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A non-typical Epileptic Reaction in a Patient with Insulinoma of the Pancreas: A Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. Author ZV designed the study and wrote the first draft of the manuscript. Authors EV, KT and AT managed the analyses of the study. Authors ZV and EV managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Extreme hypoglycemia in cases with insulinoma may be clinically manifested with epileptic reactions, most frequently described in literature as generalized tonic-clonic seizures.
We present a case of a 30-year old man who was hospitalized at the Clinic of Neurology of the University hospital "St. George" in Plovdiv, Bulgaria, because of frequent seizures (2-3/ week), manifested with impaired awareness, aggressiveness, agitation, and confusion, mainly in the morning. The patient had been initially treated in a psychiatric clinic, afterwards he was diagnosed with epilepsy and was treated with valproate without success. During his stay in the Clinic of Neurology the medical staff observed a couple of focal seizures with impaired awareness, turning head aside, fixed eyes, followed by aggressiveness, agitation, and postictal confusion. Extremely low levels of blood sugar were measured in the morning – up to 1.3 mmol/l (referent limits 4.1-5.9 mmol/l). The polysomnography recording showed episodes of high voltage synchronous slow activity in all derivations at the time of the described above seizures and hypoglycemia, which were coped with 40% glucose solution. Insulinoma in contact with the tail of the pancreas was verified as

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a metabolically active lesion on PET/CT. The patient was directed to a surgical department for biopsy and treatment. Seizures stopped after surgical resection of insulinoma.
We consider the reported case is a diagnostic challenge because of the type of clinical manifestations and the absolute necessity of highly specialized imaging investigations for etiology precision.

Keywords: Insulinoma; epilepsy; seizures; polysomnography.

1. INTRODUCTION

Insulinoma is a very rare tumor with a reported incidence of 0.5-5/1000000 [1]. It is associated with overproduction of insulin due to neoplastic proliferation of pancreatic islet β cells. It can secrete insulin in short bursts and cause fluctuation of blood glucose level. Then the patients will have intermittent neuroglycopenic symptoms, such as conscious disorder, abnormal behavior, psychiatric symptoms or convulsions, the latter usually described as generalized tonic-clonic seizures [1-6].

We present a case of a 30-year old male patient with frequent episodes with the characteristics of complex partial seizures mainly in the morning. The patient had been initially treated in a psychiatric clinic, afterwards he was diagnosed with partial epilepsy. In the process of etiology clarification insulinoma in the pancreas tail was found.

1.1 Anamnesis

The patient is a 30-year old male who was hospitalized at the Clinic of Neurology of the University hospital "St. George" in Plovdiv, Bulgaria because of frequent (up to 2-3/week recently) seizures manifested with impaired awareness, aggressiveness, agitation, and confusion, mainly in the morning; postictal confusion for a couple of minutes. The seizures were followed by confusion for a couple of minutes. The duration of complaints was 2 years. The patient was first diagnosed with Behavioral disorder by a psychiatrist. Afterwards he was diagnosed with epilepsy by a neurologist and treated ineffectively with Depakine chrono 500 mg TID for several months. Brain MRI was performed and it visualized single, symmetrical, bilateral, punctiform lesions in the subcortical white matter of parietal lobes, which were not found to be a sign of pathology.

2. CLINICAL MANIFESTATIONS AND CLINICAL COURSE

1. Physical status was normal.
2. Neurological status – no focal neurological signs were found.
3. Clinical course during hospital stay: 3 episodes (2 in the morning) with the characteristics of complex partial seizures were observed by medical staff - impaired awareness, turning head aside, aggressiveness, agitation, and confusion; postictal confusion for a couple of minutes. Extremely low blood sugar levels were measured at the time of seizures – up to 1.3 mmol/l (referent limits 2.8-6.1 mmol/l.). The clinical manifestations were coped fast by 40% Glucose Solution.

3. INVESTIGATIONS

1. Laboratory investigations: full blood count, biochemistry – urea, creatinine and ionogram in referent limits; fasting blood sugar (6 a.m.) varied from 1.3 mmol/l to 1.5 mmol/l. Similar results for fasting blood sugar were obtained later in the morning as well (9 a.m.)
2. EEG (standard vigil recording) – normal.
3. Polysomnographic recording (Fig. 1) – preserved sleep organization with normal NREM sleep quantity and slightly increased REM sleep quantity. Microarousals are distributed normally. Sleep effectiveness is normal. REM sleep latency is slightly decreased. No respiratory problems during sleep. Following an arousal from the last sleep cycle at 5:40 a.m., the patient is in a continuous state of arousals and drowsiness to N2 sleep for 1 hour, with elements of both vigilance and sleep. This period is followed by episodes of high voltage synchronous slow-wave rhythm in all derivations. The patient is agitated and confused. After the application of 40%

Glucose Solution the patient calms down, EEG is normalized.

The patient was consulted with an endocrinologist and diagnostic clarification was recommended.

4. MRI of abdominal organs – no pathological findings.
5. Somatostatin-receptor scintigraphy + SPECT-CT of abdominal organs – a suspected area with moderate expression of somatostatin receptors in the pancreas tail (Fig. 2).
6. Whole-body 18 F-FDG 6.1 mCi 70-minute PET/CT – a metabolically active lesion with malignant characteristics, in contact with the pancreas tail (Fig. 3).

3.1 Treatment

The performed surgical treatment resulted in complete coping of seizures.

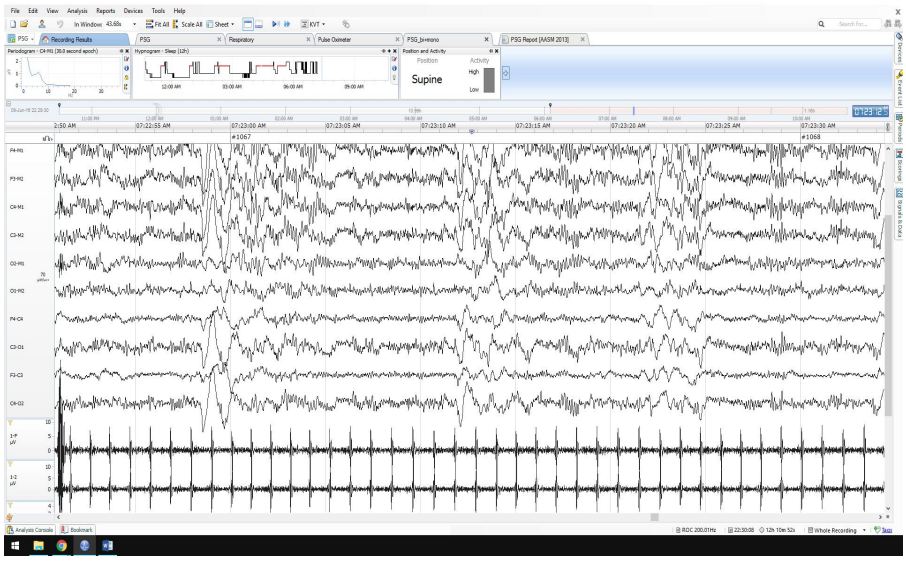

Fig. 1. Polysomnographic recording during morning hypoglycemia

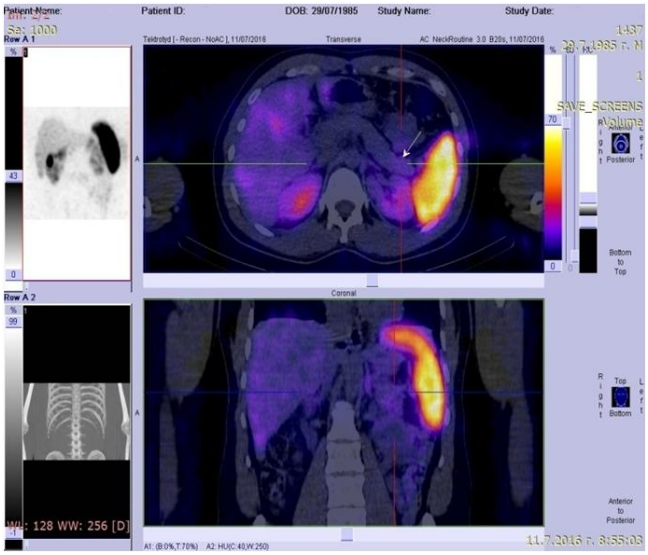

Fig. 2. Somatostatin-receptor scintigraphy + SPECT-CT of abdominal organs

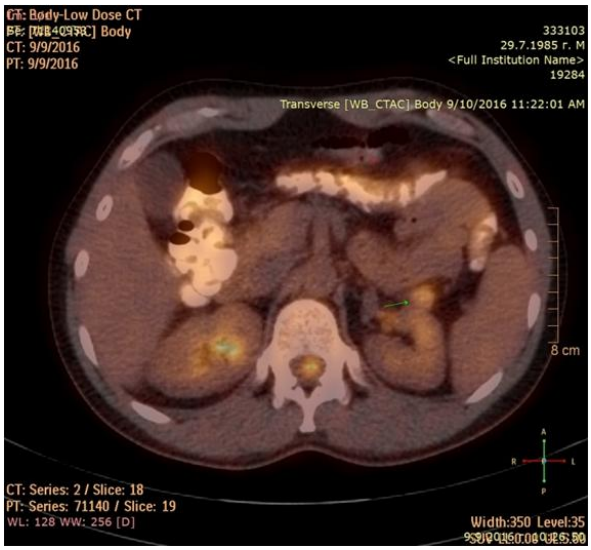

Fig. 3. PET/CT of abdominal organs

4. DISCUSSION

In most reported in literature cases with insulinoma and epileptic seizures treatment is initiated with anticonvulsants, which are not effective [1-11]. The tumor is usually diagnosed much later than the seizure onset [2,7-10], which is explained by insufficient diagnostic thinking about distinguishing the manifestations of hypoglycemia from those of epileptic seizures and psychiatric diseases [11].

In the reported case etiology clarification of epileptiform manifestations has taken more than 2 years and has gone through an initially suspected psychiatric disease and subsequent diagnosis of epilepsy. The similarity of clinical manifestations to complex partial seizures, the observed tendency for twenty-four hours rhythmicity, and the lack of pathological findings on EEG, have required a more extensive differential diagnosis for etiology precision. The results from laboratory, electrophysiological and highly specialized imaging investigations have been helpful for diagnosing insulinoma in the pancreas tail, that is the cause of fluctuations of blood sugar and clinical manifestations.

5. CONCLUSION

We consider the reported case is a diagnostic challenge because of the type of clinical manifestations and the absolute necessity of highly specialized imaging investigations.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

All authors hereby declare that the case study has been examined and approved by the appropriate ethics committee and has therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Mouaqit O, Boubouh A, Ifrine L, El Malki EHO, Mohsine R, Belkouchi A. Insulinoma misdiagnosed as epilepsy. Open Journal of Gastroenterology. 2012;2:28-30.
2. Rajadhyaksha A, Sonawale A, Chichkhede A, Zanwar A. Recurrent seizures due to pancreatic insulinoma. J Assoc Physicians India. 2016;64:86-7.
3. Kwon EB, Jeong HR, Shim YS, Lee HS, Hwang JS. Multiple endocrine neoplasia type 1 presenting as hypoglycemia due to

insulinoma. J Korean Med Sci. 2016; 31:1003-6.
4. Sharma P, Sharma S, Kalhan S, Singh BP, Sharma S. Insulinoma: A comprehensive summary of two cases. J Clin Diagn Res. 2014;8:FD05-6.
5. Kao K, Simm P, Brown J. Childhood insulinoma masquerading as seizure disorder. Journal of Paediatrics and Child Health. 2014;50:319-22.
6. Reddy MR, Ramakrishnan S, Kalra P, Saini J, Yadav R, Kulkarni GB, Kumar MV, Nagaraja D. Chronic progressive encephalopathy, intractable seizures, and neuropathy: A triad of neurological features in insulinoma. Neurol India. 2012; 60:238-9.
7. Graves TD, Gandhi S, Smith SJ, Sisodiya SM, Conway GS. Misdiagnosis of seizures: Insulinoma presenting as adult-onset seizure disorder. J Neurol Neurosurg Psychiatry. 2004;75:1091-2.
8. Wang S, Hu H, Wen S, Wang Z, Zhang B, Ding M. An insulinoma with clinical and electroencephalographic features resembling complex partial seizures. J Zhejiang Univ Sci B. 2008;9:496-9.
9. Krauß M, Berkermann H, Ghadimi M, Gaedcke J, Bürger T. Recurrent seizures of unknown aetiology. Dtsch Med Wochenschr. 2016;141:631-3.
10. Ma H, Zhang XP, Zhang Y, Lu HD, Wang JT, Zhang Y, Wu XB. Pancreatic insulinoma misdiagnosed as epilepsy for eight years: A case report and literature review. Intern Med. 2015;54:1519-22.
11. Suzuki K, Miyamoto M, Miyamoto T, Hirata K. Insulinoma with early-morning abnormal behavior. Intern Med. 2007; 46(7):405-8.

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