

# Diabetic neuropathic cachexia in a girl with type 1 diabetes mellitus

## Tip 1 Diabetes mellitus'lu kız çocukta gelişen nöropatik kaşeksi

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### Abstract

Diabetic neuropathy is a serious complication in diabetic patients, and its incidence increases by longer diabetes duration, increasing age, and poor glycemic control. Acute neuropathy hardly ever occurs in the early stages during the course of diabetes mellitus. Diabetic neuropathic cachexia (DNC) is a rare form of peripheral neuropathy characterized by profound weight loss, neuropathic pain, anorexia, depression with spontaneous resolution usually occurring within a year. The exact mechanism of DNC remains unclear. Herein, we present a case of a 17-year-old girl with type 1 diabetes mellitus for 4 years, in whom DNC developed following the rapid improvement in hemoglobin A1c after achieving a good metabolic control.

**Key words:** neuropathy, cachexia, type 1 diabetes mellitus

### Özet

Diyabetik nöropati yaş artışı, diyabet süresi ve glisemik kontrolün kötü olması ile insidansı artan, diyabetik hastaların ciddi bir komplikasyonudur. Akut nöropati diyabetin erken döneminde hemen hemen hiç görülmez. Diyabetik nöropatik kaşeksi (DNK) periferik nöropatinin nadir bir formu olup, genellikle bir yıl içinde kendiliğinden düzelen, belirgin kilo kaybı, nöropatik ağrı, depresyon ile karakterizedir. DNK'nin kesin mekanizması belirsizliğini korumaktadır. Burada, 4 yıldır Tip 1 diyabetes mellitus tanılı 17 yaşındaki bir kız çocuğunda, iyi metabolik kontrol sonrası hemoglobin A1c düzeyinin hızlı düzelmesi ile gelişen DNK olgusu sunulmuştur.

**Anahtar kelimeler:** nöropati, kaşeksi, tip 1 diyabetes mellitus

### Introduction

Patients with prolonged poor glycemic control have a higher frequency of and more severe complications than those with good control. Neuropathy is an important disability factor in diabetic patients and its incidence increases with the duration of diabetes associated with the poor glycemic control and the age of the patient . Diabetic neuropathic cachexia (DNC) is a rare syndrome first described in 1974 and seen predominantly in patients with type 2 diabetes mellitus (DM) on their sixth or seventh decade . It is characterized with substantial involuntary weight loss, anorexia, depression, severe autonomic dysfunction, and peripheral neuropathy that develop over a short period of time. The exact mechanism of acute painful peripheral neuropathy is unknown. DNC has been reported in a few cases with type 1 DM in the literature . By this way, we report a case of a 17-year-old girl with type 1 DM, who diagnosed four years before who developed an acute painful peripheral neuropathy in the setting of rapid improvement in

hemoglobin A1c after achieving a good metabolic control.

### Case report

The patient was a 17-year old female, had been diagnosed with type 1 DM four years ago. Her previous treatment consisted of intensive insulin therapy -in the form of three daily doses of insulin lispro and insulin glargine once a day- but her compliance with diabetic management was poor and last glycosylated hemoglobin level was 14%. In addition, she experienced dawn phenomenon and overweight. This time, serum total cholesterol, triglyceride, HDL-cholesterol, and LDL-cholesterol levels were 305 mg/dl, 657 mg/dl, 43 mg/dl and 131 mg/dl, respectively. At this time the age of menarche was 12 year old but she has been amenorrhea within last four years. On the examination, she weighed 70.5 kg (SDS +1.21) with a height of 159 cm (SDS -0.61). Her body mass index was 27.89 (SDS +1.41). Her total daily Insulin amount was approximately 0.75 unit/kg. She was re-educated for self-management of diabetes and

revised the insulin regimen (in the form of three daily insulin aspart and one dose insulin detemir) and adjusted. Her insulin doses were raised to 1.05 unit/kg/day. One month later HbA1c was decreased to 12.23%. Three months later after the first examination, she complained severe burning dysaesthesia, pain of the lower limbs, especially over her foot and leg, anorexia and nausea. She sometimes had been to feel palpitation. She was unable to walk and discontinued her education. She had a bad mood and sometimes cried due to her leg pain. She lost weight about 10 kg in three months. Her HbA1c level was decreased to 7.70%. Her serum total cholesterol, triglyceride, HDL-C and LDL-C levels changed to 152 mg/dl, 101 mg/dl, 46 mg/dl and 86 mg/dl, respectively. Her complaints had progressively risen. She also had generalized weakness, fatigue, nausea, decreased appetite and depressive mood. Figure 1 illustrates changes of her HbA1c levels and weight. Electrocardiography and other biochemical and hematological parameters were normal, but electromyography showed signs of mild sensory motor neuropathy. Gabapentin, tramadol, vitamin B1 and B6 were given for pain, but her symptoms resisted until her glucose levels were adjusted mildly higher. After that, her complaints gradually decreased within six months and menstrual bleeding was started. Figure 1 shows the changes of HbA1c levels and weight.

### Discussion

In patients with diabetes, studies have documented the association between the duration, degree of hyperglycemia and complications . The incidence of diabetic neuropathy increases with the poor glycemic control of the patient . Dysfunction and damage of the somatic and autonomic nervous systems leads to DNC which is reversible acute onset neuropathies and often occurs at the presentation of diabetes itself or develops following to the tight glycemic control. It is not related to the duration of diabetes or microvascular complication.

Ellenberg in 1974, in 1978 Chandler et al. and in 1983 Archer et al. described a similar acute syndrome of painful sensory neuropathy. DNC is seen predominantly in patients with type 2 DM . Acute neuropathy rarely occurs early in the course of diabetes mellitus. It has been reported more frequently in adult literature under the name of "insulin neuritis" . Our patient had been diagnosed as type 1 DM 4 years ago and she had poor metabolic control. While getting better metabolic control, insulin requirement was increased from 0.75 unit/kg/day to 1.05 unit/kg/day and in the meantime, the complaints from DNC began to emerge.

Our patient displayed a symmetrical peripheral neuropathy, weight loss and painful dysaesthesias. A case of DNC with edema which developed following the institution of tight glycemic control was reported, but our patient has no evidence of edema. In another case, symmetric lower extremity pain, parasthesias preventing daily function and acute bilateral cataract following the institution of insulin treatment were reported . Our patient did not develop any cataract or retinopathy. Llewelyn et al. described a single case of an 18-year-old girl having poorly controlled Type 1 DM for 8 years despite insulin therapy. She presented with a hemoglobin A1c of 18% and was hospitalized for continuous subcutaneous insulin infusion (9). Following the initiation of continuous subcutaneous insulin infusion, the patient's HbA1c fell to 8.5% within 6 weeks. During 6-week period, she developed severe burning in her feet, accompanied by tingling parasthesias and absent ankle reflexes. Nerve conduction studies, performed 6 weeks after beginning the continuous subcutaneous insulin infusion, showed a sensorimotor neuropathy. Sural nerve biopsy showed reduced myelinated fiber density with a loss of small myelinated axons and fiber degeneration and regeneration, but no sign of demyelination. Her pain resolved after 6 months. Archer et al. reported acute painful neuropathy in nine male patients (ages 13–61 years) with type 1 or 2 DM associated with severe weight loss, depression, and impotence (8).

DNC was present after the rapid metabolic improvement in our patient. Treatment for pain management was symptomatic, and was aiming to enable daily activities. It was significant in our patient that pain was released following the increase of target glycemic values. Therefore, we believe that intervention by changing the glycemic control is beneficial in such a patient, especially when symptoms have resisted treatment for a long time.

Possible etiologies for DNC include nerve regeneration, metabolic abnormality, hemodynamic change in the endoneurium surrounding the nerve, or immunologic reaction to insulin. When insulin therapy is initiated with rapid establishment of normoglycemia, epineural arteriovenous shunting occurs and leads to a steal effect rendering the endoneurium ischaemic . Kihara et al. have demonstrated that intravenous insulin impairs tissue oxygenation in nerves of diabetic rats when hyperglycemia is controlled (11). In our patient, rapid improvement of metabolic control or the increase of insulin amount used for metabolic improvement may play role in the physiopathology.

Depression is generally associated with this syndrome. Frequent crying episodes, insomnia, anxiety, and irritability are the characteristic features . These children can become depressed, so psychiatric support may be necessary to address these issues and help in promoting

recovery. Our patient had a bad mood and crying episodes for leg pain. We provided psychiatric support to the patient.

This case reminds us that diabetic neuropathic

cachexia in a young female patient is possible and significant diabetic complications can occur by a rapid fall in blood glucose concentrations, though the pathophysiological basis of this disorder remains unknown.

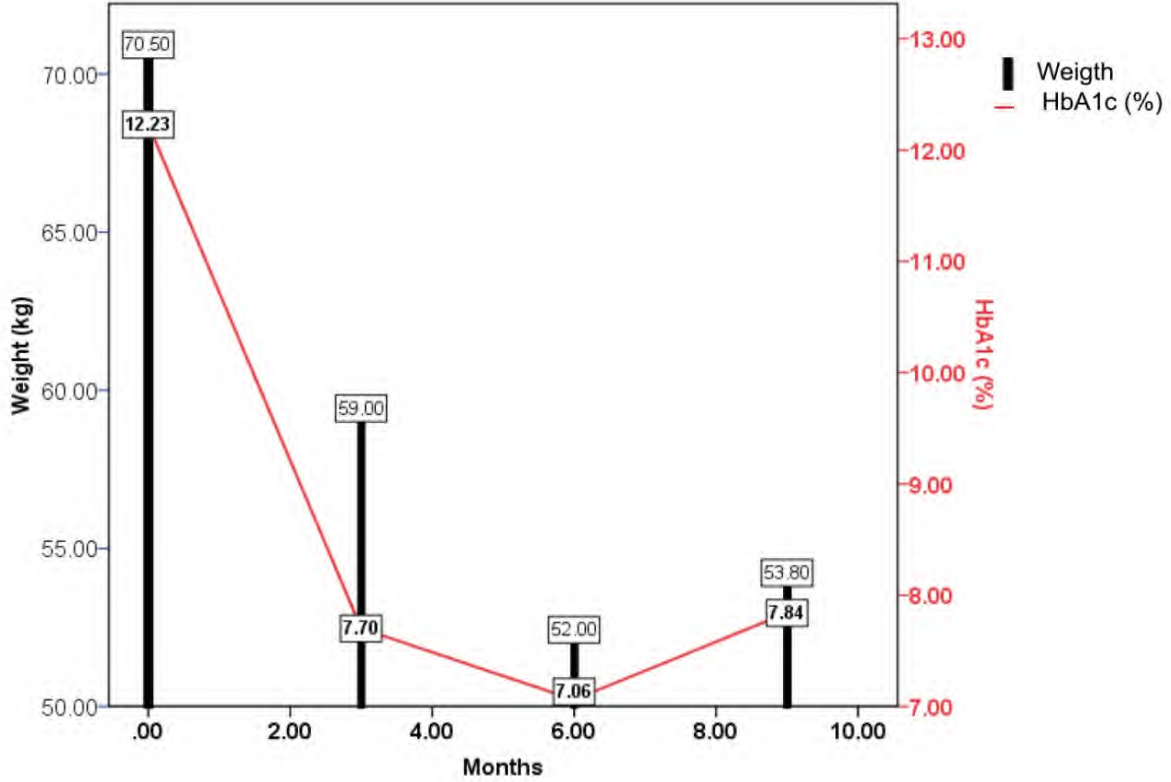


Figure 1: Changes of HbA1c levels and weight

Yazarlarla ilgili bildirilmesi gereken konular (Conflict of interest statement) : Yok (None)

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