

Intraoral and extraoral approach for surgical treatment of Eagle's syndrome: a retrospective study

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Abstract

Background

We evaluated the clinical characteristics and postoperative efficacy of 94 patients with Eagle's syndrome based on their clinical symptoms, imageological examination, and physical examination. Through the multi center clinical study of Tongji Medical College and Dalian Medical University, we found some characteristics of Eagle's syndrome in operation and imaging.

Methods

In total, 94 patients with Eagle's syndrome (treated from January 2010 to January 2019) were retrospectively enrolled. The postoperative curative effect was analysed by three surgical methods: styloid process resection through the external cervical approach, tonsillectomy + styloidectomy, and preservation of the tonsil for styloidectomy.

Results

The average length of the styloid process was 33 mm (range, 25–61 mm). The patients were followed up for 3–36 months (average, 15 months). Of the 94 patients, 20 underwent styloid process resection through the external cervical approach, 45 underwent tonsillectomy and styloidectomy, and 29 underwent styloidectomy with preservation of the tonsil. The treatment cured 45 (48%) cases, was effective in 31 (33%), and ineffective in 18 (19%). The SPSS 19.0 software was used to conduct a rank sum test, and there was no significant difference between the three types of operations ($H = 0.521$, $P = 0.771$).

Conclusions

Operation is an effective method for treating Eagle's syndrome. There were no significant differences between the effects of the intraoral and external cervical approaches. Imaging examination—especially CT scanning and 3D reconstruction of the styloid process—is very helpful for diagnosis, but not an absolute criterion for the selection of surgery protocol.

Background

Symptoms of Eagle's syndrome can be divided into two types: classic styloid syndrome and stylo-carotid-artery syndrome(1–3). Because of the variety of clinical manifestations of the disease, it can be easily misdiagnosed as chronic pharyngitis, cervical spondylosis, chronic tonsillitis, glossopharyngeal neuralgia, temporomandibular joint disorder(4, 5). The diagnosis mainly depends on the patient's clinical symptoms, physical examination (primarily via digital palpation of the tonsillar fossa), and imageological examination (X-ray and computed tomography [CT] scans of the styloid process, followed by a three-

dimensional [3D] reconstruction)(6, 7). In China, surgical treatment for this disease is generally performed by the otolaryngology department. However, the clinical symptoms of some patients are not solved through surgery(8). Therefore, this retrospective study aimed to evaluate the surgical treatment of this disease through statistical analysis of various postoperative outcome measures, providing a reference for the treatment of such patients in the future.

Methods

1. Data and patients

A total of 94 patients treated at the First Affiliated Hospital of Dalian Medical University (37 cases) and Tongji Hospital (affiliated to Tongji Medical College of Huazhong University of Science and Technology, 57 cases) from January 2010 to January 2019 were enrolled as research subjects. The patients were provided informed consent, and the relevant institutional review boards granted ethical clearance for the study. The selection criteria were as follows: 1) patients complained of either pharyngeal pain, pharyngeal foreign body sensation, unilateral neck shoulder radiation pain, or radiation ear pain; 2) a bony eminence was palpable in the fossa; 3) either the 3D CT reconstruction of the styloid process or the anteroposterior, lateral X-ray film of the styloid process met the diagnostic criteria, and the length of the styloid process was > 25 mm. Patients with severe cardiovascular diseases, neuropsychiatric diseases, and neuromuscular diseases were excluded. There were 43 men and 51 women with an average age of 45 ± 8.06 years (range, 25–68 years). The disease course was 3–60 months. Of the 94 cases, 63 had unilateral disease and 31 had bilateral diseases; 20 patients underwent styloid process resection through the external cervical approach, 45 underwent tonsillectomy as well as styloid process shortening, and 29 underwent styloid process shortening along with tonsil preservation.

2. Imageological examinations

Imageological examination was performed with a Siemens 64-slice double helix machine (Siemens, Munich, Germany). The patient was put in a supine position with the mandible raised, such that the auditory orbital line was perpendicular to the scanning table. The measurement method used was as follows(7, 9): based on the 3D CT reconstruction image, the length of the styloid process was measured as the distance between its root and the distal end. On the anteroposterior image, a perpendicular line was drawn parallel to the skull base, and the angle between the centre line of the styloid process and the perpendicular measurement line was noted as the inward angle(10). An acoustic orbital line (a perpendicular line connecting the upper edge of the outer ear hole and the lower edge of the orbit) was drawn on the reconstructed image, and the internal deviation angle was measured as the angle between the perpendicular line and the centre line of the styloid process. If the length of the styloid process was greater than 25 mm(11), the styloid process was considered too long. In this case, the angle of inclination (inward or forward) was greater than 25° and was called the abnormal azimuth angle. The anteroposterior, lateral X-ray film of the styloid process was examined as follows: for scanning, the patient (either standing or reclining) was instructed to keep their head in a lateral position and slightly tilt

their body. After scanning, the data was transmitted to a picture archiving and communication system workstation to measure the azimuth angle and the length of the styloid process using the same method as before.

3. Surgical indications

At present, patients diagnosed with Eagle's syndrome are not uncommon. Some patients with a long styloid process undergo conservative treatments such as non-steroidal anti-inflammatory drugs and glucocorticoids(12), although it has a short-term effect, can easily relapse. The inclusion criteria for surgery were the same as the selection criteria for this study. All three criteria (1, 2, and 3) were considered necessary conditions and were strictly observed, and the imaging data were carefully analysed. Although the X-ray of the styloid process could show the long styloid process bone, the 3D reconstruction of the styloid process was used as the final examination method in this study to determine surgical method. This is because the 3D CT scan of the styloid process is more accurate in displaying the details, and accurate judgement of deflection, interruption, and ossification of the hyoid ligament(10).

4. Selection of the operation method

The patients were placed under general anaesthesia and the appropriate surgical method was selected based on their oropharyngeal palpation, 3D CT reconstruction of the styloid process, and specific selection criteria: 1) for patients in whom the styloid process bone could be touched directly during intraoral palpation, or in whom the distal part of the styloid process could not be directly touched but the CT scan showed that the bone inclined toward the oropharynx and its distal part was relatively close to the oropharynx cavity (which does not cause excessive injury in the parapharyngeal space), styloid process resection was selected for transoral surgery. Whether tonsillectomy was performed depended on whether the patient's tonsil was too large to affect the surgical incision; 2) for patients whose styloid process could not be touched through the mouth or could be touched under the jaw, or when the CT scan showed that the inclination angle was not large, the external cervical approach was selected. The selection of appropriate surgical methods can lower operation time, injury, postoperative pharyngalgia, and the probability of complications.

4.1. Bimanual examination

The index finger of one hand was placed in the affected tonsillar fossa, and the other finger was placed behind or under the ipsilateral ramus of the mandible(13). Of 74 patients who underwent surgery using the intraoral approach, 49 patients could either touch the hard cord in the unilateral or bilateral tonsillar fossa, or felt tenderness, fullness, or other discomforts at the touchpoint; 10 patients could touch the hard cord behind or under the ramus of the mandible. The other 15 patients could not touch any abnormality. Specific surgical methods: 1) the tonsillar fossa approach: if the tonsil was too large and affected the exposure of the whole styloid process, tonsillectomy was performed first. Based on the position and course of the styloid process, a sickle knife was used to cut the mucosa of the palatoglossal or palatopharyngeal arch in order to separate the superior pharyngeal muscle and expose the distal styloid

process. Following this, the styloid process periosteum was cut, and soft tissues such as the muscle and styloid hyoid ligament were excised. The styloid process was covered with a small ethmoid curette from the free end and walked along the styloid process (which was clipped with a vascular clamp), then cut and removed with bone biting forceps. The cut-off length was 10–38 mm. If the residual styloid process was long, it was further stripped and shortened. During the operation, it was necessary to clamp the broken end of the styloid process with a vascular clamp as it is difficult to find once it slips. Small incisions were not sutured; however, large incisions were sutured with 3 – 0 absorbable sutures to eliminate large spaces and prevent parapharyngeal space infection. In patients with tonsillectomy, it was not necessary to suture the tonsillar fossa completely. 2) Using the palatoglossal arch- soft palate approach, the mucosa was cut between the medial sides of the opsignes on the palatoglossal arch, and the incision was extended to the upper soft palate to a length of 2.5 cm. The remaining steps were the same as those described for surgical procedure 1.

4.2. External cervical approach

An arc-shaped incision was made around the mandibular angle, approximately 1–2 cm from the posterior edge of the ramus to the inferior edge of the mandible. The length of the incision was approximately 4 cm. The skin was cut, and the layers of subcutaneous tissue and platysma were separated. The parotid masseteric fascia was exposed to protect its superficial layer and to cross the facial nerve. The mandibular angle was fully exposed, and the styloid mandibular ligament was located. The styloid process was located, and the attached mandibular ligament of the styloid process and styloid hyoid ligament was cut. The attachment site of the styloid process was approached using the ethmoid curette, and a small pair of bone biting forceps was inserted along the styloid process.

5. Statistical analyses

The SPSS software (IBM SPSS Statistics for Windows, Version 19.0; IBM Corp., Armonk, NY: IBM Corp.) was used to conduct rank sum tests, and P values < 0.05 were considered statistically significant.

Results

The average length of the styloid process was 33 mm (range, 25–61 mm). The patients were followed up for 3–36 months (average, 15 months). The visual analogue scale score was 4.37 ± 0.29 before the operation and 1.63 ± 1.12 after the operation, and the difference was statistically significant ($P < 0.05$). Surgical cure was defined as complete disappearance of symptoms, effective operation was defined as significant remission of symptoms, and ineffective operation was defined as no remission of symptoms (Table 1 for specific data). The main complaints were swallowing pain, pharyngalgia, foreign body sensation in the pharynx (with one of them) and the cure rate was 70%. Most of the above symptoms were significantly relieved after wound healing, and the main complaints were headache, earache, dizziness and tinnitus (with one of them) The cure rate was 58%, Most of the aforementioned symptoms were relieved after surgery, and the postoperative symptoms of these patients were gradually relieved after 3–6 months. A small amount of tonsillar haemorrhage occurred in 2 patients one week after the

operation, but healed without special treatment, no parapharyngeal space infection or facial nerve paralysis. Persistent dry throat and pharyngeal foreign body sensation were reported in individual cases but were tolerable without special intervention. The results of the rank sum tests were not statistically significant ($H = 0.521$, $P = 0.771$).

Table 1
The therapeutic effect of three surgical methods

THERAPEUTIC EFFECT	EXTERNAL CERVICAL APPROACH	TONSILLECTOMY + STYLOIDECTOMY	PRESERVATION OF THE TONSIL FOR STYLOIDECTOMY	TOTAL
Cure	10	20	15	45(48%)
Effective	7	15	9	31(33%)
Ineffective	3	10	5	18(19%)
Total	20	45	29	94

Case Report

Case 1

a 54 years old female patient, complained of "pharyngalgia for half a year". This case was a typical styloid process truncation with preservation of tonsils through oral approach. Through CT, we could clearly see the length, anteversion and inclination of styloid process, and judge that styloid process was toward the oropharynx cavity. Palpation through tonsil fossa also further confirms that styloid process was too long, and bone of styloid process could be seen protruding to tonsil fossa. Such typical patient was relatively rare. After operation, the wound in the mouth was very small and healed quickly. The patient's pharyngalgia was obviously improved. (Fig. 1)

Case 2

a 52 years old female patient, was admitted with the chief complaint of "foreign body sensation in throat for one year". This case was a special type. The CT of the patient showed that the styloid process bone was intact, and the styloid process was not touched in the tonsil fossa and neck. During the operation, the styloid process bone was not firmly fixed. During the operation, the styloid process could swing slightly, and the thickness was uneven, the styloid process bone was discontinuous. One month after the operation, the patient's symptoms improved. (Fig. 2)

Case 3

a 47 years old male patient, was admitted with "pharyngalgia for half a year", a case of extremely long styloid process on both sides. CT accurately measured the true length of styloid process, which was consistent with the intraoperative findings. It could be seen that CT three-dimensional reconstruction has

diagnostic value for the disease. Two weeks after operation, the symptoms improved significantly. (Fig. 3)

Case 4

a 37 years old male patient, complained of "pharyngalgia for a year", a case of styloid process bone interruption. Both styloid processes were interrupted in CT. After cutting the mucosa in the opening, the distal styloid process was not fixed. During the operation, there was a cord like interruption at the junction of the anterior and middle third of styloid process, and the bone was discontinuous. We found that if the styloid process was obviously interrupted in CT, the interruption could be seen during the operation. If the styloid process bone interruption was not shown by CT, bone interruption could occur in individual cases. The patient's symptoms relieved 3 months after operation. (Fig. 4)

Discussion

The styloid process is a slender cylindrical bone located at the junction of the mastoid and petrous part of the temporal bone. It originates from the anteromedial side of the stylomastoid foramen(14). The styloid process and its associated muscles (the styloid hyoid muscle, styloid pharyngeal muscle, and styloid tongue muscle) divide the parapharyngeal space into two parts: the anterior part (the anterior parapharyngeal space) and the posterior part (the posterior parapharyngeal space). The anterior parapharyngeal space is relatively small and contains ascending pharyngeal arteries and veins, whereas the medial side is close to the superior pharyngeal constrictor and palatine tonsil. Because of this anatomical relationship, abnormal anatomical morphology of the styloid process can cause pharyngeal symptoms such as pharyngalgia, foreign body sensation in the pharynx, and cough. The retro-parapharyngeal space is large and contains the internal carotid artery, veins, V - VI cranial nerves, and cervical lymph nodes, cause headache, neck and shoulder pain, tinnitus. Abnormal styloid process development or ossification of the styloid hyoid ligament can lead to a long styloid process (> 25 mm), abnormal angle, and abnormal shape. The normal inclination angle of the vertical line between the styloid process and the plane of the skull base plane in normal adults is approximately 25° forward and inward; if the angle is > 40° or < 20°, it is considered abnormal(7). Because the anatomical variation of the normal styloid process is relatively large, a long styloid process may not cause clinical symptoms. Therefore, both the length of the styloid process and the angle of the styloid process should be considered by clinicians. Abnormality in the styloid process shape and orientation or an abnormally long styloid process may not have the corresponding clinical symptoms of styloid process syndrome.

In this study, 94 patients were enrolled, of whom most complained of pharyngalgia, foreign body sensation in the pharynx, and neck pain. Among them, 3 patients were admitted to the hospital with otodynia as the main complaint. Most patients have been diagnosed with chronic pharyngitis, chronic tonsillitis, or otitis before surgery, Eagle's syndrome can also cause these symptoms, which makes it difficult to differentiate from other diseases that can cause neck or facial pain and abnormal sensations. Special examination—especially tonsillar fossa and neck palpation—has become the key diagnosis in

such cases. If the long styloid process can be touched or causes obvious tenderness, foreign body sensation, and fullness, the diagnosis is confirmed. Imaging technology, especially CT scanning and 3D reconstruction of the styloid process, can clearly show the styloid process length, shape, internal deflection angle, and ligament ossification(7). These examinations allow relatively accurate observation and measurement, which is of great help in guiding surgery and clinical research.

This study found that there was no significant difference in the efficacy of intraoral and extraoral approaches, but the postoperative pain lasted 5 days longer in patients who underwent surgery using the extraoral approach compared to those who underwent the intraoral approach. The average postoperative hospital stay of patients who underwent surgery with the intraoral approach was 3 days, shorter than the patients who received the extraoral approach for 3 days. A small amount of tonsillar haemorrhage occurred in 2 patients one week after the operation, but healed without special treatment, no parapharyngeal space infection, or facial nerve paralysis. Persistent dry throat and pharyngeal foreign body sensation were reported in individual cases but were tolerable without special intervention. We found that imaging examination—especially CT scanning and 3D reconstruction of the styloid process—is very helpful for diagnosis, but not an absolute criterion for the selection of surgery protocol. In some cases, although CT scans showed that the bone was continuous, it was found to be interrupted during the operation, in these cases, the effect of operation were often not very good. In some cases, CT scans showed that the styloid process was too long, but intraoperative exploration, especially in the case of sufficient exposure to the external cervical approach, the styloid process was not too long, which may be related to the incomplete ossification of styloid ligament. In this study, we set the styloid process length at > 25 mm; however, most other related studies set it at > 30 mm(15), the main consideration is that the length cut-off is not universal; some patients have a styloid process length of 25–30 mm, but their symptoms are obvious, the hard styloid process can be felt on tonsillar fossa examination, and surgical treatment is also feasible. Therefore, imaging examinations can only be used as an important auxiliary tool in clinical practice and cannot become a definitive diagnosis.

Styloid process truncation has a clear curative effect on patients with Eagle's syndrome, and can significantly reduce the symptoms of pharyngalgia, swallowing pain, cervicodynia, earache, and pharyngeal foreign body sensation(6, 16). However, the symptoms of patients with tinnitus, headache, and dizziness are not significantly relieved. The surgeon in this study is proficient in the above three surgical methods, who presents no technical differences. However, some scholars believe that the lateral cervical region approach can ensure the safety of operations and reduce the probability of postoperative complications(17), it is also suggested that the styloid process truncation should be performed through the external cervical approach because it allows clear surgical vision and less damage to peripheral blood vessels and nerves(13, 18). However, extensive facial anatomy and long operation times can easily cause epidermal nerve injuries, such as sensory abnormalities of the great auricular nerve and facial nerve branch injury(19). Open surgery should be selected when the styloid process grows towards the lateral cervical region, so as to avoid excessive operation in the parapharyngeal space, longer operation times, and higher operating costs. Through the surgical procedures used in this study, the wound in the mouth had a small internal diameter and the recovery time was short (6 days after the operation), there was no

incision or scar on the outside. Although the field of vision was smaller, the styloid process could be fully exposed in most cases(20). These selection criteria can help reduce the risk of parapharyngeal space infection and nerve injury(21). In general, it is very important to choose the appropriate surgical method according to the experience of the operator and the different pathological characteristics of the patients.

Conclusion

Surgery is an effective method for treating styloid process syndrome. However, in view of the imprecise curative effect of surgery in some cases and postoperative discomfort (dry throat and foreign body sensation in the pharynx), and the imaging examination was also inconsistent with the intraoperative findings, it is very important to strengthen the preoperative evaluation (length and angle judgement of the styloid process via imaging, digital palpation), select surgical protocols according to the symptoms. Our results show that there were no significant differences between the intraoral and extraoral surgical methods. However, based on the surgeon's proficiency in the corresponding surgical methods(22), the unnecessary operation should be avoided, the operation time should be minimized, and the adjacent anatomical structures should be avoided during surgery. Through ten years of retrospective study, we collected a large number of cases, including many typical cases, and showed them to readers through images and statistical data. It's a flaw not to record the photos of the operation in the typical cases.

Abbreviations

CT
computed tomography
3D
three-dimensional

Declarations

Ethics approval and consent to participate

This study was conducted in agreement with the Ethics Committee of Tongji Hospital Affiliated to Tongji Medical College and The First Affiliated Hospital of Dalian Medical University, All participants had provided a written informed consent

Consent for publication

All participants gave written informed consent for their personal and clinical data publication along with written consent for publication for any identifying images in this study, according to the China laws.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

Competing interests

None.

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Authors' contributions

ZBW and YL: study conception and design, acquisition of data, YKS analyzes and processes data. JW: drafting of the manuscript. All authors have read and approved the manuscript.

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guidelines

All procedures were performed in accordance with relevant guidelines.

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Figures

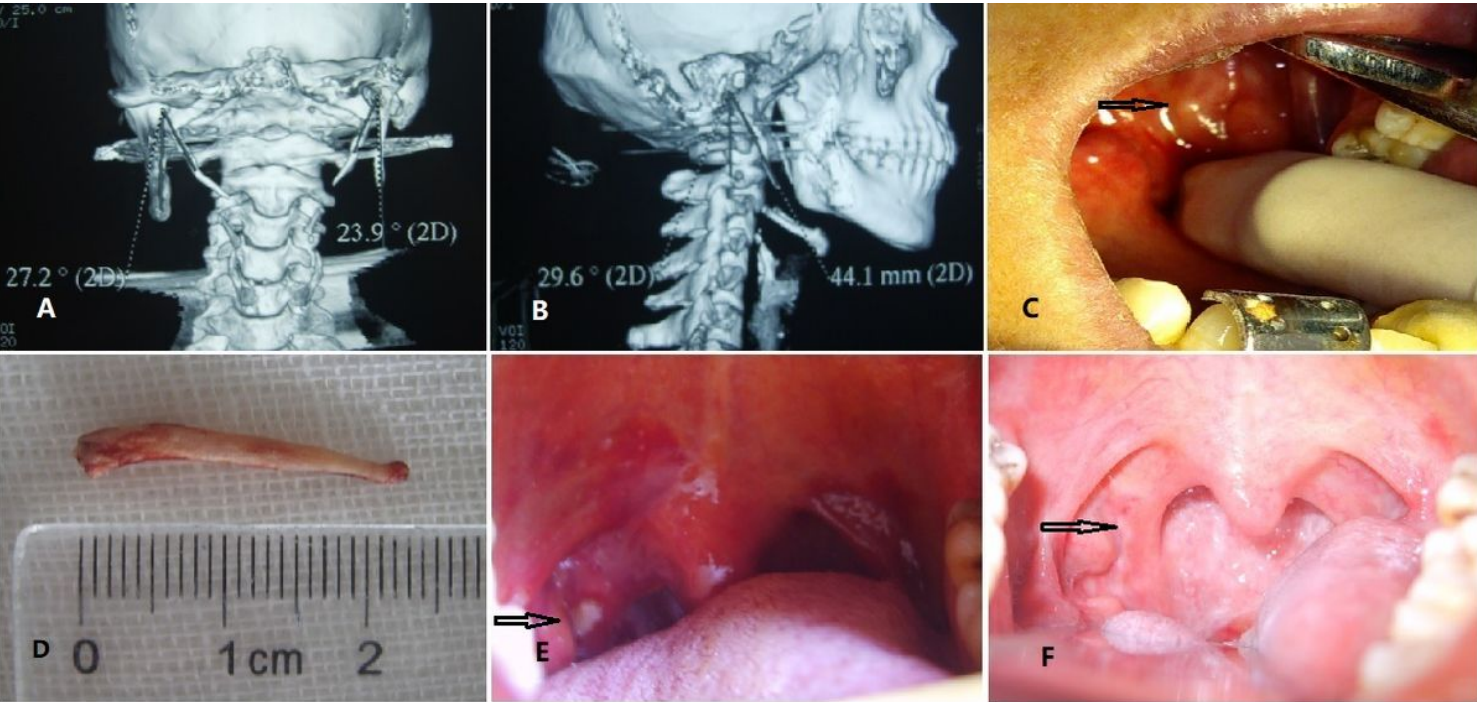


Figure 1

A and B: CT examination shows the bilateral styloid process growing downward, inward, and forward. The length of the right styloid process is 44.1 mm, the inclination angle is 27.2°, the anteversion angle is 29.6° to the superior margin of the axis, the lower segment of the right styloid process bends, and the inclination angle increases to the medial. C: During the operation, the right tonsil fossa is palpated, and a hard rod-like protrusion is found. D: The styloid process is completely truncated during the operation, and no ligament or soft tissue attachment is left around the styloid process. E and F: wound recovery on the third and sixth days after the operation. There are small wounds in the mouth with a little white membrane attached to the surface, and the surrounding mucosa is intact without redness, swelling, or congestion. The wound recovered to six days after the operation.

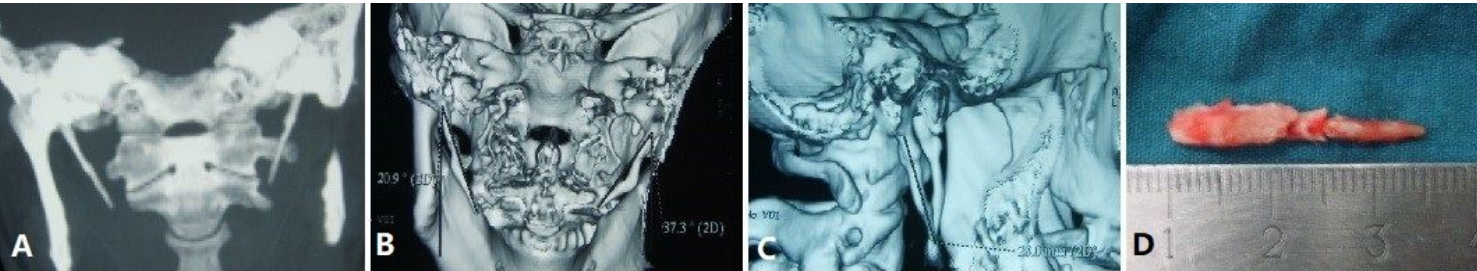


Figure 2

A-C: CT examination of the neck, the styloid process on the left side is 28 mm in length, with an inclination of 37.3° and an anteversion of 9.3° . The CT scan shows that the styloid process is continuous without interruption; D: a strip-like interruption touches during the operation.

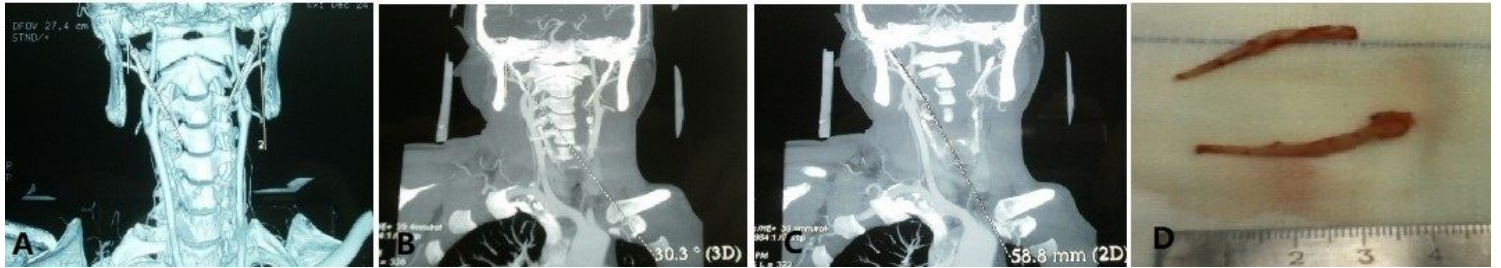


Figure 3

A-C: The length of the right styloid process is 58.8 mm and the inclination angle is 30.3° ; the length of the left styloid process is 55.6 mm and the inclination angle is 26.7° , CT can clearly show the course and length of styloid process. D: Bilateral styloid process was completely truncated during operation.

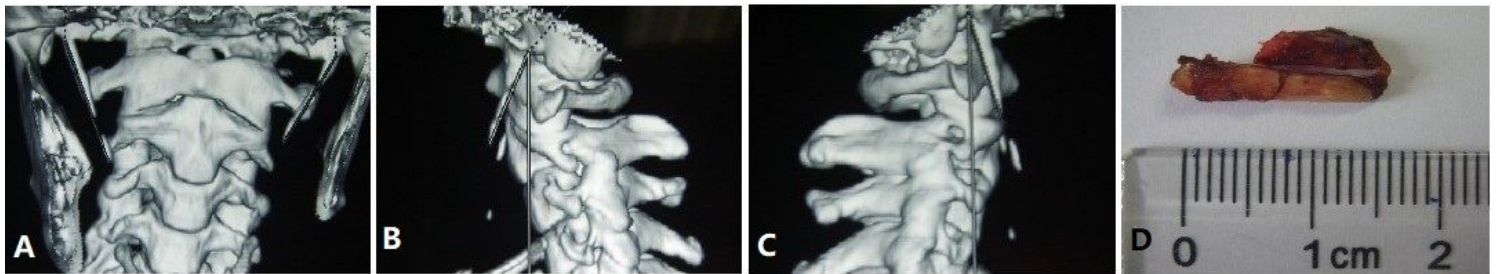


Figure 4

A-C: the left styloid process is 35.73 mm, and the right styloid process is 37.36 mm. Both styloid processes are interrupted in the CT scans. After cutting the internal mucosa, the distal styloid process is not fixed. D: A cord-like interruption is present at the junction of the anterior and middle third of the styloid process, and the bone is discontinuous.