Volume: 23, Issue: 4, October 2012 pp.: 265-271 ORIGINAL ARTICLE / ORJİNAL MAKALE

PREVALENCE OF SPINA BIFIDA IN PATIENTS TREATED SURGICALLY FOR PILONIDAL SINUS DISEASE

CERRAHİ OLARAK TEDAVİ EDİLEN PİLONİDAL SİNÜSÜ OLAN HASTALARDA SPINA BIFIDA PREVALANSI

İlker ABCI[1],
İ.Teoman BENLİ[2],
Bülent ÇAPAR[3],
Selçuk ÇAMUŞCU[4]

[1]*General Surgery Specialist, Hisar Intercontinental Hospital, İstanbul.*
[2]*Prof. Dr., Department Chief, Orthopedics and Traumatology Specialist, Hisar Intercontinental Hospital, İstanbul.*
[3]*Orthopedics and Traumatology Specialist, Hisar Intercontinental Hospital, İstanbul.*
[4]*Radiology Specialist, Hisar Intercontinental Hospital, İstanbul.*

SUMMARY:

Patients with posterior fusion defects such as spina bifida and meningomyelocele are known to have a high incidence of perianal and sacrococcygeal cutaneous problems, especially pilonidal sinus disease. Therefore, we aimed to investigate the prevalence of spina bifida and lower vertebral deformities in patients with pilonidal sinus disease.

In this study, we retrospectively reviewed conventional lumbosacral X-rays and lumbar CT scans of 25 patients treated surgically for pilonidal sinus disease at our hospital between 2005 and 2012. There were 19 male (76.0%) and six female (24.0%) patients. The mean age was 29.0 ± 6.9 (14–42) for the male patients and 27.0 ± 8.3 for the female patients. Lumbosacral X-rays and CT scans were re-evaluated by both radiologists and orthopedic surgeons, and patients diagnosed with occult spina bifida (OSB) were recorded.

A total of three patients (12.0%) with OSB were found. Two of these patients had a single-level posterior fusion defect at L5 or S1, while defects at both the L5 and S1 levels were detected in the third patient.

As a result, the OSB prevalence was found to be 12.0% when pilonidal sinus disease patients were retrospectively reviewed, which is considered to be a relatively high ratio. This finding may be due to both diseases having common genetic factors in their etiopathogenesis. In the light of these findings, we suggest that patients with pilonidal sinus disease should be screened with lumbosacral X-rays prior to surgery, to prevent possible further problems.

Key words: Pilonidal sinus, spinal dysraphism, occult spina bifida, prevalence

Level of evidence: Retrospective clinical study, Level III

ÖZET:

Spina bifida ve meningomyelesel gibi posterior birleşme defekti olan hastalarda, perianal bölge rahatsızlıkları özellikle pilonidal sinüs görülme sıklığının yüksek olduğu düşünülmektedir. Bu çalışmada bu nedenle pilonidal sinüs hastalarında spina bifida ve alt lomber vertebra deformitelerinin prevalansının araştırılması amaçlanmıştır.

Bu çalışmada 2005 ile 2012 tarihleri arasında hastanemizde cerrahi olarak tedavi edilen 160 pilonidal sinüs hastanın geriye dönük verileri incelenmiş, bunlardan 23'ünün lumbosakral konvansiyonel grafileri ve lomber BT'leri retrospektif olarak taranmıştır. Bu hasta grubundaki hastaların 19 (% 76.0)'u erkek ve 6 (% 24.0)'sı kadındır. Ortalama yaş erkeklerde 29.0 ± 6.9 (14-42), kadınlarda 27.0 ± 8.3 olarak bulunmuştur. Hastaların lumbosakral bölgeyi içeren grafileri ve BT'leri, radyoloji ve ortopedi uzmanlarınca beraber değerlendirilmiş, spina bifida okülta (SBO) olan hastalar belirlenmiştir.

Yapılan incelemede 25 hastanın 3 (%12.0)'ünde SBO saptanmıştır. Bu hastaların birinde L5 ve S1 iki seviye, birinde L-5 ve birinde S-1 omurlarda tek seviye posterior birleşme defekti olduğu belirlenmiştir.

Sonuç olarak, pilonidal sinüs operasyonu geçiren hastaların retrospektif taramasında spina bifida okülta prevalansının % 12.0 gibi yüksek bir oranda olduğu saptanmıştır. Bu bulgu her iki hastalık grubunun etiopatogenezinde ortak bazı genetik faktörlerin rol oynadığını düşündürmektedir. Bu verilerin ışığı altında pilonidal sinüs nedeniyle gelen hastalarda lumbosakral bileşkenin radyolojik incelemesinin yapılmasının, ileride gelişecek bu bölgeye ait problemlerin önceden önlenmesi açısından yararlı olacağı fikri elde edilmiştir.

Anahtar Kelimeler: Pilonidal sinüs, spinal disrafizm, spina bifida, prevalans

Kanıt Düzeyi: Retrospektif klinik çalışma, Düzey III

Address: Op. Dr. İlker Abcı, Özel Hisar Intercontinental Hastanesi, Siteyolu Sok., No:7, İstanbul.
Tel.: 0216 5241300
E-mail: cutku@ada.net.tr
Received: 1st July, 2012
Accepted: 26th August, 2012

INTRODUCTION:

Shenkin and Hurt showed a connection between the pilonidal sinus and the spinal cord in 1944 [11]. Generally, spinal deformities in the lumbosacral region are termed spinal dysraphism, and are associated with findings such as hypertrichosis in the skin and dimples[4]. It has been proposed that, if there is a disturbance during the differentiation of the neuroectoderm from the epithelial ectoderm, deformations may be common in the skin, spinal cord and spine, and also in the mesoderm surrounding these tissues[3]. However, there are also some theories involving neuroectodermal fusion defects, such as the presence of air bubbles, primary germ defects and folic acid deficiency, and the etiopathogenesis of this disease is still unknown[4].

Spinal dysraphism includes a group of complex developmental abnormalities of the bone, nerve and connective tissue components of the spinal axis. These complex abnormalities include, from most to least serious, meningomyelocele, lipomeningocele, taut filum terminale, diastematomyelia, occult spina bifida (OSB), and caudal regression syndrome (sacral agenesis or lumbosacral agenesis). In most patients, these are accompanied by mid-line hemangiomas, sacral dimples and local hypertrichosis in the sacrococcygeal region[2,4-7].

It is believed that, in any case of sacral dimples or gluteal asymmetry, spinal dysraphism must be suspected, in particular OSB[1,8,10]. For a diagnosis of spinal dysraphism, for which many publications emphasize the skin findings, conventional X-rays, computerized tomography (CT) and magnetic resonance imaging (MRI) are used. Also, pes equinovarus, convex pes valgus, lower extremity atrophy, and developmental hip dysplasia can accompany spinal dysraphism. There are many cases in the literature showing sensory and bladder dysfunction[4].

There are many publications concerning the correlation of spinal dysraphism, dermoid cysts and pilonidal sinus[1-11]. Powell reported that 7.3% of spinal dysraphism patients had skin lesions. However, there are no studies on the prevalence of pilonidal sinus in OSB patients. In this study, 160 patients who received surgery for pilonidal sinus were studied retrospectively, and 25 patients with radiological surveys were included for calculation of the prevalence of OSB.

MATERIALS AND METHODS:

In this study, 160 patients who received surgery for pilonidal sinus between 2005 and 2012 were investigated, and 25 of them had lumbosacral X-rays or lumbar CT. These scans were surveyed retrospectively. In this group, ten patients (76.0%) were male and six (24%) were female. The average age was 29.0 ± 6.9 (14–42) for the male patients and 27.0 ± 8.3 for the female patients.

On general physical examination of the patients there were no other systemic or physical findings, and a neurological examination was normal. Laboratory work revealed no pathological findings. 15 (60%) patients had excision and primary closure, five (20.0%) had a Karydakis flap and five (20.0%) had a Limberg flap.

The lumbosacral X-rays and CT scans of the patients were evaluated by radiology and orthopedic specialists, and the patients with occult spina bifida (OSB) were determined.

The level of OSB and the number of levels involved were determined. When all patients were included, the OSB prevalence, the prevalence of single-level and two-level OSB, and the OSB prevalence per level were analyzed.

RESULTS:

In the evaluation, three (12.0%) of the patients had OSB. One (4.0%) of these patients had OSB at two levels, L5 and S1, while two (8.0%) had single-level OSB (Figure-1).

In the patients with single-level involvement, one (4.0%) had an L5 and the other (4.0%) had an S1 vertebral posterior fusion defect, and when the two-level OSB patient was included, the posterior fusion defect prevalence was 8.0% for L5 and 8.0% for S1 (Figure-2).

In the patient with a single-level S1 involvement there was also sacralization, and in the patient with a single-level L5 OSB, a 42-year-old female patient, there was spondylolisthesis with 50% dislocation (Figure-3).

DISCUSSION:

Discussions are still ongoing surrounding the development of pilonidal sinus disease. The idea of late-acquired etiologies is gaining in popularity, but the co-occurrence of the disease with spinal dysraphism has given rise to the idea that a similar mechanism may play a role in the etiopathogenesis of both conditions[4]. Harris and Miller suggested that midline skin and spinal defects can develop as a result of a deficiency in neuroectodermal separation[3].

Spinal dysraphism is a group of complex abnormalities during the development of the neural axis, involving the bone and nervous tissue of the spine. These complex abnormalities can be meningomyelocele, which can cause serious neurological defects or less complicated lipomeningocele, taut filum terminale, diastematomyelia, occult spina bifida (OSB), or caudal regression syndrome (sacral or lumbosacral agenesis). In many patients, it is accompanied by findings in the sacrococcygeal region, such as midline hemangiomas, sacral dimples and local hypertrichosis[2,4-7,9].

Powell et al., in a prospective study in 1975, showed high rates of spinal dysraphism in dermal sinus patients[8]. Gibson et al. carried out a prospective study in 1995 on 95 newborns, and showed that sacral dimples, local hypertrichosis, and midline posterior hemangiomas were important indicators of spinal dysraphism. This has been accepted by many books as a consensus[4,5]. In a pediatric radiology guide, radiological studies are recommended for patients with those skin findings[7].

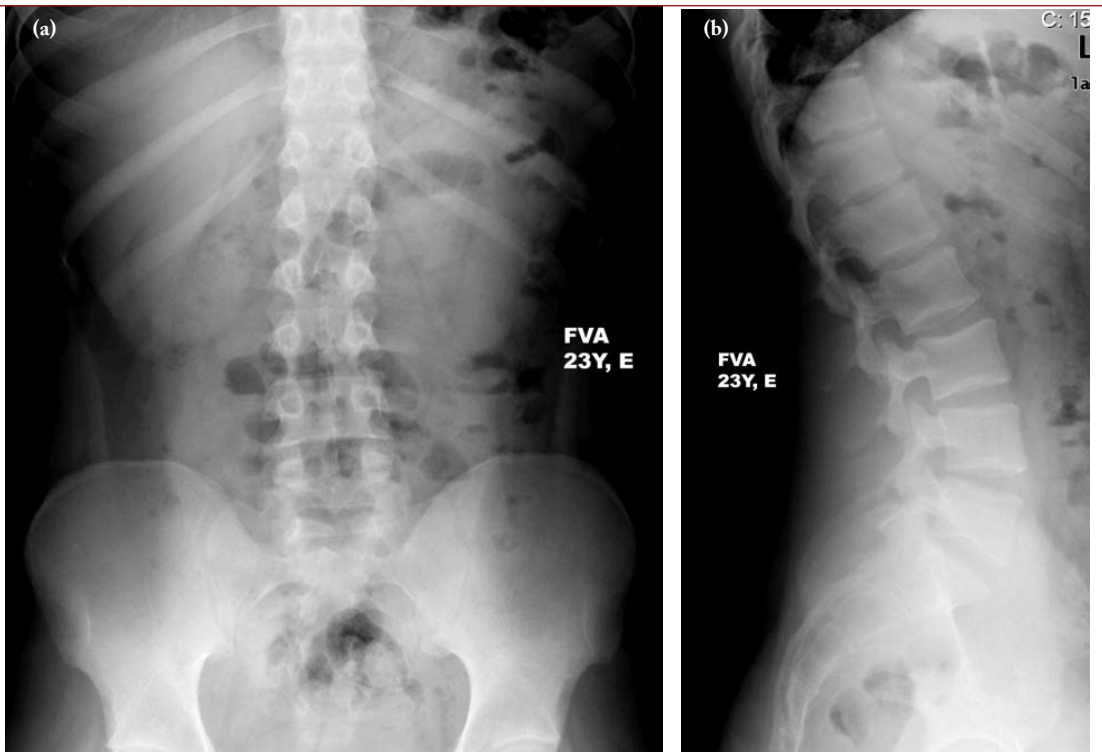

Figure-1. A 23-year-old male patient (F.V.A.). **a.** In anteroposterior lumbosacral X-rays, occult spina bifida is visible at the L5 and S1 levels, **b.** Lateral lumbosacral X-ray.

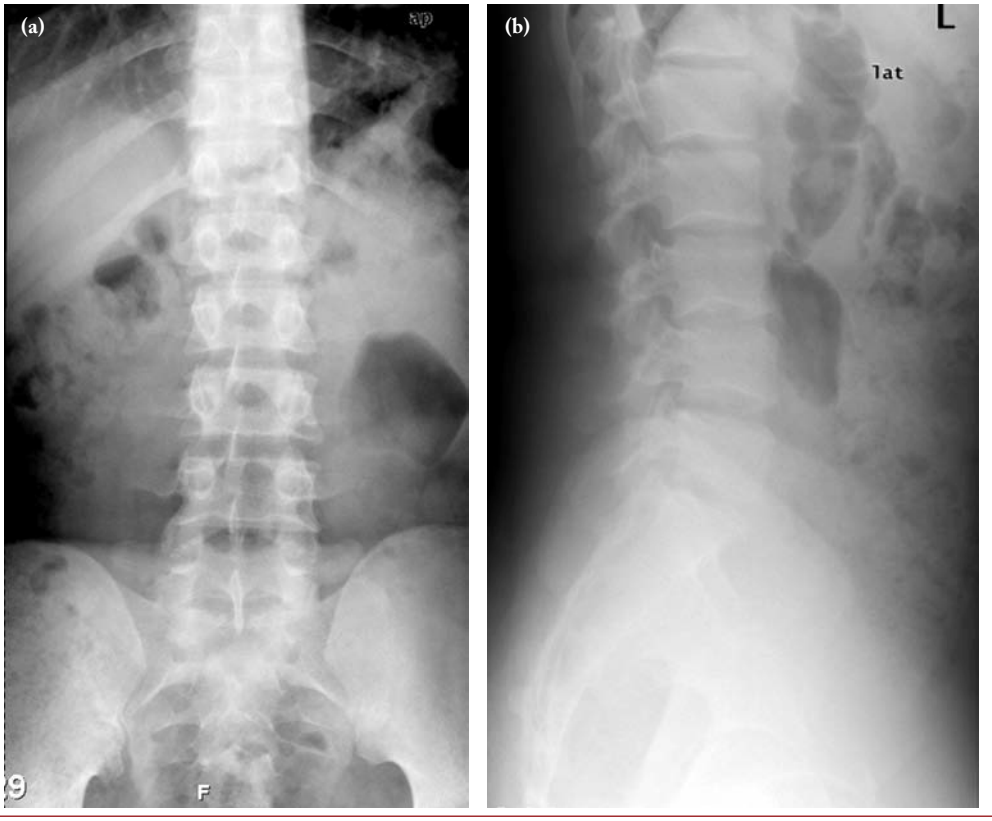

Figure-2. A 25-year-old male patient (S.A.). **a.** Anteroposterior lumbosacral X-ray showing occult spina bifida and sacralization at the S1 level, **b.** Lateral lumbosacral X-ray.

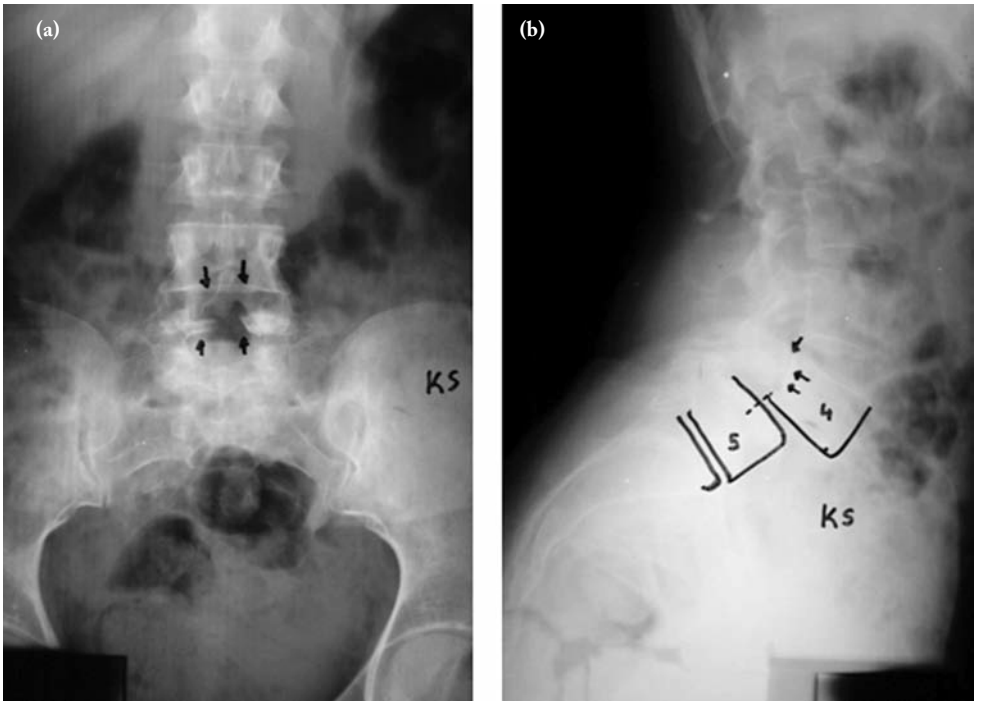

Figure-3. A 42-year-old female patient (K.S.). **a.** Anteroposterior lumbosacral X-ray showing occult spina bifida at the L5 level, **b.** Lateral lumbosacral X-ray showing 50% spondylolisthesis.

Değirmencioğlu et al., in a study in 2003 on 1000 newborns, showed that 50 babies had skin lesions, 28 had pilonidal sinus, and only three had spinal cord abnormalities with co-occurrence of pilonidal sinus[1]. Conversely, Lee et al. published a study in the same year including 5440 live births, in which only six had sacral dimples and none of the children had neurological deficit or spinal dysraphism[6]. As a result, even though there are many studies on the relationship between skin lesions and spinal dysraphism, the results are conflicting. In a study by Powell et al., the skin lesion incidence in spinal dysraphism patients was 7.3% [8].

Our study is the first study in the literature that considers the incidence of OSB among pilonidal sinus surgery patients. In our study, which included 25 cases, the OSB prevalence was found to be 12.0%.

It has been reported that OSB is generally observed at the lumbosacral junction[4]. In our study, all of the patients lesions were at the lumbosacral junction. 4% of the patients had two-level OSB and 8% of them had single-level OSB. In the group of patients with single-level involvement, the OSB prevalence was similar at the L5 and S1 levels. In one patient with single-level involvement, the development of spondylolisthesis showing 50% dislocation was discovered. This is believed to be the result of OSB influencing the etiopathogenesis of developmental spondylolisthesis.

Scatliff et al. reported neurological deficit with 16% cerebellar tonsillar ectopia and 6% hydromyelia in spinal dysraphism patients[10]. In our study, none of the patients had neurological deficit.

As a result, when pilonidal sinus patients were scanned retrospectively, the occult spina bifida prevalence was found to be high as 12%. This finding suggests that similar genetic factors may play a role in the etiopathogenesis of both diseases. In the light of these findings, radiological evaluation of the lumbosacral region of patients that are diagnosed with pilonidal sinus disease is recommended, to prevent further problems that could develop in the region.

REFERENCES:

1. Değirmenci S, Güven F, Celayir A, Demircioğlu B, Say A. Lumbosakral orta hat cilt lezyonlu yenidoğanlarda spinal kord anomalileri. *Turk Arch Pediatr* 2003; 38(2): 103-106.
2. Gibson PJ, Britton J, Hall DM, Hill CR. Lumbosacral skin markers and identification of occult spinal dysraphism in neonates. *Acta Pediatrica* 1995; 84(2): 208-209.
3. Harris HW, Miller OF. Midline cutaneous and spinal defects. Midline cutaneous abnormalities associated with occult spinal disorders. *Arch Dermatol* 1976; 112(12): 1724-1728.
4. Herring JA. *Tachdjian's Pediatric Orthopaedics*. 3rd Ed., WB Saunders Company, Philadelphia, Vol.2, 2002; pp: 1302-1310.
5. Kim DH, Betz RR, Huhn SL, Newton PO. *Surgery of Pediatric Spine*. Thieme, New York, 2008; pp: 199.
6. Lee ACW, Kwong NS, Wong YC. Management of Sacral Dimples Detected on Routine Newborn Examination: A Case Series and Review. *HK J Paediatr* 2007; 12: 93-95.
7. Pediatric and congenital imaging guidelines Spine and peripheral nervce disorders (PND), MedSolutions, NewYork, 2011.
8. Powell KR, Cherry JD, Hougen TJ, Blinderman EE, Dunn MC. A prospective search for congenital dermal abnormalities of the craniospinal axis. *J Pediatr* 1975; 87: 744-750.
9. Rauzino MS, Iskandar BJ, Oakes WJ. Occult spinal dysraphism. Contemporary *Neurosurgery* 1998; 20: 1-6.
10. Scatliff JH, Kendall BE, Kingsley DPE, Britton J,Grant DN, Hayward RD. Closed spinal dysraphism: analysis of clinical, radiological, and surgical findings in 104 consecutive patients. AJR 1989; 152: 1049–1057
11. Shenkin H, Hunt A. Sacrococcygeal sinus (pilonidal sinus) in direct continuity with the canal of the spinal cord. *Surg Gynecol Obst* 1944; 655-659.