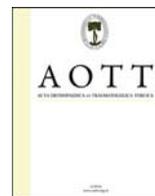


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Acta Orthopaedica et Traumatologica Turcica

journal homepage: <https://www.elsevier.com/locate/aott>

The ultrasonography evaluation of talar dysplasia as a potential prognostic factor for predicting the course and outcomes of clubfoot deformity treatment using Ponseti technique



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ARTICLE INFO

Article history:

Received 18 July 2016

Received in revised form

15 May 2017

Accepted 24 November 2017

Available online 13 February 2018

Keywords:

Ultrasonography

Clubfoot

Achilles tendon

Ponseti method

Tarsal bone

Talar dysplasia

ABSTRACT

Objective: The aim of this study was to assess the role of sonographic evaluation of Talar dysplasia in predicting the outcome of standard Ponseti method in the treatment of clubfoot deformity.

Methods: A total 23 children (15 boys and 8 girls; mean age: 18.2 ± 5.4 days (8–32)) who underwent Ponseti treatment were included in the study. Before the treatment, maximal talus length of affected and non-affected feet were measured by US and relative talar dysplasia ratio (RTDR) was calculated. The patients were categorized 2 groups according to RTDR: group A – mild and group B – severe deformity. Pirani score was used for clinical evaluation. The groups were compared in terms of number of the applied casts, need of percutaneous tenotomy of Achilles tendon (AchT) and frequency of deformity recurrence.

Results: Pirani score was 4.46 for population (4.33 for group A; 4.54 for group B). Number of casts significantly differed between groups ($p < 0.001$) and positive correlation was found ($r = 0.851$, $p < 0.001$). AchT was performed in 56% cases for group A and in 86% cases for group B; no statistically significant difference was obtained ($p = 0.162$). Recurrence occurred in 2 patients belonging to group B without significant difference compared to group A ($p = 0.502$).

Conclusion: Talar dysplasia assessment appeared as a promising prognostic factor for predicting the outcome of the Ponseti technique in treatment of clubfoot deformity.

Level of evidence: Level IV, diagnostic study.

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Introduction

Pes equinovarus congenitus (PEC, clubfoot, talipes) is the most frequent congenital structural defect of the feet with an incidence of 1–2/1000 among newborns.¹ It has already been more than 15 years since conservative Ponseti technique became a gold standard treatment method² with effectiveness estimated to approximately 90%.^{3–5} Nevertheless, the clinical follow-ups showed that not every clubfoot deformity responds to this method in a similar way; there

is a different degree of resistance to the treatment and many seemingly cured deformities have the tendency to reoccur. Up to date, there is no method which could predict when the affected feet will be responding worse to Ponseti plaster casting, eventually when the possible recurrence should be taken into account.

Nowadays, certain predictions and assessments could be stated based on the clinical examination of the feet before and during treatment. Although there are sophisticated scoring systems commonly used, especially the ones described by Dimeglio⁶ and Pirani,⁷ they represent a clinical evaluation based on the subjective assessment, which could be accompanied with a risk of possible examiners' errors or a bigger interobserver variability. Moreover, there has not been accepted unified opinion concerning clear correlation between clinical examination prior to treatment obtained by scoring systems and the outcomes of the treatment in published papers.^{8,9}

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Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

One of the possibilities how to objectively assess certain parameters of the deformity is a radiographic examination. Considering age of the patients when the treatment is initiated, we face the problem of incomplete ossification of the feet bones and the eccentric placement of ossification centres that usually appear later compared to the foot without deformity.^{10,11} Furthermore, a radiation dose in repeated radiographic examinations in children must be taken into account. The same situation occurs when considering CT examination, which, additionally, needs to be performed under general anesthesia in newborn patients. Although magnetic resonance imaging (MRI) is not burdened by the radiation dose, there is also the necessity of general anesthesia in regard to obtain the images of satisfactory quality. Additionally, the cost of this examination plays an important role as well.

Considering limitations of the different modalities mentioned above, the ultrasound (US) imaging of children feet appears to be perspective and optimal. It is widely available, even in routine orthopaedic clinics, relatively simple after training, easily reproducible and inexpensive. The recent literature already described US examination for clubfoot deformity.^{12–15} The authors often described extensive evaluation and measurements of complex parameters of deformity, which are not suitable for a daily routine orthopaedic practice. Furthermore, the US examination is often performed by radiologist and orthopaedists evaluate the deformity only in a clinical way and take care of the treatment. Yet, even these findings in most cases do not clarify the issue of US parameters of clubfoot deformity that would be suitable for the possible prediction of a course and outcomes of the treatment. Moreover, some findings appeared very often contradictory.

Thus, when searching for a suitable parameter which could be easily examined by a clinician, we worked on the presumption that tarsal bones dysplasia is accompanied with clubfoot deformity. In this respect, the most affected bone is talus¹⁶ as it is suggested by the term of the deformity - talipes equinovarus. Therefore, we focused on US examination of talus and we tried to find relationships between the talar dysplasia and (1) number of casts required for deformity correction, (2) necessity of percutaneous tenotomy of Achilles tendon (AchT) and (3) the possibility of the recurrence of the deformity. These 3 criteria were set as indicators of successfulness of the course and treatment of PEC deformity.

Materials and methods

Between January 2014 and March 2015, 23 patients (15 boys and 8 girls) with unilateral PEC deformity treated with Ponseti method in our institution were included in the prospective study. All children were treated and followed by one orthopaedic surgeon from the institution (JJ). Only children with idiopathic and unilateral deformity were enrolled in the study in regard to directly compare affected feet and their dimensions with the healthy feet without clubfoot deformity. The children with different aetiology of the deformity were excluded. None of the children was treated in a different institution prior to inclusion in our study.

Parents of all included children were fully informed and signed informed consent drawn from the authorities of our hospital and formally agreed to participate in the prospective study. Parents could have left the study anytime voluntarily without any consequences but this situation didn't occur.

Clinical evaluation

Patients were clinically examined and categorized based on the Pirani scoring system which represents clinically effective tool to assess each of the components of the deformity⁷ prior to treatment. The six components were divided into three ones related to the

hindfoot (severity of the posterior crease, emptiness of the heel and rigidity of the equinus), and the rest of them were related to the midfoot (curvature of the lateral border of the foot, severity of the medial crease and position of the lateral part of the head of the talus). Each of the components was scored as follows: 0 – no abnormality; 0,5 – mild abnormality and 1 – severe abnormality. Thus, total score could range from 0 up to 6 and Pirani score of 6 represented clinically the most severe form of clubfoot abnormality.

Ultrasound measurements

All ultrasound measurements were obtained by US machine Aloka Prosound 2 (Aloka Holding Europe, Steinhauserstrasse, Switzerland) or Mindray (Shenzhen, China) DP-50 using 7,5 MHz linear probes. The examination was performed by one orthopaedic surgeon experienced in US imaging and intraobserver variability was calculated.

From all different projections already presented in literature, the dorsal projection described by Aurel et al¹⁸ for measurement of maximal talar length was applied. Using this projection, the probe has to be placed in the sagittal position on the dorsum of the examined foot following the direction of the talar axis. For examination of the feet with PEC, the probe should be placed more laterally in order to capture the talar axis properly. Images were recorded in the habitual position of the affected foot as well as in the position when the foot was held in maximal correction.

The healthy foot was assessed first in regard to keep children calm for further measurements. The talar length was measured in millimetres (mm) (Fig. 1). The ratio of the maximal talar length of

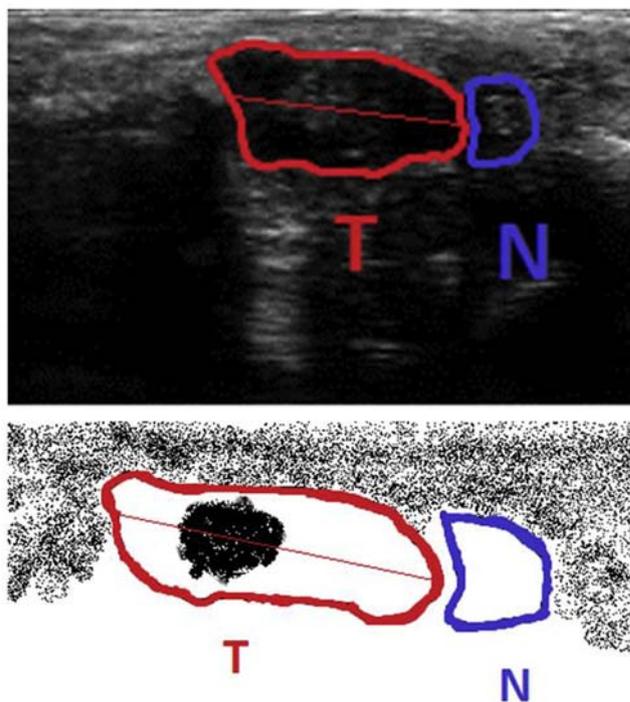


Fig. 1. Schematic demonstration of the ultrasonography examination and measurements when the dorsal projection is used (T – talus, N – navicular bone).

the affected (Taf) and not affected (Tnaf) feet was counted and it was set a criterion of the talar dysplasia (relative talar dysplasia ratio (RTDR) = Taf/Tnaf), based on which the patients were divided into 2 groups - mild form with RTDR higher than 0,75 (group A) and severe form of talar dysplasia with RTDR below or equal to 0,75 (group B).

The therapy was carried out according the principles of Ponseti treatment. The first plaster cast was applied after clinical and US examinations to correct cavus deformity component. The repeated correction plaster casts were changed with the interval of 1 week until full correction was achieved and number of casts was recorded. AchT under general anaesthesia was performed when satisfactory dorsiflexion was not achieved after the last correcting plaster cast. In patients without AchT as well as in patients after AchT the final plaster cast fixation was applied in dorsiflexion position for 3 more weeks. Afterwards, Denis – Brown splints with the plastic AFO splints set to 70 degrees of external rotation for the affected foot and 40 degrees of the external rotation for the healthy foot were applied. All parents were instructed to wear these splints in a special daily regime – first 3 months for 23 h a day and after this period for day and night sleeping time. Patients were clinically monitored in 3-months intervals.

The effectiveness and the course of the treatment were evaluated by objective criteria as follows: number of cast fixations, number of AchT and the recurrence of the deformity during the period of the study.

Statistical analysis

Descriptive statistics for population was expressed as a mean \pm SD (minimal – maximal values) for continuous variables, while categorical variables were reported in percentage (%). Normal distribution of the data was tested by Shapiro–Wilk test. Mann–Whitney test was used to compare the number of correction casts between group A and group B. Then, the Fisher's exact test was used to compare the difference between groups in terms of number of AchT and recurrences of the deformity. Furthermore, Pearson correlation was applied to define the relationship between the talar dysplasia and number of correction casts, AchT and the recurrences. All *p* values were 2-sided and a *p* value below 0,05 was considered statistically significant. Data analysis was conducted with SPSS version 20.0 (SPSS INC, Chicago, IL, USA).

Results

The patients' population consisted of 15 boys and 8 girls. Right and left feet were affected in 14 and 9 cases, respectively. The average age of the population when therapy started and US examination was performed, was $18,2 \pm 5,4$ days (8–32) and mean

Table 1
Descriptive characteristics of the population.

	Mean/Total	SD	Min	Max
Nb. of patients	23	–	–	–
Talus length – unaffected foot [mm]	18,3	1,3	15,5	21,0
Talus length – affected foot [mm]	13,6	1,2	11,5	17,0
PIRANI score	4,46	0,91	3	6
Age of therapy initiation [days]	18,17	5,38	8	32
Nb. of plaster casts	6,83	2,14	4	10
AchT [%]	17	74%	–	–
Recurrence [%]	2	9%	–	–
Follow up [months]	13,17	4,00	7	22

The table provides general information about the population which was enrolled in the study. The values are express as mean, standard deviation (SD) and minimal (Min) and maximal (Max) obtained values or as and percentage (%), if possible. (AchT – percutaneous tenotomy of Achilles tendon).

Pirani score at the beginning of the therapy was $4,5 \pm 0,9$ (3–6). General characteristics of the population are summarized in Table 1.

Normal distribution of the data was not achieved.

The average number of correction casts for group A was $4,6 \pm 0,7$ (4–6) and for group B $8,3 \pm 1,3$ (6–10), which revealed significant difference ($p < 0,001$) between the groups (Table 2). Furthermore, strong correlation was found statistically significant ($r = 0,851$, $p < 0,001$) (Fig. 2).

There was necessity for 5 (56%) AchTs in group A but, in contrary, 12 AchTs (86%) were performed in group B. No statistically significant ($p > 0,05$) difference between groups using Fisher's exact test and no significant correlation were found ($r = 0,335$, $p > 0,118$) (Table 2).

During the study, the recurrence of the deformity appeared in 2 cases (8,7%) and the process of plaster casting with AchTs had to be repeated. These 2 cases belonged to the group B representing patients with severe form of the deformity. No statistically significant difference between groups ($p = 0,502$) nor correlation ($r = 0,247$, $p = 0,255$) between the severity of talar dysplasia the recurrence occurrence were found.

The mean follow-up period was $13,2 \pm 4$ (7–22) months (Table 1).

Intraobserver variability tested on 15 consecutive patients of our population was calculated as excellent ($r = 0,92$, $p = 0,001$).

No complications were observed during treatment.

Discussion

Although PEC deformity is known for thousands of years, certainly not all the questions regarding the treatment of the deformity were satisfactory answered, and not even in this time when the conservative concept of treatment described by professor Ponseti was adopted as a gold standard method of treatment. In spite of the undeniable effectiveness of this method, we cannot effectively treat all clubfoot deformities with this procedure and about 14% of all cases present with the recurrence after the treatment.¹⁷

The US examination was described in some studies as an option in searching for objective parameters to predict the outcomes of the clubfoot deformity treatment.^{13,16,19} Despite of numerous recently published studies, there is no consensus about the validity of US assessment of measured parameters among different authors.

Some authors tried to find objective US parameters and their relationship to the severity of the deformity when measuring the angles of the tarsal bones or the bones of the lower leg.^{14,20} For example, Desai et al²¹ used talo-cuneiform angle (TCA) as a

Table 2
Comparison of the group with the mild and severe form of talar dysplasia.

	Group A	Group B	<i>P</i> value
Nb. of patients	9	14	
PIRANI score	$4,33 \pm 0,88$ (3–6)	$4,54 \pm 0,92$ (3–6)	0,502
Age of therapy initiation [days]	$19,11 \pm 5,76$ (8–32)	$17,57 \pm 5,02$ (11–28)	0,445
Nb. of plaster casts	$4,56 \pm 0,68$ (4–6)	$8,29 \pm 1,33$ (6–10)	0,001
AchT [%]	5 (56%)	12 (86%)	0,162
Recurrence [%]	0 (0%)	2 (14%)	0,502
Follow up [months]	$12,44 \pm 3,8$ (7–20)	$13,64 \pm 4,05$ (7–22)	0,487

The table shows the differences between groups divided by the relative talar dysplasia ratio (RTDR) – group A represents patients with mild form of the deformity and group B represents severe form of clubfoot deformity. Particular values were compared and statistically evaluated. Only number of plaster casts differs between groups, the rest of variables were not statistically significantly different. The values are express as mean, standard deviation (SD) and minimal (Min) and maximal (Max) obtained values or as and percentage (%), if possible. (AchT – percutaneous tenotomy of Achilles tendon).

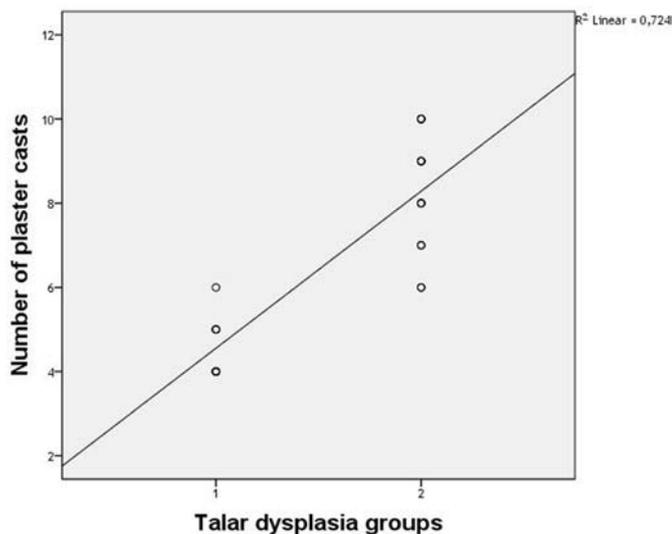


Fig. 2. The scatter plot graph represents the positive correlation found between the number of plaster casts applied during treatment and the severity of the deformity. Pearson correlation revealed significant correlation.

parameter for assessing and monitoring clubfoot deformity correction during treatment period. As described, TCA was formed by the first line drawn along the medial border of talar ossified nucleus and the second line was drawn along the medial border of the cuneiform at the anterior end of the talar nucleus. Aurell et al¹² casted doubt on measurements resulting from angles assessments due to the fact that it is difficult to obtain reliable dimensions to calculated angles in irregular-shaped bones which are, in addition, only partially ossified. Although they also tried to evaluate angles to describe the degree of clubfoot deformity, they found poor reliability of the obtained results.

Different measurements were proposed to assess the severity of the deformity. El-Adwar et al¹⁹ published paper with the set of complex measurements using medial malleolus and navicular bone distance (MMN), calcaneocuboid distance (CC) or shift of the navicular bone against talus and correlated the variables with the Pirani score before and after treatment. However, no consistent opinion was seen concerning CC as a reliable parameter throughout the literature. Aurell et al (2) concluded that CC distance is of limited use because of its small range. Although negative correlation between MMN and midfoot score was found before treatment in El-Adwar et al study,¹⁹ this finding doesn't lead us to the prediction of the response of the affected feet to the Ponseti technique treatment.

Concerning the talo-crural, the talonavicular and the calcaneocuboid joints, MMN distance was precisely evaluated and set as a possible indicator of the severity of deformity by Aurell et al¹³ although they agreed with the opinion that the simple classification of the clubfoot deformity is not possible.

Nevertheless, the MMN distance consists of several components which include a displaced navicular, an increased medial deviation of the neck of the talus, a short talar body and a forced adduction of the forefoot.¹³ As it was already observed, talar dysplasia is always accompanied with clubfoot deformity and talus presents with different shapes and eccentric ossification centres before the treatment is initiated.¹³ Due to undeniable fact that talus is affected in clubfoot deformity, we hypothesized that the talar length could play a crucial role in predicting the deformity treatment outcomes.

Up to date, the only study was published concerning the length of talus as a predicting parameter. Chandrakanth et al¹⁶ focused on US measurements of the dysplasia of tarsal bones occurring in clubfoot deformity. They reported significant correlation between

talar dysplasia and the number of casts needed to correct the deformity, no significant correlation was found between dysplasia of navicular or calcaneus and number of plaster casts. Furthermore, patients in the study with clubfoot deformity were categorized into 3 groups based on the relative talar dysplasia ratio: mild dysplasia (RTDR = 0.9 to 1.0), moderate dysplasia (RTDR = 0.8 to 0.89), and severe dysplasia (RTR < 0.8). We decided to divide patients into 2 groups with the threshold of RTDR of 0,75 because the talus length in newborns were very small and dividing patients into more than 2 groups based on RTDR would be more likely influenced by the interobserver variability, measurements errors and measurements bias. Nevertheless, we believed that distinguishing between severe and mild forms would be satisfactory for daily routine clinical practice. For talus, Chandrakanth et al¹⁶ found statistically significant correlation between the severity of the deformity and number of casts applied during treatment and no correlation between severity of the deformity and AchT, which is in accordance with our study. Although statistically significant association was not found between dysplasia of the talus and the number AchT in our study, group A needed this procedure in 56% of cases compared to 86% in group B to achieve sufficient correction which we consider as an important finding.

In comparison to Chandrakanth et al¹⁶ study in which authors didn't observe any recurrence of deformity, we reported 2 recurrences, both of them occurring in group B, although our populations were comparable in terms of number of patients. It could be explained by the fact, that our patients were followed for significantly longer time (mean 13 months) compared to 6 months follow-up period of their study. At this point, we would like to emphasize the need of intensive and regular follow-up of patients with clubfoot deformity for at least 12 months, what can be crucial time in terms of early relapses, especially in patients with severe forms of the deformity. Nevertheless, patients are monitored in our institution with the clubfoot until the skeletal maturity. Because of the small number of patients, the statistically significant correlation between the talar dysplasia and the recurrence of the deformity was not found.

In our study, only dorsal projection was chosen to be used because it offers the most accurate information about real length of the talus and it helps to obtain its maximal possible values compared to other projections. As we didn't evaluate angles, joints and soft tissue dimensions, the medial or lateral projections were not necessary to be performed. By choosing only one projection we tried to simplify the measurements to be applicable in daily routine practice and such measurements could be fast and simply performed by single experienced orthopaedic surgeon.

There are some limitations in our preliminary study. The population included is relatively small and further data collections and patients' inclusion are necessary to confirm achieved results. On the other hand, the number of patients is similar to already published studies. We did not assess interobserver variability as the measurements and evaluations were run by only one senior paediatric orthopaedic surgeon (JJ) with sufficient experiences in musculoskeletal US imaging. Because of the presence of only one experienced orthopaedics surgeon in US evaluation of talus at the time of the study, only intraobserver variability was calculated in 15 consecutive patients from the population as stated in the results. This fact brings along the advantage of unified and standard approach to all patients on one hand; on the other hand, it could be burdened by individual bias despite the fact that the US, considered as an objective method for assessment of different parameters, was used.

Conclusion

Relative talar dysplasia ratio measured during US examination of foot with clubfoot deformity appeared as a promising prognostic

parameter which could predict the future course and outcome of the treatment of clubfoot deformity with Ponseti technique. It is necessary to distinguish between severe and mild forms of talar dysplasia. Based on our results, patients with severe form of deformity would need significantly larger amount of plaster correction casts; more than 80% of them would need AchT after plaster casting and there is possibility of the recurrence of deformity (8,7%) in this group of patients compared to the patients with mild form of talar dysplasia. These findings could lead clinicians to reconsider treatment protocols and to count with suboptimal outcomes of the therapy. Nevertheless, the clubfoot deformity is a complex diagnosis and many other factor can contribute to results in terms of prognosis of the course and outcomes of the treatment. Further studies are still necessary to be done to assess the deformity from different perspectives.

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Nb.	Sex	Laterality	RTDR group	nb of casts	Age of initiation of therapy	Pirani score	Follow-up (months)	Achilotenotomy	Recidives	Talus length of unaffected foot (mm)	Talus length of affected foot (mm)	Relative talar dysplasia ratio
1	m	dx	1	4	19	3	20	0	0	18.3	14.5	0.79
2	m	sin	1	5	21	6	15	1	0	17.5	13.5	0.77
3	ž	dx	1	5	17	4	15	1	0	16	13	0.81
4	m	dx	1	4	19	4.5	14	0	0	19	14.5	0.76
5	ž	sin	1	6	32	5.5	12	1	0	18.5	14	0.76
6	ž	dx	1	5	19	3.5	11	0	0	20.5	17	0.83
7	m	dx	1	4	18	4	10	1	0	18.7	15	0.80
8	m	dx	1	4	19	4	8	0	0	17.5	14.2	0.81
9	m	dx	1	4	8	4.5	7	1	0	18.3	14.5	0.79
10	m	dx	2	6	12	6	22	1	1	19	13.5	0.71
11	ž	dx	2	10	14	3	19	1	0	18.5	13	0.70
12	ž	sin	2	7	15	5	17	0	0	17	12.5	0.74
13	m	sin	2	8	18	6	16	1	0	19	14	0.74
14	ž	dx	2	6	25	3.5	16	1	0	18.5	13.5	0.73
15	ž	sin	2	8	11	3	14	0	0	18.5	12.5	0.68
16	m	sin	2	9	23	4.5	14	1	0	15.5	11.5	0.74
17	m	sin	2	9	19	5	13	1	0	18	13	0.72
18	m	dx	2	10	20	4	13	1	0	20	13.5	0.68
19	m	dx	2	7	17	4.5	12	1	1	21	15.5	0.74
20	m	sin	2	8	14	5	11	1	0	18.5	13	0.70
21	m	dx	2	9	11	4	9	1	0	19	13	0.68
22	ž	dx	2	9	28	5	8	1	0	17	12	0.71
23	ž	dx	2	10	19	5	7	1	0	16.5	12.3	0.75