Case Report

The Outcome of Surgical Treatment for Tumors of the Craniocervical Junction

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Objective: *The authors report the clinical, radiological, and surgical findings of patients with craniocervical junction tumors surgically treated in the institution over the last 8 years.*
Material and Method: *A retrospective study was performed. Clinical, radiological, and operative data were evaluated, and follow-up information was obtained from outpatient examinations, and telephone interviews.*
Results: *There were 25 patients consisting of nine chordomas, eight meningiomas, three cysts, two schwannomas, one each of aneurysmal bone cyst, plasmacytoma, and metastasis. Twenty-nine operative procedures were performed, classified as 12 anterior, nine posterior-lateral, and eight posterior approaches. Gross total removal was achieved in 17 cases, subtotal removal in six cases, and partial removal in two cases. Re-operation was performed in six cases. Median follow-up time was 31 months. The authors found significant improvement in Karnofsky Performance Scale scores.*
Conclusion: *Appropriate surgical approaches provide successful tumor removal with less surgical morbidities, nevertheless recurrent tumors occasionally occur, and so, long-term follow-up is mandatory.*

Keywords: *Craniocervical junction tumors, Foramen magnum, Chordoma, Meningioma, Surgical approach*

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Craniocervical junction (CCJ) is a biomechanical and anatomical unit comprising the clivus, the foramen magnum, and the upper two cervical vertebrae. Tumors of CCJ may arise from osseous parts, surrounding soft tissues, or the neural structures contained within it[1]. Each lesion has a different growth pattern, with different bony destruction and neurovascular structures involvement. The complex anatomy of CCJ, along with its deep, central, and vital location, creates special problems and challenges for operative treatments of a tumor in this region. Appropriate surgical approaches for removal of tumors can prevent injury to nearby neurovascular structures and cause less instability of CCJ.

Because these are rare locations of tumors in the central nervous system and among the most formidable neurosurgical problems, the authors retrospectively analyzed the 8 year-experience at King Chulalongkorn Memorial Hospital.

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Material and Method

From January 1997 to December 2004, 25 patients with CCJ tumors were operated on in the neurological surgery division, King Chulalongkorn Memorial Hospital. The authors excluded patients undergoing only tissue biopsy and patients with tumors originating elsewhere and extending into CCJ (for example jugular foramen, upper clivus, cerebello-pontine angle, and spinal canal).

Case records, operation reports, radiological findings, and follow-up data were reviewed. For patients who could not be followed on a regular basis, the authors obtained the follow-up data by telephone interview. The clinical course was documented using the Karnofsky Performance Status (KPS) score.

Preoperative imaging consisted of plain x-rays, computed tomographic (CT) scans, and magnetic

resonance imaging (MRI) enhanced with gadolinium. Angiography was not performed routinely.

Major tumor locations were classified by anatomical relation around the CCJ (on axial plane) into; *anterior* for tumors involving anterior midline; *posterior* for tumors involving the posterior midline behind the dentate ligament; and the authors add *lateral* extension on each for tumors located between anterior midline and the dentate ligament. Tumor volume was not calculated as the geometry of tumors in this region tends to be complex and irregular.

A standard neuro-anesthetic technique was used in all patients. Somatosensory evoked potentials, when indicated, were monitored throughout the operation.

With respect to the location of the tumors, surgical approaches were classified as

1) Posterior midline approach: for accessing to a dorsal surface of cervicomedullary junction consisting of laminectomy or hemilaminectomy of upper cervical spine and a craniectomy in midline

2)Postero-lateral approach: for accessing a lateral or ventral surface of cervicomedullary junction consisted of laminectomy or hemilaminectomy of upper cervical spine and a craniectomy toward the sigmoid sinus varying on bone work for degree of tumor extension

3) Anterior midline approach:

3.1) Transbasal approach: for accessing the anterior skull base and upper clivus

3.2) Trans-sphenoidal approach: for accessing the upper two-thirds of the clivus

3.3) Transoral approach: for extradural lesion from midclivus down to the level of the C3 vertebral body and laterally for 2 cm to either side of the midline

3.3) Transmaxillary approach: for tumors that extend lateral to the occipital condyles and anterior to the pterygoid fossa behind the maxillary sinus with inferior limit at the atlas level but C3 vertebral body can also be accessed if maxilotomy is performed.

3.4) Median glossotomy and midline mandibulotomy: for further exposure down to the fifth cervical vertebra.

4) Antero-lateral (lateral extra-pharyngeal) approach: for accessing the lower clivus and upper cervical spines. It is limited in its access to the treatment of basilar invagination and intradural pathology

Extent of tumor resection was based on surgeon's observation recorded in the operative report and on postoperative radiological findings. The authors divided degree of resection into three categories: 1) gross total resection; 2) subtotal resection, in which some part of the tumor was left on vital structure, such as the vertebral artery, perforating arteries, or cranial nerves; and 3) partial resection, in which incomplete resection of tumor mass was performed.

Dural defect closure, if possible the authors completed watertight repair with fascia or pericranium, in deep region or unsecured closure the authors usually place fat graft and/or fibrin glue to prevent CSF leakage.

Spinal stability was evaluated in pre-, intra-, and post operative periods. Fusion procedures were performed if instability is evident.

Post operative complication was defined as new neurological deficit without subsequent recovery.

Post operatively, all patients underwent computed tomography in the same admission and then followed imaging examinations for surveillance on tumor recurrence.

Measurement presented in means, median, or range and standard deviation. Statistical analysis was conducted using student's t tests for paired variables. A probability values of less than 0.05 indicating statistical significance.

Results

Between January 1997 and December 2004, 25 patients were operated for craniocervical junction tumors. The average age of the patients was 40.5 years (range 7-83 yr). There were 16 women (64%) and nine men (36%): a ratio of 1.7:1. Length of stay was not evaluated as a significant number of patients remained in the hospital for non-medical issues. The follow-up period ranged from 1 to 70 months (median 31 months).

The average duration of symptoms prior to surgery was 8.4 ± 9.9 months. The most common presenting symptoms and signs were neck or head pain (64%), weakness (60%) followed by sensory deficit, sphincter disturbance, cranial nerve deficits, and respiratory dysfunction (Table 1). A combination of two or more of the aforementioned symptoms, signs, or findings was more the rule than the exception. The average Karnofsky score at presentation was 68.4.

The majority of the tumors were located in anterior (48%) or antero-lateral (44%) regions (Table 2).

Twenty-nine operations were performed, single staged operations performed in 21 cases. They consisted of six anterior, one antero-lateral, seven posterior, and seven postero-lateral (Table 3).

Two staged operations were performed in four cases due to extensiveness of the tumor or inadequate

Table 1. Summary of presenting symptoms and signs (n = 15)

Symptoms and Signs	No. of patients (%)
Pain or headache	16 (64)
Motor weakness	15 (60)
Sensory deficit	12 (48)
Respiratory dysfunction	2 (8)
Sphincter disturbance	7 (28)
Cranial nerve deficits	
Tongue weakness (XII)	4 (16)
Shoulder weakness (XI)	2 (8)
Swallowing difficulties (IX, X)	2 (8)
Diplopia (VI)	34 (16)

Table 2. Locations of tumors along the craniocervical junction (n = 15)

Location	No. of patients (%)
Anterior	12 (48)
Antero-lateral	11 (44)
Postero-lateral	2 (8)
Posterior	0 (0)

Table 3. Types of operation performed in a single-staged procedure

Types of operation	No. of operations (n = 21)
Anterior approach	
Transbasal approach	0
Transsphenoid approach	2
Transmaxillary approach	2
Transoral approach	1
Glossotomy and split mandible	1
Antero-lateral (transcervical) approach	1
Postero-lateral approach	7
Posterior approach	
SOC $\pm$ laminectomy	3
Laminectomy alone	4

first attempt approach. They consisted of transbasal approach then trans-sphenoidal approach, postero-lateral approach then transoral approach, transmaxillary approach then postero-lateral approach, and posterior midline approach then transbasal approach (Table 4).

Gross total removal was achieved in 17 cases (68%), subtotal removal six cases (24%), and partial removal two cases (8%).

Common pathological findings included chordoma (36%) and meningioma (32%). Others were arachnoid cyst, endodermal cyst, schwannoma, aneurysmal bone cyst, plasmacytoma, and metastasis (Table 5).

Gross total removals were achieved mostly in meningioma (88%), cyst (100%), schwannoma (100%), plasmacytoma (100%) and less likely in chordoma (44%), aneurysmal bone cyst (0%), metastasis (0%). In four cases that need two-staged operations, there were three chordoma and one aneurysmal bone cyst, gross total removals were successful in two cases Table 6.

Surgery related complications were as follows; new 9^{th} and 10^{th} cranial nerve deficits in five cases requiring long-term tracheostomy and nasogastric or gastrostomy tube feeding, two cases of 6^{th} cranial nerve palsy that did not improve in the follow-up period; two CSF leakage, one of which improved after CSF diversion (lumbar drainage), the other required surgical repair by fat graft and CSF diversion; two meningitis (1 case occurred after CSF leakage) that responded well to antibiotic; one hydrocephalus requiring placement of a shunt.

Mean preoperative KPS score 68.4 comparable to that of mean postoperative KPS score on discharge 70.4 ($p = 0.364$, $n = 25$).

All cases of chordoma, plasmacytoma, and metastasis received postoperative fractionated conventional radiotherapy but none in the cases of meningioma, schwannoma, cyst, and aneurysmal bone cyst. None of the patients received proton beam therapy or radiosurgery since it was not available in the institution.

Table 4. Data from 4 patients performed in a two-staged procedure

Case	Age (yrs), Sex	Approaches	Extent of resection	Pathological report
1	50, F	Transbasal then transsphenoidal	ST	Chordoma
2	19, F	Posterolateral then transoral	GT	Chordoma
3	52, M	Transmaxillary then posterolateral	GT	Chordoma
4	17, M	Posterior midline then transbasal	P	Aneurysmal bone cyst

GT: gross total; ST: sub-total; P: partial

Re-operation for tumor removals was performed in six cases (11 operations) for recurrent or residual tumors that caused neurological deficits including four chordomas, one cyst, and one aneurysmal bone cyst. All but one (1 cyst) were subtotally removed due to invasion of the tumor into vital structures. Fusion procedures were performed in four cases of chordoma, one case of aneurysmal bone cyst and one case of plasmacytoma.

There were no perioperative deaths. Five patients died during the follow-up period. The first, a patient with clival chordoma underwent transbasal then transsphenoidal approaches for subtotal tumor removal and had shunt placement due to postoperative hydrocephalus. Ten months later, she had shunt infection and expired. The second, a patient died of aspiration pneumonia 15 months following surgery for recurrent clival chordoma. The third, a patient died of disease progression 1 month following partial removal of metastatic adenocarcinoma. The fourth, a patient undergoing subtotal removal of chordoma followed by 3 more operations of recurrence. He recovered well after each operation. However, he died of pneumonia 2.5 years after the initial operation. The last patient with plasmacytoma underwent surgical removal and radiotherapy and then multiple chemotherapy courses. She died of respiratory tract infection 2 years later.

In the survival cases, excluding three cases that were lost to follow-up (median follow-up period 31 months), the authors found that mean KPS score improve significantly from 69.41 preoperatively to 85.29 ($p = 0.003$, $n = 17$) at the follow up period.

Table 5. Pathological report in 25 patients

Pathological report	No. of patient (%)
Chordoma	9 (36)
Meningioma	8 (32)
Cyst (2 arachnoid cysts, 1 endodermal cyst)	3 (12)
Schwannoma	2 (8)
Aneurysmal bone cyst	1 (4)
Plasmacytoma	1 (4)
Metastasis	1 (4)

Discussion

Because of the location, craniocervical junction tumors remain one of the most challenging tumors to surgical treatment. They contribute to approximately 5 percent of all spinal tumors and 1 percent of intracranial tumors[2]. Their rarity limits experience in their treatment. The authors' results differ from previous reports[3-8] due to varying pathological forms of tumors, definition of craniocervical region and tumor location.

There is no single symptom or neurological finding pathognomonic for a lesion in this location as in previous reports[9]. In addition, the generous size of the subarachnoid spaces at the cervicomedullary junction as well as the possibility of expansion of tumors into the high nasopharynx makes symptoms arise only after the lesions have achieved large volume[1].

Approaches to craniocervical junction tumors were once considered a "no man's land"[10]. Traditionally, a posterior midline sub-occipital approach and C-1 laminectomy has been used in the operative treatment for these tumors, however this approach provides limited access to anterior midline, primarily due to interposition of the cerebellum, brain stem, and numerous cranial nerves. The most frequently utilized anterior route is the transoral approach[11], which provides a direct access to the craniocervical junction. This approach is limited laterally by the pterygoid plates, the hypoglossal canals, the Eustachian tubes, and the width between the vertebral arteries on either side at the atlas and axis vertebrae. On the other hand, the

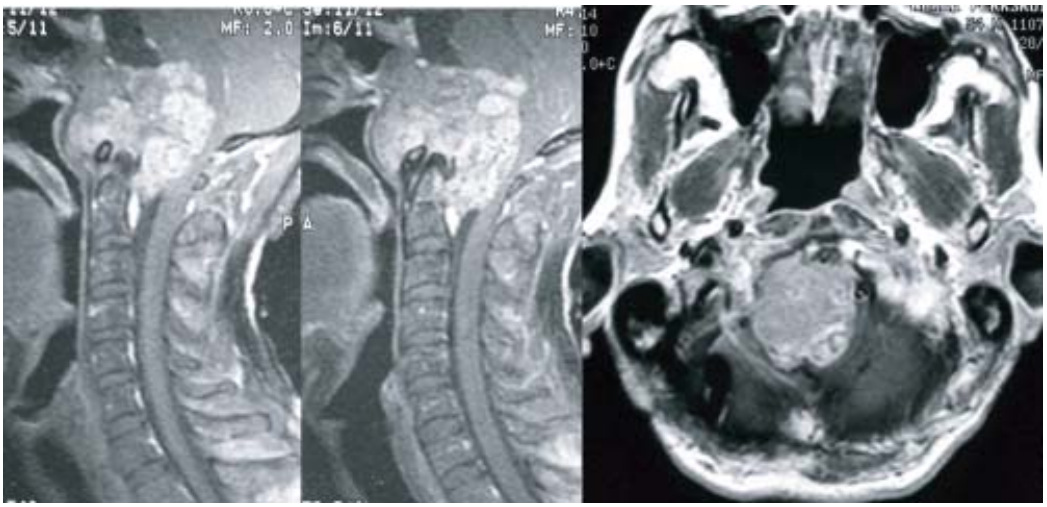
Table 6. Results stratified by pathological findings (n = 25)

Pathological report	No. of patients	2 staged operations	Gross total removal (%)	Recurrent/residual tumor operations	Fusion procedures
Chordoma	9	3	4 (44)	4	4
Meningioma	8	0	7 (88)	0	0
Cyst	3	0	3 (100)	1	0
Schwannoma	2	0	2 (100)	0	0
Aneurysmal bone cyst	1	1	0 (0)	1	1
Plasmacytoma	1	0	1 (100)	0	1
Metastasis	1	0	0 (0)	0	0

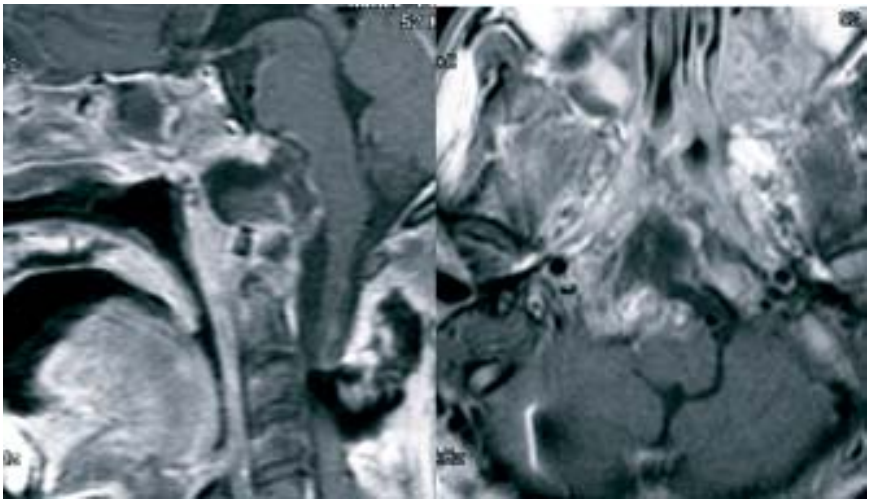
limitations of the pterygoid plates can be overcome by using a transmaxillary route[12, 13] with down fracture of the maxilla, and further extension can be obtained by an extended maxillotomy[14]. The wider exposure obtained with mandibulotomy and midline glossotomy allows the lower limit to be taken down to the C5 vertebral level[15,16]. However, these anterior approaches have disadvantages of contaminated operative field causing meningitis or a CSF fistula, difficult dural closure, and destabilization of the craniocervical junction.

Transbasal approach provides access to anterior skull base. Limitations occur by distance between optic nerves and need to work beneath sella turcica. The authors combined transbasal with other approaches for craniocervical junction tumors.

Trans-sphenoid approach[17,18] provides a short operative depth to the midline of the upper two-



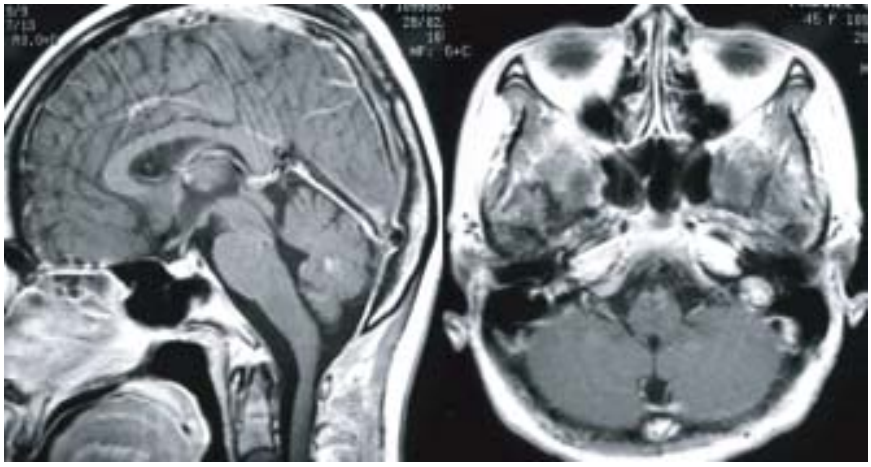
A



B

Fig 1. A 52 year old male patient presented with weakness of the right limb for 2 weeks
A: Preoperative MRI revealed a ventro-lateral extradural mass lesion on the craniocervical junction causing cord compression, the authors performed 2 staged procedures: transmaxillary approach then right posterolateral approach with gross total tumor removal and occipitocervical fusion. Pathological report was chordoma
B: Postoperative film revealed complete decompression of cervicomedullary junction
The patient's KPS score improved during follow-up of 26 months without recurrence

A



B

Fig 2. A 45 year old female patient presented with neck pain and left sided weakness
A: Preoperative MRI revealed ventro-lateral intradural extramedullary mass lesion on the craniocervical junction
The authors performed left postero-lateral approach with total tumor removal
Pathological report was a meningioma
B: Postoperative film revealed no residual tumor
The patient's KPS score improved during follow-up of 6 months with some degree of myelopathy

third of the clivus. The disadvantage is a poor access to lower part of the clivus.

A lateral extrapharyngeal route[19] is safe and effective for lower clivus and upper cervical spine. This approach has been applied mainly for extradural lesions. The surgical field is deep and limited rostrally and laterally by the internal carotid artery.

Postero-lateral approach[20-22] has been recognized for many years and referred to by different names, in essence it is one approach in which there are variations in degree of exposure and has advantage for intradural lesion[23,24]. These procedures enhance exposure of the ventral surface of the brain stem while markedly reducing the need for brain retraction. Drilling the condyle may be necessary in selected cases. Drilling less than one-half of the condyle causes no effect for cervical stability[25].

Although the authors found most tumors located in the anterior or antero-lateral regions, the approach depends on the extent of the tumor rather than the location of the tumor. The authors found that the anterior approach is highly suitable for tumors located extradurally with bony involvement. (12 in 16 operations) (Fig. 1). In cases of tumor located intradurally or

with nerve roots involvement, the authors used posterior or postero-lateral approach (13 operations) (Fig 2). Another factor we found in the authors' experience was that large tumors usually create a corridor for a simple approach and need less brain retraction.

The authors were able to achieve gross total resection in 68% of our cases at the first surgery (including staged surgery). Tumors with bony involvement have a tendency to fail gross total resection, need more operations and fusion procedures than tumors within dural cavities due to their extensive natures and destructive abilities.

Outcome KPS score in the present series did not change on immediate discharge status (68.4 vs. 70.4) but in long-term follow-up the authors found significant improvement (69.41 vs. 85.29).

In the present series, the most common complication was deficit of the lower cranial nerves, the 9^{th} and 10^{th} in particular. As other authors have also found[3,4], these deficits contributed to prolonged hospital stay for aspiration and weaning ventilator. The authors advocate for routinely preoperative evaluation for vocal cord and swallowing function and then re-evaluate in post operative phase for early management (placement of nasogastric, gastric tubes or tracheostomy).

The authors found two cases that had CSF leakage, they were both operated on by anterior approach, as watertight closure was not possible. Meningitis occurred in two cases (one case had CSF leakage) both from anterior approach procedures. Although there were no surgery-related deaths, five patients died during the follow-up period due to natural causes.

Conclusion

Craniocervical junction tumors have different pathological natures, growth patterns and neurological involvement; appropriate surgical approaches facilitate tumor removal with less surgical morbidities; nevertheless recurrent tumors occasionally occur, so long term follow-up is mandatory.

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ผลการรักษาเนื้องอกบริเวณรอยต่อศีรษะและกระดูกคอ

สุรชัย เคารพธรรม, ปกฤษณ์ จิตตภิรมย์ศักดิ์, รุ่งศักดิ์ ศิวานุวัฒน์, ไกรศรี จันทรา

วัตถุประสงค์*: เพื่อศึกษา อาการทางคลินิก ผลทางรังสีวิทยา และการผ่าตัดรักษาผู้ป่วยเนื้องอกบริเวณรอยต่อศีรษะและกระดูกคอย้อนหลังเป็นระยะเวลา 8 ปี*

วัสดุและวิธีการ*: โดยศึกษาย้อนหลังจากเวชระเบียนผู้ป่วยเนื้องอกบริเวณรอยต่อศีรษะและกระดูกคอ จำนวน 25 ราย ตั้งแต่ปี พ.ศ. 2539 ถึง พ.ศ. 2547 และติดตามผลการรักษาในเวชระเบียนผู้ป่วยนอกและโดยการสัมภาษณ์ทางโทรศัพท์*

ผลการศึกษา*: ผู้ป่วยเนื้องอกบริเวณรอยต่อศีรษะและกระดูกคอจำนวน 25 รายแบ่งเป็น เนื้องอกคอร์โดมา 9 ราย เนื้องอกเมนิงจิโอมา 8 ราย ซีสต์ 3 ราย เนื้องอกชวานน์โนมา 2 ราย แอนิวริสมอลโบนซีสต์ พลาสมาซัยโตมา และมะเร็งแพร่กระจายอย่างละ 1 ราย มีการผ่าตัดทั้งหมด 29 ครั้ง แบ่งเป็น ผ่าตัดจากทางด้านหน้า 12 ครั้ง ผ่าตัดจากทางด้านหลังเยื้องข้าง 9 ครั้ง และผ่าตัดจากทางด้านหลัง 8 ครั้ง สามารถตัดเนื้องอกออกได้ทั้งหมด 17 ราย เกือบทั้งหมด 6 ราย และตัดได้เพียงบางส่วน 2 ราย มีการผ่าตัดซ้ำในผู้ป่วย 6 ราย ค่ามัธยฐาน ของระยะเวลาในการติดตามผู้ป่วยหลังการรักษา 31 เดือนพบว่ามีการเพิ่มขึ้นของค่า คาร์นอฟกี้ เพอร์ฟอร์มแมนซ์ อย่างมีนัยสำคัญ

สรุป*: การเลือกการผ่าตัดที่เหมาะสมกับรอยโรคสามารถตัดเนื้องอกออกได้สำเร็จโดยมีผลแทรกซ้อนจากการผ่าตัดต่ำ อย่างไรก็ตามเนื้องอกบริเวณนี้สามารถกลับเป็นซ้ำได้ จึงต้องการการติดตามการรักษาระยะยาว*