



Case Report

Indirect carotid–cavernous sinus fistula following mechanical thrombectomy: A case report of a rare iatrogenic injury with progressive presentation

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ABSTRACT

Introduction: With the increasingly common operation of mechanical thrombectomy (MT) in acute cerebral infarction cases, iatrogenic CCFs were occasionally reported. All of cases reported type A CCFs, and patients were presented with either asymptom from generation of fistula to duration of postoperative follow-up or distinct presentations at once after MT.

Case presentation: A 48-year-old postmenopausal female, without history of systemic hypertension and diabetes mellitus, underwent an operation of MT outside our institution about half a year ago. An intraoperative DSA showed an iatrogenic low-flow fistula between meningohypophyseal trunk and ICA. After 4 mouths' postoperative conservative observation, patient's presentation progressed from asymptom to serious optic signs. The patient underwent *trans*-arterial interventional occlusion. On postoperative day one, visual presentations of patient relieved significantly.

Discussion: We discuss the reason for possibility of iatrogenic injury to meningohypophyseal trunk and clinical progressive presentation. A sudden swerve just beyond derivation of meningohypophyseal trunk is prone to being damaged by a misguided guide wire. The progression of clinical presentation, as a focal point in our case, is not reported in iatrogenic before, but some studys still find that spontaneous dural CCFs are inclined to occur in middle-aged or elderly women, especially in postmenopausal women, so age and sex are regarded as background factors of progressing. In addition, the change of drainage route is an immediate cause of progressive presentations.

Conclusion: We expect that when a manipulation of MT is conducted leading an iatrogenic CCF, our neuro-interventionist should maintain appropriate vigilance on sex, age, menstrual history and medical history, then take an earlier and timely interventional measure.

1. Introduction

With the increasingly common operation of MT in acute cerebral infarction cases, iatrogenic CCFs were occasionally reported. To the best of our knowledge, there are 7 reported cases [2]. Among the cases following MT, 3 patients appeared asymptomatic when they were hospitalized; 3 patients were presented with obvious clinical presentations involving chemosis, proptosis, hyperemia and cerebral nerve palsy; and one patient died 2 days postoperatively [2,3]. All of cases reported type A CCFs [2], and patients were presented with either asymptom from generation of fistula to duration of postoperative follow-up or distinct presentations at once after MT. We present a rare case of a 48-year-old

female patient with progressive clinical presentations due to an iatrogenic fistula between meningohypophyseal trunk and CS following MT. This case has been reported following the SCARE criteria [12].

2. Case presentation

A 48-year-old postmenopausal female patient, without history of systemic hypertension and diabetes mellitus, underwent an operation of MT outside our institution about half a year ago. An iatrogenic injury was generated during the procedure. Intraoperative DSA was proof of a low-flow indirect CCF draining through paired inferior petrous sinus (Fig. 1). Postoperatively, because no apparent clinical symptoms and

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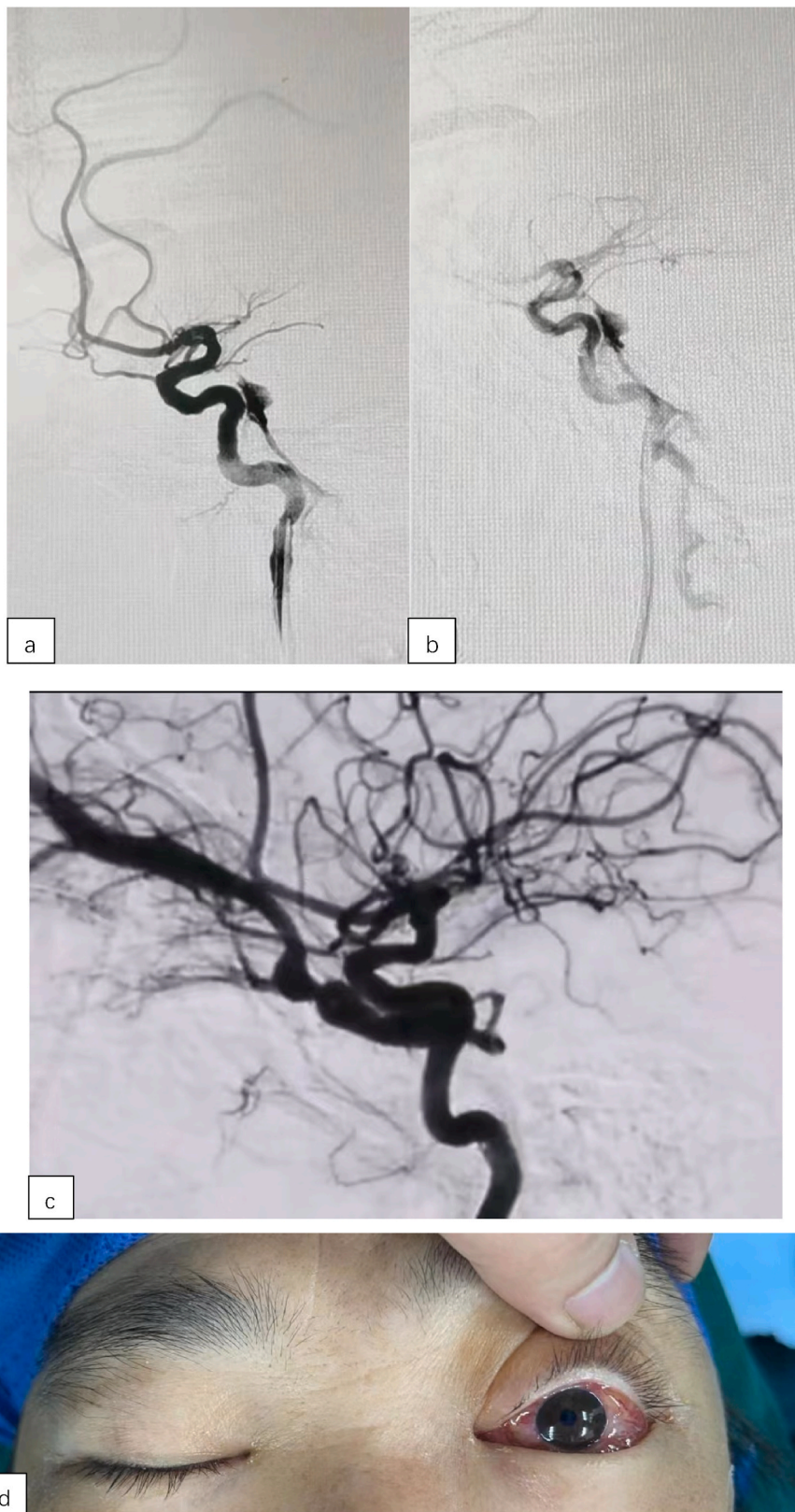


Fig. 1. Barrow type B CCF and the alternation of drainage route. a, b Selective ICA injection and lateral projection. A low-flow indirect fistula drains only through paired inferior petrous sinuses, which is the reason why our patient is asymptomatic. c, d After four months, with drainage route diversion, significant ocular symptoms were facilitated due to the thrombosis of initial route, and only drainage through paired superior ophthalmic veins was detected.

signs was presented, a conservative observation strategy was taken. 2 months ago, progressing visual impairment arose. Physical examination revealed a severe chemosis and hyperemic conjunctiva. After admission, a cerebrovascular DSA was immediately arranged and showed a clear connection between meningo-hypophyseal trunk and CS. In addition, we found a drainage route through paired superior ophthalmic veins (Fig. 1). Her treatment regimen was *trans*-arterial interventional occlusion (Class 4 operation according to our institution) by Dr. Zhang. Following conventional femoral artery puncture, a Maraton microcatheter was navigated over a microwire directly into meningo-hypophyseal trunk. An intraoperative DSA was manipulated to confirm the fistula location. Then the microcatheter was not guided into further place through microwire remodeling. Onyx liquid embolic system was injected at meningo-hypophyseal trunk. With partial liquid reflux, alternation between injection and pause was operated to possibly ensure liquid embolic system an intensive modeling. The operation was finished successfully and fistula was not detected by DSA. On post-operative day one, visual presentations of patient relieved significantly (Fig. 2). Through regular clinics follow-up of patient, her visual impairment relieved completely and physical examination manifested conjunctival injection and chemosis (–), cerebral nerve palsy (–). She remained asymptomatic at half-a-year follow-up.

3. Discussion

For better discussion and investigation about evaluation and treatment of patients with CCFs, an anatomical-angiographic classification was proposed by D L Barrow et al. According to Barrow classification, CCFs are categorized either direct fistula (type A) or indirect fistula (type B, C, D). Type A is a high-flow fistula communicating between CS and trunk of ICA. The remainder of categories are all low-flow fistulas including type B connecting CS with ICA meningeal branches, type C communicating CS with ECA meningeal branches, and type D linking CS with meningeal branches of ICA and ECA [1]. Our case reports for the first time a definite type B indirect CCF following MT between

meningo-hypophyseal trunk and CS. We deem that the fistula is caused by misguided and forcible manipulation of operator. Because there was a sudden swerve just beyond derivation of meningo-hypophyseal trunk, the acme of this position is inclined to be damaged by a mistakenly entered guide wire [5]. As a result, we suggest our neurointerventionist should operate softly and prudentially during procedure. Especially, when we advance into meningo-hypophyseal trunk or other unexpected subbranches, we need to confirm through angiography if a perforation is generated by our inappropriate manipulations.

Unlike all reported cases before, our patient's presentations became emerging and progressing after 4 months' postoperative asymptomatic and physical examination (–). The improvement of presentations is a focal point noticed by us in our report. Though progressive symptom in iatrogenic CCFs is not reported before, some studies still find that spontaneous dural CCFs are inclined to occur in middle-aged or elderly women [6,7]. Particularly the risk of incidence in postmenopausal women is 7 times higher than that in men [7]. Apart from sex and age, there are also certain risk factors including systemic hypertension, diabetes mellitus, atherosclerotic vascular disease, connective tissue disease and minor trauma [6–10]. These predisposing factors are supposed to be regarded as background of progressive symptoms. In addition, the change of drainage route is also a significant reason for progression. We think the alternation as well as the original route is predictable. The drainage route is dependent on arteriovenous relationship. Because of ICA passing, CS is divided into anterolateral and posteromedial domains communicating with each other through several input and output veins. According to the anatomical configuration, we are able to predict that anterior fistula is apt to drain anteriorly and posterior fistula is disposed to drain posteriorly. The theory can explain why our fistula initially drained posteriorly into paired inferior petrous sinus. Nevertheless, when the preferable pathway becomes thrombosed, the fistula alters to anterior drainage, leading to progressive ocular manifestations [6]. Meanwhile, most of investigators believe that the generation of dural CCFs is a response to venous thrombosis, which means to provide an output pathway [11]. On the basis of the theory, it represents when a



Fig. 2. Postoperative angiography and clinical relieve. a Selective ICA injection and lateral projection. Occlusion of fistula is complete. b An apparent clinical relieve can be seen on the second day after *tans*-arterial interventional operation.

fistula has been generated intraoperatively, thrombosis in CS may promote the opening, resulting in an increase and persistent blood flow through the fistula. Our clinical deterioration is decided by the increasing [6]. In the setting of predisposing factors, thrombosis not only leads to the alternation of drainage route but also promotes the bloodstream through the fistula, all of which are involved in motivation of progressive presentations.

Presentations related to CCF are in connection with orientation of venous drainage, the velocity of blood flow through the shunts and a venous reflux due to thrombosis [2]. It is not difficult for our experienced clinicians to diagnose either direct or indirect CCFs clinically [4]. With regard to treatment for indirect CCFs, there are several approaches including observation, conservative or domiciliary technique and endovascular intervention. Some studies consider that 70% of indirect CCFs is prone to spontaneous closure [4]. Traditionally, invasive treatment is not regarded as a preference in most of low-flow fistula. However, we aim to turn our neurologists' spotlight on elderly and postmenopausal female whether or not complicated by other predisposing factors. In such female patients, low-flow fistula is prone to progress. Thus, if fistula generated during procedure, we don't recommend these patients to undergo a conservative treatment. In case of life-threatening progression, interventional manipulation is required. In addition, we even deduce that a minor intraoperative injury of vascular wall may be regarded as an iatrogenic motivation to cause an occurrence of delayed fistula, despite there was no report before. Therefore, if there are inappropriate operational manipulations possibly damaging vascular wall, we suggest our neurointerventional operators should allow elderly female patients following MT to be followed up energetically and regularly, although perforation may be not generated at that time through angiography confirmation.

4. Conclusion

In conclusion, with incremental manipulation of MT, we should focus on CCF as an intraoperative complication. Our case presents a rare iatrogenic type B indirect CCF following MT with progressive clinical presentations. We expect that when a manipulation of MT is conducted leading an iatrogenic CCF, our neurointerventionist should maintain appropriate vigilance on sex, age, menstrual history and medical history, then take an earlier and timely interventional measure.

Ethics approval

No ethical approval necessary.

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Author contributions

Sihao Li, Changwei Zhang: Writing, review and editing of the manuscript, Sen Lin, Lunxin Liu, Changwei Zhang: Provide analysis of the angiography image, Changwei Zhang: Diagnose and treatment of patient, Changwei Zhang, Ting Wang: Supervised the writing of manuscript.

Registration of research study

Our paper is a case report; no registration was done for it.

Consent of the patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request".

Guarantor

Changwei Zhang.

Declaration of competing interest

The authors declared no potential conflicts of interests with respect to research, authorship and/or publication of the article.

Appendix A. Supplementary data

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.amsu.2022.104130>.

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