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Relevant Data From All Centers*



*Correction Of Steal Syndrome Of Native Arteriovenous Fistulas In Hemodialysis  
Patients By "Banding" Technique*



*Results Of Tipu On Patients With Primary And Recurrent Hypospadias*



*Long-Term Results Of Artificial Urinary Sphincter Implantation For The Treatment  
Of Urinary Incontinence*



*Subclinical Hyperthyroidism*



*Complication Of Felon Caused By Morganella Morgagni; Case Report*



*Aplasia Cutis Congenita Of The Scalp: Conservative Treatment After Failed Surgical  
Treatment (A Case And Review Of Literature)*



*Lately Formed Uretero-Sacro-Cutaneous Fistula And Giant Intrapelvic Urinoma Due  
To Gunshot Injury*



*Coexisting Transitional Cell Carcinoma And Leiomyosarcoma In The Urinary Bladder:  
A Case Report*

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# ASSESSMENT OF FERTILITY AND INFERTILITY IN BORON-EXPOSED TURKISH SUBPOPULATIONS 6: RELEVANT DATA FROM ALL CENTERS

Bekir Sıtkı Şaylı\* ❖ Meltem Çöl\*\* ❖ Atilla Halil Elhan\*\*\* ❖ Yasemin Genç\*\*\*

## SUMMARY

In this last article, part of a large-scale work to reveal health effects of boron exposure, findings of fertility-infertility states of subjects exposed to borates environmentally and/or occupationally in a country with the world's largest deposits are described. The study covered all centers of borate production, namely, Bigadiç county of Balıkesir; Kestelek village of Mustafa Kemal Paşa county, Bursa; Emet and Hisarcık counties of Kütahya, and Kırka town, Seyidgazi of Eskişehir, an area nearly 350 km long and 150 km wide. Drinking water piped out from springs and wells has boron concentrations 0.2 to 29 ppm (mgB/kg or mgB/l). Dust amount at work-sites was below permissible level of 10 mg/m<sup>3</sup>. The work, questionnaire-based, was realized in field as an observational one. Residents were visited at home and coffee houses in villages and public buildings in towns, and workers at facilities and ore pits without any selection. The inquiry was mainly concerned with marital state and childbearing properties of probands, and of other members in the kindred.

Infertility of the primary type among 2529 probands (participants), as a convenient sample, was 3.1%, changing from 0.0 to 6.5% regarding subpopulations from 12 centers, differences being statistically insignificant. No differences with respect to birth place and professional state as well were revealed either. Pedigree data showed the rate was 3.2% covering 14,320 marriages over 3 generations. No appreciable concentration of infertiles either in subgroups or in so-called 'borate families' in borate towns was observed. Approached as an independent test, marriages of male and female sibs of proband and his (her) spouse ranged from 2.4 to 4.2%. None of these was so far higher than found in different sets of controls and of general population over 50,000 families. Childlessness was found 1.7% among workers versus 4.3% among employees from all facilities, the difference attributable to socio-cultural grounds.

It was then once more concluded that continuous boron exposure at the present level does not affect human reproductive performance adversely primarily and secondarily over 3 generations for at least 60-70 years.

**Key Words:** Boron Exposure, Borates, Fertility, Infertility, 'Borate Families'.

## ÖZET

### Türkiye'de Bor ve Bileşiklerine Maruz Toplum Kesimlerinde Fertilite ve Infertilite Araştırması: 6. Tüm Bulgular

Dünyanın bilinen en geniş bor yataklarına sahip Türkiye'de bor ve bileşiklerine maruziyetin insan sağlığına etkisini belirlemeğe yönelik uzun vadeli bir çalışmaya ilişkin yayınların bu sonucunda bor mineralleriyle çevresel ve/veya mesleki teması bulunan kişilerin fertilite-infertilite durumları anlatılmaktadır. Araştırma bütün bor üretim merkezlerini içine almıştır. Buraları hem geniş maden yataklarının hem üretim tesislerinin yer aldıkları Balıkesir'in Bigadiç ilçesi, Bursa'nın Mustafa Kemal Paşa ilçesinin Kestelek köyü, Kütahya'nın Emet ve Hisarcık ilçeleriyle Eskişehir'in Seyidgazi ilçesine bağlı Kırka beldesi ve civarlarıdır ki aşağı yukarı 350 km uzunluğu, 150 km genişliği olan bir yurt kesimini içlerine alırlar. Ayrıca Balıkesir'in Bandırma ilçesindeki Boraks ve Asit Fabrikaları çalışanlarıyla Susurluk ilçesinin Sultançayırı, Aziziye ve Yıldız köylerinden bir grup eski madenci eklenmiştir. Bölgelerin kaynak ve kuyulardan getirilen içme ve kullanma suları bor içerikleri 0.2-29 ppm (mgB/kg veya mgB/l) arasında değişmekte olup sonucu rakam yerleşim birimleri arasında bilinenlerin en yükseğidir. Ocak ve fabrikaların toz yoğunluğu ise yasayla çizilen 10 mg/m<sup>3</sup> yoğunluk sınırını aşmamaktadır. Ankete dayalı bu gözlemsel çalışma sahada gerçekleştirilmiştir. Katılımcılarla ev ziyaretleri, kahvehane, ofis, dükkan sağlık ocaklarında, işçilerle maden ocağı ve fabrikalarda yüzyüze görüşülmüştür. Cinsel etkinliğe ağırlık veren sorular yanı sıra ailenin öteki bireylerinin evlilikleri de incelenmiştir.

Primer infertilite 2529 probanda göre %3.1 çıkmış, 12 çalışma bölgesinde değişiklik %0.0-6.5 arasında bulunmuştur (farklar anlamlı değil). Katılımcıların doğum yerleriyle meslekleri açısından herhangi fark gözlenemezken pedigr analiziyle üç kuşak boyunca 14,320 evliliğe göre primer çocuksuzlar %3.2, sekonderler % 0.4 düzeyinde kalmışlardır. Gerek bunlar gerekse bor kentlerinin 'bor aileleri' içerisinde infertilite yayılımı ortaya konamamış; bağımsız bir test olarak uygulanan proband ve eşinin erkek ve kız kardeşlerinin evliliklerindeyse %2.3-4.0 gibi benzer infertilite rakamları saptanmıştır. Hiçbiri kontrol ve genel toplum rakamlarından daha yüksek değildir. Tesislerdeki memurlar arasında saptanan % 4.3'lük infertiliteye karşın işçilerin %1.7'lik oranı sosyo-kültürel farklara bağlanmıştır.

Sonuç olarak kuşaklar boyunca en az 60-70 yıl süreyle bor bileşiklerinin ağır ve sürekli baskısı altındaki toplum kesimlerinde çocuk edinme ve evlilik durumları bakımından cinsel performansın primer ve sekonder olarak olumsuz etkilendiği savı desteksiz kalmıştır.

**Anahtar Kelimeler:** Bor Maruziyeti, Boratlar, Fertilite, Infertilite, 'Bor Aileleri'.

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Boron, ubiquitous in the human environment, is an essential micronutrient for normal plant growth and optimal crop as proved by Warington in 1923; although it is herbicide at high doses (1). In animal kingdom it does not show an adverse effect at low concentration but at higher amounts signs of acute toxication appear (2,3). In chronic exposure it exerts deleterious effects, the target organ being testis. At doses as high as 3000-9000 ppm (mg/kg or mg/l) boric acid, a boron compound naturally occurring in environment, and corresponding to 26-68 mgB/kgbw, testes are atrophied and eventually degenerated by as yet not an understood mechanism (2,3). The rats are the most sensitive animal but it is also true for mice, rabbits and even dogs (4,5).

Meanwhile two Russian writers reported that workers exposed to borate dusts and cadmium exhibited reduced sperm count and impaired sexual activity (6,7). However such observations have not been supported. And despite much work on several animal species little is known relating to human exposure. Whorton and his colleagues in the U.S.A. and I myself as well as our team here in Türkiye have published reports providing evidence that boron does not associate with an adverse effect so far as the marital status and childbearing properties of subjects exposed to borates environmentally and/or occupationally are concerned (8-10). We found the rate of childless participants was 3.17 %, and 3.0 % over three-generations. Independently, it ranged from 2.3 to 4.0 % among male and female sibs of probands and their spouses (11); none being higher than the rate revealed for the general population. Similar results have also been obtained in 'borate families' in 'borate towns' (11). More recently it has been shown that the frequency of infertility concerning borate workers only was not higher either (12).

Since the study started in 1995 has recently finished, we here give all relevant data from all borate regions of the Country, lending support that continuous exposure to the element does not interfere with human reproduction primarily and secondarily.

## Materials and Methods

### Study Areas

Türkiye is a country with the world's largest borate deposits ever discovered. It is also a remarkable producer and exporter of boron products. Two underground ore pits around Sultançayır, Aziziye and Yıldız villages of Susurluk county, Balıkesir, have been the earliest sites of borate mining. Pandermite, hence the name of Bandırma, a sodium borate species, has been exploited in 1861 and exported to Europe from 1867 till 1960. Etibank, recently Eti Holding A.Ş., established in 1935 by Atatürk, has taken borate industry over in 1978 by all means. Much work has been done with respect to geological and technical as well as trading issues of borate minerals in this country. Helvacı together with Alonso, one of the main researchers on the subject, lastly compared Turkish borates with those in Argentina (13). It has been stated that our borate salts are of superior quality.

This study covers all four regions, that is, all areas with large borate deposits, open-field and -once- underground pits, and processing facilities (Table 1).

1. Bigadiç area: From Bigadiç district where borate ore was discovered in 1950 Iskele town and Osmanca and Yeniköy villages were included. There are 4 open-field pits and 3 underground ones first owned and managed by private companies. These latter all was abandoned by 1998. The 3 settlements have already been referred to as Region I. As Prof. B. D. Culver has aptly pointed it out Iskele is "a natural human boron laboratory" together with surrounding villages. An open pit called Acep still lies side-by-side with the walls of some dwellings in Iskele. The town has a population little over 2000.

Separated by the River Bigadiç (Simav River) are Bademli, Değirmenli and Kadıköy villages far way from borate deposits and the facility, even though they too lie within the same geological zone -collectively referred to as Region II as a control of some sort in early days of the study. Many among other villages of Bigadiç and the province Balıkesir have been covered by so-called 'Bigadiç Controls'; this designation

however bears now no much value as before. In several of these villages there are inactive -left out- underground pits.

2. Kestelek area: Kestelek village is situated 22 km southeast Mustafa Kemal Paşa county, Bursa, with a borate area known since 1954. Near this small village there is an open pit and a processing plant.

3. Emet and Hisarcık areas: The two counties of Kütahya are 11 km apart each other with colemanite deposits discovered in 1956. Here Kocaçay (Emet River) runs across the valley with thermal resources aside. Underground mining has been abandoned in Kestelek and Emet-Hisarcık belt in 1990's.

4. Kırka region: The last center of borate industry active since 1968 is Kırka town of Seyidgazi county, Eskişehir. Kırka-Sarıkaya line of borates extending west is the world's largest deposit ever discovered. Likewise there are several inactive underground pits. In all these areas colemanite and ulexite are dominating major borate species -minor ones and other details can be found elsewhere (13).

Triangle formed by Sultançayırı, Aziziye and Yıldız villages have been the first quarters of production as already mentioned, and were included to evaluate late effects of boron exposure. Additionally, Bandırma Borax and Acid Plants built in 1976, were covered. Here borax, boric acid, borax decahydrate, borax pentahydrate, sodium perborate tetrahydrate and sodium perborate monohydrate are processed as refined products. These and concentrated mineral brought out here from other centers are also shipped worldwide. It has been stated that boron content of soils in Türkiye ranges from 0.75 to 4.55 ppm (mg/kg; 14).

### Boron Exposure

Second parameter was boron amount of drinking and utility waters piped out through springs and wells. For convenience study areas were divided into three: a) Those places up to 2 ppm (mgB/l); b) those up to 10 ppmB, and c) those with higher levels. The highest concentration was consistently found in Iskele-

Osmanca belt. It is 6.7-9.7 ppmB in one street fountain, and 18.5-29.0 in the other which both still are in use. There are reports showing that the amount has been measured as high as 60-90 ppmB in a well in the heart of the town – not in use any longer. In recent years fresh water from a remote spring with as little as 1.7 ppmB is pumped out to houses. Boron amounts range from 0.1 to 2.8 ppmB/l in other places, none is due to contamination. Higher levels up to 9.05 ppmB are reported in Emet-Hisarcık belt. In Kırka the concentration is 0.30-2.35 ppmB.

Another parameter was dust amount; although it turned out to be not possible to discriminate boron dusts alone. The management has routine measures in different points across a pit and facility, and these also cover dust on clothes of workers at these places. As mixed dust it never exceeds the permissible level 10 mg/m<sup>3</sup>. Yet one would expect higher concentrations, thus heavy exposure, on some occasions.

### Study Population

Anyone participated to the work was called proband, and the only criterion to be included was the presence of a marriage, no matter one spouse was a widow or divorced. For reasons explained below, study population was treated in two phases. In the first phase 2529 probands, and hence families, were ascertained (Table 1). Of these 2015 (79.6 %) were males and 514 (20.3 %) females, the excess of males being due to the nature of work. Concerning work 1459 (57.7 %) had a boron-related and the remaining 1070 (42.3%) boron-unrelated job. These corresponded to about 65 % of families in Region I, and about 45 % in Region II. There were 448 persons as 'Bigadiç Controls' composed of individuals both from Bigadiç area and far away. From Susurluk 65, from Bandırma 191, from Kestelek 166, from Emet-Hisarcık 732 and from Kırka 378 subjects were covered. Of males with a boron-related occupation 184 (7.2 % of total) and of females (at least) 53 (2.1 % of total) were retirees. Men and women worked only just a few months or years were not encountered as borate workers.



**Table 1:** Study areas and number of probands (proband families)

Province	County	Town/Village	No. probands	Total	
Balıkesir	Bigadiç	Region I*	Iskele	266	359
			Osmanca	56	
			Yeniköy	37	
		Region II*	Bademli	55	190
			Değirmenli	53	
			Kadıköy	82	
Balıkesir	Bigadiç Controls*			448	
Balıkesir	Susurluk			65	
Balıkesir	Bandırma			191	
Bursa	Mustafa Kemal Paşa	Kestelek		166	
Kütahya	Emet & Hisarcık			732	
Eskişehir	Seyidgazi	Kırka		378	
<b>Totals</b>				<b>2529</b>	

\* Not credited in later work and publications.

At the beginning Iskelè, Osmanca and Yeniköy villages have been referred to as Region I, against Region II, formed by Bademli, Değirmenli and Kadıköy villages, relatively far away from the deposits and the facility, as controls of some sort, together with 'Bigadiç Controls'—this division was not maintained any further (see Table 1).

### Methods

A questionnaire-based interview with the proband in field was the method of choice at the first phase. Details were not related only to participant himself (herself) but also to the spouse and offspring. Emphasis was mainly put onto the reproductive story; then to diseases, habits and drugs. No physical examination and laboratory tests were attempted to carry out in field but blood and urine samples were taken for different purposes (not to be given here). Pedigree data was obtained through the proband and analyzed to reveal if there was a clustering of childless families over generations, either vertical or horizontal. For statistical evaluation chi-square test was applied using SPSS for Windows 11.5 software package.

### Findings

#### Demographic Data

Age distribution of male interviewers ranged from 19 to 92 years and that of females from 20 to 79, averaging 47.7 and 46.4 years, respectively. The means for individual areas were 37.5-56.8 for the former and 34.5-52.7 for the latter subgroup, implying that a relatively young population was under discussion.

General character of it was suburban so that approximately 75% of probands were natives of settlements surrounding borate centers. Of the remaining about 20% were from a nearby county and only 5 % from bigger places, namely, cities. The same was also applied for spouses; for nearly 70% of whom were born in the same place as the proband, and 20 % in a close quarter. About 10 % of participants were from a remote place. Needless to say, all were living all around at the time of inquiry, mostly in their home towns and villages with transportation offered by the management for those who work at a borate pit/facility. Couples were relatives between 17.1-22.7% percent of the cases, excepting such urban places as Bandırma and Susurluk. Educational state was likewise rural. While many elderly has no formal education at all, a considerable number of miners had preliminary school

training, again excepting metropolitan subpopulations. Some 20 % were high-school and about 5 % university graduates. These latter were engineers and administrative people in general.

Any marriage without offspring until about the end of the second year of wedding was considered primary type of infertility, provided couples applied no birth control devices. The two-year allowance was because of military duty of males, staying away from home. And any family childless at the time of inquiry despite previous conceptions and/or births was accepted secondary infertile.

Table 2 shows figures related to the marital state of probands from all 12 borate centers. In 2261 out of 2349 fertiles the marriage was continuing whereas 77 persons were widows, and 11 separated; the rates being 89.4, 3.0 and 0.4%, respectively. From the remaining 180 childless families, 81 represented recent marriages, 79 primary - and 20 secondary-infertility with rates 3.2, 3.1 and 0.8%, respectively. Couples without children changed

Table 2: Fertile and infertile probands

Marital state	n	%
With children		
Marriage continuing	2261	89.4
Widowed	77	3.0
Separated	11	0.4
Without children		
Recent marriage	81	3.2
Primary	79	3.1
Secondary	20	0.8
<b>Total</b>	<b>2529</b>	<b>100.0</b>

from 0 % in Osmanca to 6.5 % in Kestelek, differences between communities being insignificant ( $\chi^2=15.65$ ,  $p=0.15$ ). It seemed of note that very few subjects indeed were divorced.

In order to get a closer insight for fertility-infertility state probands were first treated with their birth place and then with occupation. Although open to critics, interviewers were divided into 3 subcategories with respect to their birth place. It was cumbersome however, for no one could separate properly boron-rich soils from -poor ones. Yet they were classified for practical purposes as boron-rich, boron-poor and -necessarily- as intermediate soils (Table 3).

It was found that 969 subjects born in a high-boron community had at least one living child at the time of investigation versus 32 who had no offspring at all, the rates being 96.8 and 3.2%, respectively. Similar findings were revealed for those from a boron-poor territory and an intermediary locality as well, proving that to be born on/near borate deposits is by no means a predisposition to infertility ( $\chi^2=0.30$ ,  $p=0.86$ ).

One of the important issues with this work is the fact that individuals from high-boron regions with elevated levels of boron in fresh water has a job at a nearby pit and/or a facility, thus exposing to the element both environmentally and occupationally. The time spent at this industry changed from 2 to 34 years, averaging 12 years for an active and 20.7 for a retired miner. In some cases both spouses have involved. Because of the occurrence of probands describing few months of work, only those with a 2-year job and longer were encountered for. As it can be seen in Table 4 primary infertility stood at 2.8 % relating

Table - 3: Birth place of fertile probands and infertility

Birth place (of both sexes)	Marital state				Total	
	Fertile		Primary infertile			
	n	%	n	%	n	%
Boron-rich territory	969	96.8	32	3.2	1001	41.1
Intermediary	1133	96.4	42	3.6	1175	48.3
Boron-poor territory	250	96.9	8	3.1	258	10.6
<b>Total</b>	<b>2352</b>	<b>96.6</b>	<b>82</b>	<b>3.4</b>	<b>2434</b>	<b>100.0</b>

**Table 4:** *Boron-related occupation and infertility*

Occupation (active&retired)	Marital state				Total	
	Fertile		Primary infertile			
	n	%	n	%	n	%
Boron-related	1356	97.2	39	2.8	1395	57.3
Boron-unrelated	996	95.9	43	4.1	1039	42.7
<b>Total</b>	<b>2352</b>	<b>96.6</b>	<b>82</b>	<b>3.4</b>	<b>2434</b>	<b>100.0</b>

to 1356 miners against 4.1 % covering 996 subjects having a boron-unrelated occupation ( $\chi^2=3.3$ ,  $p=0.07$ ). It then appears clear that to have a boron-related occupation does not associate with an increased frequency of primary childlessness.

To reveal if there is a concentration of infertility, one would expect more individuals would be affected, if an agent is in operation in a given kindred or a region or a professional class of people, pedigree analysis were carried out by means of individual files. In the first generation, involving fathers' and mothers' sibs of both proband and his (her) spouse primary infertiles were found 3.5 % among 3331 couples. In the second, covering sibs and cousins of probands and their spouses, probands themselves included, there were 9105 families of which 292 had no children primarily, making 3.2 %. This was 2.7 % concerning third generation, composed of offspring of probands and of their sibs. Briefly, this unique approach, a 3-generation analysis, revealed that primary infertility among 14,320 families was 3.2 %, quite similar to that of probands themselves, indicating no a common adverse attack to marital and childbearing properties of subjects under consideration is operational for at least 60 years if one puts a 20-years span per generation. This result has also been the case through the stories of miners among Susurluk contacted. Besides it showed that no aggregation of childless families over generations have occurred remarkably. Secondary failures standing at 0.4 % indicate that reproductive performance is also not affected secondarily.

Despite several advantages this approach has an inevitable pitfall so as to lead to duplications, that is, to encounter some individuals repeatedly particularly in a small community with prevailing blood-related weddings. To prevent this, and to rule out any possible critics so far, marriages of brothers and sisters of both proband and his (her) spouse were taken into account only. Primary type of infertility was found 2.4, 2.5, 4.2 and 3.3 %, respectively, and 3.1 % concerning 6464 families in total. The differences being insignificant and findings quite similar to those mentioned above, this last independent study leaves no room for any other alternative.

Familial occurrence of childlessness was of further attack. Boron industry has created in time the so-called 'borate families' in 'borate towns'. There are such kindreds with father, mother, son, brother... being active or former borate workers. If boron exposure interferes with human fertility, one should meet with such kindreds in which two or more members sharing the same milieu would have the condition. Data from 12 borate centers showed that there was another active or retired miner in addition to proband in 171 (52.9 %) cases; two in 68 (21 %) cases; three in 27 (8.3 %) cases; four in 29 (8.9 %) cases, five in 7 (2.1 %) cases; six in 3 (0.3 %) cases; seven in 5 (1.5 %) cases; eight in 3 (0.9 %) cases and nine in 1 (0.3 %) case - about 13 % of total. Fifteen families only out of 323 kindreds were primary infertiles, making an incidence 4.6 % that was not higher than those observed across the work. This figure was 6.9 % with 4 examples relating to 58 kindreds where proband himself was not borate worker. This and the fact that among 2529 probands as well as some 15,000 within-proband

families only 75-80 couples described 1-4 members with infertility that could not be regarded significant.

To end up the presentation two more issues are to be addressed. One is pertaining to the second phase of the study. Since the conclusion drawn here might be criticized by the assumption that anyone reluctant to be questioned before the public or because of another reason or an appointment might not be covered, thus lowering the rate of infertility unduly. Although much care has been paid not to introduce a bias or make a selection, to ensure data be cleared of any suspicion all borate centers were once more visited, and computerized files of both workers and employers asked. Table 5 shows data offered by personal departments. They included marital state and related things; however no information was available on the number of abortions or dead offspring. Similarly no mention was there whether childlessness is primary or secondary.

By May 2003 there were 2375 workers and 534 employers in all facilities, and infertiles were found 1.7 % with only 40 persons among workers, and 4.3 % with 23 subjects among employers (Table 5). The difference between two subpopulations was significant and not unexpected both on technical and socio-cultural grounds ( $\chi^2=14.16$ ,  $p<0.001$ ).

**Table 5:** Fertile and infertile marriages between workers and employers in all facilities

Marital state	With children		Without children		Total
	n	%	n	%	
Worker	2335	98.3	40	1.7	2375
Employer	511	95.7	23	4.3	534
Total	2846	97.8	63	2.2	2909

These figures clearly indicate that childlessness is not a matter for both workers and employers giving services at close buildings; though borate exposure for the latter might not be comparable. As stated before this last approach

provides firm evidence that borate exposure, how heavy and continuous it is, at present levels do not affect reproduction primarily and secondarily.

### Controls

Because of the lack of reliable information concerning infertility prevalence across the country, controls were of our own, and derived from several sources as well as from seemingly boron-poor regions. Almost all have been collected at random throughout.

There were 88 families from Balya, Balıkesir, one of the earliest sites of mining in Türkiye; 81 families from Altınova, Ayvalık, Balıkesir, a sea-and-sun town, and 74 families from Çamlıdere, Ankara. Primary type of infertility was found 2.3, 4.9 and 4.0 %, respectively. Not tested statistically, an overall rate 3.8 % among 243 appeared quite comparable with those reported for borate areas. Pedigree data were also remarkable: 3.9 % childless with respect to 2215 marriages over 3 generations. Lastly we will refer to a general-population survey (15, 16): prevalence of the primary infertiles relating to over 50,000 couples gathered from almost all parts of the country hardly exceeds 4 %.

These findings, i.e., similarity between frequencies among probands, among 3-generation families, among families of probands' sibs, and among control- and general-population families do lend support to the conclusion already reached that boron exposure at the present levels environmentally and/or occupationally does not harm human reproduction primarily and secondarily.

### Discussion

The population presented here displays several features of bearing. First of all, nearly 85 % of participants are natives of settlements on or near borate deposits and processing plants. They have been living thereabouts lifetime without much moving, mainly engaged in traditional agriculture and animal husbandry no doubt for centuries. Secondly, the same holds true for

spouses, since over 90 % of them have come from the same milieu. Briefly, the population is rather homogenous, though suburban in general. Inter-marriages, running about 20 %, excepting urban communities, would add weight to this structure. It is so a convenient sample exhibiting similar features over generations and the study is an observational one, being the first of its kind.

This peculiarity finds echo in living styles, hence marital characteristics and in turn in fertility-infertility states. Theoretically, three different sets of population could be defined regarding borate exposure, either environmental or occupational or both environmental and occupational. However it was not so easy to distinguish each in practice. Primary infertility was around 3 % and the secondary 0.5 %. Figures were consistent throughout the work, and were so between different subgroups of probands, between borate regions as well as between generations, and were comparable with figures 3.2-4.6 % of controls. This low rate in subjects having borate job could not be ascribed to some workers have escaped from questioning. Number of infertiles in subpopulations was not high enough to cause any difference of meaning. Besides, clustering of the condition was not significant over generations nor in so called 'borate families' of borate towns (11), thus ruling out the operation of a common environmental agent, boron in this case, is harmful to human reproduction. Individuals from high-boron territories with elevated level of boron in waters and residing on/near deposits thus exposing to borates for years had nothing to do with generative performance. Though considered primary one would expect late fertility. In some, slightly reducing the observed rate.

As an independent test of some sort to prevent duplications unavoidable across a community with prevailing consanguineous marriages, infertility-fertility states of male and female sibs of probands and their spouses were treated separately. Again the rates ranged from 2.3 to 4.0 % without concentration of infertiles. No etiology was addressed with this study; nevertheless, because of the fact that many miners have

married before or just after the work, a supposition that exposure to borates causes infertility remains unsupported. It is then clear that the underlying basis would be the trends of marriage and childbearing. The low rate of infertility concerning workers of borate facilities only should not be underestimated.

In a conference delivered to Gynecology and Obstetrics Seminar in 1974 Baran declared the rate of women complaining from sterility was 13.4 % depending out-patients admissions (17, see 21). Later on General Director of Health Ministry spoke in 1985 that the prevalence of infertility was at least 15 %, even though a reliable study has not been available (17, see 21). In a recent publication frequency of "infertile women not under pregnancy risk" was given 6.9 % for the year 1993, and 6.6 % for 1998 (18). Although there was not a mention whether the rate represented primary or secondary failures, these last figures are rather close to those given here. In another work it has been found infertility was 2.2 % in a segment of the population (19).

Inhalation route of borates has extensively been worked out in different species, and it has been claimed that testicular atrophy is associated in rats with aerosol exposure of boric acid (6, 20). Despite systemic uptake due to swallowing particles, the most frequent and appreciable general-population exposures are from ingestion of food and of beverages (2-4). Occurrence of only 34 subjects supposedly under heavy and continuous borate pressure among 1423 miners do not support that respiratory intake is associated with human infertility.

Although not given here in detail one of the findings was the presence of families having 5 or more children in those days where birth controlling (or family planning) have not been so effective (21). Today's couples even in villages do get fewer offspring than their parents. Elderly people have apparently enjoyed having as many children as they liked or could do, implying that reproductive performance has not been affected at boron exposure at present levels for centuries.

## REFERENCES

1. Warington K. The effect of boric acid and borax on the broad bean and certain other plants. *Ann Bot* 1923; 37: 629-72.
2. Anonymous. Health Effects of Boron. *Environ Health Perspect* 1994; 102 (Suppl 7).
3. WHO (World Health Organization). International Programme on Chemical Safety (IPCS). Environmental Health Criteria for Boron 1995, Geneva.
4. WHO (World Health Organization). Trace Elements in Human Nutrition and Health 1996, Geneva.
5. ECETOC. Reproductive and General Toxicology of some Inorganic Borates and Risk Assessment for Human Beings. Technical Report No. 63, 1995, Brussels.
6. Tarasenko NY, Kasparov AA, Strongina CM. Effect of boric acid on the generative function in males. *Gigiena Truda i Professionalnye Zabolevaniya* 1972; 11: 13-6.
7. Krasovskil GN, Varshavskays SP, Borisov AL. Toxic and gonadotropic effects of cadmium and boron relative to standards for these substances in drinking water. *Environ Health Perspect* 1976; 13: 69-75.
8. Whorton MD, Haas JL, Trent L, et al. Reproductive effects of sodium borates on male employees. *Occup Environ Med* 1994; 51: 761-767.
9. Şaylı BS, Tüccar E, Elhan AH. An assessment of fertility in boron-exposed Turkish subpopulations. *Reproduc Toxicol* 1998; 12: 297-304.
10. Şaylı BS. An assessment of fertility in boron-exposed Turkish subpopulations: 2. Evidence that boron has no effect on human reproduction. *Biol Trace Element Res* 1988; 66: 409-422.
11. Şaylı BS. Assessment of fertility and infertility in boron-exposed Turkish subpopulations: 3. Evaluation of fertility among sibs and in "borate families". *ibid* 2001; 81: 255-267.
12. Şaylı BS. Low frequency of infertility among workers in a borate processing facility. *ibid* 2003; 93: 19-29.
13. Helvacı C, Alonso RN. Borate deposits of Turkey and Argentina: A summary and geological comparison. *Turkish J Earth Sci* 200; 24: 1-27.
14. Çebi H, Özkan ŞG. Solubor hakkında genel bilgiler ve tarım endüstrisindeki bor uygulamaları. *Etibank Genel Müdürlüğü Bor Araştırma Merkezi*, No. 34, 1996, Menderes, İzmir.
15. Şaylı BS. Anadolu'nun genetik yapısı üzerine çalışmalar: XII. 3. Türkiye'de fertil ve infertil evliliklerin sıklık ve öteki bazı özelliklerine ilişkin yeni bulgular. *GATA Bül* 1986; 28: 1027-1040.
16. Şaylı BS, Bilgin S. Anadolu'nun genetik yapısı üzerine çalışmalar: XVI. 4. Türkiye'de fertil ve infertil evliliklerin tipleri ve peşpeşe iki kuşak arasında karşılaştırılması. *ibid* 1041-1056.
17. Baran S ve Tandoğan T. See the reference 21.
18. Türkiye'de Ana Sağlığı, Aile Planlaması Hizmetleri ve İsteyerek Düşükler. Türkiye Nüfus ve Sağlık Araştırması. Hacettepe Üniversitesi Tıp Fakültesi Halk Sağlığı Anabilim Dalı, Türkiye Aile Sağlığı ve Planlaması Vakfı, United Nations Population Fund. (Türkiye'de Aile Planlaması Yöntem Kullanma Davranışları, Aile Planlaması Hakkındaki Görüşler ve Gelecekte Yöntem Kullanma Konusundaki Eğilimler). 2000, Ankara.
19. Sağlık Bakanlığı – Hacettepe Üniversitesi Populasyon Araştırmaları. 1998, Ankara.
20. Hubbard S. Comparative toxicology of borates. *Biol Trace Element Res* 1998; 66: 343-57.
21. Şaylı BS. Bor Bileşikleriyle Temasın İnsan Sağlığına ve Çevre Üzerine Etkilerinin Araştırılması. Ankara Üniversitesi Tıp Fakültesi Dekanlığı ile Eti Holding A.Ş. Genel Müdürlüğü arasında imzalanan araştırma projesi gereği hazırlanan nihai rapor. 2003, Ankara.



# CORRECTION OF STEAL SYNDROME OF NATIVE ARTERIOVENOUS FISTULAS IN HEMODIALYSIS PATIENTS BY "BANDING" TECHNIQUE

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Rasih Yazkan\* ❖ Ünal Sakıncı\*

## SUMMARY

**Objective:** Among complications of native arteriovenous fistulas in hemodialysis patients, symptomatic steal syndrome is uncommon, but may lead to ischemia of the hand.

**Materials and Methods:** A retrospective review of the charts of 653 patients who underwent 826 direct upper extremity arteriovenous fistulas, and 6 of whom were subsequently diagnosed as steal syndrome, was performed between January 1998 and December 2002. Six patients with chronic renal insufficiency (4 male, 2 female) with patent upper extremity native arteriovenous fistulas presented with severe hand ischemia. All patients developed severe ischemic manifestations in the form of sensory loss in 2, severe intolerable pain with impalpable pulse in 3, and cyanosis of digits and ulcerations in 1. All patients underwent banding of the native arteriovenous fistulas.

**Results:** All patients had immediate resolution of upper extremity ischemia immediately after banding procedure. Limb salvage and maintenance of a functional fistula were achieved in 100% and 83.3%, respectively, at six-months.

**Conclusions:** We claim that the banding technique reduce fistula diameter and improve distal perfusion. This technique also has the advantage of saving veins which, of course, is worthwhile in patients under haemodialysis.

**Key Words:** Arteriovenous Fistula, Arterial Steal, Hemodialysis, Hand Ischemia, Banding Technique

## ÖZET

### Hemodializ Hastalarında Nativ A-V Fibüllerin Banding ile Düzeltilmesi

**Amaç:** Hemodializ hastalarında nativ arteriyovenöz fistüllerin komplikasyonları arasında semptomatik steal sendromu nadirdir, ancak elin iskemisine neden olabilir.

**Gereç ve Yöntem:** Ocak 1998 ile Aralık 2002 tarihleri arasında, 826 direk üst ekstremité arteriyovenöz fistül operasyonu uygulanan 653 hastanın retrospektif dosya taramasında daha sonra 6 steal sendromu tanısı kondu. Patent üst ekstremité arteriyovenöz fistülü olan kronik renal yetmezlikli 6 hasta (4 erkek, 2 kadın) şiddetli el iskemisi ile başvurdu. Hastaların iskemik bulguları şu şekilde idi; 2 hastada duyu kaybı, 3 hastada nabızsız dayanılmaz ağrı, ve 1 hastada parmaklarda ülserasyon ile birlikte siyanoz vardı. Hastaların hepsinin nativ arteriyovenöz fistüllerine banding uygulandı.

**Sonuçlar:** Tüm hastalarda banding prosedürünü takiben erken dönemde üst ekstremité iskemisi düzeldi. Altı ayda, %100 hastada uzuv kazanımı sağlanırken, %83.3 hastada fonksiyonel fistülün devamı izlendi.

**Yorum:** Banding tekniğinin fistül çapını azalttığını ve distal perfüzyonu iyileştirdiğini iddia ediyoruz. Bu teknik, aynı zamanda hemodializ altındaki hastalarda kıymetli olan venlerin kurtarılmasını da sağlama avantajı sunmaktadır.

**Anahtar Kelimeler:** Arteriyovenöz Fistül, Arteriyal Steal, Hemodializ, El Iskemisi, Banding Tekniği

Native AVF (Arterio-venous fistulas) provide the best possible vascular access for chronic hemodialysis. Compared with the prosthetic bridge graft, the AVF has better long-term patency and fewer complications, including a lower incidence of vascular steal syndrome (1).

Various surgical techniques to correct steal syndrome such as ligation of the AVF, narrowing of the angioaccess, elongation of the bridge or ligation of the artery distal to the AVF plus bypass have been described (2-4). If no hemodynamically significant inflow lesion exists,

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the option for surgical technique include banding procedure to reduce AVF flow. This report presents our experience with banding technique in the six patients with chronic renal insufficiency with patent upper extremity native AVF presented with hand ischemia.

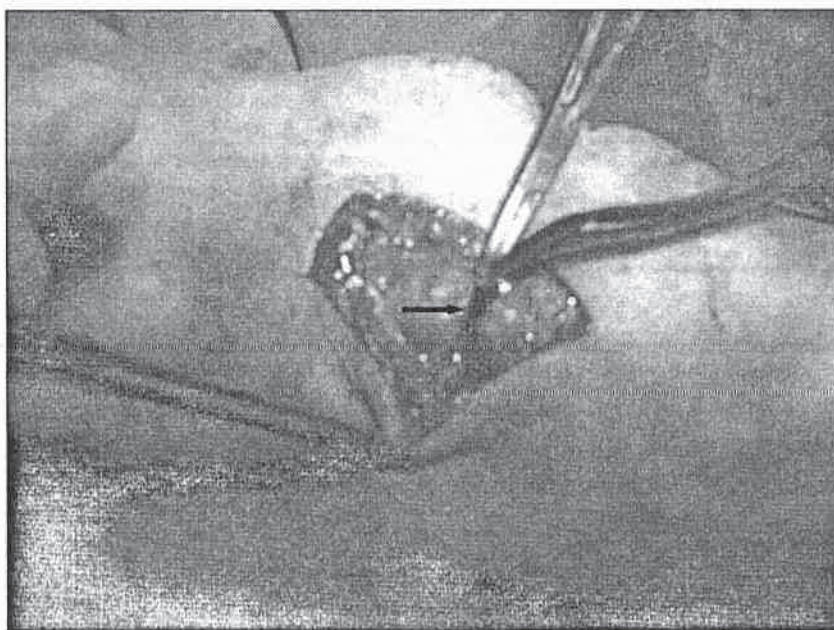
#### Materials and Methods

A retrospective review of the charts of 653 patients who underwent 826 direct upper extremity AVF, and who were subsequently diagnosed as steal syndrome, was performed at the Ankara Numune Education and Research Hospital between January 1998 and December 2002. The direct upper extremity AVF created included the following: snuff-box (n=278, 33.6%), radiocephalic (n=445, 53.8%), brachiocephalic (n=96, 11.6%), and brachio-basilic transposition (n=7, 0.8%). Hand ischemia occurred 6 (4 of brachio-cephalic and 2 of radio-cephalic) of 826 direct upper extremity AVF (0.7%). The median age of these patients (4 male, 2 female) was 52.3 years (range 35-75 years). Two patients developed ischemic manifestations immediately postoperatively, 2 in the first month,

1 after 4 months, and 1 after 1 year. All patients developed severe ischemic manifestations in the form of sensory loss in 2, severe intolerable pain with impalpable pulse in 3, and cyanosis of digits and ulcerations in 1.

Diagnosis was mainly based on clinical features: regaining of distal pulses after AVF compression, confirmed by Duplex ultrasonography (USG) examination. Monophasic flow increased to biphasic and triphasic in three and one patients, respectively.

All six patients were treated with the banding technique under local anesthesia. By a three cm oblique incision, a right-angle clamp is gently placed around the vein, special care being taken not to perforate the arterialized vein. A Teflon tape is grasped in the clamp and passed around the vein. A 2/0 silk ligature is tied beneath the right-angle clamp constricting the Teflon tape thereby bringing the caliber of the AVF down to 1/2 or 1/3 of its size without losing thrill on the AVF and palpation of the distal pulses (Figure 1).



**Figure 1:** Banding technique involves constricting the Teflon tape (black arrow) thereby bringing the caliber of the AVF down to 1/2 or 1/3 of its size.

## Results

All patients had immediate resolution of upper extremity ischemia immediately after banding procedure. Two patients became asymptomatic without restoration of a radial pulse. At 4 months postoperatively, one of the patient's painful ischemic ulcerations recovered almostly. One of them thrombosed three months after banding and its ischemic manifestations were completely resolved. Thrombectomy and graft extension to a more central vein restored patency. Limb salvage and maintenance of a functional fistula were achieved in 100% and 83.3%, respectively, at six-months.

## Discussion

Hand ischemia is rare complication after construction of an native or prosthetic bridge graft AVF. The incidence of ischemia with prosthetic bridge graft AVF varies between 2.7 and 4.3% and the incidence of ischemia with native AVF is less than 2% (4,5). In our study the incidence of hand ischemia with direct upper extremity AVF is 0.7%.

Mild circulatory insufficiency presenting as hand claudication or pain during dialysis can usually be conservatively managed by observation or by restriction of dialysis flow rates. Nevertheless there are patients developed severe ischemic manifestations necessitating surgical revision (3,4). The classic maneuver to deal with the steal syndrome is the ligation of the AVF and performance of another procedure to gain dialysis access (6). Alternatively, many of the reported techniques for reducing AVF flow have been hampered by their complexity involve the destruction of functional angioaccess sites, and

involve conversion of native AVFs to prosthetic bridge grafts (2-4). The main disadvantage of artery ligation-bypass procedures is the fact that they are more expensive than banding procedure and difficult to insert.

Another widely used technique is the so called banding. It consists of producing a stenosis in the outflow portion of the AVF, close to the anastomosis. Many variations of banding, all intended to produce a narrowing and consequent flow reduction, have been reported (6-8). The practical problem in banding technique stems from the difficulty in establishing the precise degree of stenosis required for elimination of the steal, while allowing a flow sufficient to sustain patency of the outflow fistula. The level of critical stenosis that results from the banding procedure is very important. Hemodynamic assessment is required during banding technique, but it may also be useful in pre- and intra-operative evaluation of patients undergoing therapeutical AVFs to prevent hand ischemia. Color-duplex USG is noninvasive, painless and reproducible in monitoring a vascular access. In addition, the blood flow volume measured by color-duplex USG correlates well with the blood flow of a vascular access (9). In our own experience, the amount of narrowing is determined easily in native AVFs with palpation the distal pulses and the thrill on the fistula and confirmed by Duplex USG measurement.

In conclusion, we claim that the banding technique reduce fistula diameter and improve distal perfusion. This technique also has the advantage of saving veins which, of course, is worthwhile in patients under haemodialysis.

## REFERENCES

1. Beathard GA, Settle SM, Shields MW. Salvage of the nonfunctioning arteriovenous fistula. *Am J Kidney Dis* 1999; 33: 910-6.
2. Rivers SP, Scher LA, Veith FJ. Correction of steal syndrome secondary to hemodialysis access fistulas: a simplified quantitative technique. *Surgery* 1992; 112: 593-7.
3. Schanzer H, Schwartz M, Harrington E, Haimov M. Treatment of ischemia due to "steal" by arteriovenous fistula with distal artery ligation and revascularization. *J Vasc Surg* 1988; 7: 770-3.
4. Haimov M, Schanzer H, Shaladani M. Pathogenesis and management of upper-extremity ischemia following angioaccess surgery. *Blood Purif* 1996; 14: 350-4.
5. Morsy AH, Kulbaski M, Chen C, Isiklar H, Lumsden AB. Incidence and characteristics of patients with hand ischemia after a hemodialysis access procedure. *J Surg Res* 1998; 74: 8-10.
6. Papalois VE, Haritopoulos KN, Labruzzo C, Farrington K, Hakim NS. Reversal of steal syndrome following creation of arteriovenous fistula by banding with a Gore-Tex cuff: a new technique. *Int surg* 2001; 86: 210-2.
7. West JC, Bertsch DJ, Peterson SL, Gannon MP, Norkus G, Latsha RP, Kelley SE. Arterial insufficiency in hemodialysis access procedures: correction by "banding" technique. *Transplant Proc* 1991; 23: 1838-40.
8. Khalil IM, Livingstone DH. The management of steal syndrome occurring after access for dialysis. *J Vasc Surg* 1988; 7: 572-3.
9. May RE, Himmelfarb J, Yenicesu M, Knights S, Ikizler TA, Schulman G, Hernanz- Schulman M, Shyr Y, Hakim RM. Predictive measures of vascular access thrombosis: A prospective study. *Kidney Int* 1997; 52: 1656-62.

## RESULTS OF TIPU ON PATIENTS WITH PRIMARY AND RECURRENT HYPOSPADIAS

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### SUMMARY

**Aim:** Currently, TIPU is one of the most commonly used correction methods both primary and recurrent hypospadias. In this study we aimed to compare success rate of TIPU on primary and recurrent cases.

**Methods:** In this study, totally 132 patients who had primary hypospadias with minimal or no chordee (n=83) and recurrent hypospadias with a normal urethral plate (n=49) were repaired with Tubularized Incised Plate Urethroplasty (TIPU). Patients were followed up for 34 (6-48) months postoperatively.

**Results:** Among patients with recurrent hypospadias 3 meatal stricture and 4 fistula were observed. Meatal stricture (n=4) and fistula (n=7) were seen who had primary hypospadias. None had urethral stricture. The success rate was 86,75% in cases with primary hypospadias and 85,72% in cases with recurrent hypospadias.

**Conclusions:** We concluded that the results of TIPU in both primary and recurrent cases were satisfactory with low complication rate and can be used successfully in circumcised cases.

**Key Words:** Hypospadias, Snodgrass, Tubularized Incised Plate Urethroplasty.

### ÖZET

#### Primer ve Rekürren Hipospadias Hastalıklarında TIPU Sonuçlarımız

**Amaç:** Günümüzde TIPU, hipospadyas olgularında en çok uygulanan yöntemlerden birisidir. Bu çalışmada primer ve rekürren vakalarda TIPU'nun başarı oranlarını karşılaştırmayı amaçladık.

**Yöntem:** Minimal kordili veya kordisiz ve 10'u sünnetli olan 83'ü primer ve normal üretral plate'li 49'u rekürren hipospadyaslı toplam 132 hasta TIPU yöntemi ile onarıldı. Hastalar, 2000-2003 tarihleri arasında ortalama 34 (6-48) ay süreyle takip edildiler.

**Bulgular:** Rekürren hipospadyaslı hastalar arasında 3 hastada meatal darlık, 4 hastada fistül gelişti. Primer hipospadyaslı hastalarda ise 4 hastada meatal darlık, 7 hastada fistül görüldü. Hiçbir vakada üretral darlık gelişmedi. Başarı oranları primer hipospadyaslı vakalarda %86.75, rekürren hipospadyaslı vakalarda %85.72 olarak tesbit edildi.

**Sonuç:** TIPU nun, hem primer hemde rekürren vakalarda güvenilir, komplikasyonu az ve sünnetli vakalarda da başarıyla uygulanabilen bir yöntem olduğu sonucuna varıldı.

**Anahtar Kelimeler:** Hipospadias, Snodgrass, TIPU.

It is known that more than 100 types of operation techniques have been described for the surgical treatment of primary hypospadias. The objective to be attained in all these methods is to ensure the formation of a functionally normal urethra and a cosmetically acceptable penis. Functional integrity was used to be important in the first years of hypospadias surgery which was then popular (1). Multi-staged repair methods have been described particularly for

cases with proximal hypospadias and cases with severe degree of chordee. Today there are many one-staged methods which are widely used and popularized according to the existence of chordee and the location of meatus. Among these methods, MAGPI (2) is popularized especially for cases with glandular location, perimeatal flap or onlay island flap method (3,4) for cases without chordee or with minimal chordee, and preputial tube flaps (5) for the ones with chordee (6,7). In

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recent years the search for alternative methods for the cases without chordee or with minimal chordee because of half-moon shape of newly formed meatus instead of a vertical line form with the present techniques has led to the discovery of tubularized incised plate technique. Snodgrass (8) who described the method with its most recent and developed condition emphasized the applicability of this method in all cases except for those with severe degree of chordee and unhealthy urethral plate. Also in reality, almost all of the methods: MAGPI, flip-flap or preputial flaps are suitable for special indications. However, the surgeon might find it difficult which method to choose due to varying degrees of hypospadias development in every patient. For instance, although perimeatal flap technique or onlay island flap technique is appropriate in chordee-free distal hypospadias cases with well-developed urethral plate, the presence of meatal stricture in the same condition might lead the surgeon to review the decision again. Another problem faced in hypospadias surgery especially in countries where circumcision has a traditional application is the high rate of complication and the limited number of methods for patients who need to be re-operated due to a previous unsuccessful operation.

Taking this into consideration, we would like to display our results with tubularized incised plate technique.

### Materials and Methods

Tubularized incised plate method was applied in our clinic in patients with minimal or no chordee who were primarily operated and previously operated with various techniques which were failed to be successful. Attention was paid particularly to the absence of severe degree of chordee and to the presence of a healthy urethral plate for incision in all cases. The repair was primary in 83 cases 10 of which were circumcised and recurrent in 49 cases. The site of the hypospadiac meatus was outlined in table 1. The ages of 132 cases varied between 3 and 17 years ( average 6.4 years), while the mean follow up was 44 months (3-84 months). Previous operations of patients are outlined in table 2.

**Table 1:** *The site of hypospadiatic meatus*

Site of meatus	primary	recurrent	circumcised
Glandular	18	10	-
Coronal	12	10	5
Subcoronal	25	10	3
Distal	8	10	2
Mid penil	10	9	-

**Table 2:** *The previous operation types of cases who underwent tubularized incised plate technique*

Type of the cases	Number
Recurrent Patients	49
<i>Mathieu</i>	30
<i>Magpi</i>	10
<i>Onlay Island Flap</i>	9
Primary Patients	83
<i>Circumcision</i>	10
<b>TOTAL</b>	<b>132</b>

Operations started with the application of general anesthesia subsequent to the fulfillment of routine pre-operative procedures. First of all suspension suture was applied to the glans penis and rubber clamp was placed on the penis root. The urethral plate was incised along the midline between the glans and the meatus to the corpus cavernosum. Depending on the size of the meatus, 8 or 10 F of nelatone catheter was put into the incised sulcus as to go a few centimeters into the urethra. A U-form incision was made starting from the glans 0.5 cm down to the meatus as to easily cover the stents in both lateral sides. Making a transverse circumcision starting from 1 cm below the glans, the skin was freed downwards and an artificial erection was produced. If there was chordee and if it still persisted despite correction of adhesions then dorsal plication was applied. Glans wings were freed and the use of cauther was avoided as much as possible during these procedures.

Afterwards the flap prepared above the stent was closed starting from the base of the meatus until the glans by means of continuous 5/0 or 6/0 polyglactin sutures. After the location and the width of the neomeatus was sufficiently adjusted, a subcutaneous pedicled flap was brought onto the suture line. Subsequent to the closure of the glans wings, the skin was sutured properly. Urinary diversion was applied to all patients by employing a cystofix and the urethral stent was fixed by means of suspension suture in the glans. The procedure ended following the application of compressed dressing.

The dressing procedure was repeated daily starting from the third post-operative day. The stents of the patients were removed on the 5th to 7th days (average 6.3 days). The cystofixes were clamped pursuant to the healing of the wound and they were removed on the 14th day after observing the urination of the patient. The patients were discharged after they were called for control in the first and third post-operative months.

### Results

The success rate with this method was 86,75% in primary group and 85,72% in recurrent group. Success rate in both group was not different statically ( $p > 0,5$ ). All of patients were observed excellent cosmetic results. Reobserving the urination of the patients in the first post-operative month, urinary flow rate and the postoperative complications were assessed.

There were meatal stricture in 7 patients who underwent periodic meatal dilatation and resulted in urination in normal calibration. Fistula developed in the former meatal location in 11 patients who were planned to be re-operated for the fistula 6 months later making sure that no meatal or urethral stricture were present by means of calibration by bougies with appropriate diameter. Complications in both primary and recurrent group were not different statically ( $p > 0,5$ ). There was failure of wound healing in 3 patients in whom neither urethral nor meatal problems were cured by appropriate treatment (Table 3).

**Table 3: Complications of patients after TIPU**

Complications	Number
<i>Recurrent Patients</i>	
Meatal stricture	3 (6.1%)
Fistulae	4(8.1%)
Wound opening	2 (4.08%)
<i>Primary Patients</i>	
Meatal stricture	4 (4.8%)
Fistulae	7 (8.4%)
Wound opening	1 (1.2%)

### Discussion

The main objective to be attained in all hypospadias repair procedures is to form a functional neourethra and an esthetically near normal penis. It is also of importance to attain these two major aims with minimal anesthesia, low rate of complications and a limited number of operations. Among all hypospadias repair methods so far, MAPGI is the most successful one with the lowest complication rate (2). However, MAPGI method can be used in selected cases. Hypospadias cases with scrotal or penoscrotal location and with severe degree of chordee necessitate application of specific methods (9). In the same way there are a lot of methods (6,7,9) defined for the treatment of hypospadias cases without chordee or with minimal chordee in which meatus is present in midpenil or distal location. However, it is also known that some authors have concerns about these methods (7,8). The idea that the tubularized incised plate method could be a convenient technique for the hypospadias cases defined lastly has been favored by a gradually increasing number of supporters in recent years. The most frequent question asked when this method has become popular is whether the dorsal incision bed leads to any stricture or not. However, Snodgrass suggested that no stricture developed in any of the 61 patients who he observed for a period of 15 months subsequent to the assessments of the calibration during urination as well as the urethroscopic examinations under anesthesia

(10). The appearance of neomeatus obtained in this way in the form of a vertical sulcus and quite similar to the anatomic configuration has raised much interest in the method. Some surgeons emphasize that they have been applying this method in the majority of hypospadias cases they carry out and that TIP urethroplasty is going to be the most favorable alternative to the flip-flap procedures in the future (11).

When the previous studies about this method are examined, it is seen that the rate of complications is considerably low. When the results of 9 researches with 328 distal and proximal hypospadias cases were assessed, it was seen that 18 cases developed complication (5.5%): 5 of which had meatal stricture (1.5%), 8 fistula development (2%), 4 failure of wound healing (1%) and 1 diverticula formation (0.3%). However, no urethral stricture was seen in any of the cases (10). When these rates are compared to those of other methods, they are found very reliable. The complication rate was reported as 1.2% in Duckett's 1000 cases MAPGI series (12).

Hastie also reported meatal regression in the long-term follow-up of 26 of 28 cases with MAPGI procedure (13). There are other studies stating complication rates of 3-17% with flip-flap technique (6,14) and 5 - 27 % with onlay island flap technique (4,15, 16). It is obvious that TIP technique is rather reliable in terms of complications and success rate among the studies within the same indication group.

It is stated that this method can also be applied in proximal hypospadias cases and it can

be preferred as a re-operation procedure in some selected cases (17,18,19). In our clinic, we applied TIPU to correct both primary and recurrent hypospadias and hypospadias in patients circumcised with an inappropriate method and thus having preputial tissue loss. We especially paid attention to the absence of chordee or presence of minimal chordee and a healthy urethral plate in all patients which were circumcised or operated before. This way it is seen that our results in these 132 cases considering the rate of complication and success are comparable to that of previously published studies with primary cases. In our patients, fistula formation was seen in 11, meatal stricture in 7, and failure of wound healing in 3 patients. There was no significant difference in complication rate and outcome between these groups. However, among these complications only the ones with fistula formation necessitated a second operation while the other ones were cured with appropriate treatments.

TIP urethroplasty may be indicated in the repair of distal and many proximal primary hypospadias cases without chordee or with a slight degree of chordee as well as being a good choice for secondary hypospadias cases with healthy urethral plate and for the ones with preputial tissue loss due to previous circumcision. This method seems to become more popular in the following years since it enables the formation of a functional urethra as well as a cosmetically normal meatus.

## REFERENCES

1. Beck C: Hypospadias and its treatment. *Surg. Gynecol.Obstet* 1917; 24: 511-532
2. Duckett JW: MAPGI (meatal advancement and glanuloplasty): A procedure for subcoronal hypospadias. *Urol Clin North Am* 1981; 8: 513-520
3. Mathieu P: Traitement en un temps de l' hypospade balanique et juxta-balanique. *J Chir* 1932; 39: 481-484
4. Elder JS, Duckett JW and Snyder HM: Onlay island flap in the repair of mid and distal penile hypospadias without chordee. *J Urol* 1987; 138: 376-379
5. Duckett JW: Transverse preputial island flap tecnic for repair of severe hypospadias. *Urol Clin North Am* 1980; 7: 423-431
6. Rickwood AM, Anderson PA: One stage hypospadias repair. Experience of 367 cases. *Br J Urol* 1991; 67: 424
7. Ghali AMA, El Malik EMA, Al Malki J, Ibrahim AH: One stage hypospadias repair. *European Urol* 1999; 36: 436-442
8. Snodgrass W: Tubularized incised plate urethroplasty for distal hypospadias. *J Urol* 1994; 151: 464-465
9. Mouriquand PD, Persad R, Sharm S: Hypospadias repair: Current principles and procedures. *Br J Urol* 1995; 75 (suppl 3): 9-22
10. Snodgrass W: Tubularized incised plate hypospadias repair. Indications, technique and complications. *Urology* 1999; 54: 6-11
11. Steckler RE and Zoontz MR: Stent free Thiersch - Duplay hypospadias repair with the Snodgrass modification. *J Urol* 1997; 158: 1178-1180
12. Duckett JW and Snyder HM: Meatal advancement and glanuloplasty hypospadias repair after 1000 cases. Avoidence of meatal stenosis and regression. *J Urol* 1992;147: 665-669
13. Hastie KJ, Deshpande SS and Moisey CU: Long term follow up of the MAPGI operation for distal hypospadias. *Br J Urol* 1989; 63: 320-322
14. Mustarde JC: One-stage correction of distal hypospadias and other people's fistulae. *Br J Plast Surg* 1965; 18: 413-420
15. Wiener JS, Sutherland RW, Roth DR, Gonzales ET: Comparison of onlay and tubularized island flaps of inner preputial skin for the repair of proximal hypospadias. *J Urol* 1997; 158: 1172-1174
16. Mollard P, Castagnolo C: Hypospadias: the release of chordee without dividing the urethral plate and onlay island flap (92 cases). *J Urol.* 152: 1238-1240, 1994
17. Snodgrass W, Koyle M, Manzoni G et al: Tubularized incised plate hypospadias repair for proximal hypospadias. *J Urol* 1998; 159: 2129-2131
18. Ross JH and Koy R: Use of a de-epithelialized local skin flap in hypospadias repairs accomplished by tubularization of the incised urethral plate. *Urology* 1997; 50: 110-112
19. Retik AB and Borer JG: Primary and reoperative hypospadias repair with the Snodgrass technique. *World J Urol* 1998; 16: 186-191





# LONG-TERM RESULTS OF ARTIFICIAL URINARY SPHINCTER IMPLANTATION FOR THE TREATMENT OF URINARY INCONTINENCE

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## SUMMARY

**Aim:** In this article, we aimed to report long-term results of the cases, which are treated with Artificial Urinary Sphincter.

**Introduction:** Many successful results have been obtained, following the utilization of AMS 800 Artificial Urinary Sphincter (AUS) implantation for the treatment of urinary incontinence due to pure sphincteric insufficiency.

**Materials and Methods:** We implanted AMS 800 (American Medical Systems, Minnetonka, MN, USA) to 20 male patients with pure sphincteric insufficiency. The device was placed to the bladder neck in one case and bulbar urethra in the others. The age range of our patients was between 15-74 (mean age 59) and average follow-up was 46 months (8-132 months).

**Results:** In 16 patients, incontinence was totally treated. Two patients required one pad daily or less, and two patients required more than one pad daily. Two patients were reoperated due to infection, two patients for erosion, another one for mechanical problems. Total success rate was 80%, reoperation rate was found to be 25%.

**Conclusion:** Implantation of AMS 800 Artificial Urinary Sphincter is the most effective treatment modality for the treatment of urinary incontinence due to pure sphincteric insufficiency. The success rate of the technique will increase with appropriate patient selection and surgical experience and the complication rate will decrease.

**Key Words:** Artificial Urinary Sphincter, Incontinence.

## ÖZET

**Üriner İnkontinansın Tedavisinde Artifiyel Üriner Sfinkter İmplantasyonun Uzun Dönem Sonuçları**

**Amaç:** Bu makalede Artifiyel Üriner Sfinkter implantasyonu ile tedavi edilen vakaların uzun dönem sonuçlarını bildirmeyi amaçladık.

**Giriş:** Pür sfinkter yetmezliğine bađlı idrar inkontinansı tedavisi için kullanılan AMS 800 Artifiyel üriner sfinkter (AUS) implantasyonu ile pek çok başarılı sonuçlar elde edilmiştir.

**Materyal ve metod:** Kliniğimizde 20 erkek hastaya AMS 800 AUS implantasyonu uygulandı. Alet, bir vakada mesane boynuna, diđerlerinde bulber üretraya yerleřtirildi. Hastaların yař ortalaması 59 (15-74 yař), ortalama takip süresi 46 aydı (8-132).

**Bulgular:** 16 hastada inkontinans tamamen tedavi edildi. 2 hastada günde 1 veya daha az ped ihtiyacı, 2 hastada ise günde 1'den fazla ped ihtiyacı vardı. 2 hasta enfeksiyon, 2 hasta erozyon, 1 hasta da mekanik problemler nedeniyle reopere edildiler. Total başarı %80, reoperasyon oranı %25 olarak bulundu.

**Sonuç:** AUS implantasyonu pür sfinkterik yetersizliğe bađlı idrar inkontinansı tedavisinde en etkili tedavi yöntemidir. Uygun hasta seçimi ve cerrahi deneyimin artması ile tekniğin başarı oranı artacak ve komplikasyon oranı azalacaktır.

**Anahtar Kelimeler:** Artifiyel Üriner Sfinkter, İnkontinans

Incontinence generally develops secondary trauma and surgery and due to neurogenic reasons although less frequently experienced (1). The risk of developing incontinence is between 0.4-1 % following transürethral resection (TUR)

or open prostatectomy. The risk is reported to be 2.5-12.5% following radical prostatectomy (2,3).

For reestablishing continence, the most commonly used procedures are the application of periurethral injectable materials and implantation

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of artificial sphincters. It is well known that AUS treatment can be very successful for correct indications.

In this article we report the long term results of 20 patients, which were treated with AUS between 1993-2003 in our department.

### Patients and Methods

Between 1993-2003, AMS 800, Artificial Urinary Sphincter was implanted to 20 incontinence cases with sphincteric insufficiency.

In the preoperative period, the patients were evaluated with routine blood tests, urinalysis, intravenous urography, retrograde urography, and cystoscopy and urodynamic tests. The criteria utilized for AUS implantation were the presence of sufficient bladder function, the absence of detrusor instability and the absence of previous surgical treatment for incontinence and urethral stricture.

Three patients had sphincteric urethral strictures; the stricture was treated with permanent urethral wall stent implantation in two cases and with internal urethrotomy in the other. The device was applied to the bladder neck in one patient with vesical extrophy and to bulbar urethra in all the others.

The patients did not have any intraurethral interventions during one week prior to surgery. Prophylactic antibiotics were initiated 24 hours before the surgery and were continued for one week. All the patients were catheterized during the first 24 hours; scrotal ice application was used for preventing edema in the early postoperative period. Pad utilization was recommended during the first six weeks following the operation until the activation of the device.

Following the activation of the implant, the patients were called for follow-up at one-month, three-months, six-months and one-year. During the follow-up; urinary continence and pad requirement were questioned in the patients. The number of pads that the patients needed to change at the end of the daily activities were calculated.

### Results

The duration of follow-up was between 8-132 months (average 46 months). Average patient age was 59 (15-74) years. The etiology of the 20 cases treated with AUS is outlined in Table-1. In the follow-up period, the patients were questioned for incontinence and pad requirements. 16 patients out of 20 reported total dryness (80%) with only few drops of urinary incontinence due to stress and added that they were not disturbed by this event. Two patients told

that they needed less than one pad per day (10%) and two patients needed more than one pad per day (10%). Pad use decreased from a mean of 3,5 to 0,5 units daily. ( $p < 0,001$ ) (Table 2).

AUS was retrieved from two patient due to infection and these patients were reoperated 6 months later and AUS was reimplanted. In one patient only the cuff was changed because of erosion. In one patient the pump came out from the scrotum due to erosion; after the administration of proper antibiotics, the pump was replaced to the other scrotum and no further complications developed. In one patient, the device could not be activated, we believed that this was due to needle stick during surgery and changed the device. Causes of complication and revision types are outlined in Table 3.

**Table 1:** *The etiologies of incontinence.*

Etiology	Patients
Open Prostatectomy	10 (50%)
TUR Prostatectomy	2 (10%)
Radical Prostatectomy	6 (30%)
Epispadias	1 (5%)
Extrophia vesicale	1(5%)

**Table 2:** *The success rate of AUS implantation*

Continance grades	Patients
Totally dry	16(80%)
Minimally continance	2 (10%)
Incontinence	2(10%)

**Table 3: Cause of complications and revision**

Complications	Number	Revision
Infection	2	New AUS implantation
Mechanic problems	1	New AUS implantation
Cuff erosion	1	Only cuff revision
Erosion of scrotum	1	Proper antibiotics therapy and replaced of pump in the scrotume

Two patients out of three with urethral strictures had undergone wall stent implantation and during the follow-up, the strictures have disappeared and the AUS was placed to the distal part of the stent. Third patient had undergone AUS implantation after internal urethrotomy approach.

There was no relation between the etiologies of incontinence and success of AUS implantation ( $p>0,5$ ).

### Discussion

In incontinent patients with intact bladder and urethral functioning, many different surgical techniques have been developed to protect bladder capacity and compliance while providing continence. In cases of incontinence due to sphincteric insufficiency who are resistant to medical treatment, open surgical procedures described by Tanagho and Smith or Young-Dees Leedbetter are applied. The success rate for such interventions is around 50% and strictures develop at a very high rate at the anastomosis line (4,5).

In 1976, Rosen for the first time developed a prosthesis that was placed to bulbar urethra with a pump in the scrotum; on the other hand, Scott utilized AMS 800 model artificial sphincter in 1983 firstly (6).

In patients with AUS implants, the most important problems are infection around the prosthesis, cuff erosion and mechanical insufficiency of the device. In order to prevent

infection and erosion, providing proper dissection, sufficient hemostasis, appropriate cuff selection and having the device inactive for 6 weeks are very crucial.

After conducting a metaanalysis, Hajivassiliou reported the revision rate as 30.5%; which was due to cuff erosion in 12%, infection in 4.5% and mechanical problems in 14% of the cases. Overall success rate was reported as 88% and continence rate was 73% (7). According to this study 50% of the reoperations were performed in the first 8 months and 90% in the first three years. After a follow-up of 10 years, Venn felt to retrieved the prosthesis in 37% of the cases, 56% of these were females and 23% males (8). The complication rates increased up to 25% and infection rates to 12% in cases who have received previous radiotherapy and who have been operated from the same region (9). The device has to be connected very carefully and special attention should be taken not to perforate it with the needle. Montequé et al reported a mechanical insufficiency rate of 8% in their own series (10). In our series, infection and erosion rate was 20% and mechanical complication rate was 5%. Most of the complications during the first years of our surgical experience, and we observed a decline in the complication rates during the following years. Thus, it is suggested that this expensive procedure should be performed by experienced surgeons.

Device related foreign body reactions are one of the most frequently encountered problems and reasons for revision. An urethral erosion case that resulted in stone formation in the bladder 13 months after the implantation and cases who developed peritonitis due to peritoneal irritation have also been reported in the literature (11,12).

Urethral stricture accompanying incontinence is another problematic issue. For such a problem, priority should be given to the treatment of the stricture. This can either be achieved by internal urethrotomy or the stricture can be eliminated by the placement of a permanent wall stent and an artificial sphincter can be implanted consecutively. In three patients that we treated with the latter approach, both the problems

related to the stricture and to the incontinence have been disappeared. Although one of these cases complained of mild wetting, all patients stated that they were very satisfied with the implants.

Another dimension of the surgery is its difficulties when performed on children. We performed an implantation to the bladder neck in a 15 year old boy. Postoperatif follow up was uneventfull and there was no need to change the device. However, according to the report by Barrett, the reoperation rate is 56% in children and adolescents (13). On the other hand, Kyrger reoperated 13 cases out of 32 pediatric patients that he followed for 15.4 years (14).

The success of the operation is measured by the dryness of the patient and the number of pads that are wetted. Although the satisfaction decreases in the patients who wet more than 1 pad a day, the patients all feel themselves much better after the operation compared to their previous condition. The success rate was found as 80% in our series.

The need to reoperate due to erosion is declining in both the pediatric and the adult age group due to the technological advances in the field of AUS. The success rates are reported to be higher in cases in whom narrow back cuff has been utilized (15,16). Periurethral injections that were developed as an alternative approach in recent years do not provide comparable success rates. We are also aware of the migration risks and allergic features of such materials (17). Yet, we believe that this approach is still a good choice for cases of partial incontinence because of its ease of application, low rate of complications and high chance of repeatability.

As a result of 46 months mean follow-up of 20 patients implanted with artificial urinary sphincter, we suggest that this is an intervention, which increases the quality of life in this patient group with high success rates, especially for the cases with normal bladder function and complete sphincteric insufficiency, AUS should be considered as the first choice.

## REFERENCES

1. Krede KJ, Webster GD: Evaluation and management of incontinence after implantation of AUS. *Urol Clin North Am* 1991; 18: 376
2. Gündian JC, Barrett DM: Mayo Clinic experience with the AMS 800 AUS for incontinence after prostatectomy. *Urology* 1993; 41: 128
3. Foote J, Leach GE: Postprostatectomy incontinence pathophysiology, evaluation and management. *Urol Clin North Am* 1991; 18: 229
4. Tanagho EA, Smith DR: Clinical evaluation of a surgical technique for the correction of complete urinary incontinence. *J Urol* 1972; 107: 402-11
5. Kropp KA, Angwafo F: Urethral lengthening and reimplantation for neurogenic incontinence in children. *J Urol* 1986; 135: 533-6
6. Scott FB: The artificial urinary sphincter. Experience in adults. *Urol Clin North Am* 1989; 16: 105
7. Hajivassiliou CA: a review of the complications and results of implantation of the artificial urinary sphincter. *Eur Urol* 1999; 35: 36-44
8. Venn SN, Greenwell TJ, Mundy AR: The long-term outcome of artificial urinary sphincters. *J Urol* 2000; 164: 702-6
9. Wang Y, Hadley HR: Experience with the AUS in the irradiated patients. *J Urol* 1992; 147: 612
10. Montague DK: The artificial urinary sphincter. *J Urol* 1992; 147: 380
11. Bartoletti R, Gacci M, Travaglini F, Sarti E, Selli C: Intravesical migration of AMS 800 artificial urinary sphincter and stone formation in a patient who underwent radical prostatectomy. *Urol Int* 2000; 64: 167-8
12. de Stefani S, Liguori G, Ciampalini S, Belgrano E: AMS 800 artificial sphincter: an unusual case of circumscribed peritonitis due to prosthetic reservoir infection. *Arch Esp Urol* 1999; 52: 412-5
13. Barrett DM, Parukar BG: The artificial sphincter (AMS 800). Experience in young children and adults. *Urol Clin North Am* 1989; 16: 119-32
14. Kryger JV, Spencer Barthold J, Flening P, Gonzalez R: The outcome of artificial urinary sphincter placement after a mean 15-year follow-up in a paediatric population. *BJU Int* 1999; 83: 1026-31
15. Elliot DS, Barrett DM: Mayo Clinic long-term analysis of the functional durability of the AMS 800 artificial urinary sphincter: a review of 323 cases. *J Urol* 1998; 159: 1206-8
16. Bosch JL, Klijn AJ, Schroder FH, Hop WC: The artificial urinary sphincter in 86 patients with intrinsic sphincter deficiency: satisfactory actuarial adequate function rates. *Eur Urol* 2000; 38: 156-60
17. Kershen RT, Atala A: New advances in injectable therapies for the treatment of incontinence and vesicoureteral reflux. *Urol. Clin. Of North Am* 1995; 26, 81-94



## SUBCLINICAL HYPERTHYROIDISM

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### SUMMARY

Subclinical hyperthyroidism is defined as the combination of a suppressed, usually undetectable serum thyrotropin concentration, and normal serum free triiodothyronine and thyroxine concentrations. Patients with subclinical hyperthyroidism are usually euthyroid, but we now understand that subtle symptoms or signs of thyrotoxicosis; such as malaise, tachycardia, nervousness and anxiety; may be present. The sensitivity of the pituitary gland to respond to minor elevations in serum or tissue T<sub>3</sub> and T<sub>4</sub> levels is the main pathophysiological mechanism of subclinical hyperthyroidism.

Subclinical hyperthyroidism can be classified as endogenous and exogenous regarding its cause. Patients with subclinical hyperthyroidism are at increased risk of bone loss, cardiac and neuropsychiatric abnormalities. The diagnosis of subclinical hyperthyroidism is generally made incidentally. Patients who appear to have subclinical hyperthyroidism without a clear etiology should be monitored with repeated serum free T<sub>3</sub>, T<sub>4</sub> and TSH measurements for 2 to 4 months.

In many patients with subclinical hyperthyroidism who do not have consequences of excess thyroid hormone and in whom TSH concentrations are slightly below the normal range, treatment is not necessary. Patients with symptoms of hyperthyroidism, atrial fibrillation, unexplained weight loss, osteopenia or osteoporosis, multinodular goiters would be appropriate candidates for treatment.

**Key Words:** Subclinical Hyperthyroidism, Serum Thyrotropin Concentration, Free Triiodothyronine, Free Thyroxine.

### ÖZET

#### Subklinik Hipertiroidizm

Subklinik hipertiroidizm; baskılanmış, genellikle ölçülemeyecek kadar düşük serum tirotropin konsantrasyonu ile normal serum serbest triiodotironin ve tetraiodotironin konsantrasyonlarının bir kombinasyonu olarak tanımlanır. Subklinik hipertiroidizm saptanan hastalar çoğunlukla ötiroiddir, ancak halsizlik, taşikardi, sinirlilik gibi tiro-toksikozun hafif semptomlarının bu hastalarda bulunabileceğini yeni yeni anlıyoruz. Hipofiz bezinin serum veya doku T<sub>3</sub> ve T<sub>4</sub> düzeylerindeki çok hafif yükselmelere yanıt vermedeki duyarlılığı subklinik hipertiroidizmin ana fizyopatolojik mekanizmasıdır.

Subklinik hipertiroidizm, nedenine göre endojen ve eksojen olarak sınıflanabilir. Hastalar; kemik kaybı, kardiyak ve nöropsikiyatrik bozukluklar açısından artmış risk altındadırlar. Tanı çoğunlukla tesadüfen konur. Subklinik hipertiroidizm açısından belirgin bir nedeni olmayan hastalar, 2-4 ay süreyle serum serbest T<sub>3</sub>, serbest T<sub>4</sub> ve TSH ölçümleri tekrarlanarak takip edilmelidirler.

Subklinik hipertiroidisi olan, ama tiroid hormonu fazlalığına bağlı komplikasyonları olmayan veya TSH konsantrasyonları normalin hafifçe altında olan çoğu hastada tedavi gerekmez. Hipertiroidizm semptomları, atrial fibrilasyonu, açıklanamayan ağırlık kaybı, osteopeni veya osteoporozu, multinodüler guatrı olan hastalar tedavi için uygun adaylardır.

**Anahtar Kelimeler:** Subklinik Hipertiroidizm, Serum Tirotropin Konsantrasyonu, Serbest Triiodotironin, Serbest Tiroksin.

Subclinical hyperthyroidism is an entity that is being increasingly recognized. This may be both due to the aging of the population and the development of more sensitive thyroid-stimulating hormone (TSH) assays. Subclinical

hyperthyroidism is defined as the combination of a suppressed, usually undetectable serum thyrotropin concentration, and normal serum free triiodothyronine and thyroxine concentrations. The TSH value is measured by an assay with a

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threshold of detection that is 0,3 mU per liter or less (1).

Patients with subclinical hyperthyroidism are usually euthyroid, but we now understand that subtle symptoms or signs of thyrotoxicosis such as malaise, tachycardia, nervousness and anxiety may be present. In elderly, atrial fibrillation may be the initial manifestation.

The sensitivity of the pituitary gland to respond to minor elevations in serum or tissue  $T_3$  and  $T_4$  levels is the main pathophysiological mechanism of subclinical hyperthyroidism .

Abnormal TSH levels may remain for years without clinical symptoms of overt hyperthyroidism. The rate of progression of subclinical hyperthyroidism to overt disease is at least 1 to 3 percent per year (2).

#### **Etiology and Differential Diagnosis:**

A suppression of TSH level may be due to nonthyroidal illness, steroid or dopamine administration, or pituitary dysfunction; so, these conditions must be excluded. According to its cause, subclinical hyperthyroidism can be classified as endogenous and exogenous

(Table 1). The endogenous causes of subclinical hyperthyroidism include multinodular goiter, Graves' disease (early), solitary autonomous adenoma, thyroiditis and other causes of hyperthyroidism (eg., trophoblastic tumors).

The exogenous causes of subclinical hyperthyroidism include treatment with levothyroxine, exogenous iodine exposure such as recent administration of radio contrast material (2,3) (Table 1).

A 24-hour radioactive iodine uptake (RAIU) will generally be elevated in patients with Graves' disease, multinodular goiter, and solitary autonomous nodule; but will be decreased in patients in the hyperthyroid phase of subacute, silent, or postpartum thyroiditis and in patients taking excess exogenous thyroid hormone (2).

In clinical examination, thyroid gland may be enlarged in some patients, but in most patients it is usually normal in size .

#### **Clinical Picture:**

Patients with subclinical hyperthyroidism are at increased risk of bone loss, cardiac and neuropsychiatric abnormalities.

#### **Bone loss:**

The effects of subclinical hyperthyroidism on bone mineral density are not well defined. In two cross-sectional studies, there was significantly lower bone mineral density at the femoral neck and radius than in controls (4). Postmenopausal women with subclinical hyperthyroidism appear to be at increased risk of bone loss than premenopausal women with the same condition. An analysis of 1250 subjects enrolled in 41 studies showed that in postmenopausal women, suppressive thyroid hormone therapy was associated with significant bone loss in the lumbar spine and femoral areas (4).

In subclinical hyperthyroidism, whether the changes in bone mineral density cause an increase in the fracture rate is not known. In a recent report, the risk of hip fracture was increased three-fold among women with thyrotropin concentrations of less than 0.1 mU/L compared with women who have normal thyrotropin concentrations. The same study showed a 4.4-fold increase in the risk of vertebral fracture with thyrotropin concentrations of less than 0.1 mU/L (5). These findings are suggestive of adverse effects of subclinical hyperthyroidism on the hip and spine.

#### **Cardiac abnormalities:**

Depending on the evidence from the Framingham study, the cumulative incidence of **atrial fibrillation** at 10 years among people 60 years or older is 28% for thyrotropin concentrations less than 0.1; with a relative risk of 3.1 as compared with those who had a normal serum thyrotropin concentration (6).

There is evidence that patients with subclinical hyperthyroidism have increased heart rate, premature atrial contractions, increased left ventricular mass index, and decreased left ventricular filling (7). The long-term clinical implications of these cardiac changes are not

**Table 1: Patterns of thyroid function associated with a suppressed serum thyrotropin concentration and a thyroid hormone concentration that may be normal**

CONDITION OR FACTOR	TRIIODOTHYRONINE		THYROXINE	
	FREE	TOTAL	FREE	TOTAL
Endogenous subclinical hyperthyroidism (associated with Graves' disease or nodular goiter)	Upper end of normal range	Upper end of normal range	Upper end of normal range	Upper end of normal range
Exogenous subclinical hyperthyroidism (associated with levothyroxine therapy)	Normal	Normal	Upper end of normal range	Upper end of normal range or elevated
Nonthyroidal illness	Normal, low, or elevated	Normal or low	Normal, low, or elevated	Normal, low, or elevated
Drug therapy				
Dopamine	Normal	Normal	Normal	Normal
Corticosteroids	Normal	Normal	Normal	Normal
Amiodarone	Normal	Normal	Usually elevated but may be at upper end of normal range	Usually elevated but may be at upper end of normal range
Central hypothyroidism	Normal or low	Normal or low	Low end of normal range or low	Low end of normal range or low

known, but they are concerning, and further studies are warranted (8) (Figure 1).

#### Neuropsychiatric abnormalities:

Impairment of the quality of life (assessed with a questionnaire) (7), feeling of fear, hostility, and an inability to concentrate are the suggested results of the studies on neuropsychiatric abnormalities in patients with subclinical hyperthyroidism (2).

The Rotterdam prospective study showed that among 1843 participants with endogenous subclinical hyperthyroidism who were 55 years of age or older, a serum thyrotropin concentration of less than 0.4 mU/L was associated with a three-fold increased risk of dementia and Alzheimer disease particularly if thyroid peroxidase antibodies were present (9).

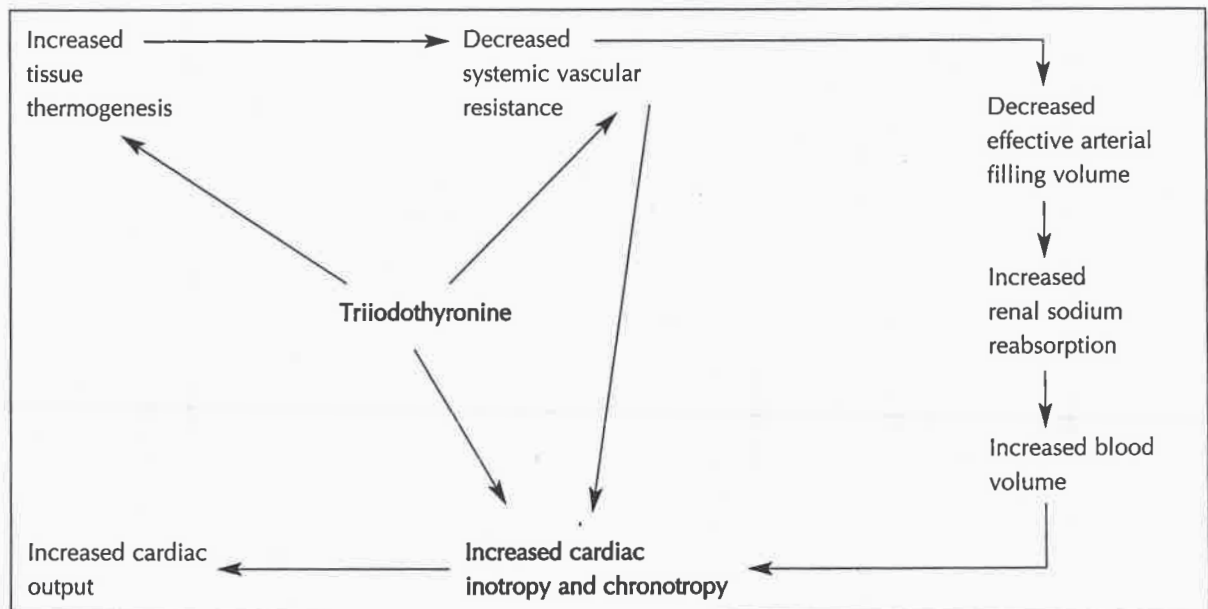
#### Diagnosis:

The diagnosis of subclinical hyperthyroidism is generally made incidentally. Patients who

appear to have subclinical hyperthyroidism without a clear etiology should be monitored with repeat serum free  $T_3$ ,  $T_4$ , and TSH tests for 2 to 4 months (10). A normal or elevated serum thyrotropin concentration at this time suggests recovery from nonthyroidal illness or the hypothyroid phase of thyroiditis.

If the low serum TSH level persists; a 24-hour RAIU, a thyroid scan, and/or a thyroid sonography to assess patients for nodules and heterogeneity are indicated. Detection of thyroid antibodies may be helpful in assessing the presence of autoimmune thyroid disease in selected patients. Thyroid fine-needle aspiration biopsy may be indicated if there are palpable nodules, or nodules identified by sonography. As patients with subclinical hyperthyroidism are at increased risk for cardiac abnormalities and osteoporosis, a bone mineral density of the hip and spine, electrocardiography, and occasionally 24-hour Holter monitor for documentation of arrhythmias may be indicated (2, 4, 6, 8) (Figure 2).

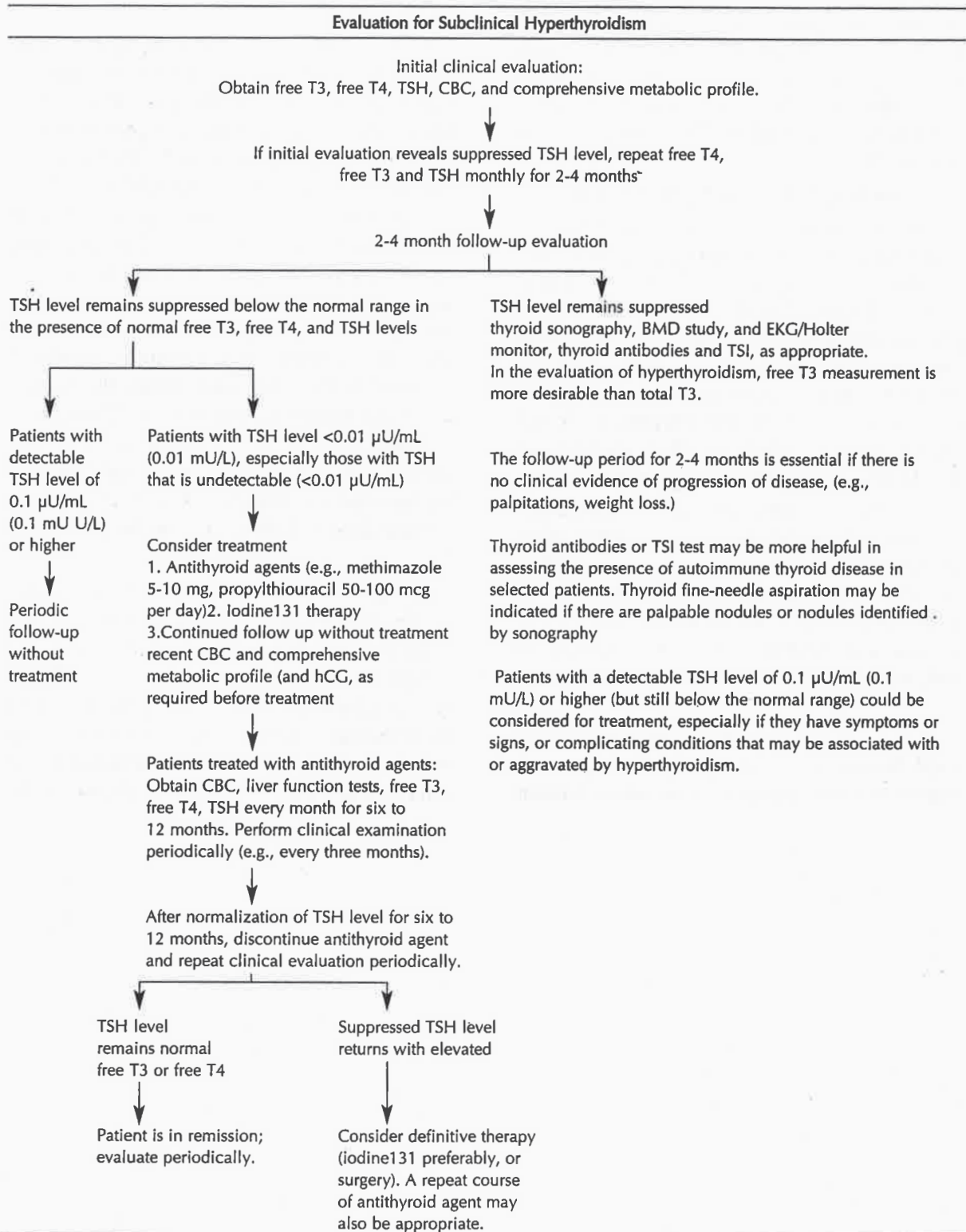
Figure 1: Effects of Thyroid Hormone on Cardiovascular Hemodynamics.



The diagram shows the way in which triiodothyronine increases cardiac output by affecting tissue oxygen consumption (thermogenesis), vascular resistance, blood volume and heart rate.

Adapted from Klein and Lavey.

**Figure 2:** Evaluation and treatment of subclinical hyperthyroidism. (T3 = triiodothyronine; T4 = thyroxine; TSH = thyroidstimulating hormone; CBC = complete blood count; RAIU = radioactive iodine uptake; BMD = bone mineral density; ECG = electrocardiography; TSI = thyroid-stimulating immunoglobulins) Adapted and modified from Am Fam Phys 2002; 65 (3)



**Treatment:****Exogenous Subclinical Hyperthyroidism**

Except the patients with prior thyroid cancer in whom thyrotropin suppression is desired, the thyroxine dose should be reduced in patients with exogenous subclinical hyperthyroidism. Even after the dose reduction, thyrotropin concentrations may remain suppressed for six to eight weeks (1).

**Endogenous Subclinical Hyperthyroidism**

No consensus exists about the management of endogenous subclinical hyperthyroidism. The American Association of Clinical Endocrinologists (AACE) recommends that all patients with subclinical hyperthyroidism should undergo periodic clinical and laboratory assessment to determine individual therapeutic options. If a sustained TSH suppression ( $< 0.1$  microIU/mL) is established, then management should be based on an individual program (10).

In many patients with subclinical hyperthyroidism who do not have complications of excess thyroid hormone and in whom TSH concentrations are slightly below the normal range, treatment is not necessary; but thyroid function tests must be monitored periodically for early recognition of overt hyperthyroidism (Figure 2).

Patients with symptoms of hyperthyroidism, atrial fibrillation, or unexplained weight loss, women with osteopenia or osteoporosis, patients

with multinodular goiter would be appropriate candidates for treatment. The treatment options include antithyroid drugs or radio iodine (10) (Figure 2).

In some patients, clinical response to a six to 12 month trial of low-dose antithyroid agents is very good. Because of the low dosage, side effects such as bone marrow suppression and hepatotoxicity are rarely seen. Methimazole at a daily dose of 5 to 10 mg or propylthiouracil at a daily dose of 50-100 mg can be preferred (2). In women who have child-bearing potential, treatment with propylthiouracil would be more appropriate since methimazole is a category D drug and contraindicated during pregnancy (11). The dose of antithyroid agent can be modulated to maintain the TSH level within the normal range, and discontinued after six to 12 months of therapy. If there is a recurrence after this treatment; a second trial of antithyroid drugs can be initiated or definitive treatment with an ablative dose of iodine-131 can be considered (2).

If a patient with subclinical hyperthyroidism has the potential of developing osteoporosis, and if the periodic follow-up of the patient is not possible, ablative therapy with iodine-131 should be considered again. In patients with multinodular goiter, the incidence of hypothyroidism after iodine-131 therapy is low as compared to patients with Graves' disease (1-3).

## REFERENCES

1. Toft AD. Clinical Practice. Subclinical hyperthyroidism. *N Engl J Med* 2001; 345: 512-16.
2. Diane KS, Kenneth DB. Subclinical Hyperthyroidism: controversies in management. *Am Fam Phys* 2002; 65: 431-38.
3. Vahab F. Adverse effects of subclinical hyperthyroidism. *The Lancet* 2001; 358: 856-57.
4. Uzzan B, Campos J, Cucherat M, Nony P, Boissel JP, Perret GY. Effects on bone mass of long term treatment with thyroid hormones: a meta-analysis. *J Clin Endocrinol metab* 1996; 81: 4278-89.
5. Bauer DC, Ettinger B, Nevitt MC, Stone KL, the Study of Osteoporotic Fractures Research Group. Risk for fracture in women with low levels of thyroid-stimulating hormone. *Ann Intern Med* 2001; 134: 561-68.
6. Sawin CT, Geller A, Wolf PA, et al. Low serum thyrotropin concentrations as a risk factor for atrial fibrillation in older patients. *N Engl J Med* 1994; 331: 1249-52.
7. Biondi B, Palmieri EA, Fazio S, et al. Endogenous subclinical hyperthyroidism affects quality of life and cardiac morphology and function in young and middle-aged patients. *J Clin Endocrinol Metab* 2000; 85: 4701-05.
8. Fadel BM, Ellahham S, Ringel MD, Lindsay J, Wartofsky L, Burman KD. Hyperthyroid heart disease. *Clin Cardiol* 2000; 23: 402-8.
9. Kalmijn S, Mehta KM, Pols HAP, Hofman A, Drexhage HA, Breteler MMB. Subclinical hyperthyroidism and the risk of dementia. The Rotterdam Study. *Clin Endocrinol (Oxf)* 2000; 53: 733-37.
10. AACE Thyroid Task Force. American Association of Clinical Endocrinologists medical guidelines for clinical practice for the evaluation and treatment of hyperthyroidism and hypothyroidism. *Endocr Pract* 2002; 8: 457-69.
11. Haddow JE, Palomaki GE, Alan WC, et al. Maternal thyroid deficiency during pregnancy and subsequent neuropsychological development of the child. *N Engl J Med* 1999; 341: 549-555.



## COMPLICATION OF FELON CAUSED BY MORGANELLA MORGAGNI; CASE REPORT

S. Sinan Bilgin\* ❖ S. Eren Olcay\*\* ❖ A. Mehmet Demirtaş\*\*\*

### SUMMARY

A felon is abscess on the pulp of any digit and it differs from the other abscess in body with local anatomic compartments of pulp. Without appropriate therapy felon can cause widespread necrosis.

*Morganella morgagni* is found in natural flora of gastrointestinal system and is a rare cause of infection without a predisposing factor. It appears as a cause of nosocomial infection or superinfection on a base of immune suppression. Infection caused by MM is generally a slowly progressive ongoing process with remissions and attacks.

Herein the authors report a felon caused by MM. Patient without an underlying disease or an immunosuppressive condition. Delay in seeking further intervention resulted in necrosis of her distal and middle phalanx and extensive surgery indicated.

**Key Words:** Felon, Hand, Infection.

### ÖZET

**Morganella Morgagni Tarafından Meydana Getirilen Felon Komplikasyonu Vaka Sunumu**

Felon parmak pulpasının apsesi olup, pulpanın septalardan oluşmasından dolayı vücuttaki diğer apselerin genel davranışlarından ayrılır. Erken evrede antibiyotik tedavisi yeterli olurken ilerleyen vakalarda cerrahi direnç gerekir.

Gastrointestinal sistem florasının doğal bir üyesi olan *Morganella morgagni*, nadiren tek başına enfeksiyon kaynağı olup genellikle immün baskılanmış kişilerde, hastane enfeksiyonu ya da süperenfeksiyon olarak karşımıza çıkar. *Morganella*'nın sebep olduğu enfeksiyon genellikle sessiz, dönem dönem ataklar ve remisyonlarla seyreder.

Makalede yayınlanan olgu *Morganella* enfeksiyonu için herhangi predispozan etkenin bulunmaması, felon'un zamanında ve yeterli tedavi görmediği için sonucun gereğinden daha geniş cerrahi girişim ile sonuçlanması açısından önem kazanmıştır.

**Anahtar Kelimeler:** Felon, El, Enfeksiyon.

A felon is abscess on the pulp of any digit and it differs from the other abscesses in body. Fingertip pulp is divided into numerous small compartments by vertical septa that stabilize the pad. Infection occurring within these compartments can lead to abscess formation, edema, and rapid development of increased pressure in a closed space. This increased pressure may compromise blood flow and lead to

necrosis of the skin and pulp. Failure of adequate infection treatment can result in serious complications(1,2).

*Morganella Morgagni* (MM) is a gram negative, facultative, anaerobic non-lactose fermenting microorganism. It is found in natural flora of gastrointestinal system and is a rare cause of infection without a predisposing factor (3-5). It appears as a cause of nosocomial infection or

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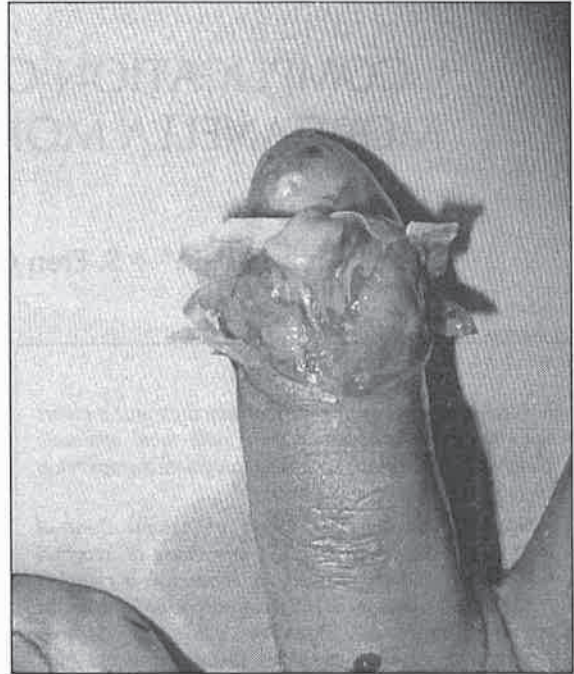
superinfection on a base of immune suppression (3). Infection caused by MM is generally a slowly progressive ongoing process with remissions and attacks.

Herein the authors present a case of complicated felon on distal phalanx caused by MM without a predisposing factor. Inadequate therapy to make way for destructive infection, ends with amputation.

### Case

A 77 year old woman was referred to our clinic because of infection in 2nd and 3rd digits of right hand, during her treatment due to dyspnea secondary to kyphosis in Department of Respiratory Disease. From the history, she had hyperemia and induration in the distal phalanx and nail bed of 2nd and 3rd fingers of right hand, which begun 3 months ago. Her complaints were increased gradually and she was given an oral antibiotic by a clinical practitioner and was offered local wound care. By the 10<sup>th</sup> day of treatment nail of second digit had been fallen and a purulent drainage from the nail bed was begun. By the 10<sup>th</sup> day of drainage since a new nail was begun to grow the patient was stopped oral antibiotics. One month later the infection was recurred with necrotic changes in distal phalanxes. She was offered three phased bone scintigraphy with a diagnosis of osteomyelitis in an orthopaedic clinic, but she was hospitalized because of dyspnea secondary to kyphosis and was consulted to our clinic.

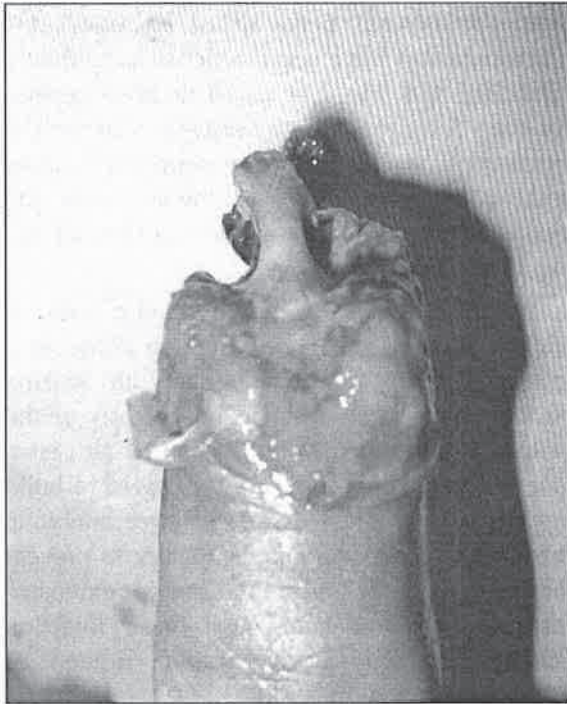
Physical examination of the patient revealed severe infection signs of phalanx 2<sup>nd</sup> digit of right hand. Pulpa and nail bed was sensitive. Digit had edema and indurations, with areas of necrosis and purulent drainage. Soft tissue had crepitation on palpation (Fig.1,2). Infection signs of 3rd finger was milder with hyperemia but without edema and drainage. Hand X rays revealed patchy lytic and destructive lesions of distal phalanx of second digit. One day later 2nd finger of right hand was operated with the diagnosis of complicated felon. During the operation necrotic tissues were debrided, distal and a part of middle phalanx was found to be so destructed that could not be preserved (Fig. 3). Thus the phalanxes was



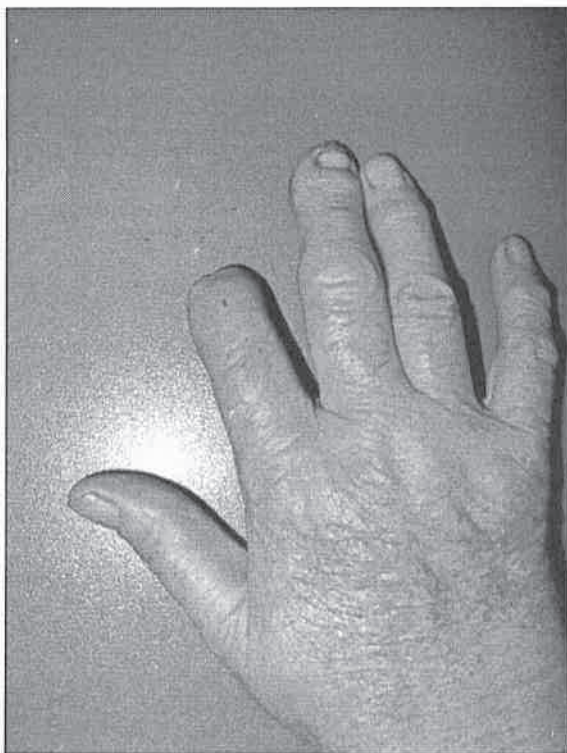
**Figure legend 1:** Figure shows severe infection and patchy necrosis in distal phalanx of second digit. Dorsal view.



**Figure legend 2:** Volar view of digit.



**Figure legend 3:** Destructed bone shown in figure. After necrotic tissues were debrided.



**Figure legend 4:** One month later after surgery no signs of infections were seen, the patient was cured.

amputated from the non destructive bone, permitting free drainage

Microbiological investigations revealed MM infection of 2nd finger and the microorganism was sensitive to quinolones. Patient was given oral antibiotics and non steroidal anti-inflammatory agents for 15 days. 30 days after the operation patient had no signs and symptoms of infection and wound was healed totally (Fig. 4). At the time after one year from the surgery no sign of the recurrence is observed.

### Discussion

A felon is a subcutaneous abscess of the distal pulp of a finger or thumb. It differs from other types of subcutaneous abscess in that multiple vertical fibrous trabeculae or septa divide the pulp into several small compartments (1). The expanding abscess break down the septa and can extend toward the phalanx and produce osteomyelitis, or it can extend toward the skin and cause necrosis (1,2,6).

Staphylococcus Aureus is the most common cause of felon but gram negative organisms have been reported in immunosuppressed patients (7). In early stages, infection may resolve spontaneously particularly with antibiotics. Later, felon requires repeat drainage and debridements as well as intravenous antibiotics over an extended period of complete resolution (8). Most common complication is osteomyelitis involving the diaphysis of distal phalanx and most serious complication is acute tenosynovitis which may result from contagious spread of infection.

MM was first isolated by Morgan in 1906 from cultures taken from children who had diarrhea (3). It is a gram negative, facultative, anaerobic, non-lactose fermenting rod belonging to the family enterobacteriaceae (3-5,9). It is found in the flora of gastrointestinal system. It's rarely the primary invader but produce disease as superinfection in areas previous infected by other organisms.

The urinary tract especially in old catheterized patients is most commonly involved but the bacteria may lead sepsis, pneumonia, wound infections, meningitis and some fatal

infections (3-5). MM may cause culture positive septic arthritis, leading few articular or systemic symptoms or signs of infection and an indolent course of disease with period of remissions and exacerbations of clinical manifestations (3). Infection is generally the disease of the immunosuppressive patients, patients with long term urinary catheterization, diabetes, rheumatoid arthritis, systemic lupus erythematosus, alcoholism, corticosteroid treatment, intravenous medications, surgically intervention and malignancy (3,4,10).

To the best of our knowledge MM infection of soft tissue and bone without a predisposing factor is not reported before.

Herein, the authors report a felon caused by MM, patient without an underlying disease or an immunosuppressive condition. Remissions and exacerbation periods in the history is consistent

with the general behavior of bacteria. This microorganism has a good response to antibiotic treatment and does not cause necrosis general manner. Since the clinical behavior of bacteria is remissions and attacks long term appropriate antibiotic treatment should involve both the acute attack and the remission period till the organism is eradicated

Felon generally require antibiotic with or without drainage, Authors think to come to a conclusion of the therapy; delay in seeking further intervention resulted in necrosis of the patient distal and middle phalanx. If abscesses are not drained and pressure is allowed to build up necrosis is unavoidable. Effective antibiotic treatment and drainage is mandatory to prevent complications of felon and avoid extensive surgery. Certain patients with finger infection should be referred to a hand surgeon or orthopaedic surgeon.

## REFERENCES

1. Robert JN. Acute infections felon. In: Green DP, Notchkiss RN, Pederson WC. Green's operative hand surgery. 4<sup>th</sup> Edition. 1999: 1036-37.
2. Lewis RC Jr. Infections of the hand. Emerg Med Clin North Am. 1985; 3: 263-74
3. Kartz LM, Lewis RC, Borenstein DG. Successful joint arthroplasty following proteus morgani (morganella morgani) septic arthritis: a four year study. Arthritis Rheum. 1987; 30: 583-85.
4. Samonis G, Anatoliotaki M, Apostolakou H, Souglakos J, Georgoulas V. Fatal septicemia and meningitis due to morganella morgagni in a patient with hodgkin's disease. Scand J Infect Dis. 2001; 33: 553-55
5. Carmora F, Fabregues F, Alvarez R, Vila J, Cararach V. A rare case of chorioamnionitis by morganella morgagni complicated by septicemia and adult respiratory distress syndrome. Eur J Obstet Gynecol Reprod Biol. 1992; 45: 67-70.
6. Newfield RS, Vargas I, Huma Z. Eikenella corrodens infections. Case report in two adolescent females with IDDM. Diabetes Care. 1996; 19: 1011-13.
7. Connolly B, Johnstone F, Gerlinger T, Puttler E. Methicillin-resistant Staphylococcus aureus in a finger felon. J Hand Surg (Am) 2000; 25: 173-75.
8. Roberge RJ, Weinstein D, Thimonas MM. Perionychial infections associated with sculptured nail. Am J Emerg Med. 1999; 17: 581-82.
9. Yolken RH. Proteus, providencia, and morganella species. In: Murray PR, Baron EJO, Pfaller MA, Tenover FC, Yolken RH. Manual of clinical microbiology, 7<sup>th</sup> Edition 1999: 491.
10. Bagel J, Grossman ME. Hemorrhagic bullae associated with morganella morgagni septicemia. J Am Acad Dermatol. 1985; 12: 575-76.



# APLASIA CUTIS CONGENITA OF THE SCALP: CONSERVATIVE TREATMENT AFTER FAILED SURGICAL TREATMENT (A CASE AND REVIEW OF LITERATURE)

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Özerk Demiralp\* ❖ Burak Kaya\*

## SUMMARY

*A case of cutis aplasia with a full thickness defect involving scalp and cranium is presented. After a failed surgical procedure healing was obtained conservative local treatment with the use of silvadene cream.*

**Key Words:** *Aplasia Cutis Congenita, Surgical Treatment, Conservative Treatment.*

## ÖZET

### *Aplasia Cutis Congenita*

*Makalede skalp ve kraniumda tam kat defekt mevcut olan bir "Aplasia cutis congenita" vakası takdim edilmektedir. Başarısız cerrahi tedaviyi takiben uygulanan konservatif lokal tedavi ve silvadene krem kullanımıyla iyileşme sağlanmıştır.*

**Anahtar Kelimeler:** *"Aplasia Cutis Congenita", Cerrahi Tedavi, Konservatif Tedavi.*

Cutis aplasia represents congenital absence of all the skin layers, and it may occasionally extend through the bone and dura of the skull (1).

Aplasia cutis congenita is a rare entity. Just over 500 cases have been reported (2). Clinically, the neonate presents one or more sharply circumscribed surface areas where skin has failed to develop. The defect is usually small and solitary, but extensive defects have been reported, resulting in early death. Approximately 80% of these defects occur on the scalp, especially at the vertex, and the remaining 20% over the lower and upper extremities in that order of frequency (3). The lesions may be ulcerated with a red glistening base, eczematous, bullous, or atrophic at birth. They heal spontaneously from the periphery over the course of 1-3 months

to leave a smooth, yellowish, hairless, and papery scar (4).

Cordon in 1767 first described the disease in an extremity and later Campbell in 1826 described it in the scalp. In 1828, Billard reported scalp involvement with an underlying skull defect (1). The etiology of aplasia cutis congenita remains uncertain, but at least one factor in the developmental defect is genetic. Most cases appear sporadically (4). There have been reports of clusters occurring in the same family pedigree, the disease being transmitted in an autosomal dominant or recessive manner or on occasion as an X-linked disease. This indicates an undoubted genetic component to the condition (5). Other proposed etiologies include teratogens, an arrest skin in the skin development in embryonic life,

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an intrauterine vascular abnormality (including placental infarcts and solitary umbilical arteries), intrauterine trauma, early rupture of amniotic membranes or amniotic bands (4). Examination of the placenta and the umbilical cord may aid in the diagnosis (4). It is important to recognise this as a sporadic intrauterine event unrelated to birth trauma or forceps delivery (6). Other associated abnormalities include cleft lip and/or palate, abnormal ears, hydrocephalus, brain malformations, microphthalmus, meningocele, congenital heart disease, tracheoesophageal fistula, omphalocele, anorectal malformation, absent or polycystic kidney, duplication of cervix or uterus, vascular anomalies, nail aplasia or dysplasia, absent, fused or accessory digits, simian creases, epidermolysis bullosa like lesions, Golt's syndrome (focal dermal hypoplasia), trisomy-13, intestinal lymphangiectasia and pyloric atresia. Association with *moya moya* disease has been reported (4,7).

The first attempt to analyze the histopathologic features of the lesion was made by Gross et al in an extensive review of the literature and a detailed study of their own cases in 1957. The membrane covering the brain consists of a very thin, flattened layer of cuboidal, nucleated cells set in a regular manner in a single layer. This histological study of the skin bordering these lesions revealed a gradual transition from normal skin to a corium that was deficient in elastic fibers. In the intermediate zone, the dermal appendages are found to be absent, rudimentary or malformed, decreasing in numbers centripetally more or less proportionately with the attenuation of elastic fibers. Centripetally also, the epidermis thins out to one or two rows of flattened epithelial cells. The collagen is compact, with little adipose tissue present. Sweat and sebaceous glands as well as the elastic fibers are absent (8).

### Case Report

Our patient is a 2900 gram white boy of 38 weeks' gestation. Pregnancy, labour and delivery courses were uneventful. No abnormal family history was noted. At birth, he was found to have big bullae over the scalp. A large round skull

defect at vertex 6x8 cm was detected. The defect including scalp and skull which was represented only by a thin membrane layer exposing underlying brain and sagittal sinus. The thin membranous layer quickly became covered with a dark dry eschar. The infant was otherwise normal, including pediatric neurological examination.

The wound was dressed with aqueous Betadine solution to avoid the possible complication of infection, maceration and subsequent haemorrhage. The eschar became well demarcated and started to become macerated 10 days after birth (Fig. 1). At the 12 days after birth the eschar was removed. We planned to cover the defect by using two temporo-occipital scalp flap after delaying procedure (Fig. 2a-2b). Immediately after delaying procedure a gradual necrosis appeared at the distal edges of the both flaps. We cancelled second stage of the planned operation and decided to go conservatively. After this failed surgical treatment, necrotic areas and defect area were treated with Silvadene and gauze dressing. With this conservative local treatment the wound epithelialized gradually (Fig. 3a-3b-3c). Although there was a great risk of bleeding from the exposed sagittal sinus, it did not occur. And also we did not notice any sign or symptom indicating a possible meningitis or other local or systemic infections. By 45 days the wound had completely epithelialized and contracted (Fig. 4).

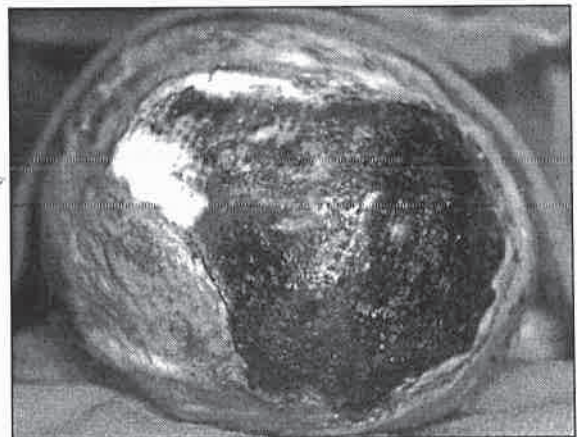
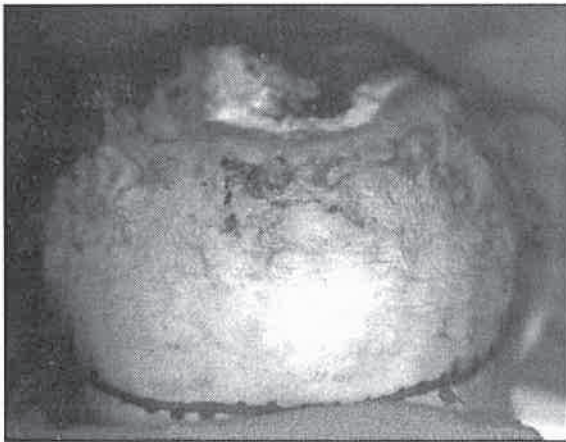
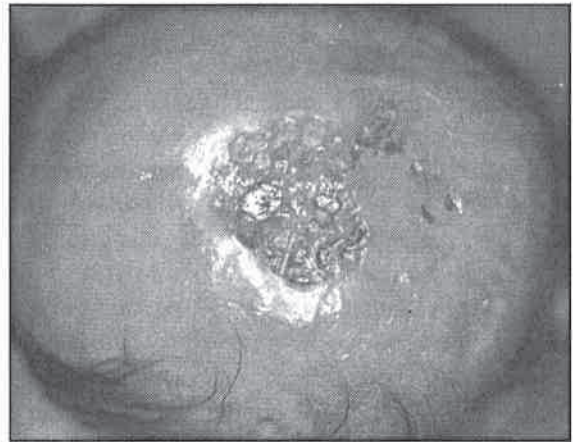


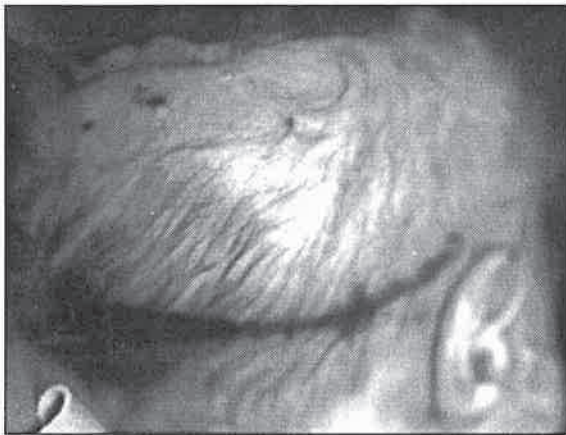
Figure 1: The child at 12 days after birth, with a full thickness scalp and skull defect.



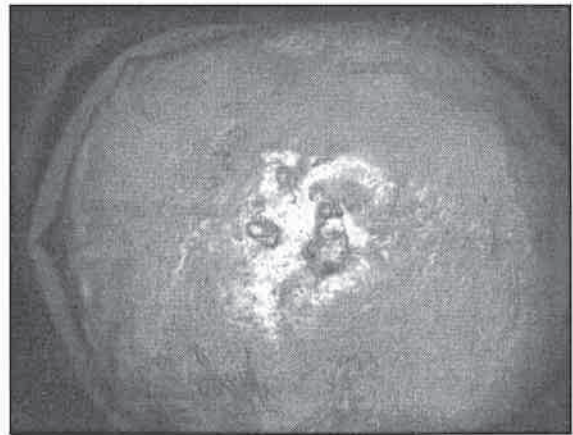
**Figure 2a:** Preoperative planning of two temporo-occipital scalp flaps (posterior view)



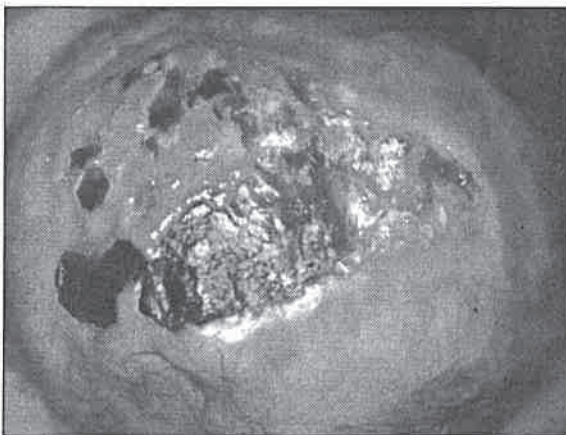
**Figure 3b:** The child at postoperatively 20<sup>th</sup> day.



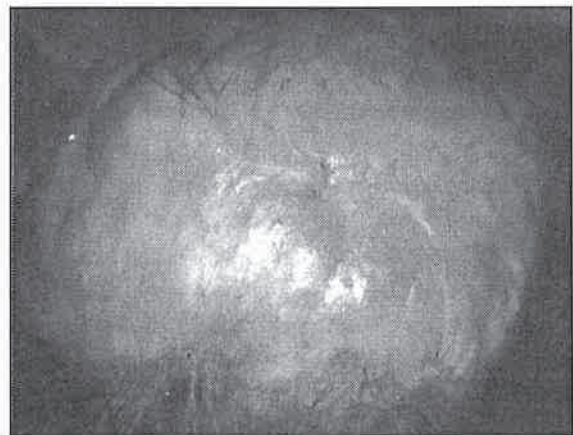
**Figure 2b:** Preoperative planning of two temporo-occipital scalp flaps (lateral view).



**Figure 3c:** The child at postoperatively 30<sup>th</sup> day.



**Figure 3a:** The child at postoperatively 10<sup>th</sup> day.



**Figure 4:** The child at 45 days of following conservative therapy.



### Discussion

Since Billard's description of the scalp and skull defects in 1828, multiple treatment regimens for aplasia cutis congenita of this area have been suggested. The stated aims of all these regimens have been to avoid meningitis, bleeding, and trauma to the unprotected brain in these children. Historical mortality figures for aplasia cutis congenita range from 20 to 55 percent. These deaths were primarily a result of associated congenital defects or bleeding from the sagittal sinus secondary to surgical intervention or drying eschar and separation (1,9). Treatment choices may be either conservative or operative.

The superficial lesions heal spontaneously under conservative antibacterial dressing. Conservative approaches involve regular dressing to keep the defect evenly moist, prevent desiccation and allow spontaneous epithelialization to occur. The operative treatments include primary closure, skin grafting, local scalp flaps with or without tissue expansion, free flaps and cranial vault reconstruction using bone grafts.(1,10)

Both conservative and operative managements are well accepted. The goal of conservative treatment is to allow granulation and healing by secondary intention whilst avoiding desiccation and eschar formation. Reports include saline dressing, continuous saline drips (8), betadine solution (11), bacitracin ointment (12) and silver sulphadiazine (1) dressing. Some authors have suggested conservative treatment in very large defects considered too large for local scalp flap coverage (10). The advantage of conservative management is the avoidance of potential operative risk to a newborn infant (13).

Conservative management may carry potentially fatal risks of infection and sagittal sinus haemorrhage. Therefore, definitive early wound cover is essential. Operative treatments have ranged from simple closure to scalp flaps as the most common reconstruction. Tissue expansion has been described although this technique may not always be possible. Split rib cranioplasty has been used to reconstruct the cranial vault in aplasia cutis congenita (13,14).

Split thickness skin grafting is simple and effective but will not provide adequate brain protection in the long term. Local flaps in the neonatal stage appear to be less reliable than expected. If employed, they should be delayed and transposed only after viability is confirmed. A skin biopsy of the adjacent skin may be helpful to confirm normal histology (15). Full-thickness flap coverage of the defect is critical and homograft application could allow adequate time for proper planning and delaying the flaps. Fluorescein, used to evaluate flap viability is definitely helpful (8).

Unrepaired bone defects proved to be the main source of problems in achieving long-term primary healing. The absence of dural cover and skull support allowed the growing brain to exert expanding forces on the local flaps and the intervening scar tissue causing recurrent breakdown. The weakness of the bony defect was associated with a progressive conehead deformity (13).

Use of a split rib cranioplasty and free latissimus dorsi muscle flap to reconstruct a complex defect of aplasia cutis congenita had been demonstrated. Microvascular free tissue transfer with split rib cranioplasty should be considered for very large defects of aplasia cutis congenita particularly in severe forms where brain is exposed or as a salvage procedure for reconstruction when conservative treatment or local scalp flaps have failed (13).

The philosophy of conservative management is taking advantage of the ability of rapid regeneration in the newborn (16). Silvadene therapy for cutis aplasia provides an antibacterial dressing that resists eschar drying and separation while allowing for epithelialization and bony ingrowth of the skull to occur. Furthermore, this conservative method of treatment eliminates the operative risks that may be encountered by the neonate (1).

We believe that the failure of our scalp flap was due to the abnormality of the adjacent abnormal skin features and inadequate blood supply. Scalp flaps in aplasia cutis congenita

have also partially or totally failed in other reported earlier cases. Local scalp flaps in the neonatal stages appear to be less reliable than expected. If employed they should be delayed and transposed only after viability is confirmed.

We believe that conservative treatment of aplasia cutis congenita have less complication rate and more reliability on neonates. Finally conservative management of aplasia cutis congenita may be considered even in large defects.

## REFERENCES

1. Wexler, A., Harris, M., Lesavoy, M. Conservative treatment of cutis aplasia. *Plast. Reconstr. Surg.* 86:1066, 1990.
2. Uppal, R. S., Moss, A. H. L. Aplasia cutis congenita associated with a lipoma. *Br. J. Plast. Surg.* 53:350, 2000.
3. Croce, E. J., Purohit, R. C., Janovski, N. A. Congenital absence of skin (Aplasia cutis congenita). *Arch. Surg.* 106:732, 1973.
4. Chitnis, M. R., Carachi, R., Galea, P. Familial aplasia cutis congenita. *Eur. J. Pediatr. Surg.* 6:100, 1996.
5. Fullana, F., Gonzales M., No M., Huc O., Gonzales-Mastre V. Aplasia cutis congenita of the scalp in five successive generations of one family. *Plast Reconstr Surg.* 95:214-215, 1995
6. Lynch P.J., Kahn E.A. Congenital defects of the scalp. *J. Neurosurg* 33:198, 1970
7. Glasson, D. W., Duncan, G. M. Aplasia cutis congenita of the scalp: delayed closure complicated by massive hemorrhage. *Plast. Reconstr. Surg.* 75:423, 1985.
8. Vinocur, C. D., Weintraub, W. H., Wilensky, R. J., et al. Surgical management of aplasia cutis congenita. *Arch. Surg.* 111:1160, 1976.
9. Islamoglu, K., Ozgentas, E. Aplasia cutis congenita of the scalp: excessive bleeding and reconstructive problems. *Ann. Plast. Surg.* 47:213, 2001.
10. Yang, J. Y., Yang, W. G. Large scalp and skull defect in aplasia cutis congenita. *Br. J. Plast. Surg.* 53:619, 2000.
11. Schneider B.M., Berg R.A., Kaplan A.M. Aplasia cutis congenita complicated by sagittal hemorrhage. *Pediatrics* 66:948-950, 1980
12. Six E.G., Kelly D.L. Jr., Conservative management of aplasia cutis congenita: case report. *Neurosurg.* 8:233-235, 1981
13. Theile, R. J. W., Lanigan, M. W., McDermant, G. R. Reconstruction of aplasia cutis congenita of the scalp by split rib cranioplasty and a free latissimus dorsi muscle flap in a nine month old infant. *Br. J. Plast. Surg.* 48:507, 1995.
14. Argenta, L. C., Dingman, R. O. Total reconstruction of aplasia cutis congenita involving scalp, skull, and dura. *Plast. Reconstr. Surg.* 77:650, 1986
15. Koshy, M. S., Waterhause, N., Peterson, D. Large scalp and skull defect in aoplasia cutis congenita. *Br. J. Plast. Surg.* 54:276, 2001
16. Yilmaz, S., Apaydin, I., Yenidunya, O., et al. Conservative management of aplasia cutis congenita. *Dermatol. Surg.* 23:402, 1997.

# LATELY FORMED URETERO-SACRO-CUTANEOUS FISTULA AND GIANT INTRAPELVIC URINOMA DUE TO GUNSHOT INJURY

Sezgin Yağcı\* ❖ Ali Avcı\* ❖ Lütfi Tahmaz\*\* ❖ Mutlu Sağlam\*\* ❖ A. Fuat Peker\*\*\*

## SUMMARY

The ureteral injuries due to gunshot are rarely seen complication. We determined uretero-sacro-cutaneal fistula absces formation and giant intrapelvic urinoma following 40 th day of gunshot injury. Ureteral injury due to abdominal gunshot injury is repated between 2.2-5 percent and due to genitourinary gunshot injuries 17 percent. In our study, we discussed the management and clinical diagnosis steps in a patient with gunshot ureteral injury.

**Key Words:** Ureteral Injury, Complications, Giant Urinoma

## ÖZET

**Ateşli Silah Yaralanmasına Bağlı Geç Dönemde Oluşan Büyük İntra Pelvik Ürinoma ve Üretero-Sakro-Kutaneal Fistül**

Ateşli silah yaralanmaları sonucu saptanan üreteral yaralanmalar nadir bir komplikasyondur. Biz ateşli silah yaralanmasından 40 gün sonra apse formasyonu, üretero-sakra-kutaneal fistül ve intrapelvik bölgede geniş bir ürinoma saptadık. Penetran üreteral travma abdominal penetran ateşli silah yaralanmalarından sonra %2.2-5 oranında, penetran genito-üriner travmalarda ise %17 oranında rapor edilmiştir. Biz sunduğumuz olguda ateşli silah yaralanmasına bağlı penetran üreteral travmalı bir hastada klinik evaluasyonumuzu ve tedavi seçeneklerimizi rapor ettik.

**Anahtar Kelimeler:** Üreteral Travma, Komplikasyonlar, Büyük Urinoma

Ureteral injuries usually result from penetrating abdominal trauma or iatrogenic causes. The reported incidence of penetrating ureteral injuries is 2.2 to 5 % of all abdominal missile wounds, 17 % of all penetrating genitourinary trauma (1). The total incidence of ureteral injury has been reported to be less than % 1 of all urologic trauma (2). The diagnosis of ureteral injury is often delayed due to the critical condition of the patient following gun shot injury (3,4). Delay in diagnosis also appeared to contribute to an increased morbidity in a patient

with a penetrating injury. We report a case with 40 days delayed diagnosis of ureteral injury and a large urinoma formation following exploratory laparotomy.

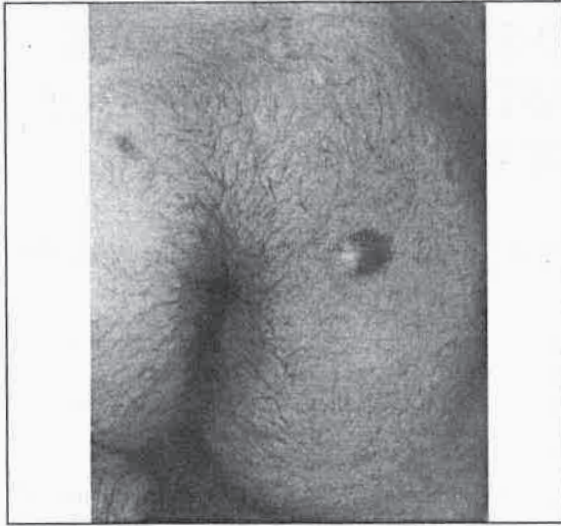
## Case Report

A 21-year-old man with 40 days ago performed laparotomy due to right iliac and umbilical region gun shot injury was referred from his local physician complaining of shaking chills, high fever, malasia, right gluteal swelling and

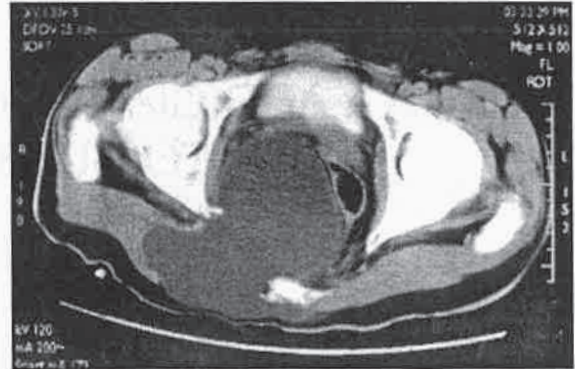
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\*\*\* GMMA Department of Urology, Prof. Dr.



**Figure 1:** Physical examination revealed fistula and gluteal swelling.



**Figure 2:** Computed pelvic tomography (CT) showed intrapelvic giant urinoma

watery discharge. Physical examination revealed fistula and wide gluteal swelling ( Figure 1) due to subcutaneously enlarged urinoma. Laboratory tests were showed leucocytosis and Klebsiella pneumonia in blood. Intravenous pyelography (IVP) showed a poorly functioning right kidney. He had a terminal colostomy located in left iliac region. Computed pelvic tomography (CT) showed intrapelvic giant urinoma extending to right gluteal skin through the sacral deformity due to gunshot injury (Figure 2). Retrograde pyelography revealed distal right ureteral extravasation near intramural ureter. 1200cc urine was drained from gluteal fistula tract. We did not try to place a double J catheter retrogradly to avoid ureteral avulsion. In the second step, we performed right percutaneous nephrostomy as a diversion. Tree months later, 1 cm ureteral stricture located in the early extravasation region was shown antegradly. Reconstruction with psoas-hitch and ureteroneocystostomy. was performed.

### Discussion

Nearly all patients with penetrating abdominal trauma should undergo exploratory laparotomy (1). However the true extend of injury in missile wounds is not immediately obvious

due to blast effect even with relatively low velocity bullets (5). The kinetic energy of such a missile transmits an explosive reaction with in the wound resulting in extensive damage to the surrounding tissues. This type of missile passes through the tissues nearly but creates a cavity 30 to 40 times it's size because of exertion of high pressure on adjacent tissues. The violence of expansion of missile tract disrupts and devitalizes tissue, blood vessels and bones at a distance from the path of the missile (6). The possibility of ureteral injury should be considered not only in the form of laceration or transection but also contusion alone, which might lead to future extravasation. Contusion damages the intima of small blood vessels in the ureter and produces thrombosis, ischemia and delayed necrosis resulting urinary leakage

(7-9). The extravasated urine causes lipolysis and stimulate on intense fibrous reaction which forms a thick wall. The lipolysis may be due to a mass effect as no direct effect of the urine on adipocytes has even be demonstrated (10).

We report the case with lately diagnosed giant intrapelvic and cutenously enlarged urinoma. To ourknowledge, this is the first case as a likely giant and lately formed urinoma ( 40 days later from gunshot ureteral injury). Urologist deal with trauma should not forget the blast effect of a missile, and should be aware of urinoma formation following gunshot injury despite exploratory laparotomy.

## REFERENCES

1. M. Al-Ali and Haddad LF.: The late treatment of 63 overlooked on complicated ureteral missile injuries: The promise of nephrostomy and role of autotransplantation. *J Urol*; 1996; 156: 1918-1920.
2. Laberge I, Hemsy YL, Dadoun G and Belad G: Avulsion of ureter by blunt trauma. *Urology*; 1979; 13: 172.
3. Campbell EW, Filderman NS, Jacobs SC: Ureteral injury due to blunt and penetrating trauma. *Urology*; 1992; 40: 216-20.
4. Tahmaz L ve ark.: Missed ureteral injuries following surgical exploration with percutaneous nephrostomy and drainage treatment, *Ulus Travma Dergisi*; 2000; 6: 284-287.
5. Carlton CE, Scott R, Guithre AG: The initial management of ureteral injuries: a report of 78 cases. *J Urol*; 1971; 105: 335.
6. *Emergency War Surgery: NATO Handbook*, Washington, DC. United States Printy Office, 1975: page 9.
7. Orkin LA: Trauma to the ureter. Pathogenesis and management. Philadelphia: FA Douis(o); 1964: 217-25.
8. Rohner TJ Jr: Delayed ureteral fistula from high velocity missiles. Report of 3 cases *J Urol*; 1971; 105: 63.
9. Case AS: Ureteral contusion and delayed necrosis from gunshot injury. *Urol*; 1978; 12: 195.
10. Rohners, Tuchschnid Y, Graber P: Perirenal urinoma. *Ann Urol (Paris)*; 1994; 28: 259-64.



# COEXISTING TRANSITIONAL CELL CARCINOMA AND LEIOMYOSARCOMA IN THE URINARY BLADDER: A CASE REPORT

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Gamze Numanoğlu\*\* ❖ N. Aydın Mungan\*\*\*

## SUMMARY

*Smooth muscle neoplasms of the urinary bladder are rare, and the majority of them are leiomyosarcomas followed by leiomyomas of conventional histopathological type. Simultaneous occurrence of transitional cell carcinoma and sarcoma as two separate primary tumors in the bladder is very rare. A new case of coexisting leiomyosarcoma and transitional cell carcinoma of the urinary bladder is presented.*

**Key Words:** Transitional Cell Carcinoma, Multiple Primary Tumors, Composite Tumor, Leiomyosarcoma.

## ÖZET

### Karsinom-Leiomyosarkom Birlikteliği

*Mesanein düz kas neoplazileri nadir görülmektedir. Bunların büyük çoğunluğu leiomyosarkom ve leiomyomdur. Mesanede iki ayrı primer tümör olarak transisyonel hücreli karsinom ve leiomyosarkom birlikteliği çok nadir görülen bir durumdur.*

*Mesanede transisyonel hücreli karsinom ve leiomyosarkom birlikteliği olan yeni bir olgu sunulmuştur.*

**Anahtar Kelimeler:** Transisyonel Hücreli Karsinom, Multiple Primer Tümörler, Leiomyosarkom.

Smooth muscle neoplasms of the urinary bladder are rare, and the majority of them are leiomyosarcomas followed by leiomyomas of conventional histopathological type. Simultaneous occurrence of transitional cell carcinoma and sarcoma as two separate primary tumors in the bladder is very rare. A new case of coexisting leiomyosarcoma and transitional cell carcinoma of the urinary bladder is presented.

## Case Report

A 65-year old man presented with a three-month history of painless gross hematuria, dysuria, urinary frequency and nocturia. An intravenous pyelography revealed normal upper urinary tract and a filling defect on the left lateral wall of the bladder. Pelvic computed tomography

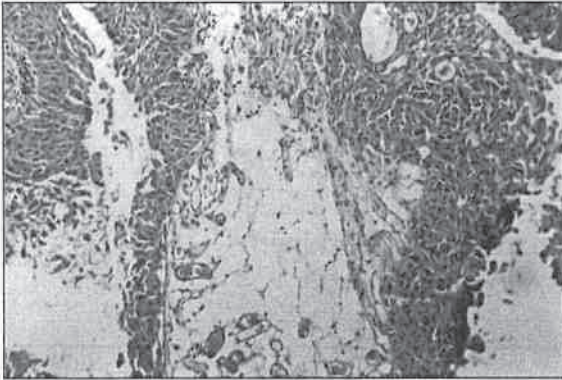
(CT) showed two solid papillary masses measuring 4x5 cm and 2x2 cm on the posterolateral wall of the bladder. No retroperitoneal adenopathy or metastases were found. Cystoscopic examination revealed a 4x5 cm polypoid, broad-based tumor on the left-posterior wall of the bladder. In addition, a 2x2 cm smooth polypoid mass with intact surface was determined above this tumor. These tumors were resected transurethrally and intravesical mitomycin-C was administered as a single instillation. Pathological examination revealed T1, grade 2 papillary transitional cell carcinoma and focal muscle invasive leiomyosarcoma, respectively (figure 1 and figure 2). Transitional cell carcinoma was composed of a central fibrovascular core that is covered by 8-10

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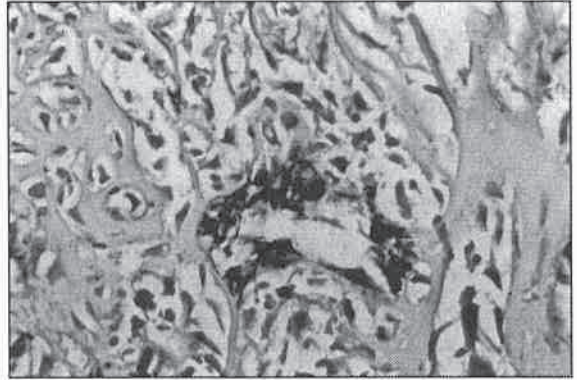
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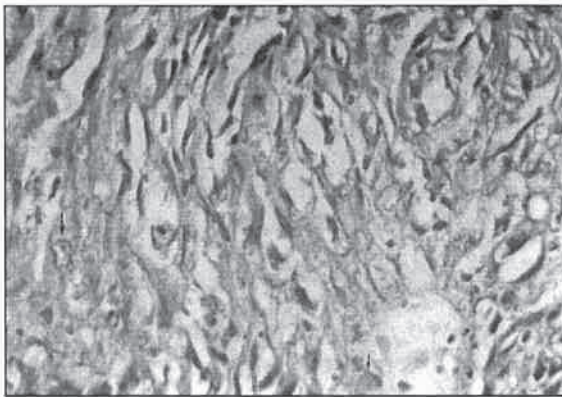




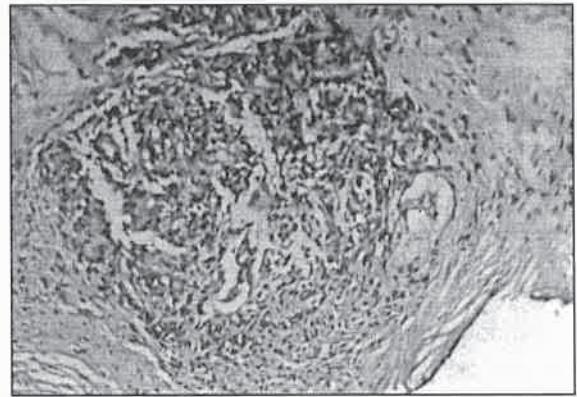
**Figure 1.** Transitional cell carcinoma with a central fibrovascular core. These cells had hyperchromatic nuclei (H&E X20).



**Figure 3.** Smooth muscle actin positive cells in focal leiomyosarcoma (SMA Ab1 Clone 1A4-Neomarkers X40).



**Figure 2.** Leiomyosarcoma; pleomorphism in spindle cells (H&E X40). Vesicular nucleus with distinct nucleolus and hyperchromatic nucleus (long arrow) and plumed nucleus (short arrow).



**Figure 4.** Cytokeratin 8 negative cells in focal leiomyosarcoma (Keratin 8 Ab3 Clone 35βH11-Neomarkers X20).

number of transitional cells. They had hyperchromasia of nuclei and crowding of cells. Leiomyosarcoma was composed of spindle cells that is blunt-ended, pleomorphic and had a prominent mitotic rate (3/40xHPF). The cells grew in long directional streams or fascicle and in immunohistochemical study it was painted positive with smooth muscle actin antibody (SMA Ab1 Clone1A4-Neomarkers) and negative with cytokeratin 8 (Keratin 8 Ab3 Clone 35βH11-Neomarkers). This component infiltrated in normal muscle tissue. Based on this pathological finding, a radical cystoprostatectomy with ileal

conduit diversion and pelvic lymphadenectomy was performed. Pathological examination of the cystectomy materials showed tumor-free bladder, granulomatous inflammation of the bladder and prostate, reactive hyperplasia of the lymph nodes. He was in good health 6 months after the cystectomy.

#### Discussion

The coexistence of two or more primary urinary bladder tumors of different histologic types are very rare. To our knowledge, 7 cases of coexisting sarcoma and transitional cell carcinoma of the urinary bladder have been

reported in the literature (1-6) . Of these 7 cases, the carcinomas were transitional cell carcinoma in all cases and the sarcomas were leiomyosarcoma in 6 cases and osteogenic sarcoma in 1 case. These cases differ from cases of carcinosarcoma in that the sarcomatous element and carcinomatous element do not admix (6). Karl T.K. Chen suggested that "unlike the dismal outcome of most cases of bladder carcinosarcoma the prognosis of cases with multiple primary tumors may be favorable especially if the coexisting tumors are all well differentiated"(6). Because of T1, grade 2 transitional cell carcinoma and focal muscle

invasive leiomyosarcoma a radical cystectomy was performed in our case. Sarcomas of the bladder may arise in patients with a history of radiation therapy and cyclophosphamide therapy (7,8). We could not discern any aetiologic or predisposing factor. Leiomyosarcomas may sometimes be amenable to treatment with partial cystectomy, but survival results may be compromised when conservative operations are performed for large or extensive tumors (9). Alabaster et al reported urethral recurrence after excision of leiomyosarcoma of the bladder (10). Our patient is under periodic control by urtehral wash cytology at 3-monthly intervals.

## REFERENCES

1. Powers JH, Hawn CVZ, Carter RD. Osteogenic sarcoma and transitional cell carcinoma occurring simultaneously in the urinary bladder: Report of a case. *Journal of Urology* 1956; 76: 263-269.
2. Hejtmanjik JH, Klatt WW. Co-existing carcinoma and sarcoma of the bladder. *Journal of Urology* 1960; 84: 320-321.
3. Mackles A, Immergut S, Grayzel DM, Cottler ZR. Carcinoma and sarcoma of bladder: Report of unusual simultaneous occurrence of both tumors. *Journal of Urology* 1948; 59: 1121-1126.
4. Uemura M, Nishimura K, Hirai T, Kanno N, Mizutani S, Miyoshi S, Yoshida K, Kawano K. Leiomyosarcoma and transitional cell carcinoma in the urinary bladder: a case report. (In Japanese) *Hinyokika Kyo* 2002; 48: 159-162.
5. O.Özteke, A.Demirel, N.E.Aydın, L.Memiş. Bladder leiomyosarcoma: Report of three cases. *International Urology and Nephrology* 1992; 24: 393-396.
6. Karl T.K Chen. Coexisting leiomyosarcoma and transitional cell carcinoma of the urinary bladder. *Journal of Surgical Oncology* 1986; 33: 36-37.
7. Sigal SH, Tomaszewski JE, Brooks JJ, Wein A, LiVolsi VA . Carcinosarcoma of bladder following long-term cyclophosphamide therapy. *Arc Pathol Lab Med* 1991; 115: 1049-1051.
8. In Sook Seo, Steve A. Clark, E.H. Jhonson. Leiomyosarcoma of the urinary bladder. *Cancer* 1985; 55: 1597-1603.
9. Swartz DA, Johnson DE, Ayola AG, Watkins DL. Bladder leiomyosarcoma: a review of 10 cases with 5-year follow-up. *Journal of Urology* 1985; 133: 200-202.
10. A.Michael Alabaster, Willis P. Jordan, JR. Mark S. Soloway, Ronald M. Shippel, Joseph M. Young. Leiomyosarcoma of the bladder and subsequent urethral recurrence. *Journal of Urology* 1981; 25: 583-585.

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