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Case Report

Idiopathic axillary web syndrome as a rare cause of shoulder pain: A case report

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ABSTRACT

Idiopathic axillary web syndrome (IAWS) is a rare clinical condition which presents as a fibrotic cord leading to pain and limitation of movement. A 39-year-old female patient with shoulder pain, axillary swelling and mild limitation of motion was diagnosed with IAWS. She participated in physical therapy and was started acemetacin 60 mg b.i.d. After two months of follow-up, the patient's symptoms completely resolved. In conclusion, this case emphasizes the importance of early diagnosis and treatment of IAWS and contributes to the limited knowledge in the literature.

Keywords: Axillary web syndrome, idiopathic, physical therapy, shoulder pain.

Axillary web syndrome (AWS) often occurs after surgical excision of axillary lymph nodes due to breast cancer surgery. It is a clinical condition that causes pain in the form of cord-shaped bands extending from the axillary region to the arm, limitation of movement and functional limitation and may negatively affect the quality of life.[1] Although the pathophysiology has not been clearly established, it is thought to be related to the formation of inflammation, thrombosis and fibrosis involving lymphatic and venous structures as a result of surgical trauma. However, the number of idiopathic AWS (IAWS) cases in the literature is limited.[2-6] Although imaging modalities can be used to support the diagnosis, the diagnosis is mainly based on clinical evaluation and physical examination. However, it is not always possible to directly visualize cord structures with imaging modalities such as ultrasonography and magnetic resonance.[7] The main goal of treatment is to reduce pain, increase range of motion, decrease functional limitation and improve quality of life.[8]

In this case report, we present a rare case of IAWS without any history of surgical intervention, trauma, or radiotherapy, and discuss the clinical features, diagnostic process, and treatment approach in light of the existing literature.

CASE REPORT

A 39-year-old female physiotherapist presented to our clinic with complaints of left shoulder pain and axillary swelling for three days. There was no history of trauma to the shoulder or upper extremity. The patient did not report any neck pain or paresthesia related to her left shoulder symptoms. Systemic laboratory tests were performed and were within normal limits. Physical examination revealed a cord-like and firm fibrous structure extending from the left axilla to the medial aspect of the arm (Figure 1). There was no tenderness to palpation. The active range of motion of the left shoulder was flexion to 170 degrees and abduction to 170 degrees. The patient reported the Visual Analog Scale (VAS) pain intensity of 5 (0 = no pain;

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Figure 1. The axillary web in the left axilla.

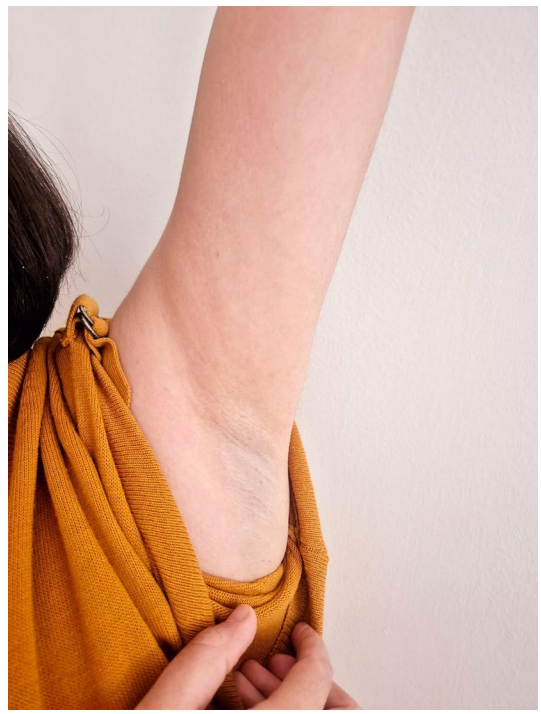

Figure 2. Follow-up at two months.

10 = very severe pain). Doppler ultrasonography did not detect blood flow within the cords, and no pathology suggestive of thrombosis was observed. Routine blood tests, including sedimentation rate (ESR), C-reactive protein (CRP), and complete blood count, were within normal limits. Additionally, chest X-ray, mammography, and breast ultrasound showed no pathological findings. The patient was diagnosed with IAWS and was prescribed acemetacin 60 mg b.i.d. as a non-steroidal anti-inflammatory drug treatment. A 15-session physical therapy program was planned, consisting of therapeutic ultrasound, transcutaneous electrical nerve stimulation (TENS), shoulder range of motion exercises, stretching and strengthening exercises, and massage. Following the treatment, her symptoms dramatically improved, the patient's shoulder range of motion was entirely restored, and VAS score was 1. At two months of follow-up, the fibrotic cord had completely resolved (Figure 2). A written informed consent was obtained from the patient.

DISCUSSION

Axillary web syndrome most commonly develops after lymphadenectomy or lymph node biopsy during breast surgery.[1] Its pathophysiology is usually thought to be related to processes of inflammation and fibrosis resulting from damage to lymphatic and venous structures caused by traumatic or surgical interventions.[8] Surgical trauma during axillary lymph node dissection has been suggested to cause lymphatic injury, particularly by affecting the lymphatic drainage pathways extending from the axilla to the medial aspect of the arm. Histopathological examinations in such patients have revealed dilated lymphatic structures containing fibrin clots, as well as thrombosed superficial veins. Additionally, lymphatic and venous stasis developing after surgery may contribute to localized hypercoagulability and fibrin deposition, thereby facilitating the formation of fibrotic bands.[1] However, IAWS is an extremely rare clinical condition which occurs without a history of trauma, surgery, or radiotherapy.[2-6] Moskovitz et al.[1] reported the incidence of AWS as 6% in patients after axillary lymph node dissection. Leidenius et al.[9] reported this rate as 72% after axillary dissection and 20% after sentinel lymph node biopsy. The exact incidence of IAWS remains unknown; however, its sporadic nature and the limited number of reported cases suggest that its prevalence is significantly lower compared to postoperative AWS.[2-6]

Although imaging modalities can be used for evaluation, it is difficult to visualize the cord

structures, so the findings may not always be diagnostic, regardless of the imaging modality used. Therefore, the diagnosis is based on the patient's symptoms, clinical observation and examination.[7] Although ultrasonography can be used as an adjunct to physical examination in the diagnosis of AWS, its contribution to visualizing cord structures remains controversial.[10] In the study conducted by Leduc et al.,[7] AWS cords were evaluated using ultrasound in 15 patients and cords could be detected in 12 of these patients. As a result of the ultrasound findings of these 12 patients, they defined a total of four different axillary web findings as hypoechoic or anechoic linear appearance with hyperechoic walls in hypodermic location, hyperechoic linear appearance in hypodermic location, hypoechoic linear appearance at the dermo-hypodermic junction and hyperechoic thrombus in a hypoechoic or anechoic structure in hypodermic location. At the same time, magnetic resonance imaging (MRI) was used to visualize the cord structures by placing a catheter containing visible gel. At the end of the study, although ultrasound imaging was reported to be more successful than MRI, it was concluded that both imaging modalities were challenging in identifying cord structures. In the study conducted by Koehler et al.,[11] ultrasound findings of 17 patients who developed AWS within 12 weeks after breast surgery were evaluated by a blinded radiologist. Accordingly, no structure directly associated with AWS could be identified on ultrasonography. This was hypothesized to be related to factors such as microlymphatic stasis or fibrin/protein deposition in the interstitial space, which are not visualized by ultrasound. Consequently, no specific structure or abnormality characteristic of AWS could be visualized, and it was concluded that current high-frequency ultrasonographic techniques may be insufficient for the diagnosis of AWS. In our case, a reactive lymph node measuring 23x8 mm was observed in the axillary region. The skin and subcutaneous tissue appeared normal on imaging. No pathology was found in the subcutaneous tissue, and no hyperechoic or hypoechoic appearance corresponding to the chordae reported in previous studies was detected. Thus, we consider the role of imaging techniques in AWS to be more relevant for differential diagnosis rather than providing direct diagnostic confirmation.

Although there is no standardized treatment approach in the literature, conservative physical therapy methods have been reported to be beneficial. Despite being a self-limiting, non-progressive

TABLE 1
Summary of previously reported idiopathic axillary web syndrome cases: Clinical characteristics and treatment approaches

	Year	Age (year)	Chief complaint	Initial VAS/LANSS	Post-treatment VAS/LANSS	Treatment	Imaging method
Demir et al.,[2]	2017	40	Shoulder pain, limited ROM	VAS: 8	VAS: 2	Oral analgesics, exercise, physiotherapy	-
Puentes Gutiérrez et al.,[3]	2020	67	Itching and stinging in axillary region	LANSS: 14	LANSS: 5	Shoulder stretching exercises, massage	-
Dündar Ahi et al.,[4]	2022	29	Shoulder pain, limited ROM	VAS: 5	VAS: 1	Oral analgesics, exercise, physiotherapy, massage	Ultrasound (Hypoechoic cord-like structure)
Cüce et al.,[5]	2023	27	Tension and pain in axillary region	VAS: 7	VAS: 1	Oral-topical analgesics, exercise, physiotherapy	-
Marya et al.,[6]	2024	24	Shoulder pain, limited ROM	-	-	Soft tissue mobilization, lymphatic massage, stretching exercises	Ultrasound (No significant findings)

VAS: Visual Analog Scale; LANSS: Leeds Assessment of Neuropathic Symptoms and Signs.

condition that may resolve spontaneously, patients often require treatment for pain management and to improve mobility, as IAWS can significantly impact daily living activities.[1,4,5,12] According to a systematic review and meta-analysis evaluating the efficacy of physical therapy in AWS after breast cancer, therapeutic exercises were the most effective treatment in reducing pain, increasing range of motion and functional improvement. Manual therapy, scar massage and myofascial release may be beneficial, but the effect of these interventions was found to be more limited than that of exercises.[12] Demir et al.[2] applied 15 sessions of physical therapy including range of motion exercises, hot pack, shoulder girdle muscle strengthening exercises, stretching exercises for shoulder abduction and flexion, and myofascial release massage to the patient diagnosed with IAWS. At the end of the first month following treatment, the patient's initial VAS score of 8 decreased to 2. Shoulder range of motion, which was initially limited to 140° in abduction and 150° in flexion, had returned to full range. In addition, the previously visible axillary cord had completely resolved. Dündar Ahi et al.[4] performed a physical therapy program including 15 sessions of infrared therapy.

Furthermore, TENS, therapeutic ultrasound, range of motion and strengthening exercises, shoulder flexor and abductor stretching and myofascial release massage for a patient diagnosed with IAWS. In the post-treatment follow-up, pain decreased, full shoulder range of motion was achieved and the axillary cord was completely resolved.[4] Similar to the literature, we applied a total of 15 sessions including shoulder range of motion, stretching and strengthening exercises, TENS, therapeutic ultrasound, and massage. Although the fibrotic band was not completely resolved after treatment, a significant reduction in symptoms was achieved. Table 1 summarizes previously reported cases, including age, symptoms, treatment modalities, baseline and post-treatment pain scores and imaging methods.[2-6]

In conclusion, IAWS should be considered in the differential diagnosis of a patient with unexplained shoulder pain and limitation of motion without a history of trauma or surgery. Therefore, in patients presenting with shoulder and arm pain, axillary examination should be considered as part of the clinical and medical evaluation process.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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