Case Report

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Idiopathic axillary web syndrome: a case-based review of an unusual disorder

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ABSTRACT

Axillary web syndrome (AWS), which frequently develops following axillary lymph node dissection, can also be caused by several other conditions, such as infection or strenuous exercise. In recent years, idiopathic cases have also been reported. We report the case of a 27-year-old female who presented with pain, tension and a taut rope-like structure in the left armpit area. She also had a limited shoulder range of motion in abduction and forward flexion. We diagnosed the patient with idiopathic AWS based on a detailed clinical history and manifestations. The patient completely recovered without any sequelae by therapeutic interventions, including nonsteroidal anti-inflammatory drugs and physical therapy. Through a systematic review of the literature, four additional cases of idiopathic AWS were identified. This study aimed to investigate the similarities and differences between idiopathic and typical AWS cases.

Keywords: Axillary web syndrome, idiopathic, physical therapy

INTRODUCTION

Axillary web syndrome (AWS) is a skin disorder characterized by a visible taut cord within the axillary skin, usually detected by inspection and/or palpation. This cord often originates in the axilla and can extend to the elbow, forearm, and even the hand. In symptomatic cases, the most common complaints include pain, feeling of tension, and a limited range of motion (ROM) of the shoulder joint (1).

AWS is commonly known as a complication that occurs after axillary lymph node dissection (ALND), with an incidence rate of 6%–86% following ALND. AWS usually develops in the early postoperative period, but it is likely to occur several months or years later and even recur (2). The most common indication for ALND is breast cancer, followed by melanoma. In such patients, AWS, despite being a self-limiting disorder, induces additional morbidity (1). Apart from ALND, cases of AWS due to causes such as granulomatous inflammation after folliculitis or epidermal inclusion cysts have also been reported (3,4).

In recent years, four interesting cases of AWS that were apparently "idiopathic" and had no attributable cause have been reported around the world (5-8). In this report, we present the clinical characteristics and treatment of an idiopathic AWS case in light of the few idiopathic cases reported in the literature.

CASE

A 27-year-old, normal-weight female engineer was admitted to our clinic with complaints of pain and tension in the left armpit, which had started approximately three weeks earlier, and a taut rope-like structure in the left armpit area, which she had noticed one week earlier. The pain started in the left armpit, spread to the medial forearm, and increased with overhead activities. Her visual analogue scale (VAS) pain score was 7 (0=no pain; 10=worst pain) at the time of admission. She had no history of trauma, surgery, rash, or infection in the axillary region. Close examination revealed a non-erythematous subcutaneous cord-like structure originating in the left axillary region and extending from the medial to the middle portion of the left arm (**Figure 1**). There were no signs suggestive of infection in the axillary region, and there were no skin lesions or lymphadenopathy. The neurological examination was normal, and peripheral

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pulses were palpable, with equal volume in both upper extremities. Active and passive shoulder abduction and forward flexion were limited to 120°, while other movements were within normal limits. The last part of her left elbow extension was mildly painful (2/10 VAS scale). Laboratory parameters (complete blood count [CBC], biochemical parameters, tumor markers, serology, erythrocyte sedimentation rate [ESR], and C-reactive protein [CRP]) were within normal ranges. No biceps tendon pathology or muscle tear was detected on a shoulder ultrasound (US) examination. Left upper extremity Doppler US was negative for superficial or deep venous thrombosis.

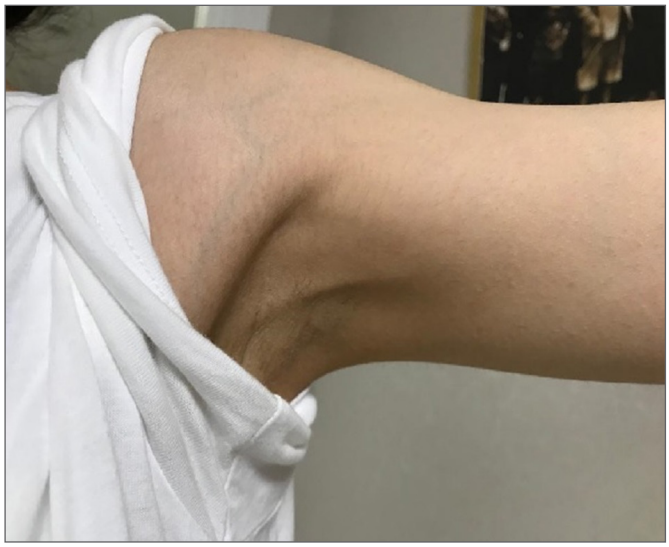

Figure 1. Idiopathic AWS, a band-like structure extending from the axilla to the antecubital fossa in the left arm.

Based on these signs and symptoms, the patient was diagnosed with idiopathic AWS and was initiated on oral meloxicam (7.5 mg/day) and topical nimesulide. She also received concurrent physical therapy five times per week for two weeks. Physical therapy interventions comprised superficial heat therapy, therapeutic ultrasound, transcutaneous electrical nerve stimulation (TENS) treatment, axillary myofascial release, and ROM and stretching exercises. The patient was then provided with an individualized, written, daily home exercise program, which was prescribed for the next 20 days.

The patient was seen for follow-up one month later. She reported that her shoulder pain had almost ceased (1/10 VAS scale), and the subcutaneous cord had completely disappeared. Additionally, her shoulder and elbow ROM were within normal limits without pain or discomfort. Informed consent was obtained from this patient.

DISCUSSION

In this case report, we describe the clinical features, diagnostic approach, and management of a patient with idiopathic AWS. To date, there have been only four reported cases of idiopathic AWS similar to that of our patient in the world (5-8) (**Table 1**). In the other four cases, the duration of symptoms ranged from two days to two months, and three of these cases presented with typical symptoms of AWS, such as a feeling of tension and pain in the axillary region, as in our patient (5,6,8). Interestingly, the remaining patient—a 67-year-old female AWS patient in France—had neuropathic-like upper extremity pain complaints, such as stinging, burning, and itching, and the patient's cord in the axillary region occurred later during follow-up (7).

Potential risk factors associated with the development of AWS after ALND include low body mass index, young age, more aggressive and extensive surgery, radiotherapy or chemotherapy, and hypertension (9). In the four cases reported in the literature, the ages of the patients ranged from 29 to 67 years, and no associated risk factors, including the factors mentioned above, were identified in the patient histories and physical examinations (5-8). Although the exact pathogenesis of AWS remains unclear, it has been reported that the processes occurring secondary to varying degrees of damage in veins following ALND, particularly in lymphatic vessels, and the processes occurring during the tissue healing phases, particularly during the proliferative phase, are effective in the formation of a visible taut cord (1,10). Moskovitz et al. (11) evaluated a large cohort of AWS patients and reported that a patient with stage IV invasive breast cancer and fixed axillar metastasis developed AWS due to the

Table 1. Summary of five cases of "idiopathic" AWS

Author	Age (yr) / Sex	Affected upper extremity	Symptom duration	Symptoms	Baseline pain severity	Management
Demir Y. et al., 2017	40 / M	Right	2 days	Shoulder pain, limitation of ROM in shoulder	VAS : 8	Oral analgesics, exercise, and physiotherapy
Tetik B. et al., 2019	41 / M	Right	2 weeks	Feeling of tension and pain in the axillary region	-	Oral-topical analgesics, exercise, and physiotherapy
Puentes Gutiérrez AB et al., 2020	67 / F	Right	2 months	Sense of itching, and stinging in the axillary region	LANSS : 14	Shoulder stretching exercises and massage
Dündar Ahi E. et al., 2022	29 / F	Left	3 days	Shoulder pain, limitation of ROM in shoulder	VAS : 5	Oral analgesics, exercise, physiotherapy, and massage
Cüce İ. et al., 2023	27 / F	Left	3 weeks	Feeling of tension and pain in the axillary region	VAS : 7	Oral-topical analgesics, exercise, and physiotherapy

ROM: Range of motion, VAS: Visual Analogue Scale, LANSS: The Leeds Assessment of Neuropathic Symptoms and Signs Scale.

disruption of normal lymphatic flow despite undergoing no surgery. In contrast, in the patient reported by Lee et al., the cord developed following the emergence of an epidermal inclusion cyst and consisted of band-like fibrotic tissue, with no accompanying lymph duct or sclerosing veins observed (4).

The diagnosis of AWS is easily made by patient-reported symptoms, visual inspection, and palpation of the cord in the axilla, upper extremity, or trunk (1). US can be used alongside physical examination for diagnosis, but its contribution is controversial in the context of visualizing the structure of the cord (12). Ahi et al. (8) reported in a case of idiopathic AWS that the cord was visualized as a hypoechoic band under the skin with US.

Management of AWS comprises physical therapy-based conservative treatment aimed at relieving pain and restoring full ROM, as well as education and medication. Additionally, it is known that AWS may resolve spontaneously without any treatment (1,9). Nevertheless, physical therapy may provide faster resolution of the cord than patients experience when they do not receive physical therapy (2). In a prospective study, 56 patients with AWS underwent physical therapy, and at the assessment 3 months after ALND, 54 had complete recovery, and 2 had residual signs of the disease (13). To our knowledge, there is not yet a definitive guide for the scope and duration of physical therapy in AWS. Physical therapy management of AWS often includes manual therapy, therapeutic exercises for restoration of ROM restrictions in the affected shoulder joint, and physical modalities, such as heat or cold therapy. According to our understanding of the literature, all of these interventions can be delivered safely, especially in idiopathic AWS cases.

CONCLUSION

It should be recognized that AWS may rarely develop idiopathically, regardless of undergoing ALND. Accordingly, an extensive awareness of the development of idiopathic AWS is essential since it can be easily diagnosed by clinical evaluation alone. However, additional diagnostic tools may be required for differential diagnosis. The clinical signs, symptoms, and management of idiopathic cases are similar to those of typical AWS cases that develop after ALND.

ETHICAL DECLARATIONS

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying image.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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REFERENCES

1. Yeung WM, McPhail SM, Kuys SS. A systematic review of axillary web syndrome (AWS). J Cancer Surviv 2015; 9: 576-98.
2. Koehler LA, Haddad TC, Hunter DW, et al. Axillary web syndrome following breast cancer surgery: symptoms, complications, and management strategies. Breast Cancer (Dove Med Press) 2019; 11: 13-9.
3. Zhang Q, Tan C. Axillary web syndrome following granulomatous inflammation after folliculitis. Eur J Dermatol 2016; 26: 314-5.
4. Lee KC, Chang YW, Chen CP. Axillary web syndrome following epidermal inclusion cyst: a case report and literature review. Clin Exp Dermatol 2019; 44: 64-6.
5. Demir Y, Güzelküçük Ü, Kesikburun S, et al. A rare cause of shoulder pain: axillary web syndrome. Turk J Phys Med Rehabil 2017; 63: 178-80.
6. Tetik B, Songür K, Aşkın A. Aksiller web sendromu: omuz ağrısının gözden kaçan nedeni. Cukurova Medical Journal 2019; 44: 255-9.
7. Puentes Gutiérrez AB, García Bascones M, Puentes Gutiérrez R, et al. [Idiopathic axillary web syndrome]. Rehabilitacion (Madr) 2020; 54: 68-72.
8. Dündar Ahi E, Ozen S, Saraçgil Coşar SN. Idiopathic Axillary Web Syndrome: A Case Report on a Rare Entity. J PMR Sci 2022; 25: 119-22.
9. Dinas K, Kalder M, Zepiridis L, et al. Axillary web syndrome: Incidence, pathogenesis, and management. Curr Probl Cancer 2019; 43: 100470.
10. Koehler LA, Hunter DW. Lymphspiration: The Axillary Web and Its Lymphatic Origin. Lymphology 2016; 49: 185-91.
11. Moskovitz AH, Anderson BO, Yeung RS, et al. Axillary web syndrome after axillary dissection. Am J Surg 2001; 181: 434-9.
12. Mullen LA, Harvey SC. Review of axillary web syndrome: What the radiologist should know. Eur J Radiol 2019; 113: 66-73.
13. Torres Lacomba M, Mayoral Del Moral O, Coperias Zazo JL, et al. Axillary web syndrome after axillary dissection in breast cancer: a prospective study. Breast Cancer Res Treat 2009; 117: 625-30.