reports

Case Report

Ketogenic Diet and Physical Exercise on Managing Tarlov Cysts: A Case Report

Roberto Cannataro [1,†], Cherubino Di Lorenzo [2,†], Mario Iazzolino [1], Maria Cristina Caroleo [1,3], Luca Gallelli [1,3], Giovambattista De Sarro [3] and Erika Cione [1,4,*]

[1] Galascreen Laboratories, University of Calabria, 87040 Rende, CS, Italy; rcannataro@nutrics.it (R.C.); iazzolinomario@yahoo.it (M.I.); mariacristinacaroleo@virgilio.it (M.C.C.); gallelli@unicz.it (L.G.)
[2] Department of Medico-Surgical Sciences and Biotechnologies, Sapienza University of Rome Polo Pontino, 04100 Latina, LT, Italy; cherub@inwind.it
[3] Department of Health Science, University of Magna Graecia, 88100 Catanzaro, CZ, Italy; desarro@unicz.it
[4] Department of Pharmacy, Health and Nutritional Sciences, University of Calabria, 87036 Rende, CS, Italy
* Correspondence: erika.cione@unical.it
† These authors contributed equally to this work.

Abstract: Tarlov Cysts is a pathological condition, with low incidence, characterized by a painful component with a strong impact on quality of life. The therapeutic options are surgery or analgesics and/or anti-inflammatory medications; however, the condition is still without resolution. Herein, we are reporting a case of a woman who expressly followed a low-calorie ketogenic diet program for 3 months. In addition to the change in diet, an appreciable decrease of weight (−5 kg) and body circumferences were recorded; there was also a marked improvement (evident from the questionnaires administered) in the quality of life, of sleep, and in the perception of pain. It is interesting to note how, in conjunction with the Christmas period, upon leaving the ketogenic regime, there was a recurrence of symptoms, confirming the beneficial effect of the low-caloric ketogenic diet at least on the management of pain and, very likely, on inflammation.

Keywords: ketogenic diet; inflammation; pain management; sleep quality; QOL

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1. Introduction

Tarlov Cysts (TCs) are benign, extradural spinal cystic lesions of the nerve root sheath at the dorsal root ganglion, filled by cerebrospinal fluid (CSF), and are usually developed between the endoneurium and perineurium in the lumbosacral spine. TCs etiology is unknown, however, and both a congenital and secondary to trauma or inflammation origin were postulated. The anatomopathological hallmark is that the nerve root is embedded into the membranous connective tissue cyst wall, or its cavity, and often has micro-connections to the subarachnoid space, with liquid circulation between the cyst and spinal CSF. Some of these saccular lesions have a ball-valve mechanism that leads to the expansion of pressure and volume of the cysts. Due to the increase of volume, TCs can compress adjacent nerve fibers, causing pain and other symptoms. Since there are frequently multiple cysts, the clinical presentation can be complex with a multi-nerve compressive symptomatology; in most cases, TCs are fully asymptomatic and their finding is usually accidental when performing lumbosacral spine imaging or postmortem autopsies. In some cases, cysts erode bone structures or induce a caudal syndrome. Conservative treatment by analgesics, anti-inflammatory drugs, and physical therapy is the first therapeutic choice, while the indication of a surgical procedure for symptomatic TCs is still disputed, but it should be attempted to reduce pain, although there is a risk of cyst recurrence [1]. There is no specific, accepted therapy for individuals with symptomatic TCs. Herein, we present a case of a woman who underwent to ketogenic diet and ameliorate chronic pain symptoms linked to this condition.

2. Case Presentation Section

A 56-year-old woman was self-referred to us to start a ketogenic diet for the purpose of weight loss, but she also heard about a possible positive effect on pain related to the diet. At the first examination, her BMI was 35.6 (height 158 cm/weight 89.1 kg). She had an history of major depressive disorder, although it was not pharmacologically treated because of the low compliance to the treatments and the limited benefit she experienced by drugs. Moreover, she was affected by a severe dorsal in the tract between D8 and D11 and lower limb neuropathy due to the presence of multiple bilateral TCs localized in the tract between L4 and S2, with evidence of sacral bone erosive phenomena (see Figure 1). Neurological symptoms included: tingling, numbness, pins and needles, stabbing and shooting pain, stress incontinence, lack of coordination, sleep problems due to night-time pain, muscle weakness, difficulty in limb movements, and episodes of falling. A recent neurological examination evidenced antalgic posture, bilateral steppage gait, and reduced reflexes from the patellar down; Lasègue's test was bilaterally positive at 45°. Upper motor signs, as clonus and Babinski sign, were absent. Under a therapeutic point of view, the patient was at the beginning addressed to physiotherapy, but with very limited benefits. Moreover, because of organizational issues, the patient was unable to follow treatments regularly, resulting in a low compliance to the treatments and the choice to stop them. In a neurosurgery consultation, she did not receive the indication to undergo the surgical procedure. Therefore, the only available treatment was a pharmacological one. Gabapentin up to 2400 mg/day and pregabalin up to 450 mg/day were ineffective on the symptoms. Duloxetine and amitriptyline were not tolerated by the patient. Additionally, different types of opioids gave her different side effects, inducing their discontinuation. The daily use of indomethacin lead to gastritis and she stopped the treatment. At the time of consultation, she regularly consumed 200 mg/day of celecoxib to reduce the pain, and, on demand, acetaminophen alone or in combination with codeine. Moreover, she was on the waiting list for a further neurosurgical consultation.

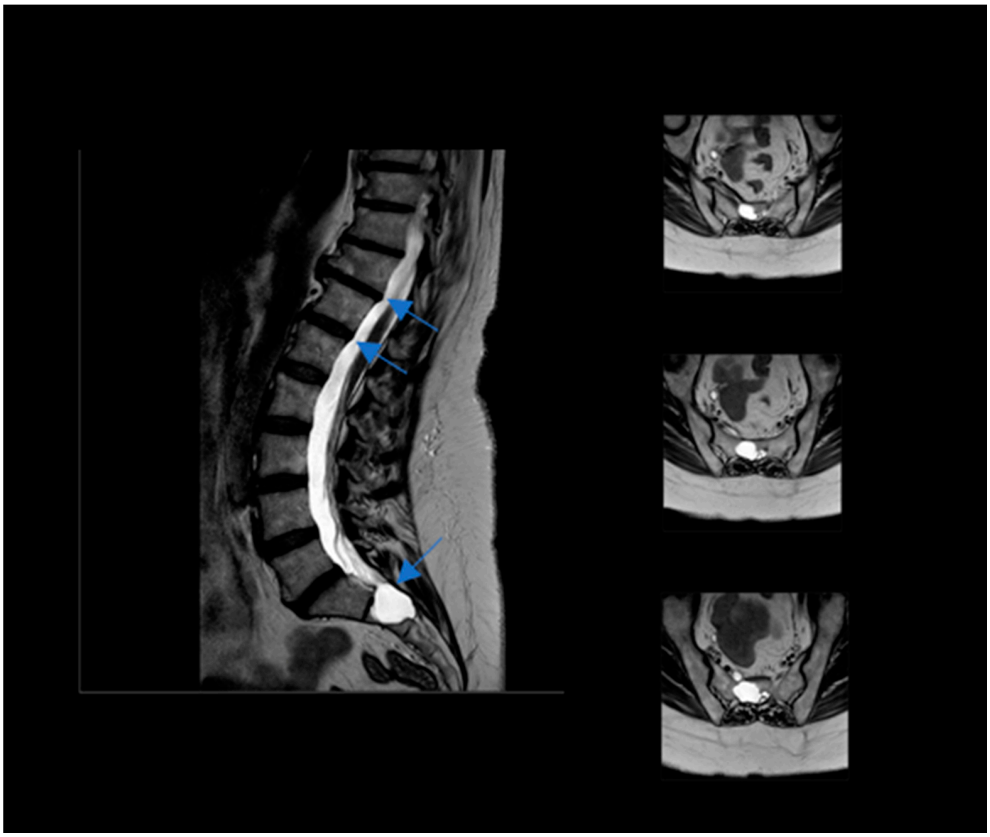

Figure 1. Nuclear Magnetic Resonance of the patient Tarlov's Cyst indicated by blue arrow.

2.1. Data Collection and Questionnaires

A few days before our first evaluation, she completed a self-reported Hamilton Depression Rating Scale (HDRS) [2], Western Ontario and McMaster Universities Index (WOMAC) [3], Sleep Quality Scale (SQS) [4], RAND-36 [5], and VAS [6]; the questionnaires were administered every month.

2.2. Bioimpedance Analysis

The bioimpedance analyzer used for bioimpedance analysis (BIA) was a BIA 101 Anniversary, Akern, Florence, Italy [7,8], and was used with alternating current at a frequency of 50 kHz. The accuracy of the BIA instrument was validated before each test session, following the manufacturer's instructions. Measurements were made on a medical bed isolated from electrical conductors. Briefly, the patient was in the supine position with legs (45° compared to the median line of the body) and arms (30° from the trunk) abducted. Alcohol was used to clean the skin and two electrodes were placed on the right hand and two on the right foot. Resistance (R) and reactance (Xc) parameters were divided by standing body height in meters. Phase angle (PhA)was calculated as the arctangent of $Xc/R \times 180^\circ/\pi$. Body fat percentage was calculated via Bodygram™ software (Akern, Florence, Italy).

2.3. Ketogenic Program

The subject had asked to follow a low-calorie ketogenic diet (KD) program, which was largely considered favorable by us; in fact, a low-caloric ketogenic nutritional plan is used not only for the purpose of slimming, but also to support the therapy of various pathologies such as neurological ones [9]; in particular, it has recently been proposed that KDs can help to improve migraine [10], lipedema [11], polycystic ovary [12], sarcopenia [13], and even some cancers [14]. The program generally provides a very limited intake of carbohydrates, less than 30 g per day or less than 5% of total calories. We choose a caloric deficit of 200–250 kcal that comes from a 14-day food diary where we applied a similar protocol in previous papers [11] to better adapt the nutritional program; the nutritional program was set to have a carbohydrate amount that was less than 20 g per day, where the ratio between proteins and fats is between 1:1 and 1:2, and we chose a moderate-high protein intake to preserve muscle mass as much as possible. We did not exclude any kind of food, so milk and dairy products, meat, and even gluten are included (the latter in any case in a reduced amount being linked to cereals or keto-specific food); we introduced the following as nutritional supplements: omega3 fish oil (4 g per day of supplement, giving around 2.4 g of DHA + EPA), ascorbic acid (1 g per day into 2 times), and vitamin D (2000 IU per day) as we noted a deficiency. All supplements are from 4+ Nutrition, Padua—Italy. Table 1 details the daily plan.

The patient was instructed on how to prepare food, but also, in this case, with few limitations; for example, even fried foods were not forbidden. The caloric intake was set at 1200 kcal, where 25–33% of that was proteins, 58–65% was fats, and 5% or less was carbohydrates. Protein sources included every kind of meat (veal, beef, pork, chicken, turkey), fish and seafood, and eggs, particularly egg white and milk derivates. We suggested to limit the intake of tomatoes, eggplant, and sweet peppers due to the not-negligible carbohydrate amount; beyond that, we strongly suggested to choose seasonal vegetables. As expected, we completely prohibited tubers, squash, legumes, and any kind of cereal or similar grain (excepting the very small amount at breakfast). The main source of fat was EVO (butter or mayonnaise was permitted as a substitute) and nuts or seeds. The patient had continuous support via text messaging via mobile phone, and we saw her every month in our facility. Few dietary supplements have been used in a targeted way as too often they are used improperly and/or are not scientifically justified, which can sometimes lead to important side effects [15]. In particular, the omega3 fish oil, in our opinion, has a beneficial action, given its proven anti-inflammatory action [16].

Table 1. Scheme of KD plan.

KD Plan	
Breakfast	A small biscuit (15 g—5 g on net carbohydrate)
	1 espresso coffee
1–2 Snacks (during the day)	40 g of seasoned cheese
	20 g of nuts
	Vegetables at will (considering those with reduced amount of carbohydrates)
Lunch	150 g of cod
	Vegetables at will (considering those with reduced amount of carbohydrates)
	20 g of EVO
	Vinegar and spices at will
Dinner	150 g of mozzarella cheese
	Vegetables at will (considering those with reduced amount of carbohydrates)
	20 g of EVO
	Vinegar and spices at will
Strongly suggested to have at least 2 L of water per day, even sweetened soft drink	

2.4. Ketosis Assessment and Unwanted Effects of the KD

Ketosis status was checked weekly in urine (self-administered) via Bayer Keto-diastix (Bayer—Germany) and monthly in blood via a Wellion Galileo Glu/Ket blood glucose meter (Wellion—Austria). With the latter, we recorded a value that was always more than 0.7–0.9 mmol/L at every test; only after Christmas time was it at 0.1 mmol/L. We checked the usual side effects reported following a ketogenic diet: frequent urination, fatigue, hunger, confusion, anxiety and/or irritability, and sweating. Sometimes she reported: constipation, difficulty in focusing, and muscle soreness. However, none of these occurred frequently.

2.5. Physical Exercise

After a careful functional evaluation of the range of motion and proprioceptive capacity, a training program was started. The goal of the first phase was to improve aerobic capacity, with a particular focus on proprioception and simple motor screens, where the loads were extremely limited. After the first five weeks, the program increased to a greater intensity, both from the aerobic and strength point of view. For the entire duration of the program, fundamental importance was given to stretching and the ability to coordinate movements. An example of the training patterns, can be found in the Supplementary Materials of the first mesocycle (Tables S1 and S2) and second mesocycle (Tables S3 and S4).

3. Results

In terms of weight, the result is not striking: there was a loss of 3.1 kg. It is noted, however, that a clear improvement in body composition, as can be seen from the percentage of fat and body circumferences (See Table 2), occurred; this is certainly due to the nutritional program rich in proteins, but, above all, to the effectiveness of the training, which can be seen is in positive progression (just consider the second mesocycle which certainly has a greater intensity and level of difficulty than the first, but was well-tolerated by the subject).

Table 2. Anthropometric parameters.

Parameters	September 2021	October 2021	November 2021	December 2021
Weight [kg]	89.1	86.7	86	85
Bf	43.2	41.4	39.7	38.9
PhA [°]	5.6	5.9	5.8	5.8
Circumference [cm]				
waist	107	105.8	103.8	102.8
hip	111	109.8	109	108
arms (dx and sx average)	39.5	36.5	36.2	36.2
coulotte	117	115.7	114.5	113.5
thigh (dx and sx average)	67.8	65.4	64.8	64.2
calf (dx and sx average)	40.5	39.5	39.5	39.3

This result is also confirmed by the decrease in body circumferences, which would suggest a greater decrease in weight, compensated by an improvement in the muscular condition, despite a low-calorie program. The most important result, in our opinion, comes from the questionnaires that show a clear improvement. In fact, at the third administration, there is an improvement in sleep quality by 57%, the WOMAC and VAS scales relating to pain are improved by 64% and 54%, respectively, the HDRS scale with a score of 8 returns to normal, as illustrated in Table 3.

This means a significant impact on the quality of life in general (see result from RAND-36 in Figure 2). It is very important to note that the fourth administration, operated on 27 December and, therefore, happening after an intake of carbohydrates and thus stopping the state of ketosis, shows a rapid deterioration in the general conditions.

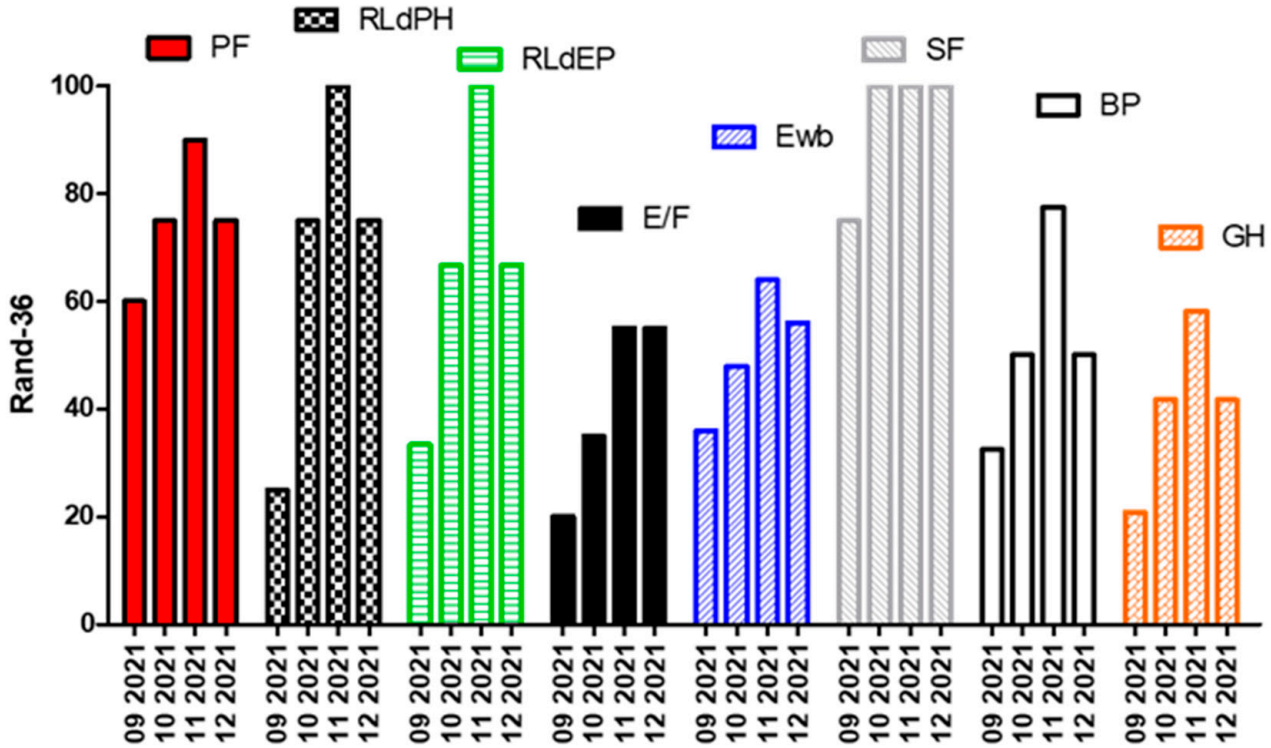

Figure 2. Rand-36 questionnair results: PF, Physical Functioning; RLdPH, Role Limitations due to Physical Health; RLdEP, Role Limitations due to Emotional problems; E/F, Energy/Fatigue; Ewb, Emotional well-being; SF, Social Functioning; BP, Body Pain; GH, General Health.

Table 3. Pain assessment and quality of life.

Recording Data	WOMAC	QS	VAS	HDRS
September 2021	90	44	9.8	23
October 2021	62	29	7	15
November 2021	32	19	4.5	8
December 2021	50	27	6.5	11

4. Discussion

Overall, the ketogenic diet was well-tolerated and the classic side effects were practically absent. Although the period is limited to three months, the result in terms of improved quality of life is important; this could be due to the anti-inflammatory and pain-modulating effect of the KD. The case we report highlights a beneficial effect of the KD on a particular pain condition, being the TCs. It is well established that KDs can be useful for several neurological conditions [9]; however, the pathophysiological basis of pain is fully different, since in migraine it has been proposed that pain comes from the sterile vascular inflammation induced by different neural and metabolic triggers [17], whereas in TCs, pain seems to be due to a mechanical compression of nerve roots, a mechanism that appears to not be modifiable by ketones. However, it has been observed that a KD can exert an anti-inflammatory action that can explain the observed outcome [11]. A further ancillary mechanism of action calls into cause two observed changes in the clinical phenotype of the patient's syndrome. In fact, she experienced an improvement of sleep and mood. There is a bidirectional relationship between chronic pain and sleep disorders [18], and it has been observed that there is an ameliorative effect of a KD on sleep disorders [19]. We can hypothesize that the KD-induced sleep improvement played an additive role on the general benefit experienced by our patient. Likewise, mood disorders and pain conditions are also in a very close relationship [20], and KDs can improve the emotional disorder by different mechanisms of action [21]. Thus, for the KD induced improvement of depressive symptoms, we can also suppose an additional benefit on the patient's global condition. Tempting to speculate about an alternative way to explain the unexpected effect we observe, we can hypothesize that the KD can lead to a modification in cortical pain processing of the patient. Consistent with the literature [22–24], a KD can influence the cognitive cortical response to painful stimulation. Besides that, thanks to the supervised and progressive practice of a physical activity that includes, in particular, proprioceptive aspects, joint mobilization, and stretching, as well as a component of strength, there is an improvement in daily functions; for example, the subject reports much more ease in doing housework, which contributes to the overall positive result. The BIA data and the measurement of body circumferences show an improvement in muscle mass, which is very important in the general management of the condition. It is interesting to note that concomitant with the Christmas period, when the patient, for obvious reasons, did not follow the ketogenic program for three days (ketonemia at 0.1 mmol/L), recorded a recurrence of symptoms, which decreased when returning to ketosis (ketonemia 0.7 mmol/L)

5. Conclusions

The effect of a KD should be investigated more in depth while monitoring classic inflammatory markers, such as CRP or interleukins, or miRNAs linked to the inflammatory state [25]. Similarly, the application of the KD should be evaluated on a larger sample, to evaluate not only any side effects, but also compliance.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/reports5020012/s1. Table S1. First mesocycle day 1, Table S2. First mesocycle day 2, Table S3. Second mesocycle day 1, Table S4. Second mesocycle day 2.

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Data Availability Statement: Data is contained within the article and in the supplementary material. The data presented in this study are available in our hardware.

Conflicts of Interest: The authors declare no conflict of interest.

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