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Atypical presentation of axillary web syndrome (AWS) in a male squash player: a case report

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Axillary Web Syndrome (AWS), also known as lymphatic cording, refers to a condition in which a rope-like soft-tissue density develops in the axilla. It usually appears in the 5 to 8 week period following breast cancer surgery and can lead to shoulder pain and restricted motion. We present a case of AWS in a male squash player with no history of breast cancer or surgery following a period of intense exercise. This case highlights the rare presentation of AWS in a male patient and raises awareness for the health care practitioner who may not suspect this condition in this population.

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KEY WORDS: chiropractic, axillary web syndrome, lymphatic cording

Le syndrome des cordelettes axillaires (AWS), également connu sous le nom de thromboses lymphatiques, se réfère à une condition dans laquelle des tissus mous denses comme une corde se forment dans la région de l'aisselle. Ceci apparaît généralement dans 5 à 8 semaines après une chirurgie du cancer du sein et peut entraîner des douleurs à l'épaule et limiter les mouvements. Nous présentons un cas d'AWS chez un joueur masculin de squash, sans antécédents de cancer du sein ou de chirurgie du sein, après une période intense d'exercice. Ce cas met en évidence la rare présentation de l'AWS chez un patient masculin et sert à sensibiliser le professionnel de la santé qui ne pourrait pas soupçonner cette condition chez cette population.

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MOTS CLÉS : chiropratique, syndrome des cordelettes axillaires, thromboses lymphatiques

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Patient consent was obtained for the use of clinical information and imaging with respect to this case report.

Introduction

Axillary Web Syndrome (AWS), also known as lymphatic cording, refers to a condition in which a rope-like soft-tissue density develops in the axilla.[1] A systematic review by Yeung et al. reported many synonymous names for AWS including axillary variant of Mondor disease, cording, lymphatic cord, cording lymphoedema, webbing, axillary web cord, vascular string, lymphatic thrombosis, axillary string, and fiddle-string phenomenon, among others.[2] The cording can extend further down into the arm and forearm and is made taut and painful by shoulder abduction.[1] AWS is typically the result of surgical treatment for breast cancer such as sentinel lymph node biopsy (SLNB) or axillary lymph node dissection (ALND), however the exact pathophysiology is currently not well established.[2,3] While AWS is often self-limiting, it can cause psychological and physical morbidity up to and beyond the typical resolution period of 5 to 8 weeks.[4] There are several risk factors for AWS which include type and extent of surgery, age, BMI, ethnicity, oncological treatment and healing complications.[2] We present an atypical presentation of AWS in a male patient with no history of breast cancer or surgery in which the cording appeared following an intense round of squash. To our knowledge, this is the first case in the literature to report this syndrome following a bout of high intensity exercise. We will also highlight the signs, symptoms, and risk factors associated with AWS to raise awareness of this condition as it is not commonly seen in general practice, and briefly discuss treatment options for manual therapists.

Case Presentation

A 38-year-old competitive squash player and commercial pilot presented to our clinic noting the recent appearance of a subcutaneous cord along the medial portion of his right arm extending into the axilla, approximately 10cm in length (Figure 1). He did not relate any other contributing factor to the appearance of this cord. The cord appeared 8 days earlier following a particularly aggressive game of squash 24 hours prior. The cord was only visible with the arm abducted to at least 60 degrees and he described it as moderately tender to touch and mildly uncomfortable at rest, and did not cause him any functional limitations. The tissue appeared to be adhered to the skin at the distal end and deformed the skin in a crinkling appearance when the arm was moved (Figure 2). The patient related an active lifestyle including competitive squash, kite sailing, jogging, and beach volleyball. He has a family history of renal cancer in his father, hepatic cancer in his paternal grandmother, and metastatic cancer

Figure 1.
Thin, compressible cord of soft tissue within the subcutaneous tissue of the axillary region.

in his maternal grandmother. He was normotensive, does not take medications for any condition, and has a history of minor knee, back injuries, and cervicogenic dorsalgia that were managed conservatively. The patient reported no significant history of right shoulder or arm injury. He does not use recreational drugs or drink alcohol to excess. Upon physical examination, the patient is 5'4" tall with a medium athletic build and normal posture. He had normal distal pulses, and bilaterally symmetrical sensation, motor response, and reflexes of the upper limbs. He did not have any signs or symptoms of thoracic outlet syndrome. On abduction of the right arm to 90 degrees, a fibrous subcutaneous cord was observed extending 10 cm from the axilla along the medial aspect of the arm. On palpation, the cord was extremely thin, like butcher's string, and collapsed under minimal pressure.

The patient underwent one treatment of manual therapy including soft-tissue mobilization and friction-type myofascial release as well as advice on how to perform self-massage at home, which he only completed a few times by his account. The soft-tissue therapy was applied over the skin of the cord, while the patient brought the shoulder into extension at 90 degrees of abduction. The patient was travelling for the subsequent 4 weeks and could not return for further treatment. At 2 weeks, the patient contacted the practitioner and noted no longer feeling any discomfort from the cording. One month later the cord was no longer visible at rest however a small amount of cording was still visible when the patient flexed his biceps muscle in an abducted position (Figure 3).

Discussion

The diagnosis of AWS is primarily made based on reported symptoms of pain and restricted motion, visual inspection and palpation of the axilla, upper extremity, and trunk for signs of cording.[3] A prospective study by Lacomba *et al*. reported an incidence rate of 48.3% in patients undergoing ALND however the range of incidences reported in the literature span from 0.6%-85.4%.[2] There are no reports of incidence rates available for males not undergoing breast cancer treatment. The natural history of this condition is reported to range from 3 weeks to 3 months,[5,6] although persistent symptoms have also been reported up to one year later.[7] Although it is not currently well understood, the most commonly cited pathophysiological process reported in the literature is damage to the lymphatic and venous systems during removal of lymph nodes for the treatment of breast cancer.[1,2,8] During the

Figure 2.
The tissue appeared to be adhered to the skin at the distal end and deformed the skin in a crinkling appearance when the arm was moved.

surgical process, increased lymphatic fluid coagulation may result in lymphatic thrombosis, which will lead to an accumulation in the lymphatic vessels, forming visible cords.[2] Lymphatic fluid has the capacity to spontaneously coagulate, although this process is increased in the presence of thrombokinase, a plasma protein. Thrombokinase is elevated post-surgery, but can also be present due to inflammation from tissue damage.[9] The present patient did not have breast cancer or surgery so the inciting mechanism is not clearly understood. One plausible explanation was a particularly intense bout of squash the day prior to the appearance of cording. The high force associated with overhead motions in his dominant racquet hand may have lead to damage of the venous and lymphatic vessels, as it has been suggested that external compression can lead to localized lymphatic thrombosis.[10] The patient is also a commercial pilot with a flight log of 54 hours in the month preceding the onset of cording at 35,000 feet with the cabin pressurised to 6000 feet. As it is known that high altitude travel can disrupt the venous system[11] It is possible that this may also have contributed to disruption of the lymphatic system, however this is purely speculative.

There are several risk factors reportedly associated with AWS including the type of surgery, BMI, age, ethnicity, axillary metastasis, oncology treatment, and healing complications.[2] Surgically, it appears that more aggressive surgical techniques such as ALND increase the incidence rate of AWS.[2,3,8] Studies have reported that a higher BMI is protective against AWS,[4,8] however it has been suggested that excess subcutaneous adipose tissue makes the visual diagnosis of AWS more difficult[4], therefore making it appear to be a protective effect. Since being older is often associated with a greater BMI, this may explain the association between AWS and younger age. Being a younger female also appears to increase your risk of AWS, however, this population is more likely to be undergoing breast cancer treatment.[3,4,8] Finally, one study reported increased prevalence of symptomatic AWS in African-American patients compared to caucasian patients.[4] Other than a low BMI and young age, the present patient did not demonstrate any of the associated risk factors for AWS, highlighting the rarity of his presentation.

The primary symptoms of AWS are pain, feelings of tightness in the axilla, and associated restricted range of motion, particularly in abduction, although not all patients with AWS will be symptomatic.[2] Our patient had mild pain at rest and moderate tenderness of the cord and due to his low percentage body fat, the visual inspection

Figure 3.
One month later, without further intervention beyond the first visit of soft-tissue mobilization and advice on stretching and self massage.

made for a clear diagnosis. His symptoms did not lead to any functional limitations or restrictions of activities of daily living.

In a systematic review by Yeung *et al*., the authors noted nine level IV studies for manual therapy techniques including myofascial release, scar massage, and manual lymphatic drainage.[2] Myofascial release treatments involving manual fixing of the cord while stretching the tissue resulted in increases in range of motion and decreases in pain.[12,13] It was also often reported that a painless audible "popping" sound would be heard during treatment, which is hypothesized to be the breakdown of the lymphatic cording.[13] Our patient underwent one treatment consisting of myofascial release and soft-tissue mobilization as well as weekly self-massage. He noted a 50% improvement in reduction of cording and perceived tightness after one treatment. After one month the cording was no longer visible at rest with only slight visualization during active biceps contraction. Due to the wide range of reported natural resolution periods in the literature[5–7], it is uncertain whether the treatment expedited recovery or if the condition spontaneously resolved. Low-level evidence has suggested between 5-10 treatments over 3-10 weeks[13,14], however, more high quality research is needed to make stronger manual therapy recommendations.

Summary

This case highlights the rare presentation of AWS in a competitive squash player with low risk factors for developing this condition. While it is plausible that frequent air travel combined with an intense bout of overhead exercise may have caused damage to the lymphatic vessels and subsequent cording, the exact mechanism cannot be determined. This case illustrates an unusual and potentially confusing patient presentation of a benign condition that was managed with manual therapy and self-massage. An awareness and understanding of this condition will provide the practitioner with confidence in providing effective conservative care.

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