CASE REPORT OF IPSILATERAL CEREBELLAR DIASCHISIS IN AN ADULT: EVIDENCED FROM BRAIN PERFUSION SCAN

Yuan-Ling Chang[1,2,3], Li-Yin Chen[1], Shin-Tsu Chang[1,3,4]

Abstract

Damage to the cerebrum sometimes causes a rare phenomenon known as 'ipsilateral cerebellar diaschisis' (ICD). Most documented cases of ICD are in children less than six years of age. Here we report a rare case of ICD, evidenced from single-photon emission computerized tomography, in a middle-aged patient due to subarachnoid hemorrhage and intracerebral hemorrhage. We will discuss the possible mechanism behind ICD in an adult and provide a summary of all documented ICD cases. We aim to highlight the importance to survey for ICD, as it might be an indicator of prognostic outcome.

Key Words: ipsilateral cerebellar diaschisis, subarachnoid hemorrhage, intracerebral hemorrhage, single-photon emission computerized tomography, Kernohan-Woltman notch phenomenon

INTRODUCTION

Diaschisis was initially coined in 1914 by Dr. Constantin von Monakow, although the concept was first brought up by Dr. Charles-Édouard Brown-Séquard in the late 1800s.[1] The term refers to a focal disturbance in the brain, at a distance from the original site of injury, but producing transneuronal suppression of function and metabolism, theorized to be due to the connection via white matter tracts.[1,2]

Damage to the cerebrum sometimes causes a phenomenon known as 'crossed cerebellar diaschisis' (CCD), signalling a decrease in blood flow and metabolism contralateral to the damaged supratentorial area.[3] However, cerebrum damage sometimes causes a lesser-known phenomenon, 'ipsilateral cerebellar diaschisis' (ICD), a much rarer entity with few reports.[4-9] According the few reports on ICD, most are documented in children less than six years of age. This time, we are reporting a case of a 49-year-old male with a seemingly mature neurologic system prior to his incident before developing ICD.

CASE PRESENTATION

We report a case of a 49-year-old, left-handed male, who appeared to be in fine health, with no known systemic diseases, prior to 2021/03/01.

On that day, there was sudden onset of conscious disturbance (E1V1M1) accompanied

Correspondence: Dr. Shin-Tsu Chang
Department of Physical Medicine and Rehabilitation, Kaohsiung Veterans General Hospital; No. 386, Dazhong 1st Rd., Zuoying Dist., Kaoshiung 813414, Taiwan[4]
Phone: +886-935-605578; E-mail: ccdivlaser1959@gmail.com
Department of Medical Education and Research, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan[1]
Department of Anesthesiology, Kaohsiung Armed Forces General Hospital, Kaohsiung, Taiwan[2]
Department of Physical Medicine and Rehabilitation, Tri-Service General Hospital, School of Medicine, National Defense Medical Center, Taipei, Taiwan[3]

by massive vomiting. At the Emergency Department of the local hospital, emergent computed tomography (CT) scan revealed diffuse subarachnoid hemorrhage (SAH) with intraventricular hemorrhage (IVH) and hydrocephalus. He underwent an emergency operation of external ventricular drainage for decompression with intracranial pressure monitor insertion. However, follow-up brain computed tomography angiography the next day revealed anterior communicating artery aneurysm, and transcatheter arterial embolization was performed on 3/4.

Follow-up CT scan was performed on 3/6, which disclosed progressive brain edema with delay hemorrhage and midline shift. Emergent right frontotemporal craniotomy with removal of intracerebral hemorrhage (ICH) and external ventricular drain monitor was inserted bilaterally. The third brain CT on 3/15 revealed resolution of hematoma, thus left ventriculo-peritoneal shunt and right cranioplasty was performed on 3/18.

Patient was transferred to our hospital and was in a chronic bed-ridden state of total dependence with no drastic improvement in mental alertness. Tc-99m ethyl cysteinate dimer single-photon emission computerized tomography (SPECT) was performed and conflicting data was obtained: Moderate hypo-perfusion in the right lateral zone of the cerebellum (white arrow) was noted, yet along a lower tans-axial cut, decreased perfusion uptake in right frontal, temporal, and occipital regions, as well as decrease in the right basal ganglion/thalamus (red arrow) was disclosed, which favors ICD. (Figure 1) But with a higher trans-axial cut, there was decreased perfusion uptake in the left frontal and parietal (blue arrow), which indicates CCD. Patient is now undergoing bedside rehabilitation, but no overt changes in mental state is noted.

DISCUSSION

The SPECT examination provided us with a perplexing conundrum, as a higher trans-axial cut favors cerebellar suppression to be contralateral, while a lower-trans-axial cut favors the suppression to be ipsilateral. As mentioned in the case presentation, we deemed the presence of ICD more significant than CCD, due to the more extensive damage to the right cerebrum. However, it is curious that our patient developed ICD, as he is an adult male with a matured neurological system prior to his A-com rupture. To our knowledge, there has been very few documentations of adults developing ICD as opposed to CCD.

The neural mechanism for ICD is not fully understood and most prior reports indicated that ICD was more likely to present in children before the age of six, the reason was proposed to be due to differences in maturation of the corticopontocerebellar fibers during childhood.[5] Only the reports by Lenzi GL, et al. and Chang ST, et al. provided cases that were in adulthood.[4,9] (Table 1)

We had considered the possibility of cerebral suppression due to mechanical damage from initial trauma. However, although A-com artery is at the base of the cerebellum and relatively close to the cerebellar, it still doesn't explain why the suppression is mainly ipsilateral rather than diffuse.

We can first take a look at Patronas NJ, et al.'s study, where five normal subjects were recruited as the control group. In their study, even CCD subjects had a lower ipsilateral cerebellar metabolism in comparison with the control group. As the spinocerebellar input is largely ipsilateral, they had suggested that this phenomenon could be related to decreased spinocerebellar or other ascending input, possibly due to hemiparesis and subsequent patient's decrease usage of one side.[10] Even though our case differs from Patronas NJ, et al.'s, in that our subject presents with ICD more predominantly, the mechanism they proposed for ipsilateral hypometabolism in the cerebellar could still be applicable.

To delve more deeply into the reason behind our patient's ICD, we can take notice of the two major systems connecting the cerebral cortex with the cerebellum, the cortico-ponto-cerebellar

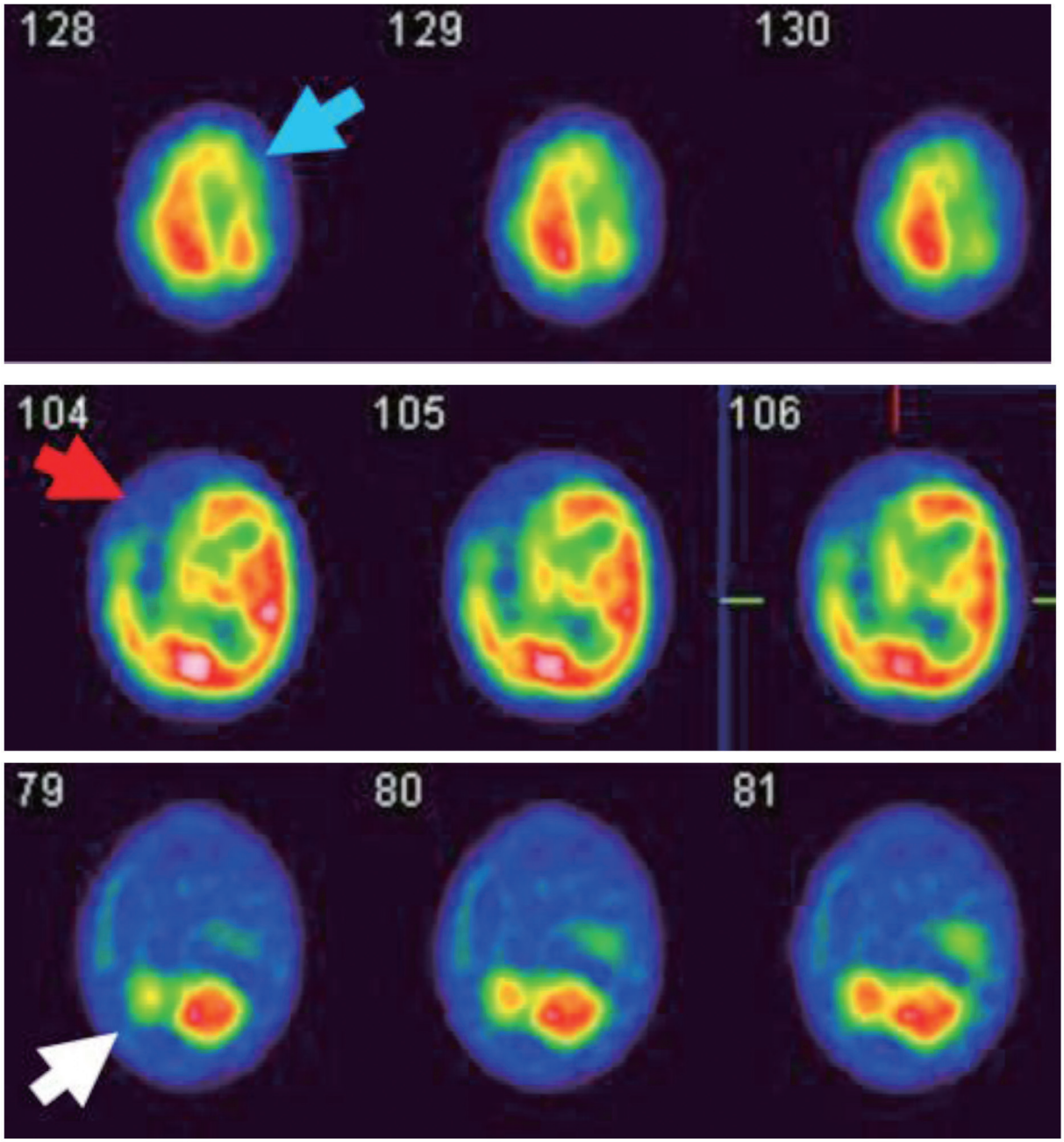
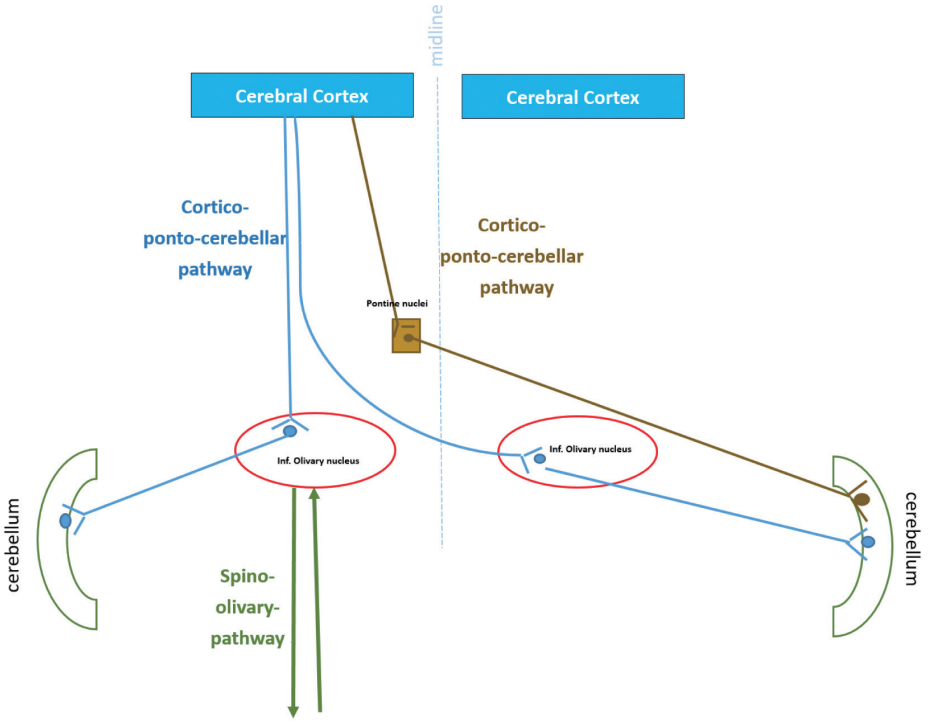


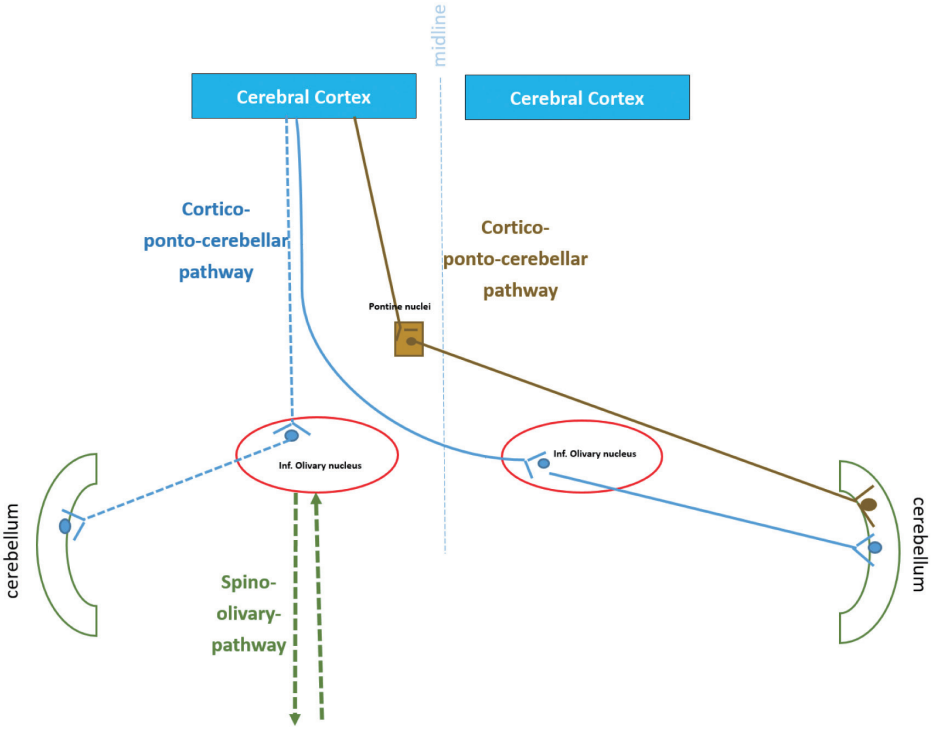
Fig. 1. SPECT brain images.

- Upper panel, A trans-axial view of frontal and parietal lobes showing decreased uptake in the left side (blue arrow), favoring CCD.
- Middle panel, A trans-axial view of the central portion of the brain showing decreased uptake in right frontal, temporal, and occipital regions, as well as right basal ganglion/thalamus (red arrow). Although the upper panel favors CCD, however, due to the right cerebrum having more extensive damage than the left cerebrum, the presence of ICD being more significant than CCD was determined.
- Lower panel, A trans-axial view of the cerebellum showing decreased uptake in the lateral zone of the right cerebellum (white arrow).

*CCD: crossed cerebellar diaschisis / ICD: ipsilateral cerebellar diaschisis



Normal neural pathways



Possible suppressed pathways in our patient with ICD:
suppressed pathways are indicated as dotted lines

Fig. 2. Schematic illustration of involved neural pathways of cerebellar diaschisis.

Table 1. Review of past ICD cases from major databases

	Year	Case No./ Age	Case Condition	Finding of ICD	Finding of CCD	Diagnostic Tool
Our Case	2021	1 case, 49 y/o male	Spontaneous A-com rupture with SAH and ICH	Prominent finding	Secondary finding (see explanation in Fig.1)	Tc-99m SPECT
Lenzi GL, et al.[4]	1982	15 cases, 36 y/o-88 y/o	acute, completed, ischemic cerebral infarcts	3 cases: 2 had previous infarcts in the other cerebral hemisphere, and the third had an infarct in the anterior temporal lobe.	6 cases	PET and oxygen-I5 steady-state inhalation technique
S, Hamano, et al. [5]	1991	25 cases, 10 mo.- 14 y/o	Hemiplegia→7: cerebral palsy, 18: acquired brain injury	5 cases: 2 with cerebral palsy, 3 with early acquired brain injury before three years of age	5 cases: acquired brain injury after seven years of age	N-isopropyl-p-I-123-iodoamphetamine SPECT
S, Hamano, et al. [6]	1993	55 cases; age range: 1 week to 18 years	Hemiplegia→8: infarction, 20: cerebral palsy, 11: ICH,4: Moyamoya disease, 9: Acute hemiplegia in childhood, 1: Brain abscess, 1: Measles encephalitis, 1: Cerebral contusion	10 cases: 3: acute subdural hematoma during infancy, 1: acute hemiplegia in childhood, 1: brain abscess, 1: moyamoya disease, 4: cerebral palsy of probable perinatal onset. All brain damage occurred before the age of four years	6 cases: brain injuries after 7 years, 5 months of age	123I-IMP SPECT
Sztriha, L., et al.[7]	1996	14 cases, age range 11 months - 11 years,	Hemiplegia→7: hemiplegic cerebral palsy (5: Birth asphyxla, 2: unknown), 1: unknown, 1: Stroke and congenital heart disease, 3: hemiconvulsion-hemiplegia-epilepsy syndrome, 2: trauma	3 cases: All birth asphyxla with hemiplegic cerebral palsy Insult during perinatal	1 case: traumatic brain injury, 4 years-old	CT and 99mTc-HMPAO SPECT
Chakravarty, A.[8]	2003	3 cases, age range: before 1 y/o	Infantile Hemiplegia Syndrome	1 case of ICD (ipsilateral cerebellar atrophy), insult appeared pre- or perinatal	2 cases of CCD (crossed cerebellar atrophy), insult appeared <1 y/o	MRI
Chang, ST, et al.[9]	2005	1 case, 23 y/o male	Left subdural hemorrhage after head injury; subsequent scoliosis	Prominent finding	none	99mTc-HMPAO SPECT

and cortico-olivo-cerebellar tracts. While both are predominantly crossed, the ascending olivocerebellar input to the inferior olivary nucleus, also known as the spino-olivary tract, is both crossed and uncrossed.[9] Our patient's ICD could be related to the suppression of the uncrossed portion of the spino-olivary tract, though through what mechanism is still unclear.

One important detail about our patient is that he is left-handed and, according to his sister, his whole family is predominantly left-handed. Left-handedness has been associated with less lateralization of the brain than right-handers,[11] with some research suggesting that the dominant hemisphere inhibits the non-dominant hemisphere in right-handers, but less inhibition occurs in left-handers.[12] Perhaps the decrease lateralization of our patient's cerebral hemispheres contributed to his development of ICD in lieu of the more common CCD. However, as previous reports of ICD didn't document the handedness of their patients, more data is needed to support this speculation.

It is also possible that our patient could have both CCD and ICD. CCD is one of the most common forms of diaschisis and the most well studied, with the main mechanism proposed to be interruption of cerebrocerebellar pathways due to damage of the predominantly excitatory cortico-ponto-cerebellar projections.[2] Studies have suggested that CCD is a prognostic factor to the functional recovery of a patient, especially concerning motor functions.[13] Nonetheless, the Kernohan-Woltman notch phenomenon cannot be ignored.

We believe that ICD could be a prognostic factor as well. However, the exact prognostic nature of ICD still needs further investigation. In the event that our patient has an ICD coupled with CCD, this explains our patient's limited recovery and severity of disease.

CONCLUSION

Research on the prognostic properties of crossed cerebellar diaschisis (CCD) and ipsilateral cerebellar diaschisis (ICD), two entities under the 'diachisis' umbrella, are still being carried out. Our case report concerning ICD with the possible coupling of CCD, adds to the limited literature surrounding the topic. Hopefully this will shed more light on this enigmatic neurological phenomenon and inform physicians in clinical settings that ICD might be a prognostic indicator as well.

References

1. Carrera E, Tononi G. Diaschisis: past, present, future. Brain. 2014 Sep;137(Pt 9):2408-22. DOI: 10.1093/brain/awu101. Epub 2014 May 28.
2. Jiménez-Caballero PE. Crossed cerebellar atrophy: Update. J Neurosci Rural Pract. 2012 Sep;3(3):235-6. DOI: 10.4103/0976-3147.102588.
3. Pantano P, Baron JC, Samson Y, et al. Crossed cerebellar diaschisis. Further studies. Brain. 1986 Aug;109 (Pt 4):677-94. DOI: 10.1093/brain/109.4.677.
4. Lenzi GL, Frackowiak RS, Jones T. Cerebral oxygen metabolism and blood flow in human cerebral ischemic infarction. J Cereb Blood Flow Metab. 1982 Sep;2(3):321-35. DOI: 10.1038/jcbfm.1982.33.
5. Hamano S, Nara T, Nozaki H, et al. Crossed cerebellar diaschisis demonstrated by SPECT in hemiplegic children. No To Hattatsu. 1991 Jan;23(1):58-64. Japanese.
6. Hamano S, Nara T, Nakanishi Y, et al. Secondary changes in cerebellar perfusion (diaschisis) in hemiplegia during childhood: SPECT study of 55 children. Pediatr Neurol. 1993 Nov-Dec;9(6):435-43. DOI: 10.1016/0887-8994(93)90022-5.
7. Sztriha L, al Suhaili AR, Prais V, et al. Regional cerebral blood perfusion in children with hemiplegia: A SPECT study. Neuropediatrics. 1996 Aug;27(4):178-83. DOI: 10.1055/s-2007-973783.
8. Chakravarty A. MR evaluation of crossed and uncrossed cerebral-cerebellar diaschisis. Acta Neurol Scand. 2003 Jul;108(1):60-5. DOI: 10.1034/j.1600-0404.2003.00039.x.
9. Chang ST, et al. Occurrence of subtle scoliosis in accordance with development of ipsilateral cerebellar diaschisis on brain SPECT in a patient with head injury. Ann Nuci Med Sci. 2005.18(3):173-179. DOI: 10.6332/ANMS.1803.007.
10. Patronas NJ, Di Chiro G, Smith BH, et al. Depressed cerebellar glucose metabolism in supratentorial tumors. Brain Res. 1984 Jan 16;291(1):93-101. DOI: 10.1016/0006-8993(84)90654-1.
11. McManus IC, Van Horn JD, Bryden PJ. The

Tapley and Bryden test of performance differences between the hands: The original data, newer data, and the relation to pegboard and other tasks. Laterality. 2016 Jul-Nov;21(4-6):371-396. DOI: 10.1080/1357650X.2016.1141916. Epub 2016 Feb 8.
12. Tzourio-Mazoyer N, Petit L, Zago L, et al. Between-hand difference in ipsilateral deactivation is associated with hand lateralization: fMRI mapping of 284 volunteers balanced for handedness. Front Hum Neurosci. 2015 Feb 6;9:5. DOI: 10.3389/fnhum.2015.00005.
13. Sin DS, Kim MH, Park SA, et al. Crossed Cerebellar Diaschisis: Risk Factors and Correlation to Functional Recovery in Intracerebral Hemorrhage. Ann Rehabil Med. 2018 Feb;42(1):8-17. DOI: 10.5535/arm.2018.42.1.8. Epub 2018 Feb 28.

同側小腦失聯的成人個案：腦部灌流掃描診斷

張元齡[1,2,3]，陳俐因[1]，張幸初[1,3,4]

摘要

腦傷有時候會造成一個罕見的現象稱作「同側小腦失聯」。大部分同側小腦失聯的個案都發生在年齡小於六歲的兒童中，但我們這次要報告的是一個蜘蛛膜下腔出血和腦內出血造成的中年男子的同側小腦失聯，由單光子射出電腦斷層掃描診斷。我們會討論成人同側小腦失聯的可能機轉，並提供一個目前所有可查到文獻中的同側小腦失聯表格摘要。我們希望能強調要做同側小腦失聯的臨床檢查，因爲可能是一個重要的預後指標。

關鍵詞： 同側小腦失聯，蜘蛛膜下腔出血，腦內出血，單光子射出電腦斷層掃描

通訊作者：張幸初醫師
813414 高雄市左營區大中一路 386 號；高雄榮民總醫院復健部 [4]
電話：07-342-2121；傳真：07-342-2288；E-mail：ccdivlaser1959@gmail.com
高雄榮民總醫院教學研究部 [1]
高雄國軍總醫院麻醉部 [2]
三軍總醫院國防醫學大學復健部復健部 [3]