CASE REPORT

Epilepsy under my skin?

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SUMMARY

A 44-year-old male patient was admitted to the hospital for observation after an unwitnessed syncope. Physical examination revealed skin purpura and bilateral tongue haematoma. Laboratory studies were unremarkable. Radiological imaging showed no abnormalities of the vasculature, signs of thrombosis or brain anomalies. Biopsy of a purpuric lesion revealed extravasation of erythrocytes. After excluding several causes of both syncope and purpura, the typical location of these thoracocervicofacial purpura, the tongue haematoma and an elevated prolactin level (which came back later) led to the diagnosis of an epileptic seizure. The patient was referred to the neurology department for follow-up. Within 3 weeks, the purpura were completely resolved, and the patient remained free of seizures during follow-up. In case of an unwitnessed syncope, an epileptic seizure should be carefully considered and thoracocervicofacial purpura can be the pivotal manifestation leading to this diagnosis.

BACKGROUND

Syncope is a frequently encountered problem at the emergency department and has a variety of causes.¹ Epileptic seizures are the cause of syncope in 5%–10% of all patients.^{1 2} It is important to recognise this diagnosis, since epilepsy is a serious condition that often requires precautions, treatment and close monitoring. In this case, we show that a careful and well-executed physical examination at presentation is essential in syncope, since some rare findings can lead to the correct diagnosis.

Thoracocervicofacial purpura can appear after an epileptic seizure due to extravasation of erythrocytes caused by an elevated intrathoracic pressure. They are a rare, but typical finding in epileptic seizures and may be the only objective sign at presentation. As far as we know, only six case reports described this finding.^{3–8} We report a case of an unwitnessed syncope in which thoracocervicofacial purpura turned out to be the pivotal sign of epileptic seizure, in addition to two other supportive findings: a bilateral tongue haematoma and an elevated serum prolactin level.

CASE PRESENTATION

A 44-year-old male patient, with a medical history of depression and obstructive sleep apnoea syndrome, was admitted to the hospital for observation after a syncope. He was found unwell on the couch after an unwitnessed event. Since he regained consciousness, he was experiencing dyspnoea and thoracic pain. The patient had no preceding complaints

before the syncope, as far as he remembered. He had not taken any medication or recreational drugs. Physical examination showed normal and stable vital signs, including respiratory rate, oxygen saturation and examination of the lungs/thorax. However, he had thoracocervicofacial purpura (figures 1 and 2), bilateral tongue haematoma (figure 3) and a small conjunctival haemorrhage. There were no signs of trauma or asphyxia. Neurological examination was normal, except for retrograde amnesia for the episode.

INVESTIGATIONS

Laboratory studies of complete blood and platelet count, inflammation parameters, coagulation profile, serum electrolytes and liver function tests revealed no abnormalities. ECG showed no signs of cardiac ischaemia or conduction abnormalities. CT and ultrasound of the cervical and thoracic structures showed normal cervical and thoracic vessels without signs of obstruction (including pulmonary embolism) or malformation. MRI of the brain showed no abnormalities either. Later on, serum prolactin turned out to be elevated (0.66 U/L; normal value: 0.10–0.50 U/L). Biopsy of one of the purpuric lesions revealed extravasation of erythrocytes and no signs of vasculitis.

DIFFERENTIAL DIAGNOSIS

Extensive investigations excluded several causes of both the syncope and the thoracocervicofacial purpura, but the simultaneous manifestation of these problems suggested a common cause. A tonic-clonic epileptic seizure can induce both unconsciousness and thoracocervicofacial purpura. Supportive of this diagnosis were the typical location of the purpura, the retrograde amnesia and the tongue haematoma, and an elevated serum prolactin level in a sample that was obtained at the emergency department and that was reported later on.^{5–8}

OUTCOME AND FOLLOW-UP

The patient was diagnosed with a tonic-clonic epileptic seizure. He was referred to the neurology department for further analysis, monitoring and follow-up. No specific treatment was initiated, but he was prohibited from driving. The patient quickly recovered, and no new symptoms or seizures occurred during hospital admission. Within 3 weeks, the purpura had completely resolved, and in the year after admission, the patient remained free of seizures without specific treatment.

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Figure 1 Thoracocervicofacial purpura. A sharp demarcation can be seen on the chest above a horizontal line between the axillae.

DISCUSSION

In our patient, the typical localisation of the purpura, the tongue haematoma and the elevated prolactin level led to the diagnosis of an epileptic seizure.

Purpura originate from extravasation of erythrocytes which in turn has several causes.⁹ In our patient, there was no coagulation problem, platelet dysfunction or signs of vasculitis. Increased intrathoracic pressure caused by extreme coughing, vomiting, trauma, asphyxia and thoracic compression can also lead to an extravasation of erythrocytes in the thoracocervicofacial area.^{10 11} As there was no evidence for any of these causes to explain the purpura in our patient in combination with other findings supportive of the diagnosis of an epileptic seizure, we concluded that the thoracocervicofacial purpura were due to intense contractions of the chest and abdominal muscles with a closed glottis during the tonic-clonic phase.^{3 4}

The diagnosis of epilepsy was supported by the bilateral tongue haematoma which has a specificity of 96%–100% and a sensitivity of 20%–33% for this diagnosis.^{12 13} Additionally, supportive was the elevated serum prolactin which has a specificity of 96% and a sensitivity of 62% for epileptic seizures in the context of a syncope.^{8 14 15} The epileptic seizure disrupts the regulation of the hypothalamus which results in the release of prolactin by the pituitary gland.⁸ Prolactin levels should be measured as soon as possible as they peak within 10–20 min, but they remain elevated during the first 2 hours.^{8 14–16} Of note,

Figure 2 Detailed image of the purpura.

Figure 3 Bilateral tongue haematoma.

most studies on the diagnostic value of prolactin have focused on samples that were taken within 10–20 min after the event,^{8 14–16} and it is advised to take the sample within that time span.¹⁴

In conclusion, in the context of a syncope, thoracocervicofacial purpura may be a pivotal sign of epilepsy. In addition, a tongue haematoma and an elevated serum prolactin contribute to the diagnosis of an epileptic seizure.

Learning points

- ▶ In case of an unwitnessed syncope, an epileptic seizure should carefully be considered and investigated, since epilepsy is the cause in 5%–10%.
- ▶ Thoracocervicofacial purpura are a rare, but typical finding in epileptic seizures and can be an important symptom that leads to the diagnosis of an epileptic seizure.
- ▶ Elevated serum prolactin levels can contribute to distinguishing psychogenic from epileptic seizures and are therefore useful to diagnose epilepsy in case of an unwitnessed syncope.

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