pacinian corpuscle, which we removed for histopathological examination. In addition to performing Z-plasties, we transplanted split-thickness skin grafts taken from the medial plantar region. No drain was inserted.

Some characteristics of the two cases are presented in the Table 1. Both patients received general anaesthesia and a pneumatic tourniquet was applied to their extremities. No systemic antibiotic therapy was administered, and the presence of Pacinian corpuscles did not change the operative management in either case. There were no complications and outcomes were uneventful.

3. Discussion

Skin sensations depend on several different kinds of receptors: basket endings are nerves around the hair follicles of the skin, and they relay touch signals when the hairs are moved; free nerve endings are receptors for touch, temperature and pain; and there are deep-lying receptor cells for pressure and touch [10].

Although at rare intervals reports of the existence of Pacinian corpuscles in some tissues, or their hyperplasia and hypertrophy, appear in the literature [12], today we still cannot say that certain tissues do not contain the corpuscle or that some diseases cannot be associated with it. A paper published in 1994, to emphasise the rarity of the condition, stated that only 13 cases had been published [13]. In 1998 the authors of another paper, stating that only 29 cases existed in the literature, argued that these cases neither be considered as real tumours nor as connected with neurofibromatosis [14].

Among many anatomical studies relating to Pacinian corpuscles is a cadaveric study in which six fresh human cadavers were dissected and interphalangeal and metacarpophalangeal joints were analysed for Pacinian corpuscles. The palmar capsule was found to include consistently many more free nerve endings than the dorsal and lateral capsules. In the same document it was emphasised that the majority of the encapsulated endings were Pacinian corpuscles [15]. When hypertrophied, a Pacinian corpuscle may include 35–60 concentric lamellae with an increased diameter of the corpuscle (e.g. 1.6–3.2 mm), whereas normal control specimens show 13–15 lamellae [13]. From the article of Stark et al. we learn that the total mean number of Pacinian corpuscles found in the hand was 300, reported as 44–60% in the fingers, 23–48% in the metacarpophalangeal area and 8–18% in the thenar and hypothenar regions. The corpuscles were smaller in palmar skin overlying the distal phalanx [19].

Pacinian corpuscles have rarely been reported in the lamina propria of the human urinary bladder [16], their possible function being likened to the modulatory function of the noradrenergic fibres in the Pacinian corpuscles of cats [6]. A single enlarged Pacinian corpuscle was described in the mature adipose tissue of human buccal mucosa when an excision had been performed to relieve pain following trauma [17]. Among other rare localisations of the Pacinian corpuscle was the vulvar region of a 27-year-old woman [18]. A case has been reported of glomus tumour Pacinian hyperplasia in the index finger treated by surgical excision [20], and it was stated that Pacinian corpuscle hyperplasia should be considered in the differential diagnosis of digital, palmar or foot pain [21–23]. Hypertrophy of Pacinian corpuscles associated with finger pain and decreased sensation experienced by a 24-year-old woman has also been

Table 1 – Summary of the features of the two cases

Case	Diagnosis	Location of PCs	Complaint	Surgery	PCs seen
1. 72-year-old man	Dupuytren's contracture	Palmar fatty tissue	Inability to extend 4th fingers	Palmar fasciectomy	>10
2. 55-year-old woman	Type 2A burn contracture	Subepineurally in proximal phalanx	Inability to extend right 2nd to 4th fingers, pain	Z-plasty, skin graft	1

PC, Pacinian corpuscle.



Fig. 1 - Bilateral Dupuytren's contracture of 72-year-old man.



Fig. 2 - Intraoperative appearance of palmar fascia and surrounding fatty tissue including cluster of Pacinian corpuscles.



Fig. 3 - Close-up view of Pacinian corpuscles.

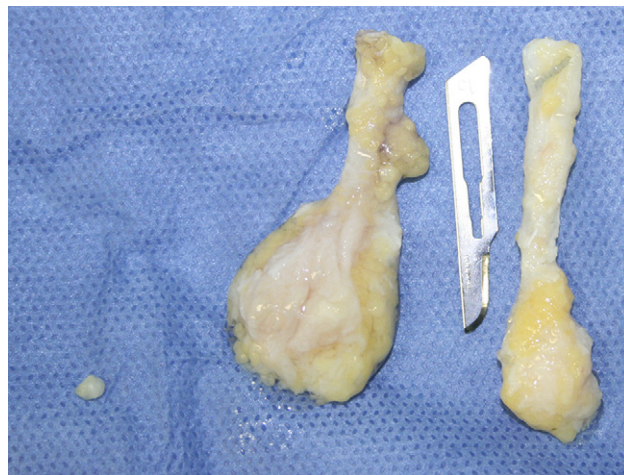


Fig. 4 - Resected materials with surgical blade between to aid comparison. The smallest material comprises Pacinian corpuscles isolated from the right hand specimen, which is seen in the middle.

documented [24]. It was suggested in some case reports that pain may be a result of compression of the nerve by enlarged Pacinian corpuscles [25,27]. Proliferation of the Pacinian corpuscles may also be associated with Raynaud-like symptoms causing marked glomus-type changes in the arteriovenous anastomoses of the fingertip [26].

Coexistence of Pacinian corpuscle hyperplasia and Dupuytren's contracture has been mentioned a few times in the literature [28-30]. In the Dupuytren's contractures that we treated, we did not have any preoperative question in our minds as to the presence of hyperplastic Pacinian corpuscles. In the first case, of bilateral Dupuytren's contracture, two teams operated simultaneously. The team operating on the right hand observed small, rice-shaped, pearl-coloured corpuscles in the fatty tissue (Figs. 1-6), whereas the team working on the left hand did not. The reason for the discrepancy may be the greater amount of fatty tissue found around the contracted palmar fascia on the right side than on the left. Most of the reports cited in this article emphasised that the Pacinian corpuscles were identified in fatty tissues,



Fig. 5 - Type 2A burn contracture of the left hand.



Fig. 6 – Appearance of palmar fatty tissue with rice-sized nodules at epineural level, palmar side of finger, following burn contracture release with Z-plasties.

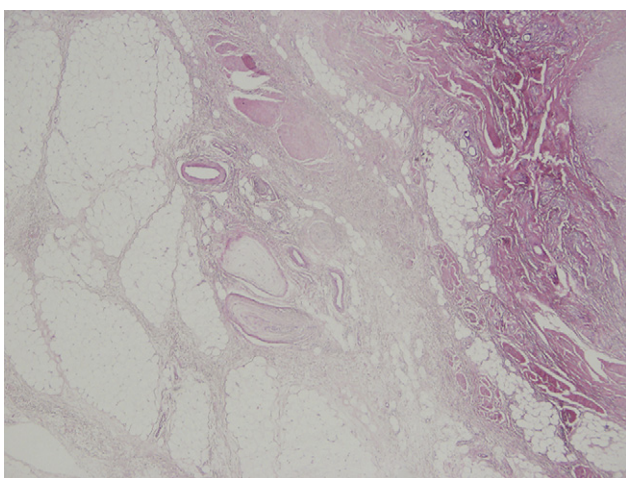


Fig. 7 – Normal Pacinian corpuscles (haematoxylin and eosin, 40×).

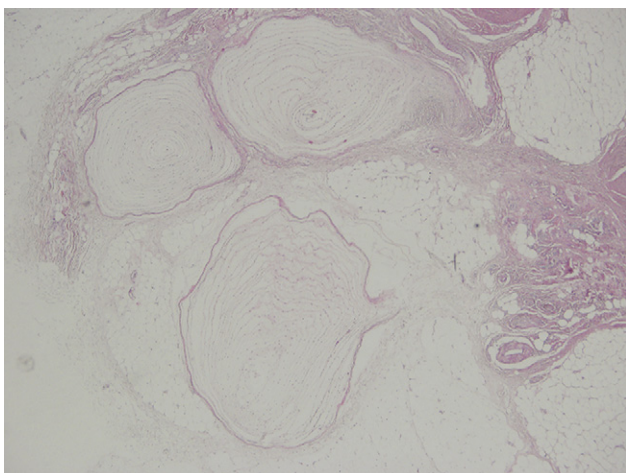


Fig. 8 – Hypertrophic Pacinian corpuscles in same specimen as Fig. 7 (haematoxylin and eosin, 40×).

except for the case of a Pacinian corpuscle found in the lamina propria of the bladder, where no hyperplasia was suggested [16]. Histopathological examination of tissues dissected from the hands revealed the existence of Pacinian corpuscles on both sides (Figs. 7 and 8). One of the three articles [28–30] concerned with Dupuytren's contracture compared the number and sizes of Pacinian corpuscles in the hands of 5 people with Dupuytren's contracture and of 17 people without Dupuytren's contracture; on gross observation, Pacinian corpuscles from the Dupuytren's cases were larger and more numerous and, when measured microscopically, the corpuscles in diseased fascia were larger and composed of more layers [30].

Type 2A burn contracture denotes a neglected burn contracture that can be treated by only skin surgery and does not affect tissues other than the skin [31]. We have been trying to cope with burn contracture problems for more than 15 years, but this report is our first observation of an enlarged Pacinian corpuscle under a burn contracture as a unique case.

A classification of Pacinian corpuscle enlargement was proposed by Rhode and Jennings and described by Calder et al. [32]. According to that classification there are four types: type A, a single enlarged subepineural corpuscle; type B, a grape-like cluster of Pacinian corpuscles attached to the nerve by a fine filament; type C, a series of enlarged corpuscles located in tandem; type D, many hyperplastic corpuscles located along a digital nerve. It seems that our first case, associated with Dupuytren's contracture, can be considered as type B (Fig. 2) and our second case, associated with burn contracture, can be considered as type A (Figs. 5 and 6).

4. Conclusion

No longer can hypertrophy of a Pacinian corpuscle be considered a rarity; however, to our knowledge, this is the first report of coexistence of burn contracture and Pacinian corpuscle hypertrophy.

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