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AN ASSESSMENT OF REASONS FOR PSYCHIATRIC ILLNESS AS PERCEIVED BY CLOSE RELATIVES OF PSYCHOTIC INPATIENTS *

Gülören Ünlüoğlu** • Rüçhan Kartallar*** • Ladiper Gürakar****

SUMMARY

Objective: *Psychiatric patients in Turkey usually live with their families and both the patients and their relatives are influenced by each other. To investigate and evaluate the relatives' thoughts and opinions about the reasons for psychiatric illness has gained much importance, since the relatives are also included in the treatment programs for the patient's well-being. To reach the relatives may become easier if we know what they think and know. Therefore , we raised the following questions for investigation: 1) What is the order of importance in relation to reasons for mental illness? and 2) Is there a difference between the males' and females' opinions?* ***Method:*** *A form with 14 possible reasons for mental illness was prepared and given to 60 male and 60 female relatives of psychotic inpatients to be filled out in the order of importance. The difference of opinions between the sexes was assessed with chi-square test.* ***Results:*** *The order of importance selected by 120 subjects in relation to reasons for mental illness was: "the difficulty in interpersonal relationship," "financial difficulties," "environmental pressure" and "death of a loved one." For males, "evil eye" was the second reason for illness, whereas for females the same reason was surprisingly in the 12th row.* ***Conclusion:*** *The findings of this study encouraged us not only in planning other more detailed studies but also working with relatives of psychotic patients more closely.*

Key Words: *Relatives of psychotic patients, reasons for mental illnes*

To be hospitalized with the diagnosis of "schizophrenia" is indeed very dramatic and traumatic not only for the patient but also for his/her family and relatives. Therefore, the doctor or the members of the therapy team should deal with both the patient and the family.

In Turkey when a patient is hospitalized, usually the relatives come to see him/her during most of the visiting hours and they constantly and continuously want to be in touch with the doctor to find out how their patient is doing. The second question they ask, if not the first, is: "What caused his/her illness?" Although the question is simple and much is known about it, the answer is very complex and all of what should be known is not yet complete and certain.

When we review the etiology of schizophrenia, we see Arieti (1), Day and Senrad (2), Kaplan et al. (3), Angermeyer and Klusman (4) mentioning the importance of traumas; infections; intoxications; acute and chronic brain syndromes; tumours; imbalance of electrolytes; various deprivations and external life events; developmental stages in human life; age, gender and to perform its related roles as males and females; to be married, divorced or single; having their impacts in peoples' lives contributing to their illness. Weisman and Paykel (5), Ünal (6), Periman and Karen (7) emphasize the importance of loneliness as one of the causes of mental illness. Race; job/profession; dependencies or abuse of alcohol and drugs/substances; IQ; constitution; heredity and genetics could play some role on mental illness (8, 3). According to Beiser (9) Meyer and Bean (10), climate, weather conditions, seasons; living in rural or urban areas; migration; war; natural disasters; socioeconomic and cultural events affect peoples' mental health. In the last decades the quality of life a child experiences, has gained much importance and various combinations of family network like "scapegoating," "double bind", "level of expressed emotion" have been discussed (11, 12, 13)

* This article was presented as a poster at the Xth World Congress of Psychiatry, 23-28 August 1996, Madrid, Spain

** Prof. of Psychiatry in Medical School of Ankara University

*** Psychologist, Ph.D. in the Department of Psychiatry, Medical School of Ankara University

**** Social Worker, M.S. in the Department of Psychiatry, Medical School of Ankara University

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as reasons for mental illness. Engel (14), Melges (15), Kaplan et al. (3) mention a bio-psycho-social model, intrapsychic conflicts and the environment's importance on the subject.

In the last 3 decades, the relatives of psychotic patients have gained importance in making the patients' treatment plans because of various reasons: 1) To get information concerning the patients; 2) To get a chance to observe their interaction with family members; 3) To understand and support them if they need it since they also get their share of distress, sorrow or anger; 4) To find out what they know about mental illness and 5) To prepare them for the discharge since the patient will return home. If a more suitable atmosphere for the patient's well being could be created, the relapses could be diminished or even prevented.

Therefore, since some time, working with relatives gained momentum and started in the West and scarcely all over the world. In fact, a study performed in Greece by Alivistos and Lyketsos (16) showed that the relatives of psychotic patients believe that psychological factors are the causes of mental illness. They stated that 26% of all relatives find intrafamilial events and 26% traumatic events outside the family and 15% genetics as the reasons for mental illness. Rose (17) studied the relatives of patients who had long stays in Veterans Hospitals and found some differences between the thoughts of close relatives and relatives of far kinship. While close relatives thought external or somatic factors as the causes for illness, the others felt "lack of love, trust and understanding" was the cause.

OBJECTIVES OF THE STUDY

Our team found it important to search the answers of the following questions: 1) What the relatives of our psychotic patients think and see as the reasons of mental illness; 2) What would be the order of importance according to them if a list of possible reasons could be prepared and given for filling, and 3) Would there be any difference between what men and women think?

METHOD

This is a descriptive study which was done in the psychiatry department of Ankara Medical School with the relatives of psychotic inpatients. During the first 3-5 days of their admission, patients' relatives (usually a family member) are interviewed by the social worker who obtains the necessary information about the patients. This interview would be the best time to reach the relatives.

Subjects:

The sample consisted of 120 relatives, 60 of which were males and 60 were females. They were accepted for the study regardless of their ages, marital status, educations, professions and their kinships to the patients.

Data Collection:

Following the interview, each subject was given a form including a list of possible reasons for mental illness to be marked in the order of importance by writing 1, 2, 3 ... on the blank space in front of the cause. The following is the list of 14 possible causes for mental illness:

... Disturbance/difficulty in interpersonal relationships.
... Economic/financial difficulties.
... Environmental pressures.
... Infectious/other somatic diseases.
... Accidents/traumas.
... Load of work/studies.
... Evil eye's effect (cultural belief among some people).
... Black love (a concept meaning hopeless and deep love in Turkish culture).
... Loss of a loved one.

... Marriage	... Beating
... Divorce	... Heredity
... Unemployment	...Other: .....

Instrument:

This list was prepared by the study team after the review of literature on etiology of schizophrenia as mentioned in the introduction, and interaction of 10-15 years with relatives of psychotic patients including their impressions plus some cultural points of view. The possible causes were expressed as-simple and understandable - as possible.

Processing:

The sums of responses given by 60 male and 60 female subjects in the order of importance concerning each cause for mental illness were recorded with numbers and percentages (Table 1). However, since the number of responses were very few following the 7th row, they were reflected under the heading "others."

The totals of all responses given by the subjects to each cause of mental illness were tabulated and com-

parison between male and female subjects was made to find the statistically significant difference (Table 2). For the assessment chi-square test was used.

FINDINGS

Distribution of all responses to 14 causes of mental illness in accordance with the order of importance given by 60 male and 60 female relatives of psychotic inpatients is reflected in Table 1.

Distribution of the totals of male and female responses given to each cause of mental illness is reflected in Table 2.

Table 1: Response Distribution for Each Mental Illness Cause Given by Psychotic Inpatients' Relatives

1. Disturbance/Difficulty in Interpersonal Relationships

OI*	Male	%	Female	%	Total
1	20	33.3	41	68.8	61
2	10	16.6	6	10.0	16
3	3	5.0	4	6.6	7
4	2	3.3	1	1.6	3
Total	**35**	**58.2**	**52**	**87.0**	**87**

2. Economic/Financial Difficulties

OI	M	%	F	%	Total
1	6	10.0	4	6.6	10
2	13	21.6	16	26.6	29
3	4	6.6	6	10	10
4	1	1.6	5	8.3	6
5	1	1.6	2	3.3	3
6	0	-	1	1.6	1
Total	**25**	**41.4**	**34**	**56.4**	**59**

3. Environmental Pressures

OI	M	%	F	%	Total
1	3	5.0	1	1.6	4
2	4	6.6	13	21.6	17
3	9	15.0	13	21.6	22
4	2	3.3	3	5.0	5
5	0	-	3	5.0	3
6	1	1.6	0	-	1
7	2	3.3	0	-	2
Total	**21**	**34.8**	**33**	**54.8**	**54**

4. Infectious/Somatic Diseases

OI	M	%	F	%	Total
1	1	1.6	1	1.6	2
2	2	3.3	1	1.6	3
3	2	3.3	6	10.0	8
4	2	3.3	2	3.3	4
5	3	5.0	2	3.3	5
6	2	3.3	3	5.0	5
7	1	1.6	3	5.0	4
Others	2	3.3	1	1.6	3
Total	**15**	**24.7**	**19**	**31.4**	**34**

* Order of Importance

5. Accidents/Traumas

OI	M	%	F	%	Total
1	3	5.0	2	3.3	5
2	1	1.6	0	-	1
3	3	5.0	1	1.6	4
4	0	-	7	11.6	7
5	1	1.6	1	1.6	2
6	1	1.6	0	-	1
7	0	-	2	3.3	2
Others	3	5	2	3.3	5
Total	**12**	**19.6**	**15**	**24.6**	**27**

6. Load of Work/Studies

OI	M	%	F	%	Total
1	6	10.0	4	6.6	10
2	6	10.0	4	6.6	10
3	0	-	5	8.3	5
4	1	1.6	6	10.0	7
5	0	-	5	8.3	5
6	1	1.6	0	-	1
7	0	-	1	1.6	1
Others	1	1.6	2	3.3	3
Total	**15**	**24.7**	**29**	**47.9**	**44**

7. Evil-eye's Effect

OI	M	%	F	%	Total
1	2	3.3	0	-	2
2	1	1.6	2	3.3	3
3	2	3.3	2	3.3	4
4	2	3.3	1	1.6	3
5	9	15.0	3	5.0	12
6	10	16.6	1	1.6	11
10	0	-	1	1.6	1
Total	**26**	**43.1**	**10**	**16.4**	**36**

8. Black-Love

OI	M	%	F	%	Total
1	3	5.0	4	6.6	7
2	2	3.3	2	3.3	4
3	0	-	4	6.6	4
4	1	1.6	2	3.3	3
5	2	3.3	5	8.3	7
6	0	-	1	1.6	1
7	1	1.6	1	1.6	2
Others	4	6.6	3	4.9	7
Total	**13**	**21.4**	**22**	**36.3**	**35**

9. Loss of a Loved One

OI	M	%	F	%	Total
1	1	1.6	4	6.6	5
2	5	8.3	4	6.6	9
3	4	6.6	5	8.3	9
4	3	5.0	6	10.0	9
5	3	5.0	5	8.3	8
6	2	3.3	0	-	2
7	1	1.6	1	1.6	2
Others	3	5.0	2	3.3	5
Total	**22**	**39.4**	**27**	**44.7**	**49**

10. Marriage

OI	M	%	F	%	Total
1	1	1.6	1	1.6	2
2	0	-	0	-	0
3	1	1.6	1	1.6	2
4	2	3.3	0	-	2
5	0	-	1	1.6	1
6	0	-	2	3.3	2
7	0	-	1	1.6	1
Others	2	3.3	3	5.0	5
Total	**6**	**9.8**	**9**	**14.7**	**15**

11. Divorce

OI	M	%	F	%	Total
1	0	-	0	-	0
2	1	1.6	1	1.6	2
3	1	1.6	1	1.6	2
4	1	1.6	1	1.6	2
5	4	6.6	1	1.6	5
6	0	-	3	5.0	3
7	1	1.6	4	6.6	5
Others	3	5.0	6	10.0	9
Total	**11**	**18.0**	**17**	**28.0**	**28**

12. Unemployment

OI	M	%	F	%	Total
1	4	6.6	0	-	4
2	1	1.6	4	6.6	5
3	5	8.3	2	3.3	7
4	3	5.0	5	8.3	8
5	1	1.6	1	1.6	2
6	1	1.6	4	6.6	5
7	2	3.3	3	5.0	5
Others	2	3.3	6	10.0	8
Total	**19**	**31.3**	**25**	**41.4**	**44**

13. Beating

OI	M	%	F	%	Total
1	1	1.6	0	-	1
2	1	1.6	1	1.6	2
3	1	1.6	2	3.3	3
4	3	5.0	2	3.3	5
5	0	-	3	5.0	3
6	1	1.6	1	0.6	2
7	1	1.6	2	3.3	3
Others	13	21.5	7	11.6	20
Total	**21**	**34.5**	**18**	**28.7**	**39**

14. Heredity

OI	M	%	F	%	Total
1	1	1.6	1	1.6	2
2	1	1.6	4	6.6	5
3	1	1.6	2	3.3	3
4	0	-	0	-	0
5	2	3.3	2	3.3	4
6	1	1.6	2	3.3	3
7	1	1.6	0	-	1
Others	3	5.0	4	6.6	7
Total	**10**	**16.3**	**15**	**24.5**	**25**

Table 2: Distribution of Sums of Responses for Each Cause

Causes of Mental Illness	M	%	F	%	Total	%	x^2 (Df *=1)
Difficulty in relating	35	58.2	52	87.0	87	72.5	3.322
Economic/Financial Difficulties	25	41.4	34	56.4	59	49.1	1.373
Environmental Pressures	21	34.8	33	54.8	54	46.6	2.667
Infectious/Somatic Diseases	15	24.7	19	31.4	34	28.3	0.471
Accidents/Traumas	12	19.6	15	24.6	27	22.5	0.333
Work/Study Load	15	24.8	29	47.9	44	36.6	4,455**
Evil eye effect	26	43.1	10	16.4	36	30.0	7.111***
Black Love	13	21.4	22	36.3	35	29.1	2.314
Loss of loved one	22	36.3	26	44.1	48	40.0	0.333
Marriage	6	9.7	9	14.6	15	12.5	0.6
Divorce	11	17.8	17	27.9	28	23.3	1.286
Unemployment	19	31.2	25	41.3	44	36.6	0.818
Beating	21	34.5	18	28.1	39	32.5	0.231
Heredity	10	16.1	15	24.5	25	20.8	1.000
Total	**252**		**323**		**575**		

* Degree of freedom**p<0.05***p<0.01

DISCUSSION

The findings of this study show that there were 575 responses from 60 male and 60 female relatives of inpatients to 14 alternatives for mental illness. (Table 2) The number of responses female relatives gave was higher than those of male subjects. Males gave 252 whereas females gave 323 responses. The average of male responses was 4.2 and females was 5.2. This finding is not surprising because in our culture according to our observations, females are more loquacious and males are more activity oriented.

The two possible causes for mental illness which they agreed upon in the order of importance were "difficulty of interpersonal relationships "as the first, and "marriage" as their last choice. The importance of interpersonal relationships and family relationships are long being studied and various recommendations are given to improve the relations and communications (18, 19, 20, 21). Why "marriage" was the last choice for mental illness is understandable, since among the traditional groups marriages are considered sacred and at times people believe that a psychiatric patient can get better or well if he/she could get married. "Shall we have him/her married so that he can get well?" is one question professionals hear very often from the relatives.

"Financial difficulties" is often the second choice of female subjects whereas "effect of the evil eye" is the second choice of men. The difference between sexes for "evil-eye" is statistically significant (p<0.01). For women this alternative is in the 12th row in the order of importance. Male subjects of this study were found more educated, in comparison to females, which is indicative of males having more opportunity to encounter realistic thinking. Demographic characteristics of this group were presented in another study (6). Therefore, one could have expected the females' response to this alternative to be higher. Since the opposite is true, one thinks "Could it be possible that the men of our society are more traditional?" or "Could this be a way of saying that they are not responsible for their relative's illness. It is the evil-eye." In a study performed by Rose in 1959 (17), close relatives tried to connect the illness to external factors. Nevertheless, schooling is not enough to change some beliefs even if they don't seem rational. This topic needs further studying.

Another difference between males and females which was statistically significant (p<0.05) was "Load of work/studies" (Table 2). Work and study are outside the home. Still, only 24% of males but 50% of females accepted this as a possible cause for mental illness. Maybe females are more sensitive to their loved one's getting tired or exhausted, or could it be that this time they are the ones externalizing the issue? At any rate, this also needs further studying.

Although it is not statistically significant, the sum of the responses given to "beating" is interesting. For men it is in the 5th order of importance whereas for women it is only in the 9th row (Table 2). In Turkish society, usually women get the "beating" and man is the aggressor. Recently the media has taken the sub-

ject of battered women in hand to prevent or at least to diminish it. Could it be that women accept it as a cultural norm or have they become so desensitized that they don't even mention it? Why do men respond more? This is also another question needing further studying.

"Environmental pressures" is in the 3rd order of importance for women and 5th for men and "financial difficulties" is in the 2nd row for women and 3rd for men (Table 2). These are close findings although one could expect "financial difficulties" and "environmental pressures" to have more impact on male responses since they are the ones who face these difficulties more in comparison to women.

However, our findings are parallel to findings of Engel (14), Arieti (1), Freedman et al. (22), Kaplan et al. (3). "Profession and Heredity" results are like Kety's (8) and Ünal's (23).

Another topic of interest in "Black-Love" which also reflects a cultural belief. The findings show that both males and females have agreed on its being in mid rows of importance, 7th cause for females and 8th cause for males which could be considered somewhat important.

This study shows that to investigate the thoughts and beliefs of relatives of mental patients in important. The more the professional know about the family system, the better they can understand their relationships, interactions and dynamics between members and they can make more effective treatment plans.

CONCLUSION

This study indicates that further research encompassing socio-cultural and economic aspects of the subject must be taken in hand in order to be able to help the patients and their relatives since schzophrenia is also a family disease like cancer (24). And also professionals should try to work more closely with relatives of psychotic patients.

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DAY HOSPITAL PRACTICES IN PSYCHIATRY CLINIC OF UNIVERSITY OF ANKARA*

Gülören Ünlüoğlu** • Aykut Özden*** • Çiğdem Soykan**** • Züleyha İdil*****

SUMMARY

Objective: *Although Day Hospital (DH) approach is extensively used and its effectiveness has long been accepted for various psychiatric disorders all over the world, in Turkey it has been used only within the last decade and publications are rather scarce. Since Psychiatry Department of Ankara Medical School is one of the first training centers in Turkey where DH approach is used as a treatment modality, we would like to present our DH experience of six years.* ***Method:*** *This study was performed as a retrospective file research of the 81 patients admitted to our DH between 1988 and 1994. Their recent conditions related to relapse were investigated from the follow up files.* ***Results:*** *The findings show that in a follow-up period of 2-6 years, DH patients relapsed less than the ones who did not enter the DH program, and it was even better for the patients who received group therapy after their discharge from DH.* ***Conclusion:*** *These results and our observations led us to conclude that, DH approach is an effective and convenient modality, especially for the chronic psychotic patients, with the inclusion of their families in the program.*

Key Words: *Day Hospital, psychosis, rehabilitation*

Deinstitutionalization movement of the '60s has pushed treatment of patients with schizophrenia into a post hospital era (1). In recent years, treatment of these chronic patients became primarily brief hospitalization, crisis intervention and medication, followed by inconsistent and inadequate psychosocial support services (2). On the other hand, with a growing economical crisis in most of the countries all over the world, long hospitalizations became out of fashion, and even, prohibited (3). Studies showing the effectiveness of brief hospitalization rationalized this trend firmly (3). However, as a consequence, a "revolving door syndrome" occurred; "admission-discharge-readmission", which is very demoralizing for the patients, their families and, naturally, the clinicians (2). Moreover, there are "the chronic psychosis (CP) patients" who are incapacitated in their interpersonal relationships. Neither brief, nor long hospitalizations seemed suitable for their needs. Day hospitals (DH), home based cares and community treatments are now trying to fill this gap (4).

DH approach in psychiatry is established long ago and it has been widely used in many psychiatric disorders, especially in CP, with success (5). DH is favored both by patients (6) and their families (7) and at the same time it is more economic for health care providers (8, 9). DH approach provides a valuable opportunity for those CP patients who need longer treatments. Clinicians who are skeptical on the efficacy of DHs, like Creed et al. (10, 11) reported a low readmission rate even for the *acutely* psychotic patients treated in DH.

In many psychiatry clinics CP patients are mostly followed in outpatient units, where a patient can not get time more than 30 minutes once a month or two. Knowing the main loss in CP is in their functioning, a treatment approach aimed at only in reduction of the acute psychotic symptoms will not cover all problem

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** Professor of Psychiatry, M.D., Department of Psychiatry, Unuversity of Ankara, Medical School
*** Psychiatrist, M.D., Department of Psychiatry, Unuversity of Ankara, Medical School
**** Psychologist, Ph.D., Department of Psychiatry, Unuversity of Ankara, Medical School
***** Psychiatric Nurse, Department of Psychiatry, Unuversity of Ankara, Medical School

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areas. CP patients frequently present problems like; difficulty in responsibility taking and initiative, lessening of self esteem, self confidence, self care and social competence, thus lowering their quality of life which medication alone would not be of great benefit. Sometimes such problems lead to further withdrawal and deterioration. DH approach has been introduced to stop and even to reverse this process. It is shown that, with DHs, not only psychopathology and relapse rate reduces, but also social functioning improves (12).

Psychiatry Clinic of University of Ankara, Medical School (UAMS) is one of the leading centers in DH practices with an eight years of experience. Here we would like to present our experiences on this field. We would also want to emphasize the role of continuous follow-ups on the prevention of relapses, even after the completition of the DH program.

DAY HOSPITAL PRACTICES IN PSYCHIATRY CLINIC OF UAMS

Psychiatry Clinic of UAMS is located on the eastern front of Ankara, as a separate building. We have several inpatient wards for various patient populations like psychotics, substance dependents and psychoneurotic patients, and three outpatient clinics for child, adolescent and adult patients respectively. DH program has been founded in 1988 as a part of the clinic, and its team has been formed by two psychiatrists (a professor and a resident), a psychologist, a social worker and two psychiatric nurses.

The program continues from September to the end of May and the patients attend for 3 days a week from 8.15 a.m. till 4.00 p.m.. On the fourth day, a previously planned social activity is carried out. The patients are referred from outpatient services or from inpatient wards, at the time of discharge. The DH team always evaluates the patients before admission to the program and uses the following selection criteria:

1) To be a residual CP patient who needs rehabilitation (However, we sometimes accepted non CP patients who also needed rehabilitation, like borderline personality disorder)

2) Not to have mental retardation or severe physical disability

3) To be a resident of Ankara or have a place to live/stay in Ankara

4) To have motivation for the program (even if limited)

5) To afford the program costs.

Patients who fulfill these criteria are included in the program and during the process, various patients can drop out or can be referred to the inpatient wards. We always try not to include new patients once the program reaches its halfway. We usually start with 12-14 patients and try to keep this number almost the same.

Our DH program included; group psychotherapy (45 minutes each, twice a week), individual psychotherapy (45-60 minutes, once a week), occupational therapy (60 minutes, three times a week) and music therapy sessions (45 minutes, twice a week). We also perform family counseling sessions and family meetings, where level of expressed emotions are explored and psychoeducation is emphasized (13). Also, social hours, where there are recreational activities, problem solving and discussion hours take place. Naturally, patients continue to take their medications which is considered a very important matter.

Our group psychotherapy sessions mostly follow supportive and interactional approach. All patients are included into the groups and the sessions are led by the resident psychiatrist, the psychologist and one of the nurses from the team. Main therapeutic factors observed in these groups are; universality, altruism, cohesiveness, instillation of hope and imparting information. Individual therapy sessions also follow the supportive-didactic approach, because these patients mostly have fragile egos and could not tolerate confrontation easily. As the cohesiveness between the patients and treatment team increases, and their strengths increased, we became more able to be confrontative and they became more tolerable and less defensive to these confrontations and criticism.

In the psychoeducation hours, we present information on psychiatric medication, psychiatric disorders and various other topics of interest. This hour is generally favored by the patients and it seems to increase the compliance to medication. In problem solving hours, we try to solve their daily problems altogether. Similarly, in the discussion hours we again try to solve problems together, but this time we present the problems or situations to them in terms of raising questions such as "What could be some of the ways of having someone do something we want ?" or "The ways to express positive and negative feelings". These techniques were aimed at improving alternative thinking and evaluation of solutions. Since we also use role playing and homework assignments we can call this

approach as cognitive-behavioral. We also used psychodrama in an informal way and we think that it could be implanted in rehabilitation programs with success.

Finally, patients arrange a social hour once a week, where they can sing, dance, tell jokes, ask puzzles and eat and drink fancy food. They also can visit each other's houses in groups, can go to movies together, and sometimes team members join them in these activities. These activities provide them the necessary social relations that they have excluded for some time and motivate them to plan, review and actualize programs diminishing their withdrawals.

In 1992 with growing experience, we as members of the therapy team, changed the name of our program from Day Hospital to Day Club, which was welcomed by our patients and perceived as having a promotion from "patient status" to "club member status". Then, we launched into a very structured, token economy system, which has been used by other clinicians perfectly, for a long time (14). In this system, patients collect tokens from various activities, behaviors and even for their grooming. They are seperated to different levels according to their performances and have different privileges. Although this approach may seem difficult for CP patients, our patients could comply with it successfully gaining considerable benefit from it.

On the other hand, we also witnessed the need for a post-rehabilitation activity, specially for the most withdrawn, troubled and unemployed patients or who had an undesirable social environment. After a successful rehabilitation program, it became obvious that most of the patients could be reintegrated back into their social lives, but with some patients, it was quite unlikely to reach this point with an eight months of therapy. Although some problems could be solved and various ones could be covered successfully, some problems never die. Therefore, we decided to follow up some of our selected patients with the use of group therapy sessions, which is used extensively in CP patients with success before (15). These groups were somewhat similar to ones they enter while in the DH program, except that they are performed once a week. We started with 12 patients from the 1992 group and until now had 17 patients, of whom 5 of them left the group or dropped out for various reasons (16).

Here we would like to present the general characteristics of our DH patients and compare them in regard to relapse prevention. We investigated the files of all patients (n=81) admitted to our DH program in its first 6 years, between 1988 and 1994. Their relapse rates are investigated from the follow up charts. The anti psychotic drug doses are expressed as chlorpromazine equivalents and the diagnosis of relapse was made using the DSM-IV criteria for schizophrenia (17). We only considered the relapse rates of schizophrenics and schizo-affective patients (n=73).

We also seperated the patients discharged from the DH program into two groups; one followed with weekly group therapy sessions, and the other in our outpatient service. Then, we compared their relapse rates with chi-square test.

RESULTS AND DISCUSSION

In the first 6 years 81 patients were admitted to the DH program, which makes 13.5 patients per year. The number of patients who had left the program before it ended was fairly low; 11 (13.6%) in 6 years. This finding may be due to careful selection of patients by the team and/or good compliance and cohesiveness of the patients.

Table 1 and 2 outline socio-demographic characteristics of the patients. The mean age of the subjects is 33. They are mostly singles (63%) and live with their parents (74.1). The number of male and female patients are close and their average year of education is 10.7. For their occupational characteristics, the majority of the patients are unemployed, among them housewives and young girls are included, who are followed by the civil servants and/or workers. As one can see only a few of our patients are living alone (2.5%). Their families do not leave them alone which may be in contrast to other western cultures forming an interesting support system which needs to be investigated further.

In Table 3, duration of illness, length of stay in the DH and the mean anti psychotic doses are outlined. They have a mean of 11.3 years of illness which indicates a long and chronic course and they use 478.4 mg / day anti psychotic (as chlorpromazine equivalent), which is rather high. Although neither we accepted, nor no one referred acute psychotic patients, it is obvious that our patients were not untroubled mild cases. Their mean length of stay in the DH was 135.7 days (4.5 months). Since the length of stay was shorter in the beginning years and with the addition of the early termination and drop outs, the mean dropped to 4.5 months instead of 7 or 8 months.

Table 4 reflects the diagnoses of DH patients. As we can see, most of the patients have CP (n= 76, 93.8%) and a minority has other disorders (major depression and borderline personality disorder). This

Table 1: SOCIO - DEMOGraphic Characteristics of the Patients Admitted to the day Hospital Program - 1

	PATIENTS (n=81)	
	n	%
GENDER		
Male	42	51.9
Female	39	48.1
MARITAL STATUS		
Married	19	23.5
Single	51	63.0
Divorced / Widowed	11	13.6
HOUSEHOLD		
Spouse and children	17	20.9
Parents	60	74.1
Spouse and parents	2	2.5
Alone	2	2.5
OCCUPATION		
Unemployed	34	42.0
Civil servants / Worker	26	32.1
Retired	9	11.1
Student	6	7.4
Other	6	7.4

Table 2: Socio-Demo Graphic Characteristics of the Patients Admitted to the Day Hospital Program-2

	PATIENTS (n=81)			
	Mean	Standard Deviation	Minimum	Maximum
AGE	32.9	8.9	18	59
EDUCATION YEARS	10.7	2.9	5	17

Table 3: Duration of Illness, Length of Stay in the Day Hospital and The Mean Anti Psychotic Doses*

	PATIENTS (n=81)			
	Mean	Standard Deviation	Minimum	Maximum
DURATION of ILLNESS (years)	11.3	7.4	1	35
DOSE of ANTI PSYCHOTICS** (mg / day)	478.4	329.6	50	1600
LENGTH of STAY IN THE DAY HOSPITAL (days)	135.7	61.3	30	243

* Chlorpromazine equivalent.
** n=74 (8 patients did not take anti psychotics)

is compatible with the aim of our DH. We want to accept mainly CP patients. There are various DH approaches outside, for substance dependence patients, geriatric populations or adolescent psychiatric patients (18), but the demand from the clinicians and patients made us start with CP patients.

In the last table (Table 5), the relapse rates of schizophrenics and schizo affective patients (total n= 73) who were followed with a weekly group therapy or in an outpatient service are compared and outlined. Overall relapse rate for a mean of 3 years was 45.2% (33 patients). In another study performed in our hospital relapse rate for outpatient CP patients was found 51%, but for 18 months (19). Similar findings were also found in the literature, too; Stirling et al. (20) found 53% relapse rate for 18 months. As a result, our

Table 4: Diagnoses of the Patients of the Day Hospital

	PATIENTS (n=81)	
	n	%
SCHIZOPHRENIA	62	76.5
SCHIZO AFFECTIVE DIS.	11	13.6
PARANOID DISORDER	3	3.7
BORDERLINE PERS. DIS.	3	3.7
MAJOR DEPRESSION	2	2.5

result is favorable and seems better than some other studies. We believe that, if all patients could continue DH program longer than the usual 4-8 months, or could come to all appointments given by our outpatient unit the relapse rate would have been lower. However, we know that relapse rate is influenced by several factors other than receiving therapy, and thus, it is hard to make direct conclusions from our results.

When we compare these patients (n=73) in regard to receiving and not receiving group therapy after discharge, we find that relapse is lower in the patients who regularly attend group therapy sessions. This result shows that the benefits of a rehabilitation program can be maintained by continuing group therapy sessions after DH. We believe that group therapy sessions lower the relapse rate by several different but interrelated ways, like; therapeutic factors, enhanced compliance to medication and identification of prerelapse symptoms and intervening before a full blown relapse occur. Ünlüoğlu and Sayıl (13) Ünlüoğlu and Tuncay (21) found low readmission rates for CP patients, when families were included in group therapy sessions. Relatives themselves stated that they benefit even from the first group session they attend (22). McKay (23) also found better outcome in DH patients who continued with self-help groups. Our post-DH has also turned to be a self-help group since May, 1996. Now it goes on with 11 patients, even better than expected.

On the other hand, we know that there is methodological difficulties in estimating the relapse rate. We may have missed some relapses since they might not have been recorded. The patient might have had a relapse but had not come to our clinic, so it is not recorded. We believe that symptom levels, social skills, quality of life, levels of withdrawal or self adequacy may be more suitable factors to evaluate the benefits and efficacy of DHs than relapse rate. Though not tested systematically, we have observed improvements in all of these areas among our patients. In another study conducted in our DH, Soykan et al.(24) found better symptom levels and functioning in discharged DH patients, compared with discharged inpatients, assessed with BPRS and GAFS.

CONCLUSION

Here, we wanted to present our DH experiences with CP patients. We have witnessed that, although it is a somewhat tiring work, it is obviously rewarding. These patients were mostly "difficult patients" of whom conventional treatments had failed or presented relapse frequently. Sometimes they made their primary clinicians feel hopeless. In fact, DH team had to face all these difficulties, too. With a structured and harmonious team work, such problems resulting from the patients could be overcome. Another key to success was to be realistic and modest on the target symptoms or improvements with these patients. A too ambitious or hurried approach might scare the patients and increase their feelings of inadequacy and cause early drop outs and more withdrawals.

DH approach helped the patients increase their responsibility taking, social skills, interpersonal relations and prevented them from relapses. These findings were most obvious in the patients who continue their relation with the DH by group therapies. Among these patients, discharge from the DH was like an attenuation, not termination, as they kept coming once a week. Termination may be a very harsh definition for some schizophrenics, and they may find this period too difficult to bear. They may not cooperate with the outpatient unit, where clinicians rotate regularly, and so the patients meet with a new clinician almost everytime they come. This might have reduced their compliance and consequently increased their relapse rates. Another solution might be to organize an outpatient facility for the discharged DH patients where they can continue their treatment with their previous doctor or another therapist whom they know from their DH therapy team regularly. Their separation anxieties could also be handled by working with them to help them adapt themselves to the new therapist before discharge. It is our impression that if such services could be more widespread in Turkey, the results for CP could be more helpful. We plan to continue our work in a more structured and organized way.

Table 5: Comparison of Rates After Discharge, in Regard to Group Psychotherapy (GP) Attendance

	FOLLOWED WITH GP (n=17)		NOT FOLLOWED WITH GP (n=56)		TOTAL (n=73)		STATISTICS	
	n	%	n	%	n	%	x^2	D.S.*
RELAPSE	3	17.6	30	53.6	33	45.2	7.3	$p < .01$

* Degree of significance

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THE RELATIONSHIP BETWEEN CLINICAL PATTERNS, EXTERNAL AND ENDOGENOUS FACTORS IN HAND ECZEMA
A Statistical Evaluation With Hierarchical Log-Linear Analysis Method

Aynur Akyol* • Hatice Erdi* • Yasemin Yavuz** • Ayşe Boyvat*

SUMMARY

In this report, 83 hand eczema cases were investigated in order to asses the relationship between contact sensitivity, atopy, localization and sex. Hierarchical analysis with these 4 variables revealed a three-way interaction between localization, atopy and contact sensitization. A positive correlation was found between atopy and positive patch test in palmar and finger eczema cases and an inverse relationship of these factors was noted in dorsal hand eczema cases. We also noted a three- way interaction between atopy, localization and sex, such that male patients with palmar eczema and female patients with dorsal hand eczema had a higher rate of atopy positivity.

In previous studies the correlations between the clinical patterns of hand eczema and their etiology was evaluated and no distribution of eczema was found to be typically allergic,irritant or endogenous . We believe that more studies should be carried out to clarify the relationships between the etiologic factors and the clinical patterns for a better diagnostic approach in patients with hand eczema.

Key words: *Atopy, contact dermatitis, hand eczema, localization, sex*

Hand eczema is one of the most common dermatologic diseases. Several factors such as age, sex, atopy and occupational exposure contribute to this disease (1,2). The disease is more common among women with a higher prevalence at young ages (3,4). Atopy is one of the most common causative factors (3,5). Irritant contact dermatitis, various types of atopic hand eczema and allergic eczematous contact dermatitis may deveilop in patients with atopy (3,4,6).

Since irritants, allergens and endogenous factors can all produce similar clinical manifestations elucidation of the etiology of hand eczema can be quite difficult. A possible combination of different etiologic factors should also be taken into consideration during examination of the patients. Assessment of the etiologic factors is of utmost importance for a better diagnostic and therapeutic approach in these patients.

Therefore, this study was designed to evaluate the relationship between localization, sex, atopy and contact sensitivity in various types of hand eczema.

MATERIALS AND METHODS

Eighty three patients with various types of hand eczema were included in this study. For each patient data regarding age, sex, occupation, contact with allergens and irritants and atopy were registered. The cases with personal or family history of atopic dermatitis, bronchial asthma and allergic rhinitis were regarded as positive for atopic history. The cases without atopic history were regarded as (-) and grouped accordingly.

The distribution of eczema on the hands was divided into the following 4 patterns: 1. Palmar pattern 2. Dorsal pattern 3. Finger localization 4. All over the hands. For the cases with hand eczema localized to more than one region, the most affected region was taken into consideration and classified accordingly. As the number of cases with eczema on all parts of the hands was considerably small they were not included in statistical evaluation.

* Associate Professor, Ankara University School of Medicine, Department of Dermatology
** Research Assistant, Ankara University School of Medicine, Department of Biostatistics

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Table 1: The distribution of patch test results and atopic findings according to localization and sex

	PATCH +						PATCH -						TOTAL		
	Atopy +			Atopy -			Atopy +			Atopy -			Atopy +	Atopy-	Total
	F	M	T	F	M	T	F	M	T	F	M	T			
Palmar r.	4	4	8	7	2	9	10	2	12	15	4	19	20	28	48
Finger r.	2	-	2	1	1	2	3	1	4	5	7	12	6	14	20
Dorsal r.	-	-	-	2	3	5	4	-	4	2	2	4	4	9	13
Generali.	-	-	-	1	-	1	-	-	-	1	-	1	-	2	2
Total	6	4	10	11	6	17	17	3	20	23	13	36	30	53	83

F: Female, M: Male, Palmar r.: Palmar region, Finger r.: Finger region, Dorsal r.: Dorsal region, Generali.: Generalized

The patients were patch tested with The European Standard Series of allergens (Chemotecnique Diagnostics AB, Sweden) in Finn Chambers® (Epitest, Finland) on Sconpor ® (Norgesplaster). The allergens were applied to the upper back and left in place for 2 days. The tests were read at 48 and 96h. Only (++) or stronger reactions were considered to be of allergic nature.

Hierarchical log- linear analysis method was used to evaluate the interactions of sex, atopy, localization and contact sensitivity in these patients.The explanatory variables included in the log- linear model were; sex (male- female), atopic history (±), localization (palmar, dorsal and finger region) and patch test results (±).

Hierarchical log- linear analysis with these 4 variables revealed the presence of two three-way interactions; one between localization, atopy and contact sensitization and the other between atopy, localization and sex ($G^2 = 3.58195$, DF= 6, P= 0.733). Statistical analysis was performed with SPSS For Windows Backward Elimination program.

RESULTS

Eighty three patients with hand eczema, 57 women and 26 men, 14 to 78 years of age(mean age 31.5) were enrolled in this study. Palmar pattern was observed in 48 patients (58 %), finger localization was seen in 21 patients(25 %) and dorsal involvement was noted in 13 patients (16 %).Only one patient had eczema all over the hands and due to the small number of the patients it was not included in statistical analysis. Thirty patients (23 females, 7 males) had an atopic history. Twenty-seven patients (17 females, 10 males) had positive patch test reactions to one or more allergens tested. The distribution of patch test results and atopic history according to localization and sex are shown in table 1.

Hierarchical log- linear analysis with these 4 variables revealed the presence of two three-way interactions; one between localization, atopy and contact sensitization and the other between atopy, localization and sex. Statistical analysis revealed a positive correlation between atopy and positive patch test in palmar and finger eczema cases but a significantly stronger relationship between atopy and patch test was observed in finger localization(odds ratios= 3) than in palmar localization(odds ratio=1.407). In dorsal hand eczema cases, an inverse relationship was noted between atopy and positive patch test (odds ratio=0.1) (Table 2).

A significant interaction between atopy, sex and localization was seen in our patients. In female patients the rate of atopy positivity didn't vary considerably with localization but the highest rate of atopy positivity was observed in patients with dorsal hand eczema. In male patients the rate of atopy positivity varied significantly with localization and the highest rate of atopy positivity was noted in patients with palmar eczema (Table 3) . Odd's ratios of the relationship between sex and atopy at certain localizations are shown in table 4.

Table 2: The Odds' ratios related to atopy and patch test

	Odds' ratios
Palmar region	1,407
Finger region	3
Dorsal region	0,1

Table 3: The rate of atopy positivity in different localization in females and males

	Atopy +				Atopy -				Total	
	Cases		Probality		Cases		Probality			
	F	M	F	M	F	M	F	M	F	M
Palmar Region	14	6	0,39	0,50	22	6	0,61	0,50	36	12
Finger Region	5	1	0,45	0,2	6	8	0,55	0,8	11	10
Dorsal Region	4	0	0,5	0	4	5	0,5	1	8	5

DISCUSSION

Hand eczema is a very common dermatological condition. Better diagnostic approaches and preventive measures are needed for this frequently disabling skin disease. Etiologic factors have to be evaluated in detail to have better diagnostic tools.

In our series of 83 hand eczema patients females predominated males by the ratio of 2.2/1. Women are in contact with great quantities of soap, detergents and water and they deal with the house work and child care at young ages. Our observation is in correlation with the previous reports in the literature (7,8,9).The frequency of the clinical patterns noted in our patients were as follows; Palmar pattern was observed in 48 patients (58 %), finger localization was seen in 21 patients(25 %) and dorsal involvement was noted in 13 patients (16 %). In previous studies palmar pattern was also the commonest.

Exogenous and endogenous factors contribute to the evolution of clinical manifestations. Among these various etiologic factors atopy is the most common one. Atopy can be found in various types of hand eczema including irritant and allergic eczematous contact dermatitis (3,5,7,10,11). In our study atopic history was found in 30 patients (36%).

Contact sensitivities to certain allergens have been searched in atopics and conflicting reports have been reported (12,13,14,15). Recent studies show that atopic patients have a significant rate of sensitization and should be patch tested (13,15). In our study contact sensitivity to one or more allergens tested was observed in 27 patients (17 females, 10 males). Ten of the 30 patients with atopic history also had positive patch test reactions. The high proportion of patients with positive patch test results among atopic patients indicates the necessity of evaluation of atopic patients for contact sensitivity.

Table 4: The Odds' ratios of the relationship between sex and atopy at a certain localization

	F/M
Palmar region	0,63
Finger region	6,6
Dorsal region	11

In our study a significant interaction between atopy, sex and localization was found in our patients. In female patients the highest rate of atopy positivity was observed in patients with dorsal hand eczema and in male patients the highest rate of atopy positivity was noted in patients with palmar eczema.

No association between clinical patterns of hand eczema and etiology was found in previous studies (7,10,16). Using hierarchical log- linear analyses with 4 variables , we showed a positive correlation between atopy and positive patch test in palmar and finger eczema cases.The relationship between finger localization and atopy was significantly stronger than the relationship between palmar pattern and atopy. In dorsal hand eczema cases, an inverse relationship was noted between atopy and contact sensitization. We believe that further studies on larger series should be designed to clarify the relationships between the etiologic factors and the clinical patterns of hand eczema for a better diagnostic approach.

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THE EFFECT OF HYPERTHERMIC PERFUSION CHEMOTHERAPY ON HEMODYNAMICS, BLOOD GASES AND SERUM TUMOUR NECROSIS FACTOR (TNF) LEVELS

Çiğdem Tezcan* • Mustafa Bayar** • Aslı Dönmez*** • Oya Özatamer*

SUMMARY

Hyperthermic perfusion chemotherapy (HPC) is a method which prevents the recurrence of the tumour and has beneficial effects on survival length. During this procedure the effects of extracorporeal circulation, temperature changes and possible increased activity of TNF may cause undesired disturbances in hemodynamic variables.

Thirteen patients were received HPC with extracorporeal circulation under general anaesthesia. The effect of this therapy on hemodynamic parameters, arterial blood gases, body temperatures and serum TNF levels were investigated.

There were no significant changes in MPAP (mean pulmonary artery pressure), PCWP (pulmonary capillary wedge pressure), CVP (central venous pressure) and heart rate throughout the study. However MAP (mean arterial pressure) decreased significantly at 30th minute of HPC. At this stage both arterial and pump TNF levels increased (11.35+5.15 and 60.38+13.96 respectively).

The elevation of TNF with hyperthermia and cytotoxic agents in systemic circulation and its undesired hemodynamic effects can be prevented by hemofiltration. Thus this chemotherapy method can be used safely unless patients are fully monitorized.

Key Words: *Blood gases, hemodynamics, hyperthermic perfusion chemotherapy, TNF levels*

Hyperthermic perfusion chemotherapy using extracorporeal circulation is one of the treatment methods for malignancies. By this method cytotoxic agents can be used safely at higher doses and the efficacy of these agents are enhanced by hyperthermia (1-5). Although it is reported that hyperthermic perfusion chemotherapy (HPC) is ten times more effective than systemic use of chemotherapeutics complexity of the process has evoked controversies about its convenience[5,6].

Tumour necrosis factor (TNF), which is mainly activated from the leukocytes and immune regulatory cells, is known to be responsible for hemorrhagic necrosis in some solid tumors and nowadays there is a wide interest on the antitumoural effects of intravenously administered TNF-a (7,8). As both hyperthermia and cytotoxic agents stimulate the release of TNF and increase its activity (1,9), the serum concentration of this factor may be increased during hyperthermic perfusion chemotherapy.

TNF is responsible for various hemodynamic and metabolic disorders. It impairs vascular endothelial permeability, provokes the release of toxic oxygen radicals and may cause multiple organ insufficiency at high concentrations (1,3). It also leads to systemic arterial hypotension by its effects on a-arginine metabolism (1).

Patients undergoing hyperthermic perfusion chemotherapy require general anaesthesia. The combination of the effects of extracorporeal circulation, temperature changes and possible increased activity of TNF may cause undesired disturbances in hemodynamic variables during this procedure.

* University of Ankara, Faculty of Medicine, Department of Anesthesiology and Reanimation
** University of The Euphrates, Faculty of Medicine, Department of Anaesthesiology and Reanimation
*** Başkent University Hospital, Department of Anaesthesiology and Reanimation

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The present study was undertaken to investigate the effects of hyperthermic perfusion chemotherapy on hemodynamic parameters, arterial blood gases and serum TNF levels.

MATERIALS AND METHODS

After obtaining approval from the Hospital Ethics Committee 13 patients aged between 20-65 (50.60±4.09) who were scheduled on pelvic hyperthermic perfusion chemotherapy under general anaesthesia were enrolled in this study. Patients with cardiopulmonary or metabolic disorders were not included in the study.

The patients received premedication with diazepam 10mg and atropine sulfate 0,5mg intramuscularly approximately 45 minutes (min) before induction of anaesthesia. Upon arrival in the operating theatre, a 5 lead electrocardiograph was attached to the patients and electrocardiogram was monitored throughout the surgery. 16 gauge catheter was placed in an arm vein for fluid infusion and drug administration.

A radial arterial catheter was inserted under local anaesthesia for continuous monitoring of systemic arterial pressure and to obtain blood samples for TNF and blood gases analysis. In addition, a 7F balloon-tipped flow directed thermodilution pulmonary artery catheter was percutaneously introduced into pulmonary artery via the right internal jugular vein for measurements of central venous and pulmonary arterial pressures. Temperature changes were monitored by rectal and oesophageal probes.

Thiopental 6mg/kg was used for anaesthesia induction and endotracheal intubation was achieved with succinylcholine 1.5mg/kg. 50% N20 in oxygen and 1-1.5% isoflurane were used for maintenance of anaesthesia under controlled ventilation. Vecuronium was used for further muscle relaxation.

Heart rate (HR), mean arterial pressure (MAP), mean pulmonary artery pressure (MPAP), pulmonary capillary wedge pressure (PCWP), central venous pressure (CVP), oesophageal and rectal temperatures were continuously monitored throughout the study by a Hewlett-Packard oscilloscope monitor (Model 78353B).

The hemodynamic parameters and temperature changes were recorded at the following stages.

Stage 1-Just before induction of anaesthesia (baseline)
Stage 2-15 minutes after intubation (before cross-clamping)
Stage 3-30th minute of perfusion and cytotoxic agent administration
Stage 4-15 minutes after declamping (postperfusion)
Stage 5-15 minutes after extubation

At these intervals arterial blood samples were taken for blood gas analysis and serum TNF measurements. At the 30th minute of perfusion and cytotoxic agent administration (Stage 3) blood samples for TNF measurement were taken both from radial arterial catheter (TNF-arterial) and extracorporeal pump line (TNF-pump). Blood samples for TNF were collected into EDTA containing tubes, immediately centrifuged and plasma was stored at -700C until analyzed. TNF analysis by ELISA method was done in Hematology Laboratory.

The patients were placed on a thermostatically controlled cooling mattress during the procedure. A. iliaca and v. iliaca were canulated and a roller pump with a membrane oxygenator was used for extracorporeal circulation. The flow rate was adjusted to 1lt/min. The perfusate was heated up to 41-420C and cisplatin (100mg) and adriamycin (50mg) was added to perfusate for hyperthermic perfusion chemotherapy. The perfusion chemotherapy was continued for 60 minutes. Hemofiltration was performed at the end of this period.

All values are expressed as mean±standart deviation. Statistical significance of the parameters were determined using Wilcoxon-Signed Rank Test. Differences were considered significant when $p<0.05$.

RESULTS

Hemodynamic variables (Table 1):

There were no significant changes in HR, CVP, MPAP and PCWP throughout the operation. MAP decreased below the baseline values 15 minutes after induction of anaesthesia (Stage 2). The reduction of MAP was greater at the 30th minute of perfusion and chemotherapy (Stage 3). Although it was still lower than the baseline values after declamping and extubation (Stages 4 and 5), the difference was not statistically significant.

Table 1: Hemodynamic parameters and blood gas analyses

	Stage 1	Stage 2	Stage 3	Stage 4	Stage 5
Heart rate (beats/min)	95.7±17.6	96.2±24.7	110.1±27.0	93.2±22.4	96.0±14.3
MAP (mmHg)	105.4±8.6	95.6±11.7#*	80.5±14.1*	93.4±15.1#	99.3±13.7#
MPAP (mmHg)	21.6±3.7	24.1±3.6	19.7±3.3	22.9±2.4	21.3±2.3
PCWP (mmHg)	16.3±4.1	20.0±3.6	15.7±1.7	17.8±2.7	16.0±2.8
CVP (mmHg)	12.9±2.2	15.2±2.6	13.3±2.9	13.5±2.2	11.9±2.3
PaO2 (mmHg)	89.4±8.7	224.8±62.4*	230.0±47.6*	234.0±51.9*	87.7±7.3
PaCO2 (mmHg)	35.3±0.9	34.5±4.2	33.6±2.7	35.7±2.3	35.4±1.2
SpO2 (%)	97.1±1.5	99.1±1.1*	99.2±1.0*	98.9±1.3*	96.9±0.9
pH	7.42±0.03	7.4±0.05	7.37±0.05	7.35±0.03*	7.4±0.03*
HCO3 (mmol/lt)	22.15±1.2	21.3±1.9	19.7±2.7*	19.6±2.4*	21.5±1.3

Stage 1 = Just before induction of anaesthesia (baseline), 2= 15 minutes after intubation (before cross-clamping), 3= 30th minute of perfusion and cytotoxic agent administration, 4=15 minutes after declamping (postperfusion), 5= 15 minutes after extubation.
* $p < 0.5$ in comparison with values at baseline, # $p<0.05$ in comparison with values at stage 3.

Temperature (Figure 1):

Both oesophageal and rectal temperatures began to decrease 15 minutes after induction of anaesthesia (Stage 2) and remained below the baseline values throughout the study. Although the temperatures increased after the removal of the cooling mattress and tracheal extubation (Stage 5) there was still a significant difference from baseline values.

Blood gas analysis (Table 1):

Beginning with the 30th minute of the hyperthermic perfusion (Stage 3), the blood pH and bicarbonate levels were below the baseline levels throughout the study but these reductions were not clinically important and no additional therapy was required. There were no alterations in arterial carbondioxide pressures during the study. PaO2 and SpO2 increased significantly after induction of anaesthesia due to the controlled mechanical ventilation with 50% oxygen.

Tumour Necrosis Factor (TNF) (Figure 2):

Serum TNF levels were not altered with anaesthesia. At the 30th minute of the hyperthermic perfusion and chemotherapeutic administration (Stage 3) TNF levels obtained from arterial blood and perfusate blood were both significantly higher than the TNF levels at other stages.

DISCUSSION

There are three basic advantages of hyperthermic perfusion chemotherapy (HPC) method: 1- neoplastic cells are more sensitive to hyperthermia than normal cells (2), 2- drug dose used can be 10 times higher than the dose administered systemically 3- the existance of a synergic action between hyperthermia and some drugs (6). In this method, normal circulation is replaced by extracorporeal circulation.

HPC requires general anaesthesia. Although several studies have demonstrated the beneficial effects of this method on survival rate or survival length by preventing recurrence of the tumour

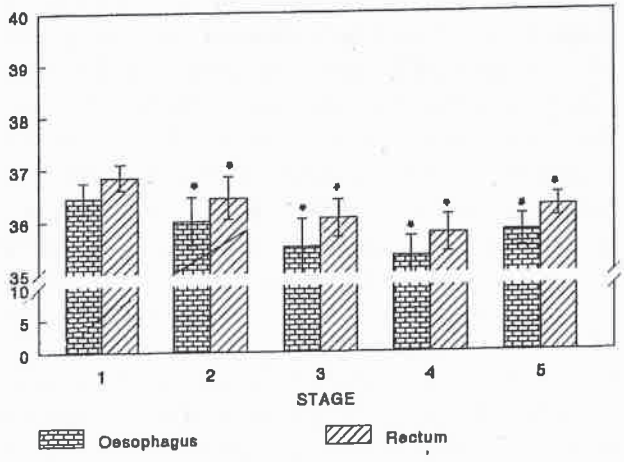

Fig. 1. Rectal and oesaphageal temperatures.
* $p<0.05$ in comparison with values at baseline.

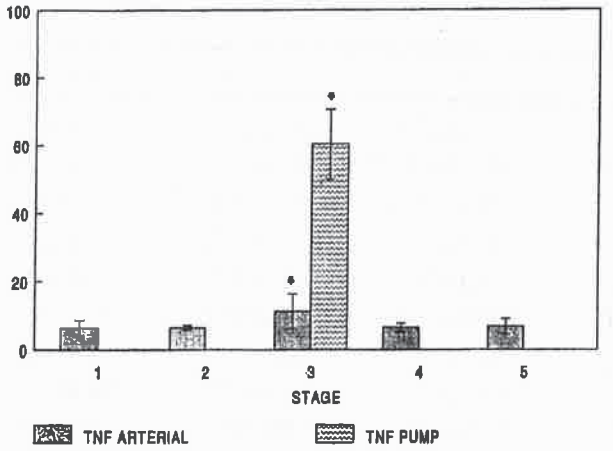

Fig. 2. Tumor Necrosis Factor (TNF) levels.
* $p<0.05$ in comparison with values at other stage.

(2,5,10-14), the effects of this therapy on circulatory status, the relation between TNF levels and HPC have not been studied.

TNF release and efficacy is increased by hyperthermia, chemotherapeutic agents, hemorrhage, trauma and by stimulation of the adrenergic system (1,9,15). Different anaesthetic agents have different effects on TNF levels. Shimado et al (16) have not observed any increases in plasma cytokine levels during prolonged anaesthesia with ketamine and penthobarbital.

In our study, anaesthesia induction with thiopental did not alter serum TNF levels. TNF levels were elevated at the 30th minute of HPC. At this period TNF level measured in the perfusate (TNF-pump) was approximately 5 times higher than the level measured from systemic arterial blood (TNF-arterial) (60.38 ± 13.96 and 11.35 ± 5.15 respectively). The levels returned to baseline levels just after HPC was completed and the cross-clamp removed. This decrease indicates the effectiveness of hemofiltration. Our results support that, hyperthermia and cytotoxic agents are important triggering factors in TNF release (1,9,15). Neither anaesthesia nor surgical manipulations altered serum TNF levels in our patients.

There is no data about the relation between HPC, TNF levels and hemodynamic changes in human. Fawcett et al (8) have reported the hemodynamic changes in a patient undergoing isolated limb perfusion with recombinant TNF-a melphalan. They found that at 30 minutes of recombinant TNF-a administration TNF-a levels reached peak levels in mixed venous blood (74.2u/ml) while arterial TNF-a concentrations remained below 17u/ml. The marked difference in plasma TNF-a concentrations between arterial and mixed venous blood can be explained by its clearance through the lungs. At this period there has been little change in MAP and MPAP but cardiac output increased markedly with concomitant decreases in systemic vascular resistance and remained increased for 24 hours. Arterial and mixed venous TNF-a concentrations returned to normal values 70 minutes after administration. This leads to the supposition that another mediator may be responsible for the hemodynamic changes (8).

In our study, there were no significant changes in MPAP, PCWP, CVP and HR throughout the study period but MAP decreased at 30th minute of HPC. At this period TNF levels were found to be higher than baseline values in arterial and pump blood samples. The reduction of MAP was not clinically important. Treatment with iv saline infusion proved sufficient to normalize MAP. This reduction may be due to the increased TNF levels as TNF is known to cause hypotension by effecting a-arginine metabolism (7).

Hyperthermia, induced with this method may lead to some undesirable effects on hemodynamics. Importance of complete temperature monitorization during HPC has been emphasized (4,6). Shime et al (17) reported the effects of hyperthermia on hemodynamics. They used icebags around patients' neck and head for external cooling and observed that during continuous hyperthermic peritoneal perfusion while blood temperatures increased significantly, MAP and SVR decreased, HR and cardiac index (CI) increased. In contrast, rectal and oesophageal temperatures decreased during HPC in our patients. Showing that body temperature may be effectively preserved by using external cooling mattress instead of icebags.

CONCLUSION

Our results show that hyperthermic perfusion chemotherapy elevates TNF levels, but extracorporeal circulation can limit leak of TNF to systemic circulation. Body temperature elevation and undesirable effects of this elevation on hemodynamics can be prevented by using cooling matresses. In conclusion; hyperthermic perfusion chemotherapy by extracorporeal circulation can be used safely unless patients are fully monitorized.

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FEMALE LARYNGEAL CANCER

Mehmet Umut Akyol* • Oğuz Öğretmenoğlu* • Faruk Ünal* • Şefik Hoşal*
Bülent Sözeri*

SUMMARY

Laryngeal Cancer is clinically thought to be a disease of male gander. During the period from 1970 to 1994, 422 patients with squamous cell carcinoma of the larynx were treated of whom 18 were females (4.26 %). Our experience with female laryngeal cancer, which is not well documented in literature was presented and discussed.

Key Words: *Female, Laryngeal Carcinoma*

Laryngeal carcinoma accounts for 1-2 % of all malignancies of the body with the exception of basal cell and squamous cell carcinoma of the skin. This percentage changes among different countries without following any geographical pattern (1). In general carcinoma or larynx has a significant preponderance of males over females, but female to male ratio shows wide variations among different countries. It was reported to be as high as 1:5 in U.S.A. and as low as 1:30 in Finland (2,3).

The incidence of laryngeal cancer has been increasing both in men and women, but the increase is more pronounced in women. In U.S.A. there was a considerable decrease in sex ratio (men : women) between the years of 1956 through 1976; 14.9 : 1 to 4.6 : 1 (4). There are different factors blamed for the increase of female laryngeal cancer, but there is one factor regarded as a significant contributor, the increasing smoking habit of women.

In spite of the narrowing breach of incidence between two sexes, laryngeal squamous cell carcinoma in females is not well documented. In this respect we believe it would be of interest to reveal the cases of female laryngeal cancer.

MATERIAL AND METHODS

The records of 422 patients with squamous cell carcinoma of the larynx treated in the department of Otolaryngology - Head and Neck Surgery at the Hacettepe University between 1970 and 1994 were reviewed retrospectively (5). Eighteen of the 422 patients were females (4.3 %). These patients were analysed with respect to age, smoking history, anatomical localization of the tumor, treatment modalities, and prognosis. The data was also compared to that of the male patients with laryngeal squamous cell carcinomas of the same period.

RESULTS

Of the 422 patients with laryngeal SCC, treated between the years 1970 and 1994, 18 were female. The age of the female patients ranged from 31 to 68, with a mean of 54 years (Table 1) Fifteen patients had supraglottic (83%) and 3 had pure glottic (17%) tumors. There were no subglottic tumor localization.

According to American Joint Committee for Cancer Staging Classification (6), 4 patients had stage I (22%), 3 had stage II (17%) tumors. Stage III lesions constituted the largest group with 9 patients (50%), and 2 patients had stage IV disease (11%).

Thirteen of the 18 women were heavy smokers. They had been smoking more than 1 package per day for an average of ten years.

One of the patients denied smoking tobacco and in four patients data about smoking were missing.

Five of the patients were treated with surgery alone, 5 received only radiation therapy. Four patients received combination of surgery and radiotherapy (RT), and in one of them chemotherapy was added to the treatment. Two patients underwent endolaryngeal stripping followed by RT. One patient could not be

* University of Hacettepe, Faculty of Medicine, Department of Hear-Nose-Throat, Ankara

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operated because of severe cardiac problems, and she was given RT and chemotherapy. She died of disease in 6 months with the progression of the tumor. One patient did not accept any treatment (Table 2).

The follow-up period ranged from 2 to 9 years. Current status was not available in 2 patients.

The patients treated with surgery alone had stage II and III tumors. All of them were alive and free of disease during the follow-up period (Table 3). Only 1 of the 5 patients treated by RT alone were free of disease for 4 years and remaining 4 patients developed tumor recurrence in 2 years time (Table 4). Combination therapy were used in stage II and stage IV tumors. Two were free of disease for 4 years, one died of disease and one patient was not available for follow-up (Tablo 5).

Table 1: Age distribution of the patients

Age	Patients (n)
31-40	4
41-50	2
51-60	6
61-70	6
Total	**18**

n: number of patients
mean age: 54.4. years

Table 2: Treatment modalities

Treatment	Patients (n)
Surgery only	5
RT only	5
Surgery +/- Chemo +/- RT	6
RT + Chemo	1
No treatment	1
Total	**18**

RT: radiation therapy, Chemo: chemotherapy

Table 3: Follow-up, surgery alone

Patient	TNM / stage	Complication	Follow-up
#1	T3N1M0 3	-	3 years, tumor free
#2	T2N0M0 2	-	2 years, tumor free
#3	T3N1M0 3	-	9 years, tumor free
#4	T2N1M0 3	hypocalcemia	8 years, tumor free
#5	T2N1M0 3	flep necrosis	7 years, tumor free

Table 4: Follow-up, radiation therapy alone

Patient	TNM / stage	Follow-up
#1	T1N0M0 1	2 years, with tumor
#2	T2N1M0 3	2 years, tumor free
#3	T2N1M0 3	died of disease, 1 year
#4	T2N0M0 2	recurrence, 1 year
#5	T1N0M0 1	4 years, tumor free

DISCUSSION

The SCC of the larynx is clinically thought to be a disease of the males. Among different cancers rising in the body, laryngeal carcinoma has the most noticeable difference of incidence between two sexes excluding genital cancers. The obtained findings in our series revealed that the ratio of female to male patients with laryngeal carcinoma is 1:22. When compared with other countries this ratio is very low. The ratio according to gender in U.S.A. is 1: 4.6 (Female to male), in Australia 1: 8, in Japan 1: 9.6, in Iceland 1: 12, in Norway 1: 13, and in Sweden 1: 8. (3,4,7,8) A similar proportion to our series was observed in Croatia, which is 1: 24 (9). In Finland, there is a conspicuous difference between two sexes and the ratio is 1: 30 (3).

72 % of the female and 91 % of male patients in our series were heavy smokers. There is no doubt about the role of smoking in the development of laryngeal carcinoma (7,10,11). In our opinion one reason for the high male / female ratio in our country must be the low incidence of smoking among female population as a natural result of the social status. Unfortunately there is no available statistical information about smoking habit of population and sub-populations in Turkey.

Among the 18 female patients, the majority of tumors located within supraglottic larynx (83.3%) and the remaining arose in glottic larynx (16.7%). Iwomoto (8) reported that 68% of female laryngeal carcinomas were located within supraglottic larynx.

Derienzo (2), however, found that glottic carcinomas represented 65% of cases in 71 female patients. The localisation of tumors in females correlates with males in our series in which supraglottic carcinomas account for 73% of cases, followed by glottic (26%) and subglottic (1%) tumors (5). This may be related to racial and genetic differences or it may be explained by the age of the patients as the supraglottic carcinoma occur in younger age (9,12,13,14). In our series the mean age was 54.3 years.

Table 5: Follow-up, combined treatment

Patient	TNM / Stage	Treatment	Follow-up
#1	T3N1M0 3	surgery + RT	-
#2	T3N2M0 4	RT + surgery + RT	4 years, tumor free
#3	T1N0M0 1	stripping + RT	-
#4	T1N0M0 1	stripping + RT	3 years, tumor free
#5	T2N1M0 3	surgery + RT	died of disease, 4 years
#6	T2N2M0 4	surgery + RT	4 years, tumor free

Both in male and female patients the most common stage of the disease in the time of diagnosis was stage III. The number of the female patients in stage IV however, was less, and in stage I was higher than the male patients. This is probably because of the fact that women seek for treatment earlier than men. This fact is also the reason of the better prognosis in female patients.

Overall 5 year survival rates for stage II and III tumors given in literature are 51-85% and 48-59% respectively (5,13,15,16). Five of our patients with stage II and III tumors treated with surgery alone had no tumor recurrence in a follow up period of 2 to 9 years. Only one patient was living tumor free for 4 years while one patient with stage I, 2 patients with stage III and one patient with stage II tumors were either dead or were living with tumor, in the group of patients treated with radiation therapy alone. In the group of patients treated with combined therapy, there were 2 cases of stage I tumors treated with stripping alone but due to the microinvasion in histopathologic diagnosis RT was added and both of them were living disease free. There were 3 other cases with advanced disease in the same group. One of them died of disease with distant metastasis and one was lost to follow up. The last case was a stage IV cancer treated with pre-operative RT + radical surgery + post-operative chemoradiotherapy and was living for 4 years with no apparent disease.

These results show that the group of patients treated with surgery alone has better results than that of treated only with RT. Even in advanced cases we can increase the survival rates with combination therapy.

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PHASE CONTRAST MICROSCOPY AND COULTER COUNTER FOR EVALUATING THE SOURCE OF HEMATURIA AFTER EXTRACORPOREAL SHOCK WAVE LITHOTRIPSY (ESWL)

Adil Gökalp* • Çağatay Öktenli** • Murat Dayanç* • Yaşar Özgök* • Bedreddin Seçkin*
Mete Kilciler * • Abdülgaffar Vural*** • Doğan Erduran*

SUMMARY

After ESWL, to localise the source of hemorrhage in 31 patients with hematuria, urine samples were examined by phase contrast microscopy. Ten of the 31 patients had isomorphic red cells of more than 80%; 5 patients had dysmorphic red cells of more than 80% and 16 patients had mixed hematuria. Urine samples taken from same patients (group1) and 35 patients (group 2) with hematuria due to kidney stones were compared using a Coulter counter for urinary red cell mean corpuscular volume and red cell volume distribution curves (RCVDC). We found that red cells had larger volume measurements in most of patients, except 3 patients in group 1 and 2 patients in group 2 who had smaller volume measurements. RCVDC were glomerular type in 3 patients in group 1 and 2 patients in group 2, non-glomerular type in 10 patients in group 1 and 12 patients in group 2 and mixed type in 18 patients in group 1 and 21 patients in group 2. Although there was no significant difference between hematuria after ESWL and hematuria due to kidney stone, we cannot exclude a direct effect of shock waves on renal tissue.

Key Words: *Hematuria, kidney stone, lithotripsy, phase contrast microscopy, coulter counter*

ESWL has rapidly become a non-invasive method of treating kidney and ureteral stones, since 1980 (1). Side effects of ESWL include hemorrhage, edema, tubular necrosis and subsequent fibrosis in the kidney (2).

The appearance of red cells in the urine may be the result of disease of any segment of the urinary tract (3, 4). Urinary red cells are usually small and morphologically abnormal in glomerular hematuria and slightly enlarged and morphologically intact in non-glomerular hematuria (5). The source of hemorrhage can be determined either by Coulter counter or phase contrast microscopy (PCM) of urine taken from patients with hematuria after ESWL. The use of PCM for examination of urine to differentiate glomerular hematuria from non-glomerular hematuria was first described by Birch and Fairley(6) in 1979. In 1986, Shichiri et al (7). described the determination of mean corpuscular volume (MCV) of urine red cells with a cell counter as a new method to distinguish glomerular and non-glomerular hematuria. Coulter counter which is a non-invasive screening test may be use to detect the MCV and RCVDC of red cells of urine in these patients (8, 9).

PCM accentuates the interfaces of surfaces of slightly differing refractive indexes. Accordingly, it enhances the ability to see and identify most cells and casts, many of which might be missed with bright light microscopy (10).

PATIENTS AND METHODS

Patients with hematuria after ESWL (Table 1, group 1)

Fresh, midstream urine samples taken from 31 patients with macroscopic hematuria immediately after ESWL*. Before the ESWL, we selected patients who have no hematuria. The mean age of patients was 29.6 years (ranged between 20-45). Twenty of the 31 patients were men.

The patients received 1 session shock wave treatment with the Lithostar Plus. The number of shock waves varied from 1400 to 4000 (mean 2593) and mean intensity was 17,7 kv. The left kidney was treated in 13 patients and right kidney in 18.

* Department of Urology, Gülhane Military Medical Academy, Ankara
** Department of Internal Medicine, Gülhane Military Medical Academy, Ankara
*** Department of Nephrology, Gülhane Military Medical Academy, Ankara

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Table1: Clinical characteristics and results in 31 patients with hematuria after ESWL

Patient No	Age	Sex	Treated Kidney	Shocks	kV	MCVU	RCVDC
1	32	F	Left	2000	18.1	88.3*	Non-Glomerular
2	38	M	Right	4000	19	90.1	Non-Glomerular
3	24	F	Left	3000	19	92.2	Mixed
4	33	F	Right	3000	16	111.2	Mixed
5	20	M	Right	1400	15.4	82.3	Non-Glomerular
6	28	F	Right	4000	19	85.7	Mixed
7	20	M	Right	4000	19	56.2	Glomerular
8	21	M	Right	2000	19	94.7	Mixed
9	20	M	Right	1600	16	83.7	Mixed
10	45	M	Right	2000	16.3	92	Non-Glomerular
11	27	F	Right	4000	18.1	55.7	Glomerular
12	43	M	Left	1800	19	103.1	Non-Glomerular
13	34	M	Right	2000	18	86.1	Mixed
14	36	F	Left	2200	16.3	85.3	Mixed
15	26	M	Left	1800	16.6	88.9	Mixed
16	44	M	Right	2500	16.9	100	Non-Glomerular
17	21	F	Right	3200	19	95.8	Non-Glomerular
18	29	M	Left	2400	18.1	85.8	Mixed
19	26	M	Left	2200	16.3	79.3	Mixed
20	43	F	Left	2700	17.5	101.1	Non-Glomerular
21	21	M	Right	3000	16.5	82	Mixed
22	20	F	Right	2200	18.1	91.5	Mixed
23	37	M	Left	2400	18.1	103.5	Non-Glomerular
24	36	M	Left	3000	19	92.8	Mixed
25	22	M	Right	3000	17.8	94.6	Mixed
26	22	M	Left	1800	17	78	Mixed
27	41	F	Right	3000	18.5	87.5	Mixed
28	33	M	Right	3200	19	89.4	Non-Glomerular
29	28	M	Left	1800	18.1	90.8	Mixed
30	26	F	Left	2000	16.6	86.6	Mixed
31	23	M	Right	3200	19	57.4	Glomerular

*fl=femtoliters
MCVU=Urine red cell mean corpuscular volume
RCVDC=Red cell volume distribution curve

Patients with hematuria due to kidney stones (Table2, group2)

Fresh, midstream urine samples were collected from 35 patients with hematuria due to kidney stones. Of the 35 patients, 27 had obvious macroscopic hematuria and 8 had microscopic hematuria (≥3+ by dipstick -Ames Multistix 10 SG, Bayer Diagnostigcs-, high power field >5 of red cells). The mean age of patients was 27.4 years (ranged between 20-41). Twenty three of the 35 patients in group 2 were men.

*Second generation Siemens Lithostar Plus Lithotriptor

PHASE CONTRAST MICROSCOPY

10 ml, fresh, midstream urine samples taken from 31 patients with macroscopic hematuria after ESWL. One drop of urine was transferred to a labelled glass slide. A coverslip was placed on the specimen. Slides were screened by (Nikon AFX-IIA) PCM at a magnification of x400 for red cells.

The morphology was classified according to the numerical criteria of Fassett et al (11). If more than 80% of the red cells in a specimen showed the morphological appearances described by Fairley and Birch (12) it was recorded as dysmorphic, indicating glomerular hemorrhage, if more than 80% of the red cells were undistorted and uniform in size and shape it was recorded as isomorphic, indicating non-glomerular hemorrhage; if a more even proportion of the red cells were dysmorphic and isomorphic it was recorded as mixed.

COULTER COUNTER

10 ml, fresh, midstream urine samples which obtained from patients in group 1 and group 2 were

Table 2: Clinical characteristics and results in 35 patients with hematuria due to kidney stones.

nt No	Age	Sex	Hematuria	MCVU	RCVDC
1	27	M	Macroscopic	96.8=	Non-glomerular
2	36	M	3+*	90.1	Non-glomerular
3	23	F	Macroscopic	88.4	Mixed
4	24	M	Macroscopic	88.6	Non-glomerular
5	33	M	Macroscopic	82.1	Mixed
6	37	M	Macroscopic	83.9	Mixed
7	20	M	3+	100.3	Non-glomerular
8	26	F	Macroscopic	89.7	Non-glomerular
9	23	M	Macroscopic	89.8	Mixed
10	39	F	Macroscopic	90.1	Mixed
11	32	M	Macroscopic	88	Mixed
12	21	F	Macroscopic	55.8	Glomerular
13	20	M	3+	90.2	Non-glomerular
14	22	M	Macroscopic	94.3	Mixed
15	30	F	Macroscopic	89.7	Mixed
16	25	F	Macroscopic	85.1	Mixed
17	20	M	Macroscopic	100	Non-glomerular
18	37	M	Macroscopic	95.3	Non-glomerular
19	29	M	Macroscopic	88.5	Mixed
20	28	M	3+	102.7	Mixed
21	34	M	3+	87.2	Mixed
22	41	M	Macroscopic	90.5	Non-glomerular
23	31	F	Macroscopic	90.9	Mixed
24	24	F	3+	98.6	Non-glomerular
25	20	M	Macroscopic	80.3	Mixed
26	28	M	Macroscopic	87.6	Mixed
27	36	F	Macroscopic	54.7	Glomerular
28	22	M	3+	94.7	Mixed
29	27	M	Macroscopic	92.3	Mixed
30	20	F	Macroscopic	89	Non-glomerular
31	25	F	Macroscopic	89.3	Mixed
32	23	F	3+	90.6	Mixed
33	34	M	Macroscopic	88.2	Mixed
34	22	M	Macroscopic	95.2	Non-glomerular
35	20	M	Macroscopic	90.1	Mixed

*by dipstick =fl:femtoliters
MCVU=Urine red cell mean corpuscular volume
RCVDC=Red cell volume distribution curve

centrifuged in a Hettich EBA 3S Centrifuge at 1500 rpm for 5 min. The supernatant of the centrifuged urine specimens was removed with a pipette and remaining 0,5 ml gently resuspended in 9,5 ml Isoton III (standart diluent, coulter reagents) buffer. The pellet was resuspended in 10 ml of diluent injected directly into the red cell counting chamber for counting in an electronic particle-size analyser by Coulter counter MaxM (Coulter Electronics, Inc., Hialeah, Fla., USA). The machine was first calibrated with the commercially available control and the parameters set for evaluation of red cells as outlined in the Coulter counter Owner's Manual (13). The pre-diluted sample, is drawn into the blood-sampling valve. The "prediluted sample mode" was used for urinary red cells analysis. Data showing cell-size distribution are shown on the video data terminal and transmitted to the printer and X-Y recorder to transcribe distribution curves. The histograms of cell volumes were drawn as distribution curves on the printer-plotter, which was connected on

line to the analyser. A RCVDC, red cell count and calculated MCV were recorded. Particles that size at 20 femtoliters (fl)or less were automatically excluded from calculation of MCV (3). On this basis, MCV between 40 and 180 fl describe as diagnostic for glomerular and non - glomerular sources of hematuria. Amongst diagnostic measurements, MCV between 40 and 59 fl classify as "glomerular" and from 60 to 180 fl define as "non-glomerular" (14). Glomerular RCVDC are somewhat skews and peaks in the microcytic range. Non-glomerular RCVDC are relatively normal in appearance and usually peaks in the normocytic range, although a few peaks in the macrocytic range (3). Fig 1 shows the characteristic distribution curves of glomerular, non-glomerular, and mixed hematuria.

STATISTICAL ANALYSIS

Data are expressed as mean ± SD. Statistical analysis were performed using Mann Whitney U, chi-square, Kruskal-Wallis Anova tests, and Pearson's correlation coeficient where appropriate. The criterion for statistical significance was $p<0,05$.

RESULTS

Ten of the 31 patients had dysmorphic red cells of less than 20% and isomorphic red cells of more than 80%; 5 of the 31 patients had dysmorphic red cells of more than 80% and isomorphic red cells of less than 20% and 16 of the 31 patients had mixed hematuria, defined as the simultaneous presence of glomerular and non-glomerular red cells in the same urine sample and in about the same proportion.

Dysmorphic red cells were detected as cells with an irregular outline, some membrane protrusions, irregular deposits of dense cytoplasmic material around the cell membrane, variations in size such as ghost cells and a variety of cells of odd appearance, particularly those with a "budding yeast" like or "mickey mouse ears" appearance may be seen (10). Isomorphic red cells had a regular shape and diameter, often revealing their usual biconcave aspect.

We found that red cells had larger volumes in most of patients, except 3 patients in group 1 and 2 patients in group 2 who had smaller volumes as it has been at red cells with glomerular origin. RCVDC were glomerular type in 3 patients, non-glomerular type in 10 patients and mixed type in 18 patients in group 1. RCVDC were glomerular type in 2 patients, non-glomerular type in 12 patients, mixed type in 21 patients in group 2.

DISCUSSION

Several studies (15,16,17) found gross and microscopic morphological defects in shock wave-treated kidneys. While in the acute phase renal edema and vascular and tubular injury predominate, focal areas of interstitial fibrosis of cortex and medulla persist (18). The stones are reported to be fragmented less than two mm in size (19). The most current side effect associated with ESWL is macroscopic hematuria due to the renal parenchymal lesion and not to movement of stone fragments during treatment (2,20,21) Jaeger et al (16) found in all cases of their series of canine kidneys that did not have kidney stones, macroscopic hematuria occured after ESWL.

In study of Recker et al (1) , rats subjected to high doses of 1000 or 2000 shock waves showed slight to moderate glomerular hemorrhage acute after ESWL. This was partially caused by the rupture of glomerular capillaries or fracture of the Bowman¢s capsule near an interstitial hemorrhage. Clinically apparent hematuria, too, is caused in a variety of different ways; penetration and drainage of parenchymal hematoma into the calyceal system, penetration of interstitial hemorrhage into nephronal tubuli, rupture of glomeruli and hemorrhage into proximal tubuli, urothelial erosions and disruption of small vessels of the renal pelvis (17). Likevise, Weber et al (17) indicates that cortical (interlobular) venular injury occurred often and usually produced a grossly visible hemorrhage. Rupture of larger arcuate and interlobar veins was not frequent, but resulted in more significant hemorrhage, extending into the subcapsular and/or pelviocaliceal space. Rupture of small cortical arterioles and glomerular capillaries was also frequently encountered.

In our study, we tried to find out source of hematuria in patients by using PCM and Coulter counter for examining of urine red cell morphology. PCM examination has been suggested as an easy, non-invasive test and provides a direct approach in distinguishing between glomerular and non-glomerular hematurias (22,23,24). Neverthless, this method is not able to localise the origin of hematuria; the differentiation between glomerular and tubular disease is not possible using PCM to look at red cell morphology (25) . Likewise, based on automated size analysis of urinary red cells, glomerular and non-glomerular hematuria

according to whether the cell volume was smaller or larger, respectively, than that of simultaneously sampled peripheral blood red cells (26). The presence of dysmorphic red cells or of cells with low MCV is believed to indicate glomerular hematuria, isomorphic red cells, or cells with a normal MCV are thought to originate from non-glomerular sources (27). Except this, Coulter counter reveals distincly different RCVDC for glomerular and non-glomerular hematuria (28,29,30).

Mixed and non-glomerular types of red cells were obtained from most of patients in our study. Reports (11,31,32,33) have emphasized that mixed type should always be considered glomerular in origin. As a valuable point to mention some of these reports (11,31) shows that kidney stones cause mostly mixed hematurias. Furthermore, it was reported in several studies (32,34) that glomerular type red cells could be found in hematuria due to kidney stones.

The shape of the RCVDC and the MCV can be altered by the presence of other debris i.e. non-cellular particles, other cells and bacteria. In patients with hematuria, particles, other than red cells, are likely to be count in the Coulter counter (30). The accuracy of this test depends on the degree of hematuria and the amount of debris present (29).

There is two suggested ways for the reason of dysmorphism of urinary red cells (35). The first stage is the initial trauma received during passage through the glimerular basement membrane and tubules resulting in damage to the red cell surface. This may cause a reversible change in shape. The second stage occurs when such damaged cells pass through the hostile hypotonic environment of the distal tubule. This causes the red cell not only to become hypochromic but also deformed. If the sodium concentration of the distal segment is above 75 mmol/l, most cells even with prior mechanical trauma, can return to their original shape and fail to become dehemoglobinised, thus masking their source of origin. If osmotic stress is minimal, fewer cells will be deformed, depending on their fragility and the degree of mechanical trauma. This may produce a mixed picture (35). Urine osmolality has influences on red cell morphology (36,37,38). Turitzin et al (39), had also found a strong correlation between low urine osmolality and a pattern similar to glomerular hematuria.

Blood samples were subjected to shock waves up to a frequency of 500 by Jaeger et al (16) . As a result a mild degree of hemolysis occured. In vitro the ESWL caused hemolysis with a RCVDC of mixed pattern when 500 shocks were used and a glomerular pattern when 1000 shocks were used (39).

Although there was no significant difference between hematuria after ESWL and hematuria due to kidney stone, we cannot exclude a direct effect of shock waves on renal tissue. There was a significant correlation between number of shock waves and glomerular hemorrhages. In some cases, glomerular type red cells might be explained with low urine osmolality, rupture of glomerular capillaries and effects of shock waves on red cells. However, source of non-glomerular type of red cells are probably draining of hematomas resulted from ESWL or larger vessells extending into the pelviocaliceal space and urothelial erosions. Mixed types may be resulted from effects of osmotic stress in the tubules, mechanical traumas and shock waves on red cells.

In conclusion, our findings suggest that ESWL induced hematurias are usually resulted form damage to renal tissue rather than movement of calculus particles though the urinary tract.

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URETERAL REPLACEMENT WITH GORE-TEX TUBE GRAFTS

Sümer Baltacı* • Gökhan Özer** • Elif Özer*** • Tarkan Soygür** • Ömer Beşaltı**** • Kadri Anfarta*****

SUMMARY

For evaluating the role of Gore-Tex (polytetraflouroethylene) as an ureteral substitude in dogs after resection of a 5-8 cm-long middle segment, five mongrel dogs underwent resection of the middle segment of the left ureter and segmental replacement of the resected part with segments of Gore-Tex over a double-j cahtheter. The double-j stents were removed at 21 days and the dogs were evaluated by excretory urography and abdominal ultrasonography 12 weeks after surgery. They were then sacrificed ant the ureters and the kidneys examined both grossly and microscopically. There was radiologic evidence of advanced hydronephrosis on the left side in all 5 dogs. Grossly, the left kidneys showed marked pelvic and proximal ureteral dilatation and atrophy of parenchyma. Severe stricture at the anastomotic sides and marked fibrous tissue around the prostheses were found. No celluler lining was found in the Gore-Tex lumen. We concluded that, there is no potential for ureteral replacement by Gore-Tex tube graft.

Key Words: *Ureter, ureteral replacement, gore-tex*

In patients with an extensive loss of the ureter various techniques, including extensive mobilization of the kidney with downward displacement (1), use of the Boari flap or vesicopsoas hitch (2) are available to bridge the gap. There are however, occasions on which the above procedures are not adequate to bridge the defect of the ureter for anastomosis. In such cases, ileal interposition, renal autotransplantation or if the other kidney and ureter is present transureterouretrostomy has been used (2,3,4,5,6). There are however, drawbacks for each technique. Various synthetic materials including Vitallium (7), Tantalum (8), Silicone (9) and Teflon (10) have also been tried in an effort to replace diseased ureter since 19842, but they have failed due to the lack of peristalsis and problems with infection and stricture formation. Restoration of ureteric continuity after transection of the ureter has recently been tried but failed as the restoration occured as a process of repair and not by regeneration of uretral wall (11). However, in 1982, Varady et al reported that the use of explanded polytetraflouroethylene (Gore-Tex) as a material for segmental ureteral replacement was encouraging, although anastomosis of this material to bladder was unsuccessful (12). Ungortunately, no other papers have been published using Gore-Tex as a ureteral substitute in dogs.

MATERIALS AND METHODS

Five mongrel dogs weighing 12-15 kg were used for the experiments. They were operated under general anesthesia induced by Xyalacin HCI (2 mg/kg), Ketamin HCI (15 mg/kg); endo-tracheal intubation was not used.

The animals were placed supine and a left paramedian incision was performed. The left ureter was identified and brought into the wound.

Two holding sutures of 3/0 chromic catgut 5 to 8 cm apart from each other were placed on the ureter. The ureteral segment between the holding sutures was excised. Both ureteral ends were spatulated and appropriately prepared to reanastomosis with Gore-Tex tubing. We intubated the Gore-Tex tube (6 mm in diameter)and the two ends of the ureter with a 5-F

* Specialist in Urology, Ankara University School of Medicine
** Assistant in Urology, Ankara University School of Medicine
*** Assistant in Pathology, Ankara State Hospital
**** Assistant in Surgery, Ankara University School of Veterinary Faculty
***** Professor in Urology, Ankara University School of Medicine

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double-j cahteter. Anastomosis was completed with interrupted 5/0 chromic cat-gut sutures using binocular loupe (Fig. 1). The right ureter was not disturbed in any animal. The abdominal muscles were closed in one layer with a 3/0 absorbable suture and the skin closed with a subcuticular 3/0 absorbable suture. Parenteral antibiotics were given for 7 days. Postoperatively, the animals were allowed free access to water and feeds immediately. The double-j stents were removed at 21 days via on open cystotomy.

The dogs were evaluated 12 weeks following surgery. The evaluation include excretory urography and abdominal ultrasonography. They were then sacrificed for necropsy. Both kidneys and ureters jere removed for histological examination. The anastomotic sites were examined grossly and histopathologically. Sections were stained by haemotoxylin and eosin.

RESULTS

All dogs survived for the duration of the experiment and had normal serum creatinine levels at the time of sacrifice. In all cases, normal renal function was evident on the right side but there was radiologic evidence of advanced hydronephrosis on the left (Fig. 2).

The right reno-ureteral unit of these five animals were both macrospocially and histologically normal. However the left kidneys showed marked pelvic and proximal ureteral dilatation and atrophy of parenchyma with enlargement of the whole kidney. Nephrocalcinosis and severe stricture at the anastomatic sites were evident (Fig. 3). The ureteral mucosa proximal to the Gore-Tex tubing revealed squamous metaplasia. We found no cellular lining in the Gore-Tex lumen. Marked fibrous tissue was found around these prostheses and no smooth muscle regeneration was seen (Fig. 4).

DISCUSSION

On occasion, the urologic surgeon is faced with dilemma of replacing all or part of ureters. Many substitutes, including synthetic materials have been tried, but for a variety of reasons, including bio-incompatibility, stone formation, infections, the lack of peristaltic activity, stricture formation and hydronephrosis, these substitutes have not been successful (7,8,9,10,13).

Varady et al reported that Gore-Tex, an experimental form of polytetrafluoroethylene was substitute

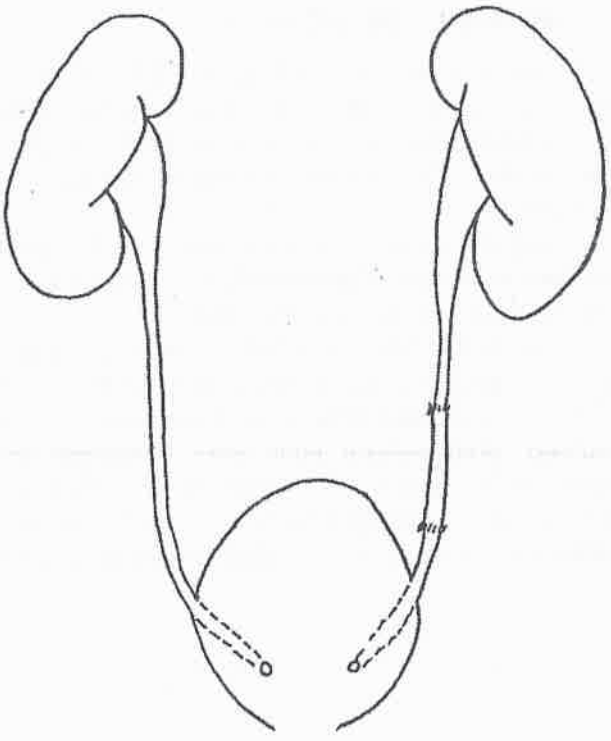

Fig. 1. Unilateral resection of a 5-8 cm segment of the left ureter and replacement with Gore-Tex tubing.

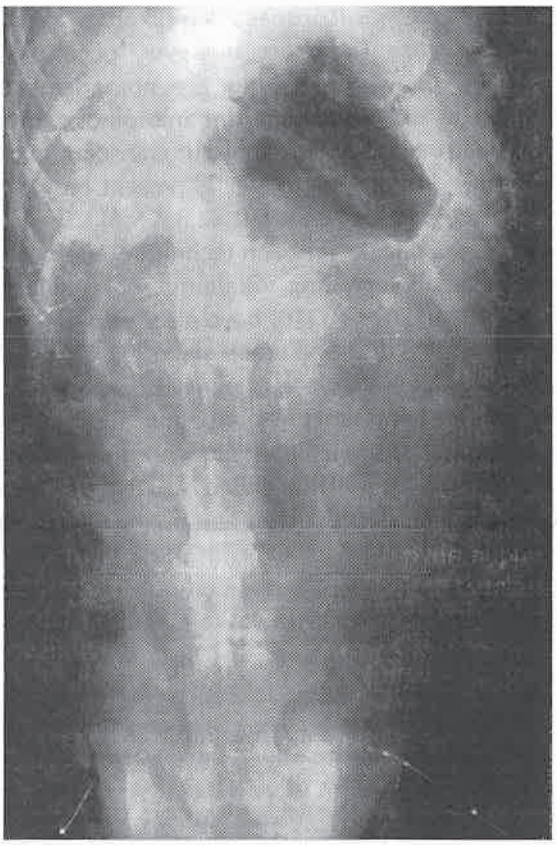

Fig. 2. Urographic demonstration of left hydronephrosis.

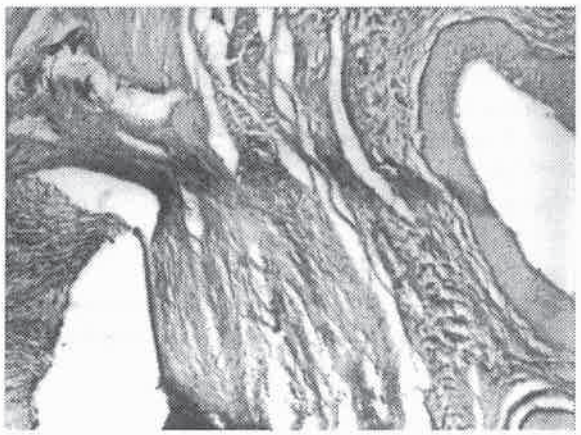

Fig. 3. Microphotography: Severe stricture at the anostomotic side is seen (H&E, x50).

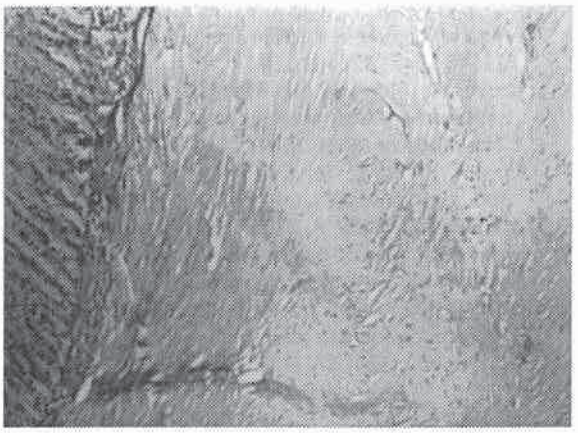

Fig. 4. Microphotography: Marked fibrous tissue around the prostheses without smooth muscle regeneration is seen (H&E, x50).

to bridge gaps between segments of ureter, although anastomosis of this synthetic material to bladder was found unsuccessful due to infection and migration of the prosthesis (12). At a mean implantation time of 7.5 months they obtained normal renal function at the side they performed segmental ureteral replacement in all 6 mongrel dogs. However no other study using Gore-Tex as a segmental ureteral replacement material has been reported. Gore-Tex is the preferred material in vascular surgery due to its nonwetting and nonclotting surface when used in the arterial and venous beds. It allows development of a neoendothelium on its luminal surface and it has excellent sewing and handling properties (12,14). In this study, we tried to reestablish the role of Gorge-Tex as a segmental ureteral substitute. Comparing our study with Varady et al's there were 2 modifications exit. First we performed unilateral segmental replacement and followed the other side as a control unit. Second, althouh Varady et al did not use stents we, like others, intubated the Gore-Tex tube and the two ends of the ureters with a double-j catheter for 21 days to prevent stricture at the anastomatic sites while acting as a guide for the regeneration of the ureteral wall.

Although, Varady et al obtained normal postoperative IVPs at the site they performed segmental ureteral replacement in all their dogs (12), we were disappointed to obtain hydronephrosis after 12 weeks in all our 5 dogs. The cause of the obstruction may lie in a stricture of the anastomatic site or an aperistaltic segment in the urinary tract. However, Stern et al have shown fluoroscopically that by preservation of the renal pelvis and a few centimeters of proximal ureter, urine could be propelled successfully into and through a prostheses (15). So, an aperistaltic segment is unlikely to be the cause of the obstruction. Although, like Tachibana et al (13) and Persky et al (16), we used double-j catheter as an ureteral splint catheter to reduce the stricture at the anastomatic sites we found severe stricture at the anastomatic sites as the cause of the obstruction in our study. On the other hand, like Tachibana et al (13) and Varady et al (12), we detected fibroblastic granulation around the Gore-Tex graft and this might also be the cause of the obstruction.

Although Tachibana et al (13) reported 5 to 7 layers of transitional cells on the inner surface of the collagen grafts, we like Varady et al (12) found no cellular lining in the Gore-tex lumen and agreed them that without supporting network to supply nutrients, luminal epithelium could not survive in the urinary tract.

The results presented in this paper, contradictory to the results of Varady et al, indicate that there is no potential for ureteral replacement by Gore-Tex tube graft.

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SPONTANEOUS DISSECTION OF LEFT MAIN CORONARY ARTERY EXTENDING INTO ANTERIOR DESCENDING AND FIRST DIAGONAL ARTERIES IN YOUNG WOMAN WITH SURVIVAL: A RARE CASE OF MYOCARDIAL INFARCTION AND CARDIOGENIC SHOCK

Ender Semiz* • Selim Yalçınkaya* • Deniz Kumbasar* • Necmi Değer*

SUMMARY

Spontaneous left main coronary artery dissection (SLMD) is quite rare. We report a case of SLMD extending into mid-left anterior descending and mit-first diagonal arteries in a young woman presenting with acute antero-lateral myocardial infarction and cardiogenic shock. Aggressive medical treatment including thrombolytics and intra-aortic balloon pumping (IABP) were all that could be done. She has been well and uneventful for a 5 month follow-up. To our knowledge, our patient with SLMD is the unique cardiogenic shock survivor with medical treatment in the literature. Thrombolytic therapy and IABP, probably due to augmented coronary perfusion pressure, might be used to maintain late patency of SLMD.

Key Words: *Spontaneous coronary dissection, left main coronary artery, acute myocardial infarction*

Spontaneous coronary artery dissection (SCAD) is an unusual pathology. Precise incidence, etiology, pathogenesis, ant treatment could not have been clearly established (1). Pretty first described SCAD in 1931 (2), in a 42-year-old woman who died following chest pain. Since 1931, only 129 cases of SCAD have been reported in the English literature (3-9), of which 92 (75%) were in women and 33 (25%) in men. Left anterior descending artery (LAD) was involved in 80% of cases, the majority of the rest involved the right coronary artery (RCA) and a small number of circumflex (10). Left main coronary artery (LMC) was involved approximately in 15% of cases (1,11). Although usually fatal, there are reports of successful medical and surgical intervention. In this article, we report the clinical course of a patient with SLMD extending into mid-LAD and mid-first diagonal arteries, presenting with non-fatal acute myocardial infarction and cardiogenic shock.

CASE REPORT

A 46-year-old woman was referred from another health center to our emergency with a diagnosis of acute antero-lateral myocardial infarction for which she received thrombolysis. She had been previously well, until she felt sudden retrosternal chest pain associated with shortness of breath. She was clammy and pale with a heart rate of 146 beats per min with a blood pressure of 50/0. Her electrocardiogram revealed an acute antero-lateral infarct. Cardiac enzymes were consistent with acute myocardial infarction (peak CPK 1174 U, MB fraction 282.5 U). An intra-aortic balloon pump was inserted and intravenous dopamine was started with a diagnosis of cardiogenic shock. Twenty minutes later she went into ventricular fibrillation and she was defibrillated. She was bolused with 100 mg of lidocaine and started on an intravenous drip at 3 mg per min. Her blood pressure was improved to 130/70 and heart rate was normal. With intravenous heparine ant nitroglycerine administraiton, she was well till the next day when she began to express angina. She was taken to the catheterization laboratory with a diagnosis of post-infarction angina. Coronary arteriography showed a longitudinal dissection flap originating in tle LMC

* Akdeniz University School of Medicine, Department of Cardiology

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extending into mid-LAD and mid-first diagonal arteries (Figs 1,2). The other coronary artery segments were completely normal. Left ventriculography revealed apical dyskinesis (Figs 3A, B). Following informed consent, percutaneous revascularization was decided to be attempted. A 0.014 inch coronary guidewire (ACS) was introduced via the ostial LMC but could not be advanced from mid to distal LAD. The procedure was given up and the patient was taken to the intensive coronary care unit. Urgent coronary artery bypass surgery (CABG) was refused by the patient. Medical follow-up was begun. Arterial sheats and intraaortic balloon catheter were removed approximately 85 hr from time of admission. The patient had no angina for 18 days while treating with aspirin 320 mg PO daily, mononitrates 40 mg PO bid, diltiazem 30 mg PO qid, and low-molecular-weight heparine (nodroparine) 15.000 U SC bid. She was discharged from the hospital. Repeat coronary arteriography, to learn whether the dissection healed or not, was performed after 3 months. Angiograms revealed the similar apperance of dissection of the previous ones. She was symptom-free. Radionuclide imaging study with exercise stress testing done prior to discharge showed an anterior and apical defect consistent with infarction but no evidence of ischemia. Following discharge, she has been well and uneventful for a 5 month clinical follow-up.

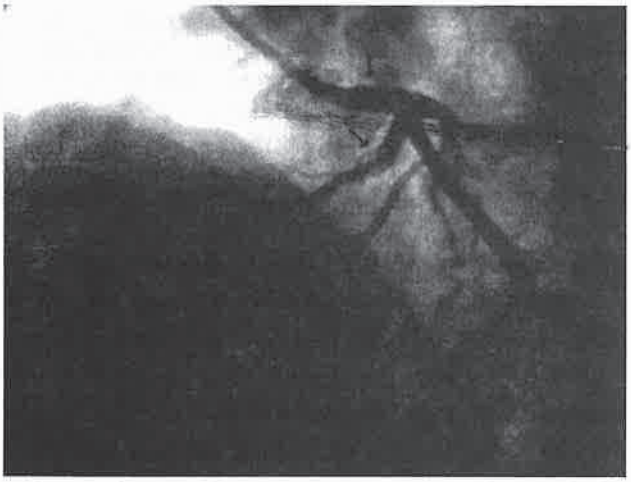

Fig. 1. Selective coronary angiography, left anterior oblique view with cranial slant, demonstrating the dissection originating in the left main coronary and extending into the left anterior descending and the first diagonal arteries (arrows).

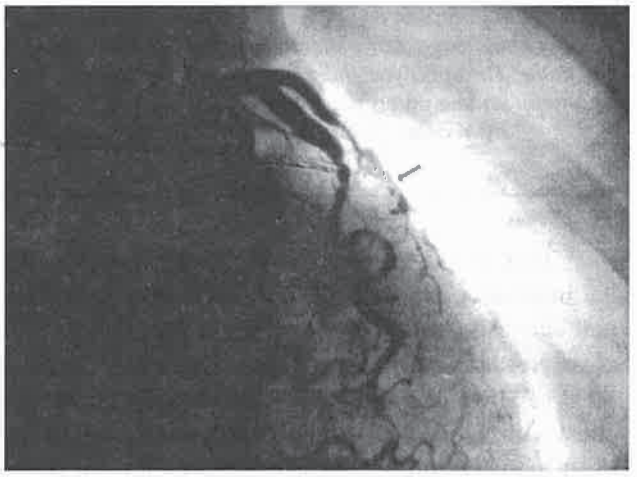

Fig. 2. Selective coronary angiography, right anterior oblique view, revaling the dissection originating in the left main coronary and extending into the left anterior descending and the first diagonal arteries (arrows).

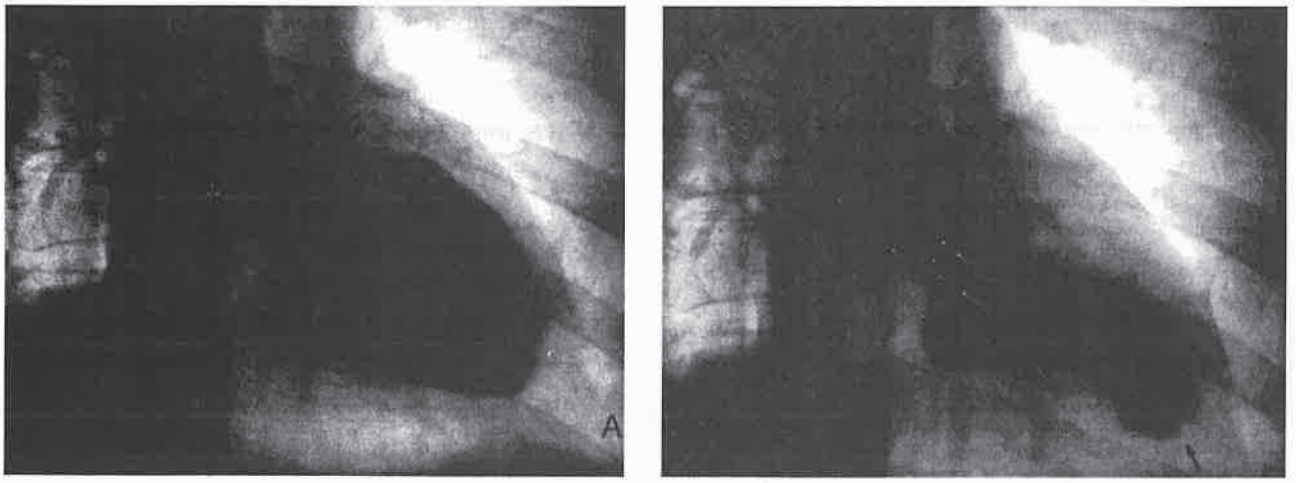

Fig. 3. End-diastolic (A) and systolic (B) left ventriculograms, right anterior oblique projection, revealing apical dyskinesis (arrow)

DISCUSSION

SCAD is a rare condition seen particularly in young women with few or no risk factors for coronary heart disease (1). Except a few cases diagnosed at coronary arteriography, most of them are diagnosed at autopsy (12). Sudden death, which occurs in 75% of patients, has been the most common presentation (1). The average age of women and men is 44 (range 17-69) and 47 (range 18-63) years, respectively (5). The etiology of SCAD is controversial. It has been associated with pregnancy and the postpartum period. Alterations in the protein and acid mucopolysaccharide content of the media in the arteries and collagen degeneration have been postulated to be the mechanism (13,14). Renin release from the chorion has been implicated to lead coronary spasm and dissection (15,16). It has also been reported that vasa vasorum rupture and hemorrhage could be the primary cause in triggering dissection (17,18). Cystic medial necrosis was shown in some cases (19,20). Arteriosclerotic heart disease, Marfan Syndrome, Kawasaki disease, systemic lupus eritematosus, the use of oral contraceptives, blunt chest trauma and exercise were reported to be other possible etiologic risk factors (21-25). Our patient had none of these factors.

SCAD is usually diagnosed by angiography. True and false lumen separated with a radiolucent line can be seen in some cases. The diagnosis has also been made by intravascular ultrasound (26) and transesophageal echocardiography (27) in rare case.

Despite medical or surgical therapy was believed to be the only two options in the treatment of SCAD, deployment of coronary stents guided by ultrasound has also been proposed (28). Some authors reported good results with medical therapy alone (4,7,22), while some recommended CABG for all patients (1,29). Treatment with thrombolytics in 5 cases has been reported (5). Thrombus may lyse in the false lumen allowing the true lumen to expand and coronary blood flow may restore. Our patient had also been administered intravenous streptokinase 1.500.000 U before admission to our emergency. Aortic counter-pulsation, due to augmented perfusion pressure, has been shown to improve late patency of the occluded coronary artery in patients with early failure of thrombolytic therapy (30). Intraaortic balloon pumping (IABP) in the early hours of infarction might be another contributing factor to maitain the vessel patency in our patient.

The greatest caution to treat the LMC dissection must be paid. It may result with sudden death, myocardial infarction, cardiogenic shock or intractable arrhytmias (1,31,32). CABG is often technically challenging due to the difficulty in ensuring bypass into the true lumen (1). If the acute infarction is largely completed, as in our case, delaying surgery to allow time for the dissection to heal and clinical reassessment of the need for surgery may be considered. Table 1shows the previously reported survivors of SLMD that had been diagnosed antemortem. It is noteworthy that all of the patients are women. Our case is the second example of survivor for SLMD that extends both into mid-LAD and mid-first diagonal artery. To our knowledge, our patient who has been surviving with medical treatment is the unique one in the literature, presenting with acute antero-lateral myocardial infarction and cardiogenic shock. Emergency CABG offers the only realistic chance for survival in a dissection involving LMC. However, patients who completed an acute event in the absence of significant stenosis at the angiogram and who has no ongoing ischemia may benefit from medical therapy temporarily.

In conclusion, SCAD as the cause of an acute myocardial infarction should be ruled out in healthy women with no risk factors for ischemic heart disease. Coronary arteriography should be considered early to diagnose the problem. Otherwise could not be urgently revascularized because of any reason, medical therapy including thrombolytics and IABP may also be considered as an alternative for the treatment of SLMD, although a 5 month clinical follow -up might be a short time period to decide whether this approach, to what extent, would lengthen the survival of the patient or not.

Table 1: Spontaneous left main coronary artery dissection survivors*

Reference	Age	Vessel location	Therapy
Razavi (1975)	31, F	LMC, LAD, LCX	CABG
Chokron (1983)	47, F	LMC, LAD	Medical
	34, F	LMC	Medical
Vicari (1986)	33, F	LMC, LAD, LCX	Clot extrusion
Keon (1987)	41, F	LMC	CABG
Thayer (1987)	39, F	LMC	CABG
Boyd (1988)	33, F	LMC	Clot extrusion
Himbert (1991)	41, F	LMC	Medical
Alvarez (1991)	34, F	LMC, LCX	CABG
Ellis (1994)	35, F	LMC, LAD, LCX	CABG
asalodos (1994)	40, F	LM, LAD, 1st DA	Medical
Leclercq (1996)	49, F	LMC, LAD, LCX	Medical
Present Case	46, F	LMC, LAD, 1st DA	Medical

* Abbreviations: LMC = Left main coronary artery, LAD = Left anterior descending artery, LCX = Left circumflex artery, DA = Diagonal artery, CABG = Coronary artery bypass surgery.

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THE DEVELOPMENT OF PULMONARY HYPERTENSION IN A CHILD WITH PORTAL HYPERTENSION: AN UNUSUAL ASSOCIATION

H. Ercan Tutar* • Nurten Girgin** • Aydan Kansu*** • Semra Atalay**** • Nahide Altuğ*****
Ayten İmamoğlu******

SUMMARY

The development of pulmonary hypertension in patients with portal hypertension is a rare association. The exact cause of pulmonary hypertension in these patients remains obscure at present. We present details of a patient with this association who eventually died from pulmonary hypertension and acute right-sided heart failure. We further discuss the etiologic and pathophysiologic mechanisms responsible for this rare entity.

Key Words: *Childhood, portal hypertension, pulmonary hypertension*

The association of pulmonary hypertension and portal hypertension was first described by Mantz in 1951 (1 review). It is a rare association with a prevalence of 0.25% to 2% in autopsy and clinical series (2-4). Most patients reported in the literature have been adults with liver cirrhosis caused by a variety of etiologies (1,3,4). Limited numbers of pediatric cases most of them with extrahepatic portal hypertension have been reported (5-8). We now present details of a boy who developed severe pulmonary hypertension during the course of idiopathic portal hypertension.

CASE REPORT

He was born at term to healthy parents after an uneventful pregnancy and delivery. He appeared to be normal until age two years, when he was admitted to Social Security Hospital because of hematemesis. He had been hospitalized and blood transfusion was given. In 1986 at three years of age he had second episode of hematemesis and he had been hospitalized at the same hospital. He had undergone splenectomy. We couldn't obtain any medical record belonging that period.

He remained asymptomatic until age seven years, when he was admitted to our hospital, because of the third episode of hematemesis. The chief findings on physical examination were; growth retardation, pallor and a grade 2/6 soft systolic ejection murmur at the upper left sternal border. Laboratory studies revealed; hypochromic, microcytic anemia, normal liver function tests including alanine amino transferase (ALT), aspartate amino transferase (AST) and prothrombin time. Abdominal ultrasonography (USG) showed minimal coarsening of liver parenchyma and a patent portal venous tree. A grade 2/4 esophageal varices were demonstrated by endoscopy. Hepatitis B virus markers were negative, and serum alpha 1-antitrypsin, alpha fetoprotein, ceruloplasmine and, urinary copper all were found within normal limits. Chest x-ray and electrocardiography were normal. Light microscopic examination of liver biopsy showed slight hepatocyte degeneration, minimal fibrosis into the parenchyma without evidence of cirrhosis. Propranolol was given at a dose of 1 mg/kg/day and oral iron therapy was started. He was followed periodically as an outpatient

* Dept. of Pediatric Cardiology, Ankara University Medical School, Resident
** Dept. of Pediatric Gastroenterology, Ankara University Medical School, Professor
*** Dept. of Pediatric Gastroenterology, Ankara University Medical School, Specialist
**** Dept. of Pediatric Cardiology, Ankara University Medical School, Associate Professor
***** Dept. of Pediatric Cardiology, Ankara University Medical School, Specialist
****** Dept. of Pediatric Cardiology, Ankara University Medical School, Professor

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with 3 to 6 months intervals until age 11 years and remained asymptomatic. Physical examination revealed only slight growth retardation and his liver function tests were within normal limits. At 11 years old, ultrasound examination of the abdomen combined with Doppler was reported as showing "minimal coarsening of liver parenchyma, periportal fibrosis, patent portal venous tree, but dilated coronary vein with hepatofugal blood flow".

He remained out of follow-up until October 1995. At 12 years of age, he was readmitted to our hospital because of hematemesis and cough. He suffered from easy fatigability and exertional dyspnea for the last 6 months. Physical examination revealed slight growth retardation, a soft systolic ejection murmur at the upper left sternal border, and a fixed splited second heart sound with accentuation of the pulmonic component. Chest roentgenogram showed increased prominence of the main pulmonary artery (PA) segment, with normal overall heart size (Fig. 1). An ECG showed right axis deviation and right ventricular (RV) hyperthrophy. An echocardiogram demonstrated an enlarged RV, a dilated main PA and its major branches, a minimal pulmonary valvar regurgitation and pulmonary hypertension. Right and left heart catheterization was carried out and simultaneous pressure recording were done. The chief findings were; elevation of RV pressures, severe pulmonary hypertension without evidence of intracardiac shunt (Pressures (mmHg); RV: 80/12, PA: 80/37, Aorta: 102/72). Right ventricular angiography revealed a large RV and markedly dilated central pulmonar vessels. Twenty-four hours Holter recording of the patients was normal. Arterial blood gas analysis was normal. Ventilation-perfusion scintigraphy of the lung demonstrated no evidence of thromboembolism. Complete blood count, liver function tests were within normal limits. A Doppler USG of the abdomen showed periportal fibrosis, a dilated coronary vein which had hepatofugal blood flow. The liver biopsy revealed a granular degeneration of hepatocytes, a minimal parenchymal fibrosis without evidence of cirrhosis. Propranolol therapy was stopped and captopril was started at a dose of 1 mg/kg/day because of pulmonary hypertension. Three months later, he had syncopal episode at home and he was hospitalized. Six days after his admission he had second syncopal attack during micturition after complaining of severe chest pain. We noted a grade 3/6 holosystolic murmur at the

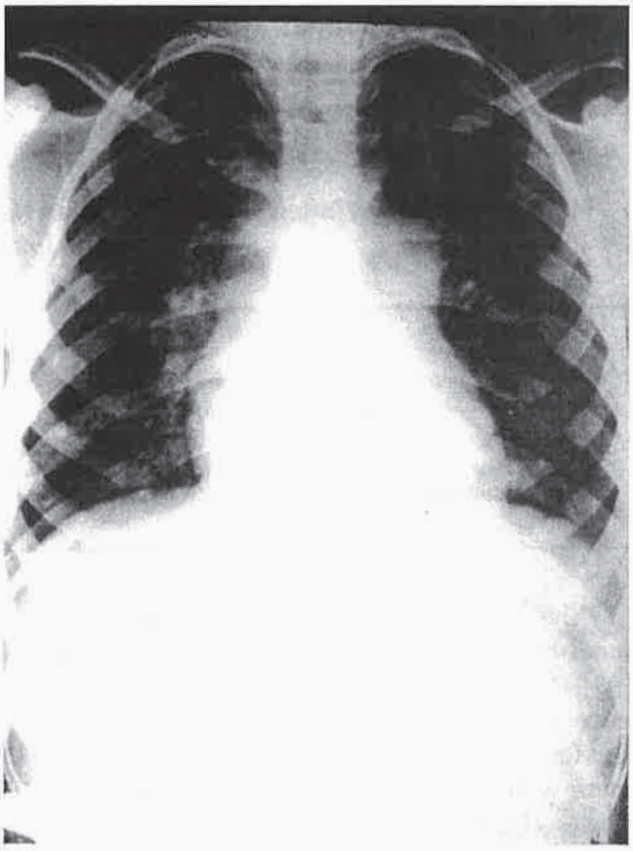

Fig. 1. Chest roentgenogram from patient when pulmonary hypertension was diagnosed showing increased prominence of the main pulmonary artery segment.

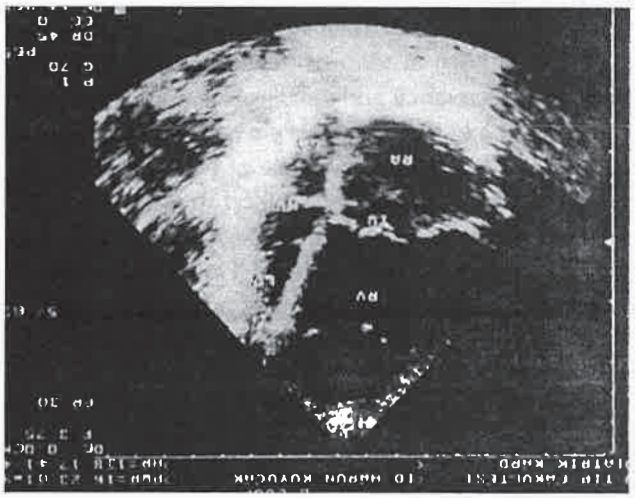

Fig. 2. Apical four-chamber two-dimensional echocardiogram from patient shows markedly dilated right heart chambers with a shift of the interventricular septum toward the left
RA: right atrium, RV: right ventricle, LA: left atrium, LV: left ventricle, TV: tricuspid valve, MV: mitral valve.

lower left sternal border which had not been audible before. An ECG showed inferior and widespread anterior subendocardial ischemia in addition to previous ECG findings. Chest x-ray showed enlarged cardiac silhouette. An echocardiogram revealed markedly dilated right heart chambers with a shift of the ventricular septum toward the left (Fig. 2), a grade 2-3/4 tricuspid regurgitation, minimal pulmonary regurgitation and, a dilated central pulmonary arteries. A biochemical survey showed increased CK-MB, AST and LDH values. We thought that the patient had acute right heart failure with myocardial ischemia. We stopped captopril treatment because of hypotension and we started digoxine and furosemide because of right heart failure. Two days after his syncopal attack, he died suddenly after complaining chest pain and breathlessness. Resuscitation was unsuccessful. Postmortem needle biopsy of the lung was not informative about the degree of pulmonary hypertension and the liver biopsy findings was not different from the previous biopsies.

DISCUSSION

The development of pulmonary hypertension in patients with portal hypertension has been observed occasionally in adults but rarely in children. Most of the reported patients has been diagnosed after death (1,3,6,7). In adults, the cause of portal hypertension is mostly liver cirrhosis (1,3,4), but in the childhood cases congenital or early infantile obstruction of the portal vein is the most common cause of portal hypertension (6-8). Cases with other etiologies (e.g. idiopathic portal hypertension, Budd-Chiari syndrome) were also reported (9). We couldn't find any cause for portal hypertension, thus we used a term "idiopathic" portal hypertension in this patient. Whatever the underlying etiology of portal hypertension, the development of pulmonary hypertension is a rapidly fatal complication with a mean survival of 15 months after diagnosis (1). Especially in children with syncope the course is very rapid, usually leading to death within a few months (6). Such a rapid progression was seen in the present patient. Almost invariable, portal hypertenson either preceded or was diagnosed concurrent with pulmonary hypertension in these patients. The average interval between the onset of portal and pulmonary hypertension was reported 5.7±4.8 years (1). Our patient's symptoms related to pulmonary hypertension were begun 9 months ago before his death. When he was 7 years of age, a grade 2/6 systolic ejection murmur at the upper left sternal border was noted on his medical record. We assume that it might be a first clinical sign of developing pulmonary hypertension, even his chest x-ray and ECG was normal at that time. The causative mechanism of pulmonary hypertension is not certain at present. The hypothesis that some unknown vasoconstrictive or toxic substances originating from the splanchnic region may reach the pulmonary vasculature through the portosystemic shunts and cause vasoconstriction and/or endothelial damage of the pulmonary arterial system seems to be a more likely explanation (1,5-7). Substances blamed include serotonine, histamine, thromboxanes, angiotensin I, elastase and various dietary factors (5-7,10). Postmortem pothologic examinations of the pulmonary arterial tree suggest that thrombosis, whether in situ or embolic may contribute to the pulmonary hypertension observed in patients with portal hypertension. Portal vein thrombus may even be a source of thromboemboli via collateral venous return (7,8).

Although, despite these explanations why does not pulmonary hypertension develop in all patients with portal hypertension is not known. The answer could be the known genetic predisposition for the process (6).

Although rather rare, patients with portal hypertension are at risk of developing pulmonary hypertension. Therefore, these patients should be routinely evaluated by ECG, telecardiography and echocardiography at regular intervals even they are asymptomatic.

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IDIOPATHIC PULMONARY HEMOSIDEROSIS AND ARTHRITIS (Case Report)

Serpil Savaş* • Ayşe Küçükdeveci* • Yasemin Keskin** • Ayşe Sertçelik***
Gülay Dinçer*** • Tansu Arasıl****

SUMMARY

Idiopathic pulmonary hemosiderosis (IPH) is a rare condition of unknown etiology. Four cases of IPH associated with arthritis were reported before. In this paper, we report another patient with IPH and arthritis.

The clinical picture of the present case, with asymmetrical, transient, migratory and nondestructive arthritis, differs from the previously reported patients who all had rheumatoid-like arthritis.

Key Words : *Alveolar hemorrhage, idiopathic pulmonary hemosiderosis, rheumatoid arthritis*

Idiopathic pulmonary hemosiderosis is manifested by the clinical triad of hemoptysis, diffuse pulmonary infiltrates, and iron deficiency anemia (1). IPH has been identified as a cause of alveolar hemorrhage in a small number of cases, mainly by exclusion criteria (2). Exclusion criteria are collagen vascular diseases (SLE, Goodpasture's syndrome and systemic vasculitides), infection, coagulopathy and haemodynamic dysfunction (3,4). After the first report about the relationship between IPH and arthritis in 1962 (5), similar 3 cases were reported by Ognibene (6), Smith (7) and Lemley(8).

Another patient with IPH and arthritis will be presented here and clinical picture will be discussed.

CASE REPORT

We describe a 30-year old man who had arthritis since age 10 and who developed IPH triad at the age of 23. He had a history of monoarticular, recurrent and migratory arthritis of the large joints. He had been admitted to the hospital for dispnea, tachicardy and hemoptysis when he was 23. He had had no history of respiratory infection, allergies, coagulopathy, renal or cardiac disease then. Physical examination had shown ralles and reduction of the inspiration sounds, systolic murmur on apex and aortic valve and arthritis of the right knee. Iron deficiency anemia had been detected with Htc 26%, serum iron 10.0 microgram /dl (normal 60-150), and total iron binding capacity 415 microgram/dl (normal 250-400). Urine analysis had been normal. Westerngreen erythrocyte sedimentation rate had been 40 mm/h, rheumatoid factor, antinuclear antibody and antids-DNA being negative. Immune globulins had been found normal. Sputum and the cultures had been negative. Chest radiograph had demonstrated bilateral hilar enlargement and a diffuse reticulonodular pattern with an intrinsic restrictive defect in pulmonary function tests. Wedge resection of lingula had shown alveoli filled with hemosiderin laden macrophages and fibrosis of alveolar septae (Fig. 1). No immunglobulin and complement deposition had been found on the alveolar basal membrane. The diagnosis of IPH had been made then. He had received a short course of systemic prednisolone therapy (64 mg/daily) with a complete cure of all symptoms and signs in a few days. During the past 7 years until admission to our department, dispnea and hemoptysis had never recurred but the joint complaints had persisted. Asymmetrical swelling of his large joints had appeared almost twice monthly and had resolved spontaneously in a few days. When he

* Resident, Ankara University Medical School Department of Physical Medicine and Rehabilitation.
** Associate Professor, Ankara University Medical School Department of Physical Medicine and Rehabilitation.
*** Professor, Ankara University Medical School Department of Physical Medicine and Rehabilitation.
**** Professor, Ankara University Medical School Department of Pathology.
Ankara University Medical School Departments of Physical Medicine and Rehabilitation and Pathology.
Ankara

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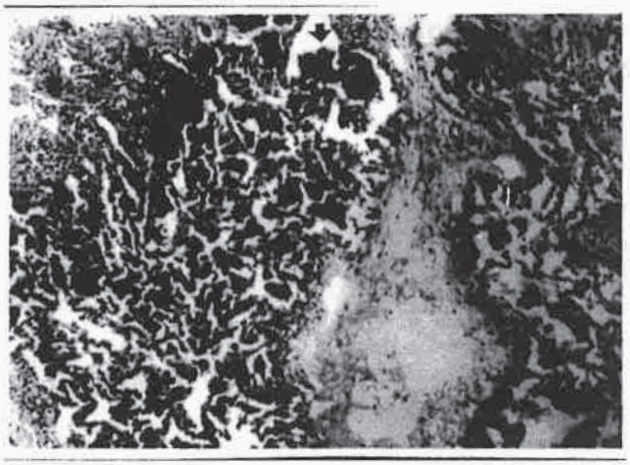

Fig. 1. Wedge resection biopsy specimen of the lung. Alveoli filled with hemosiderin laden macrophages and fibrosis of alveolar septae (H&E x100).

was admitted to our unit in October 1995, his examination was remarkable for swelling, tenderness and erythema of the right elbow. Examination of other joints were normal with no limitation of movement and no deformity. Laboratory data of note included hematocrit 44%, hemoglobin 14.7 gr/dl, WBC 8700/mm3, serum iron 100 microgram/dl, total iron binding capacity 300 microgram/dl,Westerngreen erythrocyte sedimentation rate was 70 mm/h, ASO 200 IU/ml units, CRP 5mg/l , Latex RF , antinuclear antibody, ANCA, cryoglobuline were negative. Immunglobulins were normal. Circulating immune complex was 0.60 (normal 0-5). C3 was 1.09 gr/l (normal 0.5-0.9), C4 was 0.43gr/l (normal 0.1-0.4). Protein electrophoresis included Albumin 35.8% (normal 56-68), Alpha 1 globulin 8.3% (normal 2.5-4.5), Alpha 2 globulin 24.6% (normal 9-14), Beta globulin 15.6% (normal 8-12), Gamma globulin 15.6% (10-18). Urine analysis fibronogen, renal and liver function tests were normal. A reticulonoduler pattern was detected both by chest x-rays (Fig. 2) and chest computerized tomography (Fig. 3). Knee, foot, ankle and hand X-rays were normal showing no erosions, no osteoporosis, no sclerosis. While he was in the hospital he developed arthritis of his right knee (Fig. 4). 175 mg Indomethasine was begun daily. An arthroscopic biopsy was taken from the right knee. Histopathologic findings showed no iron deposits with many papillary vascular proliferation, infiltration of the histiositic and mononuclear cells in the fibrostoma . He developed arthritis of his right ankle after 3 days following the biopsy and it resolved in a week. Chloroguine (250 mg/daily) was started and the patient was taken to follow up.

DISCUSSION

The etiology of IPH remains unknown, but immune mechanisms are thought to be involved (9). There are case reports of IPH in patients with coeliac disease , autoimmune hemolytic anemia, rheumatoid arthritis, Ig A monoclonal gammapathy and autoimmun thyrotoxicosis (10). The clinical triad of IPH occurs in many autoimmune disorders such as Goodpasture's syndrome, SLE, systemic vasculitides and these diseases show the same nonspesific pathological findings of intraalveoler hemosiderin laden macrophages and interstitiae fibrosis (8). IPH has been identified mainly by the exclusion criteria (3,4) and

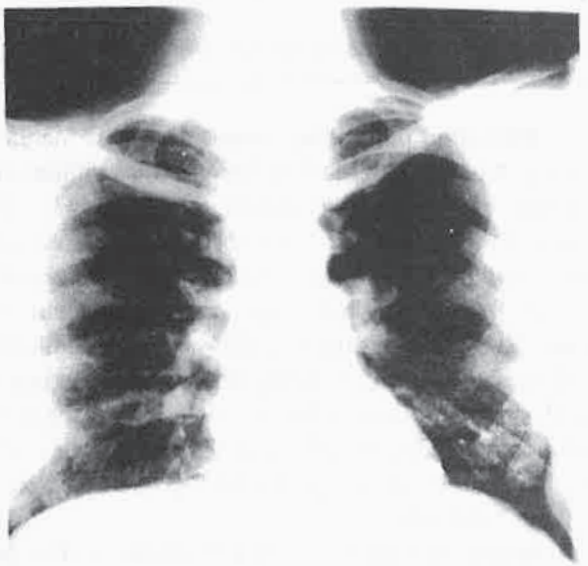

Fig. 2. Chest roentgenogram showing a reticulonodular pattern.

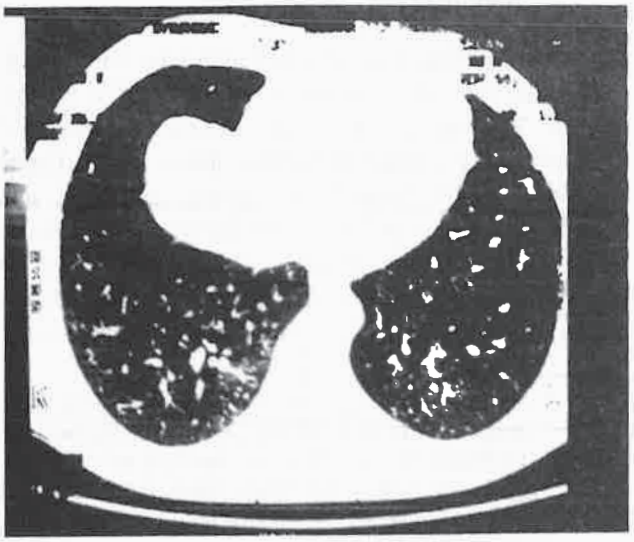

Fig. 3. Chest computerized tomograpy showing a reticulonodular pattern.

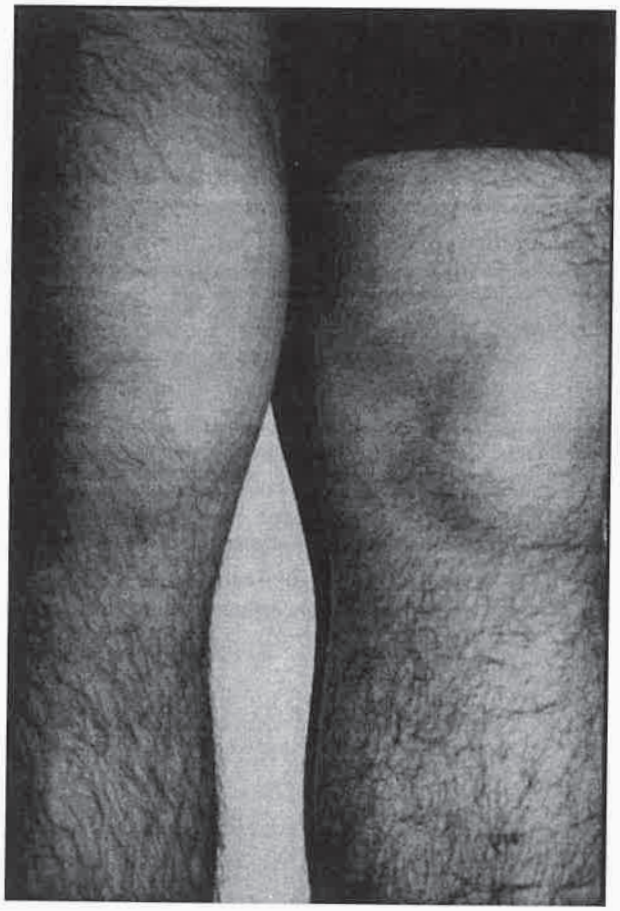

Fig. 4. Arthritis of the right knee.

the use of this term should be restricted to those cases in which there is neither extrapulmonary disease nor the presence of more well defined histopathology such as vasculitis or granulomata (1,5).

Our patient had IPH and arthritis together. The diagnosis of IPH was made by the clinical triad and the histopathological findings of the lung . He had no other systemic disease and there were no immun deposition on the alveoler basal membrane. He have had only one haemoptysis attack. It is known that IPH has a variable course and long term spontan remissions of respiratory symptoms, such as our case, were also reported before (11).

Four previous cases who had rheumatoid or rheumatoid-like arthritis with IPH has been reported (8). Present case differs clinically from those patients with asymmetrical, transient, migratory and non-destructive arthritis. Large joints were frequently involved and the attacks had responded to periods of rest. He described trauma before the onset of attacks.

Corticosteroids, immunsupression and plasmapheresis had been used in the treatment of IPH (9,10, 11 ,12). But the evaluation of the results is difficult because of the high incidence of spontaneus remissions of the respiratory symptoms (9,10). Chloroguine 200 mgr. daily was started the patient, the duration and the number of attacks were reduced. We prefered to use chloroquine because of its lack of toxicity compared with immunesupressive regimens that have been used in IPH (9).

Several clinical syndromes such as palindromic rheumatism, Behçet's syndrome, Familial Mediterreanean fever (FMF) and intermittent hydrarthrosis are characterised by intermittent inflammatory synovitis, with complete resolution during the intervals between attacks (13,14). Residual joint damage occurs only rarely.

Our patient's attacks resembles palindromic rheumatism. However there is no signs of systemic disease such as hemoptysis in palindromic rheumatism. In Behçet's syndrome , arthritis is a minor manifestation with uveitis, phlebothrombosis, and vasculitis playing major roles. Our patient had no major manifestations of Behçet's syndrome. Joint involvement of FMF is more likely to be expressed as arthralgia but there may be synovitis with acute painful effusions in large joints, such as knee, ankle, hip or elbow. Each FMF attack is accompanied by fever and one or more inflammatory manifestations including peritonitis, pleuritisvor pericarditis. Our patient had no fever during attacks and also no symptom of peritonitis or pleuritis. In intermittent hydrarthrosis recurrent effusions occur in the third to fifth decade. There is usually minimal discomfort or signs of inflammation and it is usually monoarticular. Laboratory studies are normal including ESR even during attacks. Our patient's laboratory and the clinical findings were not consistent with intermittent hydrarthrosis.

Diferantial diagnosis of this recurrent arthritis attacks might also include rheumatic fever, hemochromatosis,sarcoidosis and amyloidosis. However neither his clinical picture nor laboratory findings were not consisted with these diseases.

We conclude that this case is not only a rare occurrence of IPH and arthritis together but it is unique because of the characteristics of the arthritis completely differing from the previously reported four cases.

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DE NOVO ACUTE MYELOBLASTIC LEUKEMIA WITH SQUAMOS CELL LUNG CANCER (Case Report)

Muhit Özcan* • Harun Akar** • Önder Arslan*** • Meral Beksaç**** • Osman İlhan*

SUMMARY

Although secondary leukemia risk is enhanced following cytotoxic therapy for lung cancer,the coexistence of AML and squamous cell lung cancer is very rare. We report here a patient who suffered from de novo acute myeloblastic leukemia (AML) and squamous cell lung cancer simultaneously. After the achievement of complete remission for AML , lung cancer was treated with radiotherapy.Exposure to asbestos,known as an etiologic agent both for lung cancer and AML, is also interesting in this case.

Key Words: *Acute myeloblastic leukemia, asbestos, squamous cell lung cancer*

Epidemiological studies suggest that environmental, occupational and genetic factors play a role in the pathogenesis of AML. The overall annual incidence is 2.3 per 100000 with an increasing incidence related with age (1). Although reports of acute leukemia secondary to cytotoxic therapy for solid tumors are not less common (2), the coexistence of both malignancies is very rare. We describe here a case of de novo AML with squamous cell lung cancer.

CASE REPORT

A 67-year old male heavy smoker was admitted to the hospital because of malaise, weight loss, cough, sputum, hemoptysis and dyspnea. There was a three-months history of fatigue and dyspnea. He was from the rural part of Turkey where the frequency of environmental exposure to asbestos is high. He had a history of chronic obstructive pulmonary disease and hypertension. One of his children had died of Ewing sarcoma.

Physical examination revealed pallor and hepatomegaly. The breath sounds were diminished at the left base and expiratory phase was long. The hematological examination results were as follows; hematocrit 21.1% , WBC 1,8x109/ L, platelet count 108 x109/L. There were 15 % blasts and rouleaux formation on peripheral blood smear. Erythrocyte sedimentation rate was 130 mm/hr.

The bone marrow smear revealed 78 % blast cells.Cytochemical examination demonstrated strong positivity for myeloperoxidase. PAS reaction was negative. The diagnosis of AML (M2) was made. The immunophenotyping revealed HLA DR: 75 %, CD 34 : 64%, CD 33: 47%, CD 13:41% , CD 7: 21% positivity in blast cells which confirmed the diagnosis of AML. Left costophrenic angle was blunted with left lower lobe infiltrate upon chest x-ray. Computed tomography (CT) of the thorax revealed a solid lesion in the left lower lobe with atelectasis and minimal pleural effusion. Bronchoscopy showed a vegetating mass. Transbronchial biopsy was performed and squamous cell lung cancer was diagnosed. The stage of the lung cancer was T2NOMO.

It was decided to treat the leukemia first. The patient was treated according to ADE protocol. The regimen consisted of Cytosine Arabinoside 100 mg/m^2 /12 hr. on the 1st-10th days, Daunorubicine 50 mg/m^2 on 1st., 3rd. and 5th days, Etoposide 100 mg/m^2 between 1st-5th days. During the treatment and aplasia period, three units of platelet from random donors and ten units of whole blood was trans-

* Associate Professor, Dept of Hematology, Ankara University Medical School
** Resident, Dept of Hematology, Ankara University Medical School
*** Senior Fellow, Dept of Hematology, Ankara University Medical School
**** Professor, Dept of Hematology, Ankara University Medical School

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fused. The major reason of the frequent transfusion requirement was hemoptysis. The patient refused surgery for lung cancer and radiotherapy was applied in a dose of 5000 cGy starting on the 28th day of the leukemia treatment, while the WBC count was 5.6x109/L and platelet count was 63x109/L. On the same day, bone marrow examination revealed hematological remission.

The mass lesion was still present with an improvement of atelectasis and pleural effusion disappeared in the next CT scan after completion of radiotherapy. On the 57th day, the patient was discharged with the hematological parameters as follows; WBC count 5x109/L and platelet count 86x109/L and bone marrow aspirate showed 2% blast cells.

Post-remission treatment was not given due to the patient's incompliance and the patient was lost to follow up.

DISCUSSION

The secondary leukemia risk is enhanced following cytotoxic therapy for lung cancer. The unusual presentation of our case includes occurrence of two malignancies simultaneously. The coexistence of leukemia and another malign process was first reported approximately 100 years ago. The second patient who was an Alaskan miner with chronic lymphocytic leukemia plus non-small cell lung cancer had been reported by Moertel (3).In their excellent review of the literature up to 1957, a total of 194 cases was collected which were examples of leukemia and lymphoma associated with another primary malignant lesion. Among 2137 patients with leukemia (acute or chronic) who were seen at Mayo Clinic from 1944 to 1953, there were 52 patients who had an additional primary lesion. In 27 of them, the malignant lesion was diagnosed simultaneously with the diagnosis of leukemia. However no coexistence was reported between acute myeloblastic leukemia and lung cancer in that article. In the review of literature, we found four other similar cases reported and successfully treated. One of them included the successful treatment of concomitant acute myeloblastic leukemia and adenoid cystic carcinoma of the palate by Handa et al (4). In that case, authors preferred to remove the tumor after induction therapy of AML. In the other report, Sutedja treated their patient with AML and squamous cell lung cancer with photodynamic therapy following ARA-C/Daunorubicine regimen and autologous bone marrow transplantation (5) . Sheridan reported a patient with simultaneous occurrence of de novo acute myeloid leukemia and diffuse infiltration of bone marrow by carcinoma cells, most likely of breast cancer origin (6). Louvet et al. described a case with adenocarcinoma of the lung and AML (7). According to above data, our case seems to be the third patient who suffered from lung cancer and AML simultaneously.

All sites and histologic types of lung cancer are reported in asbestos-exposed workers (8). Significant concentrations of asbestos was detected in the bone marrow of two cases of AML described by Kishimoto et al (9). In this study, no asbestos bodies were detected in the bone marrow of individuals from the control group with only lung cancer and a causal relationship between exposure to asbestos and AML was suggested. Kagan had identified 13 asbestos workers with lymphoplasmacytic neoplasms (10) . These findings strongly supported the previous observations that asbestos might be a lymphoid system carcinogen.

Association of pleural mesothelioma with non-Hodgkin's lymphoma after exposure to asbestos was reported by Tondini (11) and Hughes (12).

Apart from the rare coexistence of AML and lung cancer, the presence of exposure to asbestos, an etiologic agent both for lung cancer and AML, is an interesting observation.

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PERINEAL ECTOPIC TESTIS
Analysis of Three Cases

Ayhan Karabulut* • Ertan Batislam** • Levent Peşkircioğlu** • Uğur Altuğ***
Cankon Germiyanoğlu*** • Demokan Erol****

SUMMARY

Perineal ectopic testis is an extremely rare condition. Herein we reported three different cases with perineal ectopic testes admitted in last eight years and reviewed the published literature. We also discussed the etiopathogenesis of perineal ectopic testis.

Key Words: *Perineal ectopic testis*

While maldescended testis is the most common anomaly of the male genitalia, about 5% of the cases present as ectopia and only 1% of the testes are located perineally. There are occasional reports of perineal ectopic testes in the literature with less than 200 cases of unilateral disease and only a few about bilateral involvement (1).

CASES

Case 1 : M. Y. , a 13 years old male patient presented with an absent left testicle. His right testicle was of normal size and consistency. The missing left testicle was palpated as a mass of 1.0 x 1.2 x 1.5 cm in the perineum (Figure 1). No accompanying anomaly was found on physical examination and routine urological work-up.

The patient underwent left orchidopexy in September 1985. Histologic features of the ectopic testes were consistent with the age of the child.

Case 2 : B.Ö. F., was a four years old boy with empty hemiscrota. His testicles were palpated in the perineum (Figure 2). A bilateral orchidopexy was performed in August 1987. Histologic examination revealed normal findings.

Case 3 : Y. K., 10 months old boy was referred because of an absent right testicle. On physical examination the left hemiscrotum and its contents were found to be normal, while the missing right testicle was palpated as amass of 0.5 x 0.8 x 1.2 cm at the right side of the perineum (Figure 3). An orchidopexy was performed in April 1992. Biopsy of the ectopic testicle revealed histologic findings consistent with the age of the patient.

DISCUSSION AND CONCLUSIONS

Perineal ectopic testis is a rare congenital anomaly. In 1786 John Hunter first described the condition. Only 105 cases were reported until 1949 (2). Since than, it seems that about one case per year have been reported up to 1990 (1, 3, 4, 5).

The term ectopic testis is used for those testes that have descended normally through the external inguinal ring but misdirected afterwards to take a perineal, prepenile, transverse scrotal, femoral or umbilical position ultimately. Therefore, an ectopic testis is different from a cryptorchid testis that has been arrested at some point along its true line of descent. An important feature is borne in this descriptive differentiation; in contrast with the undescended gonad, the ectopic testis displays normal histology. Thus, it is important to localize an ectopic testis and resume it into its normal localization.

* *Urology resident.*
** *Urologist.*
*** *Assoc. Professor of Urology.*
**** *Chief, Assoc. Professor of Urology.*
The Ministry of Health Ankara Hospital, Clinic of Urology, Ankara, Turkey.

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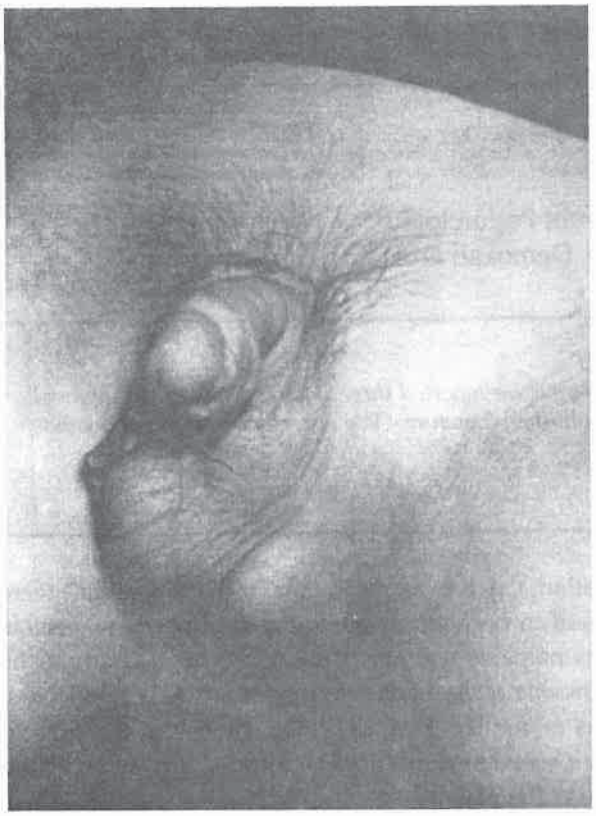

Fig. 1. Left testicle located in the perineum.

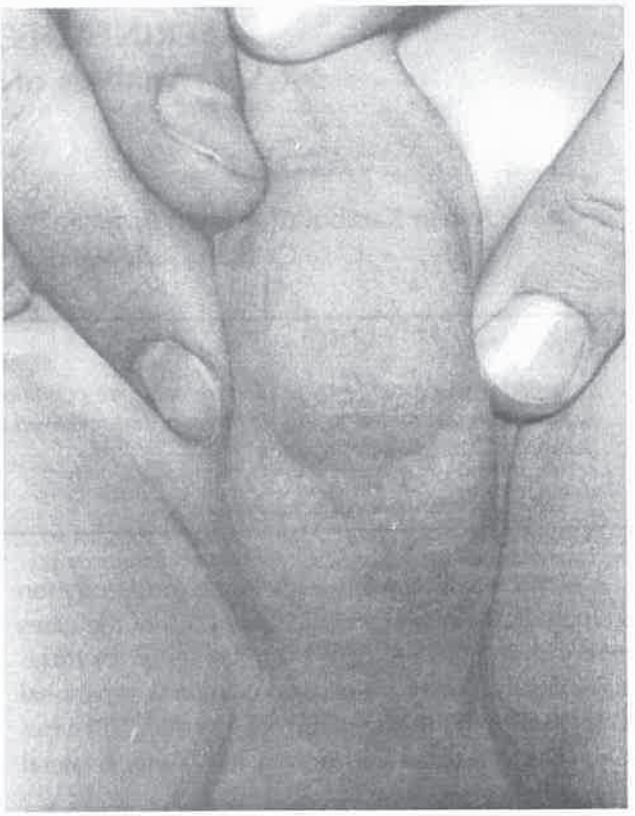

Fig. 2. Bilateral perineal ectopic testes.

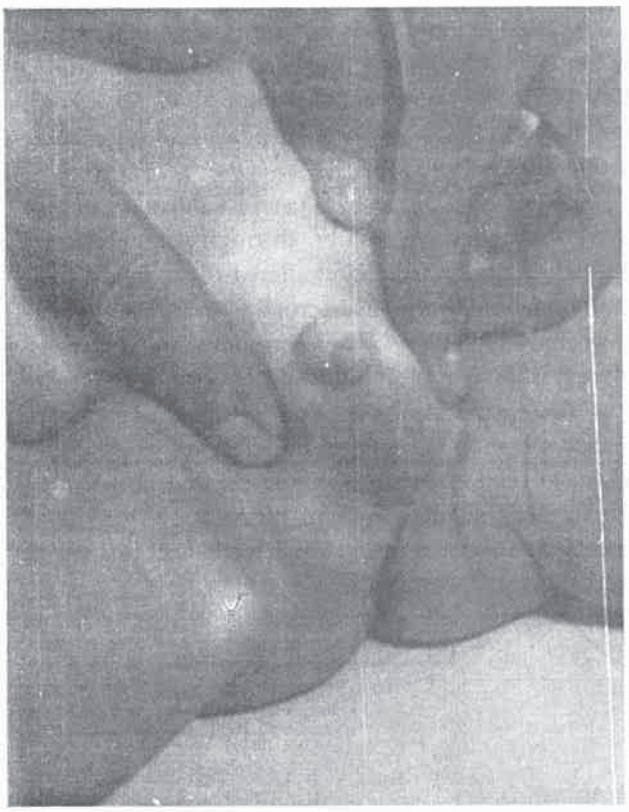

Fig. 3. Right testicle located in the perineum.

The mechanisms underlying the anomaly have been discussed for years especially when the condition seemed even more rare with small numbers of reported cases (6, 7). The well-documented different gubernacular insertions appear as a logical explanation for the perineal descent of a normal testicle.

It is clear that there is no role for hormonal therapy and the treatment is surgical. Delayed treatment will probably cause a compression atrophy in the testis. Such a case was reported by Dieckmann et al in 1988 (8). It is difficult to draw, from that four-years-old case, a conclusion such as ectopic testes differ from undescended testes only in terms of topography and these two groups are similar in histology. This presumption holds only true for superficial inguinal ectopia in which testicular histology resembles that of a cryptorchid testis. In fact, this histological similarity has lead to the conclusion that superficial inguinal ectopia should be regarded rather as a variant of undescended testis instead of ectopia (9).

It seems that ectopic testes will continue to be reported in the literature. The photographic demonstration of these cases has been mentioned by Wong et al. The authors recommend drawing up the thighs forcefully in order to render the testicle visible as a localized bulge (10). This approach is logical for a good documentation.

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SPINAL INTRAMEDULLARY DERMOID TUMORS REPORT OF TWO CASES

Nurullah Yüceer * • Serdar Uğraş ** • Mehmet Bahadır Güven *
Metin Orakdöven *** • Nejmi Kıymaz **** • Ömer Anlar ***** • Burhan Adak ******

SUMMARY

We presented two cases of the spinal intradural intramedullary dermoid tumors (dermoid cysts). Two cases were admitted to the hospital with a history of backache, weakness of the lower extremities and urinary retention. Myelography and computed tomography with contrast were performed to the patients. Myelography showed subtotal obstruction. Computed tomography with contrast media demonstrated low attenuation values. Both intramedullary tumors were subtotally excised by microsurgical techniques. Histopathological examinations confirmed the dermoid tumor. Urinary retention was improved after the operation. Weakness of the lower extremities was also gradually recovered in two cases.

Key Words: *Intramedullary tumor, dermoid tumor, myelography, computed tomography, surgery*

Dermoid tumors (dermoid cysts) occur throughout the scalp, skull, brain, middle ear cavity and spinal cord but almost never in the vertebrae (1,2). Dermoid tumors may be associated with dermal sinuses, tufts of external hair, and focal skin pigmentation (3,4). These tumors may be associated with other malformations of the spinal column such as spina bifida (1).

Spinal dermoid tumors consist of less than one percent of all spinal tumors (5). The medical literature concerning spinal dermoid tumors consists largely of isolated case reports (3,6,7,8,9,10,1112,13). These tumors may be either extramedullary or intramedullary (1,2).

In this article, we presented two cases with spinal intramedullary dermoid tumor and discussed in the light of lrelated literature.

CASE REPORTS

CASE 1

A 20-year-old female was admitted to the Neurosurgical Department with 5 months history of backache, pain in the right leg, difficulty in walking, and urinary retention.

Neurological examination: There was a mild paresis (1/ 5) in her left leg. Reflexes were active and symmetrical without pathological reflexes.

Radiological examination: Radiographs of the thoracic (Th) and lumbar (L) spine were normal. Myelography revealed a lesion at the level of Th11 - L2 and an intramedullary tumor was suspected (Fig. 1). A computed tomographic (CT) scan with contrast showed the tumor with low attenuation values at the level of Th11-L2 (Fig.2).

Operation: A laminectomy from Th11 to L1 was performed and dura was opened. The spinal cord was found to be distended without pulsation. Myelotomy was made about 3 cm size between Th11 and L1. Yellowish fluid was drained. The tumor was smooth or slightly nodular, and encapsulated. The tumor had a lot of hairs. Subtotal resection of thoracolumbar intradural intramedullary tumor was accomplished using microsurgical techniques.

* Assistant Professor, Department of Neurosurgery, University of Yüzüncü Yıl, School of Medicine, Van
** Assistant Professor, Department of Pathology, University of Yüzüncü Yıl, School of Medicine, Van
*** Neurosurgeon, Department of Neurosurgery, State Hospital, Van
**** Resident, Department of Neurosurgery, University of Yüzüncü Yıl, School of Medicine, Van
***** Assistant of Professor, Department of Neurology, University of Yüzüncü Yıl, School of Medicine, Van
****** Assistant of Professor, Department of Physical Therapy Rehabilitation, University of Yüzüncü Yıl, School of Medicine, Van

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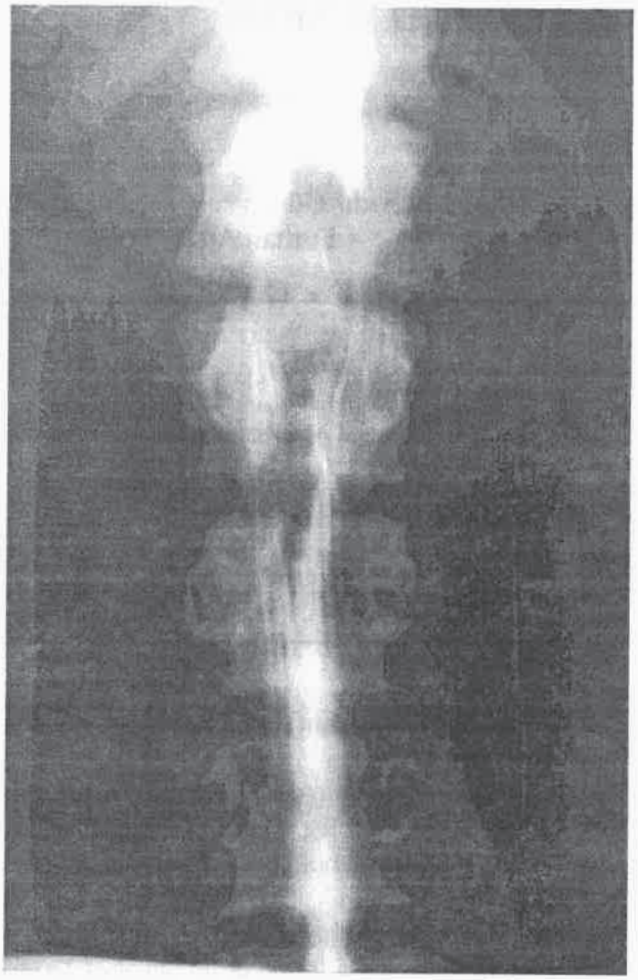

Fig. 1. Myelography, anteroposterior view, demonstrates a fusiform enlargement of the spinal cord at the level of Th11 - L2.

Histopathological examination: There were sebaceous glands and neural tissue under the stratified squamous epithelium with horny layer.

Postoperative course: The patient's monoparesis in the left lower extremity increased after the operation (2 / 5). Urinary retention improved. Physical therapy and rehabilitation was given to the patient 15 days after the operation. Monoparesis was gradually recovered within three months after the operation.

CASE 2

A 22-year-old man was admitted to our clinic with six months history of backache, weakness in the legs and urinary retention.

Neurological examination: There was paraparesis (3 / 5) in his lower extremities.

Radiological examination: Radiographs of the thoracal and lumbar vertebrae were normal. Myelography demonstrated an intramedullary lesion

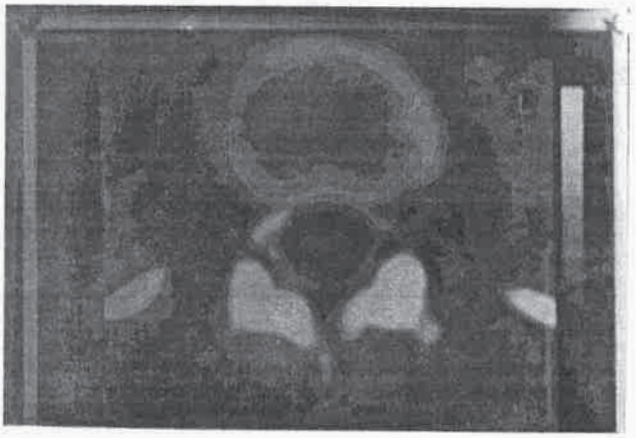

Fig. 2. Computed tomographic scan with contrast media demonstrates the tumor with characteristically low attenuation values.

at the level of Th12 - L2 (Fig.3). CT scan with contrast media showed the tumor with low attenuation values containing several high - density areas at the level of Th12 - L2 (Fig. 4).

Operation: A laminectomy from Th12 to L2 was performed. Dura was opened. A 2 cm myelotomy centered over the lesion was made. Yellowish fluid was drained. We detected a lot of hairs. The tumor was nodular, and encapsulated. The capsule was partly calcified. Thoracolumbar intradural intramedullary tumor was subtotally removed by microsurgical techniques.

Histopathological examination: There were sebaceous glands and neural tissue under the stratified squamous epithelium with horny layer (Fig.5).

Postoperative course: The immediate postoperative course was uneventful. Urinary retention improved after the operation. The patient's paraparesis was gradually recovered six months after the operation.

DISCUSSION

Spinal intramedullary dermoid tumors have been reported at all levels. But, dermoid tumors are seen the most frequently in the thoracic and upper lumbar regions (1,2,4,10,14,15). Dermoid tumors are seen more often in the first and second decades (1,16,17) as in our cases. Dermoids lack sex predilection (1).

The symptoms of these tumors are back and leg pain, and / or slowly progressive impairment of spinal cord function (monoparesis, paraparesis), sphincter dysfunction, and sensory disturbance.

Plain films, myelography, and CT scan and magnetic resonance imaging (MRI) may be used in diag-

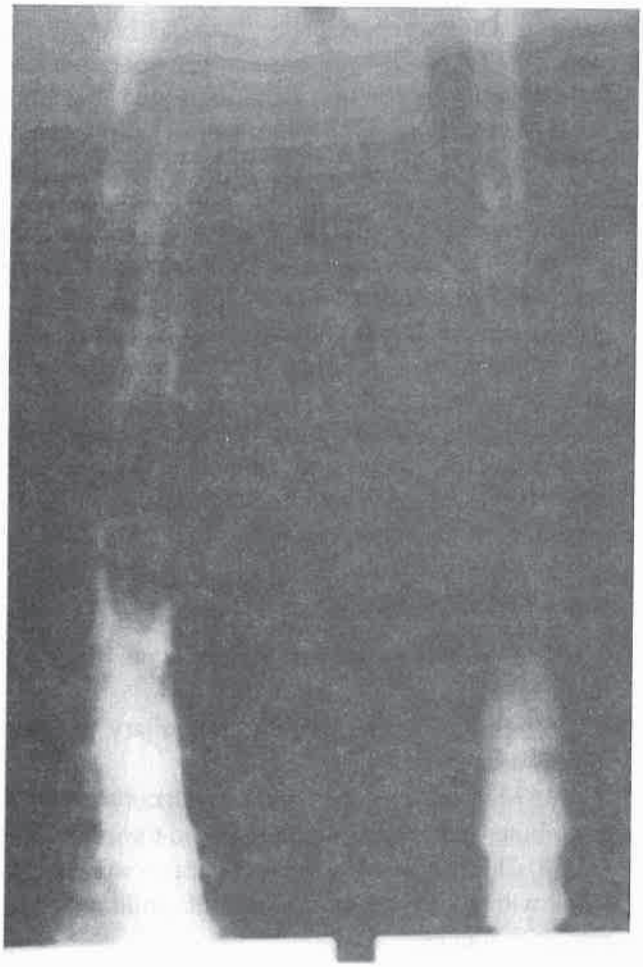

Fig. 3. Myelography revealed an intradural intramedullary tumor subtotally obstructing at the level of Th12 - L2.

nosis of the spinal dermoid tumors. The plain film findings include local widening of the vertebral canal at the level of the lesion, scalloping of the posterior margins of the vertebrae, and flattening of the pedicles. These tumors are usually quite large when they present. Myelography characteristically demonstrates a block or evidence of a significantly compromised thecal sac. The lesion has a characteristic smooth margin and may be either extramedullary or intramedullary. CT scan demonstrates a low density lesion obliterating the vertebral canal and failing to enhance after intravenous contrast injection (4).

Myelography and CT scan can demobstrate the dermoid tumor in most cases, but MRI is now the imaging modality of choice. In our study, myelography revealed subtotal obstruction due to the thoracolumbar intramedullary tumor (Patient 1 and 2). In addition, CT scan showed low attenuation values

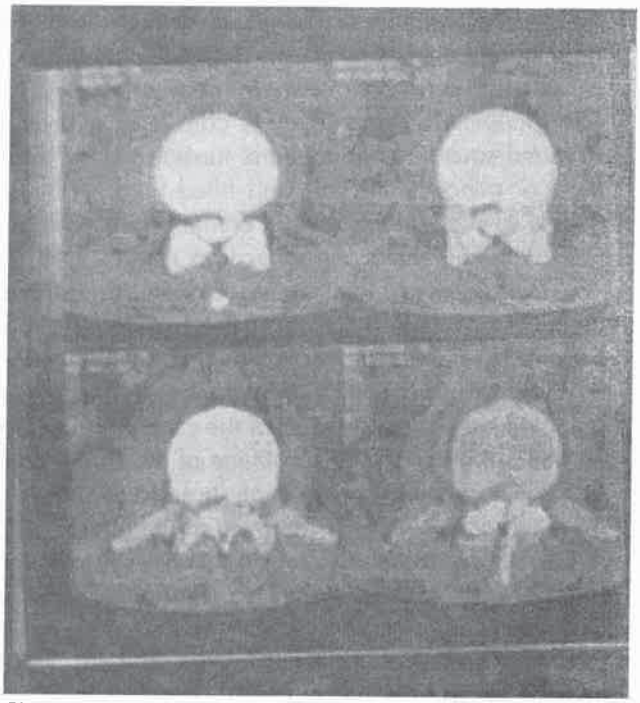

Fig. 4 . Computed tomographic scan shows an intramedullary tumor containing several high - density areas.

(Patient 1 and 2), and high density areas (Patient 2).

Spinal dermoid tumors must be differentiated from the other spinal intramedullary tumors such as ependymoma, astrocytoma, epidermoid cyst, lipoma. The low attenuation values of dermoid can be a useful aid to diagnosis when CT scan is used.

The therapeutic goal in the treatment of dermoid cysts is complete surgical excision of both the cyst lining and its contents. Macroscopically the dermoid

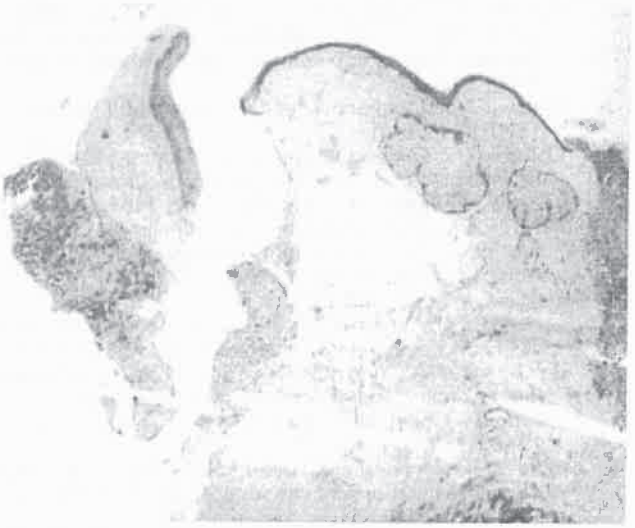

Fig. 5. Histopathological examination: It is seen sebaceous glands and neural tissue under the stratified squamous epithelium with horny layer.

cysts are of variable size and are smooth or slightly nodular and encapsulated. The capsule may be partly calcified. Teeth are rarely encountered (1,2,15). Microscopically dermoid cysts consist of a layer of stratified squamous epithelium surrounded by a thin layer of connective tissue and filled with keratinous debris. Also, the dermoid cyst contains pilosebaceous units with hair shafts and sebaceous glands (2,15,18).

The success of total excision, of course, depends on the location and extent of the individual tumor. Complete removal of the spinal intramedullary dermoid tumors can be difficult in cases with the cyst lining that invaginates the roots of the cauda equina or is densely adherent to the substance of the spinal cord. Use of the operating microscope should allow more of these tumor to be totally removed in the future.

In the present study, we subtotally excised spinal intradural intramedullary dermoid tumor. Decompression was provided. We did not perform total tumor excision to avoid from additional neurological deficits.

During removal of the spinal intramedullary dermoid cysts can contaminate the surgical field and spillage of the cyst content into the subarachnoid space. Seeding of the subarachnoid space with the irritating products from the dermoid cyst can cause severe chemical meningitis. Postoperative aseptic meningitis in the dermoid tumors occurs more frequently when removal of the cyst has been incomplete (8). But, we did not observe postoperative aseptic meningitis in our two cases who were treated by subtotal excision. Operative complications which result from damage to spinal cord and vascular structures are not unique to the surgical removal of intramedullary dermoid tumors. Prolonged and often permanent sphincter disturbance is the most common complication in cases where a spinal intramedullary dermoid tumor has been excised. In this study, there was also sphincter disturbance in our cases and postoperatively improved.

Malignant change in the dermoid tumors is very rare (1,2,9,18). Keogh and Timperley (9) reported a case of invasive atypical hidradenoma arising in a dermoid cyst of the spinal canal.

CONCLUSION

The following results according to our cases may be concluded;

(1) Spinal intradural intramedullary dermoid tumors are rare.

(2) Myelography and CT scan with contrast media contribute to diagnosis of the dermoid tumors.

(3) Clinical recovery in these patients may be provided with subtotal removal without additional neurological deficits.

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