Article

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Posterior fossa decompression with tonsillectomy in 104 cases of basilar impression, Chiari malformation and/or syringomyelia

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ABSTRACT

The prime objective in the surgical treatment of basilar impression (BI), Chiari malformation (CM) and/or syringomyelia (SM) is based on the restoration of the normal cerebrospinal fluid (CSF) dynamics at the craniovertebral junction through the creation of a large artificial cisterna magna. A small suboccipital craniectomy has been emphasized to avoid caudal migration of the hindbrain structures into the vertebral canal. Nevertheless, the results showed downward migration of the hindbrain related to that type of craniectomy. The authors present, otherwise, the results of 104 cases of BI, CM and/or SM, whose surgical treatment was characterized by a large craniectomy with the patient in the sitting position, tonsillectomy, large opening of the fourth ventricle and duraplasty with creation of a large artificial cisterna magna. A significant upward migration of the posterior fossa structures was detected by postoperative magnetic resonance imaging.

Key words: basilar impression, Chiari malformation, hindbrain herniation, tonsillectomy, craniovertebral decompression, syringomyelia.

Descompressão da fossa posterior com tonsilectomia em 104 casos de impressão basilar, malformação de Chiari e/ou siringomielia

RESUMO

O principal objetivo no tratamento cirúrgico da impressão basilar, malformação de Chiari e/ou siringomielia fundamenta-se na restauração dinâmica do líquido cefalorraqueano ao nível da transição craniovertebral e criação de ampla cisterna magna. Uma craniectomia suboccipital de pequenas dimensões foi proposta para evitar a migração caudal de estruturas rombencefálicas no canal vertebral. Entretanto, os resultados evidenciaram migração caudal do rombencéfalo. Os autores apresentam, por outro lado, os resultados de 104 casos de malformação de Chiari e/ou siringomielia, nos quais o tratamento cirúrgico se baseou em ampla craniectomia com o paciente em posição sentada, tonsilectomia, abertura ampla do quarto ventrículo e enxerto dural com consequente criação de ampla cisterna magna. Uma significante migração cranial das estruturas da fossa posterior foi detectada pelo emprego pós-operatório da ressonância magnética.

Palavras-Chave: impressão basilar, malformação de Chiari, herniação rombencefálica, tonsilectomia, descompressão craniovertebral, siringomielia.

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Basilar impression (BI), Chiari malformation (CM) and syringomyelia (SM) are the most frequent neurodysplasia at the craniovertebral junction. BI was originally described by Ackermann[1] (1790) in Cretins from the Alps. At that time, he described the small size of the posterior fossa, the elevation of the clivus and

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the projection of the border of the foramen magnum into the posterior fossa. In 1857, Virchow[2] introduced the term platybasia and in 1876[3] the denomination "basilar impression".

Chiari[4,5] (1891, 1895) described four types of cerebellar anomalies. Type I (14 cases) was characterized by downward displacement of the cerebellar tonsils and the medial portions of the inferior cerebellar lobes, which accompanied the medulla into the cervical spinal canal. Type II (seven cases) showed downward displacement of the portions of the cerebellum (1891), and portions of the inferior vermis (1895), pons, medulla and, at least, a part of the lengthened fourth ventricle, which reached the disc C4-C5, into the enlarged cervical spinal canal. In type III (one case), the hydrocephalic cerebellum, pons and medulla were inside a cervical meningocele (*hydroencephaloceles cerebellaris cervicalis*), through a spina bifida of the first three cervical vertebrae. In type IV (two cases), there was hypoplasia of the cerebellum without herniation of cerebellar structures into the spinal canal.

With respect to SM, Simon[6] (1875) introduced the term hydromyelia to designate the dilation of the ependymal canal by cerebrospinal fluid (CSF) and kept the term SM related to cavities that developed independently of the central canal of the spinal cord. It has been unanimously agreed - as can be seen in the present-day literature - that both are in different stages of the same pathological process. However, Finlayson[7] stated that hydromyelia is considered to be a congenital disturbance due to an incomplete regression of the fetal ependymal canal, whereas SM can be congenital or acquired.

The high prevalence of BI associated with CM in the Northeast of Brazil was reported by many authors[8-13], although there is no suitable known reason for this fact.

The main objective of this paper is to analyse the surgical results brought about by the use of a large craniectomy and tonsillectomy with the patient in the sitting position in 104 cases of BI, CM and/or SM.

METHOD

This study is based on a retrospective review of the patiens, 61 men and 43 women with occipitovertebral malformation, 67 (64.4%) of whom with BI, 102 (98%) with CM and 51 (49%) with SM. All patients underwent "osteodural-neural decompression" previously described by Gonçalves da Silva et al.[10]. This study was approved by the Ethics Committee of the Hospital Unimed, João Pessoa, Brazil.

As far as to surgical technique is concerned, the patients underwent a "osteodural-neural decompression" with large inferior occipital craniectomy in the sitting position, laminectomy varying from C1 to C3 level depending on the tonsillar herniation, dural opening in Y format, dissection of arachnoidal adhesions between the cerebellar tonsils, medulla and spinal cord, large opening of the fourth ventricle, intrapial aspiration of the cerebellar tonsils, suture of the residual pial sacs to the lateral dura mater in ascending position and, finally, a dural grafting was performed with the use of bovine pericardium creating, in this way, a large artificial cisterna magna.

RESULTS

One hundred and four patients with BI, CM and/or SM were presented. The clinical symptoms observed in the preoperative and follow-up examination are showed in Table 1, the clinical signs in Table 2 and the surgical findings in Table 3. In Table 1 and Table 2 were excluded

Table 1. Clinical symptoms observed in 95 cases of BI, CM with tonsillectomy and SM.

Symptoms	Cases/Number	R	%	A	%	U	%
Headache	42	42	100	–	–	–	–
Pain in the neck	64	64	100	–	–	–	–
Vertigo	55	50	90.9	–	–	5	9.0
Dysphagia	33	31	93.9	–	–	2	6.0
Nasal reflux	20	16	80	–	–	4	20
Rhinolalia	42	–	–	–	–	42	1000
Weakness of limbs	72	33	45.8	29	40.2	10	13.8
Paresthesia of limbs	52	28	53.8	10	19.2	14	26.9
Gait disturbances	44	18	40.9	14	31.8	12	27.2
Anhidrosis	12	5	41.6	1	8.3	7	58.3
Hyperhidrosis	13	5	38.4	1	7.6	7	53.8
Syringomyelic aches	18	7	38.8	1	5.5	10	55.5

R: regressed; A: amelioration; U: unchanged; BI: basilar impression; CM: Chiari marformation; SM: syringomyelia.

Table 2. Clinical signs observed in 95 cases with tonsillectomy.

Signs	Cases/Number	R	%	A	%	U	%
Lesion of the Vth nerve	27	24	88.8	–	–	3	11.1
Nystagmus	42	15	35.7	–	–	27	64.2
Abolition of gag and palatal reflexes	54	8	14.8	1	1.8	46	85.1
Lesion of the XIth nerve	51	18	35.2	–	–	33	64.7
Lesion of the XIIth nerve	3	2	66.6	1	33.3	–	–
Hypotony of superior limbs	50	8	16.0	–	–	42	84.0
Spasticity	49	35	71.4	7	14.2	7	14.2
Paresis of limbs	69	31	44.9	25	36.2	13	18.8
Hyperreflexia	73	8	10.9	–	–	65	89.0
Hoffmann's sign	30	20	66.6	–	–	10	33.3
Babinski's sign	28	14	50.0	–	–	14	50.0
Rossolimo's sign	22	10	45.4	–	–	12	54.5
Unsteady gait	44	18	40.9	14	31.8	12	27.2
Hypopallesthesia	72	3	4.1	1	1.3	68	94.4
Syringomyelic dissociation	43	12	27.9	13	30.2	18	41.8
Fasciculation	4	1	25.0	1	25.0	2	50.0
Atrophy	40	9	22.5	4	10.0	27	67.5

R: regressed; A: amelioration; U: unchanged; BI: basilar impression; CM: Chiari marformation; SM: syringomyelia.

six patients without follow-up and three patients who died. All the cases were followed with clinical studies and craniovertebral MRI for a mean of three years and a range of two months to 20.5 years. The age at surgery ranged from 18 to 74 years, with a mean of 40.7 years.

Sixty-seven out of 104 cases were associated with BI, 102 cases with herniation of the cerebellar tonsils, 74 with herniation of the brainstem, 51 with SM and 1 with syringobulbia, of which 22 patients presented a communication between the fourth ventricle and the hydromyelic cavity. As to SM and/or CM cases, the authors observed 55 cases of blockage of the foramen of Magendie. It was also observed 17 cases of impacted cisterna magna, ten of which were associated with SM without tonsillar herniation by the MRI in the supine position, but the tonsillar herniation appeared with the patient in the sitting position. In five other cases there was not association with SM, the tonsillar herniation also appeared with the patient in the sitting position in three cases and, finally, in two cases was not detected tonsillar herniation with the patient in the sitting position.

The length of the herniated tonsils varied from the end of C1 to the end of C3 level, following the caudal migration by C1 (18 cases), C1 and C2 (23 cases), C1 and C3 (six cases), C2 (31 cases), C2 and C3 (15 cases), and, finally, C3 (nine cases).

The vascular network anomalies in 69 cases were characterized by the missing of the posterior inferior cerebellar artery (PICA) on both sides in four cases, and missing on the left side in two cases, a left hypoplasic PICA in six cases, a left missing and a right large PICA in four cases, a left large and a right hypoplasic PICA in six cases, a right large and a left hypoplasic in seven cases, a right missing and a left hypoplasic PICA in two cases, and, finally, looping sign on the PICAS in 38 cases.

Table 3. Surgical findings observed in 104 cases of BI, CM and SM.

Findings	Cases/ Number	%
Thinning of the occipital bone	30	28.8
Thickening of the occipital bone	23	22.1
Thickening of dural and Atlanto-occipital ligament	19	18.2
Pulseless dura mater	53	50.9
Arachnoiditis	51	49.0
Impacted cisterna magna	17	16.3
Blockade of the forame of Magendie	55	52.8
Herniation of the brainstem	74	71.1
Tonsillar herniation	102	98.0
Impacted cisterna magna without Tonsillar herniation	2	1.9
Tonsil cyst	5	4.8
Communication of the fourth ventricle and the hydromyelic cyst	22	21.1
Vascular network anomaly	69	66.3
Syringobulbia	1	0.9
Syringomyelia	51	49.0
Basilar impression	67	64.4
Hydrocephalus	4	3.8

BI: basilar impression; CM: Chiari marformation; SM: syringomyelia.

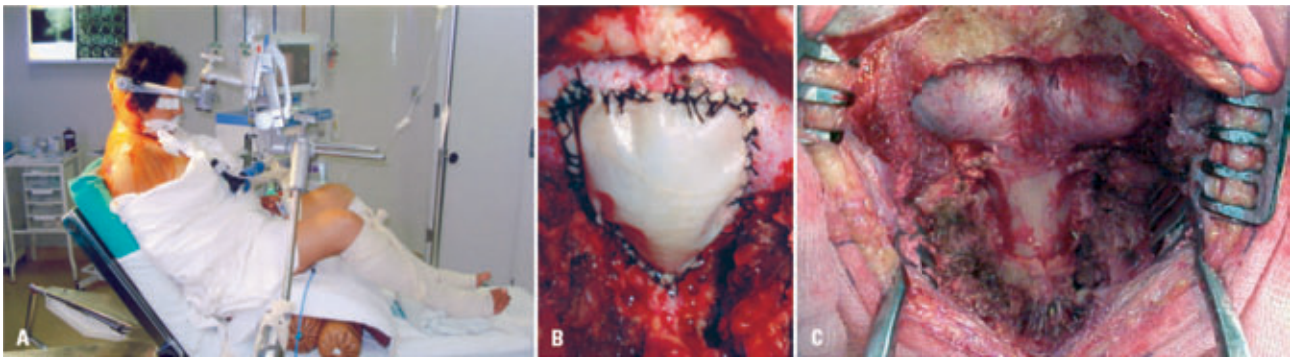

Fig 1. [A] Patient in the sitting position. [B] Duraplasty with creation of an artificial cisterna magna. [C] Large craniectomy of the posterior fossa.

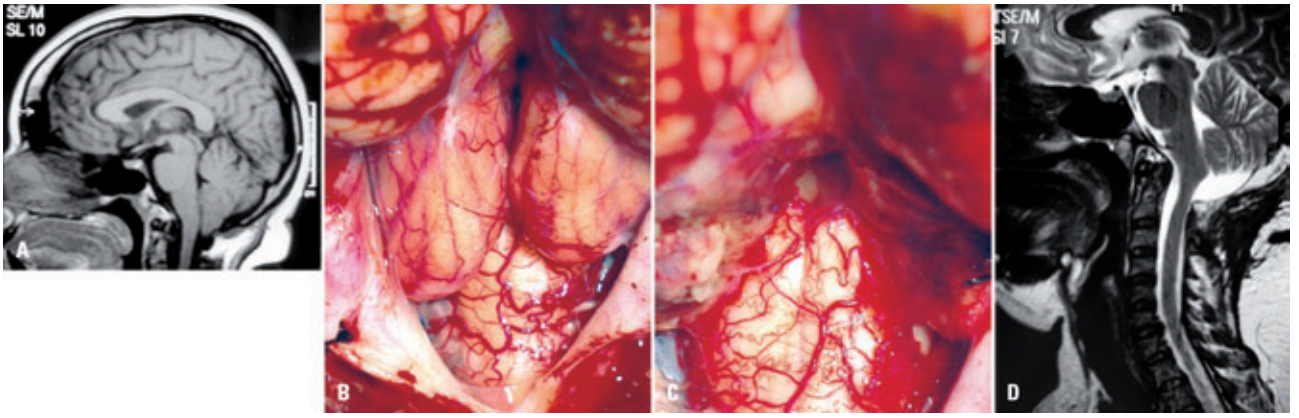

Fig 2. [A] Preoperative MRI demonstrating CM at the level of C1. [B] Extensive herniation of the cerebellar tonsils with the patient in sitting position. [C] Tonsillectomy and large opening of the fourth ventricle. [D] Postoperative MRI showing creation of a large artificial cisterna magna.

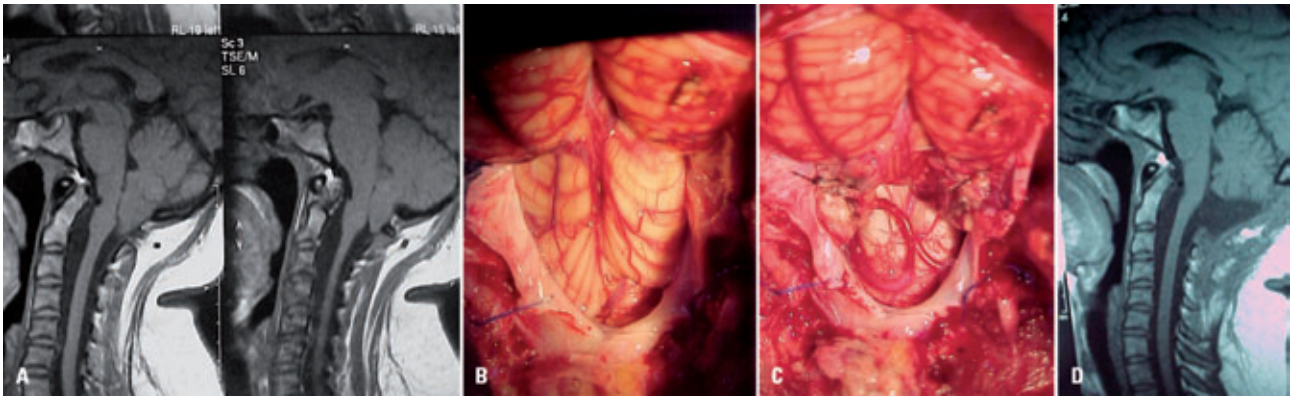

Fig 3. [A] Preoperative MRI depicting basilar impression (BI) and Chiari malformation (CM) at the level C1-C2. [B] More herniation of the cerebellar tonsils in the sitting position at the level of C3. [C] Tonsillectomy, large opening of the fourth ventricle and looping sign on the PICAS. [D] Postoperative MRI showing a large artificial cisterna magna.

DISCUSSION

The most frequent craniovertebral malformations of clinical importance are characterized by BI, CM and SM. BI was originally described by Ackermann[1] in Cretins from the Alps. He observed a small size of the posterior fossa, an elevation of the clivus, a low topographic situation of transverse sinuses and the projection of the border of the foramen magnum into the posterior fossa, which could not contain one-third of the hindbrain.

Primary BI is a neurodysplasia caused by an abnormal development of the axial neuroskeleton. It frequently comes in association with other congenital anomalies such as atlas assimilation, Klippel-Feil syndrome, Sprengel's deformity, CM, platybasia and SM as reported by

many authors[8-13]. Its association with others deformities is directly related to the theory of embryogenetic error. In addition, secondary BI is an anomaly of the basioccipital bone related to other abnormalities such as osteomalacia, osteitis deformans, osteogenesis imperfecta and Paget's disease.

The CM, on the other hand, is a caudal rhombencephalon abnormality resulting from the downward migration of the inferior cerebellar portions, mainly the tonsils (CM I) or herniation of the medulla and the caudal portion of the fourth ventricle (CM II), except the pons[4,5].

With respect to SM, Simon[6] introduced the term hydromyelia to designate the dilation of the ependymal canal by CSF and kept the term SM to cavities that developed independently of the central canal of the spinal cord. It has been unanimously agreed - as can be seen in the present-day literature - that both are in different stages of the same pathological process. However, hydromyelia is considered to be a congenital disturbance due to an incomplete regression of the fetal ependymal canal, whereas SM can be congenital or acquired[7].

Many theories have been presented in an attempt to explain the genesis of CM. Experimental studies conducted by Marin-Padilla[14] revealed that in CM, the basiochondrocranium is smaller due to an underdevelopment of the basioccipital bone. Due to a small posterior fossa, the ulterior development of the cerebellum will cause a herniation of the cerebellar tonsils. According to Marin- Padilla[14], the development of CM happens as a result of para-axial mesodermal insufficiency after the neural tube closure.

Regarding the surgical technique, all patients of our casuistic of 104 cases were operated in the sitting position which facilitates the surgical procedure. Gardner and Goodall[15] used this type of position for the operation of 13 out of 17 patients with CM.

The extension of the craniectomy varies in the literature. Because of the small size of the posterior fossa caused by BI and/or CM, especially when both anomalies are present, we prefer to use a large craniectomy which extends cranially to the transverse sinuses - frequently situated lower than those in normal persons - and laterally to 3-4 cm from the midline. The main reason for this large opening is to increase the volume of the posterior fossa and to create a large artificial cisterna magna, commonly absent in cases of hindbrain herniation, permitting, in this way, an upward migration of the herniated portions of the cerebellum and brainstem.

Batzdorf[16] and Duddy and Williams[17] stated that the herniation of the cerebellar structures and brainstem, as observed in the postoperative period, are attributed to a large craniectomy. A small craniectomy, on the contrary, might have prevented a decompression from maintained the blockage of the CSF flow and perpetuating, in this way, the craniospinal pressure dissociation.

Duddy and Williams[17], however, using a small craniectomy, revealed a frequent downward migration of the cerebellum and brainstem, and pointed this out as a cause of poor results. These authors observed a downward migration of the posterior fossa structures in 53% of their patients, whereas no change was noticed in 41% and a cranial migration of the hindbrain was observed in only one case.

Sahuquillo et al.[18] compared the results obtained in 10 cases in which a small craniectomy was performed, with other 10 patients that underwent an extensive craniectomy. An upward migration of the cerebellum and brainstem was observed in all the last patients, while in those which a small craniectomy was performed there was a downward migration in 7 patients.

The volume of the posterior fossa is notoriously reduced in cases of BI and/or CM as firstly described by Ackermann[1]. In recent times, however, some authors[19,20] using X-ray, computed tomography and magnetic resonance imaging (MRI) examinations have also demonstrated that the posterior fossa volume, in the presence of BI and CM, is smaller than those observed in normal people. Milhorat et al.[21] verified a decrease of 13.4 mL in the total volume of the posterior fossa and 40% (10.8 mL) in the CSF volume of this region.

Regarding the cerebellar tonsils, the majority of the neurosurgeons[21-26] leave them intact, performing especially the opening of the fourth ventricle, as we performed in 192 cases of our casuistic. Other surgeons[10,12,23], as observed in recent publications, recommend the dissection of the arachnoidal adherences of the tonsils and vessels and opening of the fourth ventricle. Furthermore, they recommend tonsillectomy as a protection against slump, eliminating the compressive effect over the medulla and spinal cord, as described by Williams[25], as well as it contributes to the creation of a large artificial cisterna magna, which facilitates an upward migration of the hindbrain structures. Williams[25] also recommended to remove part of the tonsils to ensure that the pathways are maximally opened. On the other side, Batzdorf[26] recommends lightly diathermying the pia mater over the surface of the tonsils. Williams[25] admitted that creating an artificial cisterna magna is better effected by a more substantial removal, causing no morbidity after evacuation.

Iskandar et al.[27] reported on five pediatric cases of SM without hindbrain herniation. All patients improved after undergoing posterior fossa decompression. Kyoshima et al.[28] described four similar cases with a good recovery of the patients after the decompressive operation. The au-

thors named the full cisterna magna as "tight cisterna magna" and designated Iskandar's description as "Chiari 0 malformation". Gonçalves da Silva et al.[29] named this type of cisterna magna as "impacted cisterna magna".

The authors observed the presence of the "impacted cisterna magna" in 17 out of the 104 cases. In ten consecutive patients with SM, no hindbrain herniation was detected by MRI in the supine position, but the herniation of the cerebellar tonsils was observed in all of them during the surgery in the sitting position[30]. In other five patients without SM there was no tonsillar herniation showed by MRI in the supine position, however, in the sitting position, they migrated caudally in three cases and, finally, in the other two patients, the cerebellar tonsils did not herniate.

Not only the cerebellar tonsils but also the brainstem and cerebellum have the tendency to migrate downward in the sitting position. In this way, we observed herniation of the cerebellar tonsils in 102 (98%) patients and downward migration of the brainstem in 74 (71.1%).

SM was observed in 51 (49%) patients, of which there was a communication between the fourth ventricle and the hydromyelic cyst in 22 cases (43.1%). Syringobulbia was detected in only one patient (0.9%). Blockage of the foramen of Magendie was observed in 55 (52.8%) patients, mainly caused by the presence of a dense membrane or adhesions between the cerebellar tonsils. Gardner and Goodall[15] described in many cases obstruction of the foramen of Magendie by a membrane representing an unperforated rhombic roof.

The vascular network anomalies in 69 cases were characterized by the missing of the left PICA in two cases, absence of both PICAS in four cases, a missing left PICA and a right large PICA in four cases, a left hypoplasic PICA in six cases, a left large PICA and a right hypoplasic PICA in six cases, a right large PICA and a left hypoplasic PICA in seven cases, a right missing PICA and a left hypoplasic PICA in two cases and, finally, looping sign on the PICAS in 38 cases.

As to postoperative complications, it was observed transient respiratory distress syndrome in two cases that resulted in death in one of them. Four cases of CSF fistula were treated with the use of external ventricular drainage with complete closure of the CSF leakage in two patients and one associated with hydrocephalus underwent a ventriculoperitoneal shunt (VPS). The authors observed pseudomeningocele (PM) in five cases, from these two associated with hydrocephalus underwent VPS, one patient underwent a lumboperitoneal shunt, other patient was reoperated and in just one case of PM, the reoperation was not indicated because of postoperative MRI did not reveal compression signs of the hindbrain structures. We have detected others complications such as hypertensive cerebral haemorrhage, epidural hematoma, hiccups in two cases, from these one lasted two days and the other lasted 20 days ceasing the hiccups only with the use of cyclobenzaprine, hypertensive pneumocephalus in two patients associated with hydrocephalus treated with VPS and, finally, two cases of hallucination lasted 24 hours in one patient and three days in another.

The causes of postoperative death in three patients should not happen in two. One patient with respiratory distress by the intubation was used a forced mistaken retroflection of the head, which provoked lesion of the brainstem and the consequent death. An other had a hypertensive cerebral haemorrhage and died, and by the last one was observed a epidural hematoma recurrance, in which failed the intubation and the patient died.

Taking into account, however, the promising clinical outcomes as seen in the present study, we have felt encouraged to justify our choice of "osteodural-neural decompression" associated with tonsillectomy as used in this group of patients.

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