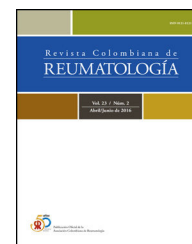




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Case Report

Tachon's syndrome: Case report and systematic review of the literature

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ABSTRACT

Corticosteroid injections are an interesting therapeutic alternative in rheumatology but are not without adverse effects. We aimed to resume the clinical, diagnostic, and therapeutic characterizations of a rare systemic effect of corticosteroid injections named Tachon's syndrome (TS). We reported a case of an 80-year-old woman who experienced acute thoracic pain and profuse sweating 5 min after a betamethasone shoulder injection with a normal physical examination. The most common presenting symptoms in the systematic review were chest and back pain as well as sweating. Symptoms mostly occurred suddenly within 1–5 min following corticosteroid injections. Although clinical features are sudden and stressful, the outcome is often excellent. The risk of occurrence is minimal. Diagnosis demands the exclusion of other medico-surgical emergencies. Patients should be reassured about the benignity of this condition despite its sudden distressing symptoms.

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Síndrome de Tachon: reporte de un caso y revisión sistemática de la literatura

RESUMEN

Las inyecciones intraarticulares con corticosteroides son una alternativa terapéutica interesante en reumatología, pero tienen algunos efectos adversos. Se buscó resumir las características clínicas, diagnósticas y terapéuticas de un raro efecto sistémico de dichas inyecciones denominado síndrome de Tachon (TS). Se presenta el caso de una mujer de 80 años con dolor torácico agudo y sudoración profusa a los 5 min de la inyección intraarticular con betametasona en el hombro, con un examen físico normal. Los síntomas más frecuentes en la revisión sistemática fueron dolor torácico y lumbar, así como sudoración. La mayoría de los síntomas aparecieron repentinamente entre uno y 5 min después de las inyecciones intraarticulares con corticosteroides. Aunque las características clínicas son

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repentinas y estresantes, el resultado suele ser excelente. El riesgo de aparición es mínimo. El diagnóstico impone la exclusión de otras urgencias médico-quirúrgicas. Los pacientes deben estar tranquilos acerca de la benignidad de esta condición, a pesar de sus síntomas de angustia repentina.

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Introduction

Intra-articular corticosteroid injections are one of the most widely used local therapy in rheumatology. This treatment has several general adverse effects that are generally well tolerated.¹ In rare cases, glucocorticoid infiltrations can cause excruciating lumbar, dorsal, and/or thoracic pain. This uncommon entity, known as Tachon's syndrome (TS), is a tribute to the first rheumatologist who reported this syndrome.² Few reports of this complication were found in the literature.²⁻⁴ We reported a case of a female patient who experienced similar symptoms after an intra-articular corticosteroid injection of the shoulder. We also systematically reviewed case reports and case series of TS in order to summarize the clinical and therapeutic characteristics of TS.

Case description

An 80-year-old female with no medical history and without known allergies had experienced mechanical pain in her right shoulder for 3 months. On examination, the range of motion of the right shoulder was limited in active mobilization but painfully recovered in passive mobilization. The Jobe and palm-up tests were positive. The remainder of her physical examination was normal.

Radiography of the right shoulder showed moderate degenerative changes in the glenohumeral joint without any calcification or erosion of the adjacent cortex. Ultrasound of the right shoulder joint revealed a rupture of the supraspinatus tendon associated with cleavage of the biceps. Laboratory evaluation included a normal blood count and normal liver and renal function tests. After local anti-inflammatory, analgesic, and functional physiotherapy treatments failed, an intra-articular injection of corticosteroid was indicated. After skin disinfection using alcoholic betadine, without local anesthesia, we carried out an injection of 5 mg/1 mL of betamethasone suspension (Diprostone®) in a pre-filled syringe in the right glenohumeral joint. There was neither a blood test during the pre-injection aspiration, nor resistance during the injection. Five minutes later, the patient experienced acute thoracic pain associated with chest oppression and profuse sweating. There was no lumbar or dorsal pain, nor skin lesions or pruritus. Blood pressure was 110/70 mmHg and heart rate was 97 beats per minute. The blood glucose level from the fingertip was 0.96 g/L. Heart and pulmonary auscultation were normal. An electrocardiograph showed no abnormalities. Chest radiography was unremarkable. Instantly, we elongated the patient with an elevation of

the lower limbs. The patient spontaneously improved within 15 min with a total resolution of thoracic pain. However, she remained in observation due to persistent fatigue for about an hour. A registration of the case was recorded in the national pharmacovigilance database under the number 407321 on August 14, 2021.

Methods

The systematic review was performed based on the Preferred Reporting Item for Systematic Review and Meta-analysis (PRISMA) statement recommendation.

Search strategy

We searched the PubMed and Science Direct databases for studies up to 2005 using the search items ("Tachon's syndrome") AND ("corticosteroid" OR "glucocorticoid" OR "corticoid") AND ("side effects" OR "adverse effects"). No restrictions were imposed. We also did manual research on reference lists of retrieved relevant articles.

Study selection

In this review, we included prospective or retrospective descriptive case reports and case series conducted on TS. We excluded papers investigating similar conditions to TS and papers not published in English or French.

Data extraction

The information extracted from the included articles is as follows: Name of the first author, publication year, number of cases, age, molecule injected, site of injection, occurrence delay, duration, and type of symptoms.

Ethical considerations

This study complies with the regulations on bioethical research, and it was approved by the Hospital local medical Ethic Committee.

Review of the literature

We reviewed the literature in order to identify cases of TS. We were unable to find extensive reports of this rare syndrome in the literature. Three articles were eligible. Up to 322 cases of TS were reported in the literature (Table 1). The

Table 1 – Studies assessing Tachon's syndrome.

Authors/year	N	Gender	Mean age (years)	Molecule	Site of injection	Occurrence delay	Symptoms duration	Symptoms
Berthelot et al. ² 2005	318	-	-	Cortivazol (67%) Hydrocortisone (25%) Betamethasone (7%) Paramethasone acetate (1%)	Lumbar epidural (39%) Upper limb (30%) Lower limb (24%) Other locations (7%)	1–5 min (78%) <1 min (22%)	5–15 min (51%) 1–5 min (31%) >15 min (15%) <1 min (3%)	Anxiety (87%) Pain in lumbar region (84%) Shortness of breath (64%) Erythema (64%) Profuse sweating (41%) Agitation (29%) Pain in dorsal region (25%) Transient cough (23%) Abdominal pain (20%) Transient hypertension (15%) Paleness (10%) Hypotension (8%) Diarrhea (3%) Headache (3%)
Hajjioui et al. ³ 2007	2	Male	47	Prednisolone acetate	Epicondyl	2 min	20 min	Back pain Chest oppression Erythematous rash of the face
		Male	57	Prednisolone acetate	Lumbar epidural	Instantly	1 h	Back and chest pain Profuse sweating Erythematous rash of the face
Rekik et al. ⁴ 2017	2	Female	64	Cortivazol	Shoulder	2–3 min	20 min	Low back pain Chest tightness Erythematous rash of the face Profuse sweating
		Female	43	Cortivazol	Lumbar epidural	2–3 min	15 min	Sweating Chest and lumbar pain Facial erythema Headache

most common presenting symptoms were chest and back pain as well as sweating. Symptoms mostly occurred suddenly within 1–5 min following corticosteroid injections. Symptoms lasted often less than 15 min. Cortivazol was the most provider molecule of TS. Mostly, TS followed injections into the lumbar epidural space. No recurrences were recorded in the studies included.

Discussion

Intra-articular corticosteroids can be considered as an additional necessity in the course of treatment of painful and inflamed joints, tendons, and/or ligaments. However, these local treatments may be associated with risks and complications. These risks include sepsis, skin atrophy, tendon rupture, and many other adverse effects.⁵ In rare cases, corticosteroid injections may cause unexpected reactions. One of these uncommon complications is TS. The observation presented here can be integrated with this syndrome. There was no other etiology explaining the condition. Symptoms, which consisted

of excruciating chest pain and oppression, occurred within 5 min of corticosteroid injection with a spontaneous resolution. According to the French method,⁶ the partial imputability scores were: C3 for the chronological imputation (score C) and S3 for the semiological imputation (score S). The global imputation (score I) was I4.

TS was first described by the French rheumatologist “Gérard Tachon” who reported his experience with several cases.² TS' occurrence has been estimated to 1 event per 8000 injections or 6.5 years of practice.² The rarity of published reports of TS can be explained by the benign nature of this event, its frequent favorable outcome, and maybe its confusion with allergic reactions.²

Physiopathological mechanisms remain unknown. The allergic mechanism does not seem involved in TS. Indeed, there was no asthma in the medical history of all patients. The urticarial lesions were also rare among patients (2%) as well as recurrence of TS (13%).³ However, TS could be explained by the accidental introduction of the product in a vein after its breach occurring during the injection. Other frequent symptoms, such as sweating, pallor, hyper or hypotension, diarrhea,

may be related to the involvement of the sympathetic and parasympathetic systems.³ Furthermore, several authors suggested that TS may be considered as the venous part of Nicolau's syndrome.⁷ Both syndromes are uncommon and cause extreme anxiety to the patient and the physician. The difference between the two syndromes lies in the fact that the drug substance is injected directly within a blood vessel in Nicolau's syndrome, causing thus acute arterial thrombosis and livedoid dermatitis. However, in TS, the suspension enters the bloodstream through breaching of a large vein.² This explains the delay of symptoms in TS as well as the more frequent occurrence after injections in areas rich in venous plexus.²

The clinical features of TS may be similar to those observed in transfusion incompatibility reactions.⁸ The difference between the conditions remains in the delay of the occurrence of symptoms; TS occurs within 1–5 min following the injection whereas transfusion incompatibility reaction is slower to happen.² Several clinical features are likely to occur in TS. However, previous reports showed that lumbar pain was the most frequent symptom (84%).² Berthelot et al.² reported that the most frequent site associated with TS was the lumbar epidural injection. However, this side effect was not reported by recent studies.^{9,10} Although all molecules of corticosteroids available for intra-articular injections may cause a TS, it has been reported that cortivazol was the most frequent molecule associated with TS.² The diagnosis of TS remains difficult since symptoms are not specific. It is essential to exclude other conditions that may present with the same symptoms, especially medico-surgical emergencies.⁴ TS is usually accompanied by a spectacular procession of symptoms that may alarm the physician and distress the patient. However, it often resolves rapidly and spontaneously. Although none of the 318 French cases of TS experienced any recurrence,² TS may recur in subsequent injections in 14–60% of cases.⁴

Conclusion

Although TS is a rare and benign condition, doctors performing corticosteroid injections should be aware of this side effect which imposes the exclusion of other emergencies. Further studies are needed to fully understand the pathogenesis of this condition.

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Conflicts of interest

None.

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