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Case Report

Extra nuchal-type fibroma associated with repetitive blunt trauma during religious activities

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ABSTRACT

Nuchal-type fibroma (NTF) is a rare, benign subcutaneous tumor that usually arises from the posterior neck. NTF is histologically characterised by dense collagen bundles and sparse fibroblasts. Only four trauma-related cases have been previously published. Herein, we present a case of extra NTF with histopathology, and six palanquin porters by using snowball sampling technique in ethnographic field research. A palanquin is a type of human-powered transport carried upon the shoulders mostly seen in religious processions. All individuals (mean age, 26.8 years) displayed similar shoulder masses measuring up to 12 cm in the greatest dimensions. They averaged approximately 8.1 years of palanquin-carrying work each. We believe that long-term, heavy shoulder weight bearing of palanquins in religious dance performances may attribute to the incidence of extra NTF. This study reviews all literature of trauma-associated NTF through PubMed database, and highlights the association between repetitive blunt trauma and the development of NTF.

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Introduction

NTF represents a rare, fibrocollagenous tumor that typically originates from the cervicodorsal region [1]. Extra NTF occurs at extra-nuchal locations. The goal of the study is to investigate the prevalence of extra

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NTF caused by repetitive blunt trauma during religious shoulder-bearing activities. Between February 19, 2015 and March 5, 2015, research participants were recruited from a Taoist temple procession in Kaohsiung City, Taiwan. As observer-as-participants, we used the snowball sampling technique to identify potential respondents with history of participation in the palanquin-carrying ritual linked with our previously pathologically confirmed case of extra NTF. Also, we used PubMed database to retrieve all literature of trauma-associated NTFs in comparison with our present cases.

Case report

A 24-year-old male presented to our plastic surgery department with a firm, painless mass over his right shoulder without restriction of movement. The lesion had gradually increased in size over time. He asked for surgical removal of the mass due to aesthetic concern. The patient had been a religious palanquin porter for more than eight years (Fig. 1). He denied any other local or systemic pathology. Physical examination revealed an erythematous, ellipsoid, solid, and immobile lesion 9×7 cm in size.

Harboring the suspicion that repetitive blunt trauma might associate with the lesion, we implemented the snowball sampling strategy in ethnographic study, and subsequently identified additional six palanquin porters with similar shoulder masses. The lesions were observed over the palanquin shaft's friction areas (Fig. 2). Axial T1- and T2-weighted MRI examinations revealed a low-signal-intensity subcutaneous mass overlying the right posterolateral side of the trapezius muscle. There was no evidence of bone invasion (Fig. 3).

A complete excision of the tumor was performed under general anesthesia. At the section level, the tumor was found to involve the dermal layer with a whitish, fibrotic tissue attached to the surface. The tumor's gross appearance was reddish-tan in color, firm, homogenous, non-encapsulated, and poorly circumscribed. Microscopic examination revealed a poorly demarcated lesion composed of thick collagen bundles, and interspersed adipose tissue with variable-sized adipocytes. Entrapped traumatic neuromas were observed. The sclerosing lesion was paucicellular, and contained CD34-positive, β -catenin-negative fibroblasts infiltrating subcutis. The adipocytic immunostaining result of CDK4 was negative. The pathological findings of the subcutaneous mass were consistent with the diagnosis of NTF (Fig. 4). At six-month follow-up, the patient still maintained normal range of motion of the right shoulder without local recurrence. The patient had also discontinued participation in the palanquin-carrying ritual.



Fig. 1. Photograph showing eight devotees holding deity statue on palanquin that weighs approximately 180 kg in religious dance routines during a Taoist festival.

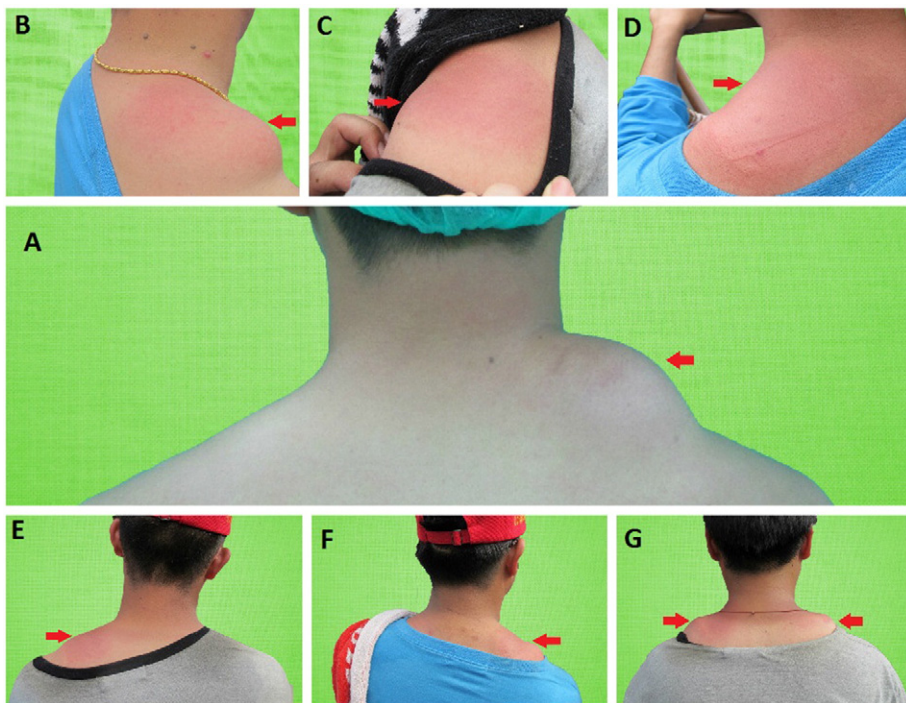


Fig. 2. Photographs showing trauma-associated shoulder masses (arrows) of pathologically confirmed extra NTF (A) and the six participants in the palanquin-carrying ritual (B to G).

Discussion

Eight shoulder masses were analysed among seven patients (Table 1). The patients were healthy young males, aged from 21 to 32 years (mean, 26.8 years). Each patient had the same background history of chronic shoulder masses. A palanquin holding deity statue (approximately 180 kg) is carried by eight porters in

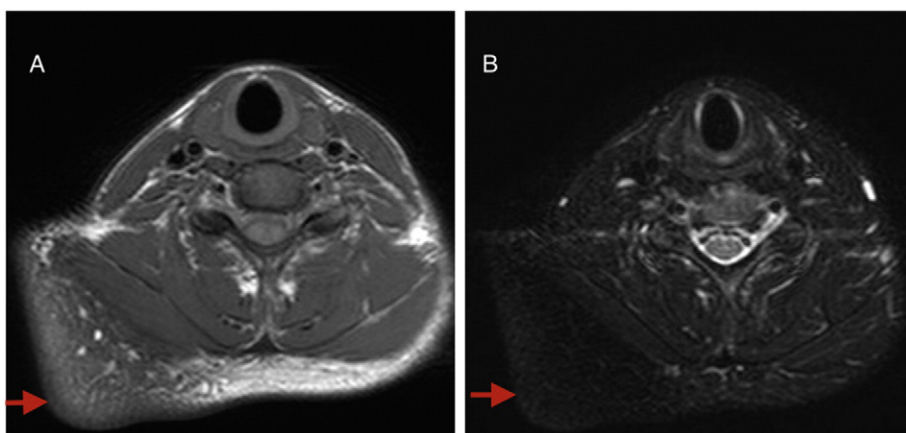


Fig. 3. A. Axial T1-weighted spin echo MR image demonstrated a low-signal-intensity subcutaneous mass (arrow) overlying the posterolateral side of the trapezius muscle. B. Corresponding axial T2-weighted short-tau inversion recovery (STIR) revealed the same subcutaneous mass with low signal intensity (arrow).

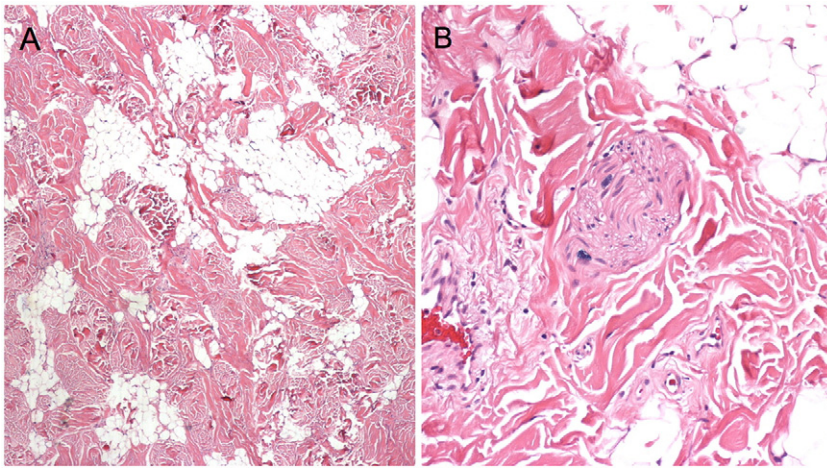


Fig. 4. A. Histology of NTF showing randomly oriented dense collagen bundles that are mixed with interspersed adipose tissue and variable-sized adipocytes. Haphazard CD-34 positive fibroblasts infiltrate the dermis and subcutis (hematoxylin and eosin, $\times 100$). B. Proliferative entrapped nerve fibres within the sclerosing tissue resembling traumatic neuroma are also readily observed (hematoxylin and eosin, $\times 200$).

religious dance routines. Each participant in the ritual must support at least 22.5 kg on their shoulders. While dancing, the wooden shaft placed on their shoulders will repetitively impact the participants' shoulders. The average duration of each participant's weight bearing history was 8.1 years. The patients' masses all were similar in appearance. They were erythematous and indurated, measuring from 7.5 to 12 cm in the greatest dimensions (mean, 9.1 cm).

Since NTF was first introduced by Enzinger and Weiss [1], 55 cases of NTFs and 44 cases of extra NTFs have been reported. NTF has been associated with diabetes mellitus, Gardner's syndrome, scleredema, and trauma. To date, only 11 cases with antecedent trauma have ever been documented to implicate in the development of NTF (Table 2) [2–5]. Collectively, the mean age of trauma-associated NTFs was 31.2 years, and the maximal sizes of the lesions ranged from 4.8 to 12 cm. The durations were between 2 and 12 years (mean, 11.3 years). To the best of our knowledge, this is the first published study of extra NTF involved in chronic blunt trauma in religious practices.

MRI is an efficient modality of choice for differential diagnosis of NTF because it readily defines the fibrous lesions. NTF is characterised by its low cellularity with dense fibrous network and minimal water content [6]. It usually shows ill-defined, low signal intensity in T1- and T2-weighted images. Histopathologically, elastofibroma is a part of the differential diagnosis because of its attribution to chronic trauma. Nevertheless, the examination reveals an absence to scanty elastic fibres of the tumor. Fibrolipoma, unlike NTF, usually shows encapsulation without nerve entrapment [2,6,7]. In addition, negative beta-catenin and positive CD34 expression permit the exclusions of desmoid-type fibromatosis and collagenous fibroma, respectively.

Table 1
Demographics of the present cases.

Patient	Age (years)/sex	Duration	Symptoms	Location	Size (cm)	Gardner's syndrome	Diabetes mellitus
A	24/male	8 years	No	Right shoulder	9 \times 7	No	No
B	30/male	12 years	No	Right shoulder	12 \times 10	No	No
C	26/male	7 years	No	Left shoulder	10 \times 8	No	No
D	32/male	10 years	No	Left shoulder	8.5 \times 7.0	No	No
E	21/male	4 years	No	Left shoulder	7.5 \times 6.0	No	No
F	28/male	8 years	No	Right shoulder	9.5 \times 7.0	No	No
G	24/male	8 years	No	Right and left shoulders	8.5 \times 6.5; 7.5 \times 7.0	No	No

Patients are listed in the same order shown in Fig. 2.

Table 2

Literature review of trauma-associated NTF.

Source	Cases	Age (years)/sex	Size (cm)	Location	Duration	Trauma type	GS	DM	Scleredema
Banney et al. (2000)	1	53/M	NR	Posterior neck	30 years	Organic solvent exposure	NR	NR	Yes
Karonidis et al. (2007)	1	38/M	3.5	Posterior neck	Several years	NR	No	No	No
Sraj et al. (2008)	1	20/M	6.0	Ankle	5 years	Sustained minor contusion	NR	NR	NR
Linos et al. (2011)	1	54/M	4.8	Buttock	>2 years	Surgeries; medical problems	Yes	No	No
This report (2015)	7	21 to 32/M	From 7.5 × 7.0 to 12 × 10	Right shoulder (4); left shoulder (3)	4–12 years	Repetitive blunt trauma	No	No	No

M: male; F: female; GS: Gardner's syndrome; DM: diabetes mellitus; NR: not reported.

Dermatofibroma differs from NTF because the overlying epidermis usually shows a pseudoepitheliomatous hyperplasia and hyperpigmentation of basal cell layer [8].

Palanquin-bearing is a common folk religious ritual worldwide; such cultural consistency reflects that the prevalence of palanquin porters' shoulder masses may be underestimated due to underreporting. NTF has benign clinical course, and is often misdiagnosed. We would like to emphasise the association between repetitive, blunt shoulder trauma and the development of NTF, and bring such awareness to clinicians.

Conflict of interest statement

The authors declare that there is no conflict of interest regarding the publication of this paper.

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