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CASE STUDY

Diagnostic difficulties in a paediatric patient with shoulder pain: Parsonage-Turner syndrome – Case Report

Bartłomiej Syzdoł[1,A-D], Wiktoria Sielwanowska[1,C-D], Anna Maria Rzewuska[1,C-D], Monika Żybowska[1,C-D], Magdalena Chrościńska-Krawczyk[2,A-B,E-F]

[1] *Student Scientific Society at the Department of Paediatric Neurology, Medical University, Lublin, Poland*
[2] *Department of Paediatric Neurology, Medical University, Gębali 6, 20-093 Lublin, Poland*
A – Research concept and design, B – Collection and/or assembly of data, C – Data analysis and interpretation, D – Writing the article, E – Critical revision of the article, F – Final approval of the article

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Abstract

Parsonage-Turner syndrome (PTS) is a rare neurological disorder characterized by severe unilateral pain in the shoulder. Muscle weakness and wasting can be also observed. The etiology is unknown. The study presents the case of a 15-year-old male with increasing paresis of the right upper limb. The patient reported pain and dizziness. Romberg's test and finger-to-nose test were positive. There was an abduction disorder of the right glenohumeral joint. MRI showed the cerebellar vermis with a slightly increased intensity. After starting the treatment, the cerebellar symptoms disappeared quickly, but the problems with the shoulder persisted. An ultrasound of the right brachial plexus showed oedema lesions. EMG revealed signs of damage to the right brachial plexus. Rehabilitation was recommended. After 10 months, the boy's clinical condition improved. PTS is a disease that presents great diagnostic difficulties. Doctors must take this syndrome in the differential diagnosis of shoulder pain.

Key words

shoulder pain, brachial plexus, Parsonage-Turner Syndrome

INTRODUCTION

Parsonage–Turner Syndrome (PTS) is a rare autoimmune disease of the peripheral nervous system that presents with pain in the shoulder or arm, usually unilateral.

The annual incidence ranges from 1.6 – 3 per 100,000 people, but the actual incidence may be 10 times higher due to under diagnosis. Most cases occur between the third and seventh decades of life [1], and the disease affects males more than females (2:1) [2].

The etiology of Parsonage-Turner Syndrome remains unknown. Several antecedent events have been reported. Possible triggers include: infections (44%): Epstein-Barr Virus, parvovirus B19, cytomegalovirus, human immunodeficiency virus, Coxsackie B, Leptospira, Mycobacterium tuberculosis, Yersinia, Salmonella; exercise (17%); surgery (14%), peripartum (9%); vaccination (4%): tetanus, hepatitis B, COVID-19; stress (4%); trauma (4%); among others (4%) [1, 3, 4, 5].

The characteristic clinical symptom is acute, severe neurogenic pain in the shoulder or arm (90–95%), most often unilateral. There is muscle weakness and atrophy. The pain may last for days or weeks and frequently can awaken patients from sleep [1, 3, 6, 7].

The muscles supplied by the upper brachial plexus are most often affected. The muscles commonly affected include the serratus anterior, deltoid, supraspinatus, infraspinatus, biceps and triceps. The most common nerves affected in PTS are the long thoracic, suprascapular, axillary, musculocutaneous, median and radial [1, 7].

The diagnosis of PTS remains predominantly clinical and is difficult to make in the early stages. Standard tests, such as blood tests, Computed tomography (CT) and magnetic resonance imaging (MRI) are used to exclude other diseases. Diagnosis of PTS may be supported by electromyography (EMG) to localizing and determining the extent of the injury. Radiographs and MRI of the shoulder may identify other causes of shoulder pain. No specific treatments have been proven to reduce neurologic impairment or improve the prognosis of PTS. However, studies conducted in recent years confirm the effectiveness of using steroids in the acute phase of the disease. Surgery may be required if muscle weakness continues. [1, 3, 4, 6, 8].

CASE REPORT

A boy aged 15 years and 11 months was admitted to the Department of Paediatric Neurology due to increasing paresis of the right upper limb and pain mainly at night. At home, the boy had a fever (39.5 °C) and had vomited. On admission, the patient was in good general condition, reported headache, dizziness and tingling in the area of the right shoulder. Physical examination showed a positive Romberg test and a positive finger-to-nose test, abduction disorders of the right glenohumeral joint, muscle weakness in the proximal part of the right upper limb. The patient denied any injuries.

CT and CT angiography examinations of the head showed no changes. MRI of the head revealed inflammatory changes within the cerebellar vermis. (Fig. 1)

Address for correspondence: Bartłomiej Syzdoł, Student Scientific Society at the Department of Paediatric Neurology, Medical University, Gębali 6, 20-093 Lublin, Poland
E-mail: bartlomiej.syzdol@gmail.com

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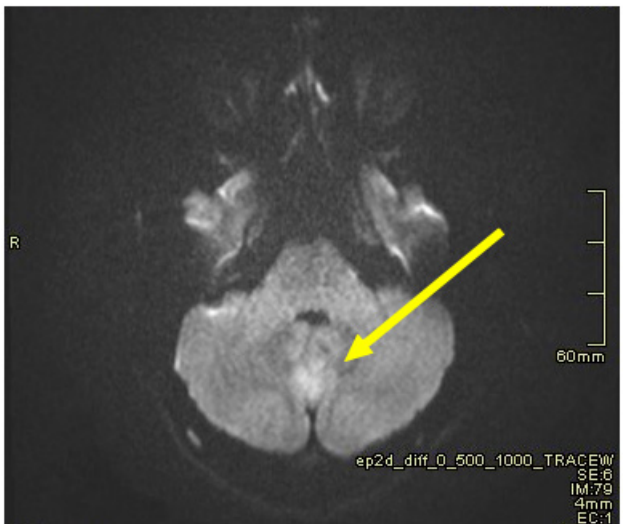


Figure 1. Slight increase in signal intensity visible within the cerebellar vermis

No changes were detected in the radiographs of the right humerus, left and right brachial plexuses.

Due to the very different etiology of inflammation, a wide panel of tests were performed. In laboratory tests, there was a slight increase in ESR (Erythrocyte Sedimentation Rate) – 19 mm / h (reference range: 2 – 15 mm / h), neutrophils – 8330/µl (reference range: 2,500 – 7,000/µl) and D-Dimer – 789 ng / ml (reference range: <500 ng / ml). Tests for Cytomegalovirus (CMV), Epstein-Barr virus (EBV), herpes simplex virus (HSV), toxocarosis, toxoplasmosis and Lyme disease were performed. Increased concentration of IgG-EBV – 56.34 S / CO (sample/cut off, positive result >= 1.00 S / CO) was detected, without the presence of IgM antibodies. Panel of active substances, including testing for the presence of synthetic marijuana, 3,4-methylenedioxypyrovalerone (MDPV), cathinone, oxycodone, lysergic acid diethylamide, mephedrone, amphetamine, benzodiazepines, cocaine, morphine was negative.

Based on the clinical presentation and MRI changes, Ceftriaxone, Acix (acyclovir) and Diflucan (fluconazole) were included in the treatment. After starting the treatment, cerebellar symptoms disappeared quickly while the soreness of the right shoulder, abduction problems in the glenohumeral joint and muscle weakness in the proximal part of the right upper limb continued.

On the 3rd day of hospitalization, the patient was consulted by a rehabilitation doctor. Neurological examination showed no disturbances of deep sensation, balance disorders and gait disturbances. Flaccid paralysis was present in the proximal right upper limb (muscle strength in the hand and forearm was slightly weakened in relation to the left upper limb). Restricted active movements during flexion, extension and abduction in the glenohumeral joint (Lovett Scale 3-). Tendon reflexes in the lower limbs (knee, ankle) – symmetrical. Tendon reflexes of the upper limbs: symmetrical on both limbs from the brachioradial muscle, slightly weakened in the right upper limb from the triceps muscle. Physiotherapy was administered to strengthen the shoulder girdle and right upper limb. Electrostimulation and kinesiotaping of the right upper limb were recommended.

After two weeks, a control MRI of the head, EMG and US of right brachial plexus were performed. A regression of changes described in previous MRI was found.

Right upper extremity nerve conduction examination revealed:
- damage to the axillary, musculo-cutaneous, suprascapular and long thoracic nerves in the form of prolonged terminal latency, decreased response amplitude;
- correct conduction in the median, ulnar and accessory nerves.

The examination showed signs of damage to the posterior and lateral cords of the right brachial plexus.

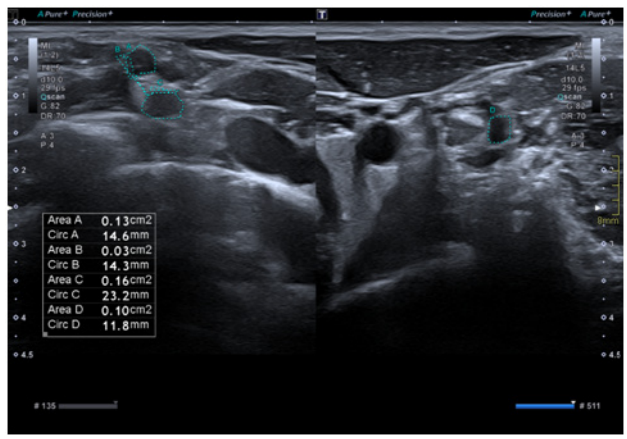
Ultrasound (US) examination of the right brachial plexus was performed and showed a slightly reduced echogenicity of the C5-C8 roots and a slight decrease in the abdominal mass of the deltoid muscle at the attachment to the acromion. Due to the suspicion of inflammatory changes and based on consultations with a radiologist, a viral etiology was suspected. Aciclovir was included in the treatment and further physiotherapy recommended.

A panel of onco- and anti-neuronal antibodies was made and showed no significant abnormalities.

Three weeks after the previous examinations a control EMG was performedwhich showed little progression compared to the previous examination.

US of the brachial plexus was also performed (Fig. 2; Fig. 3) and showed thickened C5 and C6 roots of the plexus, with reduced echogenicity and obliteration of the structure – the cross-sectional area of the roots in the range of 0.13 – 0.16 cm sq – progression of changes compared to the previous examination.

Oedema lesions of the suprascapular nerve leading from the C5 root – a nerve with reduced echogenicity, thickened to



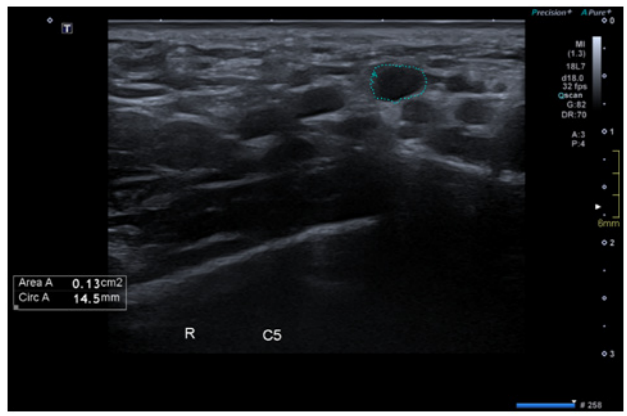


Figure 2 and Figure 3. US of the brachial plexus almost two months after the onset of symptoms

0.03 cm sq. Oedema changes with thickening and decreased echogenicity, visible in the proximal part of the upper trunk after joining the C5 and C6 roots; cross-sectional area of the upper trunk in the area of the altered part 0.13 cm sq.

Middle and lower trunks and cords of the right brachial plexus with correct echostructure.

The muscles of the shoulder girdle with a slightly increased echostructure, with features of atrophic changes, a decrease in the weight of the deltoid muscle at the attachment to the acromion (thickness 2 mm vs 4 on the left side). Similar changes occurred in the supraspinatus, infraspinatus and teres minor muscles.

The changes in the brachial plexus showed the PTS with the involvement of the C5, C6 roots and the upper cord of right brachial plexus. The diagnosis was made almost two months after the onset of symptoms. Five pulses of 750mg Solu-Medrol were made Clinical improvement was not achieved. Further physiotherapy was recommended.

After two months, a control MRI of the head and US of the brachial plexus were performed. There was a normalization of signals in all sequences of the cerebellar vermis, and visible regression of changes described in previous MRI and US examinations. Abduction and elevation of the right upper limb were present.

Ten months after the onset of the first symptoms, the patient was admitted to the Department for a control MRI of the head and US of the brachial plexus. On physical examination, the muscles of the right upper limb were slimmer, abduction and elevation of the right upper limb and normal muscle strength were present. There was a significant improvement in the clinical condition.

In the performed MRI of the head (Fig. 4) and US of the brachial plexus – regression of the changes were seen.

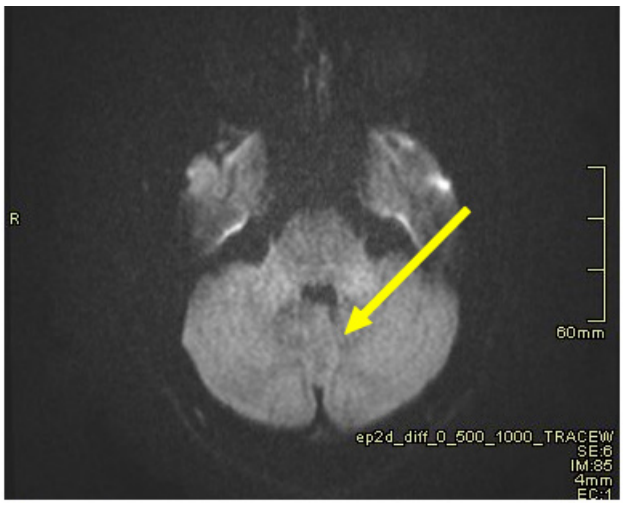


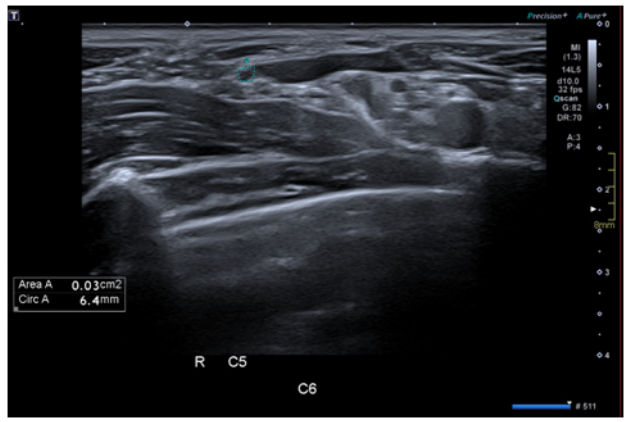
Figure 4. Control MRI of the head 10 months after the onset of symptoms

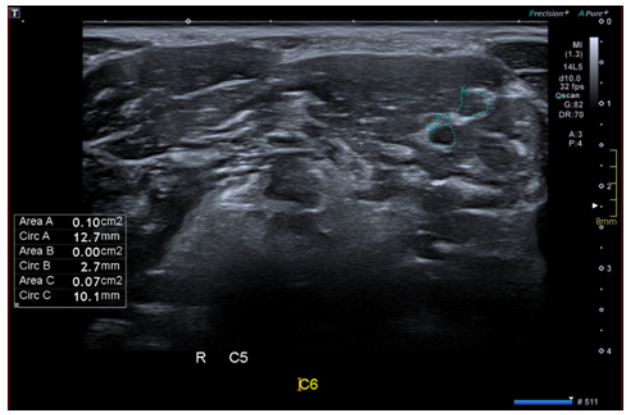
US of the right brachial plexus (Fig. 5, Fig. 6) showed C5 and C6 roots of the plexus slightly thickened, with reduced echogenicity and obliterated structure – root cross-sectional area in the range C5: 0.1 cm sq., C6: 0.07 cm sq – regression of changes compared to the previous examination. Slight reduction in the echogenicity of the suprascapular nerve leading from the C5 root – a nerve with a thickness of 0.03 cm sq – as in the previous examination.

Upper, middle and lower trunks, as well as the cords of the right shoulder plexus with correct echostructure, not thickened, with preserved continuity. Subclavian part of the plexus with correct echostructure.

Muscles of the shoulder girdle: supraspinatus, infraspinatus and deltoid with features of moderate atrophic changes – with a reduction in the weight of the abdomens (by nearly half the thickness of the abdomens) and a moderate increase in their echogenicity – denervative changes.

Figure 5 and Figure 6. Control US of the right brachial plexus 10 months after the onset of symptoms





CONCLUSIONS

Based on the clinical picture and additional examinations, the boy was diagnosed with Parsonage-Turner syndrome. In the described case, quickly implemented pharmacological treatment contributed to the resolution of cerebellar symptoms and changes visible in the MRI, but the shoulder pain remained. Intensive rehabilitation procedure has contributed to patients functional improvement just four months after the onset of the first symptoms. The steroid therapy used did not bring the expected results. Ten months after the initial symptoms there was a significant improvement in the patient's clinical condition.

The etiology of PTS remains unknown. In the literature there are reports of a disease, vaccination or trauma which had occurrred in the recent past in a patient who developed PTS [4]. Inflammatory changes seen in the MRI of cerebellar vermis may have been such an event, but based on the tests performed, the etiological factor has not been established.

Physiotherapy is important in long-term recovery from this condition and recommended after the resolution of acute neurogenic pain [3].

PTS causes great diagnostic difficulties; therefore, doctors should take this syndrome into account during differential diagnosis of shoulder pain, weakness and sensory abnormalities. Treatment should be individualized to each patient. Multiple case studies and reports suggest that corticosteroids can be effective [6]. Pharmacological treatment alone may not be sufficient, so it is worth using pharmacotherapy combined with physical therapies.

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