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## I. RADIOLOGY

# HIRSCHSPRUNG DISEASE – THE CONTRIBUTION OF THE RADIODIAGNOSTIC CASE REPORT OF ONE RARE VARIANT WITH COMPLICATIONS

**M Popescu, CM Popoiu**

Universitary and Emergency Hospital for Children “Louis Turcanu” from Timisoara

### Abstract

The following describes the case of a female suckling who, at 6 weeks of life, has been hospitalized at the

Infant Surgery Clinics within “Louis Turcanu” Emergency Hospital from Timisoara.

Both her anamnesis and clinical aspect determined the clinician (surgeon) to demand a series of investigations such as abdominal radiological exams which showed modifications (resulted from the presence of some hydroaeric levels).

The radiological exam with contrast substance confirmed our suppositions regarding the diagnostic of Hirschsprung Disease (HD). Moreover, a thorough analysis of the patient’s radiological aspect determined the radiologist to diagnose her with a rare form of Hirschsprung Disease, more precisely the ultra – short segment HD.

Infectious complications as enterocolitis and septicemia, forced the clinician (surgeon) to resort a two times surgical intervention.

**Key words:** Hirschsprung Disease (HD), congenital aganglionosis, water-soluble contrast enema (barium enema), ultra - short segment HD.

### Introduction

Harold Hirschsprung published the classic description of the congenital megacolon in 1886. Hirschsprung Disease (HD) is characterised by the absence of the myenteric and submucosal ganglion cells in the distal digestive tract. The disease results in decreased motility in the affected bowel segment (5). This causes a blockage. Intestinal contents build up behind the blockage, causing the bowel and abdomen to become swollen (8). Hirschsprung Disease causes about 25% of all newborn intestinal obstruction (8).

HD is regarded as a neurocristopathy because it involves a premature arrest of the craniocaudal migration of vagal neural crest cells in the hindgut at

weeks 5-12 of gestation to form the enteric nervous system (5). The aganglionic, aperistaltic bowel segment effectively prevents the propulsion of the fecal stream, resulting in dilatation and hypertrophy of the normal proximal colon (5).

Hirschsprung Disease (congenital aganglionosis) is caused by a single gene mutation of the RET proto-oncogene on band 10q 11.2 (3). As a congenital disorder, HD is manifested mostly in the first several weeks of life (5).

HD is estimated to occur at a rate of 1 case per 5000 live births (5). Males are affected more often than females, with a ratio of 4:1 (5).

HD is sometimes associated with other inherited or congenital conditions such as Down syndrome (2,5).

Tests used to help diagnose HD may include: abdominal x-ray, anal manometry, barium enema and rectal biopsy.

### Anamnesis and clinical findings

Female suckling with habitual constipation is hospitalized at 6 (six) weeks at Arad County Hospital between May 21, 2008 and May 23, 2008. The patient is hospitalized with the following diagnoses: hemolytic anemia (infectious?), post-transfusion reaction (hemoglobinuria, hematuria, icterus) overlapped with the initial anemia, invasive acute enterocolitis, dynamic ileus.

The patient is transferred at the University and Emergency Hospital for Children from Timisoara where initially is hospitalized in the Clinic no.3 Pediatrics. On May 23, 2008 the clinician observes: general state profoundly altered, no fever, intense pale teguments with sclerotic and tegument icterus. Between May 24, 2008 and May 28, 2008 the clinician observes and notes the state of the abdomen: distended, flat and relaxed, discrete distended, distended, distended. She is then transferred at the Surgery Clinic where repeated abdominal radiographic

examinations revealing modifications, the barium enema is recommended. On June 2, 2008 this examination was performed and the radiologist has offered the diagnostic – congenital megacolon; on June 11, 2008 the patient has undergone a operation when

the aganglionic segment was resected and the sigmoid colostomy was performed. The patient was discharged with the pathology (congenital megacolon, acute enterocolitis, severe sepsis with enterococcus, severe secondary anemia) removed.

### Paraclinic findings

#### A. Laboratory tests:

##### a. Blood cells count:

Date:	Arad,	Timisoara	29.05.2008,	08.06.2008
Hemoglobin g%	4,6		6,2	
Erythrocytes/mm <sup>3</sup>	1.690.000		2.080.000	4.310.000
Leucocytes/mm <sup>3</sup>			24.400	7.230

##### b. Serum electrolytes level:

Date:	Timisoara	29.05.2008,	03.06.2008
Na mmol/l		129	128
K mmol/l		4,3	4,5
Ca mmol/l		1,23	1,24
Cl mmol/l		93	92

##### c. Cultures: blood cultures

Date:	Timisoara	02.06.2008
		enterococcus

#### B. Radiological aspects:

##### a. Plain abdominal radiography – Dates: 24.05.2008; 27.05.2008; 28.05.2008

Marked dilatation of the bowel (small and large bowel).

Air-fluid level. No gas in the pelvis (rectum) (fig. 1).

##### b. Barium enema - Date: 02.06.2008

Reduced caliber of the terminal rectum (with a tubular aspect: 43mm/length and 12 – 16 mm/width) followed by a transition zone to an enlarged caliber of the proximal rectum and sigmoid. Dolicosigma (fig. 2).

#### C. Histopathological exam – Date: 16.06.2008

Congenital megacolon.



Fig. 1. Marked dilatation of the bowel (small and large bowel).  
Air-fluid level. No gas in the pelvis (rectum).



Fig. 2. Reduced caliber of the terminal rectum (with a tubular aspect: 43mm/length and 12 – 16 mm/width) followed by a transition zone to an enlarged caliber of the proximal rectum and sigmoid. Dolicosigma.

### Discussions

Normally, as a baby grows in the womb, bundles of nerve cells (ganglia) begin to form between the muscle layers along the length of the colon. This process begins at the top of the colon and ends at the bottom (rectum). In children with Hirschsprung Disease, this process does not finish and the ganglia do not form along the entire length of the colon. Other times a longer portion may be affected (7).

Aganglionosis begins with the anus, which is always involved, and continues proximally for a variable distance (2). The precise mechanism underlying the development of Hirschsprung disease is unknown (2).

Hirschsprung Disease (HD) can be classified by the extension of the aganglionosis as follow (5):

1. Classical HD (75% of cases): The aganglionic segment does not extend beyond the upper sigmoid.
2. Long segment HD (20% of cases).
3. Total colonic aganglionosis (3-12% of cases).

Some rare variants include the following:

4. Total intestinal aganglionosis.
5. Ultra – short segment HD.

Ultra – short segment HD is characterised by a few centimeters of aganglionic bowel in the rectum, adjacent to the anus (2).

Water-soluble contrast enema is the key study for diagnosis (6). Lateral views of early rectal/sigmoid colon filling are the most important images. The normal rectum should always be equal or of larger caliber than the sigmoid colon: the rectum/sigmoid ratio. In HD rectum/sigmoid ratio is reversed (6).

About 10% children may present with diarrhea caused by enterocolitis, which is thought to be related to stasis and bacterial overgrowth. This may progress to colonic perforation, causing life threatening sepsis(2).

The case in discussion can be classified as a rare form of HD, more specifically, the ultra – short HD, with a 4-5 cm length stenotic segment (emphasized by water soluble contrast enema), affecting not only the anal channel (1) but also the distal part of the rectum. The rectum/sigmoid ratio has been reversed.

Both the enterocolitis and septicemia, proved through positive hemoculture analyses, confirmed the fact that this case can be considered as belonging to the rare group of 10% cases - of HD – diagnosed with infectious complications (2,4).

Under these circumstances and taking into account the type of disease (HD associated with septicemia infection), the two times therapeutic surgical intervention was selected.

### Conclusions

1. In Hirschsprung Disease (HD), water soluble contrast enema is the key for the diagnostic. The rectum/sigmoid ratio is reversed.

2. The case into discussion can be classified as a rare form of HD, ultra – short HD.

3. The infectious complications have determined the surgeon to choose a two times surgical intervention.

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**References**

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|--|---|
| <ol style="list-style-type: none"><li>1. Andronescu A – Anatomia copilului; Editura Didactica si Pedagogica – Bucuresti – 1966.</li><li>2. Steven L. Lee, MD and coauthors, Mar 30, 2006 – Hirschsprung Disease.</li><li>3. David M. Manuel, MD and coauthors Aug 3, 2007 – Hirschsprung Disease.</li><li>4. Frederic N. Silverman, M.D. Caffey's Pediatric X-ray Diagnosis, Year Book Medical Publishers, 1985 – Megacolon.</li></ol> | <ol style="list-style-type: none"><li>5. Ciro Yoshida, Jr., MD and coauthors, Mar 31, 2004 – Hirschsprung Disease.</li><li>6. American Trend in Radiology, Teaching Files, 2008 – Hirschsprung Disease.</li><li>7. Mayo Clinic Staff, Nov. 10, 2006 – Hirschsprung's Disease.</li><li>8. Medline Plus, Medical Encyclopedia, 2008 – Hirschsprung's Disease.</li></ol> |
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## II. NEONATOLOGY

### SISTEMIC CONGENITAL SYPHILIS CLINICAL AND BIOLOGICAL STUDY

**Marioara Boia, Daniela Iacob, Aniko Manea, Mirela Margineanu**

„Louis Turcanu” Children’s Emergency Hospital Timisoara, Romania

#### Abstract

One of the multisystemic infection transmitted to the fetus via the placenta is the congenital, syphilis caused by *Treponema pallidum*. High incidence of the disease led to routinely screening for all pregnant women. Clinical signs in neonatal period appear in the first 5 weeks of life, but signs of the disease may occur late, after the first 2 years of life. Diagnosis based on neonatal serologic testing is complicated by the transplacental transfer of maternal Ig antibodies, which can cause a positive test in the absence of infection. Three significant cases of congenital systemic syphilis treated in the Clinic of Neonatology are presented in this paperwork.

**Key words:** congenital syphilis, neonatal diagnosis.

#### Introduction

Congenital syphilis is a multisystemic infection caused by *Treponema pallidum* and transmitted to the fetus via the placenta.

The rate of transmission is higher in women with primary and secondary syphilis than in those with tertiary syphilis. Up to 40 % of pregnant women with untreated primary syphilis have presented spontaneous abortions.

High incidence of the disease led to routinely screening for all pregnant women.

Two-thirds of the new born with syphilis are asymptomatic at birth. Clinical signs of disease can occur during fetal period, neonatal period or later in childhood with an additional perinatal mortality of 25% - 30% cases. Without treatment, in severe forms of the disease, intrauterine death occurs in 25% of cases with an additional perinatal mortality of 25% - 30%.

Clinical signs in neonatal period appear in the first 5 weeks of life and they are: ulcerative skin lesions on the palms and sole (occur in severe forms of the disease and they are highly contagious),

hepatosplenomegaly, anemia, jaundice, hydrocephalus, lymphadenopathy, mucopurulent rhinitis, meningitis and mental retardation.

In septicemic forms, X-ray examination reveals metaphase bone destruction and periosteal reaction.

Signs of the disease may occur late, after the first 2 years of life such as: frontal bossing, micrognathia, the palate pointed arch, Hutchinson’s triad, saddle nose, rhagades, optic atrophy which leads to blindness.

Because most infants born with congenital disease are free of clinical symptoms at the time of birth, final diagnosis is determined by laboratory tests. Most used are serological tests and direct fluorescent antibody test.

Diagnosis based on neonatal serologic testing is complicated by the transplacental transfer of maternal Ig antibodies, which can cause a positive test in the absence of infection. However a neonatal titer > 4 times the maternal titer would not generally result from passive transfer and diagnosis is considered confirmed or highly probable. Therefore evaluating the new born baby must follow these steps: historical data on maternal infection, physical examination, hemoleucogram, treponemic and nontreponemic serological tests, cardio-pulmonary and long bones X-ray and liver tests. *T. pallidum* can be identified in skin lesions, umbilical cord, placenta or autopsy.

#### Material and method

Study was conducted in the Clinic of Neonatology on three cases of congenital systemic syphilis which have been hospitalized in the same period. In the following we will present significant data of the three cases.

#### CASE 1

Patient MD, male, aged 2 weeks, delivered at term by normal vaginal route with green slimy amniotic fluid, IA at 5’-8, Bw = 1980 g.(birth weigh)

The patient was admitted with extremely severe condition, intense jaundice, facial cyanosis, marked abdominal distention, hepatosplenomegaly, repeated crises of apnea and cyanosis requiring oxygen mask. On the left lower limbs he presented a erythematous macular rash with a diameter between 0.5 -2 cm and with the tendency of spreading to the rest of the body.

Serological tests confirmed the suspicion of congenital syphilis and haematochemical investigations emphasize thrombocytopenia with leukocytosis and hepatocytolitic syndrome (TGO=123U/L, TGP=112U/L). All cultures were sterile.

After about a week of treatment (antibiotic + penicillin, etamsylatum, calcium, vitamins, plasma, dexamethasonum, arginine, aspatofort) CRP and aminotransferase values begin to decline slowly, jaundice gradually decreases in intensity, new born had a significant weight gain and clinical status in evolution was satisfactory (w = 3300g).

### CASE 2

Patient K.S.- male new born baby, aged 1 day, weighing 2860 g, delivered at term by caesarean intervention, IA = 9, G1, P1.

On clinical examination the patient was noted to have a extremely severe condition, jaundice, perioronazal (facial) cyanosis, labored respiration with retraction of the intercostal muscles, ritmic heart sounds, pulse=130 beats / min. The abdomen was soft, it protruded during inspiration, the edge of the liver was palpable approximately 2.5 cm below the right costal margin. Anterior fontanelle (2/3 cm) was normotensive.

Biochemical investigations have confirmed the diagnosis: systemic congenital syphilis (leukocytosis, thrombocytopenia, CRP positive, VDRL and TPHA positive, increased LDH, hiperbilirubinemia).

Abdominal ultrasonography revealed increased liver volume, gallbladder with bold walls, normal biliary tract and normal spleen volume.

Treated with penicillin-10 days, in association with other antibiotics, and with ursodeoxycholic acid, calcium, vitamins and plasma, evolution is greatly improved with the exception of transaminase that increase progressively reaching a maximum at about a month of hospitalization, as follows: TGO = 535 U/l.

After this time, jaundice decreases in intensity and the liver size is progressively reduced.

### CASE 3

Patient V.I. , Female, aged 1 day, delivered at term by normal vaginal route, with green amniotic fluid,

G2, P1, gestational age-33/34 weeks, polihidramnios, Bw = 2440 g, IA=5 at 1 minute, 6 at 5 minutes.

It was admitted in the first days of life with severe condition, cyanosis, petechial elements on the legs and body, saddle nose. Balanced cardio-pulmonary, pulse 132 beats / min, SaO<sub>2</sub>-99%, abdominal distension caused by gas accumulation, the liver was palpable to the right iliac tank. Eruptive pustular papules on the abdomen and thorax.

Biochemical investigations (high leukocytosis, thrombocytopenia, hepatocytolitic syndrome: TGO = 210 U/L, TGP = 21 U/L, elevated inflammatory tests) and serological tests (VDRL, TPHA-positive) confirmed the diagnosis.

In evolution remain thrombocytopenia, leukocytosis, hepatocytolitic syndrome and intra-infectious anemia, beginning to improve after about 2 weeks from the onset.

### Results and discussion

Although the literature does not mention an increased incidence of septicemic congenital syphilis, our clinic has faced, in a short period of time, 3 cases with similar clinical and biological features and also with a relatively good evolution despite data quoted in the literature that emphasizes a bad prognosis : death in 40% of cases.

Among the pathognomonic features of the disease, common to our patients we mention:

- Intense jaundice, with elevated bilirubinemia. At first unconjugated bilirubina and in evolution, installed colestasis led both to increased conjugated bilirubin and also elevated levels of gGT and FA.
- Hepatomegaly accompanied by high levels of hepatic enzymes which after approximately one month start to decline under established treatment.
- Elevated values of inflammatory tests had been also present at all three patients and they slowly decreased under antibiotic therapy.

Treatment was conducted in accordance with the current protocols: penicillin for 10 days. It also has been done etiopathogenic treatment of concurrent infections, correction of acidobasic and hidroelectrolitic imbalances and correction of haematological disorders.

Evolution of the patient was initially serious: status toxicoseptic, needs for oxygen mask, positive serology, biological values considerably raised but then slowly becoming favorable, with improving of general status, jaundice remission and normalization of transaminases. The haematological parameters have also been corrected reaching values that corresponded to the patient's age.

### Conclusions

1. The most commonly form of disease encountered in medical practice are asymptomatic, incidentally discovered by serologic test for syphilis.
2. All the cases of manifest syphilis had a serious evolution requiring special care and treatment.

3. Although the status of patients was extremely severe, joining various pathology and highly modified laboratory indices, the evolution under treatment was favorable.

### References

1. Alford CA Jr, Cronin congenital and perinatal infections. in Avery GB editor, Neonatology, Pathophysiology and Management of the newborn, Philadelphia, JB Lippincott, 1987
2. Babcock D.S., Cranial ultrasonography of infants, Baltimore, Williams and Wilkins, 1981
3. Bale JF, Sato Y, Eisert D, Progressive postnatal subependymal necrosis in infant with congenital cytomegalovirus infections, *Pediatr Neurol*, 1986, 2, 367-370
4. Bains MK, Hosseini-Ardehali M. Palatal perforations: past and present. Two case reports and a literature review. *Br Dent J*. 2005;199:267–269. [PubMed]
5. Chawla W, Pandit PB, Nkrumach FK, Congenital syphilis in the newborn, *Arch Dis Child*, 1988, 63, 1393-1394
6. Dykes FD, Ahmann PA, Lazzara A, Cranial ultrasound in the detection of intracranial calcifications, *J Pediatr*, 1982, 100, 406-407
7. Platou RW, Hill AJ jr, Ingraham NR jr, Early congenital syphilis. Treatment of 252 patients with penicillin, *JAMA*, 1947, 133, 10
8. Lugo A, Sanchez S, Sanchez JL. Congenital syphilis. *Paediatr Dermatol*. 2006;23:121–123. doi: 10.1111/j.1525-1470.2006.00194.x.
9. Gurlek A, Alaybeyoglu NY, Demir CY, et al. The continuing scourge of congenital syphilis in 21<sup>st</sup> century: a case report. *Int J Paediatr Otorhinolaryngol*. 2005;69:1117–1121. doi: 10.1016/j.ijporl.2005.03.007.
10. Tikhonova L, Salakhov E, Southwick K, for the Congenital Syphilis Investigation Team. et al. Congenital syphilis in the Russian Federation: magnitude, determinants and consequences. *Sex Transm Infect*. 2003;79:106–110. doi: 10.1136/sti.79.2.106. [PubMed]
11. Li, Y.; Gonik, B. Is congenital syphilis really congenital syphilis. *Infect Dis Obstet Gynecol*. 2006. pp. 1–4. Article ID 81629.
12. Humphrey MD, Bradford DL. Congenital syphilis: still a reality in 1996. *Med J Australia*. 1996;165:382–385. [PubMed]

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### III. PEDIATRICS

## DOES THE BURNOUT SYNDROME EXIST AT THE PAEDIATRICIANS FROM ARAD? - A PILOT STUDY -

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#### Abstract

*Introduction:* Burnout is a psychological term that defines the long term exhaustion and lack of interest, usually in context of work. The paediatric field needs an evaluation of the burnout as it is a medical environment that involves a lot of stress.

*Objectives:* The aim of this study is to identify the levels of burnout of the paediatric consultants from Arad, using the Maslach Burnout Inventory (MBI), created by Cristina Maslach.

*Methods:* The study involved 13 paediatric consultants, who work in the Paediatric Emergency Unit and in the Paediatric Department of the Clinical Emergency Hospital, Arad. They were asked to submit their answers for the MBI self test. MBI considers burnout to be a multifactorial problem and identifies 3 subscales: emotional exhaustion, depersonalization and lack of personal accomplishment. Each subscale is measured by a different score. The higher the score on emotional exhaustion and depersonalization are, the higher the levels of burnout will be.

*Results:* The data was introduced in an Excel database and analyzed statistically. A high average was obtained for the emotional exhaustion subscale (30.23) and a moderate one for depersonalization and personal accomplishment subscales (10.37 and 35.84 respectively).

#### *Conclusions:*

1. There are high scores for emotional exhaustion, due to the large number of working hours and shifts.
2. There are moderate scores for depersonalization and personal accomplishment, which can be explained through the paediatric profile.
3. Further studies are required to obtain a more realistic view of the problem.

**Key words:** burnout, exhaustion, depersonalization, personal accomplishment, paediatrician.

#### Introduction

Burnout is a psychological term for the experience of long-term exhaustion and diminished interest (depersonalization), usually in the context of work. Burnout is often construed as the result of a period of spending too much effort at work while having too little recovery.

The paediatric field demands a quantification of the burnout as it is a medical environment where doctors experience a lot of stress.

#### Objectives

The aim of this study is to identify the emotional exhaustion, depersonalization and personal accomplishment scores of the paediatric consultants from Arad, using the Maslach Burnout Inventory (MBI), created by Cristina Maslach.

#### Method

13 paediatric doctors submitted their answers for the MBI self test. All of them work as paediatric consultants, having from 15 to 25 years of medical experience in this field, working from 46 to 72 hours per week and with an average of the monthly shifts ranging from 3 to 7.

4 of them work in the Emergency Paediatric Unit while the others work in the Paediatric Department of the Clinical Emergency Hospital, Arad, and have, also, private cabinets.

MBI identifies the most essential subscales:

- emotional exhaustion
- depersonalization
- lack of personal accomplishment.

Each subscale is measured by a different score.

A high degree of burnout is shown by high scores of emotional exhaustion and depersonalization

subscales, and by low scores of personal accomplishment (lack of) subscale.

An average degree of burnout is reflected in average scores on the three subscales.

A low degree is shown by low scores of emotional exhaustion and depersonalization and by high scores of personal accomplishment subscales. (1, 2, 3, 5).

The higher the score on emotional exhaustion and depersonalization, the higher the levels of burnout. Moreover, the lack of personal accomplishment scale measures in the opposite directions, the lower the scale, the higher the level of burnout (7).

The scores are analyzed according to the MBI standard, after more than 11.000 persons answered this questionnaire.

MBI	Low	Average	High
Emotional exhaustion	<16	17-26	>27
Depersonalization	<6	7-12	>13
Personal accomplishment	>39	38-32	<31

#### BURNOUT SELF-TEST by Cristina Maslach

It helps you look at the way you feel about your job and your experiences at work, so that you can get a feel for whether you might be at risk of burnout.

On a scale of 0 (never) to 6 (every day), state how often you feel that the following statements apply to you:

- 0- never
- 1- a few times a year or less
- 2- once a month or less
- 3- a few times a month
- 4- once a week
- 5- a few times a week
- 6- every day

#### **Emotional exhaustion score**

1. I feel emotionally drained from my work.
2. I feel used up at the end of the day.
3. I feel fatigued when I get up in the morning and I have to face another day on the job.
4. Working with people all day is a strain for me.
5. I feel burn-out from my work.
6. I feel frustrated from my job.
7. I feel I'm working too hard on my job.
8. Working with people directly puts too much stress on me.
9. I feel like I'm at the end of my rope.

#### **Depersonalization score**

10. I feel I treat some people in an impersonal manner.
11. I've become more callous toward people since I took this job.
12. I worry that this job is hardening me.
13. I don't really care what happens with some people I encounter at work.
14. I feel others at work blame me for some of their problems.

#### **Personal accomplishment score**

16. I can easily understand how people I work with feel about things.
17. I deal very effectively with problems people bring me at work.
18. I feel I'm making a difference in other people's lives through my work.
19. I feel very energetic.
20. I can easily create a relaxed atmosphere with people at work.
21. I feel exhilarated after working with people closely on my job.
22. I have accomplished many worthwhile things in this job.
23. In my work, I deal with emotional problems very calmly.

#### **Results and discussion**

The data was introduced in an Excel database and analyzed statistically. The following results were obtained for the paediatric doctors from Arad.

Table nr.1 - The obtained scores.

MBI	Low	Average	High
Emotional exhaustion	<16	17-26 <b>30.23</b>	>27
Depersonalization	<6	7-12 <b>10.37</b>	>13
Personal accomplishment	>39	32-38 <b>35.84</b>	<31

A high average can be noticed for the emotional exhaustion subscale (30.23), and a moderate for depersonalization and personal accomplishment subscales (10.37 and 35.84 respectively) (fig. 1).

The results were compared with the superior limit of the highest MBI scores admitted (fig. 2).

In comparison with a study performed on oncologists in Turkey, the results show higher scores for emotional exhaustion and depersonalization, but lower for personal accomplishment (fig.3).

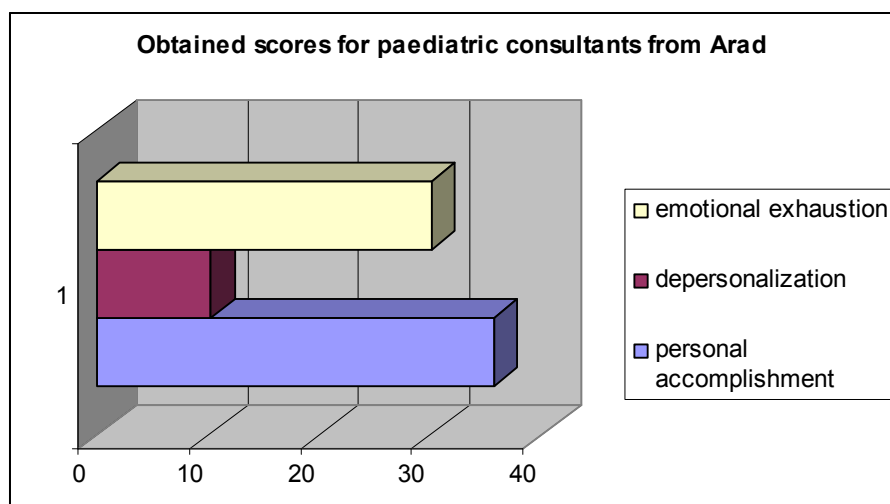


Figure 1. Obtained scores.

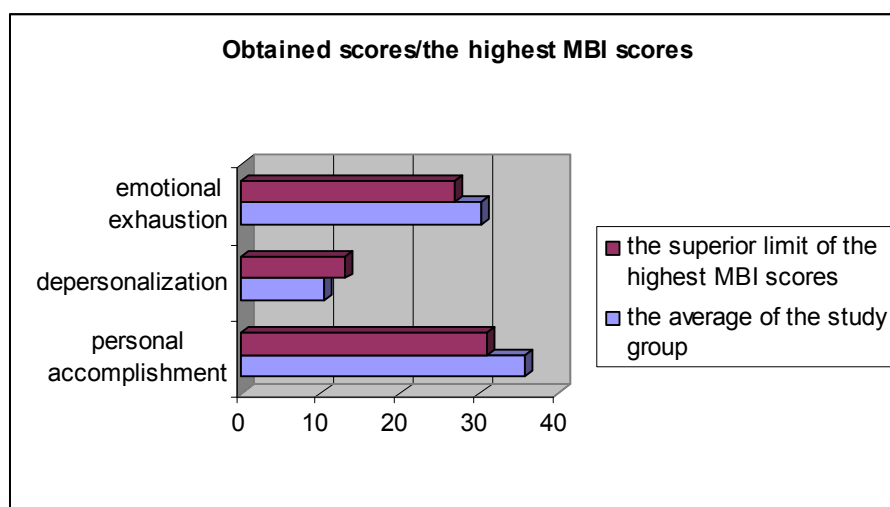


Figure 2. Comparing the obtained scores with the superior limit of the MBI scores.

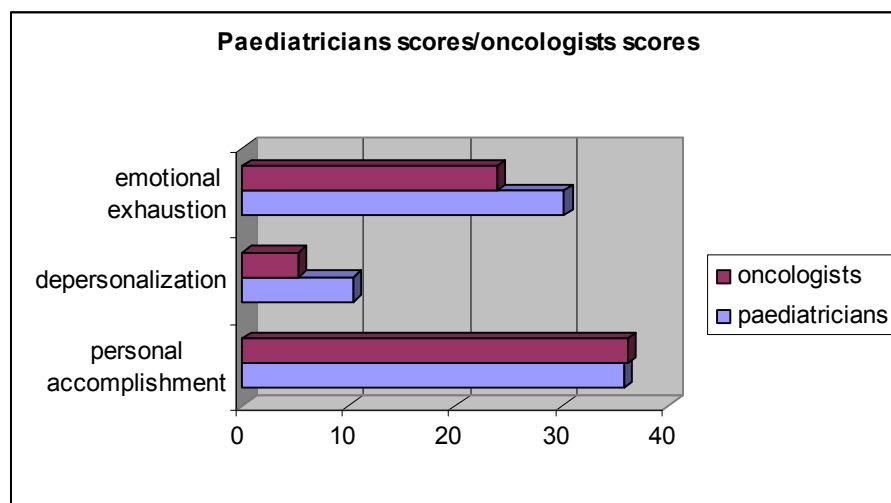


Figure 3. Comparing paediatricians scores / oncologists scores.

### Conclusions

1. There are high scores for emotional exhaustion, due to the large number of working hours and shifts.

2. There are moderate scores for depersonalization and personal accomplishment, which can be explained through the paediatric profile.

3. Further studies are required to obtain a more realistic view of the problem.

### References

1. Armstrong J, Lederberg M, Holland J. Fellows' forum: A workshop on the stresses of being an oncologist. *J Cancer Educ.* 2004;19:88-90.
2. Armstrong J, Holland J. Surviving the stresses of clinical oncology by improving communication. *Oncology (Williston Park).* 2004; 18:363-368, 373-375.
3. Holland JM, Neimeyer RA. Reducing the risk of burnout in end-of-life care settings: The role of daily spiritual experiences and training. *Palliat Support Care.* 2005; 3:173-181.
4. Holland JC. Management of grief and loss: Medicine's obligation and challenge. *J Am Med Womens Assoc.* 2002; 57:95-96.
5. Kash KM, Holland JC, Breitbart W, et al. Stress and burnout in oncology. *Oncology (Williston Park).* 2000; 14:1621-1633, 1633-1634, 1636-1637.
6. Maslach C., Schaufeli W - Historical and conceptual development of burnout.
7. Professional burnout: Recent developments in theory and research. Ed. by Schaufeli W., Maslach C. Marek T. New-York, Taylor and Francis 1993.
8. \*\*\*31st Congress of the European Society of Medical Oncology (ESMO). "Burnout is an important issue for oncology employees," lead author Senem Dubova, MD, from Ege University Medical School, in Bornova-Izmir, Turkey.

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## THE MARKERS FOR IMMUNO-GENETIC SUSCEPTIBILITY IN CHILDHOOD DIABETES

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### Abstract

Diabetes mellitus (DM) is a heterogenous syndrome characterized by a complex disturbance of the energetic metabolism, which affects the metabolism of both carbohydrates, lipids and proteins and also the other metabolisms. These disturbances result from an insulin secreting defect (the decrease of  $\beta$  cell mass / function), associated sometimes with a degree of peripheral insulin resistance. Prediction of type 1 DM, meaning the appreciation of the risk to develop the disease, raises a great theoretical and practical interest. This is based on the acceptance of the autoimmune pathogeny in most of the cases (DM type 1A) and the understanding of the progressive, stadial evolution of the  $\beta$ -cell destructive process (1). The prediction strategies are using the genetic, immunologic and metabolic markers which define the risk of the patients to develop type 1 DM.

**Key words:** susceptibility, childhood, diabetes

### Introduction

From a genetic point of view, diabetes is a complex, poligenic disease, involving numerous susceptibility genes and some protective genes, all with incomplete penetrance, reciprocally conditioning each other.

Actually, is unanimously accepted that the short prediagnosis period in type 1 DM is the top of a huge iceberg, just partially explored by the modern immunogenetic studies. These studies prefigure a stadial evolution of a variable duration (2) (months, years).

In this period of time the disease is ongoing through 6 evolutive phases:

- genetic susceptibility (3),
- precipitating event (intervention of the trigger factors),
- overt immunologic abnormalities (autoantibodies: GAD, ICA),
- progressive loss of insulin release,
- overt diabetes,
- complete islet beta cell destruction.

The genetic markers used in association with the family history shows that the risk of type 1 DM is (4):

- 1/5.000 in cases without susceptibility alleles or family history
- 1/4 if two risk alleles exist and a positive family history.

### A. Genetic markers:

In the last years numerous genes were studied (chromosomal regions); of these, two regions are mostly involved in the genetic susceptibility for type 1 DM:

- HLA region on short arm of chromosome 6 (6p21.3) noted IDDM1 and

- insulin gene region on the short arm of chromosome 11 (11p15), noted IDDM2. IDDM1 is responsible for almost 50% of the genetic susceptibility, while IDDM2 for 10-15% (5, 6, 7). Beside these two regions, genom –wide scan studies identified at least 18 chromosomal regions (noted IDDM3, IDDM4 etc.) associated with type 1 DM. For most of these regions, the susceptibility genes have not been precisely identified yet, the mechanism of their involvement in the pathogeny of the disease still remains to be clarified (8) (table 1).

The most known “diabetogenic genes” are those belonging to HLA system from the MHC region of the short arm of chromosome 6 (6p21.3) – with a major role in the immune response of the body (Fig. 1).

Presently is unanimously accepted that type 1 DM in the child is associated with:

- *DRB1\*04-DQA1\*0301-DQB1\*0302* allele and
- *DRB1\*03-DQA1\*0501-DQB1\*0201* and the decreased frequency should explain the low incidence of DM in some countries like Romania (9).

More than 90% of the diabetic patients with type 1 DM have predisposing alleles type DR3-DR4 – comparatively with 40-50% in the general population. The concomitant presence of DR3-DR4 in one patient increases the risk; actually this association is encountered in 30-50% of type 1 DM patients (compared to 1-6% in the general population).

Table I. Genome screens T1DM.

<b>IDDM1</b>	<b>6p21</b>	IDDM13	2q34-q35
<b>IDDM2</b>	<b>11p15</b>	IDDM15	6q21
IDDM3	15q26	IDDM17	10q25
IDDM4	11q13	IDDM18	5q31-q35 (IL2)
IDDM5	6q25-q27		1q42-qter
IDDM6	18q21		8q24
IDDM7	2q31	VDR, INF $\alpha$	12q12-qter
IDDM8	6q27-qter		16p11-p16
IDDM9	3q21-q25		16q22-q24
IDDM10	10p11-q11		17q24-qter
IDDM11	14q24-q31	TGF $\beta$ 1	19p13-q13
IDDM12	2q33 (CTLA4)		Xp13-p11

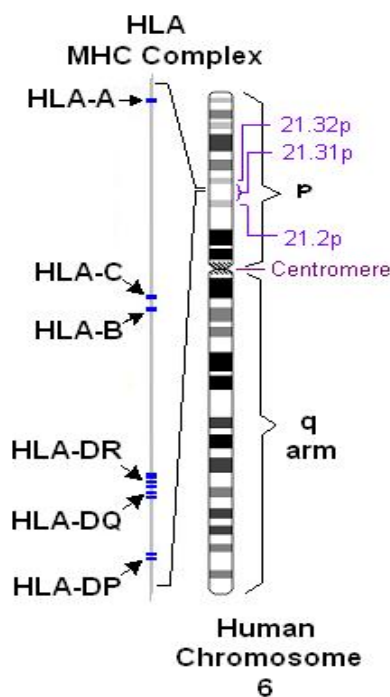


Fig. 1 - Schematic representation of HLA, projected on the short arm of chromosome 6 (Richard G Phelps and Andrew J Rees).

Numerous studies, confirm that the HLA DQ molecules have a primordial role in the predisposition to type 1 DM. DQA1\*0301-DQB1\*0302 is associated with an increased susceptibility for type 1DM in most of the populational group studied (10).

The study of the HLA-DP alleles didn't offer any certain proof concerning their involvement in the predisposition for type 1 DM.

Some HLA alleles confer protection for the occurrence of diabetes (11); we mean especially the following HLA molecules:

- DQ6 (DQB1\*0602 si DQB1\*0603),
- DQ7 (DQB1\*0301/0304),
- DRB1\*1401
- DQA1\*0201

The protection conferred is not absolute, however, less than 1% of type1 DM patients have these alleles.

These protective alleles seem to have dominance upon the susceptibility alleles.

The second region proved to be associated with type1DM is the region for insulin gene on chromosome 11 - 11p15 (IDDM2). We talk about polymorphisms from a variable zone (VNTR – Variable Number of Tandem Repeats) situated in region 5' reported to the insulin gene promotor which influences the regulatory mechanism of insulin gene transcription.

At this level 3 classes of alleles may exist. Class I haplotype are associated with Type 1 DM while those from class III confer protection (12).

The other locuses proved to be involved in the predisposition for type 1 DM, include (Fig. 2):

- The lymphoid-specific phosphatase (LYP) encoded by *PTPN22* is involved in preventing spontaneous T-

cell activation by dephosphorylating and inactivating T-cell receptor-associated Csk kinase (13). An arginine-to-triptophan substitution at codon 620 of PTPN22 was considerably reported to be associated with type 1 DM as well as other autoimmune diseases, such as rheumatoid arthritis, systemic lupus erythematosus (SLE) and Grave's disease. Genotyping of PTPN22 revealed the following alleles:

- the homozygous genotype for the T allele and the heterozygous genotype C/T is associated with an increased risk for developing type 1 diabetes
- the C/C homozygous genotype is protective against type 1 diabetes.

- The presence of the heterozygous genotype C1858T in patients with type 1 DM, increases the risk to associate other autoimmune disturbances (14).

- gene CTLA - 4 (Cytotoxic T Lymphocyte antigen) on chromosome 2q33 – corresponding to IDDM12

- gene for  $\alpha$  chain of the interleukine 2 receptor (IL2RA/CD25) on chromosome10p15.

Possibly implicated in the predisposition for type1 DM are some polymorphisms from gene ICAM -1 (Intercellular Cell Adhesion Molecule I) and the gene for Vitamine D Receptor (VDR - Vitamin D Receptor) (15).

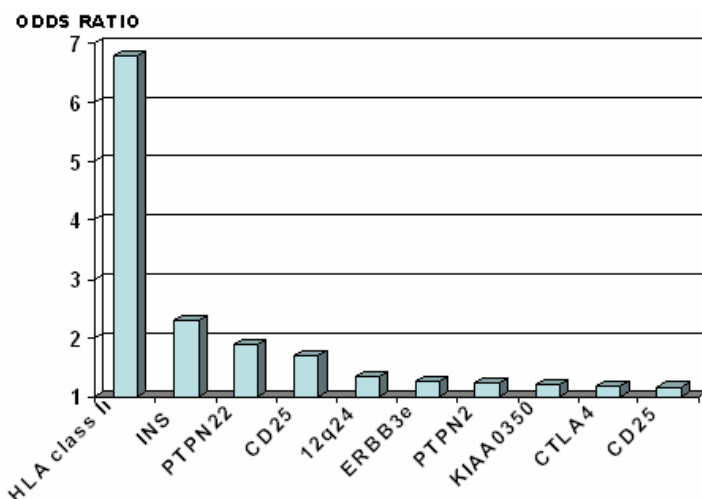


Fig. 2 - Summary of subset of confirmed loci from whole genome screens associated with type 1A diabetes (Modified from Todd et al. Nature Genetics, June 6, 2007).

### B. The markers for autoimmunity

The autoimmune destructive process of the  $\beta$  cells is a chronic process, with variable duration and evolution velocity, individualised.

Within this period, the immunologic markers might be evidenced, in the serum, including: islet cell antibodies (ICA), insulin autoantibodies (IAA), GAD65 antibodies etc.

Detection of antibodies in the serum has an important diagnostic significance, so, the high ICA is predictable for type1 DM, before the occurrence of the disease, fact that has been proved in relatives of the diabetic patients. 8-10% of these, with an increased titre of these antibodies progress towards DM within one year.

The presence of markers in association, in the serum of some subjects, both in the general population and in some belonging to subgroups with increased risk for type 1 diabetes mellitus (type 1 DM), increases the probability for developing this disease(16).

- *ICA (islet cell antibodies)* was first described as being associated with type 1 DM. ICA titre is expressed as JDF conventional units (Juvenile Diabetes Foundation).
- ICA are present in serum in 70-80% of the diabetic patients even since onset (17). Although technically difficult to perform, they remain the most sensitive marker for the prediction of the risk to develop DM, titres above 20 U JDF showing a probability of 30-40% to develop the disease in the next 5 years.
- *IAA (Insulin Autoantibodies)* are present in serum since the onset (meaning before the initiation of insulin therapy) in 50-70% of the subjects, more frequent in children than in adults. Their presence represents the proof of an ongoing  $\beta$ -cell destructive process and represents an important marker for the detection of the subjects at risk to develop type 1 DM (18).

- One of the most important autoantigenes that induces production of antibodies associated with DM is *GAD* (*Glutamic Acid Decarboxylase*) that is present in the  $\beta$  cells but also in CNS and the testicular tissue (19). The antibodies against the 65 kD peptide of *GAD* (*GADA*) are present in 70-80% of the patients type 1 DM and occur even since the prediagnostic period.
- Recently, a new family of  $\beta$  cell autoantigenes has been identified, the family of proteins *PTP* – Protein Tyrosine Phosphatase. The antibodies against a 40 kDa fragment of this protein also called *ICA512* or *IA – 2*, occur in 60-70% of the subjects with type 1 DM at the onset (20).
- There are also other cytoplasmatic  $\beta$  cell antigenes responsible for the autoimmunity in DM. Of these, the most studied were: *ICA69*, *Carboxipeptidase H*, *Ganglioside GM2-1*, *Imogen 38*, *Glima 38*, *Peripherina*, *Hsp 60* (*Heat Shock Protein 60*) etc.(21).

There are cases when healthy individuals are found with significant titres of diabetogenic antibodies that may persist years before the occurrence of clinical DM or even without developing the disease ( $\approx$  5% of the general population).

For the moment, prediction (expressed as the percentage probability of the risk to develop type 1

DM) can't offer an absolute precision (22). The association of the immunological tests and the genetic typing, more and more accesible, even in the newborn, because of the development of rapid, automatic and cheaper techniques, increases the accuracy of prediction comparatively to isolate evaluation of the humoral immunity.

In Romania there are just few studies concerning different aspects of the type 1 DM in children, but none of them prospective aiming for the evaluation of prediction and prevention in type 1 DM in the infantile population and the causal relationship *genetic predisposition – immune status – environment factors* (especially food).

There are still many questions to be answered, for instance if:

- the presence of the markers for cellular autoimmunity increase the risk for type 1 DM also in the general population,
- all subjects with autoimmune markers will develop type 1 DM,
- the detection of autoantibodies correlated with the reduction of first phase of the insulinic response increases the possibility of the disease to occur,
- the associations between antibodies increase the risk.

## References

1. Maclaren NK, Lan MS, Schatz D, Malone J, Notkins AL, Krischer J. 2003. Multiple autoantibodies as predictors of Type 1 diabetes in a general population. *Diabetologia*. 46:873-4.
2. Atkinson MA., Eisenbarth GS – Type 1 diabetes: new perspective on the disease pathogenesis and treatment, *The Lancet*, 2001, 358: 221-229.
3. Park Y., Eisenbarth GS – The natural history of autoimmunity in type 1 Diabetes mellitus. Disease, Prediction and Prevention, in Le Roith D., Taylor S.L., Olefsky J.M., - *Diabetes mellitus – a fundamental and clinical text*, 2nd Ed. Lippincot Williams and Wilkins, Philadelphia/Baltimore/ New York / London, 2000.
4. Pinhas-Hamiel O, Zeitler P. The global spread of type 1 diabetes mellitus in children and adolescents. *J Pediatr* 2005; 146: 693–700.
5. Klein J, Sato A. The HLA system. First of two parts. *N Engl J Med* 2000; 343(10):702-709.
6. Kwon OJ, Brautbar C, Weintrob N, Sprecher E, Saphirman C, Bloch K et al. Immunogenetics of HLA class II in Israeli Ashkenazi Jewish, Israeli non-Ashkenazi Jewish, and in Israeli Arab IDDM patients. *Hum Immunol* 2001; 62(1):85-91.
7. Undlien DE, Lie BA, Thorsby E. HLA complex genes in type 1 diabetes and other autoimmune diseases. Which genes are involved? *Trends Genet* 2001; 17(2):93-100
8. Sabbah E., Savola K., Ebeling T., Kulmala P., Vahasalo P., Ilonen J., Salmela P.I., Knip M. – Genetic, autoimmune and clinical characteristics of childhood and adult onset type 1 diabetes. *Diabetes Care* 2000, 23, 1326-1332
9. Ionescu Tirgoviste C., Guja C., Herr M., Cucca E., Welsh K., Bunce M., Marshall S., Todd J.A.- Lowfrequency of HLA DRB1 03-DQB1 03 and DQB1 0302 haplotypes in Romania is consistent with the country's low incidence of Type 1 diabetes. *Diabetologia*, 2001, 44, suppl. 3, B60-B66
10. Pugliese A, Kawasaki E, Zeller M, Yu L, Babu S, Solimena M et al. Sequence analysis of the diabetes-protective human leukocyte antigen-DQB1\*0602 allele in unaffected, islet cell

- antibody-positive first degree relatives and in rare patients with type 1 diabetes. *J Clin Endocrinol Metab* 1999; 84(5):1722-1728.
11. Redondo MJ, Kawasaki E, Mulgrew CL, Noble JA, Erlich HA, Freed BM et al. DR and DQ associated protection from type 1 diabetes: comparison of DRB1\*1401 and DQA1\*0102-DQB1\*0602. *J Clin Endocrinol Metab* 2000; 85(10):3793-3797
  12. Walter M, Albert E, Conrad M, Keller E, Hummell M, Todd J. A., Bonifacio E: IDDM2/insulin VNTR modifies risk conferred by IDDM1/HLA for development of type 1 diabetes and associated autoimmunity. *Diabetologia* ISSN 0012-186X, 2003, vol. 46, n°5, pp. 712-720.
  13. Meloni G.F., P. Lucarelli, M. Pellechia, et al. 2004. A functional variant of lymphoid tyrosine phosphatase is associated with type 1 diabetes.
  14. Kawasaki E., T. Awata, H. Ikegami, et. Al. 2006. Systematic search for single nucleotide polymorphism in a lymphoid tyrosine phosphatase (PTPN22) gene.
  15. Todd JA, Walker NM, Cooper JD, Smyth DJ, Downes K, Plagnol V et al. Robust associations of four new chromosome regions from genome-wide analyses of type 1 diabetes. *Nat Genet* 2007; 39(7):857-864
  16. Park Y., Eisenbarth GS – The natural history of autoimmunity in type 1 Diabetes mellitus. *Disease, Prediction and Prevention*, in Le Roith D., Taylor S.L., Olefsky J.M., - Diabetes mellitus – a fundamental and clinical text, 2nd Ed. Lippincott Williams and Wilkins, Philadelphia/Baltimore/ New York / London, 2000
  17. Vesa Eskola, Paula Vähäsalo, Hans K. Åkerblom, Mikael Knip, The Finnish ENDIT Study Group. Increased Frequency of Islet Cell Antibodies in Unaffected Brothers of Children with Type 1 Diabetes. *Hormone research*. Vol. 59. No.4.2003
  18. Schlosser M., Koczwara K., Kenk H., Strebelow M, Ziegler A.-G, Bonifacio E. In insulin-autoantibody-positive children from the general population, antibody affinity identifies those at high and low risk. *Diabetologia* 2005, vol. 48, no9, pp. 1830-1832
  19. Lindholm E, Hallengren B, Agardh CD. Gender differences in GAD antibody-positive diabetes mellitus in relation to age at onset, C-peptide and other endocrine autoimmune diseases. *Diabetes Metab Res Rev*. 2004 Mar-Apr;20 (2):158-64.
  20. Steffen G, Blanchetot C, Schepens J, Albet S, Lammers S: Multimerization of the Protein-tyrosine Phosphatase (PTP)-like Insulin-dependent Diabetes Mellitus Autoantigens IA-2 and IA-2  $\beta$  with Receptor PTPs (RPTPs). *Biol. Chem.*, Vol. 277, Issue 50, 48139-48145, December 13, 2002
  21. Martin S.: Islet cell autoantigen 69 antibodies in IDDM. *Diabetologia* 2004. pp 747.
  22. Samuelsson U, Sundkvist G, Borg H, Fernlund P, Ludvigsson J. 2001. Islet autoantibodies in the prediction of diabetes in school children. *Diabetes Res Clin Pract*. 51:51-7.

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## ACCIDENTAL ACUTE INTOXICATION WITH DENTOCALMIN IN CHILDREN – A SEVERE FORM CASE PRESENTATION

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### Abstract

The authors present the case of a child, aged one year and seven months, who was admitted to the Emergency Unit of the County Emergency Hospital Craiova, because of an accidental ingestion of Dentocalmin, with a cardiorespiratory stop. After 25 minutes of cardiorespiratory resuscitation, normal sinus rhythm is re-established; he is artificially ventilated; 48 hours after his hospitalization, he started to breath spontaneously. He is discharged after 27 days of hospitalization, with serious neuropsychic sequels.

**Key words:** Dentocalmin, acute intoxication, child.

### Introduction

Our country annually registers about 16,000 cases of intoxication in children. Intoxications with drugs, caustic household products, industrial products (antifreeze solution, petrol, and gas), and alcohol are dominant.

The highest frequency of the accidental acute intoxications is met in the age group 0-7 years (5); among them, more than 50% are caused by drugs (2).

Dentocalmin is a dental product, under the form of solution with external use. Regarding its pharmacotherapeutic action, it is a local anesthetic, an analgesic and an anti-inflammatory product. It is found under the form of bottles of 10 ml which contain: Lidocaine 2 g, Menthol 2 g and Phenol 2g (3).

### Case presentation

The child S.G.E. (F.O. 51828/ 2007, 2<sup>nd</sup> Pediatrics Clinic, Emergency County Hospital Craiova), male, aged 1 year and 7 months, Weight= 14 Kg is admitted in the Emergency Unit of the County Hospital Craiova, with a cardio-respiratory stop, on October, 20, 2007.

The anamnesis reveals that the child's state suddenly worsened, presenting – when in full health – a sleeping state followed by coma in approximately 10

minutes, after the mother administered him, by mistake, a few ml (5-6) of Dentocalmin, without using a dropping glass. The mother mistook the bottle of Dentocalmin for the bottle of Vigantol Oil (with a dropping glass), from which the child used to receive a daily 2 ml dosage. He was transported by ambulance for hospitalization.

*Heredocolateral antecedents* – young, healthy genitors; mother with higher education.

*Physiologic personal antecedents.* Single child, on term and normal delivery, W<sub>B</sub>= 3,300 G, Apgar score 10, artificial feeding when born with Milumil, Lactovit, correctly diversified when 4 months, vaccinated according to W.H.O. vaccination scheme; rickets prophylaxis with Vigantol Oil 2 drops /day; normal physical and psychomotor development.

*Pathologic personal antecedents:* 2 hospitalizations: the first when 7 months and the second when 1 year and 6 months for acute bronchiolitis.

*Life conditions:* an apartment in urban area, in a block of flats, 2 rooms, 3 persons.

*When presented in the Emergency Unit,* the child was in an extremely bad state, abolished conscience, marmorated teguments, cold and cyanotic extremities, absent peripheral and central pulse, absent spontaneous breath, mydriatic, non-reactive pupils.

After 25 minutes of cardio-respiratory resuscitation (O.T.I. with assisted ventilation, external cardiac massage, adrenaline i.v., Na bicarbonate i.v., E.V.P. with physiologic serum) the heart activity is re-established. The stages E.K.G. – initial asystoly – subsequent electromechanic dissociation and ventricular fibrillation; after 25 minutes, the child had a synusal rhythm, C.F.= 132 b/min [fig.1]. The state of the child remained severe, with an abolished conscience and mechanically ventilated.

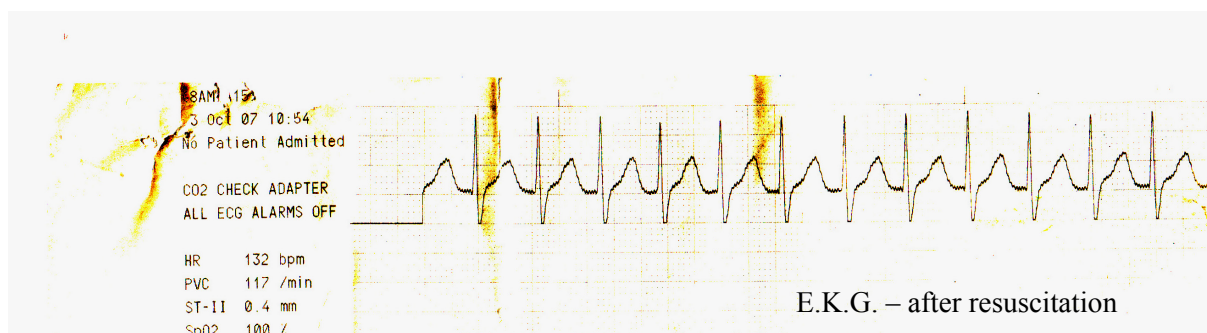
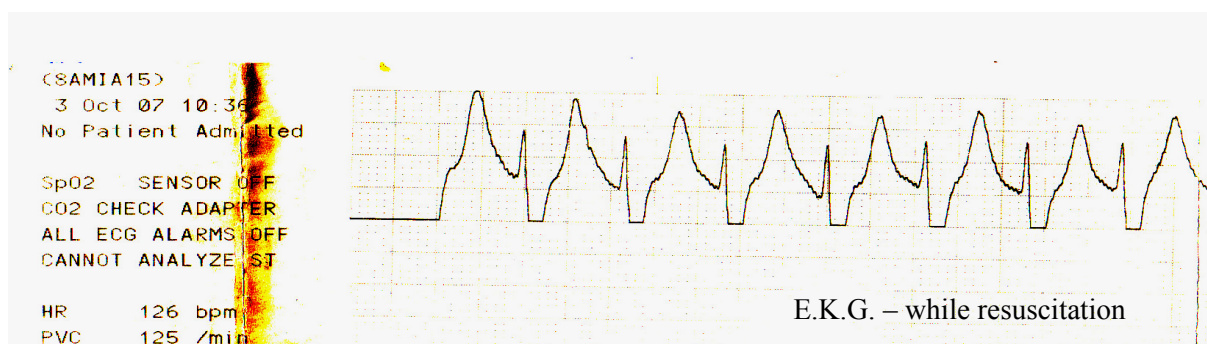
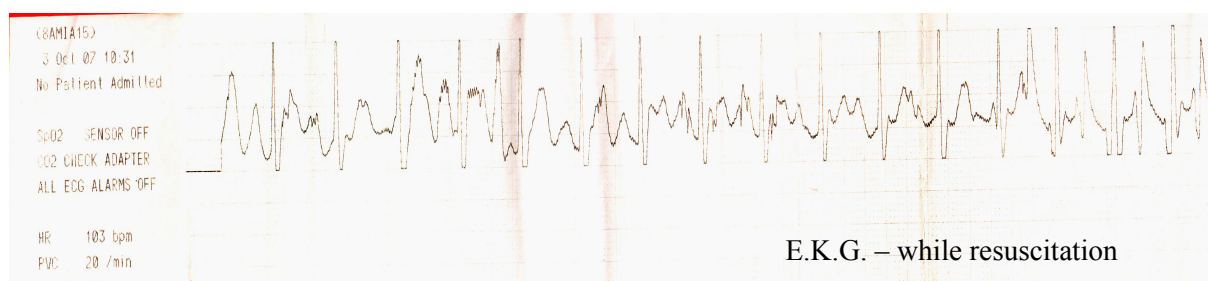
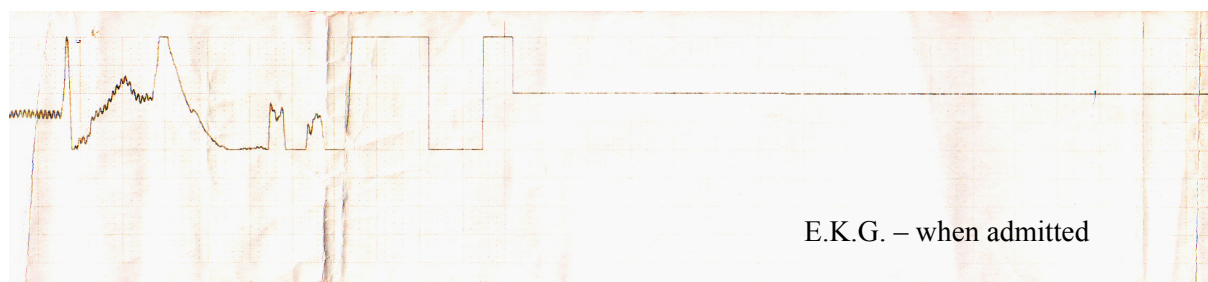
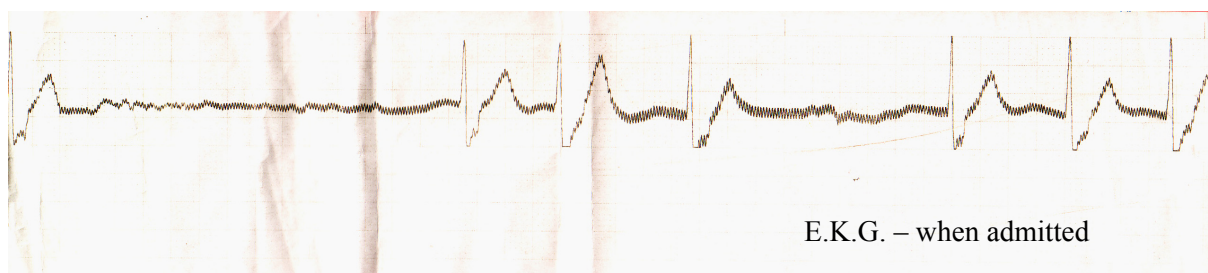


Fig. 1. E.K.G stages – initial asystoly – subsequent electromechanic dissociation and ventricular fibrillation; after 25 minutes, the child had a synusal rhythm, C.F.= 132 b/min.

He is admitted to the Intensive Care Unit where the intensive treatment goes on: assisted ventilation, gastric washing, and intravenous treatment with Manitol, Furosemid, Dexametazone, Piracetam, vitamin B<sub>1</sub>, B<sub>6</sub>, C, Tazocin.

*Investigations when admitted (20 X):*

Hemogram: Hb= 11.8 g%, T= 240,000/mm<sup>3</sup>, L= 7,500/mm<sup>3</sup>, N= 2%, S= 63%, E= 5%, Ly= 26%, M= 4%;

Glicemy = 85 mg%;

Micro Astrup: pO<sub>2</sub> 159.8 mmHg; pCO<sub>2</sub> 36.6 mmHg; pH 7.24; SO<sub>2</sub> 98.5%; BE -10.9 mmol/l; BEcf -11.8 mmol/l; CHCO<sub>3</sub> st 15.9 mmol/l; p<sub>50</sub> 26.7 mmHg; Ct O<sub>2</sub> 18.8%; CHCO<sub>3</sub> 15.5 mmol/l; Ct CO<sub>2</sub> (B) 14.3 mmol/l; SO<sub>2</sub> (C) 98.9%;

Sanguine ionogram: Na<sup>+</sup> = 143 mmol/l, K<sup>+</sup> = 3.7 mmol/l, Cl<sup>-</sup> = 110 mmol/l, Ca<sup>++</sup> = 1.033 mmol/l.

2<sup>nd</sup> Day of hospitalization – the child breathes spontaneously and efficient (sat O<sub>2</sub> 99%), he presents rhythmic cardiac sounds, C.F.= 127 b/min, slender abdomen, present diuresis; however, his future evolution is questionable, since the child alternates between periods of somnolence and agitation, opisthotonus, horizontal nistagmus, convulsions. He is fed through a nasogastric tube.

After two weeks of hospitalization, he starts his feeding per os, but the psychomotor acquisitions are lost: he does not speak, he does not walk, he reacts only to strongly painful stimuli, he presents a cerebral cry.

*Investigations performed while hospitalization:*

29 X: Hb= 8.4 g%, T= 230,000/mm<sup>3</sup>, L= 6,800/mm<sup>3</sup>, N= 2%, S= 68%, E= 3%, Ly= 10%, M= 7%, anisocytosis, poikilocytosis;

6 XI: Hb= 11.8 g%, T= 240,000/mm<sup>3</sup>, L= 8,500/mm<sup>3</sup>, N= 2%, S= 63%, E= 5%, Ly= 26%, M= 4%;

24 X: glicemy= 75 mg%;

Sanguine ionogram 24 X: Na<sup>+</sup> = 143.4 mEq/l, K<sup>+</sup> = 4.6 mEq/l;

E.g. F.O.: A.O. – normal aspect

Pulmonary X-ray: no pleural-pulmonary changes, heart in normal limits.

Skull C.T. (26 X): normal limits C.T. aspect, for native C.T. and postcontrast.

E.g. pediatric neuropsychiatry (30 X): vegetative status, reacting to strongly painful stimuli → 15 XI: Spastic tetraparesis, psychic and motor regression.

He continued to receive a treatment consisting of Diazepam, Fenobarbital, Cerebrolizin, Piracetam, Dexametazone, physiokinetotherapy.

After 27 days of hospitalization, he is discharged, balanced from the cardiac and respiratory point of view, with feeding per os, a good digestive tolerance, presenting serious neuropsychic sequels and with the following recommendations:

- to carry on medical recovery - physiokinetotherapy;

- to receive a drug-based treatment with Encephabol, Piracetam, and Vitamin B.

A year after the child was discharged (October 2008), he was admitted again in the Clinic (F.O.47113/2008) for a respiratory disease. In this period, the child followed a recovery treatment - physiokinetotherapy and drug-based treatment and progress was registered: he walks if supported, utters some words, and interacts with the surrounding persons.

### Discussions

We presented this case because of the severe intoxication produced by Dentocalmin, an apparently harmless drug.

The components of Dentocalmin have the following effects:

Phenol (phenic acid, carbolic acid), in low concentrations (0.2 -1%), has a bacteriostatic effect, and when 3-5% it has a bactericidal action, due to the protein precipitation (4).

Locally applied, in concentration of 2%, the phenol is a local anesthetic, decreasing the excitability of peripheral nerves. In solutions of 5%, it is irritant and inflammable, its great power of penetration determining deep lesions, which require a long period of healing (6).

Menthol (Mentholum) – is obtained from the mint volatile oil (natural menthol) or through synthesis (synthetic menthol). It is antipruriginous and aromatizing (6).

Lidocaine (Xiline) – is a local anesthetic with an amidic structure, which is active in all types of local anesthesia, including local anesthesia (under the form of solutions of 2-4% or Lidocaine ointment 5%). Intravenously administered or after absorption at the administrated place, lidocaine causes systemic effects: sedative, analgesics, anticonvulsive, antiarrhythmics (3).

In case of overdose (the maximum admitted dose is 4 mg/Kgc/day) or fast intravenous administration, lidocaine can cause convulsions, tachycardia, lipotimy, high blood pressure, followed by coma, bradycardia, hypotension, respiratory depression. For our hospitalized child, the maximum dose of Lidocaine admitted during 24 hours was 56 mg (3). Taking into account that he received about half of the Dentocalmin bottle content, it results that he

received approximately one dose of 1,000 mg of lidocaine, which represents twenty times more than the maximum admitted dose for 24 hours; hence the gravity of intoxication (the cardiac and respiratory stop, the coma).

Between January, 1, 2005 and April, 1, 2008, at the Antitoxic Centre of the Emergency Clinical Hospital for Children “Grigore Alexandrescu”, Bucharest, there were admitted and reported 22 cases of acute intoxication with Dentocalmin. The evolution was as follows: 15 cases with full recovery, 2 deaths, and 3 cases with serious neuropsychic sequels (7).

Following the requests of the Toxicology Department of the Emergency Clinical Hospital for Children “Grigore Alexandrescu”, Bucharest (Professor Coriolan Ulmeanu), the Drug National Agency decided, in January 2008, an urgent withdrawal of all the Dentocalmin which was found in the communitary pharmacies. This happened because Dentocalmin had to be given according to market authorization and only within hospitals (in dental offices, respectively) (1).

We have to mention that, subsequently - in September, 2008 – an infant aged 4 months was

admitted in the clinic, with a Dentocalmin intoxication; he was administered by his mother, again by mistake, Dentocalmin instead of Vigantol Oil. This time, there was registered a favorable evolution.

For the presented case, we pointed out:

- the recovery potential of the heart and lungs in children – initially healthy, which after a long period of resuscitation recovered their normal activity;
- the gravity of the child’s intoxication, requiring an initial treatment of cardio-respiratory resuscitation, followed by medical services in the Intensive Care unit and Pediatric Clinic;
- a long period of hospitalization (27 days);
- serious neuropsychic sequels and lasting treatment for neuropsychic recovery;
- mother’s psychic impact – a feeling of guilt – the one who mistook the bottle of Vigantol Oil for the bottle of Dentocalmin

## References

1. Agenția Națională a Medicamentului – Nomenclatorul Medicamentelor de uz uman, 2005, Ed. Medicală București,
2. Bates Nicola, Edwards N., Roper J., Volans S., Paediatric Toxicology, 2006, Stockolm Press, ISBN 0-333-60951-4
3. Cârlig V., Farmacologia, Ed. Aius, Craiova, 1999, Vol. I, 321-322
4. Farmacopeea Română, Ed. a X-a, Ed. Medicală București, 1998
5. Goldfrank’s Toxicologic Emergencies , Ed. a 8-a , 2006
6. Manolescu E., Farmacologie, Ed. Didactică și Pedagogică București, 1984, Ed. a II-a, p. 438
7. Ninescu G.V., Petran M., Stanca S., Ulmeanu C., Intoxicația acută cu Dentocalmin – intoxicație nouă cu un produs vechi, Al IX-lea Congres Internațional de Farmacologie, Terapeutică și Toxicologie Clinică, 11-14 iunie 2008, Sibiu, Volum de Rezumate, Editura Alma Mater, 2008.

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## STAPHYLOCOCCAL IMPETIGO WITH SEVERE SEPSIS - CASE REPORT

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### Abstract

*Staphylococcus aureus* is the most common cause of pyogenic infection of the skin, little infants being extremely susceptible. Staphylococci may enter in the blood with subsequent involvement of the organs. The development of staphylococcal disease is related to resistance of the host and to virulence of the organisms bacteria. We present the case of a five months old female infant hospitalized in our clinic for disseminated skin lesions, diarrhea with dehydration and sepsis with *Staphylococcus aureus*.

**Key words:** *Staphylococcus aureus*, impetigo, sepsis

### Introduction

*Staphylococcus aureus* causes a wide variety of infections.<sup>1,2,3</sup> It is the most common cause of pyogenic infection of the skin.<sup>4,5,6</sup> Little infants are extremely susceptible to staphylococci. A history of poor hygiene and crowded living situations are common.<sup>6</sup> Autoinfection is common and minor infection may be the source of disseminations. The intact skin and mucous membranes serve as barriers to invasion by staphylococci.<sup>4,7</sup> *Staphylococcus aureus* enters through damaged skin and is transmitted through direct contact.<sup>6</sup> Staphylococci may also enter the blood with subsequent involvement of the organs.<sup>1</sup> Impetigo is most common in children. Most children are younger than 2 years. Prognosis may be influenced by numerous host factors, including nutrition, immunologic competence and the presence of other debilitating diseases.<sup>4</sup>

### Case report

We present a five months old female infant admitted in the First Pediatric Clinic in „Louis Turcanu” Children’s Emergency Hospital Timisoara for high

fever, perioral, nasal, cervical, occipital and on the back crusting, diarrhoea, significant dehydration. She is the second child of a young healthy couple from a town area in Timis county. The pregnancy was followed up, pathological, with imminence of miscarriage in the second trimester. The child was born in term, with a weight of 2260 g and Apgar score 9. She was breastfed for 1 month and then with cow milk in excessive dilution with tea, with no sugar. She was precociously fed with potato mash from 3 months of age. Child’s hygiene was precarious.

### *The history of illness*

The patient presented diaper dermatitis for 2 months. The infant had high fever 39-40°C for five days, productive cough, diarrhoea, vomiting and erythematous rash in the perioral and cervical area, back, buttocks, perineal and thighs region..

### *Clinical findings*

The infant was seriously ill, with axial hypotonia and hyperpyrexia 40°C. The skin was pale, mottled with prolonged capillary refill time and cold extremities. There was present erythematous rash, exfoliation and crusting of perioral, cervical, perineal and flexural thighs area; purulent conjunctivitis. (fig 1, fig.2 and fig.3) She presented moderate dehydration and clinical signs of malnutrition (body weight was 4500 g). Productive cough, tachypnea 50 breaths/ minute, intercostal bulging, dyspnea, alveolar and bronchial crackles were noticed; low blood pressure (62/33 mmHg) and high cardiac rate (140/'). Watery stools, oliguria and right ear pain accentuated by pressure on the tragus were present.



Fig. 1. Clinical aspect: face lesions.



Fig. 2. Clinical aspect: cervical and back area.



Fig. 3. Clinical aspect of the child: perineal and thighs area.

#### Consultations

Chest roentgenogram noticed bilateral lobular infiltrates in the lungs.

ORL examination mentioned right ear purulent secretion.

#### Laboratory findings

Were present acute-phase reactants: leucocytosis with neutrophilia and left shift (tabel I); positive CRP, high ESR, increase serum  $\alpha_2$  globulin fractions (tab. IV); anemia and thrombocytopenia (tab.II); metabolic acidosis ( $\text{pH} = 7.21$ ,  $\text{BE} = -8$ ) and low electrolytes level. (tabel III).

Tabel I. Blood cell count.

Parameter	Value
Leucocyte/mm <sup>3</sup>	17800
Immature forms%	12
Granulocytes %	78
Lymphocytes %	10
Hemoglobin g/dl	9,7
Erythrocyte/mm <sup>3</sup>	2940000

Tabel II. Tests for coagulation.

Parameter	Value
Platelet count/mm <sup>3</sup>	87000
Bleeding time	3'30"
Coagulation time	4'20"
Prothrombin time	30"

Tabel III. Serum electrolytes level.

Parameter	Value
Na mmol/l	128
K mmol/l	3,8
Ca mmol/l	2,2
Cl mmol/l	90

Biochemical tests for liver and renal function were normal. It was present low level of seric proteins (45 g/l) with decreased albumin level (48%).

Peripheral and blood cultures were positive for *Staphylococcus aureus*. (tab. V) Germ's sensitivity was good for Rocephin and Lincomicine.

Tabel IV. Acute phase reactants.

Parameter	Value
Protein C reactiv	pozitiv
ESR mm/1h	65
Fibrinogen level g/l	4,99
Serum α <sub>2</sub> globuline %	22

Tabel V. Bacteriological findings

Specimen	Result
Blood	Staph. aureus
Occipital lesion	Staph. aureus
Otic secretion	Staph. aureus
Pharyngeal swab	Staph. aureus
Conjunctival secretion	Staph. aureus
Urine	Sterile
Stool	Negative

Skin biopsy from thigh area was performed but it was inconclusive.

#### *Medical therapy:*

- Systemic antibiotic treatment with Rocephin 0.5g/day and Gentamycin 0.02 g/day for 10 days, then Lincomycin 0.1 g/day
- Active immunization with antistaphylococcal vaccine, 7 doses
- Intravenous fluid resuscitation with isotonic sodium chloride solution; glucose 10% and electrolytes; correction of metabolic acidosis with NaHCO<sub>3</sub> 8,4%
- Oxygen and antipyretic therapy
- Parenteral nutrition with Aminosteril, Intralipid; administration of vitamins
- Diet therapy: oral glucose electrolyte solution (Gesol); carrots soup, boiled rice with 5% glucosis; low-lactose formula.

#### *Evolution*

Diarrhoea stopped after 5 days of treatment. Purulent otitis and conjunctivitis cured in 7 days, bronhopneumonia in 14 days and skin lesions healed in

4 weeks. Weight became upward after 48 hours of therapy and total increase was 1600 g.

#### **Discussions**

Severe malnutrition and iron deficiency anemia were caused by low birth weight and mistakes in infant's nourishment. Sepsis with multiple metastasis in lungs, ear and conjunctiva was caused by *Staphylococcus aureus*. The bacteria entered through damaged skin. The diarrhoea was secondary and dehydration driven to loss of electrolytes and metabolic decompensated acidosis. The appropriate therapy led to complete recovery without scarring.

#### **Conclusions**

- Staphylococcal infections have a high incidence in infants
- Malnutrition, iron deficiency anemia and lack of hygiene are factors that increase susceptibility for systemic staphylococcal infections
- Complications of skin infections are numerous and sepsis is a very serious one
- Antibiotics are the mainstay of therapy in severe staphylococcal infections

#### **References**

1. Forbes CD, Jackson WF.: Color Atlas and Text of Clinical Medicine, 2nd Edition, 1997: 35-36; 35, 38, 95
2. Berkow R, Fletcher AJ, Beers MH: The Merck Manual of Diagnosis and Therapy, Sixteenth Edition, 1992: 2416-2417
3. Bell EA: New topical offers a choice in impetigo treatment, *Inf Dis Child*, 2007, Medline
4. Behrman RE, Kliegman RM, Arvin AM: Nelson-Textbook of Pediatrics, 15th Edition, 1996: 745-8 ; 1851-1852
5. Davidovich CW, Wittler RR, Ruff ME, Bass JW, Brawning WC: Impetigo. Current etiology and comparison of penicillin, erythromycin and cephalexin therapies, 1990, *Am J Dis Child*, 12 (144), 1313-1315
6. Rashid MR, Silverberg MA: Impetigo, 2008, eMedicine
7. Johnston GA: Treatment of buloous impetigo and the Staphylococcal scalded skin syndrome in infants, *Expert Review of Anti-infective Therapy*, 2004, 2 (3): 439-446
8. King RA, de Saint Victor P: Staphylococcal scalded skin syndrome, 2006, e Medicine

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## IV. PEDIATRIC SURGERY

### DIAGNOSIS AND TREATMENT OF BLUNT SPLEEN TRAUMA

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#### Abstract

The consequence of a serious accident in children is represented by a triad associated with closed cranial injuries, abdominal or thoracic lesions and long bone fractures (9). Spleen is the most involved intraperitoneal organ in closed abdominal traumas (1,8). Splenic lesions can be isolated or may be associated with other abdominal organ lesions. Sonography and CT are the most useful diagnostic methods which reveal the presence and extension of the splenic lesions.

**Key words:** spleen trauma, sonography, nonoperative therapy

#### Introduction

The therapeutic concept in abdominal injuries, especially those of the parenchymatous organs, has dramatically changed in the last 25 years (2,3,9). The new therapeutic tactics are strictly conservative and has been possible due to the spectacular progress in the imagistic, particularly speaking of sonography and CT.

#### Material and method

This is a retrospective study of patients with splenic injury treated in our service from May 1990 until May 2000. During this period, 27 patients were admitted with splenic lesions, with 10 of them having one or more associated major lesion: pulmonary lesions – 6 patients, fractures – 5 patients, hepatic lesions – 4 patients, cranial injury – 2 patients, pancreatic rupture – 1 patient and renal rupture – 1 patient. The patient age group was confined to 5-15 years, with 16 male patients and 11 female patients. Based on etiology, most of the injuries took place during sport practice – 10 cases, followed by road accidents – 8 cases (pedestrians - 5 cases), fall from a height – 7 cases, other causes- 2. Majority of the patients (n =23) benefited of medical assistance while on their way to our hospital, 2 of whom were brought

intubated, while the other 4 patients were 4 patients were transported by there guardians. During admission in our clinic, 15 patients were hemodynamically stable, 11 were in critical state in hypovolemic shock, while one patient was brought in moribund state. Clinical examination and simultaneous re-equilibration measures were done in 11 patients who required it. The severely critical patient (with splenic and hepatic injury) was immediately transferred to the operating room.

Emergency Sonographic Examination (ESE) was done in 22 patients (all the 15 hemo-dynamically stable patients initially had only sonographic examination), while emergency CT in 15 patients (all 11 hemodynamically instable patients had emergency CT, 7 of these also had ESE). At the ESE, lesions were identified in 18 patients (81%) while at CT in 14 patients (93%). Grade IV lesions (CT classification grade I-IV) was present in 3 patients, while other 7 patients had grade III splenic lesions.

All patients with grade III and IV lesions, all hemodynamically instable patients and those with multiple lesions were at first admitted in the intensive care unit. 8 patients required blood transfusion, in volume ranging until 30ml/kg body weight. Only 2 of the 27 patients were surgically treated: the patient brought in moribund state was discovered intra-operative having both the hepatic lobes destroyed with lesions of the large veins of the liver as well as splenic rupture, while another patient had splenic fragmentation. In the latter patient, correction consisted in “approximation” of the fragmented spleen and wrapping it in a vicryl bag.

All patients, except the one who died, were followed up in evolution: clinically, the blood parameters and imagistic. Sonographic follow up was performed in all patients while CT follow up was performed only.

## Results

Only a single death was noted: the patient brought in moribund state who died on the operating table. Evolution of all other patients was favorable, without any evidence of complications related to splenic lesions. Three of the patients with pulmonary lesions necessitated pleural drainage. One patient with epidural hematoma was bored, the hematoma evacuated and the hemostasis applied, with progressive evolution without any complications. Bed rest was sustained over a variable period: 2 days in operated patients, 10-14 days in majority of patients, 14-21 days in patients with grade III and IV lesions. Discharge of patients was done after having a hemotocrite balance approximately around 7 days. Sports were restricted for a period of 2-3 months from the accident. After the discharge, the patients were followed up clinically and imagistic in our service or other clinical units until the disappearance of lesions.

## Discussions

The high incidence of infections after splenectomy has been described in numerous publications (7). The risk of sepsis after splenectomy lies between 0.5 and 20%. The risk is higher in patients under 4 years and in most of the cases in the first 2 years after splenectomy (6). On the other hand, the current diagnosis possibilities (sonography and CT) and monitoring of patients with lesions of the parenchymatous organs have made it possible in the last many years to avoid a surgical treatment in majority of patients. More than 90% of our patients were subjected to conservative treatment.

Evaluation of patients with abdominal injury can be a difficult task for the physician (5). Splenic lesions should be suspected in patients with generalized abdominal pain or pain in the left hypochondria associated with rigidity in the superior abdominal quadrant (left) and sometimes with the pain radiated to the left shoulder. Shock can be present in proportion of over 40%. In our series, 11 patients presented with shock, while another patient was brought in moribund state. Sonography is the elective imagistic tool for the paraclinical diagnosis in stable patients. Even if the lesions are not discovered in all patients at ESE, the percentage of positive results (sonographic diagnosis of the lesion) nears 100% at repeated examinations at day's 1-2 post admission. In hemodynamically unstable patients, CT with contrast substance is the obligatory examination. This examination reveals lesions in more than 90% of the cases and generally provides information more exact than sonography in

the diagnosis of associated lesions. After ascertaining the examination mode, sonography is also indicated in these patients (easily accessible, cost effective, absence of radiations could be repeated at short intervals). Our entire grade III-IV patients – grade classification I-IV (7) – were identified at the first imagistic examination. It must be mentioned that in some patients ESE determined lesser severity of lesions than the actual grade. The final extension of the lesions was correctly identified either by CT performed immediately after sonography (unstable patients) or by repeating the sonography (in stable patients) at days 2 or 3 post admission.

Effusion with peritoneal lavage (EPL) can be avoided in children when sonography and CT examination s are at hand. Rothenberg (5) published a study which showed that laparotomies done for abdominal traumas based on the results of EPL were futile in proportion of 67.5%. EPL retains its indication in multiple trauma cases when the associated lesions require urgent therapy due to shortage of time for CT.

Indications for surgical treatment are recommended in the following cases: requirement of blood transfusion exceeding amounts of more than 50% of patient's total blood volume, persistent hypotension or evidence of significant continuous hemorrhages, associated lesions that require surgery. In our series, 10 patients had grade III and IV lesions: of these only 2 were operated, while 9 required blood transfusion reaching amounts of 30ml/kg body weight. Others were stabilized by intravenous infusion of hydro-electrolyte solutions and macromolecule solutions. Bed rest was maintained over a variable period, the criteria being stabilizing hematocrite and hemoglobin and improvement in the sonographic image and from this moment another 7 days of bed rest. After this period, the patients were mobilized and were discharged the same day or the day after. American authors have suggested that hospitalization can be reduced without any negative influence on the results: patients can be discharged in 5 days after stabilizing the hematocrite (9).

In conclusion, the standard treatment of isolated splenic lesions should be the conservatory treatment in 90% of the cases. The approach should be different in cases of the presence of associated lesions which require surgery. The paraclinical examination of choice in stable patients remains sonography in cases of emergency as well as for follow up in evolution while CT examination has an absolute indication in unstable patients.

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**References**

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| <ol style="list-style-type: none"><li>1. Almond PS, Arensman RM. Abdominal trauma in children in „Pediatric Trauma: Initial care of the Injured Child“, Raven Press, Ltd., New York, 1995, pg. 77-97</li><li>2. Iuchtman M, Alfici R, Sternberg E, Trost L, Litmanovitch M. Multimodality management in severe pediatric spleen trauma. <i>Isr Med Assoc J</i> 2000, 2: 523-5</li><li>3. Lakhoo K, Bass DH, Cywes S. Blunt splenic trauma in children. <i>S Afr J Surg</i> 1991, 29: 108-9</li><li>4. Rothrock SG, Green SM, Morgan R. Abdominal trauma in infants and children: prompt identification and early management of serious and life-threatening injuries. Part II: Specific injuries and ED management. <i>Pediatr Emerg Care</i> 2000, 16: 189-95</li></ol> | <ol style="list-style-type: none"><li>5. Rothenberg S, Moore EE, Marx JA, Moore FA, McCroskey BL. Selective management of blunt abdominal trauma in children--the triage role of peritoneal lavage. <i>J Trauma</i> 1987, 27: 1101-6</li><li>6. Rowe MI, Fonkalsrud EW, O'Neill JA, Jr, Coran AG, Grosfeld JL. Abdominal and genitourinary trauma in „Essentials of Pediatric Surgery“, Mosby, St. Louis, 1995, pg. 197 – 213</li><li>7. Scorpio RJ, Wesson DE. Splenic Trauma in „Pediatric Trauma“, Mosby, St. Louis, 1993, pg. 456-463</li><li>8. Shoham N, Sweed Y. Abdominal trauma in childhood—the conservative approach in 95 cases. <i>Harefuah</i> 1999, 2: 672-7</li><li>9. Stylianos S. Controversies in abdominal trauma. <i>Seminars in Pediatric Surgery</i> 1995, 4: 116-9</li></ol> |
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## EARLY VELOPLASTY VERSUS CLASSIC URANOSTAPHILOGRAPHY

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### Abstract

In the management of the cleft lip and palate the veloplasty represents an important part of the treatment. Classic, the majority of the surgeons, practices this intervention after the age of 18 months of the child, in the same time with the closure of the hard palate. Another timing to close the hard and soft palate was introduced by Psaume and Malek in 1983: precocious veloplasty at the age of 3 months and further, the cheiloplasty and uranoraphy in the same intervention at the age of 6-7 months. In our department we practiced a modified Malek's protocol: in the same manner, the veloplasty at the age of 3 months, only the cheiloplasty of the age of 6 months and the uranoraphy, classic, after the age of 18 months. We made a comparative prospective study for a period of 10 years (from January 1995 to December 2004) using classic approach and modified Malek's protocol. From a cohort of 158 patients with cleft lip and palate, 125 (79,11%) were treated in the classic manner and 33 (20,89%) were treated with early veloplasty. The palatal suture was broken partially or totally in 10 patients (30,30%), after that to these patients was made classic uranostaphilography after the age of 18 months. The study also shows the advantages and the disadvantages of the precocious veloplasty and the necessity of a long-term study to clarify the utility of this approach.

**Key words:** cleft lip and palate, veloplasty, uranoraphy, urano-staphilography, cheiloplasty.

Cleft lip and palate (CLP) represents a frequent, complex, malformation, which poses multiple treatment problems, from birth until adulthood. This malformation requires the treatment provided by many specialties. Due to the complexity of the malformation and treatment, over a long period of time, there have been developed a large number of treatment schemes and numerous surgical methods for each treatment

step. None of the treatment plans has proved to be ideal. According to the report of the Eurocleft Project 1996 – 2000, after analyzing the reports received from 201 European centers that treat CLP, there have been noticed a large number of approaches regarding this malformation, hardly finding two resembling methods.

All the surgical interventions are aimed at obtaining a result that is esthetic and morphologically normal, or as close to normal as possible. But, in the mean time, they can also be the cause of secondary bone deformation, thus leading to new ideas for various surgical techniques, and new approaches to this complex pathology. Surgical treatment methods can be classified in two main categories:

- *Classical Timing*, presents two major, opposed, tendencies, referring to the age when the procedure that closes the hard and soft palate should be performed. Classic, the treatment begins with the lip reconstruction, and, since the year 1954, it's been applied from the age of 6 months, considering that before this age the risk of retraction of the incisive region still exists ( Petit et Psaume-Ullik) (2,3). Then, after Victor Veau, the soft and hard palate it totally closed at the age of 18 months, associating with this treatment method another orthodontic treatment at the age of 6 years. In the mean time, Schwekendiek was promoting the reconstruction of the soft palate at the age of 6-8 months, and his disciples have retained the same dates as Victor Veau, but closing the hard palate at the age of 7 or 8 years, associating with this treatment an obstruction prosthetics device from birth until later (Hotz) (2,4). Regarding these very different tendencies and treatment methods, all which cause secondary bone deformation of more or less importance, we ask ourselves if the age

when the hard palate is closed determines the secondary bone deformation, or is the cause of a different etiology (2). The necessity of associating an orthopedic treatment demonstrates that the absence of a closed hard palate is not a solution for avoiding the secondary bone deformation. It is necessary to consider the notion of scar retraction as well as the one of the abnormal muscular balance between the tongue and the upper lip (2).

- *The Inversed Sequence ( early veloplasty )*, was introduced by Psaume and Malek in 1983: early prosthetic device for restoring the role of the tongue. This is an obturation device with a velar extension which is used during the age of two months. This device will reduce the windening of the tongue, will help it's tip to lower, allowing, at the same time the correction of the palatal plates, the growth of the vomer and prepares the function of the soft palate.

The early staphyloraphy will lead to the correction of the pterygoid process with the narrowing of the cavum, and the tongue will advance forward and will lower, allowing it to play the contra-pressure role during the process of suction. The tongue will orient the growth of the upper arcade, particularly the outer margin of the division, before the lip reconstruction. Through this method a complete reconstruction of the malformation can be achieved, before the age of 7 months. (2, 3, 5, 6, 7). This method avoids transversal scar retractions and allows the tongue to oppose the convergent unfavorable forces. The growth can take place normally for the bone structure, in spite of a total surgical reconstruction, without the mandatory support of a prolonged orthodontic treatment.

### Material and Method

In the Pediatric Surgery and Orthopedics Clinic of the "Sf. Maria" Children Hospital, from Iasi, both methods for approaching the CLP treatment have been used, but in the inversed sequence, the procedure described by Malek in 1983 has been modified: staphyloraphy at the age of 3 months, cheiloplasty at the age of 6 months and then uranostaphyloraphy during between 18 and 24 months. Considering this new therapeutic approach, we have aimed at evaluating the results and comparing them with those obtained

during the classic treatment.

Between January 1995 – December 2004 the study has monitored 158 patients with CLP. 125 (79,11%) have been classical treated and 33 (20,89%) with the inversed sequence. From this 158 patients lot, 78 (49,36%) had the lesion on the left side, 28 (17,72%) on the right side and 52 ( 32,92%) had the lesion bilateral. 99 (62,65%) were boys and 59 (37,35%) girls. For the patients who were treated using the inversed sequence, only on 2 (6,06%) the protocol introduced by Malek was used: early veloplasty at the age of 3 months, then cheiloplasty, closing the hard palate, at the age of 6-7 months. For the other patients the modified protocol mentioned above was used.

### Results and Discussions

The study made in this period was a prospective study, the high difference between the number of patients treated by the two methods was due to a number of causes:

- The authors have practiced early veloplasty as many times it was possible, but the other surgeons used the classic timing
- The early veloplasty was not performed if the age of the patients exceeded 3 months and 2 weeks
- At the age of 3 months, some patients had respiratory diseases which counter indicated anesthesia, they were antipoliomielitic vaccine, they had an incomplete cardiac malformation or had counter indications for anesthesia during that time or had important anemia ( Hb under 8 g%)

The early veloplasty was practiced using the Malek technique (fig. 1) published by the author in 1983 (8, 9, 10) (fig. 2, 3).

10 (30,30%) patients, from the total of 33 which had a early veloplasty, had a broken stitch either totally or over 50%, therefore, after the age of 18 they had classic uranostaphyloraphy (fig. 4).

From the 158 patients with CLP, 34 (21,51%) with classic timing had presented fistulas, and 10 (6,32%) with inversed sequence, most of them, 24 (70,59%) and 8 ( 80% ) the fistulas was closed after another intervention after at least one year after the primary surgical intervention. In 7 (20,59%) patients and 2 (20%) two interventions were necessary, and in 3 ( 8,82%) patients ( only from the first category ) three interventions were needed.

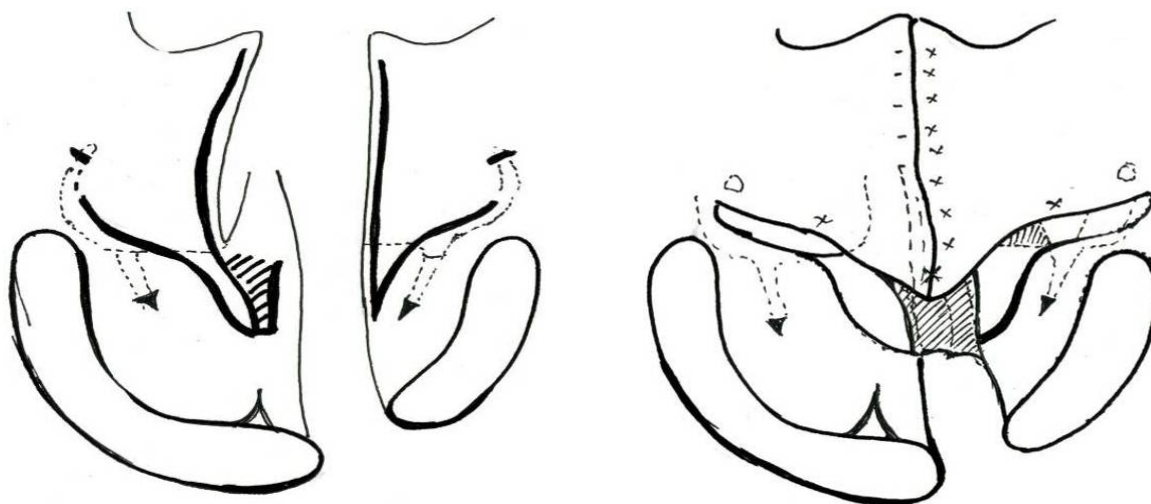


Fig. 1. Early veloplasty, Malek – surgical method.

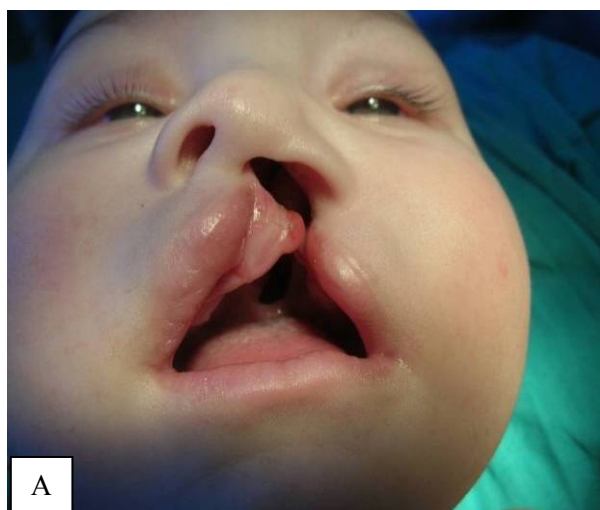


Fig. 2. A- Patient, age of 3 months, before the early veloplasty;  
B – aspect of the palate at the age of 6 months, before the cheiloplasty.

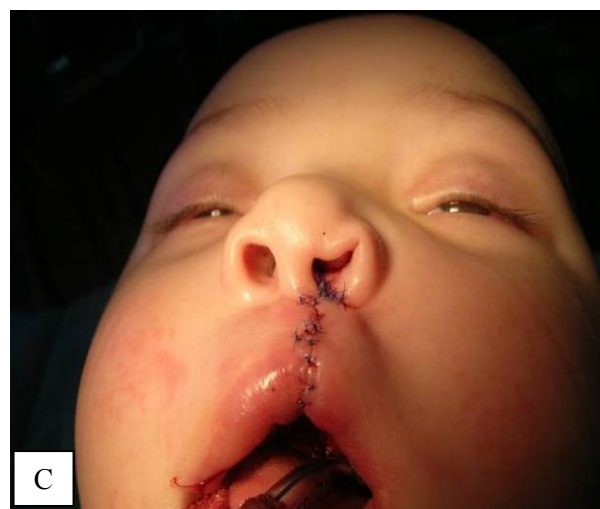
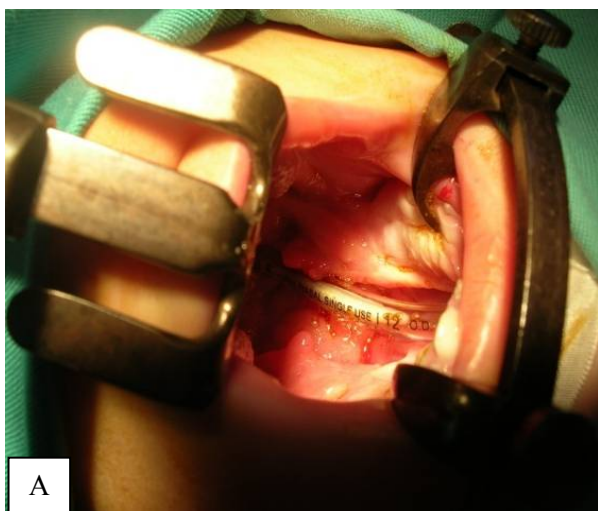


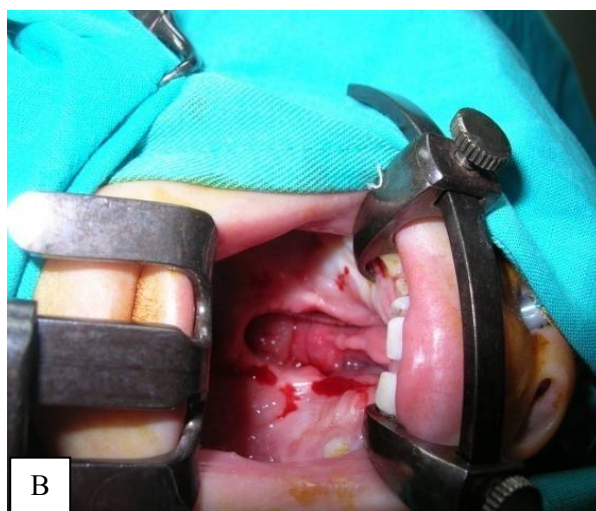
Fig. 2. C – After the cheiloplasty, immediately post operator aspect;  
D – palate aspect at the age of 20 months, before the uranoraphy.



Fig. 3. Patient at the age of 5 months, 2 months after the early veloplasty (before the chelioplasty).



A



B

Fig. 4. A – Patient at the age of 2 years, treated with classic timing (before the uranostaphilography), B – Patient at the age of 18 months, with early valeoplasty, with a partially broken suture (before uranostaphilography).

### Conclusions

The authors consider that the early veloplasty represents a progress in managing the children with CLP, because, after the early veloplasty, the remaining cleft palate on the hard palate, will shrink so much, that when the uranoraphy is performed, the cleft palate is between 0.5 – 1 cm wide. As a result, the uranoraphy will be performed a lot easier, with less complications, thus less fistulas in the palate, which in our study were three times less at the patients who had early veloplasty, the final closing was made easier, also reflected by the small number of interventions.

The family always notice right away, post

operator, improvement in the feeding of breast fed children, with no reflux, with a noticeable enhancement phonation.

There are however disadvantages. From a surgical point of view, it is true that the cleft palate is smaller and easier to close, but in the same time, the dissection for identifying the two layers ( nasal and oral ) is much more difficult, especially in the velar layer, due to the scar tissue. Also, in case the veloplasty has failed and the suture was totally broken, the uranoraphy is harder to practice due to the scar tissues and the cleft palate has a reduced mobility.

It is true that the complications rate (30,30%) was

really high, discouraging even, but we consider that this rate can be reduced by respecting certain necessary practicing conditions for all the elective surgical interventions that we sometimes skipped, but which lead to these complications. The infants who are

supposed to have surgery should have a weight correspondent to the age, and should not have anemia (Hb over 10,0 g%) and to be without respiratory diseases in that time, or less than 2 week before the operation.

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**References**

1. Bill Shaw, P. Nelson, G. Semb, V. Brattstrom, K. Molsted, B. Prahl - Anderson – The Euro- cleft Project 1996-2000, <http://books.google.com/books>
2. Berkowitz S. - A Multicenter Retrospective 3D Study of Serial Complete Unilateral Cleft Lip and Palate and Complete Bilateral Cleft Lip and Palate Casts to Evaluate Treatment, Cleft Palate-Craniofacial J 1999, Vol. 36, No. 5:413-424.
3. Farmand M. - Lip Repair Techniques and Their Influence on the Nose, Facial Plastic Surg 2002, Vol. 18, No. 3:155-164.
4. Pellerin D. – Technique de Chirurgie Pédiatrique, Ed. Masson, Paris, 1978.
5. Stricker M., Raphael B. - Le périoste dans les fentes labio-palatines, Ann Chir Pédiatrique 1983, Vol. 24, No. 4-5: 274-281.
6. Hartridge T., Illing M.H., Sandy J.R. - The Role of Folic Acid in Oral Clefting, British Journal of Orthodontics 1999; Vol 26; No. 2: 115-120.
7. Delaire J. - Intérêt de la rhinoplastie primaire. Considérations techniques, Ann Chir Pédiatrique 1983, Vol. 24, No. 4-5:286 - 296.
8. Malek R. - Staphylorrhaphie précoce - Problèmes chirurgicaux, Ann Chir Pédiatrique 1983, Vol. 24, No. 4-5: 301-304.
9. Malek R.– Traitement initial des fentes labio-palatine, Ann Chir Pédiatrique 1983, Vol. 24, No. 4-5: 256-267.
10. Malek R. - Cleft Lip and Palate: Lesions, Pathophysiology and Primary Treatment, Published by Taylor & Francis, 2001.

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## PRELIMINARY RESULTS AFTER NUSS PROCEDURE IN 5 CASES OF PECTUS EXCAVATUM

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### Abstract

Pectus excavatum (PE) is the most frequent anterior chest deformity occurring in approximately 1 in 1000 live births (1). PE is a depression of the sternum and costal cartilages. PE treatment is surgical. During time more than 50 surgical procedures were done. In 1998 Nuss et al introduced a new minimal invasive surgical procedure for PE correction in which costal cartilage resection or sternal osteotomy is no longer necessary (5). We present 5 case of PE treated by us using Nuss technique. We present you the operative technique and the results for short and medium term. No intraoperative incidences were recorded. Immediate postoperative course was good for all patients. In the 5<sup>th</sup> patient a left pleural effusion occurred 14 days from the intervention and was immediately solved by pleural puncture. Therapeutic and cosmetic results were considered good by all patients and their parents. Preliminary results indicate that Nuss procedure is safe and has excellent outcomes for PE correction in children.

**Key words:** pectus excavatum, Nuss, minim invasive, child

### Introduction

Pectus excavatum (PE) is the most frequent anterior chest deformity occurring in approximately 1 in 1000 live births (1). PE is a depression of the sternum and costal cartilages. In most of the cases the deformity is asymmetric and maximum depth point is located at the inferior part of the thorax (2). In only a third of the patients the debut of the disease is during childhood (2). More often the disease becomes apparent in the prepubertal growth spurt period. The most frequent associated diseases are: scoliosis (26%), cardiac malformations (1.5%) and asthma (5.2%) (3). Sometimes PE is a component of genetic disorders: Currarino-Silverman, Noonan and Marfan (4). The most frequent symptom is the limitation of the effort ability. An important aspect is the physic disturbances

caused by the esthetic defect. Other symptoms like: cardiac arrhythmia or chest pain are rare.

PE treatment is surgical. During time more than 50 surgical procedures were done. First attempt to correct PE was done by Meyer in 1911. The surgical technique introduced in 1949 by Ravich and modified by Adkins was in short time widely adopted and remained for almost 4 decades the main treatment method for PE. Other treatment options like: magnetic or suction elevation of sternum, sternal turnover or silicon prosthesis had poor outcomes and failed to impose as a treatment viable option.

In 1998 Donald Nuss introduced a new minimal invasive operative technique. Under thoracoscopic surveillance a rigid metal bar is inserted under the sternum trough lateral thoracic incision. The bar is introduced with the concavity facing anterior and then turned posterior in order to correct sternal bending. Cartilage resection or sternum osteotomy is no longer necessary (5). The technique was improved in the last decade by the wildy adoption of the thoracoscopy and the introduction of the lateral bar stabilizers (6). Long term favorable outcomes (95%) led to its wide adoption.

### Material and methods

Between July 2007 and September 2008 five PE patients were treated using the minimal invasive technique. The intervention represent premiere because it was performed by a team composed exclusively by Romanian surgeons. We present you the five cases, our operative technique and the short and medium term outcomes.

### The Patients

1. Male 14 years old child. Severe, symmetric PE in the lower 1/3 of sternum. Effort dyspnoea was the only symptom present. EKG and cardiac echografy are normal. Haller index (HI) is 5.98. Left pleural drainage was

- necessary for 2 days. He left the hospital 8 days after surgery.
2. Male 12 years old child. Symmetric PE. Associated disease: mitral valve prolapsed, myopia, isolated atrial extrasystole. Anamnestic effort dyspnoea was present for a least one year before. Sternum has a compressive effect on the right ventricle at the CT scan. HI is 3.82. After the intervention bilateral pleural drainage was necessary for 4 days. He left the hospital 6 days from the intervention.
  3. Male 18 years old patient. Symmetric PE. No symptoms are present. EKG shows a minor right bundle branch block. HI is 3.62. Right pleural drainage was maintained for 5 days. He left the hospital 8 days after surgery.
  4. Male 14 years old child. Left rotated asymmetric PE. Associated disease: mitral valve prolaps, dilated cardiophaty, scoliosis, pulmonary hypertension. The thoracic deformity increased significant and the effort dyspnoea accentuated during the past year. CT scan showed that sternum has a compressive effect on the right ventricle and the heart is displaced to the left. HI is 3.7. Bilateral pleural drainage was maintained for 4 days. He was released from hospital 6 days from surgery.
  5. Male 14 years old child. Cup shape PE slightly rotated to the left. The deformity increased significant during the past year. Physical exam showed easy effort fatigue. HI is 4.5. The bilateral pleural drainage was removed 2 hours from the intervention. (fig. 1)



Fig. 1 The patients.

### Pre-operative preparation

Several evaluations were performed for each patient before surgery: spirometry, cardiology consult, Rx, CT (fig. 2), EKG, cardiac echography, abdominal echography, genetic consult, ophthalmologic consult. Lab tests performed are: complete blood count, liver function tests, kidney function tests, inflammation tests, glycemia, blood electrolytes, bleeding and coagulation time.

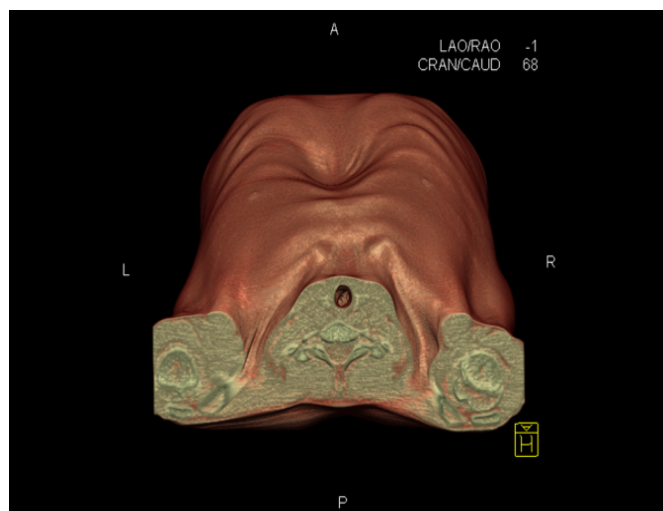


Fig. 2. CT scan with 3D reconstruction.

### Operative technique

- Before surgery the Lorenz bar was shaped to the desired shape in order to reduce the length of the intervention.

- The patient is put under general anesthesia with oro-tracheal intubation.

- The trocar for thoracoscope is inserted in 7th right intercostal space in the mid axillary line.

- Bilateral thoracic incisions are performed in the mid axillary line at the level of deepest point of the depression.

- In first 4 patients the incision was transverse while in the 5<sup>th</sup> case the incision was orientated vertical.

- Skin tunnels are raised anterior from each incision to the top of the deformity where the thoracic cavity is entered.

- When the pleural cavity is opened an iatrogenic pneumothorax is made. This pneumothorax is sufficient to form the necessary work chamber and is maintained by using low ventilation pressures.

- Under thoracoscopic surveillance the introducer is inserted in the right pleural cavity.

- Facing upwards and immediately under the sternum we slowly passed the introducer through the anterior mediastinum to the left pleural cavity.

- The assistant introduce his finger in the left pleural cavity and elevates the sternum when the

introducer is passed through the mediastinum. This maneuver increase the distance between sternum and heart.

- The assistant with his finger introduced in the pleural cavity expect and guide out the introducer.

- The introducer is than elevated and pressure applied above the sternum in order to correct the deformity.

- We attached an umbilical tape to the left end of the introducer and pulled through the tunnel by withdrawing the introducer from the right side.

- We attached the umbilical tape to the Lorenz bar and pulled the bar to the left side with the concavity facing anterior.

- After is introduced the bar is flipped with concavity facing posterior.

- Lateral stabilizer are fitted at each end and sutured to the rib cage.

- The skin is closed using non-resorbable sutures.

- We use bilateral pleural drainage in 3 cases and unilateral in 2 cases.

- On the right side we used the thoracoscope incision for drainage.

- For pain management we inserted an epidural catheter. The patient receives intravenous antibiotic, an anti-inflammatory and an analgesic drug for 5 to 6 days.



Fig. 3. Trocar introduction.



Fig. 4. Transverse skin incision in mid axillary line.



Fig. 5. Longitudinal skin incision in mid axillary line.



Fig. 6 Tunneling.



Fig. 7. The deformity is corrected by applying pressure on sternum and ribs.



Fig. 8. The bar is inserted with the concavity facing anterior.



Fig. 9. The bar is flipped with the concavity facing posterior.



Fig. 10. Lateral stabilizer are fitted at each end and sutured to the rib cage.

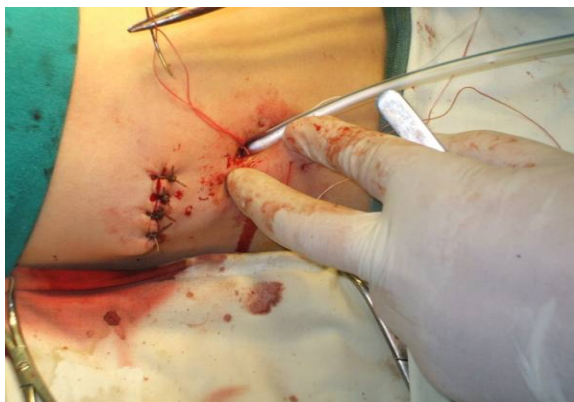


Fig. 11. The thoracoscope incision is used for right pleural drainage.



Fig. 12. Bilateral pleural drainage.

## Results

No intraoperative incidences were recorded.  
 Blood loss was minor.  
 Time of operation was between 60 and 90 minutes.  
 Postoperative course was good for all patients.  
 No complication occurred in 4 of the 5 cases.  
 In the 5<sup>th</sup> case right pleural effusion developed 14 days from the intervention and was immediately

solved by pleural puncture with no further complication.

We had no bar displacement.

Postoperative pain was minor.

Therapeutic and cosmetic results were considered good by all patients and their parents.



Fig. 13. Postoperative aspect patient 1.



Fig. 14. Postoperative aspect patient 2.



Fig. 15. Postoperative aspect patient 3.



Fig. 16. Postoperative aspect patient 4.



Fig. 17. Postoperative aspect patient 5.

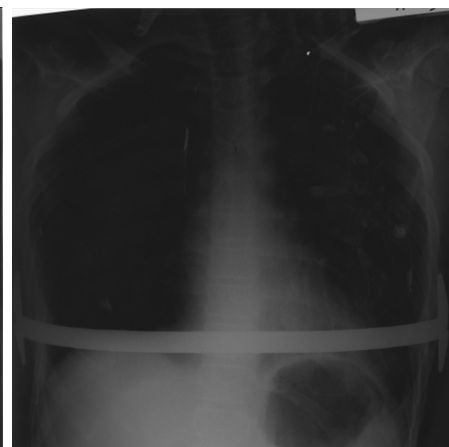
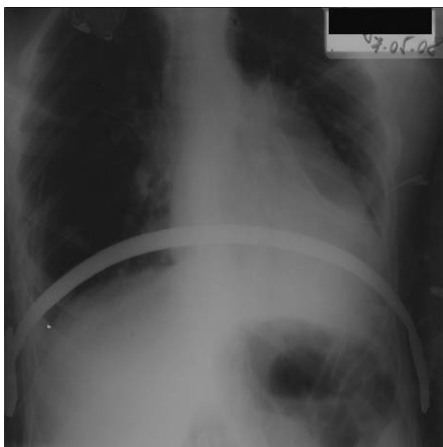


Fig.18. Postoperative Rx patient 1. Fig.19. Postoperative Rx patient 4. Fig.20. Postoperative Rx patient 5.

### Discussions

Since its introduction in 1998 Nuss technique for PE correction had stimulate the interest and was adopted by a growing number of surgeons all over the world. Previous studies have established that a HI greater than 3.1 surgery for PE should be considered (9). Indication for surgery was established in all our five case based on objective criteria ( $HI > 3.1$ ), clinical and psychological criteria. Age of the patient was also a key factor in the decision for surgery. The ideal age for PE correction is just before puberty, when the chest is still very malleable and the bar is in place during the pubertal growth spurt, reducing so and the possibility of recurrence (6). For adult

PE patient Nuss technique is still a subject of debate. Four of our patients are in pre- and puberty.

One of the main advantages of Nuss technique is the absence of the anterior thoracic incision, whom in open technique, lead in many cases to big, unaesthetic keloids. For this reasons we modified the initial technique by performing a longitudinal instead of transversal incision. In Nuss technique costal cartilage or sternal osteotomy resection is no longer necessary and operating time is significant reduced (7).

The most frequent complication cited before are: pneumothorax (6.9%), wound infection (4.5%), pericarditis (2.4%), bar displacement (1.2%) (8). None of these occurred in any of our patients. Only one patient developed a pleural effusion 14 days from surgery resolved successfully by pleural puncture.

The initial technique used CO<sub>2</sub> pleural insufflation for creating the necessary work chamber (5). We considered that the pneumothorax formed

spontaneously when the pleural cavity is opened offer sufficient space and positive pleural pressure is not necessary.

We consider thoracoscopy necessary in order to avoid heart or lung lesions. Thoracoscopy was particularly useful for the two cases where the sternum was in direct contact with the heart. The assistant introduce his finger in the left pleural cavity to expect and guided out the introducer. This adaptation of the initial technique offered a better control for introducer. An additional adaptation used by us is to elevate the sternum when the introducer passed through mediastinum increasing the space between the back of the sternum and the heart. This maneuver is achieved by introducing a finger inside the left pleural cavity through the site prepared for the left side exit of the introducer. In this way Rockitansky's subxiphoid incision becomes unnecessary (10).

One of the main problems for Nuss technique is greater postoperative pain (7). For our patient pain level was lower than that cited before. Pain management was done mainly by intravenous drugs and for short time. The epidural catheter was necessary only in 2 cases and for 3 days only.

We considered that intravenous antibiotics for 6 days are necessary for infection prophylaxis.

### Conclusions

- Preliminary results indicate that Nuss operation for PE correction is a safe surgical technique.
- Postoperative outcomes are good.
- Hospital stay length is short.
- Blood loss is minimal.
- Cosmetic outcomes are excellent, appreciated by the patients.
- We are waiting for the long times results.

### References

1. Kelly RE, Lawson ML, Paidas CN et al. Pectus excavatum in a 112-years autopsy series: anatomic findings and the effect on survival. *J Pediatr Surg* 2005;40:1275-8
2. Kelly RE jr. Pectus excavatum: historical background, clinical picture, preoperative evaluation and criteria for operation. *Semin Pediatr Surg*. 2008 Aug;17(3):181-93
3. Popoiu MC, Popoiu A. Anterior chest wall malformations in children. Diagnostic and treatment. Romania, Timisoara: Eurostampa 2002
4. Radulescu A, Adam O, Boia ES, Popoiu MC. Pectus excavatum. Diagnostic and treatment. Romania, Timisoara: Artpress 2005
5. Nuss D, Kelly RE jr, Croitoru DP, et al. A 10 years review of a minimal invasive technique for the correction of pectus excavatum. *J Pediatr Surg* 1998;33:545-552
6. Nuss D. Minimally invasive surgical repair of pectus excavatum. *Semin Pediatr Surg*. 2008 Aug;17(3):209-17
7. Fonkalsrud EW, Beanes S, Hebra A et al. Comparison of minimally invasive and modified Ravitch pectus excavatum repair. *J Pediatr Surg*. 2002 Mar;37(3):413-7
8. Park HJ, Lee SY, Lee CS. Complications associated with the Nuss procedure: analysis of risk factors and suggested measures for prevention of complications. *J Pediatr Surg*. 2004 Mar;39(3):391-5; discussion 391-5
9. Kilda A, Basevicius A, Barauskas V et al. Radiological assessment of children with pectus excavatum. *Indian J Pediatr*. 2007 Feb;74(2):143-7
10. Greeber MG. Chest wall and sternum resection and reconstruction. In Patterson et al, editor. *Pearson's Thoracic and Esophageal Surgery*. 3d ed. Philadelphia: Churchill Livingstone Elsevier; 2008. p. 1306-1328.

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## TRICHOBEZOAR WITH LARGE BOWEL OBSTRUCTION IN CHILDREN – CASE REPORT

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### Abstract

A trichobezoar is a mass of cumulated hair within the gastrointestinal tract. Stomach is the common site of occurrence. Intestinal obstruction due to trichobezoar is extremely rare. In this report, we describe one case of an atypical localization of a trichobezoar in an 11-year-old girl, who presented with large bowel obstruction. We performed an exploratory laparotomy and we diagnosed a movable intraluminal tumor of the left angle of transverse colon. Treatment consists of milking the contents to the rectum.

**Key words:** trichobezoar, children, bowel obstruction.

### Introduction

For centuries, different types of bezoars (accumulations of foreign matter) have been known to occur in the stomach and intestines of animals and humans. Trichobezoars are classically described as consisting mostly of hair, and are often noted in psychologically impaired children who also have trichotillomania (a compulsion to pull out one's hair)<sup>[1]</sup>. Rapunzel's syndrome, due to compulsive hair-chewing, results in the formation of a long-tailed trichobezoar, a documented variant of trichobezoars.<sup>[1]</sup>

Patients with bezoars often present with abdominal pain, anorexia or vomiting, anemia, and malnutrition of different degrees. Signs and symptoms of bowel obstruction or perforation may occur. Occasionally the patient is entire asymptomatic<sup>[2]</sup>.

Various imaging modalities have been recommended for detection of bezoars<sup>[3],[4]</sup>. The imaging findings are helpful in diagnosing trichobezoar. *The conventional radiography* shows a masse of opaque soft tissue in a swollen stomach<sup>[3],[4],[5]</sup> and can reveal bowel obstruction with dilated small bowel loops with fluid and air-filled loops proximal to the site of obstruction and distal collapsed small bowel. A calcified rim may delineate the edge of the bezoar<sup>[3],[5]</sup>. *The ultrasonography* shows a typical curvilinear trichobezoar with bright echogenic band, this does not allow transmitting the ultrasound waves which generate a shadow over the left upper quadrant. The high echogenicity of hair and the presence of multiple

acoustic interfaces created by trapped air and food limits the ultrasonography of the trichobezoars<sup>[3],[4],[5],[6]</sup>. Both, *the contrast radiography and the endoscopy* of the upper GI tract are the diagnostic procedures of choice for establishing the diagnosis<sup>[3],[6]</sup>. The upper GI contrast radiography confirms the existence of the trichobezoar and might detect other complications such as gastric ulcers. In addition, the upper endoscopy is definitively the diagnostic support for trichobezoar; it might be used for endoscopic retrieval of proximal small trichobezoars<sup>[3]</sup>. *The computed tomography* (CT-scan) is the most useful diagnostic tool in patients with bezoars because it reveals the localization of the bowel obstruction; it shows also a well-defined intraluminal mass of the bezoar in the transitional zone of the obstruction. A mottled gas pattern in the mass is reported characterizing the bezoar, and it is supposed to be air bubbles retained within the bezoar<sup>[6],[7]</sup>. Recently, researchers have recommended *magnetic resonance imaging* (MRI) for the evaluation of small-bowel disease. Fast imaging techniques coupled with advantages of breath holding improved MRI visualization of bezoars. Therefore, MRI is found to be better support for determining both the site and the cause of small-bowel obstructions. MRI shows the bezoar as a mass in the small bowel containing mottled and confluent low signal intensities on both T1- and T2-weighted MR images<sup>[7]</sup>.

### Treatment

Once the diagnosis is established, trichobezoar removal should be undertaken to avoid the complication of obstruction, hemorrhage, ulceration, perforation and peritonitis and to reestablish proper nutrition<sup>[2]</sup>.

The upper endoscopy of GI tract might be used for endoscopic retrieval of proximal small trichobezoars. Definitive treatment consists of exploratory laparotomy with trichobezoar removal commonly done by gastrotomy and/or enterotomy or milking of contents to the rectum. If complicated, the trichobezoars can be treated by subtotal gastrectomy

and/or intestinal resections. If trichobezoar is into the colon, then colonoscopic evacuation can be performed. Medical treatment is usually inadequate.

### Case report

We present the case of a girl R.A., 11 years'old, coming from urban area, transferred in our clinic with the diagnosis: subocclusive syndrome.

#### *History and clinical findings:*

She had a history of two weeks of colicky abdominal pain, nausea, vomiting, decreased oral intake, changes in bowel movements. At admission in the hospital she presented altered general state, puffed eyes, pale teguments, bilious vomiting, and constipation. The distended abdomen was diffusely sensitive to palpation, with no sign of peritoneal irritation. Abdominal auscultation revealed static intestinal sounds (borborygmus). Digital rectal examination revealed empty rectal ampulla, without presence of any pathological material on the hand gloves. Next day she presented signs of peritoneal irritation in the left abdomen.

#### *Paraclinical:*

The blood count showed a marked leucocytosis, high number of thrombocytes, high acute phase reactants, increased level of urea, and signs of acute dehydration with hyponatremia.

Abdominal X-ray showed multiple air-fluid levels without presence of air under the diaphragm, while the ultrasound examination was negative for any

abnormality. A positive diagnosis of intestinal obstruction was established based on the previous examinations.

#### *Treatment:*

Preoperative care consisted in gastric decompression using nose-gastric tube, parenteral nutrition, antibiotics (piperacilin tazobactam) and infusion with electrolytes.

Surgery –consisted of the following steps:

- \*median abdominal incision;
- \*abdominal cavity exploration- revealed normal stomach, duodenum and ileum, inflammatory adhesions at the first jejune loops with a small blocked perforation at 5-7cm from duodeno-jejunal junction. The thorough exploration of the large bowel revealed one intraluminal movable tumor of the left angle of transverse colon which could be milking to the rectum; this intraluminal tumor retained in this site compressed the first jejunal loop, affecting the vascular supply with secondary ischaemic perforation;
- \* lysis of adhesions, jejunoraphy in double layer;
- \*lavage of the abdominal cavity using NaCl 0.9%;
- \*double drainage of the abdominal cavity, suture of the abdominal wall;
- \*anal dilatation and pull-out the tumor which was a trichobezoar.

The postoperative care consisted of antibiotics (piperacilin tazobactam, solution of parenteral nutrition (glucosis, aminoacids), electrolytes, antalgic drugs.



Fig .1. The trichobezoar.

### Discussions

Bezoars are foreign bodies in the lumen of the digestive tract. The lumen size increases in time by the accumulation of ingested nonabsorbable food or

fibers<sup>[3],[4],[8],[10]</sup>.The bezoar is mostly caused by the presence of indigestible substance in the lumen. Some substances encourage stickiness and concrete formations<sup>[6],[8],[11]</sup>.Bezoar occurs mainly in female

children, who chew and swallow their hair (trichobezoar), vegetable fibers (phytobezoar), persimmon fibers (diospyrobezoar), or semi-liquid masse of drugs (pharmacobezoar)<sup>[3],[4],[11]</sup>.

Trichobezoar (hairball) is a complication of trichotillomania. It consists of recurrent hair pulling, and subsequent trichophagia or mouthing of the hair<sup>[5],[8],[9],[10]</sup>.

During the time, these substances are retained by mucus and become enmeshed; this yields a mass having the shape of the stomach localization where they are usually found<sup>[8],[11]</sup>. These substances attend large size due to the chronicity and delayed investigation of the affection. The age of occurrence of bezoars has been reported to range between 1 and 56 years old<sup>[3],[5],[8]</sup>. Although about 1 of 2000 children suffer from trichotillomania, trichophagia is rarely seen, and a bezoar does not occur in all children with trichophagia<sup>[3],[10]</sup>. Reduced intestinal motility is the most quoted factor in the intestinal bezoar formation. Bezoars mostly originate at the level of the stomach<sup>[3],[5],[8],[11]</sup>, it is probably related to high fat diet causing unspecific symptoms like epigastric pain, dyspepsia, and postprandial fullness. The stomach is not able to exteriorize hair and other substance out of the lumen because the friction surface is not sufficient for propulsion by peristalsis. The bezoars might also

occur with GI bleeding (6%) and intestinal obstruction, or perforation (10%)<sup>[3],[11]</sup>. The most common sites of obstruction are the gastric outlet, or duodenum. Obstructions of distal parts of the small bowel or the large bowel are extremely rare.<sup>[3]</sup> The examination of the hair content in stool would establish the diagnosis, but usually it is not done<sup>[3],[4]</sup>. It is mandatory to perform a thorough exploration of all the small intestine and the stomach searching for retained bezoars.

### Conclusions

Trichobezoar is a rare clinical entity. Stomach is the common site of occurrence. In this case the large bowel obstruction occurred because of the trichobezoar itself which have migrated from stomach and have stopped in the splenic angle of transvers colon. The retained trichobezoar in this site compressed the first jejunal loop, affecting the vascular supply with secondary ischaemic perforation. This case was diagnosed in a complicated stage (mecano-inflammatory bowel obstruction) so the patient will have high risk to develop early and tardive postoperative complication (peritoneal abscesses, intestinal adhesions). After surgery the patient was treated in collaboration with a psychiatry doctor, too.

### References

1. Andrus CH, Ponsky JL. Bezoars: classification, pathophysiology and treatment. *Am J Gastroenterol*. 1988;83:476-478.
2. Stephen L. Gans and Edward Austin: Foreign bodies. *Ashcraft Holders- Pediatric surgery* (second edition); 86-87.
3. Shadwan A, Mohammad A. Small bowel obstruction due to trichobezoar: Role of upper endoscopy in diagnosis. *Gastrointest Endoscop* 2000;52:784-6
4. Roche C, Guye E, Coinde E, Galambrun C, Glastre C, Halabi M, et al. Trichobezoar: A propos de 5 observations. *Arch Pediatr* 2005;12:1608-12
5. Hoover K, Piotrowski J, Pierreb K, Katzc A, Goldsteinb AM. Simultaneous gastric and small intestinal trichobezoars: A hairy problem. *J Pediatr Surg* 2006;41:1495-7
6. Billaud Y, Pilleul F, Valette PJ. Mechanical small bowel obstruction due to bezoars: Correlation between CT and surgical findings. *J Radiol* 2002;83:641-6.
7. Lee JM, Jung SE, Lee KY. Small-bowel obstruction caused by phytobezoar: MR imaging findings. *AJR Am J Roentgenol* 2002;179:538-9.
8. Wang PY, Skarsgard ED, Baker RJ. Carpet bezoar obstruction of the small intestine. *J Pediatr Surg* 1996;31:1691-3.
9. Ganpathi IS, Cheah WK. Laparoscopic-assisted management of small bowel obstruction due to phytobezoar. *Surg Laparosc Endosc Percutan Tech* 2005;15:30-2
10. Salaam K, Carr J, Grewal H, Sholevar E, Baron D. Untreated trichotillomania and trichophagia. *Psychosomatics* 2005;46:362-6.
11. Chintamani, Durkhure R, Singh JP, Singhal V. Cotton Bezoar: A rare cause of intestinal obstruction: Case report. *BMC Surg* 2003;4:5.

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# FAMILIAL ADENOMATOUS POLYPOSIS (FAP): WHAT MUST BE KNOWN AND WHAT SHOULD BE DONE – CASE REPORT

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## Abstract

Familial adenomatous polyposis (FAP) is a neoplastic disorder of major concern to pediatric surgeons due its fatal adenoma-carcinoma sequence. This case report illustrates the attempt of surgery to alter and perhaps break such a sequence.

**Key words:** familial adenomatous polyposis (FAP), APC gene, extracolonic cancer.

## Introduction

Familial adenomatous polyposis (FAP), also known previously as adenomatous polyposis coli (the latter term is being now reserved for the relevant gene), albeit a rare condition, is the most common adenomatous polyposis syndrome. It is classically characterized by the early onset of hundreds to thousands of polyps disseminated throughout the colon. FAP is an important neoplastic disorder since it invariably evolves into carcinoma of the colon by the fifth decade of life if left untreated (adenoma-carcinoma sequence.)

It has an autosomal dominant pattern of inheritance due a germline mutation in the adenomatous polyposis coli (APC) tumor suppressor gene, located on band 5q21.

The incidence of FAP is constant worldwide and ranges from 1 in 6.000 to 1 in 12.000 births, with both sexes being equally involved.

## Classification and natural history

Different mutations in the APC gene define a spectrum of conditions known as APC-associated polyposis which include: (1) FAP, (2) Gardner and Turcot syndrome as overlapping variants with extracolonic manifestations, and (3) Attenuated FAP (AFAP) (or Flat adenoma syndrome.)

**1. CLASSIC FAP:** is diagnosed clinically in an individual with over 100 colorectal adenomatous polyps; or fewer than 100 adenomatous polyps and a relative with FAP. Up to several thousands have been described and the mean is 500 polyps at the time of

diagnosis. The median ages for patients to develop polyps is 16, for bowel symptoms 29 and for colorectal carcinoma is 36 years.

**2.1. GARDNER SYNDROME (GS)** is the association of colonic adenomatous polyposis, osteomas (mainly on the skull and mandible), dental abnormalities (Unerrupted teeth, congenital absence of one or more teeth, supernumerary teeth, dentigerous cysts, and odontomas), and soft tissue tumors (epidermoid cysts, fibromas, desmoid tumors). These benign extraintestinal growths occur in about 20% of individuals and families with FAP.

**2.2. TURCOT SYNDROME (TS)** is a rare combination of multicentric colonic adenomatous polyposis and CNS tumors, usually medulloblastoma. Patients develop colorectal carcinoma in young adulthood from malignant transformation of the precancerous lesions or may arise de novo in the intact intercalated epithelium.

**3. AFAP** as the term implies is considered in an individual with fewer adenomatous polyps averaging 30 and with a more proximal colonic distribution when compared to classic FAP. The median ages to develop polyps is 36 and for colorectal carcinoma is 50-55 years (10-15 years later than patients with classic FAP, but earlier than those with sporadic colorectal carcinoma.)

Gastric polyps and adenomatous polyps of the small intestine are being increasingly detected with appropriate upper gastrointestinal endoscopy.

No correlation could be established between the number of colonic polyps and the frequency of upper gastrointestinal polyps. If gastric polyps are considered to have a minimal malignancy potential, the lifetime risk of small intestine malignancy is 4-12%; being the second most common malignancy in patients with FAP. The majority of small intestine carcinoma occur in the duodenum (mainly in the periampullary region, including the duodenal papilla and ampulla of Vater.) (Table 1).

Table 1 - Upper gastrointestinal polyps in FAP.

Histology	Gastric polyps		Polyps of the small intestine	
	hamartomatous fundic-gland	adenomatous	adenomatous	
Location	fundus + body	antrum	D II + D III	periampullary region
Frequency in FAP	50 %	10 %	50-90 %	50 %

**Diagnosis - tools of the trade****History and physical exam**

- 80% of patients with FAP have a family history of polyps and/or colorectal cancer at age 40 years or younger.
- Most patients are asymptomatic until colorectal carcinoma develops. Symptoms when present include non-specific abdominal pain, a palpable abdominal mass or palpable mass on rectal examination in a young patient, change in bowel habits, a progressively installed chronic bloody diarrhea and unexplained rectal bleeding.
- Congenital hypertrophy of the retinal pigment epithelium (CHRPE), best detected by slit-lamp examination. These are minimal flat pigmented lesions of the retina, mostly multiple and bilateral, that are highly specific for FAP.

**Differential diagnosis**

Peutz-Jeghers Syndrome  
 Bannayan-Riley-Ruvalcaba syndrome  
 Cowden disease  
 Juvenile polyposis syndrome  
 Cronkhite-Canada syndrome  
 Hereditary nonpolyposis colon cancer  
 Hyperplastic polyposis  
 Nodular lymphoid hyperplasia  
 Lymphomatous polyposis  
 Neurofibromatosis type 1 (NF-1)  
 Inflammatory polyposis  
 MYH-associated polyposis

**Lab studies**

CBC - chronic bloody diarrhea and rectal bleeding are often associated with anemia.

Proteinemia- late manifestations of FAP include protein-losing enteropathy and malnutrition.

Alpha-fetoprotein (AFP).

**Imaging studies**

- Barium studies, flexible sigmoidoscopy, colonoscopy ( usually reserved for AFAP because of the proximal colonic distribution of the polyps), front and side-viewing esophagogastroduodenoscopy when FAP is

established ( in order to visualize gastric, duodenal, and periampullary adenomas.)

- Dental panoramic and skull x-ray films to detect osteomas and dental abnormalities encountered in Gardner syndrome.

- Periodic abdominal ultrasounds and CT scans to detect intra-abdominal desmoid tumors and pancreatic cancer.

**Genetic studies**

APC gene sequencing

In vitro protein synthesis assay

Linkage testing

**Histology**

Tubular adenomatous polyps predominate, and later tubulovillous adenomas may be detected as they increase in size.

**Surgical treatment and long - term management**

(1) Proctocolectomy with permanent abdominal wall ileostomy.

(2) Proctocolectomy with ileal pouch-anal anastomosis/IPAA (restorative proctocolectomy) and temporary diverting ileostomy.

(3) Subtotal colectomy and ileoproctostomy, with repeated postoperative proctoscopy at 3-6 month intervals for fulguration of residual and subsequent rectal polyps.

(4) Subtotal colectomy and rectal mucosectomy with or without a temporary diverting ileostomy and an endorectal pouch reservoir.

The ideal surgery for a benign disease with an inevitable malignant transformation is prophylactic resection of all potentially malignant tissue. If this ideal is fulfilled by proctocolectomy, aggressive pelvic dissection may leave the patient with serious sequelae (neurogenic bladder, male impotence, female infertility.) On the other hand, leaving the rectum in place requires perseverant monitoring of residual and subsequent rectal polyps, with no permanent assurance to avoid rectal carcinoma.

Regression of residual rectal polyps, in terms of number and size, has been described after ileoproctostomy with or without the use of NSAIDs (Celecoxib, Sulindac or Indomethacin.)

### Complications and prognosis

If left untreated patients with FAP have a median life expectancy of 42 years. It is true that colectomy extends such a short life expectancy, but since surgery only neutralizes the risk of colorectal carcinoma at

best, continuous monitoring of patients for developing extracolonic cancers cannot be overemphasized. (Table 2).

Desmoid tumors (diffuse mesenteric fibromatosis), concerning about 20% of patients with FAP, typically postcolectomy.

The cumulative risk for developing extracolonic cancer, mostly periampullary tumors, is 11% by age 50 years and 52% by 75 years.

Table 2 - Lifetime Risk of Extracolonic Cancer in FAP.

Site	Type of Cancer	Risk of Cancer
Small intestine: duodenum or periampulla	Carcinoma	4-12%
Small intestine: distal to the duodenum		Rare
Stomach	Adenocarcinoma	0.5%
Pancreas		~2%
Thyroid	Papillary thyroid carcinoma	~2%
CNS	Usually medulloblastoma	<1%
Liver	Hepatoblastoma	1.6% (children <age 5 years)
Bile ducts	Adenocarcinoma	Low, but increased
Adrenal gland		

### CASE REPORT

#### History and physical exam

13 y.o. male patient with a known family history of FAP and colorectal cancer. (Figure 1).

The patient sought medical attention because of recurrent abdominal pain and diarrhea over the last 4

months. The physical exam outlined a non-specific diffuse abdominal pain, both spontaneous and on palpation, with no palpable abdominal mass and no clinical sign of peritoneal involvement. Likewise, there was no palpable mass on rectal examination and no bleeding.

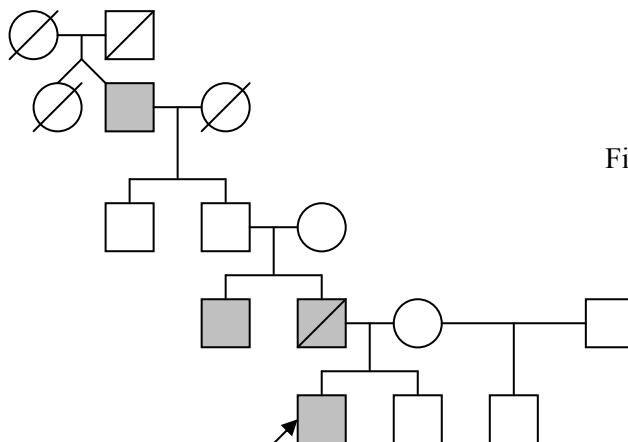
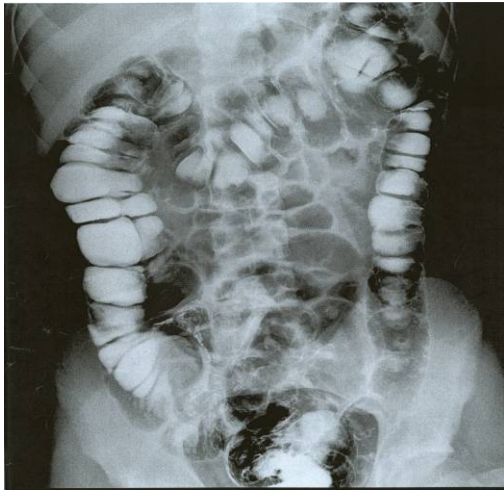


Figure 1 - Pedigree analysis.

The patient sought medical attention because of recurrent abdominal pain and diarrhea over the last 4 months. The physical exam outlined a non-specific diffuse abdominal pain, both spontaneous and on palpation, with no palpable abdominal mass and no clinical sign of peritoneal involvement. Likewise, there was no palpable mass on rectal examination and no bleeding.



#### Lab studies

CBC – normal counts.

Proteinemia – normal value.

Alpha-fetoprotein (AFP) – matched value for age.

#### Imaging studies

- Barium study of the abdomen showed multiple polyps disseminated throughout the colon. (Figure 2)

Figure 2. Barium study - multiple polyps disseminated throughout the colon.

- Colonoscopy detected hundreds of sessile polyps involving the entire colon extending from the rectum up to the cecum and hence establishing the diagnosis of FAP.

- Upper G.I. tract endoscopy revealed no gastric or duodenal tumors.

- Abdominal CT scan detected no extracolonic involvement and no desmoid tumors.

#### Histology

Biopsic polypectomy of 2 lesions was performed during colonoscopy. The first fragment was described as being a hyperplastic adenomatous tubular

polyp with minimal dysplasia, and moderate fibrosis with lymphoplasmocytic infiltrate of the chorion. The second fragment turned out to be an adenomatous tubulo-villous polyp with minimal dysplasia.

#### Surgical treatment

A prophylactic subtotal colectomy and ileoproctostomy with intraoperative diathermy of the residual polyps seemed to be the ideal procedure for this case. Avoiding to sacrifice the rectum was a satisfactory option because the patient had few rectal polyps and the concern about keeping a near-normal bowel movement pattern. (Figure 3-4)



Figure 3 – Colon subtotally resected.

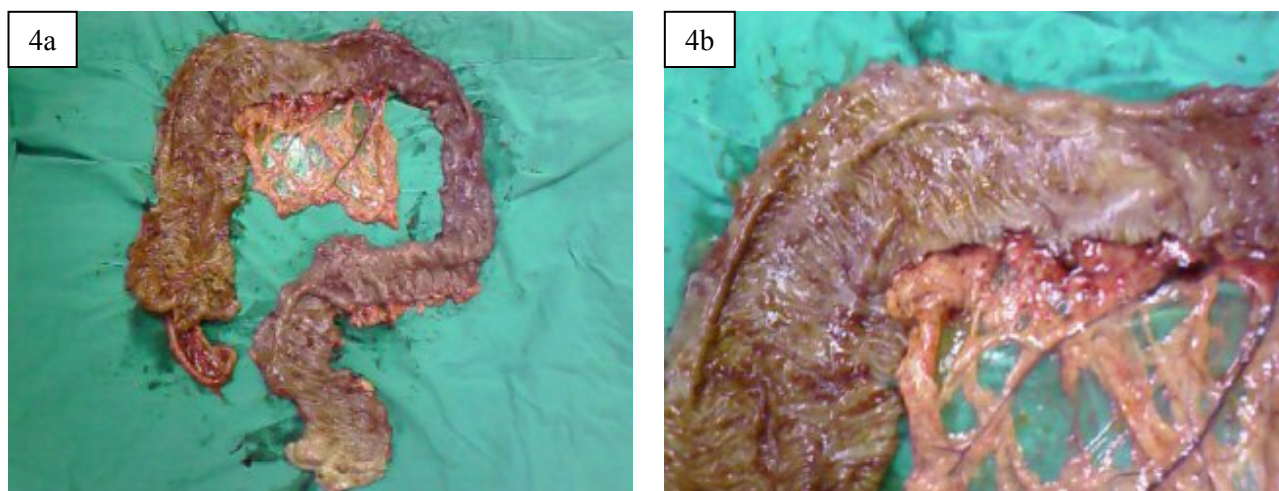


Figure 4a-4b – Resected colon was sectioned longitudinally to reveal multiple adenomatous tubulo-villous polyps involving the entire length of the colon.

#### Long-term management

1. Complete physical exam every year.
2. Stool blood testing every year.
3. Upper endoscopy at least every 4 years.
4. Flexible sigmoidoscopy every 6 months for monitoring and diathermy of residual and subsequent rectal polyps.
5. Intrarectal administration of 50 mg of indomethacin suppository once or twice daily to control the rectal remnant polyps.

#### References

1. Arensman R.M, Bambini D.A, Almond P.S, Pediatric Surgery, Landes Bioscience, 2000, 44:199-200, 52:234-236.
2. Ashcraft Keith W, Holder Thomas M, Pediatric Surgery, Second Edition, W.B. Saunders Co, ? 458-460.
3. Braun J, Dormann A, Clinical Guide Internal Medicine, 9<sup>th</sup> Edition, Urban und Fischer Verlag, 2003.
4. Goldman L, Bennett J. C, Cecil Textbook of Medicine, 21<sup>st</sup> Edition, Volume 1, W.B. Saunders Co, 2000, 139: 741-743.
5. Harold E, Calne R, Watson C, General Surgery, 9<sup>th</sup> Edition, Blackwell Science Ltd, 1998, 207.
6. Keiji H, Hideaki I, Keiichi O, Regression of Rectal Polyps by Indomethacin Suppository in Familial Adenomatous Polyposis, Report of two cases, Journal of the Diseases of Colon and Rectum, Volume 37, Number 9, September 1994, 943-946.
7. Kumar P, Clark M, Kumar & Clark Clinical Medicine, 5<sup>th</sup> Edition, W.B.Saunders, 2002, 6:315-317.
8. Passarge E, Color Atlas of Genetics, 2<sup>nd</sup> Edition, Thieme, 2001, 326-327.
9. Russel R.C.G, Williams N.S, Bulstrode C.J.K, Bailey and Love's Short Practice of Surgery, 23<sup>rd</sup> Edition, Arnold International Students Edition, 2000, 57: 1048-1049.
10. Solomon C, Burt R.W, APC-Associated Conditions, geneclinics.org, Oct 21, 2008.
11. Wehbi M, Familial Adenomatous Polyposis, emedicine, Aug 22, 2006.

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## PARTICULAR EVOLUTION OF GIANT COMPRESSIVE PERIRENAL HAEMATOMA - CASE REPORT

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### Abstract

The development of a perirenal hematoma is rare and primarily the result of trauma, malignancy, or a connective tissue disease. Spontaneous perirenal haematomas are relatively rare and its diagnosis requires the absence of recent instrumentation, surgery or trauma. We report a particular case involving the development of a giant compressive perirenal haematoma in the absence of a major trauma history.

**Keywords:** compressive perirenal hematoma, nephrectomy

### Introduction

Spontaneous retroperitoneal hemorrhage with secondary haematoma is an uncommon condition that occurs because of bleed either from the kidney or less often from adjacent retroperitoneal structures. The most common renal causes being angiomyolipoma, and renal cell carcinoma. Vascular diseases such as polyarteritis nodosa, renal artery aneurysm, infections of kidney such as cortical abscess, pyelonephritis and renal cysts are occasional etiologic factors. Adrenal haemorrhage is seen with severe stress conditions as sepsis, burns or trauma. Patients with pheochromocytoma, adrenal carcinomas, cortical adenomas also contribute as causes of retroperitoneal haemorrhage.

Clinically, these patients have variable presentation depending on degree and duration of bleed. Nausea, vomiting, low grade fever and a decreasing haemoglobin are common findings. Mild flank and upper abdominal discomfort in case of minimal bleed to patients presenting in shock with oliguria in case of massive blood loss. Urine examination is frequently normal.<sup>[1,2]</sup> Ultrasound is a rapid non-invasive test to localize the haematoma extent and probably look at primary pathology. Subcapsular haematomas do not reach as large a size as when bleeding is into the perinephric space,

because of the tamponade effect of the renal capsule. This tamponade effect is not possible with distensible Gerota's fascia and hence the perinephric haematoma may attain very large size.<sup>[3]</sup> CT scan in case of subcapsular haemorrhage will demonstrate the haematoma confined by renal capsule with parenchymal flattening. With haemorrhage in to the perinephric space, the CT scan will reveal an abnormal soft tissues density with displacement, compression or obscurity of normal retroperitoneal structures. As the haematoma ages, the density may decrease, while contrast enhancement of a subcapsular or perinephric haematoma does not occur unless active bleeding is taking place.<sup>[4]</sup> CT scan remains the gold standard not only to diagnose the cause of bleed but also to exactly localize the extent of haematoma.

Flattened renal parenchyma compressed by haematoma is clearly seen in extracapsular haematomas. Capsular arteries remain close to the capsule in subcapsular haemorrhage. In perinephric haematoma, capsular arteries are displaced away from the capsule.<sup>[5],[6],[7]</sup>

A variety of pediatric perirenal/renal masses may be differentiated in the first time from kidney tumors on the basis of their clinical and imaging features.

Wilms tumor is distinguished by vascular invasion and displacement of structures and is bilateral in approximately 10% of cases. Nephroblastomatosis occurs most often in neonates and is characterized by multiple bilateral subcapsular masses, often associated with Wilms tumors. Angiomyolipoma frequently contains fat and is associated with tuberous sclerosis. Renal cell carcinoma is unusual in children except in association with von Hippel-Lindau syndrome and typically occurs in the second decade. Multilocular cystic renal tumor is suggested by a large mass with multiple cysts and little solid tissue. Renal medullary carcinoma occurs in patients with sickle cell trait or hemoglobin SC disease and manifests as an infiltrative

mass with metastases. Metanephric adenoma lacks specific features but is always well defined. Renal lymphoma is characterized by multiple homogeneous masses, often with associated adenopathy.

### Case report

An 8 year old boy presented at the emergency department complaining of significant left-sided flank pain. Patient's history revealed a small lumbar trauma suffered 2 weeks ago. Vital signs showed a pulse rate of 88 beats per minute, blood pressure of 110/80 mm Hg, and temperature of 36.4 °C. Clinical examination

revealed abdominal pain in the left upper and medium quadrant.

Laboratory analysis of the patient's blood was normal, except for leukocytosis (count of 19,890/ml). Urine analysis revealed presence of albumin, numerous RBC/high-powered field and 1-3 WBC/ high-powered field, high levels of vanil-mandelic acid.

Renal ultrasonography: the ecographic aspect suggested a mixed left renal tumor with secondary hydronephrosis (fig. 1,2).

The right kidney had normal echographic aspect.

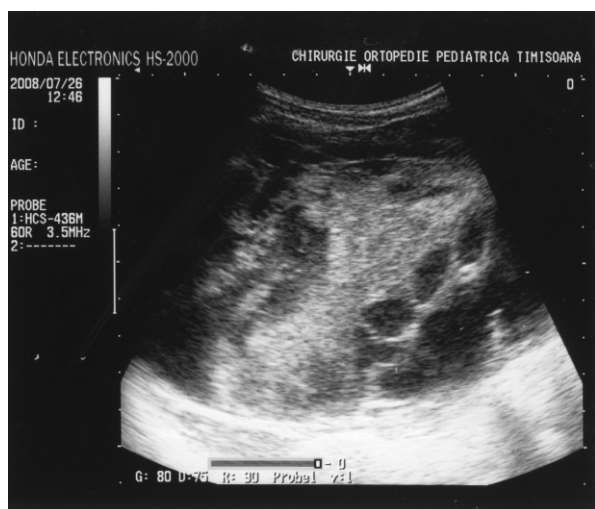


Fig. 1. Echographic aspect of left kidney.

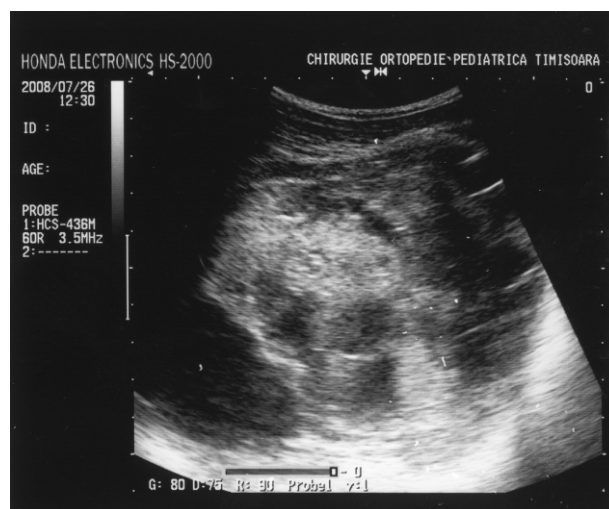


Fig. 2. Echographic aspect of left kidney.

Intravenous pyelography in the early excretory phase demonstrated normal excretion of contrast substance from the right kidney and delayed excretion of contrast material from the left kidney. Intravenous pyelography in the later excretory phase: contrast substance begins to accumulate in the dilated calyces

of the left kidney, no contrast substance was seen in the left ureter on this or other images, suggesting hydronephrosis caused by uretero-pelvic junction obstruction. The left kidney demonstrated relatively increased renal length in comparison with the right kidney.



Fig. 3. Intravenous pyelography - early excretory phase.



Fig. 4. Intravenous pyelography - latent excretory phase.

All these investigations suggested a mixed left renal tumor with secondary hydronephrosis, stage IV.

Abdominal MRI revealed left giant perirenal hematoma with secondary hydronephrosis stage IV, reduced renal parenchyma of 3 mm. (fig.5,6)

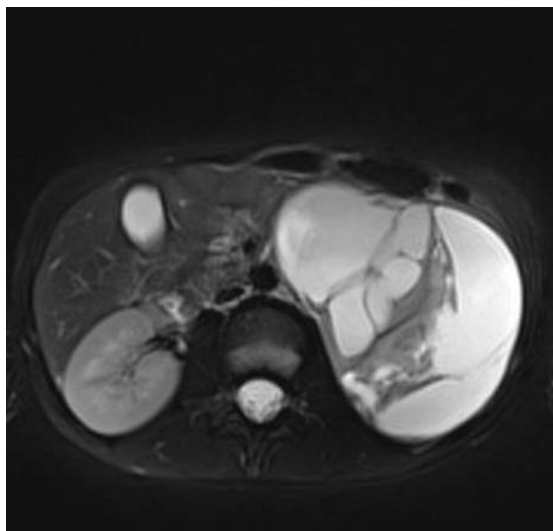


Fig. 5. Abdominal MRI.

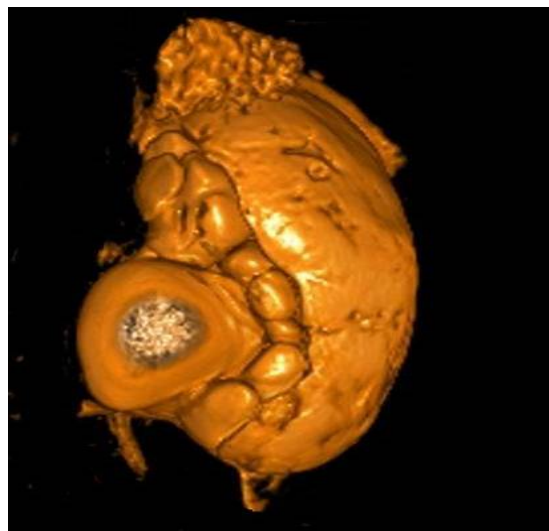


Fig. 6. Abdominal MRI (reconstruction).

*Treatment* in this case consisted of great median laparotomy with peritoneal organs exploration. The intraperitoneal organs showed no macroscopic pathologic modifications.

By drifting the left colon, an entry into the retroperitoneal space was made. The presence of a giant tumor (with clear margins, encapsulated and mixed structures) of the left kidney was evidenced, having on its surface well defined vasculature. Based on the results of the laboratory investigation correlated with the intra-operative macroscopic aspect, we performed a radical left uretero-nephrectomy within the oncological safety margins (left adrenalectomy, excision of peri-renal fatty mass and renal hilar lymph nodes). The macroscopic aspect of the excised tumor revealed some areas of haematoma alongside areas of “blood with no clots”

Post-surgery care consisted in administration of antibiotics (cephtriaxone), solution of parenteral nutrition (glucose, aminoacids), electrolytes, antalgics and non-steroids anti-inflammatory drugs. The evolution was favorable with swift digestive tolerance and regaining of the intestinal transit.

*Histopathological findings:* The post operative histopathologic results confirmed the notion of the MRI findings: perirenal haematoma.

## Discussion

The development of a perirenal haematoma is rare and primarily the result of trauma, malignancy, or a connective tissue disease.

In our case the mechanism of production of compressive perirenal haematoma remain unidentified. The absence of major trauma, clinical findings and the echographic aspects of left kidney has raised the suspicion of a Wilm's tumor. MRI showed left massive perirenal haematoma, left pelvicalyceal dilatation and a thinned renal cortex suggestive for a compressive perirenal haematoma with severe hydronephrosis. The ureter was collapsed. The hydronephrotic modifications could be produced by the progressive perirenal haematoma with compression of the upper portion of ureter. This massive perirenal haematoma was not communicating with the subcapsular parenchyma or the collecting system. The contralateral kidney was normal. In these circumstances we could not determine if perirenal haematoma was a result of a minor injury to a hydronephrotic kidney or if it was a result of a minor injury to an already existing arterio-venous renal malformation and the hydronephrosis was secondary to it.

Intraoperative we found giant perirenal haematoma which compressed the left kidney and massive destruction of kidney tissue which imposed nephrectomy. Because this patient did not show signs of acute hemorrhage, the haematoma probably appeared gradually with progressive compression and disturbances of renal blood and secondary injury of nephrosclerosis.

Histopathological examination postnephrectomy not detected tumoral changes, congenital or vascular malformations.

### Conclusion

1. The perirenal hematomas are usually a result of a severe injury and have an obvious clinical manifestation.

2. In the described case, the injury was insignificant and without any clinical signs.
3. Preoperative differential diagnosis was suggestive for perirenal haematoma or mixed renal tumor, with the destruction of the renal parenchyma (limited to 3 mm).
4. The intraoperative aspect was suggestive for a mixed renal tumor, reason for which a left nephrectomy (within oncological safety margins) was performed.
5. The positive diagnosis of left giant, compressive perirenal haematoma with secondary hydronephrosis and nephrosclerosis was established examining the macro and microscopic aspects of the excised mass.

### References

1. Machuca SJ, Julve YE, Galacho BA et al. Spontaneous retroperitoneal haematoma - Our experience. *Acias Urologicas Espinolas* 1999;23:43.
2. Brkovic D, Moehring K, Doersam J et al. Aetiology, diagnosis and management of spontaneous perirenal haematomas. *Euro Urol* 1996; 29: 302-307.
3. Kendall AR, Senayba, Coll ME. Spontaneous subcapsular renal haematoma: Diagnosis and management. *J Urol* 1988; 139: 246-250.
4. Sagel SS, Siegel MJ, Stonely RJ, Jost RG. Detection of retroperitoneal haemorrhage by computed tomography. *Am J Roentgen* 1977; 129: 403.
5. Meyers MA, Whalen JP, Evans JA. Diagnosis of perirenal and subcapsular masses: anatomic-radiologic correlation. *Am J Roentgen* 1974; 121: 523-538.
6. Pollack HM, Popley GL. Roentgenographic manifestations of spontaneous renal haemorrhage. *Radiology* 1974; 110: 1.
7. Gupta N P, Karan S C, Aron M, Pawar R, Ansari M S. Spontaneous perirenal haematoma: A case report and review of literature. *Urol Int.* 2000; 64(4): 213-5.

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The manuscript must be in English, typed single space, one column on A4 paper, with margins: top – 3 cm, bottom – 2,26 cm, left – 1,5 cm, right – 1,7cm. A 10-point font Times New Roman is required.

The article should be organized in the following format: Title, Names of all authors (first name initial, surname), Names of institutions in which work was done (use the Arabic numerals, superscript), Abstract, Keywords, Text (Introduction, Purpose, Materials and Methods, Results, Discussions and/or Conclusions), References, and first author's correspondence address.